

The Oophorectomy Decision Explorer

A decision support intervention to facilitate
deliberation and coping efforts
in women at increased risk of ovarian cancer

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Abstract

Background: Traditionally, coping and decision making have been viewed as separate concepts. However, analysis of the role of emotions during decision making in healthcare suggests that coping with health threats, and associated emotions, should be viewed as an integral part of deliberation processes. This thesis reports on the development of a framework that merges deliberation and coping processes. Subsequently, this framework is operationalised by adapting it to specifically describe deliberations about risk-reducing salpingo-oophorectomy (RRSO) by women at increased genetic risk of ovarian cancer.

Methods: A narrative review of the literature on decision making and coping theories informed the development of a novel framework that integrates theories from both fields. A multi-methods approach, which included a systematic literature search and qualitative methods, examined women's decision making about RRSO and informed adaptation of the framework to specifically describe such decisions. The adapted framework was then used to develop a decision support intervention for women at increased risk of ovarian cancer in the UK. Incremental prototypes of the intervention were reviewed by a group of stakeholders and usability of the final prototype was tested using cognitive interviews.

Results: The Coping in Deliberation (CODE) framework describes deliberations as multi-step appraisal and coping processes. The framework was successfully adapted to decisions about RRSO and used to develop a two part patient decision support intervention, consisting of a brief paper-based tool (Option Grid) and a longer website (the Oophorectomy Decision Explorer, OvDex).

Conclusions: Emotions and coping are integral parts of deliberations in healthcare and the CODE framework, which acknowledges the importance of these concepts during deliberations, can be used to guide the development of patient decision support interventions. Future research should apply this framework to other healthcare decisions. The intervention developed in this thesis requires field testing to assess its impact before implementation in clinical practice.

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List of Abbreviations

| | | |
|-------|---|---|
| CAD | - | Coronary Artery Disease |
| CASP | - | Critical Appraisal Skills Programme |
| CODE | - | Coping in Deliberation (Framework) |
| CTOCS | - | Collaborative Trial of Ovarian Cancer Screening |
| FOCS | - | Familial Ovarian Cancer Screening |
| GCOS | - | Genetic counselling outcome scale |
| HBOC | - | Hereditary breast and ovarian cancer |
| HNPCC | - | Hereditary non-polyposis colorectal cancer |
| HRT | - | Hormone replacement therapy |
| IPDAS | - | International Patient Decision Aid Standards |
| MAU | - | Multi-Attribute Utility Theory |
| NICE | - | National Institute for Health and Care Excellence |
| NPT | - | Normalisation process theory |
| OCP | - | Oral contraceptive pill |
| ODSF | - | Ottawa Decision Support Framework |
| PCC | - | Patient-centred care |
| PROM | - | Patient reported outcome measure |
| RRM | - | Risk-reducing mastectomy |
| RRH | - | Risk-reducing hysterectomy |
| RRSO | - | Risk-reducing bilateral salpingo-oophorectomy |
| SDM | - | Shared decision making |

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1 Introduction and Thesis Overview

1.1 Introduction

Previous research suggests that emotions and coping are integral parts of decision processes and that emotions influence people's predictions of how different options might affect their life (Balneaves and Long 1999, Gilbert et al. 2002, Wilson and Gilbert 2003). It has also been proposed that medical decisions are in fact coping behaviours aimed at regulating a health threat and associated emotions (Balneaves and Long 1999, Luce 2005, Power et al. 2011). To date, however, decision making theorists have largely ignored the role of emotions and coping in healthcare decision making. Patient decision support interventions are tools designed to facilitate decision making processes, which should ideally be grounded in theory (Charles et al. 2005, Durand et al. 2008). As coping is an integral part of decision making, coping theories could be used alongside decision making theories to support the development of such interventions. However, as decision making theorists have largely ignored the role of emotions and coping, so have decision support developers largely ignored coping theory (Elwyn et al. 2011b). In fact, many decision support interventions have been found to be atheoretical (Durand et al. 2008). There is a need for a framework that considers emotions and coping as intrinsic parts of deliberation processes and that can be used as a basis for the development of decision support interventions. This thesis aims to propose such a framework to describe deliberations about preference-sensitive healthcare choices and to guide the development of more holistic decision support materials.

This chapter will:

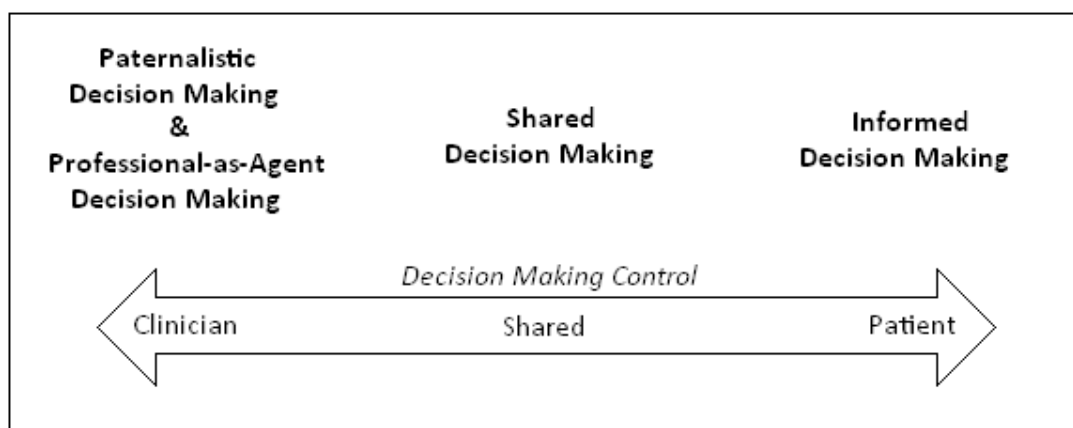
- i. provide a definition of shared decision making and patient decision support interventions
- ii. give an overview of coping theory and coping interventions
- iii. offer an introduction to ovarian cancer risk and risk management options
- iv. outline the objectives and structure of the thesis.

1.2 Shared decision making in healthcare

1.2.1 Shared decision making

Traditionally, medical decision making has been described using the 'paternalistic' decision making model, in which the clinician decides on the best course of action with very limited input from the patient (Charles et al. 1997, Charles et al. 1999, Emanuel and Emanuel 1992). Since the introduction and promotion of the idea of patient centred care (PCC), other models have been put forward that assume that both the clinician and the patient possess expertise that is important for decision making (Figure 1.1).

Figure 1.1 - Clinical decision making styles



These models describe approaches to decision making that require the exchange of information. The clinician is seen as the expert regarding treatment options and their expected efficacy, while the patient is considered the expert on their personal values and on how any treatment option might affect their life (Charles et al. 1997, Hurley et al. 1992). In the 'Informed', as well as the 'Professional-as-Agent', decision making models, information is transferred from one party to the other, so that either the patient or the physician possess both types of expertise (i.e. relevant medical knowledge and patient values). After considering this knowledge, either the patient or the professional can then make a decision independently. In 'Informed' decision making, the clinician provides medical information and empowers the patient to make a decision, whereas in 'Professional-as-Agent'

decision making, the patient provides information about their preferences and empowers the clinician to make a decision. However, according to Charles et al. (1997) neither of these models are truly representative of 'shared' decision making (SDM).

SDM has previously been defined as "an approach where clinicians and patients share the best available evidence when faced with the task of making decisions, and where patients are supported to consider options, to achieve informed preferences." (Elwyn et al. 2012, page 1361). SDM requires information sharing between two or more parties, at a minimum including a patient and their clinician, followed by making a shared decision that is ideally agreed by all parties (Charles et al. 1997, Elwyn et al. 2000). Generally, the clinician provides medical information on the condition and on the risks and benefits of available options, including probabilities and uncertainties, while the patient contributes their view on how they perceive those risks and benefits in the context of their personal goals and values (O'Connor et al. 2004). However, clinicians also have values that they bring to the consultation and similarly, patients may possess medical knowledge that they would like to discuss with the clinician (Charles et al. 1997). During a shared decision, medical information and values may therefore be exchanged in either direction. Both parties then discuss their preferences and reach an informed, value-adjusted decision together (Charles et al. 1997, Charles et al. 1999).

Some have argued that SDM should ethically be done in any consultation (Schneider 1998, Stiggelbout et al. 2012); however, it is especially important in situations of medical equipoise, where no medically superior option exists and where harms and benefits of different options are balanced (Coulter 2010, Elwyn et al. 2000, Elwyn et al. 2009a). Such decisions are characterised by inherent uncertainty, as the clinician cannot make a firm recommendation as to what the best course of action for any individual patient may be without considering the patient's values with regard to the risks and benefits of the different options (Elwyn et al. 2009a). Therefore, these decisions are said to be 'preference-sensitive', i.e. depending on the goals, values and preferences of the individual patient and

requiring trade-offs between risks and benefits of different available options (Wennberg et al. 2002). Examples of such decisions include treatment options in early stage breast cancer (Kiebert et al. 1991), management of chronic kidney disease (Morton et al. 2010), participation in prenatal screening (Santalahti et al. 1998) and preventative options for breast and ovarian cancer in the context of increased genetic risk (Howard et al. 2009a).

Accepting that there is no medically recommended option can be difficult for patients, who are often more used to a paternalistic approach to medical decision making, i.e. being told what the best course of action is (Elwyn et al. 2000, Elwyn et al. 2009a). Uncertainty expressed by the clinician and being asked to play an active role in the decision making process may be perceived as a burden and cause a sense of worry, upset or even abandonment (Dresser 2012, Olthuis et al. 2013, Quill and Cassel 1995, Say et al. 2006). Patients may, for example, interpret the mention of uncertainty as a sign that the clinician lacks the necessary knowledge to make a recommendation. To counteract this, it has been noted that a clear distinction should be made between uncertainty due to a lack of clinical evidence for superiority of one option over another, and uncertainty due to a lack of knowledge on part of the clinician (Elwyn et al. 2000).

In some circumstances patients may also feel overwhelmed by the existence of choice (Dresser 2012, Olthuis et al. 2013). Therefore, considering patients' emotional states, vulnerability and willingness to participate in SDM is essential before asking them to play an active role (Olthuis et al. 2013). Unfortunately, during consultations, health professionals' assessments of their patients' desire for SDM are not necessarily accurate (Coulter 2007, Cox et al. 2007). Patients' preferences for involvement in the decision vary and may depend on the type of medical decision at hand, patient personality and socio-demographic variables, as well as the skill of the clinician (Bruera et al. 2001, Coulter 2003, 2007, Elwyn et al. 2000, Guadagnoli and Ward 1998). Hence, the extent of actual involvement may also differ, depending on the patient's preferences and the clinician's commitment to SDM (Deber et al. 1996, Deber et al. 2007, Thistlethwaite et al. 2006). Therefore,

preferences for involvement should ideally be discussed with patients prior to decision making (Coulter 2007, Cox et al. 2007). The process of SDM deliberations is outlined in detail in Chapter 2.

1.2.2 Patient decision support interventions

So-called patient decision support interventions or ‘decision aids’ are tools that have been developed to support and, to a degree, standardise the process of SDM by providing evidence-based medical information, teasing out preferences and stimulating discussion (O'Connor et al. 2004, Thistlethwaite et al. 2006). In recent years, the use of decision support interventions has become more widespread with the promotion of PCC and SDM in many healthcare systems, including the UK (Coulter 2009a, 2010, Coulter et al. 2011, Dagnone 2009, Department of Health 2011, Elwyn et al. 2010b, Patient Protection and Affordable Care Act 2010, Secretary of State for Health 2010). Patient decision support interventions are tools designed to facilitate SDM in situations of medical equipoise (Coulter 2009a, Elwyn et al. 2000, Elwyn et al. 2009a). They should be distinguished from behaviour change interventions, which aim to modify patients’ behaviour to improve health outcomes (Elwyn et al. 2009a). Decision support interventions, in contrast, are non-prescriptive and should include a balanced representation of the risks and benefits of all available options, including the option of doing nothing, where appropriate, to help patients clarify their preferences (Coulter 2003, Elwyn et al. 2009b, Joseph-Williams et al. 2013).

It has been shown that patient decision support interventions can significantly improve not only patients’ knowledge and accuracy of risk perceptions, but also alignment between a chosen option and patients’ preferences and, ultimately, decision satisfaction (Stacey et al. 2011). Therefore, this type of decision support goes beyond the mere provision of information regarding the health problem and associated options. These interventions offer a means to directly compare options, predict future feelings and evaluate possible outcomes to facilitate preference construction and SDM (Elwyn et al. 2009a). They engage

patients with their own healthcare and empower them to play an active part in decisions about their health, thereby addressing a number of healthcare quality improvement goals (Coulter and Ellins 2006).

Numerous interventions have been developed over the years. Since the first systematic review of patient decision support interventions in 1999 (O'Connor et al.), which included 17 trials of 11 different interventions, numbers have increased dramatically and in the latest Cochrane review, published in 2011 (Stacey et al.), 86 trials of more than 70 different interventions were analysed. These interventions come in a number of different formats, ranging from paper-based booklets to video interventions and sophisticated computer-based clinical guidance programmes (Anderson et al. 2011, Kaufman et al. 2003, Ottawa Hospital Research Institute 2013, Pell et al. 2002). Most of these extensive tools are designed for independent use by the patient, either before or after a consultation. However, embedding such tools into clinical practice has been difficult and many never proceeded to full implementation following initial development and research trials (Elwyn et al. 2008, Elwyn et al. 2013b, Frosch et al. 2011, Harter et al. 2011). Chapter 6 reviews previously developed patient decision support interventions that include the option of risk-reducing ovarian surgery with a view to assessing their format, content and availability.

In parallel to extensive decision support interventions, some groups started to develop much briefer tools, which are designed to be used in clinical encounters (Breslin et al. 2008, Elwyn et al. 2004, Whelan et al. 2004). Most recently, the use of a type of brief paper-based tool called 'Option Grid' was shown to support SDM in the clinical setting, stimulating discussions and resulting in a standardised way of presenting options to patients (Elwyn et al. 2013b, Marrin et al. 2013). Although Option Grids require further research regarding their effectiveness, these tools have the potential to be embedded in clinical practice more readily than extensive interventions and may solve some of the implementation problems observed with longer, more complex tools (Elwyn et al. 2013b, Elwyn et al. 2013c).

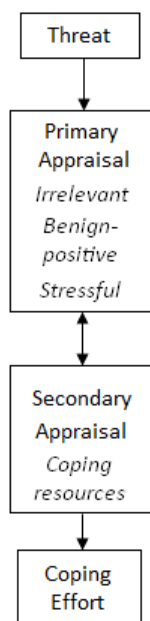
Although decision support interventions have been shown to have positive effects on patient knowledge and decision making (Stacey et al. 2011), to date there seems to have been a distinct lack of thorough use of theories in the development of patient decision support (Durand et al. 2008, Elwyn et al. 2011b). Interventions that state a theoretical basis usually refer to either 'Expected Utility Theory' (EUT) or the 'Ottawa Decision Support Framework' (Bernoulli 1954, Durand et al. 2008, O'Connor 2006). EUT is a normative theory of decision making. It describes how individuals should identify the best option based on mathematical principles (Bernoulli 1954). The Ottawa Decision Support Framework is a combination of several concepts and theories. It postulates that patients' decision needs affect decision quality, actions and impact and may be addressed through decision support (O'Connor 2006). A number of other theories have also been suggested as appropriate for use during development of decision support (Elwyn et al. 2011b). However, the emphasis has so far been on theories that describe decision making only, although additional types of theories may be applicable here, such as those that describe the process of coping. Chapter 2 provides a detailed review of decision making and relevant theories in preference-sensitive medical contexts.

1.3 Coping with health threats

1.3.1 Coping theory

In their theory of stress, appraisal and coping, Lazarus and Folkman (1984) define coping as "constantly changing cognitive and behavioural efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the resources of a person" (page 141). They describe the appraisal process in terms of two interacting phases, labelled 'primary' and 'secondary' appraisal, which result in a coping effort to manage the demands of the situation (Figure 1.2). During primary appraisal, an event's meaning is appraised as either irrelevant, benign-positive or stressful (Lazarus and Folkman 1984, Park and Folkman 1997).

Figure 1.2 - The appraisal and coping process in response to threats



Events that are appraised as stressful will result in a coping response. According to Lazarus and Folkman (1984) secondary appraisal “is a complex evaluative process that takes into account which coping options are available, the likelihood that a given coping option will accomplish what it is supposed to, and that one can apply a particular strategy or set of strategies effectively” (page 35). Once the event and possible coping options have been appraised, a coping response may be initiated. Chapter 2 provides a more detailed description of coping and associated theories.

Until recently, emotions and coping processes have largely been neglected by decision making theorists and have rarely been used to inform interventions that are designed to support deliberations (Balneaves and Long 1999, Power et al. 2011). In the context of medical decision making, coping plays an important role, as having to make a decision may be seen as taxing and/or exceeding an individual’s coping resources (Power et al. 2011). Being given a diagnosis, being told about different treatment options and being asked to get involved in SDM may thus result in a coping effort to understand the meaning and to manage the emotions and demands of the situation. Furthermore, emotions may be experienced in response

to making predictions about future outcomes of options (Gilbert and Ebert 2000, Wilson and Gilbert 2003, 2005). Therefore, emotions and coping should be viewed as an integral part of decision making (Balneaves and Long 1999, Power et al. 2011). Consequently, decision support interventions should ideally incorporate a coping component, to facilitate not only decision making processes, but also the process of coping with difficult medical decisions and associated emotions.

1.3.2 Coping interventions

To date, a wide range of interventions to facilitate coping processes in cancer, chronic disease and other contexts has been developed. Often, however, coping is not explicitly mentioned as an outcome or mediating variable in publications relating to these interventions, which can make their identification difficult (de Ridder and Schreurs 2001). Therefore psychosocial interventions, such as group therapy, problem solving training, stress management and cognitive behavioural therapy, that aim to address conceptual equivalents to coping, should be included when searching for interventions aimed to facilitate coping in these contexts (de Ridder and Schreurs 2001). Several reviews of such psychosocial interventions have found that these have positive effects on outcomes, including quality of life, self-efficacy and emotional well-being (Astin et al. 2002, Badr and Krebs 2012, de Ridder and Schreurs 2001, Scott-Sheldon et al. 2008). However, others have not confirmed these findings, especially in the context of cancer care (Coyne et al. 2006, Lepore and Coyne 2006, Meyer and Mark 1995). These discrepant results may be explained by the variety of psychosocial interventions that exist, the different healthcare contexts in which they are used and the range of outcome measures with which they are evaluated. While a detailed review of coping interventions is beyond the scope of this thesis, a brief overview of cancer-specific coping interventions is presented below.

Coping and/or psychosocial interventions in the context of cancer come in a variety of formats, however, most are face-to-face interventions either delivered in a one-on-one (Audrain et al. 1999, Schwartz et al. 1998) or group setting (Cousson-

Gélie et al. 2011, Edgar et al. 2001, Hamilton et al. 2011, Hosaka et al. 2000, Wellisch et al. 1999). There are a number of interventions concerned with facilitating or enhancing coping with a cancer diagnosis or cancer survival (Edgar et al. 2001, Hinds et al. 2000, Lee et al. 2006, McMillan et al. 2006, Radziewicz et al. 2009, Reavley et al. 2009, Weber et al. 2004), many specifically aimed at breast cancer patients (Cousson-Gélie et al. 2011, Hamilton et al. 2011, Hosaka et al. 2000, Lev et al. 2001, Maeda et al. 2008, Wengström et al. 2001). However, some have also been targeted at caregivers of cancer patients (Berila et al. 2009), while others were developed to support coping in women at increased risk of cancer (Esplen et al. 1998, Wellisch et al. 1999) or first degree relatives of women with breast cancer (Audrain et al. 1999, Schwartz et al. 1998).

As with extensive decision support interventions, implementation of psychosocial interventions that require a face-to-face format is not without its challenges, primarily due to the required staff time and training (Backer et al. 1986, Jacobsen 2009, Sin and Scully 2008). Some researchers have therefore developed cancer-specific coping interventions that are delivered in a paper- or web-based format and do not require specialist nurses or educators for delivery (Appleton et al. 2004a, Beatty et al. 2011, Phelps et al. 2006). These interventions were shown to reduce stress, enhance social functioning, improve quality of life and support coping (Appleton et al. 2004b, Beatty et al. 2011, Beatty et al. 2010a, Beatty et al. 2010b, Bennett et al. 2007, Phelps et al. 2013, Vilela et al. 2006). Paper-/web-based psychosocial interventions may be easier to implement compared to nurse/educator led programmes, as they do not require the same levels of staff time or training. To facilitate coping as well as decision making, elements of such interventions could potentially be embedded within paper- or web-based decision support tools. Alternatively, decision support tools could signpost users to relevant coping interventions. Chapter 7 describes the development of a decision support tool that includes elements of coping advice and signposting to external coping interventions with a view to support coping efforts and emotional well-being.

1.4 Ovarian Cancer Risk

The theoretical framework developed as part of this thesis will be applied to a specific healthcare context in order to determine whether it is a feasible model to guide the development of decision support interventions. Application of the framework will focus on decisions about risk management in the context of increased genetic risk of ovarian cancer, specifically decisions regarding risk-reducing bilateral salpingo-oophorectomy (RRSO). The justification for selecting this preference-sensitive decision is twofold. Firstly, ovarian cancer poses a considerable burden due to the lack of effective screening, late diagnosis and poor outcomes (Cancer Research UK 2011). This especially affects those at the highest risk for this disease, of whom up to two in five may develop ovarian cancer in their lifetime (Antoniou et al. 2003, Chen and Parmigiani 2007). Secondly, previous research has shown the inherent complexity of decisions regarding RRSO, including the role of emotions, past experiences, personal preferences and the variety of coping responses that may be employed (Fang et al. 2005, Howard et al. 2011, Howard et al. 2009a, Howard et al. 2010c, Miller et al. 1999). Hence, it has been proposed that decision support interventions may be of benefit in this context (Fang et al. 2005, Howard et al. 2009a). These factors make this decision an ideal candidate for depiction in a framework that describes appraisal and coping during deliberations and for the development of a tailored, theory-based decision support intervention that integrates deliberation and coping processes.

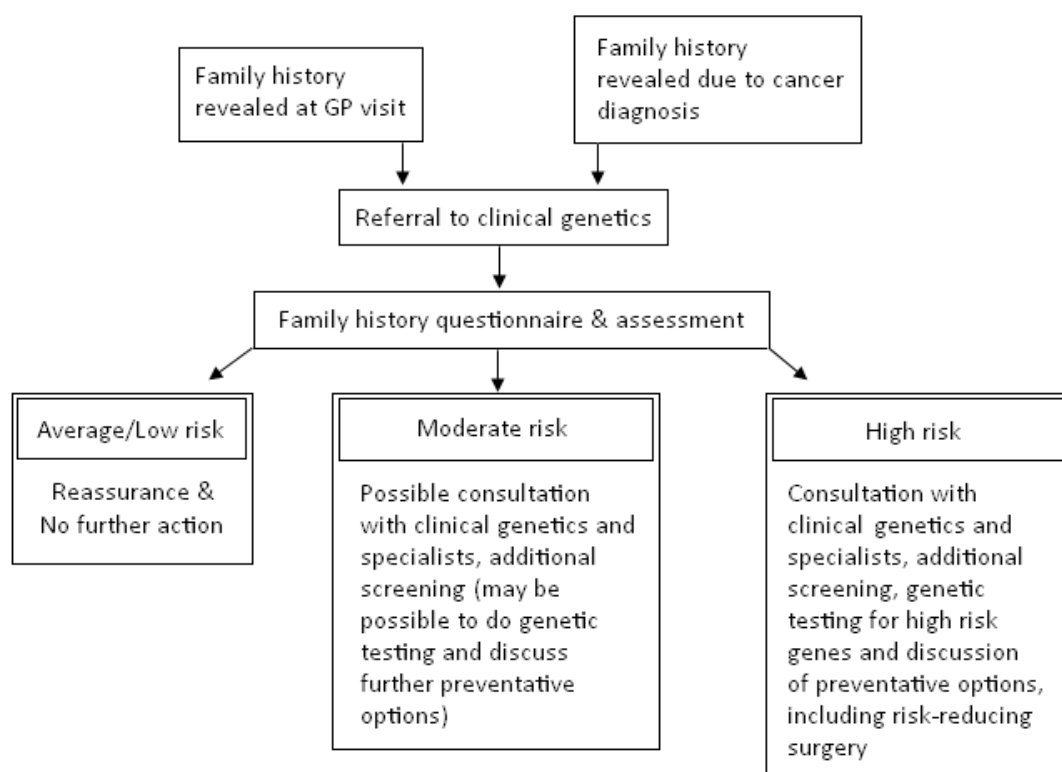
1.4.1 *Ovarian cancer risk and family history*

In the UK, ovarian cancer is the fifth most common cancer and second most common gynaecological cancer diagnosed in women (Cancer Research UK 2011). In England, approximately 6,000 new cases of ovarian cancer are diagnosed and 3500 ovarian cancer related deaths are reported annually (Trent Cancer Registry 2012). Due to the current lack of effective screening and vagueness of symptoms, these cancers are often detected at later stages (Stage III and above) and prognosis is relatively poor (Cancer Research UK 2011, Engel et al. 2002, Stearns et al. 2007). Five year survival rates for ovarian cancer are below 50% and for stage III and stage

IV disease survival rates are around 20% and 5%, respectively (Cancer Research UK 2011, Engel et al. 2002, Stearns et al. 2007). Lifetime risk of ovarian cancer in the general population is estimated to be around 2% (Cancer Research UK 2011). With most cases occurring sporadically in those over the age of 70 years, ovarian cancer in the general population is primarily a disease of older age; however, around 10% of ovarian cancers may be attributed to a family history and/or genetic mutation (Cancer Research UK 2011, Risch et al. 2001, Trent Cancer Registry 2012).

If a woman is found to have a family history of ovarian, breast or other cancers, either during a routine visit with her General Practitioner (GP) or as a result of an exploration of her family history after a cancer diagnosis, she may be referred to a clinical genetics service (Figure 1.3; Appendix 1.1). There, her family history will

Figure 1.3 - The referral and family history assessment pathway (see also: Appendix 1.1)



be assessed in detail and she will be given cancer risk estimates, for example for breast and ovarian cancer. Depending on her family history, she may be at 'average', 'moderate' or 'high' risk of ovarian cancer (Cancer Genetics Service for Wales 2006). The average, or sometimes termed 'low', risk group is estimated to have a risk that is the same or similar to that of the general population, whereas the moderate and high risk groups have an elevated risk. Moderate and high risk groups may be offered a consultation with the clinical genetics service and other specialists, as appropriate. Genetic testing may also be offered to some individuals, especially those thought to be at high risk, to determine whether a genetic mutation runs in the family (Cancer Genetics Service for Wales 2006). Risk management options, including additional screening and risk-reducing surgeries, such as mastectomy for breast cancer risk and salpingo-oophorectomy for ovarian cancer risk, may also be discussed.

Women who have relatives that have been diagnosed with ovarian, breast or a number of other cancers, may be at increased risk for ovarian cancer (Hanna and Adams 2006, Negri et al. 2003, Stratton et al. 1998). The number of and relation to affected relatives and the age of the woman herself, as well as the age of relatives at time of diagnosis, all play a role in determining whether an individual woman is at risk and in estimating her risk level (Cancer Genetics Service for Wales 2006, Stratton et al. 1998). Stratton et al. (1998) performed a meta-analysis to determine the effects of family history on risk of ovarian cancer. Overall, they found an approximate lifetime risk of ovarian cancer of 4% for women who had one first degree relative with ovarian cancer. More specifically, those whose mother was diagnosed with ovarian cancer had a lifetime risk of around 7.5%, whereas those with an affected sister had a lifetime risk of around 5% (Stratton et al. 1998). For women with more than one relative with ovarian cancer the lifetime risk was approximately 14%. It should be noted that these lifetime risks were lower for women who were over 45 at the time of their relative's diagnosis (Stratton et al. 1998). Others' estimates have varied between 2-5% for those with one first degree

relative and 7-23% for those with two or more affected relatives, depending on study design (Evans et al. 2009b, Kerlikowske et al. 1992, Negri et al. 2003).

A family history of other cancers, including breast, lung, stomach, intestine and lymphoma has also been found to confer an increased risk of ovarian cancer with corresponding odds ratios of 2.3, 1.3, 1.5, 1.7 and 2.3, respectively (Negri et al. 2003). By far the highest risk of ovarian cancer, however, is conferred by genetic mutations associated with Hereditary Breast and Ovarian Cancer (HBOC) and Lynch Syndrome (formerly Hereditary Non-Polyposis Colorectal Cancer, HNPCC) (Gayther and Pharoah 2010, Schorge et al. 2010). These mutations may be identified through genetic testing following risk assessment at the clinical genetics service.

1.4.2 Breast Cancer genes 1 and 2

Hereditary breast and ovarian cancer (HBOC) is associated with germline mutations in the Breast Cancer genes (BRCA) 1 and 2, which confer high risks of breast as well as ovarian cancer (Antoniou et al. 2003, Chen and Parmigiani 2007, Tranin 2005). The frequency of BRCA1 and 2 mutations among the population is very low at around 0.128% and 0.172%, respectively (Antoniou et al. 2000). However, in individuals of Ashkenazi Jewish descent frequency has been shown to be much higher, with a combined BRCA1/2 frequency of above 2% (Roa et al. 1996). Women with mutations in BRCA1 or BRCA2 make up the vast majority of non-sporadic, familial ovarian cancers (Antoniou et al. 2000, Risch et al. 2001).

Individual studies have produced a range of different cancer risk estimates for carriers of mutations in BRCA1/2 genes and it has been shown that heterogeneity of risk exists between families and depending on the position of the mutation on the gene (Antoniou et al. 2003, Easton et al. 1995, Evans et al. 2008, King et al. 2003, Struewing et al. 1997). In research combining several such studies, carriers of a BRCA1 mutation have on average been estimated to have a cumulative risk of breast cancer of 54%-65% and risk of ovarian cancer of 39% by age 70 (Antoniou et al. 2003, Chen and Parmigiani 2007). Carriers of a BRCA2 mutation on average have slightly lower cancer risks, with an estimated cumulative risk of breast

cancer of 45% and risk of ovarian cancer of 11-16% by age 70 (Antoniou et al. 2003, Chen and Parmigiani 2007).

1.4.3 *Lynch syndrome*

Lynch Syndrome is associated with mutations in a number of DNA mismatch repair genes, such as MLH1, MSH2, MSH6, PMS1 and PMS2 (Liu et al. 1996, Lynch and De la Chapelle 2003). While primarily associated with colorectal cancer, these mutations additionally confer an increased risk for a variety of other cancers in both sexes (Hampel et al. 2005, Vasen et al. 1996). In women, Lynch Syndrome mutations have been associated with a high risk of endometrial cancer and an elevated risk of ovarian cancer. One of the major issues with Lynch Syndrome associated cancers is the comparatively early age of onset, with the average age at endometrial and ovarian cancer diagnosis under 50 years (Brown et al. 2001, Lu et al. 2005).

Estimates for the lifetime risk of endometrial and ovarian cancer have varied between studies and exact risks depend on the type of genetic mutation, as well as other factors, such as year of birth (Barrow et al. 2009, Vasen et al. 2001, Watson et al. 2008). Average risk of endometrial cancer for all genes has previously been estimated at 28.2% (Barrow et al. 2009), 43% (Aarnio et al. 1995), 54% (Hampel et al. 2005) and 62% (Aarnio et al. 1999). Average risk of ovarian cancer has previously been estimated at 6.1% (Barrow et al. 2009), 6.7% (Watson et al. 2008), 6.9% (Vasen et al. 2001), 7.1% (Vasen et al. 1996), 12% (Aarnio et al. 1999) and 13.5% (Hampel et al. 2005). As stated earlier, the risk differs depending on the affected gene, which might explain some of the variation in pooled risk estimates, as the proportion of carriers of different mutations might vary between studies. Vasen et al. (2001), for example, found the risk of ovarian cancer by age 70 in carriers of MLH1 mutations to be 3.4%, compared to 10.4% in carriers of MSH2 mutations. Similarly, Barrow et al. (2009) estimated cumulative risk in individuals with MLH1 mutations to be 5.5%, compared to 7.5% in those with MSH2 mutations. Further

discrepancies between studies may be explained by additional factors, such as study design and birth cohort (Watson et al. 2008).

1.4.4 Ovarian cancer risk management options

Women from HBOC and Lynch Syndrome families, as well as those with no confirmed genetic mutation but a significant family history, live with an increased risk of ovarian cancer (Table 1.1).

Table 1.1 - Estimates of absolute lifetime risk of ovarian cancer

| Family history or genetic mutation | Lifetime risk of ovarian cancer |
|--|--|
| General population | ≤2% |
| One first degree relative with ovarian cancer | 2-7.5% ¹ |
| Two or more relatives with ovarian cancer | ≥7% ² |
| Confirmed BRCA1 mutation | 39% |
| Confirmed BRCA2 mutation | 11-16% |
| Lynch Syndrome family history and/or associated mutation | 6-12% |

¹risk depends on exact relationship with patient (i.e. mother, sister, daughter) and age at diagnosis

²risk depends on number of relatives, exact relationship and age at diagnosis

As the American Society for Clinical Oncology noted in its statement on genetic testing for cancer susceptibility (1996), “the [...] ability to identify individuals at highest risk of cancer holds the promise of improved prevention and early detection of cancers. It also poses potential medical, psychological and other personal risks that must be addressed [...].” (page 1730). While these risks include those which need to be addressed prior to genetic testing as part of an informed consent process, they also include issues following a positive gene test result. The American Society for Clinical Oncology therefore emphasises the need for follow-up care and discussion of effective risk management options post-testing.

Since 1996, the statement has been updated twice (2003, 2010); however, the basic principle of offering advice and guidance, as well as management options, following testing for genetic mutations remains an important aspect. For risk of breast cancer for example, management options include enhanced surveillance,

chemoprevention with Tamoxifen or risk-reducing mastectomy (Brekelmans et al. 2001, Cuzick et al. 2007, Cuzick et al. 2003, Hartmann et al. 1999, Rebbeck et al. 2004). In the case of increased risk of ovarian cancer, the management options offered to women at high risk are somewhat more limited, due to the lack of effective screening (Appendix 1.2).

1.4.4i Ovarian cancer screening: While several researchers have explored the effectiveness of screening for ovarian cancer using annual trans-vaginal ultrasound scans and blood tests for biomarker Cancer Antigen (CA)125, successful early detection (at Stage I-II) and a gain in survival for screen detected cancers has not been fully established and routine screening has not been implemented (Bell et al. 1998, Jacobs and Menon 2004, Kauff et al. 2005). In the UK, the Familial Ovarian Cancer Screening (UK FOCS) study Phase I used annual trans-vaginal scans and CA125 blood tests to detect ovarian cancer in women at $\geq 10\%$ risk, but did not find a sufficiently high proportion of early stage cancers through screening to recommend introduction of a routine screening programme for high risk women (Long and Kauff 2012, Rosenthal et al. 2013a). Phase II of the UK FOCS study increased CA125 testing frequency to 4-monthly intervals and made a range of other modifications (Institute for Women's Health 2010, Rosenthal et al. 2013a). This study has only recently (summer 2011) concluded. While women gained considerable reassurance from this screening program (Brain et al. 2012, Lifford et al. 2013), results establishing the clinical effectiveness of this type of screening are not yet available. Similarly, another trial of ovarian cancer screening in the general population, the UK Collaborative Trial of Ovarian Cancer Screening (UK CTOCS), which may provide evidence of effects of screening on mortality, has also only recently concluded (end of 2011) and has not reported results yet. Until the complete results of the UK FOCS study Phase II and UK CTOCS have been published, the recommendations from the UK National Screening Committee (2006) and the guideline published by the National Comprehensive Cancer Network (2012) do not recommend routine screening for ovarian cancer in high risk women.

1.4.4ii Risk-reducing surgery: In the absence of medically proven screening for ovarian cancer, the only active management option currently available and recommended is risk-reducing bilateral salpingo-oophorectomy (RRSO) (National Comprehensive Cancer Network 2012). RRSO involves surgical removal of both ovaries and fallopian tubes to prevent ovarian cancer. This operation substantially reduces ovarian cancer risk by up to 80% in the highest risk groups (Rebbeck et al. 2009). Additional benefits of RRSO include a substantial reduction in breast cancer risk of up to 50% if surgery is performed before the menopause (Kramer et al. 2005, Rebbeck et al. 2009). This is especially relevant in carriers of mutations in the BRCA genes, who are at high risk of breast cancer and who may choose to undergo RRSO to reduce their breast cancer risk (Metcalf 2009). Domcheck et al. (2006, 2010) showed that RRSO reduces ovarian and breast cancer specific mortality, as well as all-cause mortality, in carriers of BRCA mutations. RRSO is most effective if done at or before age 40 in carriers of BRCA1 and BRCA2, but it still confers a survival benefit if done at a later age (Kurian et al. 2010, Latosinsky et al. 2011).

However, RRSO is not without its drawbacks. The surgery itself can usually be performed as a keyhole procedure, but does have a complication rate of 4% that should be considered when discussing surgery (Manchanda et al. 2011). Additionally, surgical removal of the ovaries before age of natural menopause results in infertility and oestrogen deprivation. These effects may have far reaching medical as well as psychosocial consequences for patients. In the short-term, the sudden lack of oestrogen results in a surgically induced menopause, which may cause severe menopausal symptoms, including headaches, hot flushes, night sweats, mood swings, sleep disturbances, weight gain, depression, vaginal dryness and reduced libido (Finch et al. 2011, Finch and Narod 2011, Hallowell et al. 2012). These in turn may affect social functioning and relationships, including sexual relationships with partners, as well as body image and perception of gender identity (Finch et al. 2011, Hallowell et al. 2012).

In the longer term, it has been suggested that oestrogen deprivation may also affect bone density, cardiovascular health and cognitive functioning (Aitken et

al. 1973, Gordon et al. 1978, Parker 2010, Parker et al. 2009, Rocca et al. 2009). The lack of oestrogen following surgery increases bone density loss, thereby enhancing risk of osteoporosis and fractures (Challberg et al. 2011). Similarly, some studies have shown an increased risk of cardiovascular events in women who underwent RRSO, especially in those under the age of 45 at time of surgery (Atsma et al. 2006, Falkeborn et al. 2000, Rivera et al. 2009). However, data on the exact level of bone density and cardiovascular disease is controversial and research is still ongoing (Andrews et al. 2012, Jacoby et al. 2009). Effects of RRSO on cognitive functioning, including risk of Parkinson's and dementia, have also been noted and found to be more pronounced in women who are pre-menopausal at time of surgery (Rocca et al. 2007, Rocca et al. 2008a, Rocca et al. 2008b). Exact risk figures for these long-term health outcomes for any individual woman may depend on her age at the time of RRSO, her lifestyle and her family history of osteoporosis, dementia and cardiovascular disease.

Despite these drawbacks, RRSO in women at high risk of ovarian cancer is warranted and does confer an overall survival benefit (Domchek et al. 2006, Domchek et al. 2010, Kerlikowske et al. 1992). To mitigate the short- and long-term effects of oestrogen deprivation at a young age, women who undergo RRSO before the age of 45 are recommended to take hormone replacement therapy (HRT) until the age of natural menopause (Challberg et al. 2011, Finch et al. 2011, National Institute for Health and Care Excellence 2013). While HRT has previously been linked to an increased risk of breast cancer, it has been shown to be safe in women at high risk if taken until the age of natural menopause to supplement the sudden lack of oestrogen following surgery (Beral and Collaborators 2003, Eisen et al. 2008, Rebbeck et al. 2005).

It should be noted that, while the risk of ovarian cancer is dramatically reduced by RRSO, it is not completely eliminated. Studies have found varying residual risks of primary peritoneal cancer for women with a family history and/or BRCA mutation ranging from 0% (Evans et al. 2009a), 0.8% (Rebbeck et al. 2002), 1.3% (Domchek et al. 2006), 1.9% (Piver et al. 1993), 2% (Rabban et al. 2009) to

4.3% (Finch et al. 2006). The differences in risk estimates between studies may be explained by differing inclusion criteria, varying proportions of women with BRCA1 versus BRCA2 mutations, differing protocols for post-surgical cytology assessment (at which ovarian cancers may be identified) and varying length of follow-up (Manchanda et al. 2012b, Powell 2006). Generally, women with mutations in BRCA1 were found to be at higher risk for post-surgical primary peritoneal cancer than women with mutations in BRCA2 (Finch et al. 2006, Rabban et al. 2009). The remaining risk of primary peritoneal cancer therefore needs to be discussed with women prior to surgery.

In the case of Lynch Syndrome, the significantly elevated risk of endometrial cancer warrants a risk-reducing hysterectomy (RRH; surgical removal of the uterus), which should be discussed during consultations about risk management (Chu and Coukos 2006, Lindor et al. 2006). RRSO is usually considered as a concomitant procedure with RRH for women with Lynch Syndrome. Studies have found that RRH with concomitant RRSO is highly effective in reducing risk of endometrial and ovarian cancer (Schmeler et al. 2006a). However, a small risk of primary peritoneal cancer also remains in this population (Schmeler et al. 2010).

1.4.4iii Symptom awareness: As noted earlier, effectiveness of screening for ovarian cancer has not yet been proven and therefore RRSO is the only active management option available to women at increased risk (Schorge et al. 2010). Women who decide to delay or reject surgery are recommended to be aware of the symptoms of ovarian cancer and seek immediate professional help if any persistent symptoms develop, to improve the chances of early detection (Schorge et al. 2010). The symptoms of ovarian cancer include persistent abdominal distension, early satiety and loss of appetite, pelvic or abdominal pain and increased urinary frequency (National Institute for Health and Care Excellence 2011).

1.4.4iv Chemoprevention and other risk-reducing measures: Pre-menopausal women who do not want to undergo surgery may also explore the possibility of chemoprevention using the oral contraceptive pill (OCP). This has been shown to

reduce the risk of ovarian cancer (Cibula et al. 2011, Iodice et al. 2010). The NICE clinical guidance (2013) notes that the OCP should be discussed with women from HBOC families, however, due to its potentially increasing effects on risk of breast cancer, does not recommend the OCP purely for cancer prevention (Brohet et al. 2007). Other chemoprevention options for ovarian cancer are not available. It has been shown that parity exerts a protective effect from ovarian cancer that increases with the number of pregnancies experienced, likely due to suppression of ovulation, a similar protective mechanism as that achieved through OCP use (Cramer et al. 1983). A review of women's decision making regarding management of increased risk of ovarian cancer is presented in Chapters 3 and 4.

1.5 Thesis overview

Two research gaps are addressed in this thesis. Firstly, to date, decision making and coping have rarely been considered as simultaneous, interdependent processes. SDM in healthcare has been endorsed and promoted by various governments, but the role of emotions and coping during decision making and their possible influence on the process of SDM have been neglected. Patients who are worried or anxious and patients who do not use effective coping strategies may not feel equipped to deal with the added pressure of participating in SDM. Therefore, to advance the field of SDM, there is a need to consider how coping and decision making processes overlap. A clear understanding of how emotions and coping may influence appraisals and deliberations could shine a light on how they may facilitate, instead of hinder, SDM. Therefore, this thesis sets out to develop a novel framework that integrates the concepts of deliberation and coping in healthcare contexts.

Secondly, developers of patient decision support interventions, which aim to facilitate SDM, have also failed to use coping theory to inform the design and content of their interventions. In fact, many do not draw on theory at all, despite repeated calls for theoretical underpinning of such tools (Charles et al. 2005, Durand et al. 2008, Elwyn et al. 2011b). Interventions guided by coping, as well as

decision making, theory may be able to provide more holistic decision support than those developed to date. However, as no attempts have been made to use coping theory to guide development of patient decision support interventions, it is unclear whether use of such theories is feasible in this context. For this reason, and using decisions about RRSO in women at increased risk of ovarian cancer as an example, this thesis aims to outline and test how a framework that integrates coping and decision making theories may be operationalised to assess the content of existing interventions and to guide the development of new interventions.

1.5.1 Objectives and methods

The thesis is based on reviews of the literature and qualitative research methods, including focus groups and interviews. The objectives of this thesis are:

1. To undertake a narrative review of the theoretical literature on decision making and coping applicable to healthcare contexts.
2. To develop a novel framework that integrates decision making and coping theory to describe preference-sensitive medical decisions from a patient perspective.
3. To describe in detail the decision making and coping processes regarding risk management in the context of increased genetic risk of ovarian cancer using data from a systematic search of the literature, as well as focus groups with affected patients and interviews with health professionals.
4. To adapt the novel framework to specifically describe decision making regarding risk management in the context of increased genetic risk of ovarian cancer.
5. To operationalise the adapted framework by using it:
 - a. to assess how previously developed decision support interventions, identified through a systematic literature search, facilitate appraisal and coping processes regarding ovarian cancer risk management.

- b. to develop a novel decision support intervention for women at increased genetic risk of ovarian cancer in the UK, following the MRC guidelines for the development of complex interventions (Craig et al. 2008).
6. To assess usability of this novel decision support intervention using cognitive interviews.

1.5.2 Thesis structure

This thesis contains eight further chapters. With exception of Chapters 2 and 9, each chapter contains four main sections on background, methods, results and discussion.

Chapter 2 - Decision making and coping in healthcare: The Coping in Deliberation (CODE) framework

This chapter reviews the existing theoretical literature on decision making and coping with a specific focus on those theories applicable to preference-sensitive medical contexts. It then goes on to propose a novel framework called the 'Coping in Deliberation' (CODE) framework, which integrates decision making and coping theories, to describe preference-sensitive medical decisions from a patient perspective as a multi-step coping process.

Chapter 3 - Risk-reducing bilateral salpingo oophorectomy: A dilemma for patients

Chapter three presents a review of the literature, published between 1996 and July 2012, regarding women's decision making about RRSO. It delineates the medical, psychological, social, demographic and other considerations important and influential in women's decision making and describes women's expectations of decision support.

Chapter 4 - Patients' and health professionals' perspectives on decisions about risk-reducing bilateral salpingo-oophorectomy

This chapter reports on the findings of a focus group and interview study conducted between June 2011 and April 2012 with women at increased risk of ovarian cancer and health professionals involved in their care. It outlines considerations important during decision making about RRSO in the UK context and proposes a decision making pathway to describe such decisions.

Chapter 5 - Adapting the CODE framework: Decisions about risk-reducing bilateral salpingo-oophorectomy

Chapter five describes an adaptation of the generic CODE framework to decisions about RRSO in the context of increased genetic risk of ovarian cancer. Using the considerations identified in Chapters 3 and 4, the contents of the framework are adapted and then validated by mapping them onto a retrospective, in-depth report of one woman who went through this decision.

Chapter 6 - Applying the CODE framework: A review of decision support interventions that include the option of risk-reducing oophorectomy

This chapter applies a checklist, as well as the adapted CODE framework, as an assessment tool to evaluate previously developed decision support interventions' coverage of considerations important during appraisal of cancer risk and RRSO and of coping advice.

Chapter 7 - Developing a decision support intervention for women at increased risk of ovarian cancer

Chapter seven presents the development of a decision support intervention for women at increased risk of ovarian cancer who are considering RRSO. Based on the adapted CODE framework and informed by feedback from health professionals and women at risk of ovarian cancer, the intervention consists of an Option Grid and an "Oophorectomy Decision Explorer" (OvDex) website/booklet. It is designed to facilitate deliberation and coping processes during decision making about RRSO.

Chapter 8 - Usability testing of a decision support intervention for patients considering risk-reducing bilateral salpingo-oophorectomy

Chapter eight reports on the findings of a usability testing study using cognitive interviews conducted in March 2013. It outlines reviewers' feedback on usability, accessibility, layout and understanding of the contents of the Option Grid and OvDex and any changes made in response.

Chapter 9 – General Discussion

This concluding chapter discusses the findings of the thesis within the wider context of patient centred care and shared decision making. Key findings are summarised and methodological limitations are discussed. Possible future applications of the generic CODE framework are explored and suggestions for further evaluation, and ultimately implementation, of the decision support intervention in clinical practice are provided.

In summary, the thesis outlines the development of a novel theoretical framework that acknowledges the importance of emotions and coping during deliberations about healthcare decisions, followed by operationalisation of this framework through adaptation and subsequent application in the assessment and design of decision support interventions for decisions about RRSO in the context of increased genetic risk of ovarian cancer.

2 Decision making and coping in healthcare: The Coping in Deliberation (CODE) framework

2.1 Introduction

In the face of medical uncertainty, when there is no single recommended option, healthcare decisions are dependent on personal circumstances, values and preferences of the patient (Elwyn et al. 2009a). These preference-sensitive decisions may be complex, unfamiliar and difficult for patients, as they are required to process a wealth of new medical information and to play an active role in decisions about their own health. Deliberation and coping processes in these situations are not well understood and rarely described in detail (Noone 2002). A thorough description of the deliberation and coping processes undergone by patients throughout the deliberation process could be helpful to understand the different issues and concerns involved in preference-sensitive medical decisions and to improve the support available to patients in those situations.

There are a number of theories addressing decision making or coping processes which have been applied in healthcare settings (e.g. Bravata et al. 2002, Goldstein and Gigerenzer 2009, Henderson et al. 2006, Janis and Mann 1977, Kahneman and Tversky 1979, Lazarus and Folkman 1984, Leventhal et al. 1980, Reyna 2008, Svenson 1992). However, traditional decision making theories do not consider the relationships between decision making and coping processes, despite the interdependency of these processes in real life. More recently, researchers have begun to integrate coping and decision making theories and thereby describe more fully the processes individuals go through when making preference-sensitive medical decisions, such as treatment decisions in breast cancer (Balneaves and Long 1999, Power et al. 2011). However, there is scope for integration and further extension of these theories to produce a more detailed framework of decision making and coping in order to describe more accurately the multidimensional

interactions between these processes and to specifically highlight the questions and issues that patients face during preference-sensitive decision making.

Building on previous integrative approaches, this chapter¹ aims to (i) describe current theoretical approaches to decision making and coping in healthcare, and (ii) develop a multidimensional, interactive framework of patients' appraisal and coping responses in preference-sensitive medical decisions. The process of decision making in healthcare settings is explored, followed by a description of appraisal and coping responses to health threats. These processes are then integrated in one overarching framework that aims to describe coping in deliberations about preference-sensitive medical decisions and thereby provide a theoretical basis for developing decision support materials.

2.2 Methodology

2.2.1 Literature search strategy

The literature review of decision making and coping theories was conducted using a narrative search strategy (Baumeister and Leary 1997, Rother 2007). The narrative search strategy lends itself to integrative theorizing, thereby feeding into the process of developing a novel framework from theories identified by the review, which was the ultimate aim of this study (Baumeister and Leary 1997).

Reviews of patient decision support interventions and their use of theory were initially used to identify relevant theories that had previously been used in the development of decision support and similar interventions the healthcare contexts (Bekker et al. 1999, Durand et al. 2008, Elwyn et al. 2011b). Subsequently, a snowballing approach was used to identify further theories relevant to decision

¹ Parts of this chapter have been published as Witt J, Elwyn G, Wood F and Brain K (2012) *Decision making and coping in healthcare: The Coping in Deliberation (CODE) framework*. Patient Education and Counseling Vol. 88(2): 256-261 [Appendix 2.1]

making and coping processes in a medical context. Additional hand searches were carried out in issues of *Patient Education and Counselling* and *Medical Decision Making* published in 2008-2011, to identify novel theories not yet used in intervention development or included in earlier reviews.

Rather than being comprehensive, this approach was selected to include those theories most relevant to coping and deliberations in healthcare settings, ultimately leading to a novel framework that could form the basis for the development of decision support materials. Theories of behaviour change were excluded from the analysis, as the aim was to explore decision making in situations of medical equipoise, when the choice is preference-sensitive and depends on patient values (Elwyn et al. 2009a).

2.2.2 Framework development

Theories were summarised and emerging decision making and coping processes were integrated into a new framework. Face validity of the framework was evaluated in group meetings with members of the decision laboratory and health psychology research teams at Cardiff University.

2.3 Deliberation in preference-sensitive medical contexts

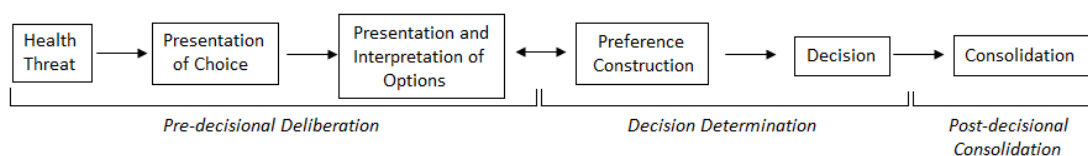
2.3.1 Overview

Many decisions regarding the best course of action in healthcare, such as what medication to prescribe for a specific disease, are mainly influenced by established medical efficacy. Although these decisions should be discussed with the patient, the health professional will usually be able to make a firm recommendation as to what course of medication or action would result in the best outcome. From a patient perspective, such decisions may not require an extensive deliberative effort.

Extensive deliberation processes will only ensue when there is more than one choice and an individual experiences conflict as to which choice might result in the most favourable outcome (Janis and Mann 1977). For a patient, this may be the case in situations of equipoise when no clear medical guidance is available, when

two or more types of treatment may be equally effective and when decisions are preference-sensitive (Elwyn et al. 2009a). The deliberation process in such situations may be described in several stages, including pre-decisional deliberation, decision determination and consolidation (Elwyn et al. 2012, Elwyn and Miron-Shatz 2009, Svenson 1992) (Figure 2.1).

Figure 2.1 - The deliberation process in preference-sensitive healthcare decisions



2.3.2 Presentation of the health threat and introduction of choice

Before the main deliberation process is initiated, the health threat is presented to the patient in the form of a diagnosis, test result or risk assessment. This is then followed by the presentation of choice, which introduces the patient to the idea that there is more than one option and that options have risks and benefits, which require the patient to consider their own values and preferences and play an active role in decision making. This is often a new and unfamiliar concept for patients in a medical context and may result in surprise, upset or even feelings of abandonment (Elwyn et al. 2000, Quill and Cassel 1995, Say et al. 2006).

Patients' reactions to a health threat (and the idea of choice) may depend on a large number of contextual influences, such as personal interpretations of the health threat, values and goals, past experiences and social support (Leventhal et al. 1980, Shaw 1999). Interpretations of information, reactions to the idea of choice and to being asked to play an active role in the decision making process also depend on the current emotional and health state of the patient (Kahneman and Tversky 1979).

For instance, a diagnosis of the common cold may be appraised as a relatively minor threat, which according to past experience will be fully cured in a few days and have little or no consequence to personal goals, therefore instilling little or no anxiety. Thus the deliberation process, if one is initiated, is relatively effortless. The patient may for example choose to take painkillers and decongestant treatment or may decide not to medicate, but whatever decision they take, they are likely to make a full recovery. In contrast, a diagnosis of early stage breast cancer will usually be appraised as a major threat, which will have far reaching consequences for one's own future and that of others in the family, thus instilling high levels of anxiety (Burgess et al. 2005). The patient may choose between two types of surgery, lumpectomy and mastectomy, both of which have similar efficacy and therefore depend on personal preferences. This is likely to be a complex and difficult deliberation process and such decisions ideally require careful consideration of the patient's values, goals and preferences, therefore warranting a discussion of choice.

Individual differences in self-efficacy or perceived control may influence a patient's perceived ability to make a decision themselves and may affect coping responses (Bandura 1977, Davison and Degner 1997, Skinner and Zimmer-Gembeck 2011). The concept of self-efficacy describes beliefs in one's ability to perform a certain behaviour or achieve a certain goal, such as making a preference-sensitive decision. Individuals with high self-efficacy may wish to be more active throughout deliberation, whereas patients with low self-efficacy may prefer a more passive role (Oron et al. 2009). Similarly, individuals with high health literacy may prefer to be involved in decision making whereas those with low literacy may not (DeWalt et al. 2007). Either preference is acceptable, and it may be beneficial to discuss preferred level of involvement at the point of introduction of choice. Early stage breast cancer patients, for example, have reported various preferences for their level of involvement in the deliberation process, which should be catered for by health professionals involved in their care to achieve informed, value-adjusted decisions (Degner and Sloan 1992, Hack et al. 2006). It has previously been shown that a match between patient preference for involvement and perceived actual

involvement is associated with better outcomes post-decision (Keating et al. 2002). However, irrespective of desired role, patients should always be informed about their options and their values and goals should be considered before a decision is made, if that decision is preference-sensitive (Levenstein 1984, Say and Thomson 2003).

2.3.3 *Presentation of options and preference construction*

An essential part of the deliberation process is the provision of information about options and the mapping of those options onto the patient's values and goals, to enable them to construct a preference. Hence, following presentation of the health threat and the idea of choice, the main deliberation process is initiated. Options are described to, and interpreted by, patients, while preferences are constructed. When faced with decisions in healthcare, patients are often given a substantial amount of information that is new, relatively complex and threatening. Both the quantity and content of this information may cause distress. It is likely that patients struggle to remember all the information they are given during a consultation (Reyna 2008). Patient preferences may therefore be based on the parts of the information that were understood and remembered, rather than exact numerical values and associations (Reyna 2008). Affect heuristics and intuition have also been considered important in preference construction, especially in situations where patients do not have much time to consider their options, for instance when parents are asked to decide about amniocentesis (Durand et al. 2011, Goldstein and Gigerenzer 2009).

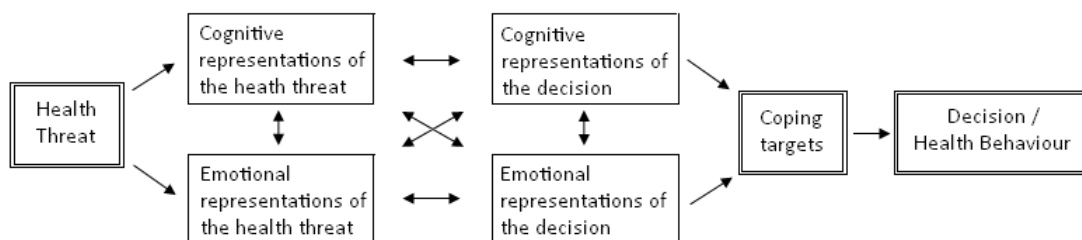
Emotions such as distress and anxiety can have detrimental effects on deliberations as they may impede understanding of medical information and careful consideration of values. As a result, they may lead to poor decisions (Janis and Mann 1977). Loewenstein et al. (2001) distinguished two broad types of emotions in decision making: *anticipatory* and *anticipated* emotions. Each phase of the decision making process in healthcare is influenced by both types of emotions. Anticipatory emotions arise in response to the immediate issue of being faced with a health threat and with the need to make a decision. These can include emotions

such as worry, fear or anxiety. Anticipated emotions are predicted emotions, which the patient expects they will experience when one or more of the possible outcomes associated with the decision occur. These may include relief, regret or anger. Both types of emotions will form during the deliberation process, in response to the health threat and the presentation of available options. For instance, a patient will emotionally react to being told about a health threat (anticipatory emotions) and may simultaneously imagine what this threat might mean for their future and how they would feel in this predicted future (anticipated emotions). Similarly, each possible option and associated outcomes will give rise to anticipated emotions during the presentation of options. However, individuals may struggle to accurately predict anticipated emotions, over- or underestimating their duration and intensity (Wilson and Gilbert 2003, 2005). Therefore, anticipated emotions can lead to biases in the decision making process, as patients may base their decision partly on inaccurate predictions of their future emotions (Kermer et al. 2006).

2.3.4 *A model of cognitive-emotional decision making*

Power and colleagues (2011) developed a framework of medical decision making which considers the importance of emotions as a motivational factor in decisions (Figure 2.2). “Cognitive-emotional decision making” distinguishes *cognitive* processes from *emotional* processes formed in response to the health threat and the decision. This distinction allows consideration of each cognitive and emotional aspect of decision making in turn, and of the motivational influence emotions have on cognitive processes. For example, emotions often influence risk estimates, as patients struggle to understand the concept of risk (Loewenstein et al. 2001). Therefore they use their emotional responses to the threat (e.g. ovarian cancer) to interpret their risk (Zikmund-Fisher et al. 2010). A numerical estimate such as ‘7 in 100 women like you will get ovarian cancer’ may not mean much to a patient. However, if that same patient has seen a relative die of the disease, then their interpretation of that risk will be formed whilst they re-live emotions of fear and loss associated with their past experience of ovarian cancer (anticipatory

Figure 2.2 - Model of cognitive-emotional decision making (adapted from Power et al. 2011)



emotions) and imagine what a diagnosis might mean for them in the future (anticipated emotions) (Dagan and Goldblatt 2009, Howard et al. 2011). Therefore, their interpretation of that risk may be high, which can in turn influence decisions relating to that risk (Howard et al. 2011). Power's (2011) model of cognitive-emotional decision making clearly distinguishes emotional from cognitive processes, emphasising the importance of emotions in the decision making process and their influence on cognitive representations of both the health threat and the decision.

Therefore, regulating both anticipatory and anticipated emotions is an important motivational target in decision making (Power et al. 2011). Strong negative anticipatory emotions may lead to avoidant behaviour, which allows patients to temporarily regulate these emotions (Anderson 2003). For example, a patient may display avoidant behaviour in response to being asked to make a decision if they feel overwhelmed by, and anxious about, the prospect of having to make a decision about their health. However, unless the situation is out of the patient's control, non-avoidant behaviours lead to better long-term outcomes (Krohne 1993, Rippetoe and Rogers 1987, Stanton and Snider 1993).

2.3.5 Post-decisional consolidation

Once preference construction has been completed to identify the preferred option, a decision can be made, either by the patient, by the health professional or in collaboration. Decisions may be final or may be revised at a later stage. Following the decision, consolidation of that decision may protect the decision maker from

regret in the future and is especially important in irreversible decisions, such as those about surgical procedures (Connolly and Reb 2005, Svenson 1992). Post-decisional consolidation may be described as a way of coping with the outcomes of a decision once that decision has been made. This should be distinguished from coping with the deliberation process itself, which is described in section 2.4. It should be noted that, if a decision is deferred or avoided (Anderson 2003), post-decisional consolidation does not occur until a decision is made.

2.4 Coping in deliberation during preference-sensitive medical decision making

2.4.1 Overview

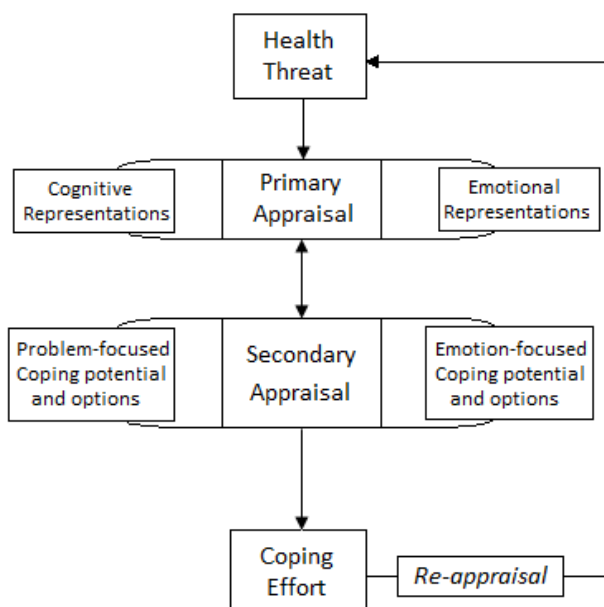
Coping describes efforts to actively manage events which are appraised as 'stressful' (Lazarus and Folkman 1984). Any event can result in a coping response, if it is appraised as having resulted in harm or loss, as having the potential to result in harm or loss (i.e. being a 'threat'), or as being a challenge (Lazarus and Folkman 1984). In other words, an event is appraised as stressful if it is incongruent with an individual's perceived global meaning, i.e. a person's goals, beliefs or sense of self (Park 2011, Pergament 1997). A single stressful event may lead to several coping responses and may also result in future events, which can stimulate appraisal and coping responses. For instance, the loss of a limb in an accident is an event which results in harm and loss and will require a coping response. Future events induced by this initial event, such as having to have several rounds of surgery to repair damage, can be appraised as stressful in their own right and therefore lead to further coping responses.

In a medical context, patients who are required to play an active role in decisions about their healthcare have to cope with a number of different potentially stressful events. As outlined in section 2.3.2, these individuals need to cope with the fact that they are either ill or at risk of disease, in which case the diagnosis or risk statement would be the 'threat' that requires a coping response. Additionally, they need to come to terms with the fact that there are several options, and that they are being asked to play an active role in decision making. Here, the burden of

having to decide may be perceived as a threat or challenge in its own right. Further still, as described in section 2.3.3, patients then have to consider the different options and all their potential outcomes in order to make that decision. These options, and risks linked to them, may be appraised as bearing the potential for loss or harm in the future and can therefore be perceived as stressful as well, requiring a coping response in their own right.

Researchers have explored appraisal and coping processes in healthcare, for example using Leventhal's Self-Regulatory Model (SRM) of illness representations (1980) and Lazarus and Folkman's transactional theory of stress, appraisal and coping (1984). Both these models suggest that illness representations are formed during appraisal of the diagnosis or health threat and ultimately determine the coping response. An integrated outline of the appraisal and coping process as described by these models is presented in Figure 2.3.

Figure 2.3 - Appraisal of and coping with health threats



2.4.2 Primary appraisal

According to Lazarus and Folkman (1984), events are appraised in two ways. During *primary* appraisal an event may be appraised as irrelevant, benign-positive or stressful. The two former appraisal types (irrelevant and benign-positive) usually do not warrant or require further appraisal or a coping response, while the latter appraisal type, which categorises an event as 'stressful', does. Lazarus and Folkman (1984) distinguish three types of 'stress appraisals': harm/loss, threat and challenge. Aldwin et al. (1996) proposed three additional appraisal types: 'hassle/annoyance', 'uncertainty' and 'worry about others', which may also require a coping response and therefore be defined as 'stressful' appraisals (Folkman 2011).

In the medical context, the patient cognitively and emotionally appraises the diagnosis or risk estimate they have been given, its severity and its relevance to their life. A diagnosis may be appraised as harm/loss, if it is already debilitating, and/or as a threat, if the patient expects future harms/losses due to the diagnosis. Being at risk for a certain disease may be appraised as a threat that carries potential for future harm/loss. If the disease has a genetic component, then it may also result in worry about others, including children, siblings and parents. As proposed by Leventhal et al. (1980), cognitive representations (or appraisals) of a health threat are based on the patient's understanding and interpretation of five dimensions of the threat: identity, cause, time line, consequences and possibilities for cure/control (Leventhal et al. 2008, Weinman et al. 1996). In other words, cognitive representations of illness are formed by assessing: what the illness is and what the illness label, e.g. 'cancer', means to the patient, what the causal mechanisms behind the illness are, how long the illness will last, what its consequences might be and whether and how the illness might be controlled or even cured (Weinman et al. 1996). Emotional representations (or appraisals) of a health threat may be linked to anticipatory and anticipated emotions induced by the threat (Loewenstein et al. 2001).

2.4.3 Secondary appraisal

Secondary appraisal is focused on what can (or cannot) be done in response to the stressful event (Lazarus and Folkman 1984). Lazarus and Folkman (1984) themselves note that the chosen terminology of primary and secondary appraisal was inadequate and potentially misleading, as it may be interpreted as a ranking of importance or indicate a sequential process. They refrain from suggesting new terms; however they stress that neither is more important than the other and that they may occur in parallel (Lazarus and Folkman 1984). Primary and secondary appraisal may also form a feedback loop, in which a quick primary appraisal and initial assessment of the most important coping resources may then be followed by a more in-depth primary and secondary appraisal (Shaw 1999).

During secondary appraisal the patient assesses the *coping potential*, i.e. *whether* anything can be done to actively deal with the threat (*problem-focused coping potential*) and/or the emotions it causes (*emotion-focused coping potential*). This is followed by a more detailed assessment of the *coping options* or resources i.e. *what* can be done to deal with the threat and associated emotions. This includes an appraisal of whether these can be implemented effectively and how likely they are to achieve the desired outcome (Lazarus and Folkman 1984). Lazarus and Folkman (1984) distinguish '*problem-focused*' coping options, designed to relieve the threat directly, and '*emotion-focused*' options, designed to regulate the emotions experienced in response to the threat (Lazarus 1993). Some coping options may be a source for both problem- and emotion-focused coping. For instance social contacts may provide informational and instrumental support (problem-focused) and at the same time be a source of love, affection and reassurance (emotion-focused) (Helgeson and Cohen 1996). Kramer (1993, O'Brien and DeLongis 1996) made a further addition to the ways of coping by describing relationship-focused coping, which is designed to moderate social relationships and is especially important in caregiver coping.

Others have proposed alternative ways to categorise coping, such as engagement (approach) versus disengagement (avoidance) coping and cognitive

versus behavioural coping, which should be acknowledged (Carver and Connor-Smith 2010, Latack and Havlovic 1992, Roth and Cohen 1986). The engagement/disengagement distinction differentiates coping which brings an individual closer to a stressor (approach) from that which allows the individual to distance themselves from the stressor (avoidance) (Carver and Connor-Smith 2010, Roth and Cohen 1986). For example, seeking social support would be defined as engagement coping, whereas distraction would be defined as disengagement coping. Similarly, the cognitive/behavioural distinction discriminates coping that occurs internally (cognitive) from that which requires external action (behavioural) (Latack and Havlovic 1992, Skinner et al. 2003). For instance, seeking social support would be defined as behavioural coping and positive re-appraisal as cognitive coping. Rather than being mutually exclusive, these ways of categorising coping overlap. Information seeking may be labelled as problem-focused, engagement and behavioural coping. Similarly, wishful thinking may be defined as disengagement and cognitive coping.

Due to this multidimensionality of coping some have gone so far as to suggest that coping should not be categorised in binary form at all, proposing a hierarchical system instead (Skinner et al. 2003). They suggest that binary approaches to categorisation are prone to misclassification and are therefore inadequate. Alternatively, they propose a hierarchical system with twelve overarching families of coping approaches (Skinner et al. 2003). Examples of families are problem-solving, information seeking, escape and support seeking. Each family has a specific function and includes several approaches (Skinner et al. 2003). Such a detailed categorisation may be most appropriate in research specifically studying individual coping responses. However, it may still be useful to use a binary categorisation in other contexts. Therefore, rather than there being one correct approach, the way in which coping is categorised becomes a question of preferred terminology, perhaps depending on the purpose of the study in which the categorisation is used. Whichever categorisation is selected, a clear statement of why it was chosen is essential.

Building on the models proposed by Power et al. (2011) and Balneaves and Long (1999), the framework presented in this chapter adopts Leventhal's (1980) distinction between cognitive and emotional representations, or appraisals, of the health threat and Lazarus and Folkman's (1984) categorisation of problem-focused and emotion-focused coping. This allows a similar breakdown of processes during primary and secondary appraisal, into cognitive or problem-focused approaches on the one hand and emotional processes on the other (Figure 2.2). Therefore coping strategies are referred to as problem- or emotion-focused in this chapter. However, it is acknowledged that other categorisations exist, and might apply, in parallel.

2.4.4 Coping responses

Once a threat has been appraised in primary and secondary appraisal, the individual selects one or more coping resources and initiates a coping response by implementing the selected strategies. This response seeks to reduce discrepancies between an appraised meaning of a specific situation and an individual's global meaning, which describes the person's understanding of themselves and the world (Park 2011, Pergament 1997). Selected coping strategies may include a mixture of problem- and emotion-focused approaches to facilitate meaning making. For instance, an individual may turn to their family for emotional support and actively seek information about the threat and available options in order to take action against it. The preferred coping strategy may be influenced by personal factors and characteristics, such as self-efficacy beliefs, past experiences and social settings (Carver and Connor-Smith 2010, Lazarus and Folkman 1984, O'Brien and DeLongis 1996, Shaw 1999, Smyth and Filipkowski 2010). Situational and temporal factors may also play a role in shaping appraisal and coping responses (Folkman 2011, Lazarus and Folkman 1984) and they may also depend on the patient's preferred coping style (Miller 1987, Miller et al. 1988). Miller (1987) distinguishes two coping styles: monitoring and blunting. While high monitors choose to seek out threat-relevant information and show low levels of distracting activity, high blunters choose to ignore or avoid threat-relevant information and show high levels of distracting activity (Miller 1987, Miller et al. 1988). Coping strategies may be

selected according to these styles. High monitors may lean towards strategies such as information seeking, while high blunterns preferably chose strategies such as avoidance and withdrawal.

Once a coping strategy has been implemented the threat may be reappraised and strategies may be adapted if the desired outcome was not achieved (Lazarus and Folkman 1984). The effectiveness of a coping strategy depends on its appropriateness or 'goodness of fit' for the situation (Lazarus 1993, Lazarus et al. 1985, Lazarus and Folkman 1984). Problem-focused approaches may be adaptive for modifiable situations, for instance when there is a choice between treatments for a medical condition, whereas emotion-focused approaches appear more effective in situations when nothing can be done, such as periods of waiting for the results of medical tests (Lazarus and Folkman 1984, Smyth and Filipkowski 2010). Coping flexibility describes the ability of individuals to adapt their coping strategy to cater for the coping demands of a specific situation (Cheng 2001).

2.4.5 The embedded decisional model of stress and coping

The embedded decisional model of stress and coping (Balneaves and Long 1999) describes the decision making process by outlining a series of questions informed by decision making and coping theory. The authors embed Lazarus and Folkman's transactional model and their illustration of system variables for stress (Lazarus et al. 1985, 1984) within Janis and Mann's conflict theory of decision making (1977). The resulting model acknowledges the transactional and multidimensional nature of decision making and coping. It explores causal antecedents, mediating processes and short- and long-term effects of a decision through a series of five questions: "What are the antecedent circumstances in making this treatment decision?", "What is at stake in making this treatment decision?", "What are my options in making this treatment decision?", "What are the possible ways of coping with this decision?" and "What are the adaptational outcomes of this decision?". These questions address different considerations important in the decision making process, including antecedents (such as personal beliefs, values and commitments), potential coping resources and temporal

influences. Balneaves and Long (1999) were among the first to integrate decision making and coping in an attempt to provide a more holistic vision of the deliberation process.

2.5 The Coping in Deliberation (CODE) framework

2.5.1 Introduction to the CODE framework

Coping with a health threat and its potential short- and long-term consequences is an integral part of the deliberation process in preference-sensitive healthcare decisions. Without appraisal and coping strategies, individuals may not be able to make informed, value-adjusted decisions, as their emotional state would hinder an effective analysis of the situation and their options (see section 2.3.3). The transactional functions of coping aid the decision maker in regulating their emotional state whilst going through the decision making process and may also allow them to devise active strategies to deal with a given threat. Therefore, integrating decision making theory and coping theory is a useful exercise, as it allows for a more holistic vision of human decision making in preference-sensitive healthcare contexts. By proposing a detailed framework of coping processes during deliberation, this chapter attempts to build on the ideas of Balneaves and Long (1999) as well as Power et al. (2011).

Balneaves and Long (1999) embedded Janis and Mann's (1977) conflict theory within Lazarus and Folkman's theory of stress, appraisal and coping (1984), proposing a model which considers antecedents, appraisal of options and potential coping resources associated with a decision. Power et al. (2011) emphasised the motivational role of emotions in decision making and made a clear distinction between threat- and decision-associated cognitive and emotional processes. The Coping in Deliberation (CODE) framework integrates and extends these ideas in the context of preference-sensitive deliberations in healthcare by proposing that each phase in deliberation (presentation of the health threat, choice, options, preference construction) results in cognitive and emotional appraisal and stimulates a coping process in its own right, the result of which can influence a patient's questions,

attitude and behaviour in other stages of deliberation. Deliberation in the CODE framework is therefore described as a multidimensional coping process, depicted in Figure 2.4 on the horizontal axis ('Deliberation') and vertical- axis ('Coping'). The framework applies to preference-sensitive decisions in healthcare contexts, meaning those which require the consideration of patient values and allow patient input as no clear medically recommended option exists.

2.5.2 Framework structure

The patient may move through the process depicted in Figure 2.4 from top to bottom and from left to right. At the start of the deliberation process the patient is presented with a health threat. During primary appraisal, they appraise this threat cognitively and emotionally by exploring questions relating to the threat itself and their feelings about it. During secondary appraisal, they appraise the coping potential and the coping options that could be helpful in dealing with this threat, for example their ability to find out more (problem-focused coping option) and the possibility of seeing a positive side to the situation (emotion-focused coping option). Then, they may select one or more coping strategies that could be helpful in dealing with this health threat. For instance they may prefer a problem-focused approach and decide to find out as much as possible about the diagnosis and their options, or they may decide to turn to their faith (emotion-focused). The background colour in Figure 2.4 indicates the question type as forming part of cognitive (light) or emotional (dark) appraisal and as being a problem-focused (light) or emotion-focused (dark) coping option. The shading depicts the overlap between the two, i.e. where a question may contribute to both cognitive and emotional appraisal and where a coping option may be both problem- and emotion-focused.

Figure 2.4 - The Coping in Deliberation (CODE) framework (Witt et al. 2012)



The coping options selected and implemented in response to the presentation of the health threat will determine the patient's reaction to, and interest in, the idea of choice and the different treatment or prevention options, which will be presented in the next stages of the deliberation process (see sections 2.3.2 and 2.3.3). For instance, a patient who feels extremely anxious after being told about a certain health threat may initially lean towards an avoidant coping behaviour to regulate the anticipatory emotion that they are experiencing. This patient may then be less interested in the presentation of choice and options, as this would require them to confront the health threat again. In contrast, a patient who feels hopeful that knowing about the health threat presented will empower them to act upon the threat may be very interested in the presentation of choice and available options, as this would cater for their preferred problem-focused coping style.

While this description depicts a relatively linear approach to decision making and coping, one has to consider that coping is interactive, multidimensional and transactional (Lazarus and Folkman 1984). Primary and secondary appraisals can occur simultaneously and individuals may revisit certain threats, reappraise their situation and readjust their coping efforts. For instance, the patient who felt anxious after initial presentation of the health threat and chose an avoidant coping behaviour may over time come to the realisation that avoidance does not reduce their worry. They may then decide that confronting the threat and learning about options available to counteract it may be more adaptive and therefore readjust their coping effort.

Individuals may also skip certain steps in the process, or first appraise a threat briefly to return later for a more comprehensive appraisal (Shaw 1999). The framework aims to portray this possibility of moving through the process in a non-linear fashion by using double-pointed arrows. Until a final decision is reached and implemented, a decision maker may re-appraise any phase of the coping and deliberation process. After considering the options, they may, for instance, reappraise the threat and decide that the risks associated with the options are

more threatening to them than the original threat itself at this point in time, therefore delaying a decision. In another example, the decision maker may initially appraise the choice as a challenge and prefer to cope by playing an active role in decision making. However, after considering the options, they may feel overwhelmed and reappraise the choice as a threat, preferring their doctor to make a decision for them instead.

2.5.3 *Adaptability of the framework*

In the CODE framework, a series of questions outline possible cognitive and emotional appraisals and possible problem- and emotion-focused coping resources that a patient might explore in response to each deliberation phase. The questions in the generic version of the CODE framework were designed to be adaptable to different healthcare decisions. Statements at the end of each column represent some of the coping strategies a patient might select, which may again be adapted to different medical contexts. The framework was designed to accommodate a wide range of preference-sensitive medical decisions, ranging from decisions about genetic testing and cancer prevention to those about choices between different medications or types of surgery. To illustrate the adaptability of the framework, consider the following three healthcare decisions and how questions within the framework may be adapted to these contexts.

2.5.3i Increased genetic risk of ovarian cancer: Women from families with a high incidence of ovarian, and some other, cancers may be at increased risk of ovarian cancer. A woman with such a family history may initially appraise her risk. To do this she may ask what ovarian cancer is, what genetic risk means and how threatening this risk is to her health. She will also respond to the threat emotionally and explore how she feels about being at increased risk, potentially drawing on past experiences of observing ovarian cancer in relatives. She may then appraise the resources that could help her deal with that risk. She may wonder whether she could seek more information, talk to her clinician or family, or try to find a positive side to the situation. During the presentation of choice she may ask why there is a

choice at all, may inquire about the timescale she has for making this decision and may wonder who she should involve. She may also explore coping resources which could help her to make this decision, such as support from her clinician and/or family and friends. Similarly, during the deliberation about options, the woman may ask specific questions about the risks and benefits of options such as risk-reducing salpingo-oophorectomy (i.e. surgery to remove healthy ovarian tissue) and the effects of surgery on her life, and she may consider follow-on decisions such as hormone replacement therapy (HRT) and alternatives such as ovarian screening (Rosenthal and Jacobs 2006). She may also emotionally appraise potential outcomes of different options, for example she might predict that she will feel a sense of loss if she chose to have surgery, because she would lose her fertility. Past experiences of others who have made this decision – for instance a sister who has opted for surgery – might help her in forming anticipatory emotions about options. She may also explore the coping resources at her disposal that could help her deal with the potential consequences of options and the decision, such as emotional and instrumental support.

The questions within the framework can be adapted accordingly to reflect these cognitive and emotional processes. It should be noted that questions and coping strategies will vary not only depending on the decision at hand, but also on situational factors and the characteristics of the individual patient. In addition to preferred coping style, other influences such as life stage, experience, self-efficacy and values will play a role (Carver and Connor-Smith 2010, O'Brien and DeLongis 1996, Shaw 1999, Smyth and Filipkowski 2010). In the example used above, a patient at risk of ovarian cancer who is aged 55 may not consider HRT, whereas a woman in her early 40s might explore that option in detail. Similarly, a single mother would not look to a partner for coping support, but might turn to her clinician, family or friends, whereas someone in a relationship might value input from her partner and explore how risk-reducing surgery would affect their relationship. A religious person might explore whether she could find strength in her faith as a coping resource.

2.5.3ii Amniocentesis: To demonstrate the flexibility of the CODE framework, consider a different healthcare decision, such as amniocentesis. Parents are required to cope with the fact that a routine test has indicated a potential problem and are now faced with the decision of whether or not to explore this further. During appraisal of the health threat to their unborn child they may ask what the current result means, how it might affect their pregnancy and who they could rely on for support. During deliberation of the options they may consider the risks and benefits of amniocentesis, ask themselves what they would do if the test confirmed an abnormality, consider termination of pregnancy and explore their coping resources for different scenarios. During preference construction they may ask whether they both agree and how likely they are to experience regret.

2.5.3iii Coronary artery disease (CAD): Another example may be selected from the context of CAD, where patients may be asked whether they would like to have a catheter investigation to check for CAD or whether they would prefer medical therapy. Patients in this situation need to appraise and cope with the fact that they are at risk of CAD and what this means for their life. They are also required to appraise their options, such as catheter investigation, asking about the risks and benefits and exploring how they might feel about these. Knowing someone who went through this procedure might help them with primary appraisal. During secondary appraisal they may explore whether they can turn to others for support or simply avoid dealing with the situation if they feel overwhelmed.

These three examples depict preference-sensitive medical decisions and illustrate the flexibility of the framework, which allows the addition, removal or adaptation of questions depending on the decision at hand. The generic framework can be revised to describe any preference-sensitive decision. Furthermore, questions may then be added, removed or adapted depending on the individual patient's coping style, life-stage and experiences, if a specific patient's decision needs to be described in detail.

2.5.4 *Links to theoretical concepts*

Incorporated in the CODE framework are many of the concepts found within both coping and decision making theories (see sections 2.3 and 2.4). The questions posed during the appraisal process were selected to represent a wide range of considerations and influences. For instance, personal values and goals, which can affect the interpretation of different options, are considered when the patient assesses the relevance of the health threat and again when they evaluate the potential effects of options on their life (Balneaves and Long 1999, Leventhal et al. 1980, Shaw 1999). Emotional responses are incorporated when patients explore how they feel about a threat, the choice and options (Loewenstein et al. 2001, Power et al. 2011).

The coping resources and strategies in the framework were selected to represent different coping strategies that patients may use, such as vigilance, avoidance and seeking social support (Henderson et al. 2006, Janis and Mann 1977). It should be noted that not all possible coping strategies are included, as this would be beyond the scope of the framework. However, any coping strategy may be incorporated when adapting the framework. Preferred coping strategies may also change over time or vary from one deliberation stage to another, as coping is transactional and multidimensional (Lazarus and Folkman 1984).

The contents of the CODE framework are not aiming to be comprehensive, rather they reflect a variety of different questions and coping options that a patient may consider whilst going through the deliberation process, maintaining the multidimensional and transactional functions of coping (Balneaves and Long 1999). The framework does not propose that one coping strategy is more effective than another, as different strategies may be appropriate in different situations (Lazarus and Folkman 1984). It therefore includes a range of strategies and allows decision makers to select any number and adapt their approach at a later time, if necessary. The number and types of questions asked, the answers to these questions, the choice of coping strategies and ultimately the decision itself will depend on the type

of health threat, the options, the patient's coping style, their preferences, past experiences, personal values and ability to forecast feelings accurately.

The framework also includes a consolidation phase following the actual decision. Consolidation is in essence a coping mechanism designed to defend a decision from regret in the future (Connolly and Reb 2005, Svenson 1992). The inclusion of this phase allows the framework to describe deliberation beyond the actual decision and acknowledges that the consequences of a decision should be an integral part of any decision making model, as patients ultimately have to live with their decision and its consequences.

2.6 Discussion

2.6.1 The CODE framework in context

Leventhal et al. (1980) were the first to consider the importance of emotions in addition to purely cognitive processes in response to threats. More recently, Power et al. (2011) distinguished cognitive and emotional representations formed in response to a health threat from those formed in response to the need for a decision. They also argued that emotions play an important motivational role during decision making. Following on from this idea, the preliminary CODE framework proposes a further distinction between the different stages within the decision and postulates that each of these stages gives rise to its own individual cognitive and emotional appraisal process, essentially adding a further degree of detail to Power et al.'s model, which only differentiated the threat and the decision. This additional distinction allows a more fine-grained analysis of the appraisals which occur during the different deliberation stages. In agreement with Power et al. (2011) and other theorists, such as Lazarus and Folkman (1984), the CODE framework considers emotional appraisal and emotion-focused coping processes to be of equal importance to cognitive appraisal and problem-focused coping processes. Both occur in parallel, influence each other and are essential for good decision making.

The embedded decisional model of stress and coping describes decision making as a series of relatively general questions (Balneaves and Long 1999). The CODE framework similarly identifies questions relating to deliberation and coping processes, but does so in a more detailed fashion. For instance, what Balneaves and colleagues (1999) cover with the question “What are my options in making this decision?”, the CODE framework explores with a series of more detailed questions about benefits and risks, effects on life and possible follow-on decisions. Similarly, Balneaves’ question “What are the possible ways of coping with this decision?” is split into a series of several individual questions during secondary appraisal, describing different coping strategies at different stages in the deliberation process, such as “Can I talk to my family/friends?”, “Can I change how I feel about this?” and “Can I transfer this decision?”. Balneaves’ questions “What are the antecedent circumstances in making this treatment decision?” and “How is the nature of the decision shifting over time?” are integrated more indirectly. The questions within the CODE framework can be adapted according to antecedent circumstances that apply to a patient. Throughout the framework, multiple questions relate to personal values and past experiences, such as “Do I have experiences that could help me imagine what it could be/feel like?” and “Is this option congruent with my goals and values?” The potential for shift over time is acknowledged by the non-linear structure, which allows decision makers to revisit stages, reappraise threats and adopt new coping strategies at any time.

Additionally, the framework includes a consolidation phase following the decision, as proposed by Svenson (1992). This allows a description of the deliberation process beyond the point of decision making. Consolidation is a way of coping with a decision and its outcome and may be especially important in irreversible decisions to avoid regret. In the case of reversible decisions, inclusion of the consolidation stage in the framework is also warranted, as these types of decisions may be revisited, starting a renewed deliberation process, if the decision maker finds that they did not make the right decision during consolidation. Similarly to Balneaves’ and Long’s (1999) model, the CODE framework explores deliberation

from the patient perspective, which may allow practitioners to develop a more detailed understanding of patients' appraisal processes during deliberations in healthcare.

2.6.2 Practice implications and future research

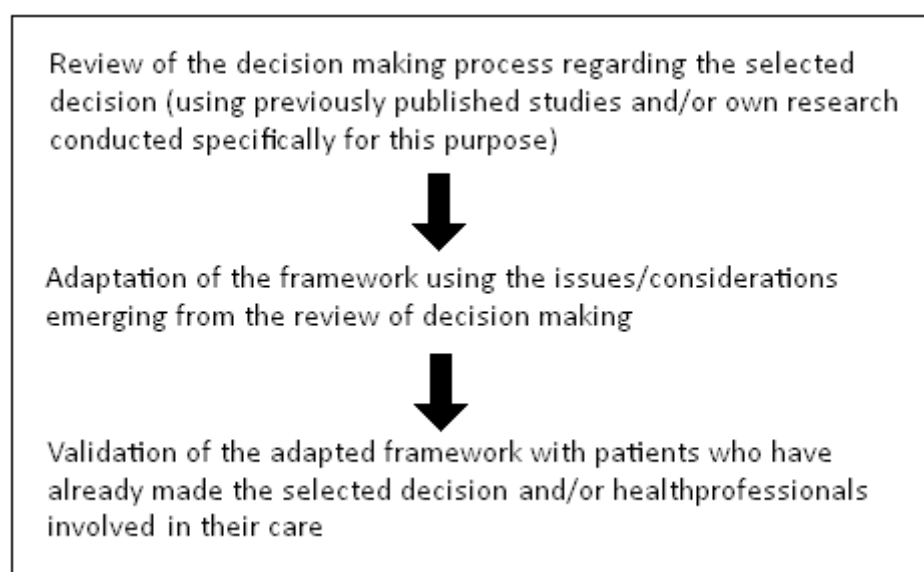
Patient-centred care (PCC) and shared decision making (SDM) are becoming increasingly important in healthcare, especially in the context of preference-sensitive decisions which require patient input (Coulter 2009a, Coulter et al. 2011, Elwyn et al. 2000, Elwyn et al. 2009a, O'Grady and Jadad 2010, Secretary of State for Health 2010, Svenson 1992). In order to provide PCC, practitioners are expected to understand their patients' needs and concerns and to address them accordingly. The preliminary CODE framework provides a description of preference-sensitive decision making from a patient perspective, which may ultimately – when adapted to specific decisions – help practitioners to better understand their patients' questions and concerns by shedding light on underlying cognitive and emotional processes.

Notably, the framework can also provide a theoretical underpinning, and potentially practical guidance, for the development of decision support interventions which are designed to support SDM. Currently, many of these tools lack a theoretical basis and may therefore be of questionable benefit to patients (Durand et al. 2008). The CODE framework was developed with this purpose in mind. It proposes specific questions at each stage of the deliberation process, which could – either directly or in adapted form – be integrated in decision support materials. A tool based on the framework would include information to support cognitive appraisal as well as coping advice to encourage adaptive coping. Interventions guided by the CODE framework – which itself is based on theoretical models of decision making and coping by Lazarus and Folkman (1984), Balneaves and Long (1999), Power et al. (2011) and others – would have a solid theory base. Therefore, it would avoid the pitfall of many currently available patient decision support interventions, which lack the theoretical underpinning that is important to

enable predictions about the likely effects of the intervention (Durand et al. 2008, Elwyn et al. 2011b). The framework may also be adapted to decisions for which interventions are already available. It could then be used to assess whether the content of these interventions addresses the issues patients explore during deliberation, as outlined in the adapted framework.

The framework clearly needs to be adapted to actual decisions in healthcare and tested with patients to explore its applicability and provide feedback on validity and potential amendments that may be needed. The adaptation of the preliminary framework to specific decisions should take into account the views of stakeholders involved in those decisions, including patients, patient organisations and health professionals, to allow for an accurate representation of questions and coping strategies patients may explore. Adaptation may be achieved by following a three step process (Figure 2.5). Once adapted versions are available, empirical evidence about the use of such versions as a guide for practitioners in consultations, an assessment tool for existing interventions and a basis for decision support development should also be gathered.

Figure 2.5 - Guide for the adaptation of the CODE framework



2.6.3 *Strengths and limitations*

The preliminary CODE framework is a theoretical description of deliberation and coping processes, purposefully generic and designed to be applicable to a range of preference-sensitive decisions. While this makes it extremely flexible and allows a variety of potential uses in different medical contexts, the framework now needs to be adapted to specific decisions and tested with patients. A thorough adaptation should firstly focus on determining considerations important in a selected decision (see Chapter 3 and 4). These considerations, alongside an understanding of the specific decision making process in the selected context (e.g. short or long deliberation time and existing barriers and facilitators), should then be used to operationalise the questions proposed in the framework (Chapter 5). Following adaptation, a retrospective investigation of one, or ideally more, actual decisions should be conducted to reveal whether decisions were described accurately by the framework, before it can then be used in further research or development work.

To date, the framework has only undergone face validity testing and has not been subjected to content validity assessment (Anastasi 1988, Carmines and Zeller 1977). This limits the certainty with which it can be assumed that each question within the framework is truly representative of the suggested associated deliberation and appraisal/coping phases. In the future, the CODE framework would benefit from content validation through a group of reviewers with expertise in decision making and coping theory. The questions may then be adapted. Similarly, the framework may also benefit from convergent validity testing to evaluate whether it maps onto validated measures of appraisal, coping and deliberation, such as the brief COPE (Carver 1997) and the decisional conflict scale (O'Connor 1995).

Few theories to date have specifically considered emotional and cognitive appraisal processes in conjunction with coping efforts when patients are making decisions about their health. The questions included in the CODE framework attempt to describe issues explored during patients' appraisals at each stage of the

deliberation process. This may provide health professionals with a basis for looking at the variety of patients' questions and concerns. The framework also draws attention to the fact that deliberation and coping processes are closely linked and therefore may help practitioners to explore coping resources together with their patients, potentially adding a new element to PCC.

In the future, the framework may benefit from an extension which allows the inclusion of patient information and coping needs, identifying specifically what help patients would like to be available to support their coping effort at each stage in the process. This would additionally strengthen the framework's value as a support tool for PCC and a guide for the development of patient decision support.

Some limitations to the applicability of the CODE framework should also be noted. The framework is only applicable to preference-sensitive healthcare decisions that require a relatively elaborate exploration of choice and options. Medical decisions that have only one clinically recommended option would not fall within this remit and are therefore not described by the framework. However, certain sections of the framework, such as issues explored during primary and secondary appraisal of the health threat, might still apply to such situations.

As discussed earlier, the questions and coping strategies in the framework do not represent an exhaustive list of possible issues explored. Instead they act as examples that can be changed and adapted to any specific preference-sensitive decision and any patient. Therefore, any number of considerations may be missing from the current framework; however it has the capacity to encompass those issues in future adaptations.

Finally, as the framework is currently purely theoretical, it cannot be assumed that it is truly reflective of deliberations in healthcare. The framework now needs to be operationalised to confirm whether it accurately describes such preference-sensitive medical decisions and may need to be amended if it is found to be inaccurate.

2.6.4 *Conclusions*

This chapter described decision making and coping as it emerges from current theory. It also presented the development of a multidimensional, process-oriented framework of patients' coping responses during deliberations in healthcare. The preliminary CODE framework has the potential to be adapted to a number of preference-sensitive medical decisions and to act as a support tool for medical practitioners and as a guide for the assessment and/or development of patient decision support interventions.

One such example of adaptation is shown in Chapter 5, which reports on the development of a CODE framework adapted to describe decision making about risk-reducing salpingo-oophorectomy by women at increased risk of ovarian cancer. This adapted framework is then used to assess currently available decision support interventions that include the option of risk-reducing salpingo-oophorectomy (Chapter 6) and to develop a new intervention for this context, specifically for women located in the UK (Chapter 7). Adaptations to other decisions may follow.

3 Risk-reducing bilateral salpingo oophorectomy: A dilemma for patients

3.1 Introduction

Women with a family history indicative of hereditary breast and ovarian cancer (HBOC) or Lynch Syndrome may be at increased risk of ovarian cancer (Chapter 1; Barrow et al. 2009, Chen and Parmigiani 2007). In the UK, women found to be at moderate to high risk of this cancer may be offered risk-reducing salpingo-oophorectomy (RRSO), which involves surgical removal of healthy ovarian tissue and has been shown to substantially reduce the risk of ovarian and breast cancer (Rebbeck et al. 2009, Schmeler et al. 2006a). However, RRSO has a number of drawbacks including risks associated with surgery, but also issues such as infertility and surgical menopause in women who are pre-menopausal prior to the procedure. Therefore the decision to undergo, defer or decline RRSO is preference-sensitive, depending on the goals and values of the individual patient (Chapter 1; Elwyn et al. 2009a). Preference sensitive decisions require the patient to play an active part in the deliberation process, become knowledgeable about available options, weigh positive and negative attributes of these options and clarify their own values in order to ultimately make a decision in line with their personal preferences. The process of preference-sensitive decision making and coping has been described previously in the Coping in Deliberation (CODE) framework (see Chapter 2 and Witt et al. 2012).

How women make decisions about RRSO in the context of familial risk has been explored in a number of quantitative and qualitative studies to date. A detailed understanding of the decision making process, and issues that are influential during this period, is essential in order to provide appropriate counselling to these women and to develop suitable decision support materials. The aim of this chapter is to summarise the knowledge generated in previous research regarding (i) the concerns and issues which are explored by women during deliberations about RRSO and (ii) what these women expect or desire in terms of

support materials or interventions designed to facilitate decision making. The search focused on women's expectations of decision support, rather than coping advice, as this project aimed to develop a decision support intervention with coping components, rather than a coping intervention per se. The results presented here may be helpful as a guide when developing materials for these women and will be mapped onto the generic CODE framework in Chapter 5 of this thesis to produce an RRSO-adapted version of the CODE framework.

3.2 Methodology

3.2.1 Literature search strategy

A systematic search of the literature about women's decision making regarding RRSO was conducted in July 2012 to identify issues considered during deliberations about this surgery. Databases (Embase, MEDLINE, MEDLINE In-Process and PsycINFO) were searched via Ovid for studies published between 1996 and July 2012. Search terms used in the title search are detailed in Table 3.1. Duplicates were removed using the Ovid de-duplicate function prior to review of the abstracts. No grey literature was included in this review.

3.2.2 Inclusion and exclusion criteria

Abstracts of identified publications were then reviewed and primary exclusion criteria applied. Publications were excluded if they were not concerned with (i) decision making processes, (ii) personal cancer risk reduction or prevention, (iii) ovarian cancer and/or oophorectomy or (iv) if they exclusively included participants who were not at increased risk of ovarian or breast cancer, such as those considering oophorectomy in the context of a planned hysterectomy for a benign indication. Increased risk was defined as risk assessed to be above that of the general population. In a secondary exclusion step, publications which did not

Table 3.1 - Search terms for title search in Ovid

| Term 1 | | Term 2 |
|--|-----|---|
| Decision\$ Choice\$ Option\$ Discussion\$ Decide Deciding Choose Choosing Perception\$ Concern\$ Question\$ Perspective\$ | AND | Risk reducing salpingo oophorectomy Risk reducing oophorectomy Prophylactic salpingo oophorectomy Prophylactic oophorectomy Prophylactic surgery Risk reducing strategies Risk reducing strategy Ovarian cancer risk Ovarian cancer screening Ovarian cancer prevention Inherited ovarian cancer Inherited breast ovarian cancer Inherited ovarian breast cancer Familial ovarian cancer Familial breast ovarian cancer Familial ovarian breast cancer BRCA Lynch Hereditary non-polyposis colorectal cancer / HNPCC |

explore (i) women's self-reported intentions, opinions and concerns or (ii) women's demographic characteristics associated with decisions about RRSO were also excluded, as the current research aimed to specifically determine factors that influence women's decisions about RRSO. Excluded in this step were papers which exclusively focused on health professionals' decision making (Plusguin et al., 2011; Stany et al, 2010), those which described decision analysis using Markov models (Armstrong et al., 2004; van Roosmalen et al., 2002) and those which did not report on original research. Publications which reported on participants who were eligible for and expressed an intention to undergo RRSO, but had not had surgery at the time of the study, were included, as it has been shown that intention to undergo RRSO does translate into actual uptake (Tiller et al. 2002).

3.2.3 Complementary search

Further publications were identified through previously published reviews with a similar, albeit wider, focus. These often explored RRSO together with risk-reducing mastectomy and/or breast and ovarian screening decisions. The initial

search identified a review on breast and ovarian cancer risk management options (Howard et al. 2009a), a review on psychosocial factors influencing decisions about risk-reducing surgeries (Fang et al. 2005) and one on predictors of breast and ovarian screening and risk-reducing surgery uptake (De Leeuw et al. 2008). An additional review on the uptake and experiences of women considering RRSO was also identified (Miller et al. 2010). References from these reviews were selected according to the criteria detailed above. All publications exclusively concerned with breast/ovarian cancer screening and/or prophylactic mastectomy were excluded, as the focus of the current review was on decisions about RRSO.

3.2.4 Appraisal and analysis

All publications were critically appraised using an appraisal checklist from the Critical Appraisal Skills Programme (CASP) collaboration (see Appendices 3.1 and 3.2) appropriate for the study's main method (qualitative or quantitative). The main results of each study were summarised and factors explored during, or associated with, decisions about RRSO were extracted. Similar to Howard et al.'s approach (2009a), the issues identified were assigned to one of five categories: 'medical/physical', 'psychological/emotional', 'social context', 'demographic' or 'other'. As the review included a large number of qualitative studies, statistical significance was not a criterion for inclusion. Additionally publications were reviewed to identify any specific expectations women have of the content and format of support materials or interventions designed to facilitate these decisions.

3.3 Results

The literature search identified 115 publications of which 97 were excluded after application of the exclusion criteria (Figure 3.1). Thirty-one additional publications were identified through a complementary search of reference lists of previously published literature reviews (De Leeuw et al. 2008, Fang et al. 2005, Howard et al. 2009a, Miller et al. 2010). The final publication list includes quantitative (n=38) and qualitative studies (n=11) (Table 3.2).

Figure 3.1 - Results of the literature search via Ovid

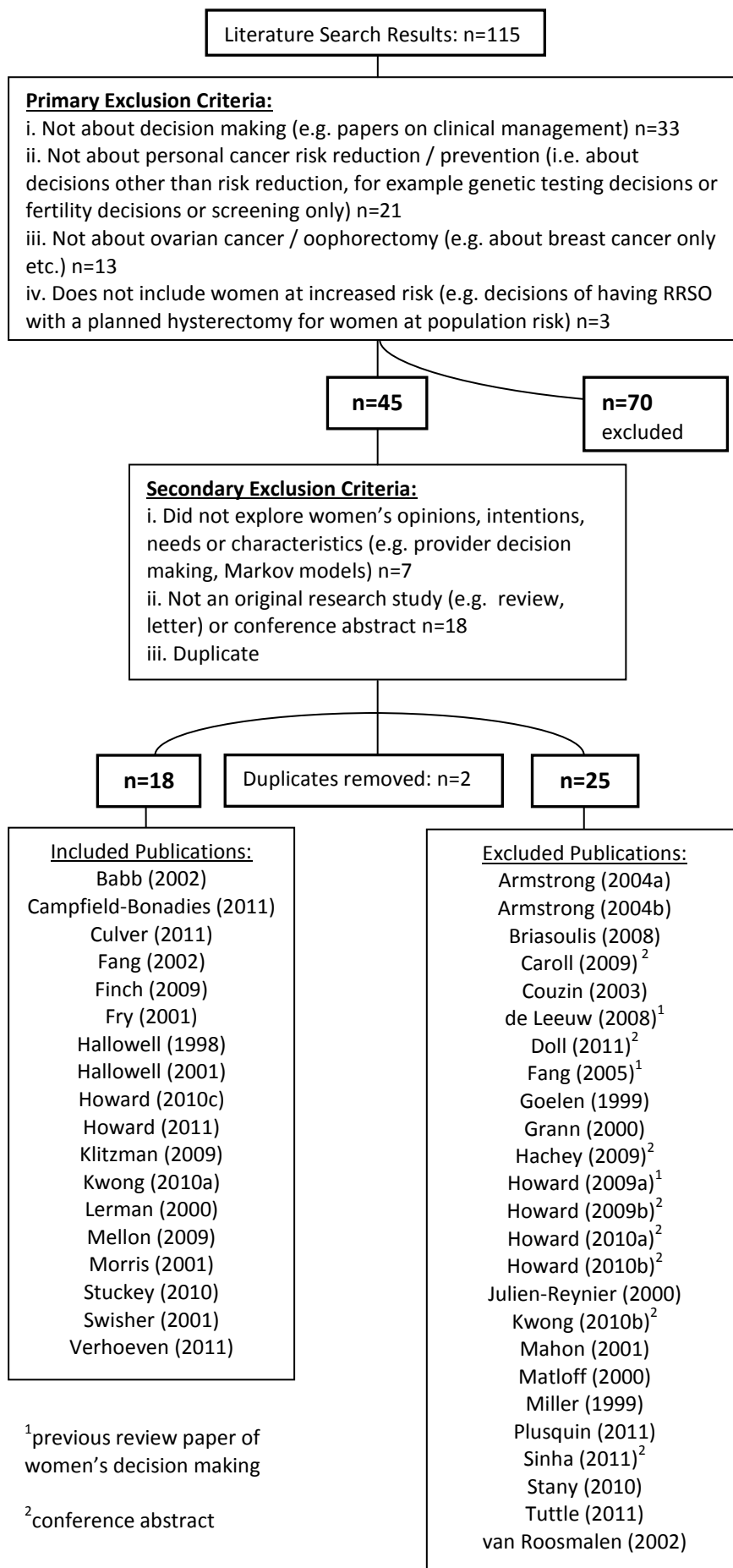


Table 3.2 - Publications included in final review (1 from Howard et al., 2009; 2 from Miller et al., 2010; 3 from de Leeuw et al., 2008; 4 from Fang et al., 2005)

| First Author | Year | Study | Mode of data collection | Study population |
|-----------------------|-------------|--------------|---|---|
| Antill ¹ | (2006) | Quant. | Structured self-administered questionnaire | 182 women at moderate (12.5-25%) or high (25-85%) risk of breast cancer who attended Familial Cancer Clinics in New South Wales / Victoria between 1999 and 2000 |
| Babb | (2002) | Qual. | Semi-structured telephone interview | 60 women who had undergone prophylactic oophorectomy (n=30) or ovarian cancer surveillance (n=30) at Washington University |
| Beattie ² | (2009) | Quant. | Medical records | 240 BRCA positive women enrolled in the Cancer Risk Program at the University of California (USA) who received genetic test results between 1996 and 2006 |
| Botkin ^{1,2} | (2003) | Quant. | Structured telephone interview | 129 women from the K2082 kindred in Utah of whom 37 were BRCA1 positive and 92 were BRCA1 negative when tested for the kindred-specific BRCA1 mutation |
| Bradbury ¹ | (2008) | Quant. | Medical Records | 88 BRCA positive women who had received clinical services at the University of Chicago Cancer Risk Clinic between 1996 and 2003 |
| Brain ¹ | (2004) | Qual. | Semi-structured interview | 10 women newly identified as being at increased risk of developing familial ovarian cancer by the Cancer Genetics Service for Wales (UK) |
| Campfield-Bonadies | (2011) | Quant. | Structured self-administered questionnaire (incl. 3 open ended questions) | 98 BRCA positive women who had undergone prophylactic bilateral salpingo-oophorectomy and received counselling at the Yale Cancer Genetic Program |
| Claes ¹ | (2005) | Quant. | Structured self-administered questionnaire and semi-structured interview | 68 women from BRCA positive families who underwent genetic testing at the Centre for Human Genetics, University of Leuven (Belgium), of whom 34 were BRCA positive and 34 were BRCA negative |
| Culver | (2011) | Qual. | Focus group | 11 BRCA positive breast cancer survivors who had elected RRSO (=1), RRM (n=1), both (n=8) or screening (n=1) and were enrolled in the City of Hope Clinical Cancer Genetics Hereditary Cancer Registry (USA) |
| Evans ² | (2009b) | Quant. | Medical records | 211 BRCA positive women assessed at the Breast Cancer Family History Clinic of the regional genetics service for the Northwest of England in Manchester (UK) [Cohort in which uptake of bilateral risk-reducing salpingo oophorectomy was examined] |
| Fang | (2002) | Quant. | Structured telephone interview | 80 unaffected women who have a first degree relative with ovarian cancer and were enrolled in the Family Risk Assessment Program at Fox Chase Cancer Centre (USA) |
| Fang ¹ | (2003) | Quant. | Structured self-administered questionnaire | 76 women with a family history of ovarian cancer enrolled in the Family Risk Assessment Program at Fox Chase Cancer Centre (USA) |

Table 3.2 cont. - Publications included in final review

| First Author | Year | Study | Mode of data collection | Study population |
|------------------------|-------------|--------------|--|---|
| Finch | (2009) | Quant. | Structured self-administered questionnaire | 127 BRCA positive women who had undergone prophylactic salpingo-oophorectomy at the University Health Network, Toronto (Canada) |
| Friebel ¹ | (2007) | Quant. | Structured self-administered questionnaire and medical records | 537 BRCA positive women tested between 1994 and 2006 in one of 17 centres in North America and Europe |
| Fry | (2001) | Quant. | Structured self-administered questionnaire | 58 women who had undergone prophylactic oophorectomy (n=30) or annual ovarian cancer screening (n=28) registered on the Family Ovarian Cancer Clinic database of Western General Hospital (Edinburgh, UK) |
| Hamilton ¹ | (2009) | Qual. | Semi-structured telephone or email interview | 44 BRCA positive women recruited from two internet sites for BRCA carriers (www.facingourrisk.org and www.youngsurvival.org) |
| Hallowell | (1998) | Qual. | Semi-structured telephone interview and observations | 41 women with a family history of breast and/or ovarian cancer who attended genetic counselling at the Cambridge Cancer Family History Clinic (UK) |
| Hallowell ¹ | (2000) | Qual. | Semi-structured face-to-face interview | 23 women who had had prophylactic bilateral oophorectomy before age 46; recruited from the UK Cancer Coordinating Committee Familial Ovarian Cancer Register |
| Hallowell | (2001) | Qual. | Semi-structured face-to-face interview | 49 women who had undergone prophylactic oophorectomy (n=23) or ovarian cancer screening (n=26) and were registered on the UK Cancer Coordinating Committee Familial Ovarian Cancer Register, the Risk Advisory Clinic at St Bartholomew's Hospital (London) or the Cancer Family History Clinic at Royal Marsden (London) |
| Howard | (2010c) | Qual. | Semi-structured face-to-face interview | 22 BRCA positive women (16 had had risk-reducing oophorectomy, 3 had had risk-reducing mastectomy) participating in a hereditary cancer program at the University of British Columbia (Canada) |
| Howard | (2011) | Qual. | Semi-structured telephone interview | 22 BRCA positive women (16 had had risk-reducing oophorectomy, 3 had had risk-reducing mastectomy) participating in a hereditary cancer program at the University of British Columbia (Canada) |
| Hurley ¹ | (2001) | Quant. | Structured telephone interview | 94 women with a family history of breast and/or ovarian cancer enrolled in the Family Risk Assessment Program at Fox Chase Cancer Centre (USA) |
| Klitzman | (2009) | Qual. | Semi-structured face-to-face interview | 32 women at risk of breast cancer of whom 8 were BRCA positive, 10 were BRCA negative, 1 had an indeterminate result and 13 were untested ; recruited through various methods via Columbia University, New York (USA) |

Table 3.2 cont. - Publications included in final review

| First Author | Year | Study | Mode of data collection | Study population |
|---------------------------------|-------------|--------------|---|--|
| Kram ² | (2006) | Quant. | Structured self-administered questionnaire | 99 women tested for a BRCA mutation at the Hadassah Hebrew University Hospital in Jerusalem (Israel) between 1995 and 2001, of whom 43 were BRCA positive and 56 were BRCA negative |
| Kwong | (2010a) | Quant. | Medical records | 31 breast and ovarian cancer index patients positive for BRCA and 41 family members of index patients positive for BRCA who participated in genetic counseling and testing in genetic centres in China |
| Lerman | (2000) | Quant. | Structured telephone interview | 216 female members of BRCA1/2 linked families enrolled in a prospective observational study from 1994 to 1997, of whom 84 were BRCA positive, 83 were BRCA negative and 49 declined to receive test results |
| Madalinska ² | (2005) | Quant. | Structured self-administered questionnaire | 846 women from a hereditary breast and/or ovarian cancer family who has sought advice about preventative measures at a gynecology department in one of eight hospitals in the Netherlands between 1996 and 2001 |
| Madalinska ^{1,2} | (2007) | Quant. | Structured self-administered questionnaire | 160 BRCA positive women over the age of 35 who did not wish to have more children; recruited between 2002 and 2004 from gynecology departments in seven of eight hospitals in the Netherlands that have a clinical genetics centre |
| Meijers-Heijboer ² | (2000) | Quant. | Medical records | 682 unaffected family members from 53 consecutive families with known BRCA mutations; recruited through the Rotterdam Family Cancer clinic (the Netherlands) between 1994 and 1998 |
| Meijers-Heijboer ^{1,2} | (2003) | Quant. | Medical record and structured interviews | 220 women with breast and/or ovarian cancer from 112 families with known BRCA mutations; recruited through the Rotterdam Family Cancer clinic (the Netherlands) before 2000 |
| Meiser ¹ | (1999) | Quant. | Structured self-administered questionnaire | 95 women with a family history of breast and ovarian cancer who approached one of 14 Familial Cancer Clinics in five Australian States between 1996 and 1999 |
| Meiser ¹ | (2003) | Quant. | Structured self-administered questionnaire | 371 women with a family history of breast cancer enrolled in the Kathleen-Cunningham Consortium for Research into Familial Breast Cancer (Australia) |
| Mellon | (2009) | Quant. | Structured face-to-face interviews and structured self-administered questionnaire | 146 women with invasive breast or ovarian cancer identified through the Metropolitan Detroit Cancer Surveillance System and 146 female blood relatives (one relative per cancer patient) |
| Metcalfe ² | (2008a) | Quant. | Structured telephone/mail questionnaire | 2677 BRCA positive women assessed for genetic risk at one of 41 centres in 9 countries |

Table 3.2 cont. - Publications included in final review

| First Author | Year | Study | Mode of data collection | Study population |
|-------------------------|-------------|--------------|---|--|
| Metcalfe ^{1,2} | (2008b) | Quant. | Structured telephone/mail questionnaire | 517 BRCA positive women assessed for genetic risk at one of 11 centres in Canada |
| Morris | (2001) | Quant. | Unclear | 83 women who underwent in-depth family history evaluation for breast and/or ovarian cancer |
| Phillips ¹ | (2006) | Quant. | Structured questionnaire (interviewer and self-administered) | 142 BRCA positive women enrolled in the Kathleen-Cunningham Consortium for Research into Familial Breast Cancer (Australia) who, between 2001 and 2005, had completed the questionnaire sent out 3 years after study entry |
| Ray ¹ | (2005) | Quant. | Structured self-administered questionnaire (incl. 2 open ended questions) | 62 women enrolled in the Hereditary Cancer Risk Database Project at the Arizona Cancer Centre (USA), who met criteria for hereditary risk of breast or ovarian cancer and had at least one breast or both ovaries remaining |
| Scheuer ¹ | (2002) | Quant. | Structured self-administered and telephone questionnaire | 251 BRCA positive individuals (includes 18 males) who received genetic test results at Memorial Sloan-Kettering Cancer Centre (USA) between 1995 and 2000 |
| Schmeler ¹ | (2006b) | Quant. | Medical Records | 106 BRCA positive women registered on the University of Texas MD Anderson Cancer Centre Clinical Cancer Genetics database who had received test results between 1996 and 2005 |
| Schwartz ² | (2003) | Quant. | Structured telephone questionnaire | 289 women at perceived high risk who had received genetic counseling and testing through the Lombardi Cancer Centre (USA) Cancer Assessment and Risk Evaluation program between 1995 and 2000 |
| Skytte ² | (2010) | Quant. | Medical Records | 306 BRCA positive women who were unaffected by cancer and has not had risk-reducing surgery and who were registered on the Danish Departments of Clinical Genetics and the Danish National Pathology Registry databases between 1998 and 2008 |
| Stuckey | (2010) | Quant. | Medical Records | 90 BRCA positive women (39 had undergone risk-reducing bilateral salpingo-oophorectomy, 13 had undergone risk-reducing mastectomy and 44 had not undergone a risk-reducing surgery) evaluated in the Cancer Risk Assessment and Prevention Program between 1998 and 2006 at the Warren Alpert Medical School of Brown University (USA) |
| Swisher | (2001) | Qual. | Semi-structured telephone interview | 60 women who had undergone prophylactic oophorectomy (n=30) or ovarian cancer surveillance (n=30) at Washington University |

Table 3.2 cont. - Publications included in final review

| First Author | Year | Study | Mode of data collection | Study population |
|-----------------------|-------------|--------------|--|--|
| Tiller ^{1,2} | (2005) | Quant. | Self-administered structured questionnaire | 129 women at potentially high risk of ovarian cancer who had discussed ovarian cancer screening and/or prevention at one of 6 Familial Cancer Clinics in New South Wales / Victoria (Australia) |
| Tiller ³ | (2002) | Quant. | Self-administered structured questionnaire | 68 women from breast/ovarian cancer families who attended one of 14 Familial Cancer Clinics in New South Wales / Victoria / South Australia / Queensland / Western Australia (Australia) between 1996 and 1999 |
| Uyei ² | (2006) | Quant. | Medical Records | 554 women who had undergone testing for a BRCA mutation at the University of Texas MD Anderson Cancer Centre between 2001 and 2005 of whom 78 were BRCA1 positive, 54 were BRCA2 positive and 410 were BRCA negative |
| Verhoeven | (2011) | Quant. | Structured questionnaire | 119 pre-symptomatic individuals (includes 43 males) recruited from the Medical Genetics Centre at the University Ziekenhuis Gent (Belgium) of whom 54 were BRCA positive and 65 were BRCA negative |
| Wagner ⁴ | (2000) | Quant. | Structured self-administered questionnaire | 90 BRCA positive individuals (includes 2 males) from 35 families with a known BRCA mutation; recruited through the genetic counselling service of the Division of Senology at the University of Vienna (Austria) |

3.3.1 *Medical and physical issues*

The main medical and physical issues explored in a number of studies were mutation status, prior cancer diagnosis, cancer risk reduction, surgical menopause, HRT and the risks of surgery (Table 3.3).

Mutation status: A number of studies considered knowledge of BRCA mutation status as a factor during RRSO decision making (Antill et al. 2006, Culver et al. 2011, Kram et al. 2006, Madalinska et al. 2005, Schwartz et al. 2003, Uyei et al. 2006, Verhoeven et al. 2011). Ray et al. (2005) showed that women felt that genetic testing was the most influential factor in their decision. Phillips et al. (2006) found that women who are aware of their positive mutation status were seven times more likely to undergo screening or risk reducing surgery than women who did not know their mutation status. Similarly, Meijers-Heijboer (2003) found that receipt of positive genetic test results increased the likelihood that women would consider RRSO as an option. Conversely, negative test results did not reduce the number of women considering surgery in this study. The authors suggest that women may be aware that a negative test result does not necessarily indicate the absence of a mutation, as there are mutations which cannot be identified with currently available methods. Alternatively, women may disbelieve negative results based on their strong family history and perceived risk (Dillard et al. 2006). It was frequently found that uptake of surgery varied between women who carry mutations in BRCA1 and BRCA2, with the former more likely to undergo surgery (Evans et al. 2009b, Metcalfe et al. 2008b, Schwartz et al. 2003). This may reflect the differential risk profiles, as carriers of mutations in BRCA1 have a higher risk of ovarian cancer than those with mutations in BRCA2. However, others did not confirm these findings (Skytte et al. 2010, Stuckey et al. 2010). Participation in screening was also associated with BRCA mutation carrier status (Schwartz et al. 2003). However, Lerman et al. (2000) found that a positive BRCA test did not lead to increased screening adherence nor higher uptake of risk-reducing surgery.

Table 3.3 - Issues associated with/considered during decision making about RRSO

| |
|--|
| <p>Medical and physical:</p> <ul style="list-style-type: none"> Mutation status Previous cancer diagnosis Cancer risk reduction Issues with surgical menopause and HRT Surgical complications Benign gynaecological issues Previous positive screening results Residual cancer risk Sexuality / loss of libido Recovery time from surgery Physical discomfort Cessation of menstruation |
| <p>Psychological and emotional:</p> <ul style="list-style-type: none"> Perceived cancer risk Cancer-related distress/worry/anxiety Beliefs in screening / detection effectiveness Family planning / fertility Perception of femininity Issues of female identity Dislike of screening methods / wish to discontinue screening Beliefs in effectiveness of surgery Attitude towards surgical procedures Fear of death Wish to reduce uncertainty Coping style General self-efficacy |
| <p>Social and family context:</p> <ul style="list-style-type: none"> Personal experiences and family history Closeness / role in relation to cancer sufferers Family obligations Sexual relationships Family communication Partner's coping style |
| <p>Demographic:</p> <ul style="list-style-type: none"> Age Parity Income Education Employment Marital status |
| <p>Other:</p> <ul style="list-style-type: none"> Physician recommendation Readiness of healthcare system Social support Effects on insurance Incidental benefits Avoidance of surgery when healthy Inconvenience of attending the clinic Possibility for alternative therapy |

Cancer diagnosis: A previous diagnosis of cancer, especially breast cancer, was associated with decisions to undergo RRSO in several studies (Beattie et al. 2009, Hamilton et al. 2009, Madalinska et al. 2005, Metcalfe et al. 2008a, Stuckey et al. 2010). One study found that the stage of breast cancer at diagnosis was influential, with early stage cancer patients opting for RRSO more frequently (Meijers-Heijboer et al. 2003). Another study found a higher rate of uptake of RRSO among affected index patients compared to unaffected family members (Kwong et al. 2010a). However, others did not find such a relationship (Tiller et al. 2005). Non-cancerous gynaecological problems were also associated with decisions to undergo RRSO in one study (Swisher et al. 2001).

Risk reduction: Several studies specifically mentioned ovarian cancer risk reduction as a contributing factor to decisions about RRSO (Claes et al. 2005, Fry et al. 2001, Hallowell 1998). One study found that reducing ovarian cancer morbidity and mortality were named as reasons by women when asked retrospectively why they had opted for RRSO (Kram et al. 2006). Reduction of breast cancer risk following RRSO was a factor considered especially important by women with prior breast cancer (Culver et al. 2011). In a hypothetical scenario, in which women at increased risk were asked to indicate their leaning towards different management options in the context of differing degrees of hypothetical risk (unknown, 10% and 60%), the number of women leaning towards surgery for risk reduction increased with higher risk (Tiller et al. 2005).

Menopause and HRT: In a number of studies issues with surgical menopause and HRT appeared to be essential considerations when making decisions about RRSO, particularly for pre-menopausal women. Often mentioned as a drawback or 'cost' of surgery, these issues were discussed by many women in qualitative studies (Brain et al. 2004, Fang et al. 2002, Hallowell 1998, Swisher et al. 2001). Ray et al. (2005) found that one of the reasons women remained undecided about RRSO was uncertainty about the menopause. Some perceived the menopause to be an important natural event that should not be induced surgically (Hallowell 1998). Many women also had concerns about HRT and its effects on their body, as well as

its potential link to increased risk of breast cancer (Fang et al. 2002, Hallowell et al. 2001, Swisher et al. 2001).

Risk of surgery: The potential for surgical complications was explored in several studies (Brain et al. 2004, Hallowell 1998, Hurley et al. 2001). Women considered risks associated with surgery as well as recovery time after surgery, both of which could impact on their final decision (Brain et al. 2004, Fang et al. 2003, Fry et al. 2001). Some women chose to avoid surgery, as they felt it was an unnecessary risk (Claes et al. 2005). Similarly, fears and concerns in relation to surgical procedures influenced some women in their decisions (Hallowell et al. 2001, Ray et al. 2005).

Other, less frequently mentioned medical or physical considerations during deliberations about RRSO also emerged from previous studies. Residual cancer risk, not only in terms of the low remaining risk of primary peritoneal cancer after RRSO, but also in terms of the risk of cancer developing elsewhere, was also an issue brought up by women. This deterred some women from considering surgery, as they saw cancer as an inevitable and uncontrollable event, which would simply occur elsewhere in the body if they removed one organ (Hallowell 1998). In contrast, abnormal screening results and discomfort during screening led women to revisit surgery as an option (Howard et al. 2010c). The effects of surgery on sexuality and sexual experience (Babb et al. 2002, Klitzman and Chung 2009), including the loss of libido and cessation of menstruation (Fry et al. 2001), and the effects of surgery, such as recovery time (Fry et al. 2001, Swisher et al. 2001) and physical discomfort (Swisher et al. 2001), were also considerations identified as playing a role in some women's decision making.

3.3.2 *Psychological and emotional issues*

Psychological and emotional issues were frequently explored in studies of decisions about RRSO. These included perceived cancer risk, cancer-related anxiety or worry, beliefs in the effectiveness (or otherwise) of screening and factors relating to a desire to have children and preservation of fertility.

Perceived cancer risk: Perceived or subjective risk refers to the understanding of a patient about their own risk and is often independent of objective or actual risk. Women at increased familial risk often tend to overestimate their personal risk or the reduction in risk achieved by RRSO (Finch et al. 2009, Lerman et al. 1995). Some women thought that their cancer risk was fully eliminated following surgery (Finch et al. 2009). Perceptions of ovarian cancer risk and risk reduction following surgery were frequently found to play a role in many women's decision making processes (Babb et al. 2002, Brain et al. 2004, Hallowell et al. 2001, Hamilton et al. 2009). Women who perceived their risk to be greater opted for surgery more frequently (Claes et al. 2005, Fang et al. 2003, Kram et al. 2006, Madalinska et al. 2007, Schwartz et al. 2003) and reported higher interest in, and intention to undergo, surgery (Hurley et al. 2001, Meiser et al. 2003). Morris et al. (2001) found that of eight women who were not considered to be at high risk after evaluation of their family history, four still chose to have prophylactic surgery due to a high perceived cancer risk.

Cancer worry/anxiety: Several studies found that cancer-related worry, distress, fear and anxiety also influence decisions about RRSO (Culver et al. 2011, Fry et al. 2001, Hallowell 1998, Hamilton et al. 2009, Howard et al. 2011). Some studies found that higher levels of cancer worry, anxiety and fear were significantly associated with decisions to undergo RRSO (Madalinska et al. 2007, Meiser et al. 1999, Schwartz et al. 2003). However, others did not find such a relationship (Antill et al. 2006, Botkin et al. 2003, Meiser et al. 2003). Retrospectively, women in a questionnaire study stated that anxiety reduction was influential in their decision making process (Kram et al. 2006) and many women noted that relief from worry and fear was an important factor (Culver et al. 2011, Fang et al. 2003, Fry et al. 2001). One study found that intrusive thoughts about ovarian cancer were positively correlated with interest in surgery, and that anxiety and uncertainty reduction were the strongest predictors of interest in RRSO (Hurley et al. 2001).

Beliefs about ovarian cancer screening: Several studies found that beliefs in the effectiveness of screening and women's perceived ability to detect symptoms

themselves were influential in the decision making process (Brain et al. 2004, Fang et al. 2002). Women who doubted the efficacy of screening were more likely to select surgery (Claes et al. 2005, Fang et al. 2003, Kram et al. 2006), whereas those who believed that screening would detect cancer early were more likely to choose surveillance (Howard et al. 2011, Madalinska et al. 2007). Similarly, those women who felt that ovarian cancer could be cured easily tended to prefer screening over surgery (Madalinska et al. 2007).

Family planning and fertility: In a number of studies the desire for children, or more children, was explored as a potential issue considered by women who had been offered RRSO (Culver et al. 2011, Skytte et al. 2010). Infertility following surgery can be particularly burdensome for pre-menopausal women and the desire for children, or more children, is usually associated with deferral or rejection of surgery (Fang et al. 2002, Fang et al. 2003). However, even women who have completed childbearing may be concerned about the loss of fertility following surgery (Swisher et al. 2001).

Femininity and female identity: Linked to the concept of fertility are the expected effects that loss of fertility will have on femininity and female identity (Babb et al. 2002, Hallowell 1998, Wagner et al. 2000). In one study women saw their fertility as an important part of 'being a woman' and voiced concerns over having their ovaries removed, fearing the loss of their female identity following surgery (Hallowell 1998).

Other, less frequently explored psychological and emotional factors included dislike of screening methods (Fry et al. 2001) and beliefs in the effectiveness and benefits of surgery (Hurley et al. 2001, Madalinska et al. 2007), both of which could result in an increased interest in RRSO. Similarly, a reduction in uncertainty was seen as a potential benefit of surgery by some women (Fang et al. 2002). Attitudes towards surgical procedures (Hallowell et al. 2001, Wagner et al. 2000) and fear of cancer and/or death (Babb et al. 2002, Swisher et al. 2001) were also found to influence women's decisions. Fear of surgical procedures was found

to lead to deferral or complete rejection of surgery (Hallowell et al. 2001, Wagner et al. 2000), whereas the wish to reduce the fear of cancer and/or death was found to be linked to decisions to undergo RRSO (Babb et al. 2002). Women's, and their partner's, preferred coping style and general self-efficacy, i.e. belief in one's competence, also played a role in decision making (Mellon et al. 2009), particularly with regards to seeking information. Women who showed higher self-efficacy and who preferred a high monitoring coping style were more likely to actively seek information about cancer risk and management options (Mellon et al. 2009, Wilson and Gilbert 2005).

3.3.3 *Social context issues*

The main social context issues addressed in studies were family history and familial obligations. However a few other issues, including parity, sexual relationships and family communication, and their impact on decisions about RRSO, were also discussed.

Family history: Several studies explored family history, both in terms of its medical and social aspects (Bradbury et al. 2008, Friebel et al. 2007, Uyei et al. 2006). Family history influenced not only the decision itself, but also the way women approached decisions about RRSO (Hamilton et al. 2009, Mellon et al. 2009). All women who had opted for surgery in a retrospective study by Swisher et al. (2001) mentioned family history as a reason for their decision, while some of the women in this study who had opted for surveillance did not. However, others did not find family history to be predictive of choice to undergo RRSO (Antill et al. 2006, Botkin et al. 2003, Scheuer et al. 2002). Personal experience of ovarian cancer in close relatives and the role women played in the life of cancer sufferers also influenced their decisions about RRSO (Babb et al. 2002, Hallowell et al. 2001, Howard et al. 2010c). One study found that women whose mother or sister had been diagnosed with ovarian cancer were more likely to opt for surgery than those who did not have a first degree relative with ovarian cancer (Metcalfe et al. 2008b); however others did not find such a relationship (Tiller et al. 2005).

Familial obligations: Studies also explored family and social obligations as factors influencing RRSO decision making (Bradbury et al. 2008, Brain et al. 2004, Howard et al. 2010c). Similar to age (see section 3.3.4), family obligations could be associated with decisions to undergo or decline surgery (Brain et al. 2004, Hallowell 1998, Hallowell et al. 2001). Some women argued that they wanted to undergo RRSO to be able to be there for their family in the long-term, while others deferred surgery as they felt that they could not cease to fulfil their familial obligations in the short-term. Additionally, the potential effects of surgery on sexual relationships were perceived by women to be a 'cost' of surgery that may put a strain on their relationship with their partner (Hallowell 1998).

A few other social context influences were discussed in previous studies. Family/partner communication and partner's coping style were found to influence women's information seeking behaviour (Mellon et al. 2009). Women who reported good family communication and who had partners displaying a high monitoring coping style were more active in seeking information about cancer risk and management options (Mellon et al. 2009, Wilson and Gilbert 2005).

3.3.4 Demographic factors

Age: Several studies explored the influence of age on decision outcomes (Brain et al. 2004, Fry et al. 2001, Meiser et al. 1999, Skytte et al. 2010, Tiller et al. 2002). Age was associated with decisions both for and against RRSO (Claes et al. 2005). Older age was frequently linked to decisions to undergo RRSO (Bradbury et al. 2008, Brain et al. 2004, Fang et al. 2003, Madalinska et al. 2007, Phillips et al. 2006, Scheuer et al. 2002, Schmeler et al. 2006b). However, it has also been shown that from age 60, uptake of RRSO decreased as women were more concerned about their daughters' risks than their own (Beattie et al. 2009).

Parity: Some studies found that parity was associated with election of risk-reducing surgery (Schmeler et al. 2006b, Stuckey et al. 2010) and one study found that having two or more children was predictive of uptake of RRSO (Skytte et al.

2010). However, others did not find parity or the number of co-habiting children to be predictive of RRSO uptake (Antill et al. 2006, Tiller et al. 2002).

Additional demographic factors assessed in several studies included marital status (Fang et al. 2002, Madalinska et al. 2007), education (Fang et al. 2002, Madalinska et al. 2005, Meiser et al. 2003), income (Antill et al. 2006, Botkin et al. 2003) and employment status (Antill et al. 2006). Most studies found that these variables did not influence the decision of whether or not to undergo RRSO (Antill et al. 2006, Botkin et al. 2003, Fang et al. 2002). One study concluded that being married and less educated were predictive of uptake of RRSO (Madalinska et al. 2007).

3.3.5 *Other issues*

A few issues that did not fall within the four previous categories were also considered during, or associated with, decision making about RRSO.

Physician recommendation: The recommendation from a physician for RRSO was an influential factor, usually associated with uptake of RRSO. This indicates that women valued their physician's opinion (Claes et al. 2005, Kram et al. 2006, Swisher et al. 2001). Others found that a physician's recommendation against RRSO was similarly influential (Fang et al. 2002).

A number of other, less frequently explored considerations included the availability of social support (Mellon et al. 2009) and the readiness of the healthcare system, i.e. perceptions of when the healthcare system could accommodate women and when referrals could be made (Howard et al. 2010c), both of which could influence women's decisions about RRSO. Incidental benefits of surgery, such as the resolution of other gynaecological problems (Hallowell 1998), and a parallel decision to undergo prophylactic mastectomy (Beattie et al. 2009, Madalinska et al. 2005) were associated with more positive attitudes towards RRSO and/or undergoing this surgery. In contrast, avoiding surgery when it was not necessary was found to be a motive to opt for screening rather than RRSO (Claes et al. 2005). The potential of RRSO to negatively affect future insurance contributions

was considered by women living in countries where health provision is regulated through insurance, such as the US (Hurley et al. 2001). A few women also felt that inconvenience of attending the clinic (Fry et al. 2001) or availability of information about alternative therapies (Klitzman and Chung 2009) were important factors in their decision making.

3.3.6 Women's expectations of decision support

Eight studies described what women expected or desired in terms of support during deliberations about RRSO (Babb et al. 2002, Campfield Bonadies et al. 2011, Hallowell 2000, Howard 2010, Ray et al. 2005, Swisher et al. 2001, Tiller et al. 2005), a summary of which is shown in Table 3.4.

Table 3.4 - Women's expectations of decision support content

| |
|--|
| <p>Information support:</p> <ul style="list-style-type: none"> Ovarian function and the menopause Personal medical history Cancer family history Cancer risk assessment / mutation status Ovarian cancer screening RRSO (procedure, recovery time etc.) Ovarian and breast cancer risk-reduction after surgery Effectiveness of surgery Residual cancer risk Surgical risks HRT Timing of surgery Types of surgery Effects of surgery (on sexuality and menopause) Effects of surgery (on heart disease and osteoporosis) Possible adjunctive surgery |
| <p>Psychological and emotional support:</p> <ul style="list-style-type: none"> Control, empowerment Future hope (alleviate fear of death) Strategies for coping with the consequences of surgery Availability of sex counselling Reduction in uncertainty |
| <p>Social support:</p> <ul style="list-style-type: none"> Availability of social support Building trust Effective communication (from doctors, friends, family) |

Unsurprisingly, women's expectations in terms of information provision to support the decision mapped well onto the issues explored by women during deliberations as discussed in previous sections. Most importantly, women wanted accessible, comprehensive information materials, which covered the areas of risk and potential mutation status, as well as management options, such as RRSO or screening (Babb et al. 2002, Klitzman and Chung 2009, Ray et al. 2005). The information required with regards to surgery included details of the procedure, recovery time after surgery, risks and effectiveness, as well as effects concerning fertility, sexuality and menopause (Babb et al. 2002, Campfield Bonadies et al. 2011, Hallowell 2000). The vast majority of women in one study indicated that they wanted as much information as possible, including good and bad outcomes of management options (Tiller et al. 2005). Background information on ovarian function, the menopause and potential use of HRT were also areas where information was requested (Hallowell 2000, Swisher et al. 2001). One study found that women desired information about insurance issues regarding both genetic testing and cost of surgery (Ray et al. 2005). Women reporting retrospectively on information they would have liked to have had before having surgery mentioned that effects of surgery on sexuality and the availability of sex counselling had been neglected in pre-surgery consultations (Campfield Bonadies et al. 2011). More than 40 per cent of women in this study also felt that further pre-surgery information about changes in heart disease, osteoporosis risk and body image would have been helpful. The use of HRT and its effects on breast cancer risk, which women were concerned about, was also a topic seen as important by women post-surgery (Swisher et al. 2001).

In addition to information content, women also desired psychological and emotional support, wanted to feel in control and to build trust with medical professionals (Babb et al. 2002). Women expected effective communication between themselves and their clinician, as well as family and friends, which to them was vital to establish rapport and an appropriate support network (Babb et al. 2002). Women also desired information about, or help with, strategies to deal with

the consequences of surgery and to support coping efforts (Howard 2010). Although most of what women expected or desired in terms of support during deliberations about RRSO referred to specific information materials, some women also voiced additional desires that might be catered for with carefully designed decision support. These include stimulating effective communication between the patient, health professionals and family, enhancing patient empowerment and sense of control, and offering coping advice.

3.4 Discussion

3.4.1 Discussion of findings

This chapter reviewed the issues considered during, and associated with, decisions about RRSO in women at increased risk of ovarian cancer. Unsurprisingly, the number of issues identified was relatively large, thus exposing the complexity of this decision and the challenges women face whilst going through the appraisal and deliberation process. By far the largest proportion of issues fell within the 'medical/physical' and 'psychological/emotional' categories.

3.4.1i Medical and physical issues: Personal medical history, such as knowledge of a positive mutation status (particularly BRCA1), prior cancer diagnosis or a benign gynaecological problem, was often found to be associated with decisions to undergo surgery (Beattie et al. 2009, Evans et al. 2009b, Hamilton et al. 2009, Kwong et al. 2010a, Madalinska et al. 2005, Manchanda et al. 2012a, Meijers-Heijboer et al. 2003, Metcalfe 2009, Metcalfe et al. 2008b, Phillips et al. 2006, Stuckey et al. 2010, Swisher et al. 2001). Hence, the finding that women reported wanting information about their risk and mutation status to facilitate decisions about RRSO was not surprising (Babb et al. 2002, Klitzman and Chung 2009, Ray et al. 2005). Dagan and Goldblatt (2009) confirmed that knowledge of risk, especially of a confirmed genetic mutation, was perceived by women as empowering. These findings suggest that women who are aware of their risk due to a confirmed mutation understand themselves as 'patients' and feel as though they have a 'diagnosis' upon which they can act (Scott et al. 2005). Hence, they may perceive

more benefits to surgery compared to those who do not know their mutation status and therefore may not have an exact risk estimate on which to base their decision. However, one study did not find a correlation between positive BRCA status and surgery uptake (Lerman et al. 2000). This study had a short follow-up time of one year post-genetic test disclosure. In contrast, Phillips et al. (2006) found that confirmation of a BRCA mutation did lead to increased uptake of surgery with a follow-up time of three years. Therefore time elapsed from point of disclosure may influence women's risk management choices at time of assessment and results should be interpreted with this in mind. Differences in uptake of RRSO between carriers of mutations in BRCA1 and BRCA2 may be explained by the differing ovarian cancer risk estimates, which are higher for carriers of mutations in BRCA1 than for those with mutations in BRCA2 (Chen and Parmigiani 2007). Therefore increased uptake by women with BRCA1 mutations, as reported in many studies, may reflect a realistic estimation of personal risk. Studies which did not find a difference in uptake combined RRSO and risk-reducing mastectomy in their analysis (Stuckey et al. 2010) or were conducted in a country with very high (>75%) uptake of surgery amongst carriers of mutations in BRCA1/2 (Skytte et al. 2010).

It should be noted that studies regarding positive mutation status and uptake of RRSO only assessed BRCA carrier status. The influence of Lynch Syndrome mutation status on decisions regarding RRSO was not explored by studies in this review. This may be due to the increased risk of endometrial and colorectal cancer associated with Lynch Syndrome, which may be of primary concern for most affected women. RRSO in this context may mainly be considered as an adjunct to risk-reducing hysterectomy (RRH) for endometrial cancer risk reduction, rather than a stand-alone procedure. It may also be due to lower rates of positive mutation tests in women from families with Lynch Syndrome, as many mutations are not detectable using currently available methods and risk reduction decisions are often based solely on family history.

Those women who had already experienced cancer themselves, and those who viewed RRSO as a solution to other gynaecological issues (as well as a means to

reduce cancer risk), may be more likely to opt for RRSO (Manchanda et al. 2012a). These patients may perceive there to be more benefits to the operation than those who have not been affected by cancer or suffered from benign gynaecological problems (Kwong et al. 2010a, Meijers-Heijboer et al. 2003, Stuckey et al. 2010, Swisher et al. 2001).

Effects of surgery on cancer risk were specifically mentioned by relatively few studies as an issue explored during decision making about RRSO (Claes et al. 2005, Culver et al. 2011, Fry et al. 2001, Hallowell 1998, Kram et al. 2006); however this issue is arguably the main reason why RRSO would be considered in the first place and is likely to play a significant role in most decisions although it was not explored by all studies. Both, the reduction in ovarian and breast cancer risk may be important for women, as many come from HBOC families and face a substantial risk of breast cancer in addition to their elevated risk of ovarian cancer.

Physical effects of surgery, particularly in relation to surgical menopause and the potential need for HRT, were frequently found to influence decisions about RRSO (Brain et al. 2004, Fang et al. 2002, Hallowell 1998, Hallowell et al. 2001, Ray et al. 2005, Swisher et al. 2001). These findings are unsurprising, as infertility and surgical menopause are irreversible and often seen as the main drawbacks to surgery by pre-menopausal women. Menopausal symptoms, which may be more severe following surgery when compared to the natural menopause, can affect women's quality of life (Elit et al. 2001, Hallowell et al. 2012). Fears of, and concerns about, the short- and long-term effects of surgical menopause are therefore an important factor influencing decisions about RRSO (Hallowell 1998, Ray et al. 2005). While HRT can relieve some of the symptoms of the menopause, women are often reluctant to take HRT due to concerns about the effects of HRT on breast cancer sparked primarily by the media coverage of a study on post-menopausal women (Beral and Collaborators 2003, Fang et al. 2002, Rossouw et al. 2002, Swisher et al. 2001). It must be noted that findings from the study which caused these concerns have now been widely discredited (Shapiro et al. 2011a, b, Shapiro et al. 2011c, Shapiro et al. 2012) and it has been shown that HRT is safe for

the vast majority of women with a family history, if it is only taken until the age of natural menopause (Eisen et al. 2008, Rebbeck et al. 2005). Therefore, it is essential that women are provided with up-to-date information regarding surgical menopause and HRT when making decisions about RRSO, especially in light of their self-reported information needs (Hallowell 2000, Swisher et al. 2001).

Similar to concerns about the surgical menopause and HRT, fear of and concerns about risks of surgery were also found to influence women's decision making (Claes et al. 2005, Hallowell et al. 2001, Ray et al. 2005). Any surgery bears risks and these may be perceived as higher by individuals considering risk-reducing surgery, i.e. surgery in the absence of clear clinical findings, as opposed to surgery as a treatment option for a current health problem. It has been previously reported that women see their decision as a weighing up of the 'dominant fear', either fear of cancer or fear of the risks of surgery (Dagan and Goldblatt 2009). Therefore it is unsurprising that surgical risks were explored in a number of studies and that women themselves also reported a need for information about risks and effectiveness of surgery (Babb et al. 2002, Campfield Bonadies et al. 2011, Hallowell 2000, Howard 2010, Tiller et al. 2005).

3.4.1ii Psychological and emotional issues: Perceived cancer risk can influence women's decision making regarding RRSO dramatically, even though it may be discrepant from their objective risk estimate (Cull et al. 2001, Mellon et al. 2008). Many studies found a high perceived risk to be associated with intention to undergo, or actual uptake of, RRSO (Claes et al. 2005, Fang et al. 2003, Kram et al. 2006, Madalinska et al. 2007, Schwartz et al. 2003). This may indicate that the emotional burden of a high perceived risk can be enough to persuade women to select surgery, despite a relatively low actual risk. This is supported by the fact that some women who have been found to be at or near population risk based on their family history still choose to undergo RRSO (Morris et al. 2001). Previous findings also suggest that emotional responses to test results are influenced by pre-existing concepts about risk and that some individuals appear to 'distrust' a negative

genetic test result (Dillard et al. 2006, Hilgart et al. 2010, Lerman et al. 1995, Michie et al. 1996).

Higher perceived risk tends to co-exist with higher cancer worry and can be resistant to change despite a low risk assessment (Brain et al. 2011). Relief from this worry or anxiety has been found to motivate women to undergo surgery (Culver et al. 2011, Fang et al. 2003, Fry et al. 2001, Madalinska et al. 2007, Meiser et al. 1999, Schwartz et al. 2003) and confirms that emotional cues, such as distress and anxiety, play a vital role in decision making processes, which may be geared towards regulating these emotions. However, a number of studies did not agree with these findings (Antill et al. 2006, Botkin et al. 2003, Meiser et al. 2003). Such contradictory findings may be explained by different measures used to assess anxiety and distress, or by the varied follow-up times in the different studies. Additionally, two of these studies were evaluating factors predictive of RRSO uptake; therefore they were not simply considering issues that may be important in the decision (Antill et al. 2006, Botkin et al. 2003). Anxiety and worry may still be an important issue in these decisions; however it may not be predictive of RRSO uptake, as some anxious or worried women may opt for screening instead, if available.

Women's beliefs in the effectiveness of screening and their own ability to detect symptoms were also explored in several studies (Brain et al. 2004, Claes et al. 2005, Fang et al. 2002, Fang et al. 2003, Madalinska et al. 2007). Those who felt that screening is an effective means of detecting ovarian cancer or that they would be able to detect symptoms themselves were more likely to lean towards surveillance. In contrast, those who perceived screening efficacy or their ability to detect symptoms to be low showed more interest in surgery. Similarly, an unfulfilled wish to have a child or issues concerning fertility meant that women would defer or decline surgery (Culver et al. 2011, Fang et al. 2002, Fang et al. 2003, Skytte et al. 2010). Perhaps more surprising was the finding that the issue of fertility remained a consideration even for those women who had completed their family (Swisher et al. 2001). These data may point towards a more emotive role of fertility

in the decision making process, which may be linked to perceptions of femininity, womanhood and body image that may be affected by RRSO (Babb et al. 2002, Wagner et al. 2000).

3.4.1iii Social context issues: Family history was found to influence decisions about RRSO in terms of the number of affected relatives and types of cancers, as well as in terms of the closeness of affected members to women and the role women played in sufferers' lives (Bradbury et al. 2008, Friebel et al. 2007, Mellon et al. 2009, Uyei et al. 2006). It is likely that women draw on their own experiences of ovarian cancer in the family, and perhaps RRSO in other relatives, to facilitate affective forecasting when making decisions about this surgery (Wilson and Gilbert 2005). Therefore, it is unsurprising that the number, closeness and outcomes of relatives' ovarian cancers would influence their decisions. Dagan and Goldblatt (2009) found that cancer outcomes witnessed in the family, particularly their mothers' experience with ovarian cancer, influenced how women perceived their own ability to cope with this cancer, thus indirectly affecting choice of risk management. Studies looking at predictors of choices of risk management strategies, however, did not find family history to be predictive of RRSO uptake (Antill et al. 2006, Botkin et al. 2003, Scheuer et al. 2002). Similar to cancer worry, family history may play a role in decision making but may be equally predictive of choice of screening and surgery, therefore failing to be a significant predictor of one or the other in these studies.

Familial obligations also played a role in women's decision making about RRSO. Some women argued that their survival was key, so that they could fulfil their familial obligations in the long-term future, and therefore selected surgery. Others decided that the recovery time associated with surgery would mean that they would not be able to fulfil their familial obligations in the short-term, and therefore declined surgery (Brain et al. 2004, Hallowell 1998, Hallowell et al. 2001). Women in the literature often considered the impact surgery might have on their family, both in terms of obligations, such as taking care of their children or elderly and vulnerable relatives, as well as in terms of their relationship with their partner.

Women reported a need for more information on the effects of surgery on sexuality and the availability of sex counselling, indicating that these issues were important to them (Campfield Bonadies et al. 2011).

3.4.1iv Demographic factors: Older age was frequently found to be associated with increased uptake (Botkin et al. 2003, Bradbury et al. 2008, Brain et al. 2004, Fang et al. 2003, Manchanda et al. 2012a, Scheuer et al. 2002, Schmeler et al. 2006b). This is unsurprising as age has an effect on many of the issues discussed above, including cancer risk and menopausal status. Awareness that ovarian cancer risk rises with age may therefore lead to increased uptake of surgery (Lifford et al. 2012). Additionally, the impact of RRSO on the life of older patients, particularly those who have completed childbearing and natural menopause, is significantly reduced, therefore potentially reducing the negative effects associated with surgery. Age may also play a role in women's decision making with regards to their mother's age at diagnosis or death, which may be seen as an important point to reach in their own lives (Dagan and Goldblatt 2009).

Parity was also found by several studies to be associated with uptake of surgery (Schmeler et al. 2006b, Skytte et al. 2010, Stuckey et al. 2010). This finding may be explained by women's sense of responsibility towards their children and a wish of wanting to avoid their children growing up without their mother or seeing their mother go through cancer treatment.

3.4.1v Other issues: Unsurprisingly, a recommendation for or against RRSO from a physician was also found to influence women's decisions (Claes et al. 2005, Fang et al. 2002, Kram et al. 2006, Swisher et al. 2001). It is logical that women would take into account or simply follow their physician's recommendation, as most lack medical expertise and may be used to a paternalistic approach to decision making about their health, readily accepting a plan of action from their physician. However, due to the preference-sensitive nature of this decision it may not be productive for a physician to make a recommendation early on in the decision making process. Rather it may be preferable to make the patient aware of her

options, including the risks and benefits, and to help her clarify her values with regards to RRSO. If the patient then still wishes the physician to make a recommendation, this should be in line with the patient's values.

The literature search found a large number of other factors that were considered during, or associated with, decisions about RRSO. These included residual cancer risk, reduction in uncertainty and the availability of social support. Most of these factors are considerations that will be explored with, or by, most patients when they go through the appraisal and deliberation process. Additionally, this study explored what women expected in terms of support during decision making and found that women reported a need for detailed information materials as well as for psychological support, empowerment, effective communication and coping advice (Babb et al. 2002, Howard 2010). The variety of factors contributing to appraisal and deliberation processes, and the support needs uncovered here and in previous reviews, highlight the complexity of decisions about RRSO and the need for additional decision support to augment that which is currently provided in clinical settings (De Leeuw et al. 2008, Fang et al. 2005, Howard et al. 2009a, Miller et al. 2010).

3.4.2 Results in context

Decisions about RRSO cannot be seen as isolated decisions. They are being made before, in parallel to or after other decisions, which are of similar complexity. These include decisions about genetic testing, other risk-reducing strategies, such as risk-reducing mastectomy or risk-reducing hysterectomy, the use of HRT and decisions about disclosure to others, such as partners, children and other family members. Another additional pressure on patients at the time of decision making about their own risk management strategies may be the need for disclosure of that risk, or even a positive mutation status, to other family members and the worry of the impact of their risk on close relatives, such as their children (Hopwood and Watson 2010).

Therefore, decisions about RRSO are often being made under conditions of high worry and low comprehension (Fang et al. 2005). It has been suggested that under these circumstances patients may not comprehend the full scope of the information presented to them and hence may be making decisions that are not fully informed and based on gist rather than verbatim memories of exact risk and effectiveness of options (Lloyd et al. 2001, Reyna and Hamilton 2001). Findings that perceived cancer risk and anxiety influence RRSO decision making support this suggestion (Claes et al. 2005, Hurley et al. 2001, Madalinska et al. 2007). Even after discussing management options at a familial cancer clinic, women were found to have a number of misconceptions about the available risk-reduction strategies and their effectiveness (Tiller et al. 2005). Evidence-based decision support interventions may be helpful in supporting patients' accurate understanding of their risk and have been shown to improve knowledge and alignment of decisions with personal preferences (Stacey et al. 2011). Furthermore, these materials can be designed for use in the patient's own home and may be shown to other members of the family, including husbands, sisters or mothers, thereby facilitating disclosure and discussions within the family about risk and potential management options.

Women make decisions about RRSO in the context of their everyday lives and consider a range of issues to do with their personal and familial circumstances, such as their age and menopausal status as well as their family and work obligations and the impact surgery might have on these. Women not only make a decision about whether or not to have the surgery, they also deliberate about when to have the surgery. Perceptions of the right time to go ahead with surgery can vary and depend on considerations such as life plans (for example, completion of family), resolution of conflicts with self and others, readiness of the healthcare system and cognitive and emotional readiness of the woman herself (Howard et al. 2010c, Lifford et al. 2012). Therefore, women may defer the decision initially and deliberations may take several years. Decision support materials, which can be accessed by women outside the clinical context, could be useful here. These allow women to come back to this decision and review the information when they are

ready before making an appointment with a physician or genetic counsellor, which could be particularly useful as women may not be seen on a regular basis at genetics centres to follow up on their decision making (Howard et al. 2010c, Tiller et al. 2003, Wonderling et al. 2001).

In order to develop adequate support materials for these women it is essential to understand the issues that influence women's decisions and what they desire in terms of information and support. The current review explored these issues and the results presented here may be helpful as a guide when developing decision support materials for these women. In Chapter 5, these results will be applied to the generic version of the CODE framework to create an RRSO-adapted version. This will then be used to develop a decision support intervention tailored to the needs of women at increased genetic risk of ovarian cancer (Chapter 7).

3.4.3 Strengths and limitations

This review offers a detailed analysis of the factors potentially considered by women during deliberations about RRSO and additionally explores what women expect and desire in terms of support during decision making. It includes a number of qualitative and quantitative research studies and therefore provides a more holistic view of issues important in the decision making process than previous reviews of an exclusively or mainly quantitative nature (De Leeuw et al. 2008, Miller et al. 2010). Furthermore, to be as inclusive as possible, the review was not limited to one specific category, such as medical or psychological issues, as others have done before (Fang et al. 2005).

A few limitations to this review should be noted. Firstly, as with any review, the quality of the studies which were included determines the strength of the conclusions that can be drawn. As this review aimed to be as inclusive as possible, the design and quality of studies varied considerably. Although all studies were critically appraised before inclusion, the differing study populations, varying sample sizes, wide range of measures, follow-up times and modes of data collection used should be noted. However, it is reassuring that the same or similar issues were

often explored in a number of different studies, thus demonstrating some consistency, despite the range of studies included, and lending credibility to the findings.

Secondly, factors found not to be statistically significant or predictive of a certain decision were not excluded. Arguably, this could lead to over-emphasis of factors that do not influence the decision either way or of issues that are only considered by a minority of patients. However, as this review was seen as the basis for further research into the issues considered by a specific cohort of patients – women at increased risk of ovarian cancer in the UK – such factors should not be dismissed at this point.

Thirdly, this review brought together the findings of studies from a number of different countries, including the US (Schmeler et al. 2006b), Canada (Howard et al. 2010c), the UK (Brain et al. 2004), the Netherlands (Meijers-Heijboer et al. 2003), Denmark (Skytte et al. 2010), Australia (Hallowell 1998) and China (Kwong et al. 2010a). Intra- and inter-country differences related to issues considered by women making decisions about risk-reducing surgery, and discrepancies in rates of uptake of such strategies, have been identified in previous studies (Julian-Reynier et al. 2000, Metcalfe et al. 2008a, Metcalfe et al. 2008c). These differences may be due to discrepant cultural norms or differences in healthcare systems (Metcalfe et al. 2008a). Countries such as Norway or the USA see very high rates of uptake, around 70%, while others, such as Poland and Italy, report much lower rates, around 50% or below (Metcalfe et al. 2008a). This could indicate that the general attitude towards risk-reducing surgery may vary between populations. Furthermore, due to differences in healthcare systems, women in the US considered insurance issues an important factor during decision making (Hurley et al. 2001), whereas in countries with state funded healthcare, such as the UK, this is not an issue women usually take into account.

Additionally to covering a number of countries, the studies in this review included a variety of patient populations, ranging from unaffected women with only

a family history to women with a diagnosis of breast cancer and confirmed genetic mutation. These differing populations may consider different issues during deliberations about RRSO. The results of this study therefore are not directly transferable to any specific at-risk population, such as women with a family history in the UK. Further research is needed to determine which of the issues identified here are of importance to women at increased risk of ovarian cancer in the UK, particularly if these may be used to guide the content of genetic counselling sessions and decision support materials for this patient cohort. However, the current review does provide a list of potential issues that could be important during deliberations about RRSO on which to base future research and analysis of results from focus groups and interviews with specific populations.

Finally, the current review was limited to women's characteristics, intentions, emotions, thoughts and considerations. Studies of the views of healthcare providers or others involved in these decisions were not included and therefore no inference can be made about what providers regard as important considerations during these deliberations. However, it has previously been shown that providers consider age and menopausal status as well as medical history, screening and family history when making decisions about risk-reducing surgery (Plusquin et al. 2011, Stany et al. 2010). This maps well onto some of the medical and demographic considerations explored by studies of women's decisions.

3.4.4 Conclusions

This chapter reports on the currently available empirical evidence regarding issues considered during, and associated with, women's deliberations about RRSO. It also explored what women expect or desire in terms of support materials or interventions designed to facilitate appraisal and deliberation processes. The variety of issues reported in this chapter highlights the complexity of decisions about RRSO. It also provides evidence that there is scope for decision support materials to be developed for women at increased risk of ovarian cancer who are trying to make these decisions.

As the number of issues raised was relatively large, and as there are concerns about the applicability of these issues to specific populations, any inferences to the UK population should be made with great care. Therefore, before developing any support materials, it is necessary to specifically investigate the issues important to women who are making decisions about RRSO in a UK context (Chapter 4). Together with the findings of the literature search, these will then be used to adapt the CODE framework to describe deliberation and coping processes relating to RRSO decisions in women at increased risk of ovarian cancer in the UK (Witt et al. 2012). The adapted framework will then be used as a theoretical basis to facilitate the development of decision support materials specifically designed to help patients who are considering RRSO (Chapter 7).

4 Patients' and health professionals' perspectives on decisions about risk-reducing bilateral salpingo-oophorectomy in the UK

4.1 Introduction

The previous chapter demonstrated that the decision making process about RRSO in the context of cancer genetic risk is complex and involves a wide range of issues that women may consider (Chapter 3; De Leeuw et al. 2008, Fang et al. 2005, Howard et al. 2009a, Miller et al. 2010). These issues can include medical, psychological, demographic and social context factors, including gene mutation status, cancer worry, age and familial obligations. They may vary between different populations and between countries, depending on cultural attitudes and norms, as well as healthcare systems (Metcalf et al. 2008a).

In the UK, healthcare is provided through the National Health Service (NHS). The service is fully tax-funded and therefore provides healthcare free at the point of access to patients. Therefore, issues around cost of surgery and impact on insurance are less important in the UK than they may be elsewhere. However, private health insurance is also available in the UK and surgery may affect life insurance premiums. As previously described in Chapter 1, women with a family or personal history of cancer may be referred to a genetics service by their GP or oncologist, where the patient's family history will be assessed and a risk estimate based on this information will be provided (Wood et al. 2003). Women may then be offered appropriate screening or risk-reducing options. In the case of elevated risk of ovarian cancer, this includes risk-reducing bilateral salpingo-oophorectomy (RRSO) from age 35. A trial examining the efficacy of ovarian cancer screening in high risk women with a trans-vaginal ultrasound once a year and CA125 blood tests three times a year has only recently concluded and results are not expected until late 2013 (Institute for Women's Health 2010). Therefore, screening for ovarian cancer is not routinely available in the NHS; symptom awareness is recommended as an alternative (UK National Screening Committee 2006).

It is important to understand the risk management options available in the UK and women's decision making and coping processes when faced with these options.

This is an essential step towards developing decision support materials specifically aimed at these women and their health professionals. The aim of this chapter is (i) to explore issues associated with decisions about RRSO specifically for women in the UK and (ii) to create a model of the preference construction pathway for women making decisions about RRSO in the UK. The results presented here will be used to adapt the generic CODE framework to specifically describe decisions about RRSO by women at increased risk of ovarian cancer (Chapter 5).

4.2 Methodology

This study received approval from the Multi-Centre Ethics Committee for Wales (Ref: WA-11-0094) and Research and Development departments at all participating centres (Cardiff, Swansea and London) (Appendix 4.1).

4.2.1 Recruitment for focus groups

Focus groups are a method of qualitative data collection that allows the researcher to explore perceptions, opinions, beliefs and attitudes with participants using a relatively informal approach that may lead to more honest, in-depth answers than would be achieved in questionnaire studies (Adams and Cox 2008, Bloor et al. 2000, Krueger 1994, Morgan and Krueger 1997, Puchta and Potter 2004). Participants may also build on each other's answers and so explore topics that they may not have thought of in an individual interview (Slaughter et al. 1999). An additional advantage of focus groups is the possibility of asking the group to find a consensus (Stewart et al. 2006). This can then be used by the researcher to create a hierarchy of issues discussed during the meeting. In the context of the current study, focus groups were deemed appropriate as they allowed detailed discussion of women's views of decision making about RRSO, as well as exploration of the relative importance of the issues that influence their decisions. Furthermore, the focus group approach was suitable for examining participants' views of, and ideas for, a decision support intervention in the RRSO context (reported in Chapter 7).

Women were identified from the databases of three centres in Wales and England (the Cancer Genetics Service for Wales based at the University Hospital of

Wales, Cardiff, and Singleton Hospital, Swansea, and the Familial Cancer Clinic at University College London Hospital). Women were eligible if they were at moderate to high risk of ovarian cancer (estimated lifetime-risk $\geq 10\%$), aged 30 to 80, and if they had not yet made a decision about RRSO. Women who had already opted for RRSO were not eligible to take part in the focus groups. Seventy-seven eligible patients were identified and invited by health professionals, who provided them with an information pack, either directly after a consultation or by post. This number represents all eligible women registered with the three centres at the time of the study. Interested patients then contacted the researcher and were invited to attend a focus group. Full written consent was obtained on the day of the meeting. Focus groups were held in meeting rooms on the premises of the Cancer Genetics Service for Wales (Cardiff, Swansea) and the Institute for Women's Health (UCL, London).

4.2.2 *Focus groups*

During the focus group, participants were asked (i) to discuss the decision making pathway, exploring catalysts and barriers to the decision, and (ii) to share their ideas for the content and format of a patient decision support intervention (reported in Chapter 7). A topic guide for the group can be found in Appendix 4.2. Discussions were guided by a moderator (Jana Witt) and supported by a moderator aid. After discussing the decision pathway, participants were asked to rank the issues raised by placing prompt cards (compiled by the moderator aid) along a scale (from least important to most important) in order to gain an understanding of the relative importance of each issue. At the end of the meeting, the focus group participants were invited to become part of a 'virtual reference group' to give them an opportunity to remain involved in the study (see Chapter 7, section 7.2.1). Focus groups were convened between January and April 2012. Participant validation was carried out by sending women a short summary of the issues raised during the focus group discussion one week after the meeting, asking them whether they felt the summary was a fair reflection of the group's views.

4.2.3 *Interviews with health professionals*

Due to the busy schedule and geographical spread of health professionals in the field of genetics, convening a focus group was not a practical method of data collection for this group. Therefore, semi-structured interviews were deemed more appropriate to explore the opinions and views of health professionals (Kvale 1996). Semi-structured interviews ensure a certain set of topics is covered during the interview, but allow the researcher to deviate from the interview guide to further explore any issues raised (Friesen 2010, Harrell and Bradley 2009, King and Horrocks 2010). In the context of this research, a semi-structured approach allowed exploration of professionals' views of women's decision making about RRSO and their ideas for a possible patient decision support intervention in this context (reported in Chapter 7), whilst also allowing issues that were important to the professionals to emerge.

A purposive sample of healthcare professionals who worked with women at increased risk of ovarian cancer, and those involved in the psychological and/or emotional support of women in this situation, was identified through personal contacts and institutional websites. Eligible professionals either worked for genetics services, hospitals or charities in England, Wales and Scotland. They were contacted directly by the researcher and sent an information sheet to explain the study. Full written consent was obtained before the interview. The researcher (Jana Witt) conducted interviews in the professionals' office or a meeting room at their workplace. One interview was conducted over the phone. Interviews focused on (i) professionals' experience and perceptions of women's decision making regarding RRSO and (ii) their views on, and ideas for, the content and format of a decision support intervention (reported in Chapter 7). An interview topic guide is included in Appendix 4.3. Interviews took place between June 2011 and February 2012.

4.2.4 *Data analysis*

All focus groups and interviews were audio-recorded with participants' consent and later transcribed for analysis. An initial coding frame was developed from the topic guides for focus groups and interviews (Boyatzis 1998, Fereday and

Muir-Cochrane 2006). After careful reading of the transcripts, the coding frame was amended as new themes and codes emerged from the data (Appendix 4.4; Boyatzis 1998, Fereday and Muir-Cochrane 2006). Sixty per cent of focus groups and over eighty per cent of interviews were double-coded and discrepancies between coders were discussed until an agreement was reached. The coded extracts were then managed and analysed in the data analysis software Nvivo8 (QSR International Pty Ltd 2008). Codes were arranged under five themes: 'Background information'; 'Needs, questions and concerns'; 'Decision making'; 'Decision support' and 'Miscellaneous'. In this chapter only data within the 'Background information'; 'Needs, questions and concerns'; 'Decision making' and 'Miscellaneous' themes are discussed. Data within the 'Decision support' theme, which includes codes discussing format and content of a possible intervention, are reported in Chapter 7. Samples of coded transcripts are included in Appendix 4.5 and 4.6.

Analysis focused on identifying factors that were perceived to influence the decision, which were categorised as either 'medical or physical', 'psychological and emotional', 'social context', 'demographic' or 'other' factors. Further, these factors were then reviewed to identify specific barriers and facilitators to decision making. 'Barriers' were defined as factors that were thought to complicate the decision, contribute to remaining undecided and lead to unnecessary delay. 'Facilitators' on the other hand were defined as factors that made deliberation easier and could lead to a decision being made, either for or against RRSO.

Quotes presented in this chapter were selected as examples of themes and codes that emerged from the data. Quotes from focus groups are denoted as P[1/2/3][S/C/L], where the number describes the participant and the final letter indicates the location (S=Swansea, C= Cardiff and L=London). Quotes from health professionals are denoted with HP. Quotes were edited to remove word repetitions and irrelevant sections. Where content was removed this is indicated by a [...].

4.3 Results

4.3.1 Response rates

Response rates for this study were low amongst patients invited through the genetics or familial cancer clinic services. Response rates to focus group invitations for the three sites in the study are shown in Table 4.1. Initial response rate across the three sites was 16.9% (13 of a total of 77 women approached), which, while disappointing, is not unusual for a focus group study, as recruitment is known to be difficult for this method (Bloor et al. 2000, Phelps et al. 2006). Final participation was three in Swansea (one participant cancelled due to personal health problems and one did not attend on the day), three in Cardiff and three in London (one was unable to attend due to family illness and one could not attend on the chosen date due to work commitments). Final participation of those who initially registered their interest was 69.2% (9 of 13). Data regarding the characteristics of non-responders and non-attendees were not collected.

All participants were registered patients with a clinical genetics service and had been assessed as being at increased risk of ovarian cancer. Patient characteristics collected during the study are shown in Table 4.2. Five women were pre-menopausal, with three reporting experiencing some menopausal symptoms (peri-menopausal) and one having completed the menopause. One participant had received a negative genetic test result before the date of the focus group, one came

Table 4.1 - Response rates to focus group invitations by study site

| Site: | Cardiff n (% of invited) | London n (% of invited) | Swansea n (% of invited) |
|---------------------------------|------------------------------------|-----------------------------------|------------------------------------|
| Total patients invited | 25 | 32 | 20 |
| Registered for study | 3 (12%) | 5 (15%) | 5 (25%) |
| Final focus group participation | 3 (12%) | 3 (9%) | 3 (15%) |

Table 4.2 - Focus group participant characteristics (self-reported)

| | Number | Percent |
|---|--------|---------|
| Menopausal status | | |
| Pre-menopausal | 5 | 56% |
| Peri-menopausal | 3 | 33% |
| Post-menopausal | 1 | 11% |
| Genetic mutation status / Lynch syndrome | | |
| Unknown status | 7 | 78% |
| Confirmed positive (BRCA1/BRCA2/Lynch) | 0 | 0% |
| Confirmed negative | 1 | 11% |
| Lynch syndrome family history | 1 | 11% |
| Prior cancer | | |
| Yes (Breast cancer) | 1 | 11% |
| No | 8 | 89% |
| Participated in UK familial ovarian cancer screening study | | |
| Yes | 8 | 89% |
| No | 1 | 11% |

from a Lynch syndrome family and seven had not been tested or had not yet received their test results. The majority of participants had taken part in the UK FOCS study (Brain et al. 2012, Rosenthal et al. 2013a). The average duration of focus groups was 79 minutes (range: 77 to 83 minutes).

Response rate for health professional interviews was 65% (11/17). Professionals included four genetic counsellors, four gynae-oncologists, one health psychologist, one clinical nurse specialist, from hospitals in England (n=3), Wales (n=6) and Scotland (n=1), and one nurse working for a national charity support line. Eight interviewees were female and three were male. The average duration of interviews was 45 minutes (range: 24 to 80 minutes).

4.3.2 Medical and physical issues

Mutation status: Confirmation of a genetic mutation was identified by women and health professionals as the main motivator for RRSO decision making. The certainty that comes with knowledge of gene carrier status and associated risk estimates was often interpreted by women as a 'diagnosis', which then acted as a trigger to opt for

surgery. This effect is clearly illustrated by one woman who had just consented to be tested.

P2S: Now if [the gene test] is a 'Yes' then I'll be seriously...

P1S: That's the decision made for you, isn't it?

P2S: Yeah and that's the conversation that I have had with my husband that if the letter comes back to say: 'Yes, you have got the gene which is more likely to develop into ovarian cancer' I'll be in [for surgery].

Another participant who had tested negative before the date of the focus group also confirmed the role of gene mutation status as catalyst for surgery.

P3L: Had I tested positive for one of the genes I was going to have surgery [...] that was my action plan.

Similarly, health professionals reported that confirmation of carrier status was associated with decision making and often interpreted as a 'diagnosis' that needed 'treatment'.

HP7: ...I suppose having a BRCA mutation confirmed kind of makes it very clear that: "This is what my risk level is. This is a diagnosis, now it's clear, might as well have the treatment"...

HP2: ...If people have a clear [...] awareness of a gene mutation that makes it very clear in their minds that they need to consider different options...

Women also interpreted a negative test result as a facilitator to their decision, as even a negative test result was believed to result in a more exact risk estimate on which to base decision making.

P3C: Because I think [genetic testing] influences so much.

P1C: That could be the end, you could stop there.

P3C: Yeah, exactly.

P1C: If you haven't got the gene, couldn't you?

P3C: Yeah.

P2C: Because if you haven't got the gene then obviously the percentage is so-and-so and that influences your decision.

However, one health professional voiced concern that women's interpretation of a negative test result may be overly simplistic, emphasising that a negative test does not necessarily mean a 'low' risk, as there are genetic variants that cannot be detected through currently available tests. Deferral of surgery based on a negative test result may therefore not be appropriate.

HP4: ...women where the test comes back negative [...] might think: "Oh, right, so the test has come back negative, that means I am not at increased risk of it, or maybe I am not at the increased risk of the BRCA1/BRCA2 type, so maybe I don't need to think about surgery yet."

Cancer diagnosis: A previous cancer diagnosis was often discussed by health professionals as an influence on RRSO decisions. Most felt it was a catalyst to wanting surgery.

HP7: ...[Patients argue:] "[I] have had chemo in the past, definitively don't want to have that again!" And so I suppose having had breast cancer in the past makes it more real and so they then opt for surgery...

Conversely, several professionals mentioned that a prior cancer diagnosis could act in the opposite way to deter a patient from opting for RRSO in order to avoid a hospital stay and/or surgery while they are healthy.

HP9: ...Particularly for women who have had cancer [...] there are two groups: there are the ones who have already been through [cancer treatment] once and cannot face having chemotherapy again [...] and there are the ones who have been ill with cancer and then find it very hard to contemplate having an operation which is not because they are sick...

Women in the focus groups did not discuss the influence of prior breast cancer on decision making as only one of the participants had directly been affected by cancer and therefore this consideration did not apply to the majority of participants.

Risk-reduction: As the main reason for recommending surgery is to reduce a woman's ovarian cancer risk, ovarian cancer risk reduction was a key issue discussed by health professionals. One professional remarked upon ovarian cancer

risk reduction as the main focus for women who do not wish to have children or have completed family planning.

HP2: ...Some individuals that I have worked with, they have been very clear that they don't want children and therefore their highest priority and the greatest value that they have attached to their decision making is reducing their risk...

Risk reduction was also mentioned when professionals spoke about balancing the positive effect of surgery on cancer risk with the potential negative effects of surgery, such as surgical risks, hospital infections and oestrogen deprivation.

HP8: ...But of course it's a balance of risk and benefit. I think that's the bottom line: How much [...] risk is there from ovarian cancer and how much benefit from removing the ovaries that are technically healthy and normal?

During the focus groups, women themselves did not discuss ovarian cancer risk reduction specifically as a factor in their decision making; however they did discuss breast cancer risk reduction. When women found out during the focus group that breast cancer risk was also reduced by RRSO, they saw this as a positive effect that could influence their decision.

P1C: I didn't know that having your ovaries removed would reduce the risk of breast cancer.

P2C: No I didn't know. By up to 50%.

P1C: And that would influence my decision certainly because I feel that in my family – even though my mother had ovarian cancer – that my risks are higher in terms of breast cancer.

Menopause and hormone replacement: For most women, surgically-induced menopause was one of their main concerns, as illustrated by one participant's comment:

P1S: I know from having discussed [surgery] that it throws you into an early menopause, which for me is what is making me stick my head in the sand a bit at the moment. [...] I think that's really what my primary concern would be.

Women were worried about psychological changes following surgery and surgical menopause and how those might affect their lives.

P1L: [...] Is [surgery] going to change your personality? Like your hormones. And are you going to turn into this mad, neurotic [...] woman that no one recognises?

Women were also aware of the physical symptoms of surgical menopause, such as hot flushes, night sweats, weight gain and skin changes, and were considering how to counteract those changes.

P2L: I was thinking: "Oh God, if I have [surgery] I need to lose weight before I have it [...] just in case everything explodes" [laughter] and "I have to invest in very rich face creams" and stuff like that. [...] I think a lot of it was physical things.

Consequently, women not only wanted to know about the exact effects of surgical menopause, but they also needed information about different management options for menopausal symptoms, including (but not limited to) hormone replacement therapy (HRT).

P3C: I think in conjunction with it there needs to be a lot more information about the menopause and different treatments, whether they are conventional treatments, alternative treatments or combinations of them. Because I think that will address a lot of women's fears because it's [...] the fear of what comes next, head first into the menopause. [...]

P2C: I agree with that. [...] A friend of mine has had her ovaries removed and she said: "[Name] I would really really [...] consider it really really a lot because", she said, "you just suddenly go into menopause and you don't know whether you're on your head or your backside."

Health professionals also picked up on the issue of early menopause as a major concern for women and it was clear that within consultations a lot of time was spent on answering women's questions and allaying their fears and concerns regarding this issue.

HP9: ...One of the most important things for those women [...] is the issue of HRT and having an early menopause and how that's going to be for them in terms of their mood and their lifestyle and their bones and their husband and all of those things...

HP7: ...For the younger ones it has to do with the menopause and what it would mean for them, what the implications are, what the menopausal symptoms are likely to be...

HP6: ...For most of them menopause is the issue. Premature menopause. So there's a clear discussion about that. If someone is really anxious we [...] make sure they meet the menopause team...

Although menopausal symptoms only affect women who have not yet completed natural menopause at the time of surgery, one professional mentioned that even older women were concerned about the menopause.

HP1: ...They are worried about what lack of oestrogen is going to do and all sorts of things. [...] Definitely the younger ones, but surprisingly also with the older women, even if they have gone through the menopause...

Women had also heard about the potential association between HRT and breast cancer risk and were concerned about this, especially in the context of an already increased breast cancer risk.

P1S: She [doctor] said: "You could go on HRT", but of course HRT then brings up the increased breast cancer and where do you pitch it?

P3S: Yeah, where is the risk?

P1S: Yeah.

Correspondingly, professionals reported their concern about women delaying surgery due to fears about HRT and breast cancer risk and appeared determined to explain to patients that short-term HRT was safe, even for individuals with a family history.

HP5: ...Women often ask about HRT and they get messages from their GPs that HRT is really bad if you have a family history of breast cancer, whereas in actual fact [...] the amount of HRT you are on after risk-reducing oophorectomy would be a lot less than your body would

produce if you still had your ovaries. So, [...] it's important for women actually to be on HRT at least up until the age they would normally have gone through menopause to protect their bones and all of those other benefits...

HP7: ...As soon as you say HRT then the next question that kind of comes up is: "Well, I thought HRT increases your risk of breast cancer?" or "Isn't it associated with breast cancer?" And so you have to unravel and undo a lot of the stuff they have heard in the media...

Health professionals understood decisions about HRT to be an integral part of the decision making process about RRSO. They felt that HRT and its implications should be discussed with women in order to make informed, value-adjusted decisions.

HP2: ...Some women may not understand very much about the implications of taking HRT and menopause and I think [...] it is very important that they [...] do process that and have good information with which to make those judgements...

Professionals felt that HRT was essential for women who have surgery before the age of 45, due to the potential negative long-term health effects of oestrogen deprivation.

HP8: ...[We discuss] hormone replacement therapy if surgery is done at an early age. Sometimes if a woman comes at 48, and they are worried then I think the issue of hormone replacement therapy becomes more flexible and debatable, but if somebody is [...] under 45, then the recommendation is that hormone replacement therapy should be discussed and offered...

HP6: ...There is a mortality issue if they don't have oestrogen until 45 and after that it's a quality of life issue. [...] but below 45 if someone says they don't want to take HRT, we would be very reluctant to undertake surgery in the absence of any contra-indication for HRT, just because they don't want to take it...

From a medical perspective, contra-indications to HRT were also an important influence on decision making, particularly with regards to prior breast cancer diagnosis.

HP11: ...Unless of course they have got breast cancer [...] in which case you can't really give them HRT, so that's [a] very difficult decision for them. Because if they are in their 30s [...] and they have got a long time before natural menopause [...] the menopausal symptoms are much much more severe [...]

HP7: ...For that younger group the most difficult will be [...] those who have had breast cancer in the past, [...] because obviously they can't have HRT. And some of them [...] can be distressed and anxious, because they've [...] had breast cancer young, they don't want to have ovarian cancer, but what are they going to do [...] if they can't take HRT?...

Women in the focus groups did not discuss contra-indications to HRT, as they either were not aware of any contra-indications affecting them or were post-menopausal and therefore did not need to consider HRT.

Risks of surgery: Some women were aware of the risks of the surgical procedure itself and the risks of acquiring a hospital infection. These participants voiced concerns about taking those risks whilst being healthy.

P11: I'm 42, it is quite early I [...] feel and putting yourself through surgery when – touch wood – there isn't actually anything wrong with me at the moment you think: Any surgery has risks, doesn't it? And I think that's what is kind of holding me back at the moment

P3S: [...] would you want to go into an NHS hospital or would you want to pay to have it done privately where they have not got all the infections that you have got in the hospitals here? Because you could just be adding to your problem.

P2S: Yeah, you could, you are quite right, yeah. That is the frightening reality of today, isn't it?

The professionals also emphasised that RRSO is a major operation that bears risks which patients will be made aware of before consenting for surgery and need to consider before their final decision.

HP8: ...As part of the discussion of surgery, you explain to them what the surgery involves, what are the potential risks of surgery, complications, anaesthetic, operative, post-operative. That comes as part of the surgical explanation...

HP10: ...It's a major operation, so that is one of the minus points [...] and there are potential complications, serious complications, so that is the minus points [...] it's essentially doing a prophylactic operation without any cancer there...

One professional acknowledged that concerns about surgical risks were a factor that could prevent women from opting for surgery.

HP6: ...For some people it's [difficult] because they never had surgery and the risk of complications of the surgery [puts them off], because they would have to sign the consent form, where we have to write all sorts of complications...

Results of screening: During the time of the current study the UK familial ovarian cancer screening (UK FOCS) study, which offered annual trans-vaginal ultrasounds and CA125 blood tests three times a year, was coming to an end. As most women in the focus groups had taken part in the UK FOCS study, they reported that an abnormal finding during one of the screens, particularly the ultrasound scan, would act as a facilitator to making the decision.

P2S: If they ever said to me: "Yes, there is a change or there is something." I wouldn't hesitate. I wouldn't even want to read up on it. You could book me in, I'll go in the next day. No, I wouldn't bat an eyelid.

P2C: If I saw something on the scan that looked untoward and they said it looked untoward then I would be a bit like: "Oh Yeah, just get rid of it now then." [...] I guess I wouldn't think about the after, I'd just think at the time: "Just get rid of it!"

One of the professionals also noted that abnormal findings during the screening could act as a catalyst for patients to consent for surgery.

HP3: ...The main precipitant to the decision, I found, has been a jog from us for whatever reason [...]. In fact that's perhaps the big benefit of screening [...] it scared quite a few people into surgery...

Other: A number of other medical and physical factors were briefly discussed by both women and health professionals during the study. These included operative

procedure, residual risk after surgery and possibility of finding cancer during surgery.

4.3.3 *Psychological and emotional issues*

Perceived cancer risk: Health professionals discussed the role of perceived risk in women's deliberations about surgery. They reported that a woman's perception of her risk of ovarian cancer could make her lean one way or the other. Perception of risk was understood as independent of the objective risk as determined by family history and/or gene carrier status.

HP9: ...often for those women who come in saying they know they definitively want to have the surgery, it's [...] their understanding of the risk or how they perceive the risk [that] is important...

HP10: ...We used to have people with very high risk disease, you know, BRCA1 positive, who wouldn't have an operation and people come with low risk disease, who really didn't need to come to clinic, who would want a hysterectomy and the works. [...] I think an individual's perception, [...] that's the thing. [...] The perceived risk...

One professional explained that while a woman's perception of her risk may make her lean towards having or not having the surgery, this may be counterbalanced by her perception of the risks of surgery.

HP4: ...It comes down to the woman's perception of her risk. She might have a relatively small risk – at least as far as we are concerned – but if her perception of that risk is really high, then she'll make a clear cut decision. [...] Whereas if we might think she's got a relatively high risk, but her perception of that risk is not so high or maybe if her perception of the cost of surgery is going to be so much higher, then she'll find it much more difficult making a decision...

Women themselves only briefly discussed their ovarian cancer risk levels. They spoke about it using the estimates they had been given in clinic; however some provided their own interpretation of the level of risk as well.

P3C: I'm sort of the high-end of medium risk. [...] because it's only my mother and her mother, that's clear that it was ovarian cancer.

P1L: They said to me that I'm 70% risk of ovarian and about 80% risk of bowel cancer, so yeah it's sort of quite high.

One participant spoke about her perceived risk of breast cancer as opposed to ovarian cancer.

P1C: I feel that in my family – even though my mother had ovarian cancer – that my risks are higher in terms of breast cancer.

Women's interpretation of their risk appeared to depend not only on the estimates provided in clinic, but also on their experiences of cancer within the family.

Cancer worry and anxiety: Both women and health professionals mentioned cancer worry and anxiety as factors influencing decision making. Women explained that their at-risk status was a constant worry to them and could not be fully understood by others, who were not in their position.

P2S: Unless you are in this bubble, the outside people don't understand. People just don't comprehend for one minute, I think, the worry, because it is a worry.

Women also described that the intensity of cancer worry was not static, but rather that it varied from day to day depending on external factors and their own state of mind.

P2S: Depending on what mood you are in, you might think: "Oh, it's actually not quite important to me today" or "That is really important!"

Similarly, professionals considered patients' emotional burden when giving advice about RRSO decisions. They felt that women who were experiencing extreme cancer worry or anxiety could sometimes benefit from surgery.

HP8: ...The quality of life is affected, so you have to take that element into consideration for surgery. I mean if she says: "I can't deal with it emotionally." [...] I think it's fair to listen...

HP1: ...Some people have a cancer phobia, haven't they? So if you've got a cancer phobia you are going to want to have [surgery] done...

Belief in screening: As most women in the focus groups had participated in the national screening study, many reported that screening had been a welcome alternative to surgery and had given them a ‘safety net’ or ‘peace of mind’.

P2C: I was happy to have the screening. While they offered me the screening I was absolutely over the moon. I thought: “Well, they can keep track of my ovaries every six months and [...] if there is anything they’ll find it.” [...] I was really, really happy to be on the ovarian screening thing.

P1C: I was as well. It’s the reassurance.

P2C: Reassurance, yeah. [...]

P3C: It felt a bit like a safety net I think.

P2S: Wrongly or rightly I always felt that it was a bit of a safety net being on the [screening] programme, because I have my blood tests quarterly [...] I was coming here every January for scans and although I knew [...] it’s not a guarantee you just think that it’s early detection. [...] I felt that I was getting a lot of benefit out of it. A little peace of mind, wrongly or rightly it did feel like peace of mind and I have felt very vulnerable since it has come to an end.

These positive feelings towards screening remained true even for those women who were clearly aware that screening offered as part of the trial was not yet medically proven and that there was a possibility of false negative results, as illustrated by two participants’ comments.

P3C: I was told by a genetic counsellor even with the trans-vaginal scan that it’s not infallible [...]. That women who have had those and CA125 tests [...] then developed ovarian cancer [...]. So it’s not 100% but it gives you a better chance, doesn’t it? Because at least you’re being monitored.

P1L: Obviously it was made clear to you [screening] was not 100% guarantee so it may not have picked up any abnormalities but [...] you felt as though at least you’re doing something positive towards checking early signs of any problems.

Professionals on the other hand often felt that the availability of screening as part of the trial might have given women a false sense of security. They wondered if

women's faith in / reliance on screening prevented them from opting for surgery, despite surgery being the only medically recommended option.

HP5: ...It was always a worry that I had that I felt [patients] were putting too much store in ovarian screening, even though we [...] always would point out to women that not only are there lots of false positives, but there are also lots of false negatives and that there's no guarantee that the screening would actually be any use. But in spite of saying that, they still put a lot of store and a lot of hope [into screening] and gained a lot of reassurance from [it]...

HP4: ...If you can have screening then no matter how useless we might say it is, or not helpful we might say it is, some women still hold on to it [...]. It's falsely reassuring if we can't say it is definitively useful...

HP7: ...it's a very mysterious relationship these high risk women have with screening I think and it's not always easy to understand, especially since the information we give is clear, the message we give face-to-face is clear, but they still [...] gain more reassurance or have a higher expectation of screening than they ought to have...

One professional spoke about a group of patients who had had negative screening experiences, such as a false negative result, and who therefore put less trust into screening. She remarked that this group had a more accurate understanding of the reliability of screening, which could be influential in their decision making.

HP6: ...Some of the patients have obviously had breast cancer before and some of the breast cancers have been missed on mammography, so then their understanding of screening is better, in the sense that it's not foolproof and I think their belief is correct [...] so it helps them make their decision...

Family planning and fertility: As RRSO leads to infertility, the implications of infertility must be fully considered before opting for surgery. Professionals emphasised the importance of women considering their family plans and confirmed that women delayed making decisions about surgery based on their desire to have children, or more children.

HP7: ...We generally standardly say that one of the criteria is that you know you have completed your family. [...] There is some who delay it on that basis that they want to have another child...

HP9: ...The issue of children, there are some women where that will stop them because [...] they haven't had children yet [...] and they may not even have a partner, but they can't quite put themselves in a position where they can no longer have that possibility...

In agreement with this, one participant confirmed that a desire to have children would take priority over cancer risk reduction.

P1L: If [...] you want more children then that's going to be your first choice, first consideration I would've thought.

The loss of fertility as a separate issue to family planning issues also played a role in health professionals' discussions of women's decision making. One professional stated that allaying concerns about fertility could lead to a deferral of decision making.

HP2: ...The anxiety I think from a lot of health professionals is [...] the [...] loss of fertility. [...]. The decision making process for ovarian cancer management is kind of pushed off because clinicians are trying [...] to help women to manage those concerns about losing their fertility with surgical interventions...

Women themselves did not discuss loss of fertility as an issue directly. This may be due to the fact that women in the focus groups reported having completed their families and therefore did not consider their fertility to be an important issue.

Femininity and body image: The psychological consequences of surgery with regards to body image and femininity were discussed by both groups of stakeholders. Women were concerned about how they might feel following surgery and whether their personality and perception of themselves might change. One woman, who had previously had breast surgery, observed that surgery could have a psychological impact, even if no changes were visible externally.

P3L: Any surgery can affect things like body image and how you feel about yourself afterwards, totally regardless of physical stuff.

Health professionals recognised the importance of femininity and body image in women's decision making processes and spoke about women's concerns and questions regarding those issues.

HP6: ...Body image [as a] barrier is a very big thing. [...] Because [...] some of the women who don't want to have [surgery] say: "Oh god, I'll suddenly become old and my skin won't be quite the same..." [...] For some people it's a huge issue...

HP7: ...Women do wonder: "How will I change if I lose my ovaries?", "Will I lose my femininity?", "Will I be less of a woman?" [...] those questions are asked...

One professional spoke about perceptions of the difference between mastectomy and oophorectomy in terms of visibility of the results and the differential impact on women's body image and perceptions of their femininity. This professional emphasised that, despite not being visible, the removal of ovaries and the womb, in the case of a full hysterectomy, could affect women's self- concept.

HP9: ...Some people that I have spoken to don't think [having oophorectomy] is as important for a woman as having her breasts removed. Perhaps because it's inside and not visible and it's quicker and for some women it's the same, but for some women it really is very important, [...] because [...] their womb is a very emotive area to do with their femininity...

Other: A few other psychological and emotional issues were briefly discussed primarily by health professionals. These included perceptions and fear of hormone replacement, which are linked to the concerns about breast cancer risk discussed earlier. Other psychological influences on decisions mentioned by health professionals related to hospital anxiety and to the effects of prior cancer.

4.3.4 Social context issues

Family history: Women at increased risk of ovarian cancer often have seen and experienced cancer in close relatives. The types of cancer their relatives suffered from influenced women's attitude towards ovarian cancer. This influence was evident both in terms of the importance women attributed to the cancer their relative had compared to other cancers and in terms of women's perception of the usefulness of RRSO. The comment below illustrates the awareness that the participant is not only at risk for ovarian cancer and may develop a different cancer even if she opted for RRSO.

P3S: Even if [RRSO] was 100% I mean I have got people in my family who have had other cancers so you'd then think: "Right, okay, well that one is out of the way."

P1S: Sods law I'll get something else now.

P3S: Yeah. "What is it going to be, what symptoms should you be looking out for now?" You know. Or "Is there some kind of surgery I can have to make sure I don't get that one either?"

Health professionals also confirmed that the type of cancer women observed in their relative could influence their perception of their own risk for that cancer and other cancers, which may be independent of their objective risk for those cancers.

HP5: ...Women whose mothers have died from breast cancer want to have risk-reducing mastectomies and women whose mothers have died from ovarian cancer want to have their ovaries out, even though the highest risk they have is to breast cancer...

Additionally, some professionals noted that a woman's relationship with a cancer sufferer and the role they played in their care could influence decision making.

HP2: ...what they do, it's often very dependent upon [...] their perceptions of what it means to be at increased risk and the closeness with which they have observed or they haven't observed the experience of cancer as a result of the high risk inherited factors...

Women were aware that the age at which a family member developed or died of cancer would be a significant time point in their own lives in terms of a requirement for decision making.

P1L: They did say because my mum was 48 when she died [...] I should be thinking of [surgery] in the next year or so really. So it's [...] difficult.

Professionals pointed out that certain events within the family might act as triggers for women to make a decision. These included a new cancer diagnosis within the family and a close relative deciding to have surgery.

HP6: ...I think somebody in the family getting ovarian cancer while you are waiting to make that decision [...] is a big thing to come and have surgery. [Or a] sibling or cousin you are close to having risk-reducing surgery...

One woman in the focus group reported having recently lost her mother to cancer. However, while she did experience this event as a catalyst for coming into the genetics service and considering RRSO, she still did not want to have surgery.

P3S: My mother has been quite ill up until recently with ovarian cancer and she has literally just died [...] but I still don't feel like [...] I want to have everything done now, so I would say that that is a good indication that I'm not going to want to have it done for the minute.

Women in the focus groups often spoke about their own experiences of cancer in their mothers or sisters. They talked about their worry that what happened to their mother or sister might happen to them, and how this might affect their children. However, although they did voice these concerns, women remained undecided about surgery.

P2L: You think about your children. I've seen my sister whose children don't have their mother and I think: "Oh for goodness sake, I'm worried about my hair getting a bit thin" and I'd much rather be there for my children.

P1L: I was eight when my mum died of ovarian cancer and I think just because I've got a 4 year old and a 13 year old and I don't want them to go through that.

Familial and work obligations: Women thought about the potential effects surgery could have on their life and ability to fulfil their duties in regard to their family and their career. While some women were primarily concerned with their familial obligations, such as the 'school run' and similar duties, others were more worried about the impact of surgery on their career.

P1L: I think it's having time off from the school run and doing all that [...] it's not like you've just got yourself to worry about, it's other people to think of.

P3L: My personal situation is that I work for a very small organisation [...], if it had been guaranteed keyhole: all well and good. If it went full works I would lose my job. [...] Obviously my life is worth more than my job but I wasn't going to lose the job unless I had to.

The question of recovery time after surgery was also discussed. Women in focus groups debated the effect longer recovery periods after different types of surgery might have on their families.

P3C: I think also the surgery technique is, whether it's keyhole or whether it's open surgery, is that keyhole is [...] far less invasive because with open surgery you can't drive for something like 8 weeks and you can't lift and you can't walk and you can't do this, that and the other, which impacts on your work, well just your life.

P2C: Getting the kids places everywhere, yeah.

P3C: Just everything, doesn't it? Whereas keyhole surgery appears to be a lot easier to recover from.

Health professionals also recognised familial and work obligations as issues that influenced decision making about RRSO. It was generally noted that the potential effect of surgery upon a woman's ability to carry out her normal life duties was an important consideration.

HP4: ...for some women [recovery time] is actually an important factor as to whether or not they decide to go ahead with surgery [...] because they might be a single mum and have children to look after, they might be a carer for somebody else who is off sick, ill, an elderly parent who they have to look after and so how long they would be off sick is important to them...

HP6: ...I think work plays into it, definitively. [...] Everybody is frightened [...], everybody is worried, not that they lose their job or anything, but about the impact of [taking time off for surgery] on other people at work. [...] Especially because it is prophylactic...

Relationship effects: Surgical removal of the ovaries and resulting infertility and surgical menopause may affect women's sex life post-surgery and thereby also change their relationship with their partner. Women were aware of the loss of libido following surgery and were concerned about the potential effect this might have on their relationship.

P3C: It's another thing: potential problems with relationships...

P2C: Exactly.

P3C: ...because of [loss of libido]. For some people it's not going to be an issue but for others it is.

P2C: For me it would be. [...] I have got a good relationship with my husband and it would cause a problem. Potentially it could cause a problem.

Health professionals also mentioned the potential of surgery to affect sexual functioning.

HP5: ...I would always bring it up as a menopausal symptom, that women have vaginal dryness and that that can make their sex life less satisfying for them. [...] patients differ in how they respond to that...

One professional was speaking about her experience of women reporting lack of libido after surgery. She remarked upon the difficulty of counselling a woman and her partner if the partner is unable to accept the changes in the patient's sexuality following surgery.

HP11: ...We get lots of patients [...] with problems with libido. [...] And they are very difficult to handle then, [...] because they might have come to terms with their lack of libido, but the partner hasn't...

Major life events: Women and health professionals also referred to other events occurring in the social surroundings of a patient at the time of decision making,

which could lead to a final decision either being made or deferred. These included positive events, such as a child graduating or getting married or a grand-child being born, which may stimulate a wish to avoid cancer at all costs.

HP9: ...I suppose a major life event for some people has been something that has [...] made them think. [...] Their son graduates from university and they think "I want to see the next one come through", some of those kinds of things will trigger that...

Similarly, some negative events may also make a decision to have surgery more likely. These may include diagnosis or death of a relative or withdrawal of a service, such as the screening.

HP1: ...If somebody has been found to have ovarian cancer and that has put them from having one member into having two, to put them in a higher risk group, [...] that's when they might phone up because they are worried...

HP7: ...Certainly since the [familial ovarian cancer screening] trial started to wind down and has now stopped, [...] there's greater interest in surgery, there is no doubt about that, yeah. Definitely there's an increase...

However, one woman remarked that despite feeling vulnerable initially when screening was withdrawn, competing life events and commitments had reduced the relative importance of making a decision about surgery to a point where she was now no closer to a decision than when screening was available.

P3S: When I found out that [the screening] was all stopping I was like: "Oh Gosh, this is it!" but then after a while [...] other things happen and it's not at the forefront until something happens or you hear about somebody that you actually think about it. Well I don't. I try not to.

In agreement with this, one professional noted that other life events, especially negative ones, may take priority over the ovarian cancer risk and therefore lead to deferral of surgery in favour of dealing with the current event.

HP9: ...When [...] a major life event happens which is a negative thing [...] it pushes [the ovarian cancer risk] back in their minds in terms of

importance [...] – if there is a tragedy in the family [...] or someone leaves them – then they no longer can think about something like surgery...

4.3.5 Demographic factors

Age: Health professionals recognised that a patient's age played a major role in decision making and issues to do with life stage could influence attitude towards surgery.

HP5: ...I think women think differently depending on what age they are and what stage of life they are at. [...] Some women might be finished [with] their family at 26, but then others might not start their families until they are 36...

HP7: ...I suppose it depends on [...] the age group. If you are near 60s or your 70s you just think: "Well actually I have had a good run of it and why should I go to theatre if I don't have to." And so they are living comfortably with that...

One professional remarked upon the difference between individuals that were familiar with their at-risk status from a young age and those who found out about their at-risk status in later life. Adjusting to the thought that medical intervention might be necessary was suggested as a particular difficulty.

HP6: ...The younger people, they know their status, so they already have a lot of medical context [and] they come for screening [...] but some of the old people who know their status only when they are 60 [...] for them it is much more difficult, because they have been really healthy and well...

Age was also mentioned in terms of the effects of surgery, which are reduced for women who opt for surgery when they have completed the natural menopause.

HP7: ...I think it is easier for women who are post-menopausal [...] I think the younger the women are the harder it is, especially if they haven't had any children yet...

Women in the focus groups did not specifically discuss age as a factor, but did refer to their own ages on occasion. One woman who had been very worried about

surgical menopause for example stated that she had now reached 50 and was therefore likely to go through the natural menopause in the near future.

P2L: I'm 50, and my thing was "Oh my God, I'm going to be plunged into menopause" but then worked out it's 90% that are going to go through it between 49 and 51 or something, so chances are it's going to happen anyway.

Other: Apart from age no other demographic factors were mentioned; however some of the women talked about 'wanting to be there' for their children, which may be an indication that parenthood or parity might be a factor influencing decisions. However, even those women remained undecided and therefore no clear conclusions can be drawn.

4.3.6 *Other issues*

Physician recommendation: Some of the health professionals clearly supported RRSO as the best management option for women at increased risk and were recommending this option to their patients.

HP7: ...I mean we are very emphatic that surgery is the thing that we recommend. And that really, you know, if you are in your 40s and certainly if you are over 50, you should be having your ovaries out...

HP6: ...Everyone of us has to die, but I feel very strong that women don't have to die of ovarian cancer. It might [be] easier to have a heart attack...

Others, however, were keen to emphasise that they engaged in shared decision making about decisions and wanted the patients to take an active role in the process. While these professionals personally felt that RRSO was the best option, they sought to avoid being 'directive'.

HP5: ...I would be very happy if all my patients who were at risk for ovarian cancer had oophorectomies. I wouldn't want to push oophorectomies on them, but rationally it is the best way to manage that risk, but ultimately it's the woman's own decision...

HP9: ...we don't tell people that they should be having this at all [...] it is very non-directive the way that we do our counselling, [...] and so we do tell them that for some people this isn't the right decision and that [...] there are complications after surgery and all of those kind of things, but it's a balance...

Women also discussed the recommendations they had been given from different health professionals. It emerged that in many cases specialised consultants had been recommending surgery to patients quite strongly, as one woman remembered:

P2C: I think the general opinion of the consultants I've had is just like: "I don't know what you're making all the fuss about, you don't need your ovaries. It's not a question of if you are going to get it, it's when, so why are you not making this decision? Just get rid of the things!" [...] That's the opinion I've had [...].

However, the same patient emphasised that she would not make this decision based solely on the consultant's recommendation.

P2C: I couldn't just have it done with [my consultant] alone just saying it, I think I'd have to have other things to back it up.

P3C: Definitely!

P2C: That wouldn't be my main decision just because the surgeon said it.

In contrast, GPs appeared to be more reluctant to recommend surgery to their patients. In some instances, this reluctance seemed to reduce women's resolve to explore surgery in more detail.

P2L: I went to my GP and she said to me: "Just really think about it." Because she had had a patient that had come back and had problems with adhesions and stuff afterwards and said that if she'd known it was going to be like that she'd never have had the operation done.

P2S: "You are just as likely to be knocked down by a bus, so do you want to take that course of action?" that's what my GP told me. [...] and when you get that off your GP you think: "Oh, I don't know if I'll consider that any further now." You know, I had gone with a serious conversation of considering it.

There was a considerable range in the recommendations that women had received from different health professionals, which contributed to their confusion about the most appropriate course of action.

Elective procedure: The fact that RRSO is an elective procedure which would be undertaken in the absence of a clear diagnosis, while a woman was well and healthy, appeared to constitute one of the main concerns for women when they were trying to make a decision.

P2S: Well, I think it's difficult to, well essentially going in for surgery of your own choice. [...] there are so many other things that could go wrong. [...] it's going in with [...] no symptoms or no issue. You haven't been diagnosed. It's quite a big step.

Women distinguished between surgery that was required as treatment for a cancer diagnosis and risk-reducing surgery, which would be done to avoid cancer developing. They felt that surgery as a treatment was an easy, clear-cut decision; however they struggled with the idea of elective surgery in the absence of a clear diagnosis.

P3S: If somebody said to you: "You have got ovarian cancer" you would literally [...] be like "Okay, yeah, no problems." You know, well 'Yes' straight away.

P1S: But you wouldn't cut your arm off if there was nothing wrong with it, would you?

Women also remarked upon the difficulty of making the decision on their own and about actively asking for surgery as opposed to being told that they needed to have it.

P1C: In the back of my mind it is: "Well, should I be looking at this?" There's nobody prompting me, I don't see anyone to look at it, so it would be me taking it forward and requesting [surgery].

Trust in the medical profession: Women discussed their relationship with clinical staff and their level of trust in their GP, which was a factor that was important, especially with regards to identification of symptoms.

P3L: I think there's another factor which is how much you trust your GP [...]. I have trust that if I go to my GP with any of the symptoms on the ovarian cancer check box, she – knowing my history – is not going to mess around. [...] So that's another comfort level for me. [...]

P1L: It's having the confidence in your first port of call really, isn't it?

P3L: Yes.

In agreement with this, one professional remarked upon women's experience of having witnessed a failure by a GP during the care of a close relative. Again, women's trust in their GP's ability to identify and act upon symptoms was seen as an important influence upon their decision about surgery.

HP1: ...I think they may just want to get on with [surgery] because of their lack of trust. If you tell somebody to think about the symptoms they may say: "Oh well that's what my mother did and look what happened!" So, they would rather not trust that, they would rather just get on and have it done...

Women also explored the expertise of different surgeons at different hospitals and how this might influence their choice of location if they decided to opt for surgery.

P3L: My genetic counsellor who I saw locally [...], she said you get the choice [of where to have surgery] and I thought: 'Well, that's no real choice! I know where I'm coming!'

P1L: Yeah, so I think if [...] in five years' time something happens and I've got to have it done [...] I probably would look back and regret not having it done here where it's under someone that's got a good reputation.

Availability of screening: Women in the focus groups generally felt that screening had provided them with reassurance and that they would be happy to continue screening if it was offered after the trial completed.

P1C: Just feeling a little bit in limbo and would go to any further [screening] if it would reassure me and potentially help others [...] I would probably be quite grateful if I could have the CA125 annually or something just for that reassurance if nothing else.

Therefore the availability of screening appeared to reduce the attractiveness of the surgery option and the current lack of screening made women reconsider, albeit not opt for, surgery.

P2S: For me [the end of screening] has made me think more [...] about surgery, but it hasn't made me opt for surgery. I don't think I am any closer to it than I was a couple of years ago, but I just feel like: 'Should I be really considering that now?' Because to me at the moment that is the only way I have got of being screened then in some respects. Putting a full stop on that avenue. But I am no closer to having it.

Other: Stakeholders additionally mentioned some other issues that might be influential when making decisions about RRSO, such as patient personality and the vagueness of ovarian cancer symptoms. Women and health professionals also discussed the availability and importance of peer support on decision making about RRSO, with most feeling that information from a peer could facilitate decisions.

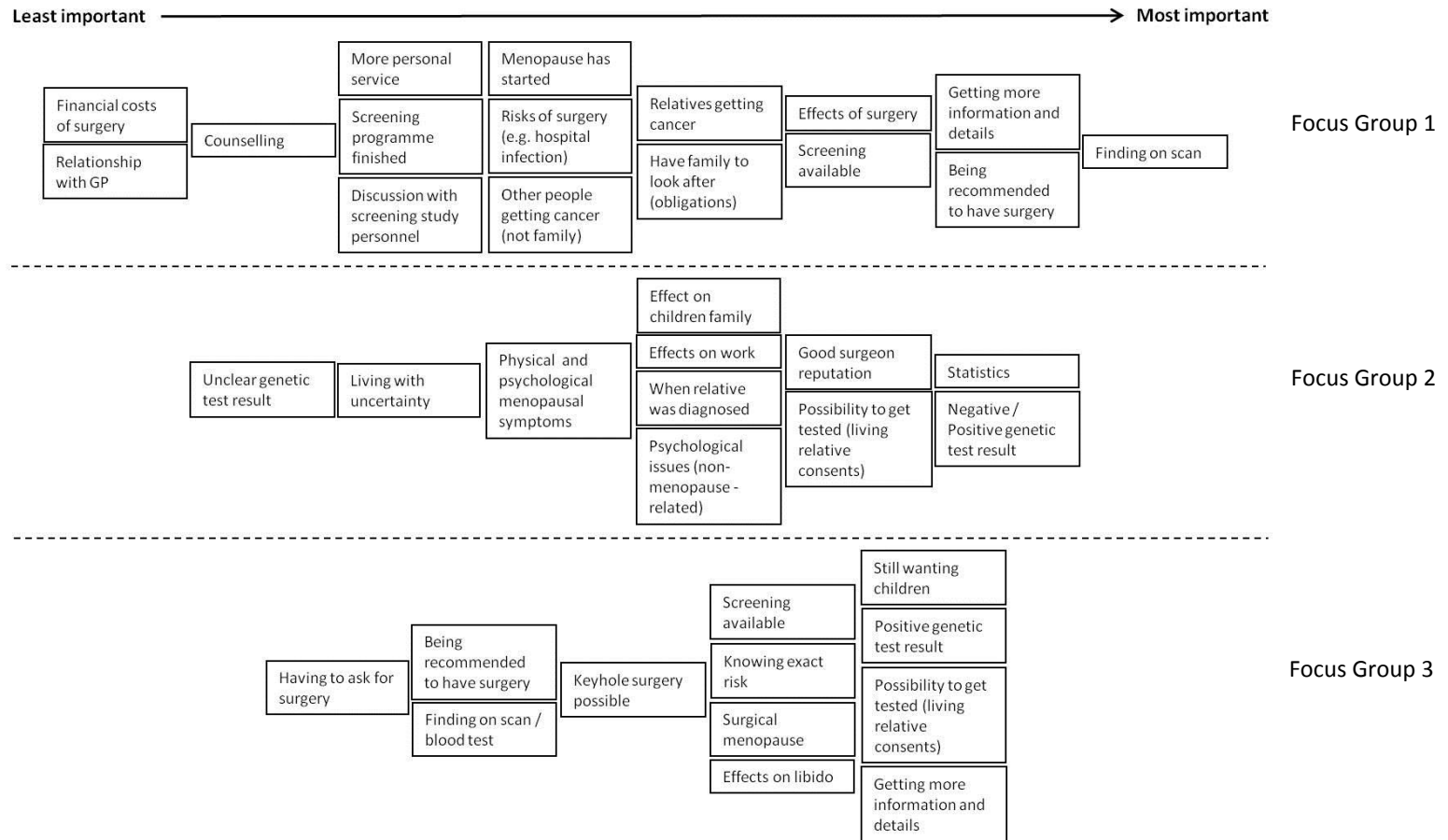
HP3: ...The other thing I suppose is what no woman has and would like at any point in making a decision is to talk to somebody who has already had it done...

4.3.7 *Relative importance of considerations*

The relative importance of different factors in the decision making process was explored with a ranking exercise during the three focus groups, in which participants were asked to collectively arrange factors mentioned during the group discussion in order of importance. Results of the ranking exercise are illustrated in Figure 4.1.

The effects of surgery and specifically the symptoms of surgical menopause were discussed in all groups. The groups differed in the importance they attributed to these effects; however all felt that they were at least moderately important. The second factor that was discussed by all groups was further information and details about cancer risk and risks of surgery. One group specified a wish for better statistical information. All groups rated the availability of information as one of the most important factors for their decision making. Two groups also discussed genetic

Figure 4.1 - Ranking exercise outcomes from focus groups



test results and exact risk estimates as important influential factors in their deliberations.

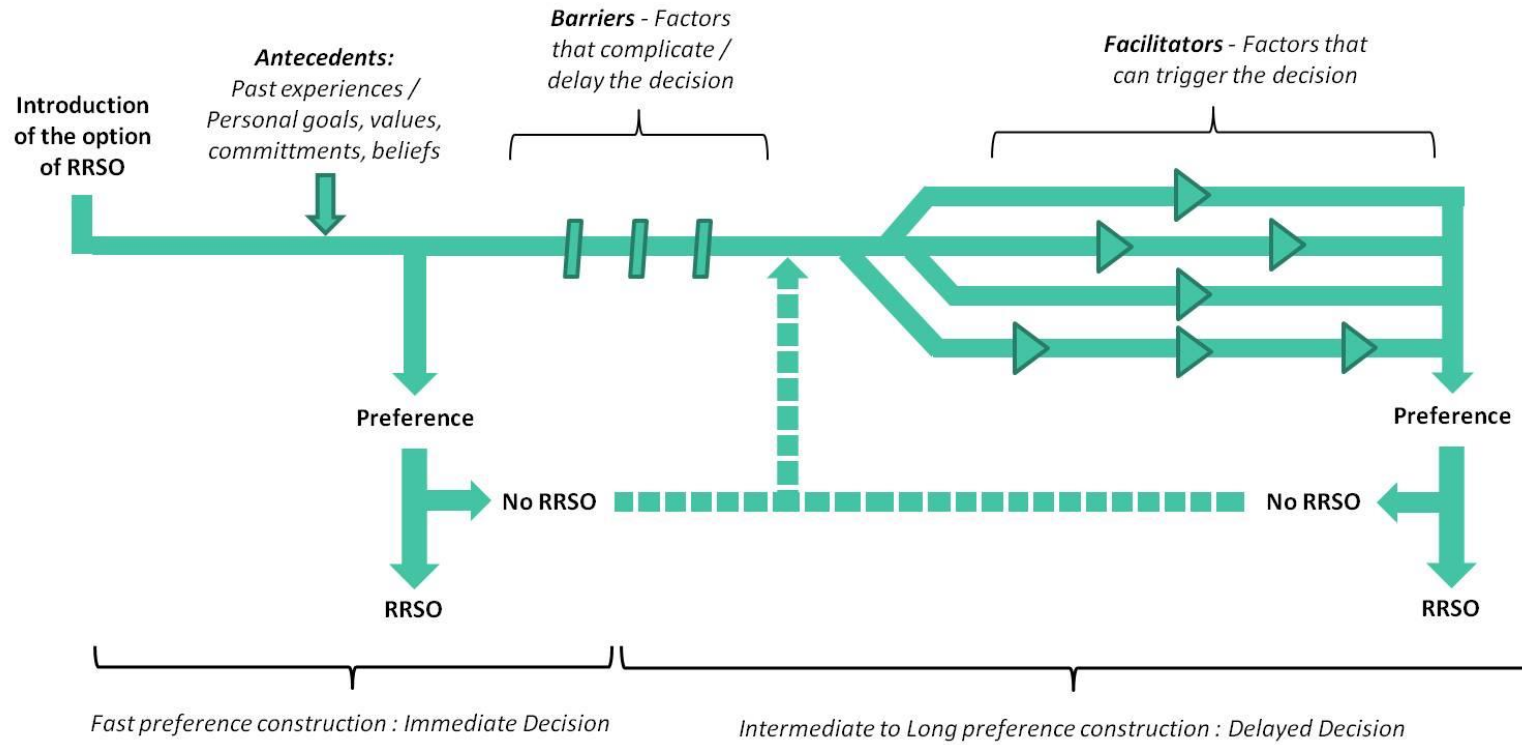
A range of other factors was discussed, including abnormal findings on screening and availability of screening. Interestingly, the former was rated very differently by the two groups that considered these. One group felt abnormal screening results were the most important factor and would result in consent for surgery, while the other group did not rate abnormal findings during screening as particularly important. In contrast, both groups felt that availability of screening and the associated possibility of knowing if something was wrong was an important factor. These two groups also rated the importance of a recommendation from a health professional very differently. Again, the first group rated it as very important and the second group as less important. The effects of surgery on the family and the ability to fulfil familial obligations were seen as moderately important factors by two groups. Other considerations discussed by individual groups included others being diagnosed with cancer, effects on work, type of surgery and numerous others, most of which were of moderate or low importance to group members.

4.3.8 The preference construction pathway

Due to the variety of factors that may influence appraisals and deliberations, the preference construction pathway women go through when making decisions about RRSO is complex. It may not only depend on the patient's values and past experiences, but also on the medical, psychological and social factors that could complicate or facilitate the decision, as highlighted by the current study. Figure 4.2 shows the preference construction process as it emerged from the data.

Once women knew about their increased risk and the option of RRSO, they would draw on their own experiences and look at their personal goals to form an initial opinion about this option. Some women were able to form a preference and make an immediate decision based on these factors alone. Health professionals particularly emphasised the role of past experience of cancer in close relatives as a reason to undergo surgery to avoid the same fate as their relative.

Figure 4.2 - The preference construction pathway during decision making about RRSO



HP3: ...Some I think have almost made their mind up straight away, because mum and gran had it, or sister had it and [they argue]: "I'm not going to have that happen to me." Near experience, that really colours it...

HP10: ...Often it's seeing a relative die of the disease [...] relatively young and I think that memory is something which drives people [...]. Mum died of ovarian cancer at 51 and they didn't have a very pleasant pathway to their death [...]. That can be a very strong motivator actually, very strong I found...

Similarly, having been directly affected by cancer and having gone through cancer treatment was identified by health professionals as a catalyst for making an immediate decision, both for and against surgery.

HP6: ...Having the breast cancer does drive people to want to have surgery [...] while for some other people the whole thing of having the breast cancer works in the opposite way [...] they have been in hospital so much that they just don't want anything for a couple of [years]...

HP9: ...Most women if they have had cancer don't want to go back through chemotherapy and the treatment side of things [...]. And there are some women where actually because of their prognosis from their breast cancer, they really want to go ahead and have their ovaries out...

Others may be able to make an immediate decision based on their personal goals and values, such as wanting to complete their family. In this case RRSO is not an option they would consider further until their goal was completed. Therefore, they would decide against surgery at the point of first discussion, but may come back to the decision later on.

In rare cases women may be suffering from hospital phobia. Under these circumstances a decision against surgery as a risk-reducing option is likely to be made very quickly. One professional's quote illustrates such a case.

HP7: ...[The patient] was just completely paralytic [...] this poor thing was just like completely falling apart and everything. [...] It was very very

extreme. She is definitely not going to have surgery, she is definitely not going to come in for an operation...

However, for many women a decision for or against surgery is not as clear cut, and is not made immediately. These women remain undecided for longer periods of time and may neither be able to fully commit to surgery nor to completely reject surgery. In these cases the deliberation process is prolonged and influenced by a range of factors that can act as barriers or facilitators to decision making (Table 4.3 and 4.4).

Table 4.3 - Barriers to decision making about RRSO

| Medical & Physical | Psychological & Emotional | Social context | Other |
|---------------------------------|---|--|---|
| Unable to get tested for a gene | Difficulty in adjusting to the reality of the situation | Familial and work obligations | Jargon / Communication problems |
| Inability to take HRT | Fear of effects of surgery / surgical menopause | Discrepancies between patient's and partner's preference | Health professional hesitation / differential recommendations |
| Concerns about surgical risks | Uncertainty about goals (e.g. wanting a family) | Competing life events | Concerns about procedure being elective |
| | Fear of or uncertainty about HRT | | Problems with follow-up / GP as gatekeeper |
| | Unresolved bereavement | | Gender of health professional |
| | Personality | | |

Table 4.4 - Facilitators to decision making about RRSO

| Medical & Physical | Psychological & Emotional | Social context | Other |
|---|--|---|---------------------------|
| Getting a genetic test result / precise risk estimate | Personal goal(s) reached | Input from family | Availability of screening |
| Natural menopause Reached / completed | Age at which relative developed cancer reached | Input from partner | New GP / Review by GP |
| Possible symptoms experienced | | Salient life event (e.g. child graduates) | Follow-up consultation(s) |
| Finding on scan | | Peer support | Personal time plan |
| Salient personal health event (other than cancer) | | Someone else gets cancer | Physician recommendation |

Many of the considerations identified by women and health professionals as influencing decisions could readily be labelled as barriers or facilitators. For instance, fear of the menopause and concerns about HRT emerged as barriers, while confirmation of a genetic mutation and physician recommendation appeared to act as facilitators. It transpired that a single facilitator may result in a decision, or several facilitators may act in synergy to drive preference construction, depending on the number of barriers in place. Facilitators may have differing effects for different individuals, for instance one woman may be able to make a final decision when she receives a positive genetic test result, whereas another might remain undecided even after receiving confirmation of a genetic mutation.

To illustrate how barriers and facilitators emerged from the data, consider the example of one woman who voiced her concerns about an early menopause. This was a barrier for her and prevented her from making a clear cut decision for surgery. She later stated that reaching the natural menopause would reduce her worries about surgery and therefore act as a facilitator; however she added that this alone would not trigger a decision for surgery.

P3S: My main thing would be the menopause [...] starting, [...] because [natural menopause] happens gradually, your body sort of gets used to what's going on, but where you have your ovaries out it literally starts straight away, doesn't it? [...]

P3S: I'm waiting for the menopause to happen naturally so that I can think: "Oh right, this is it. It's okay we can go ahead now, I don't have to worry about that bit anymore." [...]

P3S: I think if I had started the menopause then I might find it easier but I still think something else would have to happen to push me to have surgery.

The interview data from health professionals also confirmed that entering the natural menopause was a facilitator which resulted in women re-evaluating surgery.

HP6: ...as they go into the menopause [...] then they are like 'okay'. They are re-visiting it [surgery] [...] at that point, they kind of consider it again.

4.4 Discussion and Conclusions

4.4.1 Discussion of findings

The current chapter identified considerations during decision making about RRSO that may be important to women at increased risk of ovarian cancer in the UK. A model was also proposed in this chapter of the preference construction pathway during deliberations about RRSO, identifying specific barriers and facilitators which may play a role in decision making.

4.4.1i Medical and physical issues: As previously revealed in the literature search (Chapter 3; Howard et al. 2009a), medical and physical issues appeared to play an important role in decision making. Women and health professionals in the current study discussed genetic testing as an important event that could result in more accurate risk estimates on which to base their decision. This is in agreement with previous research, which found that confirmation of a genetic mutation appears to be a key event in women's decision making processes (Meijers-Heijboer et al. 2003, Phillips et al. 2006, Ray et al. 2005). It should be noted that others have not found a positive genetic test result to translate into high uptake of surgery (Lerman et al. 2000). However, this finding could be due to the relatively short follow-up time of 12 months in this study or may be caused by the lack of additional facilitators to trigger a final decision.

Health professionals spoke about the differential impact a prior cancer diagnosis might have on attitudes to risk-reducing surgery, stating that most affected women opt for surgery, while some might decline surgery to avoid returning to hospital when they are healthy. Earlier studies have mostly reported a positive association between prior cancer diagnosis and decisions to undergo RRSO (Beattie et al. 2009, Hamilton et al. 2009, Madalinska et al. 2005, Stuckey et al. 2010). However, one study did not find such a link (Tiller et al. 2005), which may

indicate that a prior cancer diagnosis may influence the decision either way, as suggested by health professionals in this study.

Ovarian and breast cancer risk reduction were mainly discussed by health professionals in the current study. Women did not specifically mention ovarian cancer risk reduction; however, previous research has found that women do consider this during decision making (Claes et al. 2005, Fry et al. 2001, Hallowell 1998, Kram et al. 2006). This discrepancy in findings may be due to the fact that women in the current study perceived ovarian cancer risk reduction to be the main and obvious reason why they might consider RRSO at all and therefore did not think to mention it specifically. Most of the previous studies were quantitative and provided women with a set of possible answers as part of a questionnaire (Claes et al. 2005, Fry et al. 2001, Kram et al. 2006), therefore it cannot be determined whether women would have discussed these issues of their own accord. Alternatively, women in this study may have experienced a higher number of breast cancers in their family and therefore felt that this was their primary concern. The women in the current study did discuss breast cancer risk reduction and its influence on their decision, similarly to other research (Culver et al. 2011).

Surgical menopause and HRT have been established as important considerations by earlier research (Brain et al. 2004, Fang et al. 2002, Hallowell 1998, Ray et al. 2005, Swisher et al. 2001). In agreement with these findings, women and health professionals in the current study discussed issues regarding surgical menopause and HRT at length, with some women citing this as their main concern.

Finally, the risks of surgery were also discussed by women and health professionals, especially in terms of their short-term effects and potential for complications. Women seemed particularly concerned about going into hospital whilst healthy and about the potential for hospital infections, which may be due to the recent media attention given to this topic. Earlier research has also found that

the potential for complications during and after surgery was a consideration addressed by women (Brain et al. 2004, Hallowell 1998, Hurley et al. 2001).

In contrast to previous studies, participants in the current study specifically discussed the impact of screening on decisions about RRSO in detail. This may be due to the involvement of women and professionals in the UK FOCS study (Institute for Women's Health 2010). Earlier research rarely reported influences of screening on decision making about RRSO. This is most likely explained by the lack of an ovarian cancer screening programme. Howard et al. (2010c), who noted that anxiety, discomfort and abnormal findings during screening may lead women to reconsider risk-reducing surgery, refer primarily to breast screening rather than ovarian cancer screening. The current study offers an insight into the potential effects of an ovarian cancer screening programme on women's decision making. In agreement with others (Brain et al. 2012, Howard et al. 2010c, Lifford et al. 2012), participants in this study reported that an abnormal screening result would lead women to revisit the surgery decision.

4.4.1ii Psychological and emotional issues: Psychological and emotional factors also played a role in women's decision making in this study, confirming the findings of the literature search (see Chapter 3; Fang et al. 2005). Perceived cancer risk was discussed by health professionals in the current study and others have previously noted that women's perceived risk was associated with decisions to undergo or decline surgery (Babb et al. 2002, Brain et al. 2004, Fang et al. 2003, Hallowell et al. 2001, Madalinska et al. 2007). Cancer fear, worry and anxiety, which are often linked to women's perceived risk, were discussed by women and health professionals. Professionals suggested that high levels of worry, anxiety or fear are linked to decisions to undergo surgery. However, despite being worried, women in the current study had not yet opted for surgery. In agreement with health professionals, earlier research has suggested that fear, worry and anxiety do play a role in decision making (Kram et al. 2006, Madalinska et al. 2007, Meiser et al. 2003, Schwartz et al. 2003). Considering previous evidence and the indecision of women in the current study despite their self-reported worry, it could be suggested

that worry or anxiety alone are not deciding factors, but that they may act as facilitators in combination with other factors.

Many of the participants in this study had been involved in the UK FOCS study and therefore discussed beliefs regarding the effectiveness of screening. In agreement with previous studies, women in the current study reported high levels of reassurance from screening despite receiving abnormal screening results (Brain et al. 2012). This could be expected, as earlier research had shown that women who perceived screening to be effective were more likely to opt for surveillance and none of the women in the focus groups had opted for surgery at the time of the study (Howard et al. 2011, Madalinska et al. 2007). Health professionals reported that women's beliefs in screening could deter them from making a decision about surgery and voiced concerns over the reassurance women gained from screening in the absence of proof of efficacy.

Women's fertility, femininity and female identity were discussed by both women and health professionals. Professionals emphasised the need for consideration of family planning and discussed the questions and concerns women voiced in the clinic regarding the effects of the surgery on the body and psyche. While women in the focus groups had completed their families and therefore did not specifically discuss fertility, they did consider the effects of surgery on their body, personality, mood and identity. Earlier research also found that these are important considerations in women's decision making (Babb et al. 2002, Hallowell 1998, Hallowell et al. 2012).

Health professionals discussed a number of further, less frequently encountered considerations. These included fear of hospitals, which had also been identified as a factor in earlier research (Hallowell et al. 2001, Wagner et al. 2000). Two other issues raised in this study - fear of HRT and psychological effects of a prior cancer - had not been specifically explored in previous research. However, both of these are linked to medical and physical discussions about the menopause,

HRT and prior cancer, and may therefore have been included in this context, rather than being explored as standalone psychological factors.

Earlier studies have highlighted a number of other emotional and psychological issues that may play a role in decision making, including coping style and self-efficacy (Chapter 3; Mellon et al. 2009). These were not mentioned in the current study. However, these are arguably relatively abstract concepts that indirectly affect decision making. Therefore, it could be argued that this study's design could not be expected to identify such factors, especially as participants were not explicitly asked about self-efficacy and coping during the focus groups.

4.4.1iii Social context issues: Participants in the current study explored a number of social context factors, namely family history, work and family obligations and effects on relationships. Women and health professionals discussed the type of cancer women had observed in relatives, which was commonly more than one, and how this influenced women's attitude toward RRSO. From health professionals' comments, it appeared that women's perceptions of their genetic risk were heightened by the emotional, rather than the genetic, bond between relatives; i.e. interpretation of their own risk depended on their experience of certain types of cancer in emotionally close relatives (Howard et al. 2011).

Women mentioned the age at which their relative developed cancer as an important time point in their life regarding the decision making. Health professionals identified a new cancer diagnosis in the family or a relative opting for surgery as facilitators to decision making. This is in agreement with earlier research identified in Chapter 3, which found that experience of ovarian and other cancers in the family played a role in decision making (Babb et al. 2002, Dagan and Goldblatt 2009, Hallowell et al. 2001, Metcalfe et al. 2008b, Swisher et al. 2001). Closeness of women to the relative who had cancer was not specifically discussed by women in the focus groups, but others found that experiencing cancer in a first degree relative may influence decision making in favour of surgery (Howard et al. 2011, Metcalfe et al. 2008b).

The impact of surgery on work and familial obligations played a major role in women's discussions about RRSO in the current study. Previous research had also found this to be an important consideration (Bradbury et al. 2008, Brain et al. 2004, Howard et al. 2010c). Health professionals in the current study confirmed that the impact on a woman's day-to-day life was an issue that was explored during decision making in clinic.

Earlier research also found that the impact of surgery on sexual relationships was an issue that women explored (Hallowell 1998). In agreement with this, women in the focus groups discussed potential problems that having RRSO might cause in their relationship, and health professionals emphasised the need to include partners in discussions about surgery.

The current study identified major life events as one additional social context factor which had not been explored specifically in previous studies. The influence of a major event, such as a graduation, wedding or death in the family, was perceived by health professionals as a facilitator to decisions. In contrast, women mentioned that other life events could reduce the relative importance of ovarian cancer worry and therefore lead to delaying the decision. Specifically considering other major life events with women when exploring the option of RRSO in clinic may be beneficial in clarifying how these may impact on their decision making and to work on a specific time plan to fit the decision into a woman's life.

4.4.1iv Demographic factors: Age was the only demographic factor specifically mentioned in the current study. It was discussed by women and health professionals primarily in the context of pre- and postmenopausal status, as postmenopausal women do not enter the menopause after surgery and do not need to consider infertility. Therefore the potential negative consequences of surgery in older age are reduced, which can make the decision for surgery easier. In agreement with this, earlier research has found that older age is frequently linked to decisions to undergo RRSO (Brain et al. 2004, Fry et al. 2001, Skytte et al. 2010). However, several health professionals mentioned that older patients chose not to

opt for surgery and this is in agreement with a previous study by Beattie et al., (2009), who found that from age 60 uptake of RRSO decreased. Therefore different age groups and associated life stages can influence the decision.

Earlier research had found that parity may be linked to RRSO (Schmeler et al. 2006b, Stuckey et al. 2010). As the current study was a qualitative study, the number of children and its influence on the decision making process for women in the focus groups is unknown. However, several women mentioned that they wanted to 'be there' for their children and family, which may indicate that having children does play a role in decision making.

Previous quantitative studies have investigated a range of other demographic factors, such as marital and employment status, education and income. However, none of those was found to be influential in decision making and participants in the current study did not explore any of these issues.

4.4.1v Other issues: A number of other factors were identified as considerations during decision making by women and health professionals. A recommendation from a health professional could act as a facilitator to decision making, either for or against surgery. Women in the focus groups quoted a whole range of recommendations they had received from their care teams and explored their reactions to those recommendations, indicating that they do have an effect on decision making. In agreement with this, earlier research also highlights the influence of physician recommendations, both for and against surgery (Claes et al. 2005, Fang et al. 2002, Kram et al. 2006). This range of recommendations indicates that there is a need for evidence-based, reliable information materials and decision support interventions to counteract confusion and facilitate decision making.

Women in the current study highlighted the fact that RRSO is an elective procedure and voiced concerns about having to actively seek this surgery by taking it forward themselves. In earlier research Claes et al. (2005) found that women who wanted to avoid surgery when they felt it was not necessary opted for screening, which may explain why participants in the current study picked up on the issue of

elective surgery. Most of the women in this study had initially opted for screening when it was available as part of a trial and therefore are likely to represent the population that perceives surgery as an extreme measure.

Trust in the medical profession was an issue discussed by participants in the current study, yet has received only limited attention in previous research about RRSO. Women's relationships with clinical staff, including their GP, genetic counsellor and gynaecologist, were remarked upon both by women themselves and by health professionals. Trust (or the lack thereof) may influence decisions either way. One professional explained that women who had seen a relative suffer from a cancer that had been missed in primary care would be reluctant to trust their GP and would want surgery. Women confirmed that trust in their GP and their ability to recognise symptoms is important. Women also discussed their trust in the expertise of the surgeons at different hospitals and how this might influence their decision in terms of where to have surgery. Trust may also be linked to adherence to health professional recommendations. The importance of trust has previously been shown to be linked to consent and treatment adherence in different medical contexts (Davies 1997, De Achaval et al. 2010, Stall et al. 1996), indicating that it is likely to play a role in decisions about RRSO as well.

As expected, a number of other factors identified in previous studies, such as the impact of RRSO on insurance contributions (Hurley et al. 2001) and the readiness of the healthcare system (Howard et al. 2010c), did not come up in the current study as they are not usually relevant to the UK population. However, some UK specific factors were explored in the current study. These included the availability of ovarian cancer screening on the NHS and/or privately. Currently, there is no routine ovarian cancer screening programme offered in the UK; however several areas might offer privately funded screening. Women in the focus groups did discuss the end of the screening trial and the potential for obtaining private screening, both of which appeared to influence their attitude towards surgery. Some women had sought information about private screening, while others had reconsidered surgery once the screening stopped. However, none had made a final

decision. The current situation regarding screening in the UK is complex, as the UK FOCS trial concluded in 2011 and results are not expected until late 2013. In the interim period, women who previously received screening were informed that they would no longer have this option until evidence emerges regarding its clinical effectiveness. However, some women may be able to obtain privately funded screening during this period, which could provide false reassurance in the absence of proven clinical effectiveness (Brain et al. 2012). Availability and beliefs in screening will therefore remain an important consideration in the decision making process until clear clinical guidance is available.

4.4.1vi Relative importance of considerations: The current study not only identified facts that might influence the decision, but explored their relative importance to women through a ranking exercise. This revealed that women frequently ranked further information, details and statistics about their cancer risk, such as a genetic test result, and the effects of surgery, including the menopause and HRT, highly, and felt these would help them in making a decision. This is in agreement with previous studies, which showed that women wanted reliable information about ovarian cancer risk and management options (Babb et al. 2002, Hallowell 2000, Klitzman and Chung 2009, Ray et al. 2005, Tiller et al. 2005).

In contrast, there was some discrepancy between groups in their rankings of 'findings on screens' and 'being recommended to have surgery', with one group ranking these very important in their decision making while the other attributed low importance to both these factors. This discrepancy may be explained by the differential experiences of group members. In the group that ranked both of these factors as very important, no one had experienced an abnormal finding on a scan or blood test, whereas in the other group two women had been recalled after an abnormal finding on a blood test. Both had been cleared and returned to routine screening after it was established that the abnormal finding was not ovarian cancer. Therefore, these women were aware that an abnormal screening result may not necessarily translate into a cancer diagnosis and may have felt less inclined to see an abnormal screen as an important factor in their decision. In agreement with the

first set of women, who felt a finding on a screen would be important in their decision, previous research has shown that women are more likely to consent for surgery after an abnormal screening result (Brain et al. 2012, Lifford et al. 2012). Therefore, this may be described as a facilitator to the decision. However, as is evident in the second group, some women may not opt for surgery despite an abnormal finding. Similarly, women in the first group had not received a strong recommendation for surgery from a health professional, with one even reporting that her GP actively discouraged her. In the group which attributed low importance to this factor, at least one member had received strong recommendations from a professional and yet still felt reluctant to decide. This demonstrates how differential experience can affect what women consider most and least important in their decision making. It also highlights the importance of trust between women and health professionals.

4.4.1vii Preference construction: According to the preference construction pathway, which emerged from the data, women may approach the decision in two ways. Some decide very quickly based on their past experiences and personal goals (immediate deciders), while others decide more slowly due to a number of barriers, which may lead to delay (delayed deciders). The first approach may resemble fast and frugal decision making based on heuristics, while the second may describe more deliberative decision making, following vigilant information seeking and a differentiation and consolidation approach (Gigerenzer and Goldstein 1996, Goldstein and Gigerenzer 2009, Janis and Mann 1976, Svenson 1992). During both these approaches, decision makers must appraise their risk and their options in order to construct a preference, whilst also regulating their emotional responses and appraising the coping resources at their disposal (Chapter 1; Lazarus and Folkman 1984, Power et al. 2011, Witt et al. 2012). For immediate deciders, this appraisal process may be fast and frugal, in accordance with heuristics (Gigerenzer and Goldstein 1996, Goldstein and Gigerenzer 2009). For delayed deciders, it may be slow and deliberative in accordance with the conflict model of decision making, differentiation and consolidation theory and a monitoring coping style (Janis and

Mann 1976, Miller 1987, Miller et al. 1988, Svenson 1992). The Coping in Deliberation (CODE) framework proposes a number of questions that decision makers may ask whilst going through the appraisal processes (Chapter 1; Witt et al. 2012). Using the considerations identified in the current study, along with the findings from the literature search (Chapter 3), adapting this framework may be useful in describing in detail the appraisal processes women go through when making decisions about RRSO (see Chapter 5).

4.4.2 Results in context

Overall, findings of the current study mapped well onto the results of previous research investigating women's appraisals of, and deliberations about, RRSO. However, a number of issues that were discussed at length among women who took part in the current study had rarely been explored in earlier research. Factors women may consider during decision making about RRSO depend on the local healthcare system and availability of options, may be influenced by cultural norms, may change over time and are likely to be influenced by current scientific developments and media reports.

The current study was conducted at a point in time at which a national ovarian cancer screening study for women at increased risk of ovarian cancer had just come to an end (Institute for Women's Health 2010). Consequently, many women in the study had participated in screening and were now faced with a situation where this option was no longer freely available. Therefore, belief in screening and the availability of screening was discussed at length by women in this study. Women who had participated in the ovarian cancer screening trial may arguably constitute a somewhat unusual population that has experienced screening in the past and is no longer offered it currently, although this may change once the results of the trial are available. In the absence of medical proof, women can now opt for private screening. This does however have cost implications. Alternatively, they can decide to have surgery or simply do nothing. Women who have experienced screening in the past may be more likely to opt for private screening to

achieve the same reassurance they experienced during the trial and may be less likely to opt for surgery because of this reassurance (Brain et al. 2012). However, it should be noted that health professionals provided anecdotal evidence of higher rates of uptake of surgery after the screening trial had stopped, indicating that some women do opt for surgery once they are faced with a decision of whether to have surgery, do nothing or pay for private screening.

Women in the current study voiced concerns about the risks of surgery, specifically the risks of hospital infections. In recent years there has been an increase on media reports of hospital infections, with a focus on pathogens such as methicillin resistant *Staphylococcus Aureus* and *Clostridium difficile*, that could influence public opinion of UK hospitals (Boyce et al. 2009). This could have resulted in women perceiving the risks of surgery and hospital infections to be relatively high during this study, which could partially explain women's reluctance to opt for surgery. The incidence of hospital infections in the UK is decreasing (Health Protection Agency 2012); however the way media report on these types of infections may contribute to a sustained high perception of these risks, which in turn influences decision making about surgery.

Another media driven issue seen in the current study was that of women's concerns about the link between HRT and breast cancer, which had been widely reported in the media (Beral and Collaborators 2003). Additionally, the discussions with health professionals revealed that there is mounting confusion regarding the safety of HRT post-RRSO in women at risk of breast cancer and women who had breast cancer, even among health professionals. Few research studies to date have studied these groups specifically and more data are needed to make firm recommendations regarding the use of HRT in these patients. However, some studies have been published which confirm that short-term use of HRT up to the age of natural menopause in patients who have not had breast cancer themselves appears to be safe. These studies suggest that, given the long-term health effects of oestrogen deprivation, HRT should be recommended to patients who have surgery before the age of 45 (Eisen et al. 2005, Rebbeck et al. 2005). Furthermore, the study

linking HRT to breast cancer risk has now also been widely criticised (Shapiro et al. 2011a, b, Shapiro et al. 2011c, Shapiro et al. 2012). However, attitudes towards HRT remain cautious.

As the above discussion has shown, issues raised by both women and health professionals in this study need to be considered in the context of current developments. They may be subject to change as new evidence becomes available. However, the salient issues frequently discussed by women and health professionals in the current study relating to cancer risk, the menopause, HRT and ovarian screening are likely to remain important, even as evidence changes. Information provided to women by health professionals and/or through decision support materials regarding these issues will need to be updated on a regular basis to ensure women can base their decisions on the latest evidence. Furthermore, psychological factors (e.g. cancer worry), social factors (e.g. family history and obligations) and other factors (e.g. physician recommendations) are likely to play a role in decision making regardless of research developments. Therefore the results of this study provide a valuable insight into issues considered during decision making about RRSO despite the fact that some of those issues may shift in their relative importance or underlying information content.

4.4.3 Strengths and limitations

The current study combined the use of interviews with health professionals and focus groups with women at risk of ovarian cancer. This approach allows for a comprehensive and, at the same time, in-depth analysis of the decision making process. Women in the focus groups explored their personal situation and decision making in detail, providing the researcher with an in-depth understanding of individual's decision making. In contrast, health professionals drew on their experience of supporting a number of women to make decisions about RRSO, allowing the researcher to obtain a relatively comprehensive overview of issues that may be considered by women. This also enabled the researcher to distinguish common, salient issues mentioned by several health professionals and by women in

focus groups, from rarer considerations, which only played a role in a minority of women's decision making. A thorough understanding of both types of considerations may be important when developing patient decision support designed to cater for all women. Additionally, considerations which were explored by several members of the focus groups were confirmed as salient issues in interviews. This validates the conclusions drawn about the most important considerations.

Unsurprisingly, many considerations were discussed by health professionals, but not women themselves. These included prior cancer, perceived risk and fertility. There are several possible explanations for this. The first is that women in the focus groups had not experienced a certain issue, such as a diagnosis of breast cancer, or that a certain consideration was no longer relevant to them, such as completion of family. Hence, these were not considerations which played a role in their decision making. This may indicate that due to the low response rate to focus group invitations, women in the current study were not representative of the range of women who may face decisions about RRSO. However, it should be noted that representativeness is not a goal of qualitative research. Another reason for the lack of discussion of seemingly salient considerations by focus group participants may be that they assumed an issue to be obvious or self-evident. For example, ovarian cancer risk reduction was not mentioned specifically in focus groups as a consideration. However, it may have been interpreted by women as the main reason why they would consider surgery in the first place, therefore not warranting specific discussion (see Chapter 3). Alternatively, women may have been reluctant to discuss ovarian cancer risk reduction, as this issue may have been too emotionally charged. However, this is unlikely as women discussed various other emotional topics during the focus groups. As the topic guide for focus groups was designed to let participants explore issues freely and of their own accord, the lack of discussion of some seemingly salient considerations may also be due to the lack of prompting by the facilitator to draw out such issues. The final explanation may be a lack of awareness of the role of emotional or psychological factors. For

instance 'perceived cancer risk', which was mentioned by several professionals, was not discussed in focus groups. This may be due to the nature of 'perceived' risk being perceived by the individual as actual or objective risk. The fact that women did not mention 'perceived' risk may simply imply that they discussed this as their actual risk.

It may be argued that this lack of discussion of several issues highlights the limitations of focus groups with affected individuals, as their contributions are limited by their own personal circumstances and experiences. However, for this reason the current study included interviews with health professionals to complement the focus group data and to present a more holistic view of the decision making process.

Despite the inclusion of a range of health professionals from different areas and specialities in this study, additional professional groups, who were not included, could have made valuable contributions to this study. For example, GPs and their view of the decision making process regarding RRSO may have been an interesting group whose views are not included in the current study. Given women's comments about recommendations received from GPs, and the importance of trust between patients and their GPs, this appears to be a group that should be considered in future research. However, including all professional groups that were potentially relevant to this study was beyond the scope of the current research.

One further limitation should be noted. While the moderator was the same in all the focus groups, different individuals were fulfilling the task of moderator aid. During the initial discussion of decision making, the moderator aid was required to make notes of the points discussed by participants, which were then used as prompts in the ranking exercise. As moderator aids changed, it cannot be assumed that the same points were picked up consistently between groups, which may reduce comparability of the ranking exercise outcomes. However, the moderator

(Jana Witt) was present at each group to ensure that the salient and most frequently mentioned points were picked up and included in the ranking exercise.

4.4.4 Conclusions

This chapter explored decision making about RRSO in a UK context, postulated a model of preference construction and identified barriers and facilitators to decision making. In agreement with other research (see Chapter 3), the range of issues considered by women in the UK was large and varied depending on personal experiences and goals. Women reported a variety of recommendations given to them by different health professionals, which resulted in confusion about which option was best. Taken together with the fact that both women and health professionals felt that more information and details on various issues would be helpful in making a decision, this provides evidence that there is a place for decision support materials targeting women at increased risk of ovarian cancer in the UK who are making decisions about RRSO.

Using the issues highlighted by women and health professionals in this chapter, together with the results of the literature search (Chapter 3), the CODE framework will now be adapted to specifically describe the deliberation and coping processes relating to RRSO decisions in women at increased risk of ovarian cancer in the UK (see Chapter 5). This adapted framework will then act as a theoretical basis for the development of the content of a patient decision support intervention (Chapter 7). The model of preference construction, which emerged from the data, distinguishes two approaches to deliberations: a heuristic approach leading to an immediate or quick decision, and a deliberative approach leading to a more protracted decision. This suggests that a two part intervention, which has the ability to support heuristic as well as more deliberative decision making, may be most effective in ensuring that both groups of decision makers reach an informed decision. The development of such an intervention is described in Chapter 7.

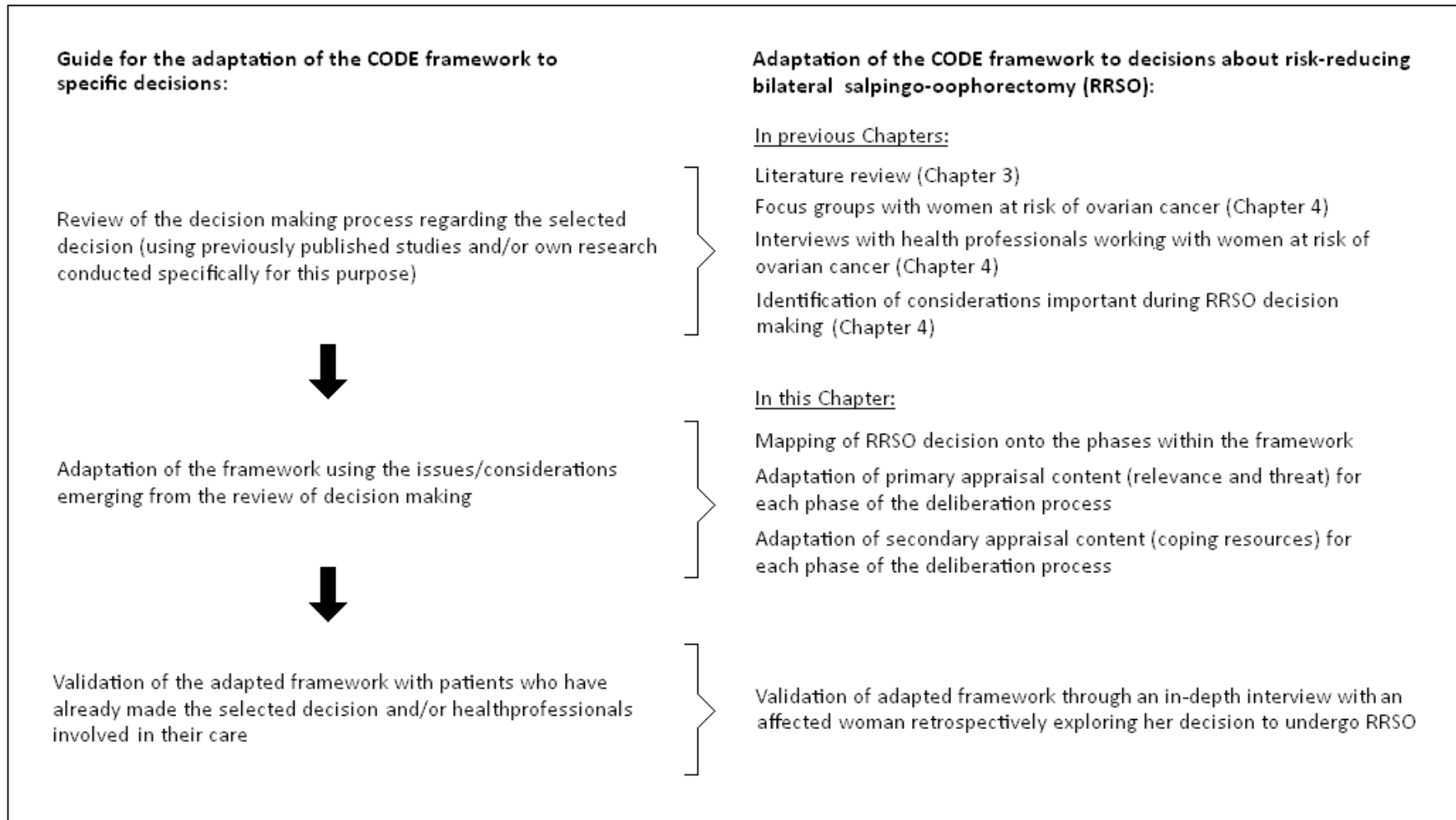
5 Adapting the CODE framework: Decisions about risk-reducing bilateral salpingo-oophorectomy

5.1 Introduction

Preference-sensitive medical decisions in situations of equipoise, when no medical treatment is clearly indicated or when treatments bear substantial risks, require the patient to play an active role in the decision making process. The preliminary Coping in Deliberation (CODE) framework attempts to describe deliberation and coping processes in such preference-sensitive situations from a patient perspective (Chapter 2; Witt et al. 2012). The framework was developed to be applicable to various situations and its flexibility allows adaptation to a number of different healthcare decisions. An example of a preference-sensitive medical decision is that of electing risk-reducing salpingo-oophorectomy (RRSO) to reduce ovarian cancer risk in women with a substantial family history and/or genetic mutation predisposing them to this cancer. As explained in Chapters 1 and 4, screening for ovarian cancer is not routinely available in the UK, as effectiveness has not been established (Menon and Jacobs 2002, Rosenthal et al. 2013a, Rosenthal and Jacobs 2006). Therefore, the only clinically recommended management option for women at increased risk of ovarian cancer is RRSO. As discussed in Chapters 3 and 4, the decision of whether and when to undergo surgery is complex and may be influenced by a number of internal and external factors. Thus, deliberation and coping processes undergone by women when making decisions about RRSO may be described using the CODE framework (Witt et al. 2012).

Following the recommendations for adaptation of the CODE framework outlined in Chapter 2 (Figure 2.5), the aims of this chapter are (i) to adapt the generic version of the CODE framework to specifically describe decisions about RRSO using findings from Chapters 3 and 4, and (ii) to map the adapted framework against a retrospective report of a decision about RRSO. Figure 5.1 outlines the framework adaptation process for RRSO decisions.

Figure 5.1 - Process of adapting the CODE framework to decisions about risk-reducing bilateral salpingo-oophorectomy in the context of genetic risk



5.2 Methods

5.2.1 *Data from literature search, focus groups and interviews*

The framework adaptation presented in this chapter is based on data presented in Chapters 3 and 4. Search strategy and inclusion/exclusion criteria for the systematic literature search were outlined in detail in Chapter 3 (see section 3.2). Factors important during decision making identified using this search strategy were also discussed in depth in Chapter 3 (see section 3.3). Similarly, recruitment to focus groups and interviews, and analysis of data collected, was described in detail in Chapter 4 (see section 4.2). The resulting themes and subthemes were also discussed in Chapter 4 (see section 4.3.2-6).

5.2.2 *Framework adaptation*

Items within the original CODE framework (Chapter 2) are generic and widely applicable. Items within adapted versions need to have a narrower, more defined focus in order to be relevant to the specific health threat and decision. The issues considered by women going through deliberations about RRSO in general (Chapter 3), and in the UK in particular (Chapter 4), were used to inform the adaptation of the preliminary CODE framework. The items within the framework were modified to specifically reflect issues that had been identified during the literature review and analysis of focus group and interview data. Additional items were added if they were supported by data and helped to portray appraisal processes in further detail. For instance, data indicated that, during primary appraisal of risk, women explore the reasons for their increased risk, especially with regards to their family history. Therefore, the generic question 'What caused this?' was adapted to: 'What caused me to have an increased risk of ovarian cancer?' and 'How does my family history influence my ovarian cancer risk?'. Any references to specific information or support needs made by women were also integrated into the items outlined in the appraisal sections of the framework. For example, women felt that evidence-based information and speaking to peers would be beneficial as potential coping resources. This was integrated into secondary appraisal sections in

the adapted framework by adding questions about the availability of further information and peer support. Some items within the generic framework did not appear relevant in the context of the RRSO decision after data analysis and were therefore not incorporated in the adapted version.

5.2.3 Face validation

The adapted CODE framework was mapped against an interview with one woman who had opted for RRSO with hysterectomy and retrospectively explored her decision. She was recruited through the Cancer Genetics Service for Wales in Cardiff by invitation from her genetic counsellor. Full written consent was obtained on the day of the interview. During the interview the woman explored in detail her decision making regarding risk-reducing surgery, including the coping resources she had used and the information she had gathered before her final decision. The interview schedule is included in Appendix 5.1. The interview began as a narrative interview with an open question regarding her decision making pathway (Friesen 2010, Kvale 1996). Prompts were available to the interviewer if required; however the woman was left to tell her story relatively freely. Towards the end of the interview a semi-structured approach was taken to discuss her needs for, and views of, decision support for women in her situation. The CODE framework was not used to guide the interview, as the patient was encouraged to explore her experience freely to minimise the influence of the research framework on her account. The RRSO-adapted CODE framework was then applied during analysis.

5.2.4 Data analysis

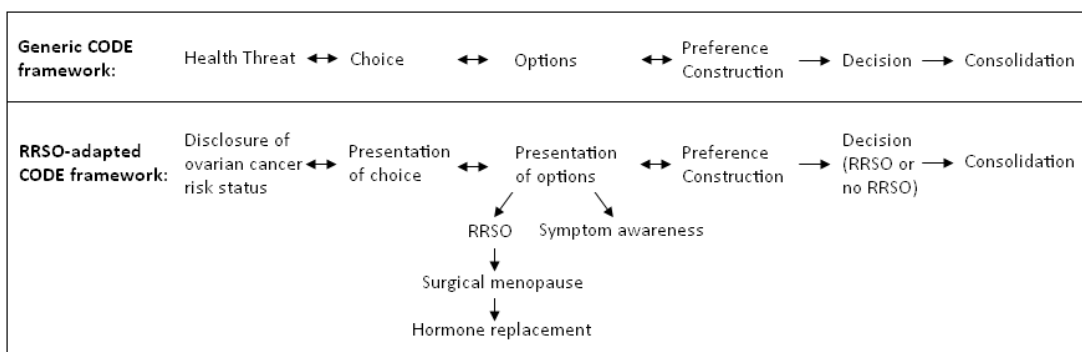
The interview was audio-recorded, transcribed and analysed thematically using a coding frame based on the adapted CODE framework (Appendix 5.2; Boyatzis 1998, Braun and Clarke 2006, Joffe and Yardley 2004). In this way, the adapted CODE framework was mapped against the experience of the patient to examine whether it adequately described the questions and coping options explored by the patient during her appraisal and deliberation process. A sample of the coded transcript is included in Appendix 5.3.

5.3 Results

5.3.1 Mapping of the RRSO decision onto the CODE framework

The CODE framework describes specific events associated with the process of preference-sensitive decision making as ‘deliberation phases’. It further proposes that each phase gives rise to a primary and secondary appraisal process and coping response. Patients at risk of ovarian cancer may experience several potentially stressful ‘events’ or go through potentially stressful ‘phases’ while deciding how to manage their risk of ovarian cancer. These can be mapped onto the deliberation phases delineated in the CODE framework. Figure 5.2 summarises how the phases in the generic framework can be adapted to decisions about RRSO.

Figure 5.2 - Deliberation phases in the generic and adapted CODE framework



The first event of interest when considering preference-sensitive decision making is the *health threat*, in this case the disclosure of an elevated risk of ovarian cancer to the patient. Cancer risk may be appraised as a threat, as it has the potential to result in significant future loss or harm (Lazarus and Folkman 1984). The perceived level of threat depends on the patient’s past experiences, goals and values. For instance, someone who cared for a relative suffering from ovarian cancer and witnessed them dying of the disease may conceive their own risk as a high level threat, whereas a woman who only knows of a distant relative who recovered from ovarian cancer might view it as a lower level threat (Dagan and Goldblatt 2009, Howard et al. 2011). Depending on the perceived level of threat, the patient may differentially appraise the coping resources needed to manage their ovarian cancer risk.

A further event in the deliberation process is the *presentation of choice*, which may be appraised as a threat or challenge, depending on the woman's experiences, values, level of health literacy and perception of control or self-efficacy (Bandura 1977, Lazarus and Folkman 1984). For example, someone who has high self-efficacy and believes in their ability to make good decisions might appraise the fact that they are being asked to play an active role in decision making as a challenge. In contrast, someone with low self-efficacy, who feels they have insufficient medical knowledge to make this decision, might appraise being asked to contribute to a decision about their health as a threat. Preferences for involvement in decision making will vary accordingly. Coping resources will again vary depending on whether choice is appraised as a threat or challenge (Lazarus and Folkman 1984).

The options available in case of increased genetic risk of ovarian cancer are RRSO and symptom awareness, as screening is currently not recommended. The *presentation of these options* may also be distinguished as a specific 'event' or phase in the deliberation process in which the patient is required to appraise each option in turn, including its risks and benefits, as well as the potential impact on her life. Options themselves may be appraised as threats. For example, RRSO bears the potential for future loss or harm due to surgical risks as well as the lack of oestrogen following surgery. Similarly, rejecting or deferring surgery means that cancer risk remains high, potentially resulting in future loss or harm in the form of ovarian cancer. Some of the individual outcomes of RRSO are very complex and may be appraised as threats in their own right, such as surgical menopause and the subsequent need for hormone replacement.

Preference construction is an essential phase in every deliberation process. At this point the patient differentiates one option as superior to the other(s) and begins to form a preference, which may later translate into a decision (Svenson 1992). At this point the potential option will be mapped onto personal and important others' goals and values, further details of the preferred option will be explored and timing considered. This phase may be perceived as threatening, as the decision maker is nearing a final decision and has to face losing the freedom of

making a different choice, particularly if the preferred option is irreversible. Once preference construction is complete, a decision for or against RRSO is made and may be *consolidated* (Svenson 1992).

Overall, the events occurring during consultations about increased genetic risk of ovarian cancer and associated clinical management options can readily be mapped onto the deliberation phases in the preliminary CODE framework (see Figure 2.4, Chapter 2). Therefore, the questions within the generic framework can subsequently be adapted to represent questions specific to the RRSO context arising in response to each phase or event during deliberations about this surgery.

5.3.2 *The threat - adapting primary appraisal content*

Primary appraisal describes the process of cognitively and emotionally appraising an event's meaning, relevance and level of threat (see section 2.4.2 Chapter 2; Lazarus and Folkman 1984). According to Leventhal's (1980) self-regulatory model of illness representations, cognitive appraisal explores the identity of the threat, its causes, timeline, consequences and opportunities for control (Diefenbach and Leventhal 1996, Lau et al. 1989, Leventhal et al. 1984, Weinman et al. 1996). In other words, during primary appraisal individuals examine what the threat is, what it means to them, what caused it, how long it will last and how it might change over time, what consequences they are likely to experience (including the potential severity of such consequences and impact on all aspects of life) and whether and how they might be able to control the threat (Weinman et al. 1996). Questions regarding possibilities for control might initiate secondary appraisal of coping resources available to manage the threat (see section 5.3.3).

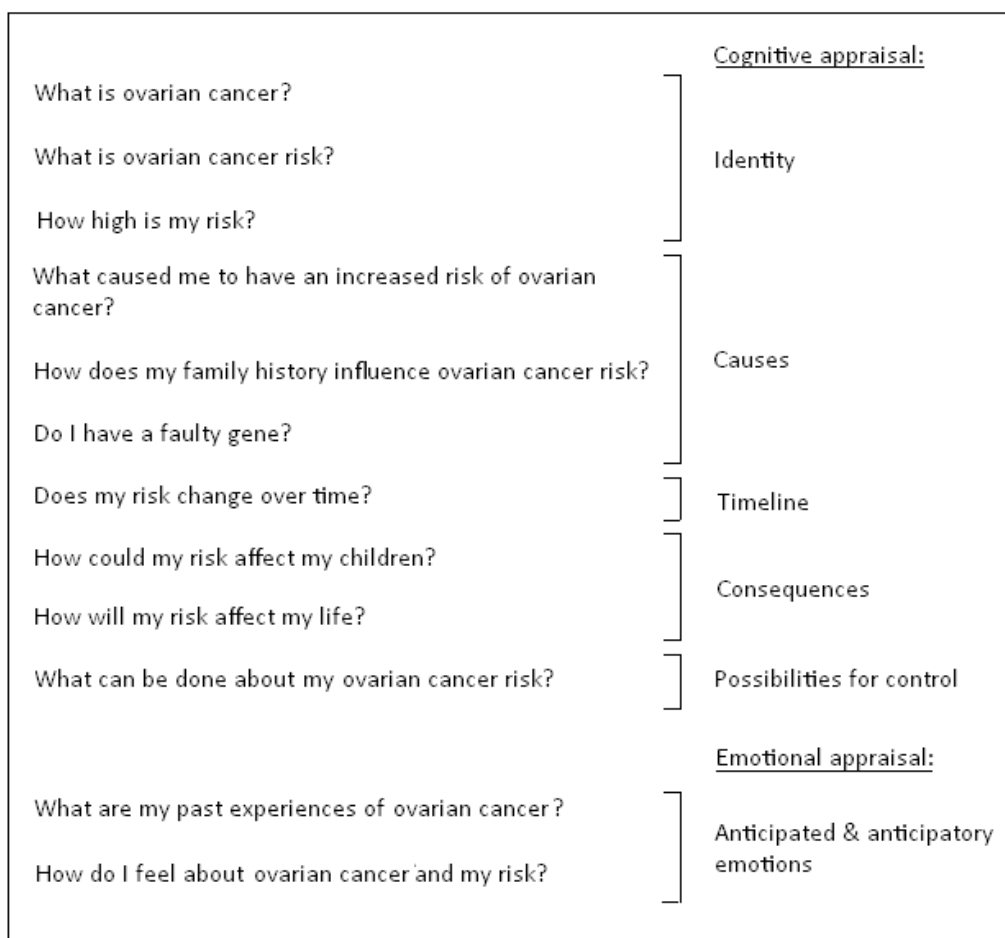
Emotional appraisal is guided by anticipatory and anticipated emotions induced by the threat and can influence cognitive appraisal processes (Loewenstein et al. 2001, Power et al. 2011). Anticipatory emotions are those emotions experienced in response to the threat itself, whereas anticipated emotions are those which the decision maker predicts they will experience if certain future

events linked to the threat occur. Both types of emotions may be based on past experiences and beliefs, such as perceived self-efficacy.

As discussed in section 5.3.1, there are four potentially stressful ‘events’ which require appraisal by patients during deliberations about RRSO: disclosure of ovarian cancer risk status, presentation of choice, presentation of options and preference construction. Below, each of these phases is considered in turn and the considerations important during the phase, as they emerged from the data presented in Chapters 3 and 4, are expressed in the form of questions.

5.3.2i Disclosure of ovarian cancer risk status: The first phase is comprised of a discussion about the patient’s ovarian cancer risk, which is influenced by her family history and genetic status. Figure 5.3 depicts the specific questions a patient might explore in response to being told she is at increased risk of ovarian cancer.

Figure 5.3 - Considerations during primary appraisal of increased genetic ovarian cancer risk



The identity of ovarian cancer and ovarian cancer risk may be explored by patients using their own ideas about the label 'ovarian cancer' and the nature of this disease, as well as information provided by the health professional and acquired elsewhere (Sivell et al. 2008). Risk is an abstract concept, which again will be appraised using the patient's view of what risk is and any information provided in consultations (Kelly et al. 2005). The clinical level of risk (average or 'population', moderate or high) will play a role in the appraisal process and perceived level of risk may be influenced by personal beliefs, experiences and external influences (Kelly et al. 2005, Lancaster et al. 2011). For instance, as found in the literature search and in focus groups and interviews, a previous cancer diagnosis or having witnessed ovarian cancer in a close relative were identified as important factors in the decision and may influence how patients perceive their own risk during primary appraisal (Babb et al. 2002, Dagan and Goldblatt 2009, Hallowell et al. 2001, Hamilton et al. 2009, Stuckey et al. 2010).

The causes of the increased risk may be explored in terms of the patient's family history and potential genetic mutation. Patients may also consider lifestyle or environmental factors that could influence their risk. This exploration of ovarian cancer risk was evident in the focus groups and interviews, as health professionals and women emphasised the importance of exact risk estimates, ideally with confirmation of a genetic mutation. Evidence from the literature search also confirmed the importance of information about gene mutation status during deliberations (Antill et al. 2006, Culver et al. 2011, Kram et al. 2006, Madalinska et al. 2005, Schwartz et al. 2003, Uyei et al. 2006, Verhoeven et al. 2011). Some patients in the focus groups felt that, if their increased risk was due to a genetic mutation, then they would be able to make a clear cut decision, as this would change their appraisal of their risk. Previous research, and health professionals in this study, found that a patient's interpretation or perception of their risk may influence decisions. This indicates that patients may base their appraisal of risk on past experiences and beliefs, rather than objective estimates (Finch et al. 2009, Lerman et al. 1995). Beliefs about ovarian cancer and perceived levels of risk may

result in emotions, such as worry and anxiety, as part of the emotional appraisal process (Culver et al. 2011, Howard et al. 2011, Lancaster et al. 2011).

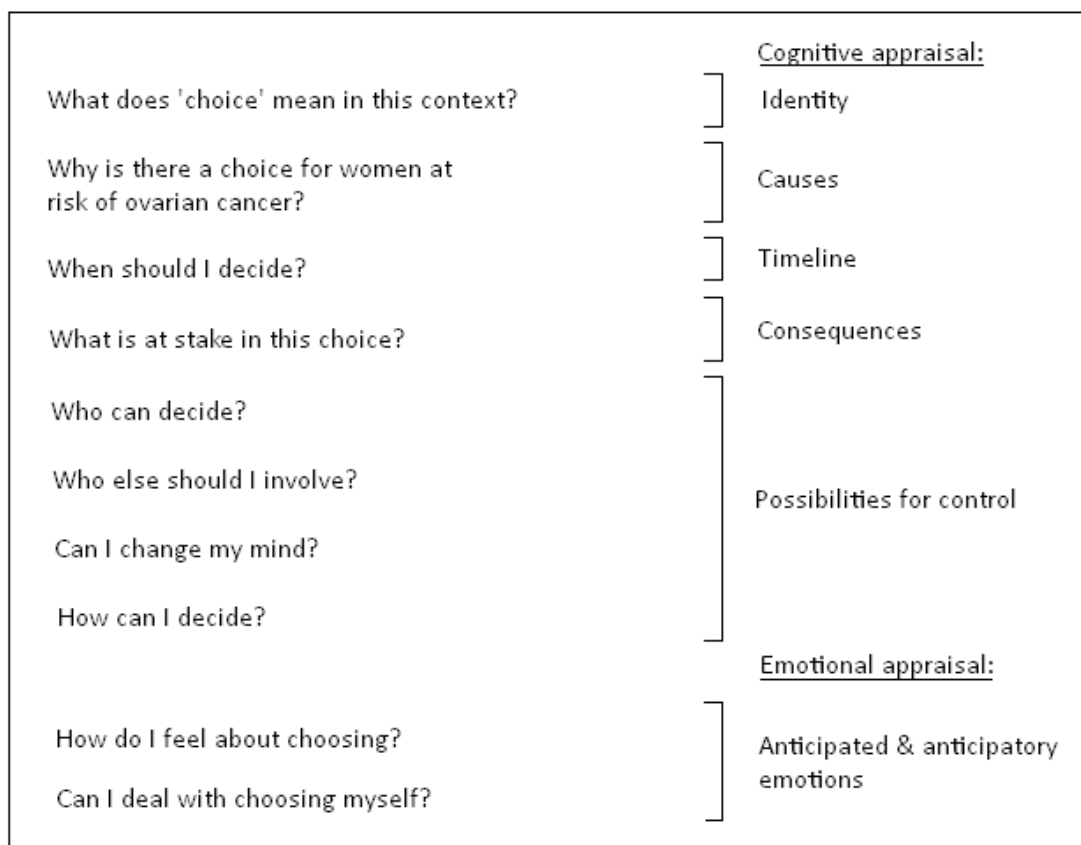
Patients may explore the timeline and consequences of their cancer risk, for example how their risk might change over time and how their risk will affect their own and their offspring's life (MacDonald et al. 2010). Health professionals involved in RRSO decisions stressed that women younger than 35 were at a relatively low risk and should not have surgery at this age; however as age increases, so does the cancer risk, and therefore professionals emphasised the importance of considering RRSO around age 40-45. In the focus groups women discussed the effects that the knowledge of their risk status has had on their life and described the lack of understanding from people in their surroundings. Women were also concerned about the effects of their own risk status on their children, both in terms of the possibility that they may develop cancer and no longer be able to fulfil their parental role, as well as in terms of the children's cancer risk. Evidence from the literature search indicated that parity may be associated with higher uptake of RRSO, which could be due to women's desire to be there for their children in the future (Skytte et al. 2010). Emotional appraisal of the cancer threat will depend on the patient's knowledge about, and past experiences of, ovarian cancer. Finally, during primary appraisal of cancer risk, women may also ask what they can do to reduce their risk, and this will prompt a discussion of choice and options.

5.3.2ii Presentation of choice: The health professional will introduce the idea that there is more than one option and may initiate a discussion about choice. Figure 5.4 delineates the questions a patient might explore in the context of increased ovarian cancer risk when faced with the fact that there is a choice and that they are being asked to play an active role in decision making. The identity of the choice, i.e. the meaning of 'choice' in the context of increased genetic risk of ovarian cancer, is determined by what the woman understands by the term 'choice' and by the number and characteristics of options available. Some women might view this decision as relatively simple and immediately prefer one option over the other, as was mentioned by health professionals, who spoke about women being

able to make a clear cut decision almost immediately due to a prior cancer diagnosis, past experiences of ovarian cancer or hospital phobia (Chapter 4). For these women a detailed appraisal of the choice may not be necessary.

Others may struggle more with the concept of choice in this context and find it difficult to settle for one option. This was illustrated by women in the focus groups, who had not been able to make a decision due to a number of barriers. For these women detailed appraisal of the choice may be necessary in order to prepare for decision making. They may want to know the cause for the choice, i.e. why there is no clear medical recommendation. They may explore the timeline of the choice to determine at what point in time a decision should or could be made (Howard et al. 2010c). Additionally, they may explore the possibilities for control over the choice by considering who (apart from themselves) could make or be involved in the choice, whether the choice is reversible or not and how they should approach this choice.

Figure 5.4 - Considerations during primary appraisal of the choice



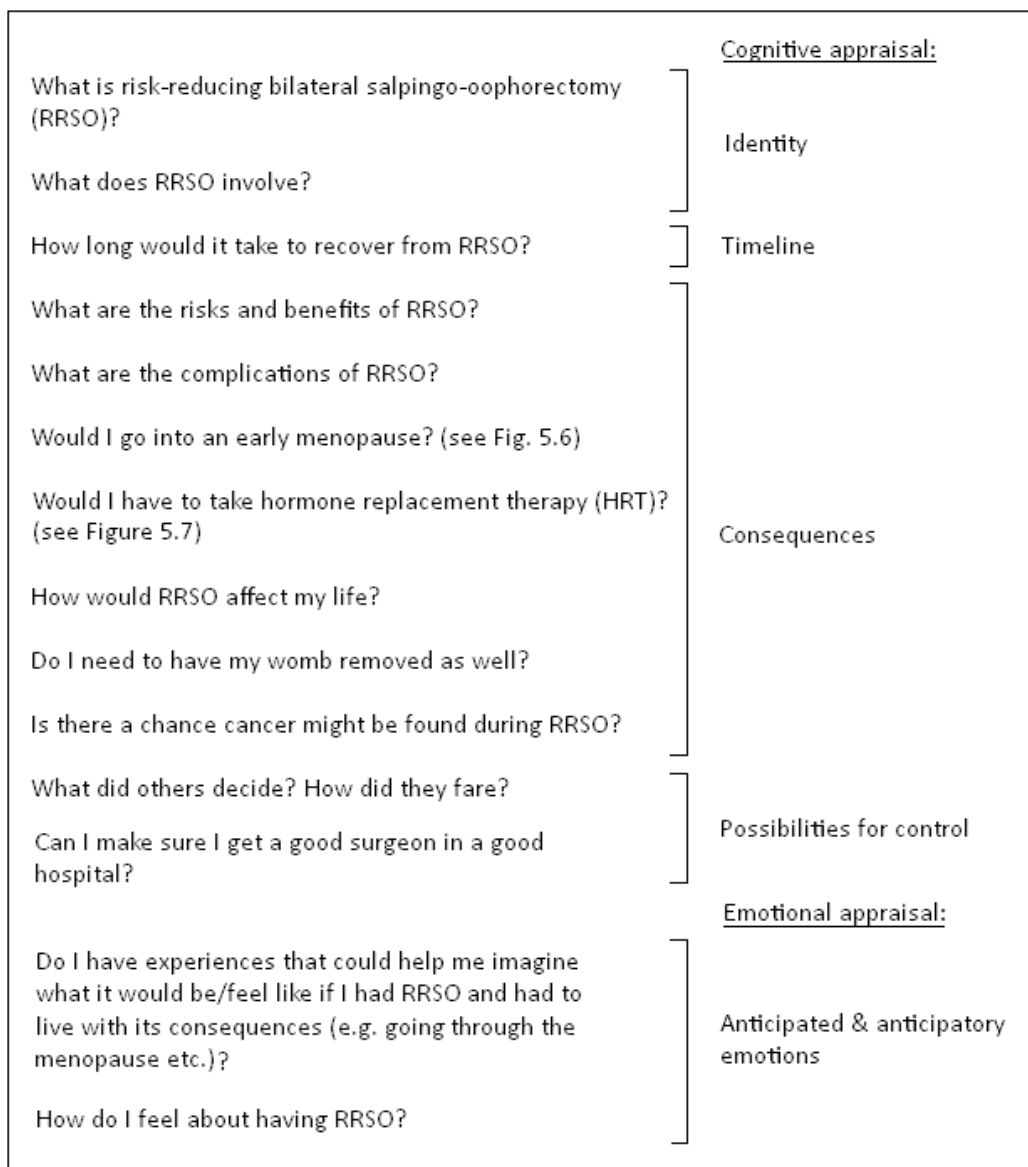
Furthermore, women may want to explore the potential consequences of making a choice and the effects those will have on their lives, such as the loss of fertility following RRSO. This will prompt a discussion of the available options and their attributes. Emotionally, the availability of choice may instil anticipatory emotions, such as anxiety about having to make a difficult decision, as well as anticipated emotions, such as an anticipated feeling of regret if one makes the wrong decision.

Considerations that play into how choice is appraised in this context include a woman's trust in the medical profession and recommendations by clinical staff. As discussed by women and health professionals, trust was an issue during decision making and may therefore also influence how women view the choice and their role in it. Similarly, recommendations from clinical staff, especially those who are strongly favouring one option, may also influence appraisal of choice; in the presence of a strong recommendation, choice may no longer seem relevant (Claes et al. 2005, Fang et al. 2002, Kram et al. 2006, Swisher et al. 2001).

5.3.2iii Presentation of options: The appraisal of options is very complex and therefore may be split into a number of sub-phases for each option. In the context of increased genetic risk of ovarian cancer, the options are RRSO and 'symptom awareness'. During the appraisal of RRSO, the decision maker may additionally need to cognitively appraise the possibility of an early menopause and of whether or not to take hormone replacement. Therefore the primary appraisal processes for the presentation of options phase presented below are split according to these different considerations.

The *surgery* itself may be appraised as a threat due to surgical risks and the impact it has on a patient's life. Figure 5.5 details questions that may be explored during primary appraisal of the surgery option. The identity of RRSO may be explored in terms of what this surgery is and what it involves. The cause necessitating RRSO would be the woman's increased risk of ovarian cancer; therefore there is no detailed exploration of the cause within the presentation of options phase. Women in the focus groups discussed RRSO as being 'elective surgery' and found it difficult to make the conscious choice for surgery when they

Figure 5.5 - Considerations during primary appraisal of risk-reducing ovarian surgery



were feeling healthy and well, appraising this as an extreme step. Health professionals emphasised that it is a major surgery and has a number of risks.

The timeline for RRSO may be explored in terms of its disruptive impact on the patient's life. Women in the focus groups discussed how surgery might affect familial or work obligations, such as going on the school run or carrying out one's job. Various such obligations may impact on women's appraisal of RRSO as a viable option (Bradbury et al. 2008, Brain et al. 2004, Howard et al. 2010c). Further, a detailed exploration of the consequences of surgery is necessary to fully appraise

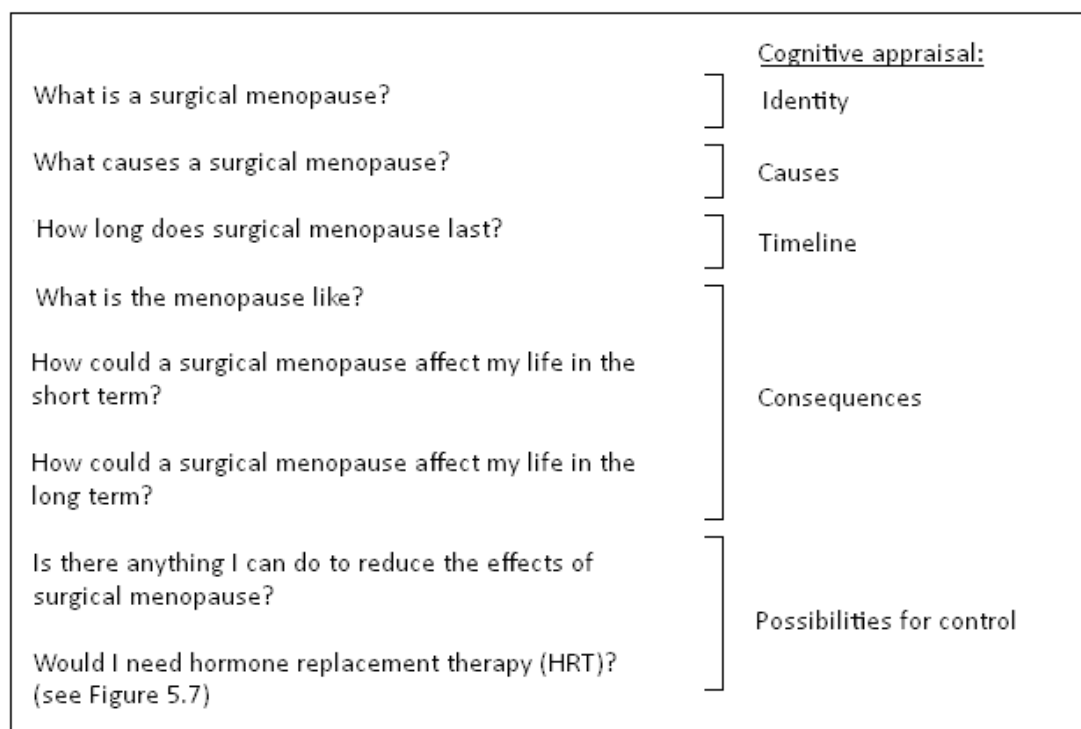
this option. In this context women may explore the risks and benefits of RRSO, possible complications, after effects and impact on their life. Women in the focus groups discussed surgical risks, particularly the likelihood of hospital infection, as well as the impact surgery may have on their family and/or career. Evidence from the literature search also highlighted risks of surgery as an important consideration (Brain et al. 2004, Fang et al. 2003, Hallowell 1998, Hurley et al. 2001). Furthermore, women may also discuss the possibility of, or need for, a hysterectomy with RRSO and may explore the likelihood of a cancer diagnosis during surgery, as was mentioned by health professionals.

In terms of possibilities for control, women may explore the scope for influencing where the operation takes place, i.e. a local versus specialist hospital unit, and linked to this, the surgeon's expertise, in order to reduce the risks of surgical complications and hospital infections. This was evident in women's discussions regarding possible locations for surgery and reputation of different hospitals and surgeons during focus groups. Women may also seek to find out how others, who have already made a decision, fared, in order to adjust their own appraisal of what to expect from RRSO, which was evident from enquiries about and discussions of peer-support.

Finally, RRSO will also be appraised emotionally based on past experiences, personal goals and values. Women may have relatives or friends who have previously had good or bad experiences of RRSO that could influence the woman's own emotional appraisal. For instance, someone who has seen a relative struggle to adjust after surgery might be more fearful than someone whose relative had a very good surgical experience and strongly recommends RRSO. Someone with hospital phobia may react with extreme anxiety in response to learning about RRSO, whereas someone who is very scared of cancer might react with relief when the option of RRSO is presented. Emotional appraisal will also include exploration of how one feels about the short- and long-term consequences of RRSO, such as the surgical menopause and the option of HRT, if appropriate.

For women who are considering RRSO before the age of the natural menopause, oestrogen deprivation following surgery is a major concern, as was clear from data obtained in the literature search, interviews and focus groups (Brain et al. 2004, Fang et al. 2002, Hallowell 1998, Ray et al. 2005, Swisher et al. 2001). Therefore, appraisal of *surgical menopause*, as part of the consequences of RRSO, may result in a number of questions (Figure 5.6). The identity, causes and timeline of a surgical menopause may be explored to understand what a surgical menopause is, why RRSO will result in a menopause and how long it would be expected to affect one's life. Women in the focus groups were especially concerned about the consequences of a surgical menopause, its potential effects on their body and their mental state in both the short- and long-term. Health professionals confirmed that the menopause was a major concern for most premenopausal women and an important consideration in terms of bone and cardiovascular health.

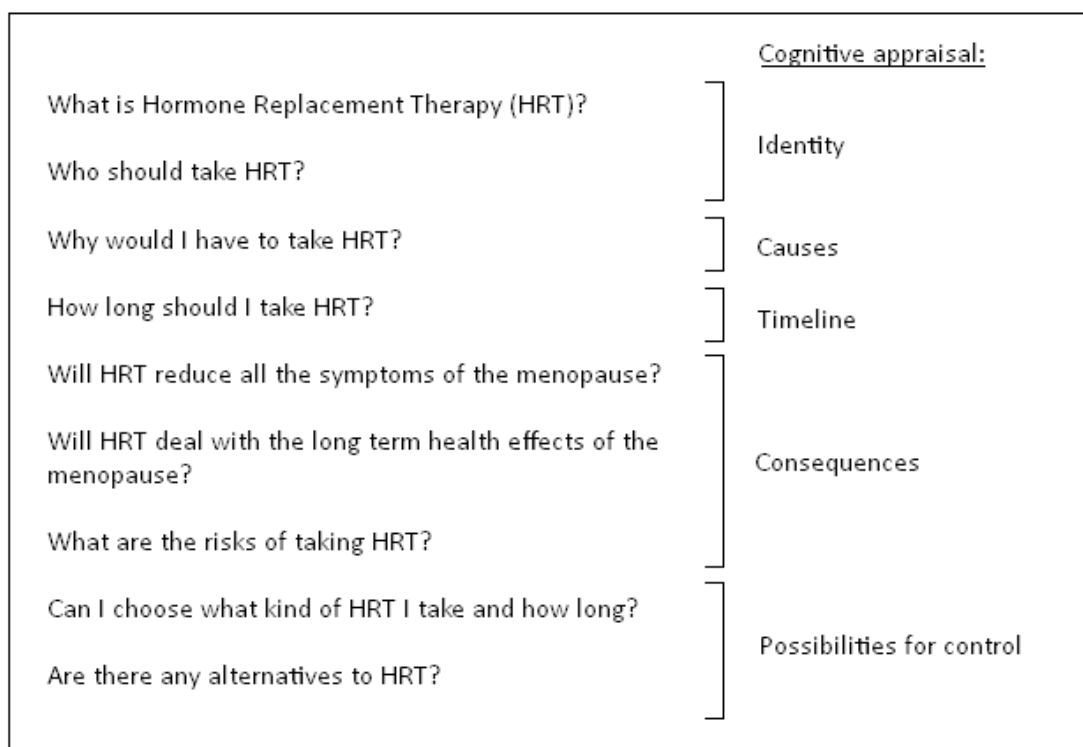
Figure 5.6 - Considerations during cognitive appraisal of surgical menopause*



*As appraisal of the surgical menopause is part of the appraisal of RRSO and its outcomes, emotional appraisal processes have not been outlined here, as these form part of the appraisal of the RRSO option

Possibilities for controlling the effects of a surgical menopause may also be explored during appraisal and may depend on perceived self-efficacy, i.e. the belief that one can cope with these effects, and coping resources. Options for control that might be considered include psychological and medical interventions, primarily *hormone replacement therapy (HRT)*. Similar to the surgical menopause, deliberations about HRT are complex and therefore may also result in a number of more detailed questions (Figure 5.7). Evidence from the literature search also highlighted questions and concerns about HRT as a major consideration during deliberations about RRSO (Fang et al. 2002, Hallowell et al. 2001, Swisher et al. 2001). Women may explore what HRT is and who would be eligible to take it, especially with a view to their own family history of breast cancer. Women in the focus groups voiced concerns about the potential for HRT to increase breast cancer risk which had been reported in the media. Women might also explore why HRT is recommended for premenopausal women and how long they would have to take it.

Figure 5.7 - Considerations during cognitive appraisal of hormone replacement therapy*

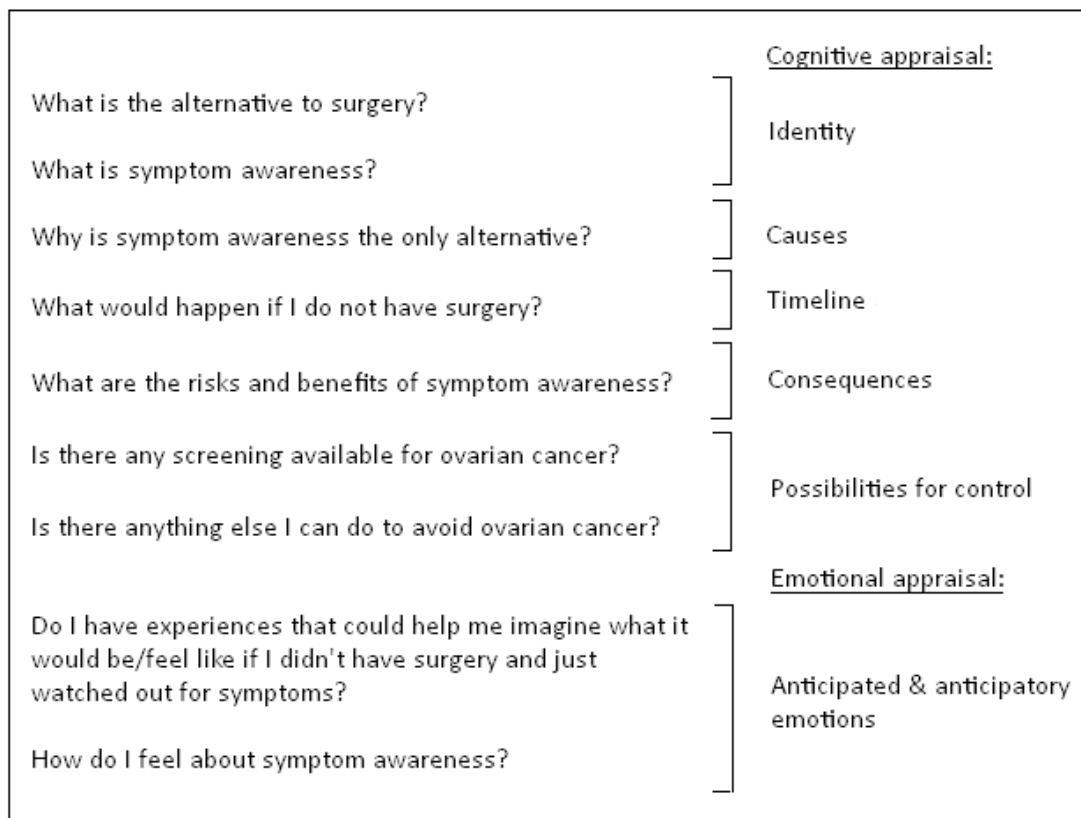


*As appraisal of HRT is part of the appraisal of RRSO and its outcomes, emotional appraisal processes have not been outlined here, as these form part of the appraisal of the RRSO option

In terms of the consequences, women may explore the risks and benefits of HRT and its potential to counteract and control the symptoms and long-term health consequences of a surgical menopause. As there are different types of HRT, women may also consider what method of delivery best suits their needs and whether they can influence the type of HRT and potentially the length of treatment they receive. Women in the focus groups were also keen to discuss alternatives to HRT, which they could administer themselves, therefore exploring options of controlling menopausal symptoms without HRT.

Finally, the second available option for women at increased genetic risk of ovarian cancer is to *reject RRSO* and become aware of the symptoms of ovarian cancer. This alternative also requires an appraisal process (Figure 5.8). Women may first explore what the alternatives to surgery are and upon being informed that the only clinically recommended alternative is symptom awareness, they may then consider this option in more detail, including looking into why this is the only alternative, what might happen if they rejected surgery and whether there might be

Figure 5.8 - Considerations during primary appraisal of alternatives to RRSO

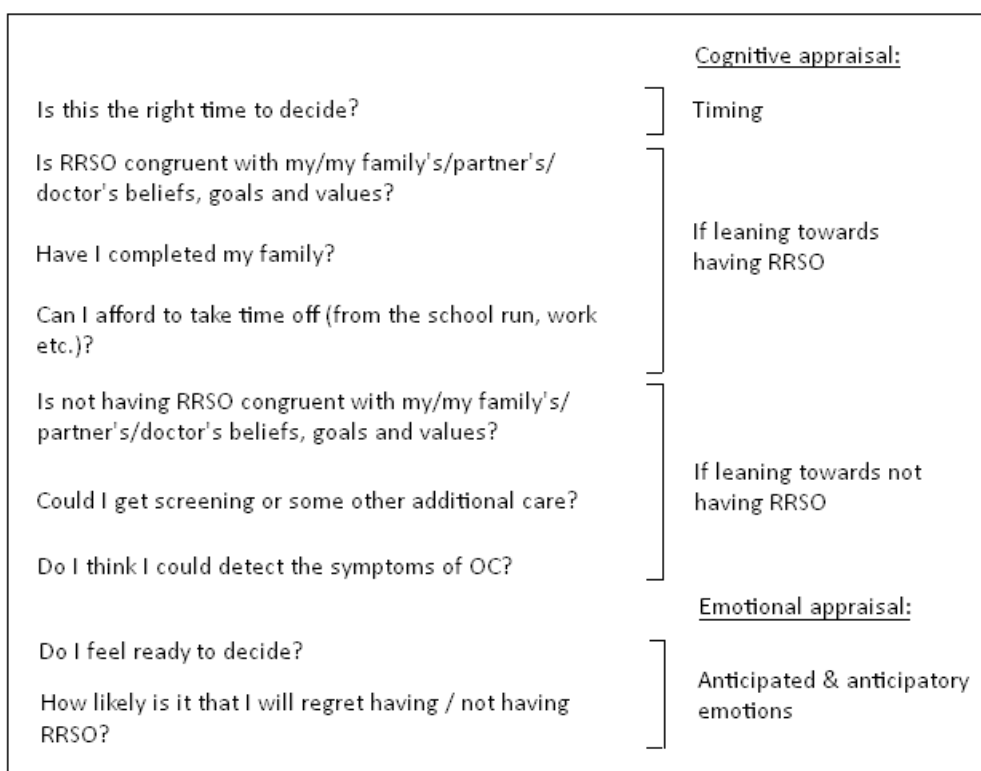


anything else they could do. Women in the focus groups discussed the possibility of privately funded screening and considered whether this might be an option. Health professionals also noted that women frequently enquired about screening and other alternatives to surgery. Evidence from the literature search also showed that belief in, and availability of, alternatives to RRSO were important considerations (Brain et al. 2004, Fang et al. 2002, Howard et al. 2011, Madalinska et al. 2007).

Similar to emotional appraisal of RRSO, the emotional appraisal of symptom awareness depends on past experiences, the patient's knowledge of symptoms of ovarian cancer and self-efficacy beliefs regarding the ability to detect those symptoms (Brain et al. 2004, Fang et al. 2002).

5.3.2iv Preference Construction: The final phase before decision determination is not readily split into the five aspects of cognitive appraisal suggested by Leventhal et al. (1980). However, during this phase patients explore important questions regarding timing and value-adjustment, before making a final choice. Questions which may be explored in this phase are suggested in Figure 5.9.

Figure 5.9 - Considerations during primary appraisal of the preferred option



Considerations which might impact on the appraisal of timing during preference construction include major life events, age and work, as well as familial, commitments. As discussed by women and health professionals, the relative importance of ovarian cancer depends on other events and responsibilities in a person's life and timing of the decision has to be right (Howard et al. 2010c). Age played a role in timing as well, as the consequences of surgery differ for different age groups. For example, a postmenopausal woman does not need to consider menopause and HRT, whereas a thirty year old may be deemed too young to have this surgery due to loss of fertility and the long-term health effects of oestrogen deprivation.

Women may also explore the congruence of a selected option with their own and important others' goals and values. In the case of surgery, one such important goal is the completion of family planning. RRSO will be at odds with a wish to have children, or more children, and therefore not congruent with an important personal goal (Culver et al. 2011, Fang et al. 2002, Fang et al. 2003, Skytte et al. 2010). The input from important others, such as partners, mothers or sisters, might also be sought at this point to seek their approval and prepare for the chosen option together. Additionally, physician recommendation may influence this part of the appraisal process, as congruence of the chosen option with a GP's or surgeon's recommendation can be reassuring for patients (Claes et al. 2005, Kram et al. 2006, Swisher et al. 2001). Practical aspects, such as ability to book time off work for the surgery or knowledge of ovarian cancer symptoms, may also be appraised at this point.

During emotional appraisal in preference construction, the woman may explore whether she feels ready to decide and attempt to predict possible regret following the decision (Connolly and Reb 2005). If a chosen option is at odds with one's own or an important other's goal or value, then this phase can be emotionally stressful. For example, if a woman is extremely anxious about cancer but has not yet completed her family, she may choose to delay surgery. This option is at odds with her desire to reduce her cancer anxiety; however it is in line with her goal of

having a child. Therefore, her emotional appraisal of any option will be difficult, as she will anticipate regret with either option. The final decision will depend on which goal is more important to her at that moment in time.

5.3.2v Consolidation: The generic CODE framework includes a consolidation phase following the decision, which has not been discussed here. The review of decision making regarding RRSO presented in Chapter 3 and the study of women and health professionals reported in Chapter 4, which act as the basis for this adaptation, focus on the pre-decisional process of appraisal of the threat and differentiation of options (Svenson 1992). An adaptation of the questions associated with these processes will allow the creation of an RRSO-adapted CODE framework which describes pre-decisional deliberation. This may act as a guide for consultations and decision support materials designed to facilitate decision making. In contrast, post-decisional consolidation occurs once a decision has been made to further differentiate the chosen option and thereby avoid regret (Svenson 1992). Hence, an adaptation of the consolidation phase was not included in the current chapter, as it is not relevant to materials that aim to support pre-decisional differentiation, such as patient decision support interventions.

5.3.3 Coping resources - adapting secondary appraisal content

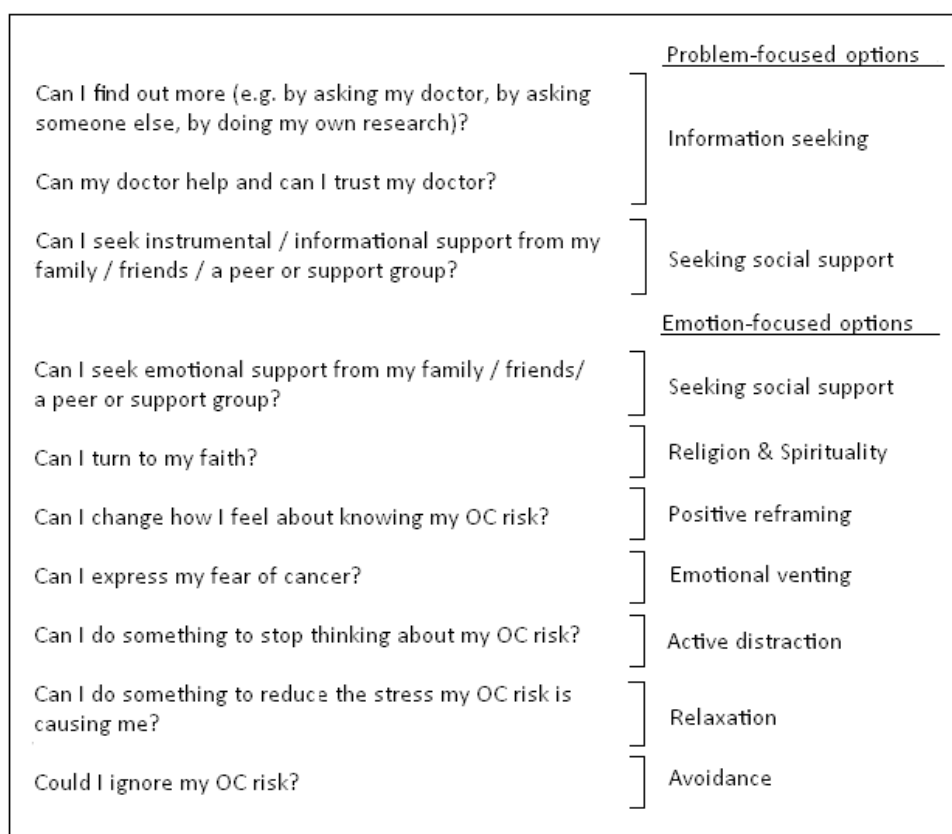
During secondary appraisal the individual appraises the coping potential (i.e. whether anything can be done) and options (i.e. what can be done) to manage a stressful event (see section 2.4.3, Chapter 2; Lazarus and Folkman 1984). Potential coping options that might be implemented in response to the threat may be defined as problem- or emotion-focused, depending on whether their primary aim is to deal with the threat directly or to regulate emotions experienced in response to the threat (Lazarus and Folkman 1984).

As previously described, there are four potentially stressful phases during deliberations about RRSO: disclosure of ovarian cancer risk status, presentation of choice, presentation of options and preference construction. Below, these phases are considered in terms of the secondary appraisal they stimulate, with a view to

exploring potential coping strategies that might be appraised by patients going through these phases. Secondary appraisal during the presentation of options and preference construction is represented as one process (section 5.3.3iii), as these two phases are closely linked and coping options explored during these phases are interchangeable.

5.3.3i Disclosure of ovarian cancer risk status: Once women are aware of their risk, they may begin to appraise the coping potential and options at their disposal to manage their risk and their emotional response to that risk. Firstly, they may assess whether there is potential for dealing with the threat directly and/or for regulating emotions. Then they may further appraise individual coping options, such as seeking information, seeking social support and using avoidance, in order to find those best suited to deal with the current threat (Figure 5.10). Potential coping options considered initially may include some that have not been appraised as currently available or suitable. These will then not be used as a resource.

Figure 5.10 - Secondary appraisal of coping options in response to disclosure of cancer risk

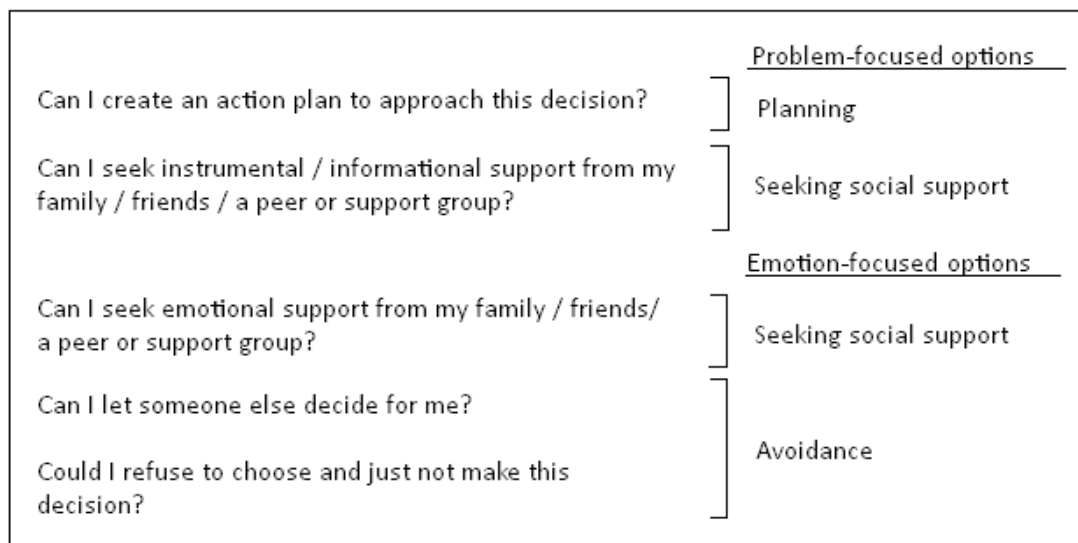


If a woman appraises that she can take a direct approach to dealing with the threat, she might explore possibilities for obtaining further information through communicating with others, such as her doctor, family or peers (Babb et al. 2002). If she also appraises that there is the need for and potential to regulate emotions in response to the threat, she might explore the possibility of finding comfort in her faith or using distraction or relaxation techniques (Pergament 1997, Phelps et al. 2006). For example, women in focus groups explored the possibilities for obtaining information and mentioned issues around trusting clinical staff. Health professionals noted that peer support would be a useful resource for women. Many situations, such as cancer risk, will be appraised as having problem-focused as well as emotion-focused coping potential and some coping options (e.g. seeking social support) may be problem- and emotion-focused at the same time, as indicated in Figure 5.10.

5.3.3ii Presentation of choice: When presented with the fact that there is choice, and that personal goals and preferences play a role in this decision, women may appraise the coping options available to help them deal with making the decision (Figure 5.11). As discussed earlier, the availability of choice may be appraised as a threat or a challenge during primary appraisal, depending on personal factors, such as self-efficacy (DeWalt et al. 2007). Depending on the outcome of this appraisal, the coping options considered during secondary appraisal may vary. Someone who appraised choice as a challenge may be more likely to consider options for approaching the decision and resources of instrumental and informational support, focusing on those coping options that directly deal with the choice. In contrast, someone who appraised choice as a threat may explore options for emotional support through social contacts or may consider avoidance options, such as transferring responsibility for the decision to someone else.

Women in the focus groups discussed their feelings about recommendations they had received and whether they could trust health professionals to provide good information and make the right decision for them (Babb et al. 2002). They also

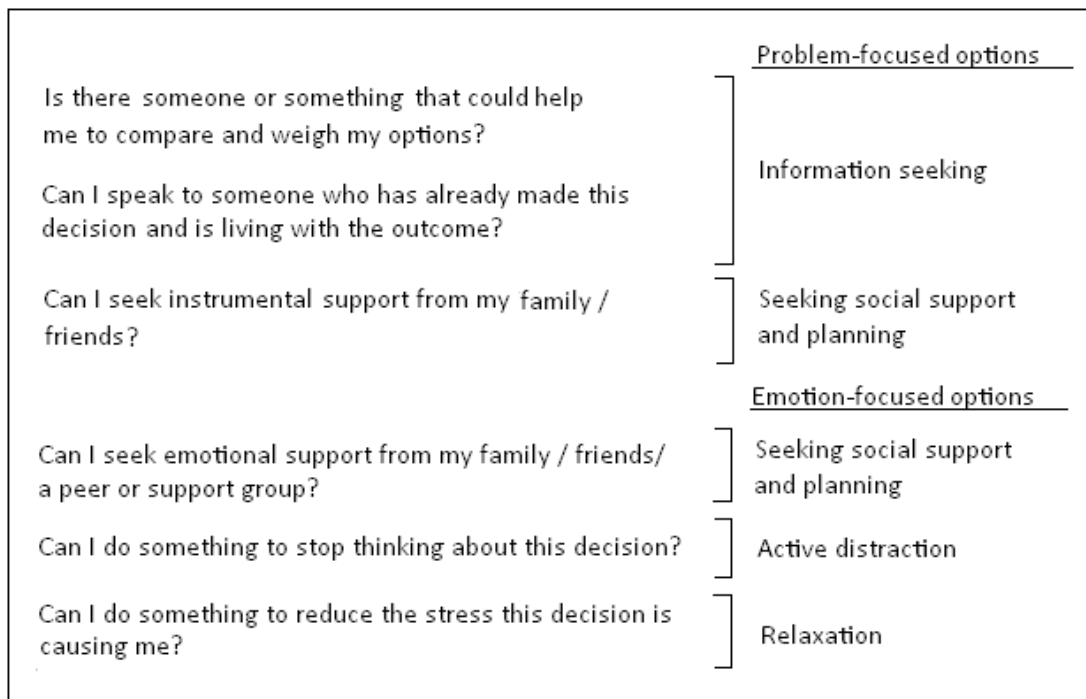
Figure 5.11 - Secondary appraisal of coping options in response to presentation of choice



explored the possibility of deferring the choice. In contrast, health professionals emphasised the importance of involving others, such as partners, and of having a personal time plan when approaching this decision (Howard et al. 2010c).

5.3.3iii Presentation of options and preference construction: During secondary appraisal of options and preference construction, patients may assess whether there is potential for dealing with the threat posed by the options directly or for regulating emotions in response to the options presented (Howard 2010). Patients may then explore individual coping options that could help them make and implement the decision and subsequently deal with its outcomes (Figure 5.12). For example, a woman who appraised that there are ways she can deal with the threat directly may explore weighing her options and seek to speak to a peer who has already made this decision (Babb et al. 2002, Kenen et al. 2007). A woman who has appraised that there are ways she can regulate her emotions might consider whether her options include social support or whether she might be able to employ distraction or relaxation techniques (Phelps et al. 2006). Instrumental and emotional support from a partner during and after decision making was an important issue discussed by women and health professionals in focus groups and interviews, respectively. Participants also discussed how best to compare options and whether there was any way of facilitating the differentiation process.

Figure 5.12 - Secondary appraisal of coping options in response to presentation of options



5.3.4 The RRSO-adapted CODE framework

The individual appraisal processes outlined in sections 5.3.2 and 5.3.3 were combined and summarised to form the content of an RRSO-adapted CODE framework that specifically describes deliberations and coping in the context of RRSO decisions (Figure 5.13). The preliminary framework includes a wide range of possible issues. The exact questions explored and the problem- and emotion-focused coping options that will be appraised, selected and used will vary depending on the individual decision maker and her personal situation. Patients may consider only a few or many of the questions and coping options proposed. They may explore questions in a similar order to which they are listed in the framework, or they may jump between phases and questions in a non-sequential manner.

The RRSO-adapted CODE framework, therefore, is not necessarily a model that accurately describes a selected patient's deliberation pathway or predicts which questions will be asked and coping options will be selected by which patient.

Figure 5.13 - The RRSO-adapted CODE framework

| Deliberation | | | | | |
|--------------|--|---|--|---|--|
| | Disclosure of ovarian cancer risk status | Presentation of choice | Presentation of options (RRSO or symptom awareness) | Preference construction | |
| Coping | Cognitive | <p>What is ovarian cancer (OC)?</p> <p>What is OC risk?</p> <p>How high is my risk?</p> <p>What caused me to have an increased risk of OC?</p> <p>How does my family history influence OC risk?</p> <p>Do I have a faulty gene?</p> | <p>What does 'choice' mean in this context?</p> <p>Why is there a choice for women at risk of OC?</p> <p>When should I decide?</p> <p>What is at stake in this choice?</p> | <p>What is risk-reducing bilateral salpingo oophorectomy (RRSO) / symptom awareness?</p> <p>Why is symptom awareness the only alternative?</p> <p>What does RRSO involve?</p> <p>How long would it take to recover from RRSO?</p> <p>What would happen if I do not have surgery?</p> <p>What are the risks and benefits of RRSO / symptom awareness?</p> <p>What are the complications of RRSO?</p> <p>Would I go into an early menopause?</p> <p>Would I have to take hormone replacement?</p> <p>How would RRSO affect my life?</p> <p>Do I need to have my womb removed as well?</p> <p>Is there a chance cancer might be found during RRSO?</p> <p>Is there any screening available for OC?</p> <p>Is there anything else I can do to avoid OC?</p> <p>What did others decide? How did they fare?</p> <p>Can I make sure I get a good surgeon in a good hospital?</p> | <p>Is this the right time to decide?</p> <p>Is RRSO / not having RRSO congruent with my/my family's/partner's/doctor's beliefs goals and values?</p> <p>Have I completed my family?</p> <p>Can I afford to take time off?</p> <p>Could I get screening or some other additional care?</p> <p>Do I think I could detect the symptoms of OC?</p> |
| | Primary Appraisal | <p>Does my risk change over time?</p> <p>How could my risk affect my children?</p> <p>How will my risk affect my life?</p> <p>What can be done about my OC risk?</p> | <p>Who can decide?</p> <p>Who else should I involve?</p> <p>Can I change my mind?</p> <p>How can I decide?</p> | <p>Do I need to have my womb removed as well?</p> <p>Is there a chance cancer might be found during RRSO?</p> <p>Is there any screening available for OC?</p> <p>Is there anything else I can do to avoid OC?</p> <p>What did others decide? How did they fare?</p> <p>Can I make sure I get a good surgeon in a good hospital?</p> | <p>Do I feel ready to decide?</p> <p>How likely is it that I will regret having / not having RRSO?</p> |
| | Emotional | <p>What are my past experiences of OC?</p> <p>How do I feel about OC and my risk?</p> | <p>How do I feel about choosing?</p> <p>Can I deal with choosing myself?</p> | <p>Do I have experiences that could help me imagine what it would be/feel like if I had / didn't have RRSO?</p> <p>How do I feel about having / not having RRSO?</p> | |
| | Problem-focused | <p>Can I find out more?</p> <p>Can my doctor help and can I trust my doctor?</p> <p>Can I seek instrumental / informational support from my family / friends / a peer or support group?</p> | <p>Can I create an action plan to approach this decision?</p> <p>Can I seek instrumental / informational support from my family / friends / a peer or support group?</p> | <p>Is there someone or something that could help me to compare and weigh my options?</p> <p>Can I speak to someone who has already made this decision and is living with the consequences?</p> <p>Can I seek instrumental / informational support from my family / friends / a peer or support group?</p> | |
| | Secondary Appraisal | <p>Can I seek emotional support from my family / friends / a peer or support group?</p> <p>Can I turn to my faith?</p> <p>Can I change how I feel about knowing my OC risk?</p> | <p>Can I seek emotional support from my family / friends / a peer or support group?</p> | <p>Can I seek emotional support from my family / friends / a peer or support group?</p> <p>Can I do something to stop thinking about this decision?</p> <p>Can I do something to reduce the stress this decision is causing me?</p> | |
| | Emotion-focused | <p>Can I express my fear of cancer?</p> <p>Can I do something to stop thinking about my OC risk?</p> <p>Can I do something to reduce the stress my OC risk is causing me?</p> <p>Could I ignore my OC risk?</p> | <p>Can I let someone else decide for me?</p> <p>Could I refuse to choose and just not make this decision?</p> | | |

Rather, it offers a dissected view of deliberation and coping processes that was constructed by an analytical process, using input from a number of stakeholders. It is a model that helps to identify the kind of support that women may require while going through the deliberation pathway. It therefore acts as a framework to help those who strive to facilitate decision making, in consultations or otherwise, to provide sufficient information, support affective forecasting and encourage coping. Hence, the RRSO-adapted CODE framework is a conceptual framework that may guide the development of support methods and materials for women at increased genetic risk of ovarian cancer.

5.3.5 *Face validation*

The RRSO-adapted CODE framework was validated through an interview with a carrier of a mutation in BRCA1 who had undergone RRSO with a risk-reducing hysterectomy in the preceding six months and had been premenopausal at the time of surgery. The patient described issues considered during decision making as well as coping strategies used throughout her deliberation process. The issues explored whilst going through the deliberation process described by the patient in this interview map onto many of the questions delineated in the RRSO-adapted CODE framework (Figure 5.14).

5.3.5i Ovarian cancer risk status: When appraising her *cancer risk*, the patient used her experience of observing ovarian cancer in her mother and breast cancer in her aunt to appraise the level of threat. Her appraisal of her own risk was high, as both her mother and aunt had died of cancer. While she acknowledged that there was a genetic component to the disease, she also felt that stress was a causal factor. This initial appraisal maps onto the questions delineated in the framework about ovarian cancer, risk, causes, family history and past experiences.

...my mum and my aunt they've had cancer after a really traumatic time in their life and [...] I just think that maybe there just must be some form of a link [...] I know that's not in any way [...] scientific but I don't know, I just think stress seemed to be [...] a crucial part of things in our family...

Figure 5.14 - Questions within the RRSO-adapted CODE framework explored by one patient (in red and bold)

| Deliberation | | | | |
|--------------|--|---|--|---|
| | Disclosure of ovarian cancer risk status | Presentation of choice | Presentation of options (RRSO or symptom awareness) | Preference construction |
| Coping | Cognitive | <p>What does 'choice' mean in this context? Why is there a choice for women at risk of OC? When should I decide? What is at stake in this choice?</p> | <p>What is risk-reducing bilateral salpingo oophorectomy (RRSO) / symptom awareness? Why is symptom awareness the only alternative? What does RRSO involve? How long would it take to recover from RRSO? What would happen if I do not have surgery? What are the risks and benefits of RRSO / symptom awareness? What are the complications of RRSO? Would I go into an early menopause? Would I have to take hormone replacement? How would RRSO affect my life? Do I need to have my womb removed as well? Is there a chance cancer might be found during RRSO? Is there any screening available for OC? Is there anything else I can do to avoid OC? What did others decide? How did they fare? Can I make sure I get a good surgeon in a good hospital?</p> | <p>Is this the right time to decide? Is RRSO / not having RRSO congruent with my/my family's/partner's/doctor's beliefs goals and values? Have I completed my family? Can I afford to take time off? Could I get screening or some other additional care? Do I think I could detect the symptoms of OC? Do I feel ready to decide? How likely is it that I will regret having / not having RRSO?</p> |
| | Primary Appraisal | <p>Who can decide? Who else should I involve? Can I change my mind? How can I decide?</p> | <p>Do I have experiences that could help me imagine what it would be/feel like if I had / didn't have RRSO? How do I feel about having / not having RRSO?</p> | |
| | Emotional | <p>What are my past experiences of OC? How do I feel about OC and my risk?</p> | <p>How do I feel about choosing? Can I deal with choosing myself?</p> | |
| Coping | Problem-focused | <p>Can I create an action plan to approach this decision? Can I seek instrumental / informational support from my family / friends / a peer or support group?</p> | <p>Is there someone or something that could help me to compare and weigh my options? Can I speak to someone who has already made this decision and is living with the consequences? Can I seek instrumental / informational support from my family / friends / a peer or support group?</p> | |
| | Secondary Appraisal | <p>Can I find out more? Can my doctor help and can I trust my doctor? Can I seek instrumental / informational support from my family / friends / a peer or support group?</p> | <p>Can I seek emotional support from my family / friends / a peer or support group? Can I turn to my faith? Can I change how I feel about knowing my OC risk? Can I seek emotional support from my family / friends / a peer or support group? Can I do something to stop thinking about this decision? Can I do something to reduce the stress this decision is causing me?</p> | |
| | Emotion-focused | <p>Can I express my fear of cancer? Can I do something to stop thinking about my OC risk? Can I do something to reduce the stress my OC risk is causing me? Could I ignore my OC risk?</p> | <p>Can I do something to stop thinking about this decision? Can I do something to reduce the stress this decision is causing me?</p> | |

She went on to describe how her emotional reaction to her aunt's cancer led her to avoid taking action to manage her own risk initially and select an avoidant coping strategy, until another event in her life reminded her of the importance of confronting her cancer risk.

...my aunt [...] lived with breast cancer [...] for eight years and she died three years ago. So after she died, me and my cousins were saying: "We really need to get back involved in the genetic counselling" [...]. But actually we didn't do anything about it, because I think to be honest it was just so awful, it was such an awful time [...]. I think we just thought: "This is just too awful to contemplate really." And then I found a lump in my breast and [...] if it wasn't for that lump in my breast maybe I might have just put my head in the sand for a little while longer...

This describes coping options explored and employed during and after appraisal of the health threat: while she first explored whether she could ignore the information, and subsequently chose avoidance to cope with the emotions in response to her aunt's death, she later came back to the decision and re-evaluated her coping options, concluding that she wanted to take action and could find out more by contacting the genetics service.

There, she was offered genetic testing for a BRCA gene mutation. The patient described her struggle with this decision, as she had already appraised her cancer risk as very high and she feared that her worries would remain, even if no mutation was found.

...first of all I decided I wasn't going to have the tests but I wanted to have a hysterectomy [...]. I know that sounds a bit all round the wrong way but [...] in my head [I] thought if I haven't got the gene obviously that would be brilliant but I'll still want to have a hysterectomy because [...] I don't trust my own mind that I'll be able to relax...

Therefore, she initially evaluated her cancer risk considering both possibilities, by talking "a lot about statistically how likely it is that I would [develop ovarian cancer] if I was BRCA1 or not BRCA1". However, she did eventually recognise the importance of confirming the genetic mutation for her children and wider family and decided to go

ahead with testing following a relative's decision to get tested. This process maps onto the exploration of cancer risk in terms of genetics and effects on offspring as detailed in the CODE framework.

...[...] they found that she [cousin] did have the BRCA1 mutation and [...] I think talking to her made me think: "Actually, what am I doing? I need to know really for my boys and I need to know for our family." [...] So then a year ago I went along and had the test and so then found out that I did have the BRCA1 mutation...

Confirmation of the mutation resulted in elevated anxiety and made her want to press ahead with surgery urgently. However, she did not have surgery until one year after receiving her test result and described how taking the time to consider the impact of the confirmed mutation, as well as carefully familiarising herself with the benefits and risks of surgery, was important before committing to the operation.

...the first panic [...] when you find out you've got the BRCA1 just makes you think: "Right, get me in, I'll have it all done, just quick as a flash [...]". But actually I think you really do need a certain amount of time just to come to terms with it all and [...] make sure you do know the in's and out's of what the operation means and what it means afterwards and what the BRCA1 gene means with regards to yourself and your whole family...

During these deliberations, she turned to support from her family to help her cope and was particularly close to her cousin, who also carried a mutation in the BRCA1 gene and was making decisions about risk-reducing options at the same time. This maps onto the coping options of instrumental, informational and emotional support from family and friends outlined in the CODE framework. While the patient acknowledged that it was sometimes difficult for others to understand her worries and she didn't "want people to think that you're just trying to make something out of nothing", she emphasised repeatedly how support from relatives helped her to cope with her cancer risk, and indeed the decision making process.

...I do speak to my cousin about it, so obviously we're all totally on the same page and my dad and our outside family are very supportive [...], but I think [...] it's harder for people who haven't directly seen the effects of ovarian and

breast cancer [...] to understand perhaps where we're coming from [...]. There is the mind set of: "Why don't you just enjoy your life and see what happens?" and I can understand that, because I've sort of thought that myself: "Let's just [...] put our heads down and just enjoy life [...]". It's just whether you really can do that or whether it would just always be at the back of your mind...

5.3.5ii Presentation of choice: The patient described how she had suspected that she was at an increased risk of cancer since her mother's diagnosis, and had already at that point started to contemplate risk-reducing surgery.

...For me, it's not been something that I've suddenly had to make a decision on [...]. I had my mum and talked about it with her and then [...] came to it sort of fairly gradually...

Therefore, the discussion of choice and being asked to play an active role in the decision making process came as no surprise when introduced by the genetic counsellor. Hence, this patient did not explore many of the questions outlined in the choice section of the CODE framework in any detail. However, she did describe her feelings of empowerment in response to being given the opportunity to decide how she would manage her cancer risk.

...obviously it would be much better if nobody had to worry about any of these things but that's not life, is it? [...] It is better to confront things head on, because this way we're getting [...] the control to be able to do what we can [...]. If you've got a bit of control about it [...] it really can [relieve] anxieties...

Describing herself as a 'worrier' she felt that any action she could take to reduce her risk of cancer would be worthwhile.

...I know what I'm like as a person and I'm a worrier and I know that I would just be always second guessing any problem that I had [...]. This is because of what happened to mum and I just would always be thinking it [...]. I do think that the mind plays such a massive role in this...

5.3.5iii Presentation of options: When discussing her surgical options, due to her concerns about cancer, the patient considered a full risk-reducing hysterectomy (RRH) with bilateral salpingo-oophorectomy, instead of RRSO alone.

...we talked about should I just have an oophorectomy or should I have the hysterectomy and I just felt that if I was going to have surgery, I might as well limit any other risks that might be there...

During appraisal of the option of RRSO plus RRH, she reported having considered many of the issues included in the CODE framework, such as risks and benefits of surgery, surgical menopause, hormone replacement, potential complications and residual risk.

...I was obviously concerned about [...] any complications and of course there is always a risk [...] with any kind of surgery...

...the other [...] concern obviously was HRT [...] and we talked about the problems of not being on HRT and we also talked about [...] the link with breast cancer [...] and the fact that really everything should be OK [...] until I was at natural menopausal age anyway. So that definitely was something that I was concerned about...

...I know that [...] there is a risk with regards to the stomach lining [...], but what I'm doing is limiting my risk and if something still happens then, well, I just know that I've done absolutely everything I can and then I've got the peace of mind and that's what I was really needing I think. [...] I think I'm just of the opinion that if I'm still going to get it, then [...] that is what is supposed to be [...] the course of my life...

While she remembered that these concerns were addressed fully and to her satisfaction at the genetics service, she acknowledged that contact with a peer who had already made the decision for surgery would have been a valuable resource. This indicates that she would have liked to seek instrumental, informational and/or emotional support from a peer to help her make her decision and to explore questions such as 'What did others decide?' and 'How did they fare?', although this option was not available to her at the time.

...I think a person to talk to would be brilliant [...] to help you clarify your own thoughts [...] against somebody who's actually been through the surgery [...]. I haven't been offered anything up here, having said that I'm not lacking anything [...]. Obviously it would be good to talk to somebody who'd been through the operation but [...] the genetic counsellors are absolutely fantastic...

5.3.5iv Preference construction: Linked to her description of herself as a ‘worrier’, the patient considered the reduction in cancer risk (which would most likely result in a reduction in her cancer worry) as the most important factor in her preference construction for surgery.

...the most important thing is the impact on life [...]. You will feel less worried about developing ovarian cancer. And then everything else that you have to cope with is absolutely fine, because if you can just limit your worries there then you will put up with anything [...] to a degree of course but [...] the balance is: not having to worry about ovarian cancer...

Drawing on her past experience of cancer, she continued to explain that having surgery would enable her to look after her children and that worries about other issues, such as the risks of surgery or subsequent oestrogen deprivation, appeared small in comparison. This shows her exploration of personal values and goals (e.g. seeing her children grow up) during preference construction.

...if I can do anything [...] then I would because I don't want my children to have to go through what I went through [...] and obviously because I want to be there...

...[I] probably wasn't really that worried about it all I think. That's because I'd just seen how awful it was with my mum and then how awful it was with my aunt and then any operations are not going to come close to those kinds of things...

The patient explained that she “was lucky [...] I was already married and had the children and I was pregnant when I was offered the test” and therefore did not have to consider the issue of family planning. This addresses the question of whether her family was complete, as outlined in the CODE framework.

During the final stages of decision making she then also described how practicalities, such as childcare and work obligations, played an important role in planning whether it was the right time and whether she was ready for surgery.

...The main things I think were practical issues of how long I was going to be in [hospital] for just from [...] the point of view of who was going to have the

children, how long I would be off work for [...] and just the kind of expectations of that...

...He [the consultant] just said: "You guys think about when is going to be practically speaking a good time for you and then we'll slot you in". So I said: "Well, actually October would be good", because [...] my oldest son was starting school and I wanted him to start and be settled before I had the op and I knew that we could rely on some grandparents to come and stay at that time...

As she had throughout her deliberation process, she used emotional and instrumental support from her family to cope with preference construction, decision making and the time after decision making. However, she did feel that others in her surroundings found it hard to understand why she wanted to have surgery. She explained how she had based her final decision on the information she had been given about her increased risk.

...It is such [...] an unusual thing to be deciding to do [...]. Sometimes [I] think: "Gosh, am I just going mad here even considering these things?" [...] People might say to you: "You wouldn't like just chop your hand off just because you might [...] get cancer in your little finger [...]". In a way they've got a point, you can't go round chopping off bits of your body [...], but at the same time [...] the statistics are so high and the evidence is so stacked that way that [...] that's what's making me make the decision, it's not just on a whim...

Finally, she explained how taking her time to approach the decision and considering the impact her cancer risk and surgery might have on her life was an important process to go through in order to make an informed, value-adjusted decision.

...when it was time for me to [...] sign on the dotted line that I was going to have it done I just felt absolutely fine about it [...]. I didn't feel I'd been pushed into anything, I'd been given all of the statistics, I'd had time to digest it all and discuss it all and talk to [husband] about it all [...] and that was [...] really important. So I didn't feel concerned at all once I'd made my decision...

Overall, this patient explored a wide range of issues and coping options outlined in the RRSO-adapted CODE framework during her deliberation process (Figure 5.14). However, some issues and coping options were not explored. For instance, she did not

appraise symptom awareness as an option in any detail, as she was relatively focused on surgery. She also did not report considering or using emotional coping options, such as religious coping or relaxation techniques. As discussed earlier, the questions in the RRSO-adapted CODE framework that are explored in any given deliberation process will vary depending on the individual decision maker and her personal situation. However, while patients may vary in their experiences, beliefs, values and personal goals, and therefore in the exact issues they will explore, the overall framework appears to describe the general deliberation process and associated appraisals relatively accurately. Additionally, this demonstrates that a single woman may explore many of the questions proposed in the framework when appraising the deliberation phases and her coping options.

5.4 Discussion

5.4.1 The RRSO-adapted CODE framework in context

The framework presented in this chapter represents the first adapted version of the CODE framework and describes in detail the deliberation process about RRSO by women at increased genetic risk of ovarian cancer.

The generic CODE framework was developed based on a number of prominent decision making and coping theories, which informed the structure and the questions amenable to adaptation within the framework. Theories included Svenson's (1992) differentiation and consolidation theory, Power's (2011) model of cognitive-emotional decision making, Leventhal's (1980, 1984) Self-Regulatory Model, Balneave and Long's (1999) embedded decisional model of stress and coping, and Lazarus and Folkman's (1984) transactional theory of stress, appraisal and coping. The adaptation of the CODE framework was therefore also guided by these theories.

In the past, Leventhal's (1980) Self-Regulatory Model was mainly applied to illness cognition in the context of chronic diseases, such as diabetes (Scollan-Koliopoulos et al. 2005) or HIV (Reynolds et al. 2007). However, some have also used this model to explore patients' representations of cancer risk (Decruyenaere et al.

2000, Kelly et al. 2005, Lancaster et al. 2011). In agreement with these studies, the current adaptation of the CODE framework using the Self-Regulatory Model demonstrates that representations of illness threats, rather than chronic conditions, may also be formed using the five dimensions of identity, cause, timeline, consequences and control.

The issues explored, and questions posed, by women (Chapter 3 and 4) during appraisal of ovarian cancer and their risk fit readily within these five dimensions. Furthermore, these dimensions could also be used to describe cognitive appraisal of the meaning of choice and management options, both of which could be seen as threats and were explored by women in a similar fashion in order to form representations of the meaning of choice and individual options. While this does not propose that Leventhal's dimensions may be used to describe cognitive appraisal of any condition or option, it shows that representations of illness, illness threats or other threats (such as treatment options) may be described using these five dimensions, which may be useful in future adaptations of the CODE framework to other decisions.

The RRSO-adapted framework includes many of the questions important to women making this decision as determined by previous research highlighted in Chapter 3. These include questions exploring the meaning of ovarian cancer and elevated risk (Howard et al. 2011, Meijers-Heijboer et al. 2003, Ray et al. 2005), the effects of surgery, surgical menopause and HRT (Brain et al. 2004, Fang et al. 2002, Hallowell 1998), the availability of support (Howard et al. 2011, Mellon et al. 2009) and the timing of surgery (Howard et al. 2010c). Therefore, the framework combines the knowledge accumulated in previous research on women's deliberations about RRSO with a study of women's decision making (Chapter 4) to propose a detailed patient-centred description of this process.

Previously, Howard et al. (2011) proposed the 'Preserving-the-self' theory to describe decision making about management options in carriers of mutations in BRCA1/2, which includes breast screening, RRSO and risk-reducing mastectomy. The

'Preserving-the-self' theory postulates that being at risk threatens four aspects of the 'self': physical health, self-identity, relationships and emotional well-being, and that women's decision making is geared toward preserving these aspects (Howard et al. 2011). Through interviews, Howard et al. (2011) explore women's deliberations and propose seven decision making approaches, such as 'weighing the pros and cons', 'taking time', 'paying attention to emotions' and 'engaging with others'. Combinations of these approaches then describe five distinct decision making styles: Snap, Intuitive, Deliberative, If-Then and Deferred style (Howard et al. 2011).

The RRSO-adapted CODE framework sits well alongside the 'Preserving-the-self' theory, as it proposes questions that patients may explore during deliberations. Howard et al. (2011) did not discuss specific questions and issues women explore during deliberations, but rather focused on the process, identifying individual decision-making approaches and styles. As previously discussed (Chapter 2), the questions within the CODE framework may differ between and can be adapted to individuals, depending on their preferred decision making and/or coping style. Therefore, the approaches and styles identified by Howard et al. (2011) may be used to determine which questions might be most important to a certain individual. For instance, a woman may select approaches representative of the intuitive decision making style, which include looking inward and reflecting on emotions (Howard et al. 2011). In this case, the questions in the framework focusing on emotional responses to the threats and emotional coping options would be most relevant. In contrast, if a woman uses approaches representative of the deliberative style, which include making sense of the numbers and weighing the pros and cons, questions relevant to cognitive appraisal and problem-focused coping, such as information seeking, might be more relevant (Howard et al. 2011). It should be noted that women may use a number of different approaches and decision-making styles throughout the process, just as women may use several coping strategies (Howard et al. 2011, Lazarus and Folkman 1984).

5.4.2 *Practice implications and future research*

The CODE framework was developed to provide a description of preference-sensitive decision making from a patient perspective in order to (i) help practitioners better understand their patients' questions and concerns, (ii) provide a theoretical underpinning for the development of decision support and (iii) act as an assessment tool to test whether existing decision support truly addresses issues patients explore during deliberation. Overall, the framework could thus support patient-centred care (PCC) and provide a theoretical underpinning for decision support materials (Durand et al. 2008, Elwyn et al. 2000, Elwyn et al. 2009a, O'Grady and Jadad 2010, Secretary of State for Health 2010).

By adapting the framework to a specific decision the relevance of the phases and questions is increased, thereby providing an even more accurate description of decision making in a specific context. The RRSO-adapted CODE framework presented in this chapter describes deliberations about increased genetic risk of ovarian cancer and associated management options. This adapted version may now be useful in assessing currently available decision support aimed at women at increased risk of ovarian cancer and in developing tailored decision support, if such support is not available or not adequate.

Future research using the RRSO-adapted CODE framework may focus on additional validation, for example content validity assessment with a group of experts and further face validity testing in focus groups or interviews with health professionals and/or patients. Furthermore, the framework may be applied to assess and develop decision support interventions. As demonstrated in this chapter, the CODE framework is readily adaptable to a specific context and the results of this research may guide further adaptations to other preference-sensitive decisions, such as catheter investigation for coronary artery disease (CAD) or amniocentesis to assess foetal health.

5.4.3 *Strengths and limitations*

The current chapter delineates the adaptation of the CODE framework to a specific preference-sensitive decision using information obtained from a literature review, as well as focus groups with patients and interviews with health professionals (Chapter 3 and 4). This approach to adaptation results in a thorough understanding of the issues important during deliberation about the specific decision (i.e. RRSO) in a specific population and context (i.e. women at increased genetic risk in the UK). Therefore the adapted version of the framework is grounded in a literature review as well as data collected from stakeholders and reflects important aspects explored by patients and health professionals. Other approaches to adaptation, limited to either a literature review or data from a small group of patients and/or health professionals only, may not be as accurate or reflective of a specific decision in a specified population/context and may therefore be less useful in supporting PCC and the development of decision support materials.

Arguably, the questions posed in the adapted framework are not exhaustive and there may be a whole range of other aspects women might explore during primary and secondary appraisal. However, by using a broad literature review in combination with interviews and focus groups with stakeholders to inform the issues included in the adapted framework, the current adaptation likely covers the most common and salient points that may be explored during deliberations about RRSO. Furthermore, while it cannot provide information about the exact questions a given individual patient might explore, it offers an overview of those issues most frequently considered and those deemed important by health professionals. Therefore, any consultation or support material which addresses the questions included within the adapted framework will likely cover the more common issues. This may leave more time to discuss any additional individual questions a patient may have after receiving the initial information.

As with the generic framework, the adapted framework attempts to describe both cognitive and emotional processes, as well as appraisal of coping options. This

comprehensive view of deliberation and coping as intricately linked processes allows a more holistic description of decision making. This means that decision support materials based on the framework may support adaptive coping and emotion regulation in addition to providing information to facilitate cognitive appraisal and affective forecasting.

The RRSO-adapted framework was validated using one retrospective account of coping and deliberation processes, which may have been subject to recall bias. However, the patient had undergone surgery only six months before the interview and was recalling events that had led up to her decision in the year preceding the surgery. Therefore, the time delay between decision making and interview was relatively short. The patient had a confirmed BRCA mutation and had completed her family, hence some may argue that her decision was comparatively easy and free from conflict. However, the patient was under 40 years of age at the time of her decision, which means she had to consider the effects of a premature menopause and whether or not to take HRT. Additionally, although she had had two children, she had to be sure her family was complete before making her final decision and also had to consider the potential effects surgery would have on her relationship with her husband. The in-depth account of her decision making shows that, despite being BRCA positive and having completed her family, this patient did take her time with making this decision and explored how outcomes would affect her life before undergoing surgery, indicating a thorough deliberation worth mapping onto the RRSO-adapted CODE framework.

Further interviews with other patients recounting their deliberation process would have been beneficial. However, recruitment was extremely difficult as many women who opt for surgery leave the genetic service and are difficult to contact thereafter. Although the current study attempted to recruit more patients, this was not successful due to the low response rate, as well as time and area restrictions. However, the interview that was conducted included an in-depth narrative account of a deliberation process, which was analysed using thematic analysis (Braun and Clarke

2006). The richness of this dataset allowed accurate mapping onto the CODE framework and provides a detailed insight into this patient's deliberation and coping strategies that may not have been possible with a larger dataset from several patients.

A prospective validation using observations and interviews with patients as they go through the deliberation process would also have added to the adapted framework's credibility. Similarly, content validity assessment with a group of experts examining whether adapted questions truly represent the suggested associated deliberation and appraisal phases would have been advantageous. However, such approaches were beyond the scope of the current study. Despite this limitation, the retrospective exploration carried out was appropriate as an initial validation procedure. It demonstrated that the patient's deliberation process mapped well onto the framework and was accurately reflected by a selection of the questions within it. The RRSO-adapted framework may benefit from further validation in the future.

5.4.4 Conclusions

In this chapter it was demonstrated that the generic CODE framework can be readily adapted to a selected preference-sensitive decision using information about salient issues important in that decision. These considerations were derived from focus groups and interviews with stakeholders, as well as a literature review (Chapters 3 and 4). Adaptations to other decisions using this approach are possible and enable the generic CODE framework to be applied to a variety of different preference-sensitive medical decisions.

The RRSO-adapted framework will be used to assess currently available decision support interventions that include the option of RRSO and their coverage of issues considered during primary and secondary appraisal (Chapter 6). Subsequently, it will be used as a guide for the development of a patient decision support intervention for women at increased genetic risk of ovarian cancer in the UK, where currently no tailored decision support outside the clinic is available (Chapter 7).

6 Applying the CODE framework: A review of decision support interventions that include the option of risk-reducing oophorectomy

6.1 Introduction

Decisions about management in the context of increased genetic risk of cancer are inherently difficult, as they affect otherwise healthy individuals who are making a decision on how to manage a potential future risk (Fang et al. 2005, Howard et al. 2009a, Miller et al. 1999). Furthermore, risk in itself is an abstract concept, which can be difficult to understand for individuals of all ages and abilities (Gigerenzer and Edwards 2003, Zikmund-Fisher et al. 2010). Women at increased genetic risk of ovarian cancer are required to appraise their cancer risk, as well as the risks and benefits associated with available management options. Additionally, they should consider their personal values and preferences and, ideally, actively participate in shared decision making (SDM) to reach an informed, preference-adjusted decision. They may be expected to do all this in a psychologically aroused state and while experiencing a number of anticipated and anticipatory emotions (Loewenstein et al. 2001, Wilson and Gilbert 2003). Therefore, in order to reach truly informed and value-adjusted decisions, regulating emotions during decision making is essential. Hence, coping with the threat and associated emotions is an integral part of deliberation processes, as suggested by the Coping in Deliberation (CODE) framework (Chapter 2; Witt et al. 2012).

Decision support interventions are tools that aim to provide information, facilitate risk communication, ease value clarification and support SDM (O'Connor et al. 2004, Thistlethwaite et al. 2006). Such interventions have been shown to have positive effects on patient knowledge, accuracy of risk perceptions and decision satisfaction by increasing alignment between the chosen option and patient values (O'Connor et al. 2009, Stacey et al. 2011). Currently several generic decision support interventions are available which may be used in a number of different medical

contexts (Ottawa Hospital Research Institute 2013). Additionally, a range of tailored patient decision support interventions has been published for a large number of medical decisions, from acne treatment and cancer care to genetic testing and weight control (Ottawa Hospital Research Institute 2013, Stacey et al. 2011). Some are designed to address the needs of specific patient populations, for instance, those with a family history of cancer who are considering genetic testing for a BRCA mutation (Wakefield et al. 2008a, Wakefield et al. 2008b) or those with early stage breast cancer who are deciding between lumpectomy and mastectomy (Sivell et al. 2012b).

The aim of this chapter is (i) to identify patient decision support interventions that include information about risk-reducing salpingo-oophorectomy (RRSO), (ii) to examine their format, design, content, effectiveness and use of theory and (iii) to map the contents of these interventions onto the RRSO-adapted Coping in Deliberation (CODE) framework (Chapter 5; Witt et al. 2012).

6.2 Methodology

6.2.1 Literature Search

For the purpose of this Chapter, patient decision support interventions were defined as “evidence-based tools designed to prepare clients to participate in making specific and deliberated choices among healthcare options” (Stacey et al. 2011, page 3).

To identify relevant decision support interventions and associated publications, a systematic literature search was carried out (Table 6.1). A title search was conducted in July 2012 via Ovid. Databases included Medline In-Process 1996 to July week 4 2012, Medline without Revisions 1996 to July week 4 2012, EMBASE 1996 to 2012 July and PsycINFO 2002 to July week 4 2012. Duplicates which had been found in more than one database were removed.

Table 6.1 - Search-terms used for title search via Ovid

| Term 1 | | Term 2 | | Term 3 |
|------------|-----|-------------------------------|-----|--------------------------|
| Decision\$ | | Aid\$ | | Ovarian cancer |
| Decide | | Tool\$ | | Oophorectomy |
| Deciding | | Intervention\$ | | BRCA |
| Choose | AND | Support | AND | BRCA1 |
| Choosing | | Explorer | | BRCA2 |
| | | Clinical guidance programme\$ | | Lynch |
| | | | | Hereditary non-polyposis |
| | | | | colorectal cancer |
| | | | | HNPCC |

6.2.2 Exclusion Criteria

The study aimed to identify original research that was concerned with the development and/or evaluation of decision support interventions that included the option of RRSO. Therefore, reviews and editorials were excluded. Furthermore, publications were excluded if (i) they were not concerned with decision support interventions, (ii) they exclusively focused on supporting decisions on genetic testing, disclosure of at-risk status or treatment of ovarian cancer or (iii) they were exclusively concerned with breast cancer prevention by screening or mastectomy.

6.2.3 Other Methods of Identification

A supplementary search was performed manually by searching the Ottawa Hospital Research Institute decision aid A-Z inventory (2013), the Informed Medical Decision Foundation website (2012) and the New South Wales Centre for Genetics Education website (2008) to identify further relevant interventions.

6.2.4 Analysis

Wherever possible an effort was made to obtain the full decision support intervention and associated materials, if it had been published and was accessible

to patients. In cases where an intervention was not publicly available, the author of corresponding publications was contacted to provide additional information.

The interventions were assessed using a specially designed data extraction form (Appendix 6.1). The form was developed using the International Patient Decision Aid Standards (IPDAS) checklist items on intervention content as a guide (Elwyn et al. 2006). The items within the checklist were adapted to assess information about ovarian cancer risk and associated management options. Additionally, the form assessed format and accessibility, as well as information on choice, value clarification content, opportunity for personalisation and effectiveness. Where interventions had been evaluated, study design and outcomes were also examined. The extraction form was then applied to two decision support interventions (Healthwise 2011b, New South Wales Centre for Genetics Education 2008) and subsequently amended to include additional items relevant to decisions on RRSO that had previously been omitted. Two interventions were assessed by two independent raters. Inter-rater consistency was >85% and any discrepancies were resolved by discussion until a consensus was reached.

6.2.5 Application of the CODE framework

The CODE framework describes decision making and coping using a number of questions (Chapters 2). It has been proposed that this framework could act as an underlying theoretical framework for decision support interventions (Witt et al., 2012). In this study, the RRSO-adapted version of the CODE framework (Chapter 5) was mapped onto the content of decision support interventions identified during the review in order to determine how many of the questions raised in the framework were addressed in each intervention (Appendix 6.2). A question was deemed 'addressed' if:

- i. there was information directly relating to the question (e.g. Question: "What is risk-reducing bilateral salpingo oophorectomy?" was addressed by

“Prophylactic bilateral salpingo-oophorectomy: Removal of both ovaries and fallopian tubes before cancer occurs” (Kurian et al. 2012, in glossary)

- ii. users were encouraged to consider or discuss an item or were made aware of an issue indirectly (e.g. Question: “Have I completed my family?” was addressed in a flowchart by the statement: “You may want children/more children” (New South Wales Centre for Genetics Education 2008, page 14)

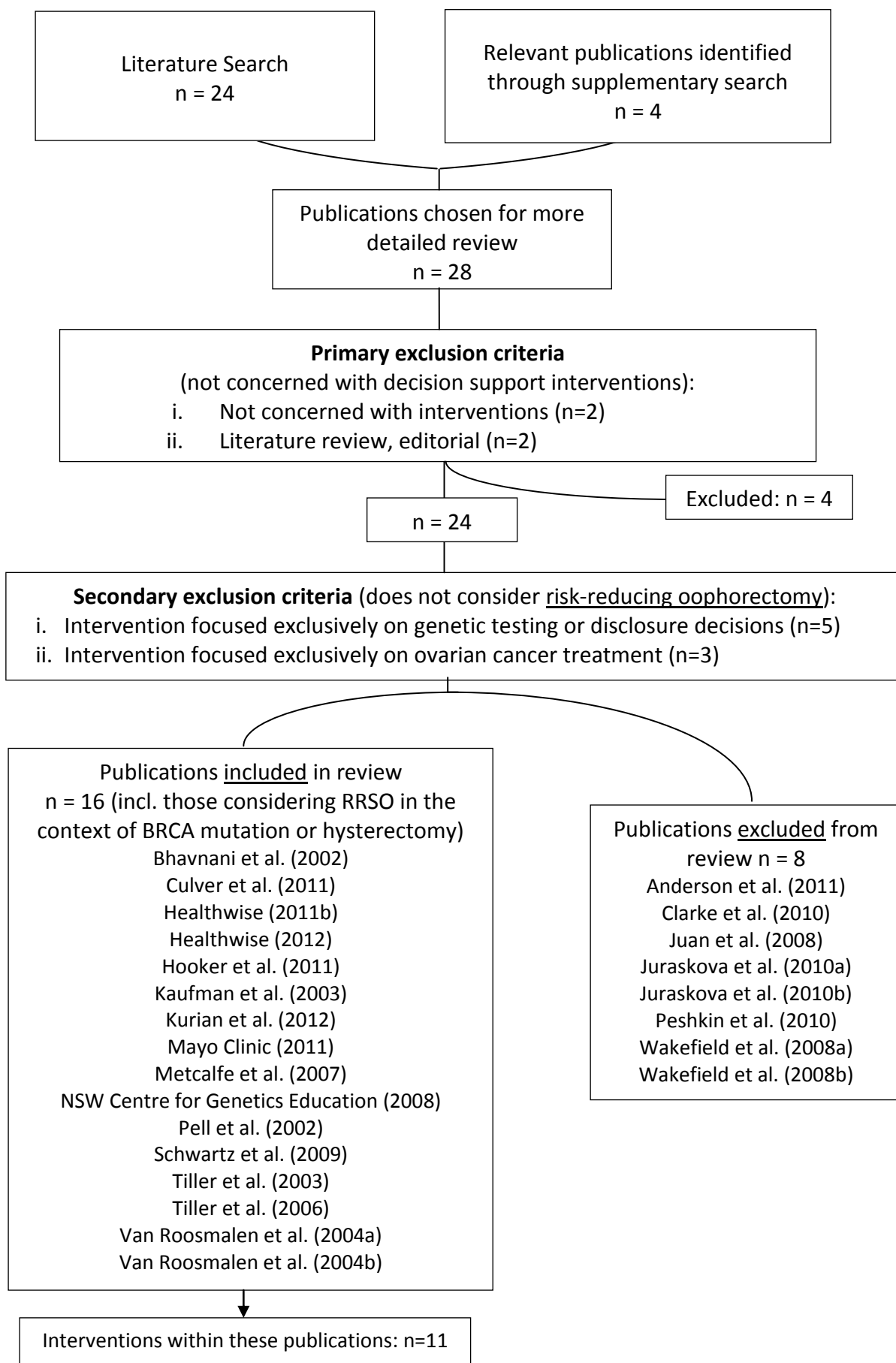
Two interventions were assessed this way by two independent raters (inter-rater consistency >80%) and discrepancies were discussed until a consensus was reached.

6.3 Results

6.3.1 Available Interventions

The literature search yielded a total of 51 results, of which 27 were duplicates found in more than one database (Figure 6.1). The results of a manual search on relevant websites (n=4) were added to the 24 publications identified through the databases to yield a total of 28 publications for further analysis. Four publications did not present original research or were not concerned with decision support interventions. Of the 24 remaining publications, five were concerned with interventions focused on genetic testing or disclosure to family members and three were concerned with ovarian cancer treatment, rather than prevention, and were subsequently excluded. A total of 16 publications remained for detailed analysis, encompassing 11 different decision support interventions that included the option of RRSO. These included interventions aimed at carriers of mutations in BRCA1/2 (Culver et al. 2011, Healthwise 2011b, Kurian et al. 2012, Mayo Clinic 2011, New South Wales Centre for Genetics Education 2008, Tiller et al. 2003, van Roosmalen et al. 2004a) and at women considering RRSO in the context of a planned hysterectomy (Bhavnani et al. 2002, Healthwise 2012, Pell et al. 2002).

Figure 6.1 - Results of the literature search



A total of six interventions were available for analysis at the time of the study (Healthwise 2011b, 2012, Kaufman et al. 2003, Mayo Clinic 2011, New South Wales Centre for Genetics Education 2008, Stanford Medicine 2012) and five interventions were analysed based on the information provided in publications and through discussions with their developers (Bhavnani et al. 2002, Culver et al. 2011, Metcalfe et al. 2007, Pell et al. 2002, van Roosmalen et al. 2004a, van Roosmalen et al. 2004b). None of the five interventions assessed through publications were available to patients at the time of the study. A summary of the 11 interventions which were included in the review is presented in Table 6.2.

6.3.2 *Format and Design*

The interventions varied widely in format and design, from booklets and videos to computerised guidance programmes and websites (Table 6.2). Most interventions (n=7) were designed to be used independently by the patient outside of clinical encounters (Healthwise 2011b, 2012, Kaufman et al. 2003, Mayo Clinic 2011, Metcalfe et al. 2007, Tiller et al. 2003, van Roosmalen et al. 2004b). Four interventions were designed to be used together with a facilitator or health professional in the clinic or on the phone (Bhavnani et al. 2002, Kurian et al. 2012, Pell et al. 2002, van Roosmalen et al. 2004a). Of these, three interventions were delivered in a single face-to-face session (Bhavnani et al. 2002, Kurian et al. 2012, Pell et al. 2002) and one intervention was delivered in three separate sessions: two face-to-face encounters and one telephone interview (van Roosmalen et al. 2004a). The latter was a SDM intervention, which differed substantially from the other decision support interventions in this study.

Five interventions were available freely online (Healthwise 2011b, 2012, Kurian et al. 2012, Mayo Clinic 2011, New South Wales Centre for Genetics Education 2008) and one was available through practitioners as a CD-Rom (Kaufman et al. 2003). The remaining five interventions were not available publicly at the time of the study.

Table 6.2 - Decision support interventions included in the review

| Format | Publication | Year | Country | Decision | Target Audience | Options included |
|--|--------------------|-------------|----------------|---|---|--|
| Online tool | Kurian | (2012) | USA | RM/RRSO/Screening in BRCA1/2 positive women | BRCA1/2 positive women | <ul style="list-style-type: none"> - Risk-reducing mastectomy and RRSO - Breast cancer screening and RRSO - Risk-reducing mastectomy without RRSO |
| Online tool | Healthwise | (2012) | USA | RRSO with hysterectomy | Women who are undergoing a planned hysterectomy | <ul style="list-style-type: none"> - Hysterectomy with RRSO - Hysterectomy alone |
| Online tool | Healthwise | (2011b) | USA | RRSO for high risk women | Women at high risk | <ul style="list-style-type: none"> - Have RRSO - Don't have RRSO |
| Online tool | Mayo Clinic | (2011) | USA | RRSO in BRCA1/2 positive women | BRCA1/2 positive women | <ul style="list-style-type: none"> - Have RRSO - Don't have RRSO (alternatives: screening, RRM, OCP) |
| Online tool | Metcalfe | (2007) | Canada | RRM/RRSO/chemoprevention | BRCA1/2 positive women | <ul style="list-style-type: none"> - Have / Don't have RRM - Have / Don't have RRSO - Take / Don't take Chemoprevention |
| Booklet and Video | Van Roosmalen | (2004b) | NL | RRSO in BRCA1/2 positive women | BRCA1/2 positive women | <ul style="list-style-type: none"> - Risk-reducing mastectomy and RRSO - Breast cancer screening and RRSO - Risk-reducing mastectomy and ovarian cancer screening - Breast cancer screening and ovarian cancer screening |
| Shared Decision Making Intervention | Van Roosmalen | (2004a) | NL | RRSO in BRCA1/2 positive women | BRCA1/2 positive women | <ul style="list-style-type: none"> - Risk-reducing mastectomy and RRSO - Breast cancer screening and RRSO - Risk-reducing mastectomy and ovarian cancer screening - Breast cancer screening and ovarian cancer screening |

Table 6.2 cont. – Decision support interventions included in the review

| Format | Publication | Year | Country | Decision | Target Audience | Options included |
|----------------------------------|--------------------|-------------|----------------|---|---|--|
| CD-Rom | Kaufman | (2003) | USA | RM/RRSO/Screening in BRCA1/2 positive women | BRCA1/2 positive women | <ul style="list-style-type: none"> - Risk-reducing mastectomy and RRSO - Breast cancer screening and RRSO - Risk-reducing mastectomy and ovarian cancer screening - Breast cancer screening and ovarian cancer screening |
| Booklet | Tiller | (2003) | AUS | RRSO for high risk women | Women at high or potentially high risk | <ul style="list-style-type: none"> - Have RRSO - Don't have RRSO |
| Decision Chart | Bhavnani | (2002) | UK | RRSO with hysterectomy | Women who are undergoing a planned hysterectomy | <ul style="list-style-type: none"> - Hysterectomy with RRSO - Hysterectomy alone - Use of HRT |
| Clinical Guidance Program | Pell / Bhavnani | (2002) | UK | RRSO with hysterectomy | Women who are undergoing a planned hysterectomy | <ul style="list-style-type: none"> - Hysterectomy with RRSO - Hysterectomy alone - Use of HRT |

Two of these targeted women in the UK who were undergoing a planned hysterectomy. These had not been published due to a change in NICE guidelines, which no longer recommended RRSO with planned hysterectomy in women not at increased risk of ovarian cancer (Bhavnani et al. 2002, Clarke 2010, National Institute for Health and Care Excellence 2007, Pell et al. 2002). Similarly, two Dutch interventions for carriers of mutations in BRCA1/2 had never been implemented in clinical practice (Stalmaier 2010, van Roosmalen et al. 2004a, van Roosmalen et al. 2004b). The fifth unpublished intervention was still being used in trials and was therefore not publicly available at the time of the study (Culver et al. 2011, Metcalfe et al. 2007).

6.3.3 *Information Content*

The scope and depth of information presented in interventions varied widely and depended on the target decision and audience (Table 6.3). Only five interventions were specifically designed to facilitate decisions about RRSO to reduce increased risk of ovarian cancer due to genetic predisposition (Healthwise 2011b, Mayo Clinic 2011, Tiller et al. 2003, van Roosmalen et al. 2004a, van Roosmalen et al. 2004b). Three of these were specifically aimed at women with a confirmed mutation in BRCA1/2 (Mayo Clinic 2011, van Roosmalen et al. 2004a, van Roosmalen et al. 2004b). Three other interventions considered RRSO as a concomitant procedure in women scheduled for hysterectomy (Bhavnani et al. 2002, Healthwise 2012, Pell et al. 2002) and three interventions mainly focused on the reduction in risk of breast, rather than ovarian, cancer (Kaufman et al. 2003, Kurian et al. 2012, Metcalfe et al. 2007). Culver et al. (2011) adapted the intervention designed by Metcalfe et al. (2007), but it remained focused on breast cancer risk reduction and was therefore considered as a single intervention.

Table 6.3 - Information content of interventions that include the option of risk-reducing salpingo-oophorectomy (RRSO)

| Content Item | RRSO to prevent ovarian cancer | | | | | RRSO to prevent breast cancer | | | RRSO with planned hysterectomy | | |
|--|--------------------------------|-------------|----------------|-----------------|------------------|-------------------------------|---------------|---------------|--------------------------------|---------------|-----------|
| | Healthwise 2011 | Tiller 2003 | Roosmalen 2004 | Roosmalen 2004* | Mayo Clinic 2011 | Kurian 2012 | Metcalfe 2007 | Kaufmann 2003 | Healthwise 2012 | Bhavnani 2002 | Pell 2002 |
| Information about the health condition | | | | | | | | | | | |
| Information about the function of ovaries | • | • | ? | | | | ? | | • | ? | ? |
| Information about ovarian cancer | | • | • | | | | ? | | • | • | ? |
| Information about symptoms of ovarian cancer | | • | ? | | | | ? | | | ? | ? |
| Risk of ovarian cancer explained | • | • | • | | | • | ? | • | • | ? | ? |
| Explanation of familial/hereditary cancer | • | • | • | | | | • | • | • | • | ? |
| Information about genetic testing | • | • | • | | | | ? | | • | ? | ? |
| Information about risk-reducing surgery | | | | | | | | | | | |
| Information about what RRSO entails | • | • | • | • | • | • | • | | • | ? | ? |
| Information about recovery time | • | • | • | | • | | ? | | • | ? | ? |
| Information about surgical menopause | • | • | • | | • | • | • | • | • | ? | • |
| Information about hormone replacement therapy | | • | • | | • | | ? | | • | • | ? |
| Information about infertility | • | • | ? | | • | | • | | | ? | ? |

Table 6.3 cont. – Information content of interventions that include the option of risk-reducing salpingo-oophorectomy (RRSO)

| Content Item | RRSO to prevent ovarian cancer | | | | | RRSO to prevent breast cancer | | | RRSO with planned hysterectomy | | |
|---|--------------------------------|-------------|----------------|-----------------|------------------|-------------------------------|---------------|---------------|--------------------------------|---------------|-----------|
| | Healthwise 2011 | Tiller 2003 | Roosmalen 2004 | Roosmalen 2004* | Mayo Clinic 2011 | Kurian 2012 | Metcalfe 2007 | Kaufmann 2003 | Healthwise 2012 | Bhavnani 2002 | Pell 2002 |
| Information on psychosocial issues after surgery | | • | • | | • | | ? | | | ? | ? |
| Information about residual risk after surgery | • | • | • | | • | • | • | | | ? | ? |
| Risks / Negative features explained | • | • | • | | • | | • | • | • | ? | ? |
| Benefits / Positive features explained | • | • | • | | • | • | • | • | • | ? | ? |
| Information about watchful waiting / symptom awareness | | | | | | | | | | | |
| Information about what option entails | • | • | | | | | | • | | | |
| Information about symptoms | | • | | | | | | | | | |
| Risks / Negative features explained | | • | | | | • | | | | | |
| Benefits / Positive features explained | | • | | | | | | | | | |
| Information about screening for ovarian cancer | | | | | | | | | | | |
| Information about what option entails | • | • | • | • | • | | • | • | | | |
| Risks / Negative features explained | | • | ? | | • | | ? | • | | | |
| Benefits / Positive features explained | | • | • | | • | | ? | • | | | |

Table 6.3 cont. – Information content of interventions that include the option of risk-reducing salpingo-oophorectomy (RRSO)

| Content Item | RRSO to prevent ovarian cancer | | | | | RRSO to prevent breast cancer | | | RRSO with planned hysterectomy | | |
|---|--------------------------------|----------------|-------------------|--------------------|---------------------|-------------------------------|------------------|------------------|--------------------------------|------------------|--------------|
| | Healthwise 2011 | Tiller 2003 | Roosmalen 2004 | Roosmalen 2004* | Mayo Clinic 2011 | Kurian 2012 | Metcalfe 2007 | Kaufmann 2003 | Healthwise 2012 | Bhavnani 2002 | Pell 2002 |
| Information about chemoprevention with oral contraceptives | | | | | | | | | | | |
| Information about what option entails | • | • | | | • | | | • | | | |
| Risks / Negative features explained | • | • | | | • | | | | | | |
| Benefits / Positive features explained | • | • | | | • | | | | | | |

*Individuals receiving this intervention had previously been given the booklet and video intervention (van Roosmalen 2003, 2004).

• = item present (either checked directly with available interventions, mentioned / shown in associated publications or through correspondence with author)

? = intervention not available for direct review, therefore not possible to determine whether item is present or absent

Generally, interventions designed for independent use by patients included information materials on the health condition and at least one management option, usually RRSO (Healthwise 2011b, 2012, Kaufman et al. 2003, Metcalfe et al. 2007, Tiller et al. 2003, van Roosmalen et al. 2004b). In contrast, interventions delivered by facilitators or health professionals either only included a minimum amount or no standard information (Bhavnani et al. 2002, Kurian et al. 2012, van Roosmalen et al. 2004a). However, information may be obtained or discussed prior to delivery of the intervention in a consultation or using another intervention (van Roosmalen et al. 2004a, van Roosmalen et al. 2004b). Alternatively, information may also be provided during the delivery of the intervention through discussions of options with the health professional (Bhavnani et al. 2002, Kurian et al. 2012, Pell et al. 2002).

The majority of patient decision support interventions included some information about the option of RRSO; however the amount and level of detail of this information differed greatly. While some included comprehensive information on operative procedures, surgical menopause and hormone replacement (Mayo Clinic 2011, Tiller et al. 2003), others only included a short definition (Kurian et al. 2012). Psychosocial consequences of surgery, such as effects on sexuality, relationships and perception of femininity, were also only discussed briefly, if at all (Mayo Clinic 2011, Tiller et al. 2003, van Roosmalen et al. 2004b). Information on other management options, such as screening or symptom awareness, was similarly varied. While some interventions contained extensive information on screening, symptom awareness and the oral contraceptive pill (Tiller et al. 2003), others merely mentioned these options briefly (Healthwise 2011b). Interventions aimed at women undergoing a planned hysterectomy included no information about alternative management options for ovarian cancer risk, as the target populations were not at increased risk for this cancer (Bhavnani et al. 2002, Healthwise 2012). Similarly, interventions mainly concerned with reductions in breast cancer risk often

did not include management options focused on ovarian cancer risk (Kurian et al. 2012, Metcalfe et al. 2007), although some did discuss these options briefly (Culver et al. 2011, Kaufman et al. 2003).

The SDM intervention developed by van Roosmalen et al. (2004a) included the least amount of information. However, this was preceded by the video and booklet intervention also developed by this group. Therefore, patients could be assumed to have received this information prior to going through the time-trade-off exercises in the SDM intervention (van Roosmalen et al. 2004a, van Roosmalen et al. 2004b).

6.3.4 Value Clarification

Most interventions included a type of value clarification exercise. This was either completed by the patient herself (n=5) (Healthwise 2011b, 2012, Kaufman et al. 2003, Metcalfe et al. 2007, Tiller et al. 2003), by the patient with guidance from a facilitator (n=2) (Bhavnani et al. 2002, Pell et al. 2002) or by a trained facilitator or health professional with input from the patient (n=2) (Kurian et al. 2012, van Roosmalen et al. 2004a). Two interventions did not appear to include a value clarification exercise (Mayo Clinic 2011, van Roosmalen et al. 2004b). However, one of these was complemented by a separate SDM intervention (van Roosmalen et al. 2004a, van Roosmalen et al. 2004b).

Most of the explicit, self-administered value clarification exercises asked patients to score, rank or weigh different features of options using sliders, five point Likert scales or shaded weights (Culver et al. 2011, Healthwise 2011b, 2012, Kaufman et al. 2003, Metcalfe et al. 2007, Tiller et al. 2003). It should be noted that the intervention by Kaufman et al. (2003), while including an explicit value clarification exercise, did not include the option of RRSO within this exercise, as it exclusively focused on decision making about breast cancer risk management.

Other interventions, including the SDM intervention, established patient values with time trade-off exercises requiring the patient to attribute values to potential future health states associated with different options (e.g. with/without RRSO, with/without HRT) (Pell et al. 2002, van Roosmalen et al. 2004a). Life expectancy and quality adjusted life expectancy were then calculated based on the information provided by patients.

The interventions by Kurian et al. (2012) and Bhavnani et al. (2002) explored patient values more indirectly than the explicit value clarification exercises in other tools. The intervention designed by Kurian et al. (2012) allowed visualisation of the effects of different management strategies on cancer incidence and life expectancy, so that individual values and trade-offs could be discussed. In the decision chart developed by Bhavnani (2002), patients were first presented with graphical, textual and quantitative information for each of four possible outcomes, then individual risk factors and their effects on outcome probabilities were discussed and women could express their thoughts and feelings about the different options.

6.3.5 Additional Outputs

Five interventions allowed patients to personalise the information or the probabilities presented by considering personal risk factors such as age, smoking and disease history (Bhavnani et al. 2002, Kaufman et al. 2003, Kurian et al. 2012, Pell et al. 2002, Tiller et al. 2003). The Australian decision support booklet clearly distinguished between pre- and post-menopausal women and informed post-menopausal women that certain sections in the booklet did not apply to them (Tiller et al. 2003). On the CD-Rom, patients could enter personal data such as age and previous cancer history to view more tailored information (Kaufman et al. 2003). The web-based intervention by Kurian et al. (2012) allowed users to tailor content by age and mutation status. In the other two interventions, personal information was elicited by the facilitator (Bhavnani et al. 2002, Pell et al. 2002).

Three interventions allowed patients to add their own reasons to the value clarification exercise (Healthwise 2011b, 2012, Metcalfe et al. 2007). Four interventions left space for personal notes, which could be used to summarise information or note down questions to ask the clinician (Healthwise 2011b, 2012, Kaufman et al. 2003, Tiller et al. 2003). One intervention included a list of nine questions to ask the healthcare provider (Mayo Clinic 2011).

The inclusion and usefulness of personal stories in patient decision support interventions is controversial (Winterbottom and Bekker 2009). While stories can add credibility, break up complex medical information and help patients imagine different outcomes and how they could affect their life, there is a danger that patients may identify with some of the patients in the stories and may base their decision on what happened to that individual patient. Furthermore, as patient stories are often biased, portraying potential outcomes in a balanced way using patient stories is also seen as problematic (Redelmeier et al. 1993, Winterbottom and Bekker 2009). Despite this on-going debate, almost half of the interventions in this study (n=5) included a form of personal story or account (Healthwise 2011b, 2012, Kaufman et al. 2003, Tiller et al. 2003, van Roosmalen et al. 2004b). Three of these presented personal stories as written quotes and two included audio-visual accounts of patients' experiences.

More than half the interventions included in this study (n=7) referenced the sources they used to enable patients to obtain further information if they wished (Healthwise 2011b, 2012, Kurian et al. 2012, Mayo Clinic 2011, Metcalfe et al. 2007, Tiller et al. 2003, van Roosmalen et al. 2004b). The Australian booklet also included contact details of genetics services throughout Australia and referred to a number of websites with further information, such as the cancer council (New South Wales Centre for Genetics Education 2008, Tiller et al. 2003). The intervention by Metcalfe

et al. (2007) additionally provided quality statements about the evidence used to support the included information and risk estimates.

6.3.6 Evaluation

Seven of the decision support interventions included in the analysis had been evaluated before publication. Four interventions were evaluated in five randomised trials (Hooker et al. 2011, Schwartz et al. 2009, Tiller et al. 2006, van Roosmalen et al. 2004a, van Roosmalen et al. 2004b) and three were piloted with small groups of women (Bhavnani et al. 2002, Metcalfe et al. 2007, Pell et al. 2002). Three interventions had been developed by commercial producers (Healthwise 2011b, 2012, Mayo Clinic 2011) and one had only been published in 2011 (Kurian et al. 2012), therefore no information was available on evaluation of these interventions.

The Australian decision support booklet developed by Tiller et al. (2003) was compared to an educational pamphlet in a randomised controlled trial with 109 women (Tiller et al. 2006). The study measured changes in knowledge, intrusive thoughts, anxiety, decisional conflict and uncertainty over three time points. Additionally, acceptability and helpfulness of the material, as well as its influence on the decision, were assessed at the final time point. The trial found higher levels of acceptability of materials in the intervention group, but no difference in perceived helpfulness when compared to an educational leaflet. There were no significant differences in any of the psychological outcomes and only small to medium effect size differences associated with knowledge between intervention and control group. The only significant outcome measure was decisional conflict, which was reduced in the intervention group 2 weeks post-intervention; however this effect was no longer significant at 6 months. Those who had received the intervention were also significantly more likely to indicate that it had influenced their decision. No differences in the actual decisions were noted.

Schwartz et al. (2009) evaluated the interactive CD-Rom developed by Kaufman et al. (2003) in a randomised trial with 214 women. The study assessed changes in decisional conflict and decision satisfaction, as well as effects on decision making in an intervention group compared to a control group that received usual care. Among women who reported being undecided at the point of randomisation, the intervention significantly increased the number of individuals who could decide on a management option. In this group, it also significantly decreased decisional conflict and increased satisfaction. In contrast, the intervention did not significantly influence these outcomes in individuals who had already made a management decision at the point of randomisation. The authors therefore suggest that the intervention may only benefit those individuals who struggle to make a decision, and not those who have already made a decision shortly after receiving their test result (Schwartz et al. 2009). Hooker et al. (2011) reported on the same trial, but focused on effects of the intervention on several measures of distress. They found that receipt of the intervention after BRCA testing resulted in higher sustained cancer-specific and genetic testing-specific distress at 1 month post-intervention. The authors suggest that sustained distress may be indicative of enhanced cognitive processing and deliberation about decision relevant information in the short-term (Hooker et al. 2011).

In a combined randomised trial of two interventions, van Roosmalen et al. (2004a, 2004b) trialled a booklet/video and SDM intervention. Measures included well-being, treatment choice and decision-related outcomes. Additionally, outcomes were assessed for the information booklet and video intervention through knowledge, perceived amount of and satisfaction with information and risk perception. Patients (n=384) were randomised after providing a blood sample for a BRCA mutation test (van Roosmalen et al. 2004b). The intervention group received the booklet and video before, and the control group after, they had received their

test result. No significant effects of the booklet/video were found on measures of well-being or decision related outcomes between groups. However, the intervention significantly improved subjective knowledge and satisfaction with the information provided and more women in the intervention group favoured surgery before disclosure of the results. Some improvements in risk accuracy in the intervention group were also noted. Once mutation status was confirmed, 88 carriers of mutations in BRCA were randomised a second time to receive the SDM intervention or usual care (van Roosmalen et al. 2004a). This intervention showed long-term significant positive effects on depression and intrusive thoughts, which declined, and general health, which improved. No significant effects were found on anxiety, treatment choice and decision-related outcomes, with the exception that the intervention group had stronger preferences and felt that they had weighed the risks and benefits of the available options.

Bhavnani et al. (2002) conducted a pilot evaluation through a qualitative study using semi-structured interviews with 29 women before and after they had used one of two interventions: a decision chart (n=10) or clinical guidance programme (n=19) with a facilitator in the clinic. Women were asked about their expectations of the intervention, the process of using the intervention, the impact it had on their decision and their reaction to the output. Most women were satisfied with the chart's output, some even felt empowered after using it, but it did not appear to change women's initial attitudes towards their preferred management option. The chart did, however, encourage communication between patients and facilitators. The clinical guidance program was more difficult to use and women appeared surprised about the definitive statement provided at the end of the intervention, which read: 'Given the information you have provided, the CGP recommends the [option]' (Bhavnani et al. 2002). When this guidance statement differed from women's preferred management option, they tended to reject the

program's advice and used it instead to consolidate their decision by arguing that there was almost no difference between the quality adjusted life expectancy gained with either option.

Metcalfe et al. (2007) developed and piloted their intervention in a non-randomised, before and after evaluation with 21 carriers of mutations in BRCA1/2. Outcomes assessed in this study included knowledge, decisional conflict, choice predisposition, cancer-specific distress and acceptability of the materials. Overall, the intervention appeared to increase knowledge and reduce decisional conflict. However, it had no effect on cancer-related distress. The majority of women reported high acceptability (Metcalfe et al. 2007). This intervention was further adapted by Culver et al. (2011).

6.3.7 Theoretical basis or framework

Three interventions were based on at least one theory, model or framework that was specified in associated publications (Culver et al. 2011, Kaufman et al. 2003, Metcalfe et al. 2007, Schwartz et al. 2009, Tiller et al. 2003). Eight interventions did not provide information on underlying theory through associated publications or the intervention itself (Bhavnani et al. 2002, Healthwise 2011b, 2012, Kurian et al. 2012, Mayo Clinic 2011, Pell et al. 2002, van Roosmalen et al. 2004a, van Roosmalen et al. 2004b).

The Ottawa Decision Support Framework (ODSF) (O'Connor 2006, O'Connor et al. 1998) was quoted as the underlying framework in all interventions that specified a theory base (Metcalfe et al. 2007, Schwartz et al. 2009, Tiller et al. 2003). The ODSF attempts to guide the design of patient decision support interventions by identifying modifiable determinants of decisions, such as inadequate knowledge, unclear values, lack of support and undesirable stressors, that should be addressed by interventions (O'Connor, 1998). Publications did not specify how the ODSF was

used to guide development (Metcalfe et al. 2007, Schwartz et al. 2009, Tiller et al. 2003).

Expectancy-value decision theories were mentioned in relation to the Australian decision support booklet (Fishbein 1963, Tiller et al. 2003). Expectancy-value decision theories postulate that values are central in determining a person's attitude towards an option and thus their final choice. Value clarification exercises in decision support interventions therefore aid decision making by helping decision makers examine their values and beliefs about the different options. However, no details on how these theories informed the design of the Australian intervention were provided (Tiller et al., 2003).

Multi-attribute Utility Theory (MAU), which is derived from Expected Utility Theory (Bernoulli 1954), was used in the development of the interactive CD-Rom intervention by Kaufman et al. (2003). MAU theory postulates that the best option is the one which maximises positive and minimises negative outcomes and can be identified by weighing different attributes of options (Carter et al. 1986, Keeney and Raiffa 1993, Torrance et al. 1982, Weber and Borchering 1993). The authors describe in detail how MAU theory was applied during development of the value clarification exercise in this intervention (Kaufman et al. 2003).

Overall, few of the interventions included in this study stated an underlying theory and in those that did, most did not report how this theory was actually applied to the development process. Furthermore, those theories which were used primarily applied to the information and value clarification components of the interventions and did not explicitly cover appraisal and coping as processes that are intrinsically related to decision making.

6.4 Applying the CODE framework

The CODE framework, which describes patient decision making and coping as interlinked processes, has been discussed elsewhere (see Chapter 2; Witt et al. 2012). In the previous chapter, this framework was applied to decisions about RRSO. The content of patient decision support interventions and materials in this study was mapped onto the questions within the RRSO-adapted CODE framework to determine which questions were addressed by these tools, and which were not. Interventions that address most of the questions proposed within the RRSO-adapted CODE framework may support decision making, as well as coping processes, in the context of increased risk of ovarian cancer, thereby providing a more holistic support tool for deliberations about RRSO.

6.4.1 Coverage of primary appraisal content

During analysis, two types of interventions had to be distinguished: those for independent use by patients and those for use with a facilitator or health professional. The former generally covered many questions posed during primary appraisal within their information content, while the latter covered far fewer questions, mainly in the 'presentation of options' and 'preference construction' phases (Figure 6.2; Bhavnani et al. 2002, Kurian et al. 2012, Pell et al. 2002, van Roosmalen et al. 2004a). However, it should be noted that, while not explicitly stated, questions may be discussed in conversation with the health professional delivering the intervention.

Interventions for women at increased risk of ovarian cancer that consider RRSO as a strategy to prevent ovarian cancer covered information regarding ovarian cancer, risk, RRSO and other management options most often, thereby supporting cognitive appraisal of the health condition and management options (Healthwise 2011b, Mayo Clinic 2011, Tiller et al. 2003, van Roosmalen et al. 2004b).

Figure 6.2 - The CODE framework assessment sheet and coverage of questions during primary appraisal by interventions in the review

| | Disclosure of ovarian cancer risk status | Presentation of choice | Presentation of options (RRSO or symptom awareness) | Preference construction |
|--------------------------|--|--|---|--|
| Cognitive | What is ovarian cancer (OC)? ^{2,3a,4,8} What is OC risk? ^{1,2,3a,7,8} How high is my risk? ^{3a,5,6,7} | What does 'choice' mean in this context? | What is risk-reducing bilateral salpingo oophorectomy (RRSO) / symptom awareness? ^{1,2,3a,4,5,6,7,8} Why is symptom awareness the only alternative? | Is this the right time to decide? ^{2,3b,4} |
| Primary Appraisal | What caused me to have an increased risk of OC? ^{2,3a} How does my family history influence OC risk? ^{1,2,3a,7,8} Do I have a faulty gene? ^{1,2,3a} Does my risk change over time? ^{2,7} How could my risk affect my children? How will my risk affect my life? | Why is there a choice for women at risk of OC? When should I decide? ^{2,4} What is at stake in this choice? ² Who can decide? ^{1,5,8} Who else should I involve? ² | What does RRSO involve? ^{1,2,3a,4,5,6,7,8} How long would it take to recover from RRSO? ^{1,2,3a,4,6,8} What would happen if I do not have surgery? ^{1,5,8} What are the risks and benefits of RRSO / symptom awareness? ^{1,2,3a,3b,4,6,7,8} What are the complications of RRSO? ^{2,3a,4,6} Would I go into an early menopause? ^{1,2,3a,4,5,6,7,8} Would I have to take hormone replacement? ^{2,3a,4,6,8,9,10} How would RRSO affect my life? ^{2,3a,3b,4,6,9,10} Do I need to have my womb removed as well? ^{2,4,8} | Is RRSO / not having RRSO congruent with my/my family's/partner's/doctor's beliefs goals and values? ^{1,2,3b,6,8,9,10} Have I completed my family? ^{2,4,6} Can I afford to take time off? |
| Emotional | What can be done about my OC risk? ^{1,2,3a,4,7,8} What are my past experiences of OC? How do I feel about OC and my risk? | Can I change my mind? How can I decide? ^{1,2} How do I feel about choosing? Can I deal with choosing myself? | Is there a chance cancer might be found during RRSO? ¹ Is there any screening available for OC? ^{1,2,3a,3b,4,7} Is there anything else I can do to avoid OC? ^{1,2,4,7} What did others decide? How did they fare? ^{1,2,3a,7} Can I make sure I get a good surgeon in a good hospital? Do I have experiences that could help me imagine what it would be/feel like if I had / didn't have RRSO? How do I feel about having / not having RRSO? | Could I get screening or some other additional care? ^{1,2,3a,3b,4,7} Do I think I could detect the symptoms of OC? Do I feel ready to decide? ^{1,2,3b,8} How likely is it that I will regret having / not having RRSO? |

Intervention number indicated in superscript if item is addressed (items that were not addressed in any of the interventions indicated in red):

RRSO to prevent ovarian cancer:

- 1 – Healthwise 2011
- 2 – Tiller 2003
- 3a – Roosmalen 2004b
- 3b – Roosmalen 2004a
- 4 – Mayo Clinic 2011

RRSO to prevent breast cancer:

- 5 – Kurian 2012
- 6 – Metcalfe 2007 & Culver 2011
- 7 – Kaufman 2003

RRSO with planned hysterectomy:

- 8 – Healthwise 2012
- 9 – Bhavnani 2002
- 10 – Pell 2002

In contrast, interventions focused on RRSO as an option to prevent breast cancer included less information about effects of surgery on ovarian cancer risk, although some did make reference to the reduction in risk of ovarian cancer following RRSO (Culver et al. 2011, Kaufman et al. 2003, Kurian et al. 2012, Metcalfe et al. 2007). Similarly, interventions for women undergoing a planned hysterectomy only included limited information on ovarian cancer risk or alternative management options, as these interventions were not aimed at women with an increased risk of ovarian cancer (Bhavnani et al. 2002, Healthwise 2012, Pell et al. 2002).

Overall, coverage of questions during primary appraisal of the health threat and available options was adequate, with the majority of interventions addressing at least one, and frequently more, questions posed in the framework. Most supported cognitive appraisal processes by providing factual information. Fewer supported emotional appraisal by encouraging patients to envisage how their risk and potential outcomes of options might affect their life and to consider their own experiences and feelings. None explained why watchful waiting / symptom awareness is the only official alternative management option to RRSO. However, this may be due to the fact that none of the interventions aimed at high risk women was developed for the UK population (which currently has no ovarian screening programme) and guidelines for ovarian cancer screening and chemoprevention may differ between countries. Therefore, while other management options may be recommended in some countries, the UK guidelines currently do not recommend ovarian screening or chemoprevention (National Institute for Health and Care Excellence 2011, 2013).

Coverage of questions regarding choice was less comprehensive. Only five interventions addressed questions posed within the primary appraisal of choice section of the framework (Healthwise 2011b, 2012, Kurian et al. 2012, Mayo Clinic 2011, Tiller et al. 2003). The decision support booklet by Tiller et al. (2003) gave an indication of when surgery is most effective, listed disadvantages, encouraged

patients to involve others, such as genetic counsellors and family members, and provided a step by step guide to reaching a decision. Conscious appraisal of emotional responses to the choice was not encouraged by interventions in this review; therefore, content was again biased towards cognitive appraisal.

Preference construction was supported by interventions that included explicit value clarification exercises and thereby helped patients to assess congruence between options and values, to consider timing and to appraise readiness to decide (Bhavnani et al. 2002, Culver et al. 2011, Healthwise 2011b, 2012, Metcalfe et al. 2007, Pell et al. 2002, Tiller et al. 2003, van Roosmalen et al. 2004a). Items within most of these value clarification exercises exclusively covered medical or physical attributes of option(s), such as reduction in cancer risk and side effects (Bhavnani et al. 2002, Pell et al. 2002, Tiller et al. 2003, van Roosmalen et al. 2004a). Psychological and emotional aspects, such as worry, anxiety and body image, were included less frequently (Culver et al. 2011, Metcalfe et al. 2007). The intervention by Kurian et al. (2012) showed the effects of different management strategies, but did not explicitly ask the patient to attribute values to these outcomes. The intervention by Kaufman et al. (2003) included a value clarification exercise; however, as stated earlier, it did not include options for risk reduction of ovarian cancer.

6.4.2 Coverage of secondary appraisal content

Coverage of questions posed during secondary appraisal of cancer risk, choice and options was poor (Figure 6.3). While most interventions included information about ovarian cancer, risk and available options, enabling patients to find out more by themselves and referring patients to health professionals for further discussion, only one encouraged seeking informational, instrumental and emotional support from family and friends (Tiller et al. 2003). None touched on emotion focused coping options such as religious coping, positive reframing, relaxation or meditation.

Figure 6.3 - The CODE framework assessment sheet and coverage of questions during secondary appraisal by interventions in the review

| | Disclosure of ovarian cancer risk status | Presentation of choice | Presentation of options (RRSO or symptom awareness) & Preference construction |
|----------------------------|---|---|--|
| Problem-focused | Can I find out more? ^{1,2,3a,4,6,7,8} Can my doctor help? ^{1,2,3a,4,5,6,7,8} Can I trust my doctor? | Can I create an action plan to approach this decision? ^{1,8} | |
| Secondary Appraisal | Can I seek instrumental / informational support from my family / friends / a peer or support group? ² Can I seek emotional support from my family / friends / a peer or support group? ² Can I turn to my faith? | Can I seek instrumental / informational support from my family / friends / a peer or support group? ² Can I seek emotional support from my family / friends / a peer or support group? ² | Is there someone or something that could help me to compare and weigh my options? ^{1,2,3b,5,6,8,9,10} Can I speak to someone who has already made this decision and is living with the consequences? |
| Emotion-focused | Can I express my fear of cancer? Can I do something to stop thinking about my OC risk? Can I do something to reduce the stress my OC risk is causing me? Could I ignore my OC risk? | Can I let someone else decide for me?^{1,8} Could I refuse to choose and just not make this decision? | Can I seek instrumental / informational support from my family / friends/ a peer or support group? ² Can I seek emotional support from my family / friends / a peer or support group? ² Can I do something to stop thinking about this decision? Can I do something to reduce the stress this decision is causing me? |

Intervention number indicated in superscript if item is addressed (items that were not addressed in any of the interventions indicated in red):

- | | | |
|--|---------------------------------------|--|
| <u>RRSO to prevent ovarian cancer:</u> | <u>RRSO to prevent breast cancer:</u> | <u>RRSO with planned hysterectomy:</u> |
| 1 – Healthwise 2011 | 5 – Kurian 2012 | 8 – Healthwise 2012 |
| 2 – Tiller 2003 | 6 – Metcalfe 2007 & Culver 2011 | 9 – Bhavnani 2002 |
| 3a – Roosmalen 2004b | 7 – Kaufman 2003 | 10 – Pell 2002 |
| 3b – Roosmalen 2004a | | |
| 4 – Mayo Clinic 2011 | | |

Two interventions allowed users to create an action plan using their preliminary decision as a starting point to decide about the next steps they would take (Healthwise 2011b, 2012). These aids also emphasised that patients could let their doctor make a recommendation for them.

Interventions with explicit value clarification exercises indirectly supported coping by providing a means of comparing and weighing options, thereby addressing the corresponding question posed during secondary appraisal in the option and preference construction section of the framework. The intervention by Kurian et al. (2012) allowed users to directly see the effects of having/not having RRSO on survival and therefore also allowed comparison of options, although individual patient values were not considered explicitly. None of the interventions provided information about peer support and possibilities to hear from or contact others who have gone through the decision.

6.5 Discussion

6.5.1 Discussion of findings

In the current chapter, eleven decision support interventions were identified that included the option of RRSO. These were examined with regards to their format, design, information content, value clarification components, effectiveness, and use of an underlying theory or framework. Interventions varied widely in their format and design as well as in the types and detail of information included. Two basic formats of interventions were distinguished: those designed for independent use by patients, and those delivered by trained facilitators or health professionals. Previous research has found that most interventions available to date fall within the former category (Elwyn et al. 2010a). Therefore, it is not surprising that the majority of interventions in this study were comprised of self-administered booklets, websites or other types of decision support materials. Among interventions delivered by trained personnel, Elwyn et al. (2010a) make a further distinction, differentiating between interventions for use in face-to-face clinical encounters and

interventions delivered across several socially-mediated encounters. The former may include interventions such as those described by Bhavanani et al. (2002) and Kurian et al. (2012), which are administered in face-to-face encounters. The latter may include interventions such as that trialled by van Roosmalen et al. (2004a), delivered over a number of encounters, which may be face-to-face or via phone (Elwyn et al. 2010a).

6.5.1i Information content: The information provided within these formats differed greatly. Interventions designed for independent use by patients tended to include more comprehensive information on the decision and management options, plus additional materials, such as patient stories. Interventions delivered by trained personnel usually only included a minimum amount of information. These types of interventions are often preceded by counselling sessions or other written materials that provide information relevant to the decision. Their primary aim, therefore, is not to support the exchange of information, but rather to facilitate the preference construction stage of decision making (Charles et al. 2005). However, some have argued that a requirement for a minimum amount of information in all patient decision support interventions would be advantageous to achieve a level of standardisation (Elwyn et al. 2009b, Joseph-Williams et al. 2013).

6.5.1ii Value clarification: Most interventions in the current study included a type of value clarification exercise to elicit patient preferences. Interactive value clarification exercises allow patients to become more engaged and to actively attribute their personal values to different outcomes (Llewellyn-Thomas 2009). This can help to visualise values, both for the patients themselves and others, such as their clinician or family (Llewellyn-Thomas 2009). Explicit value clarification exercises have been shown to lead to more informed, value-adjusted decisions, hence it is not surprising that many interventions included a means of value clarification (Stacey et al. 2011).

6.5.1iii Evaluation: A number of interventions had been evaluated in randomised trials and small pilot studies. A popular outcome measure for trials of

patient decision support interventions is decisional conflict (Charles et al. 2005, O'Connor 1995). Three studies in the current analysis used this as the primary outcome measure (Metcalf et al. 2007, Schwartz et al. 2009, Tiller et al. 2006). All found a significant reduction in decisional conflict post-intervention. This is in accordance with the most recent Cochrane review of patient decision support interventions, which concluded that decisional conflict was reduced by interventions (Stacey et al. 2011). However, reduction in decisional conflict was no longer significant at 6 months post-intervention in one study (Tiller et al. 2006) and was only significant in the group of patients that was undecided at randomisation in another study (Schwartz et al. 2009). This indicates that there may be limitations to the effects of decision support interventions on decisional conflict. Decision regret may be a more appropriate measure of the longer-term effects of patient decision support interventions (Joseph-Williams et al. 2010).

Information transfer is another popular outcome to evaluate intervention effectiveness, most commonly measured using knowledge assessments (Charles et al. 2005). Three studies included a measure of knowledge (Metcalf et al. 2007, Tiller et al. 2006, van Roosmalen et al. 2004b). Two of these found significant improvements in knowledge post-intervention (Metcalf et al. 2007, van Roosmalen et al. 2004b), whereas one study showed small to medium improvements that were significant at 2 weeks post-intervention, but did not persist 6 month post-intervention (Tiller et al. 2006). Overall, these findings are in agreement with those of the Cochrane review, which showed that decision support interventions increase knowledge (Stacey et al. 2011).

Psychological outcomes, such as distress, anxiety and intrusive thoughts, were assessed in four studies. Tiller et al. (2006) found no significant effect of their decision support booklet on psychological outcome measures, including anxiety and intrusive thoughts. Similarly, Metcalf et al. (2007) found no significant effect of their intervention on cancer-specific distress. Van Roosmalen et al. (2004a) showed that their SDM intervention led to significant long-term reductions in depression,

intrusive thoughts and general health, but not anxiety. Hooker et al. (2011) found that a CD-Rom based intervention (Kaufman et al. 2003) led to sustained higher levels of distress 1 month post-intervention. The results of the 2011 Cochrane review confirm these contrasting findings, as the authors were unable to establish fully whether patient decision support interventions had significant effects on anxiety, distress or intrusive thoughts, indicating that the evidence for such effects is variable (Stacey et al. 2011).

6.5.1iv Theoretical basis or framework: Only three of eleven interventions explicitly mentioned one or more theories that had guided development (Kaufman et al. 2003, Metcalfe et al. 2007, Tiller et al. 2003). This is in agreement with a previous study analysing theory use in a set of patient decision support interventions included in a meta-analysis (O'Connor et al. 2007), which found that only 34% were grounded in theory (Durand et al. 2008). Due to this apparent lack of theories to guide goal setting, development, testing and evaluations of patient decision support interventions, assessing and explaining the impact of these tools is difficult (Charles et al. 2005, Durand et al. 2008). Without a theoretical basis or framework, selected outcome measures may not accurately measure concepts linked to the goals set out during development of the intervention (Charles et al. 2005). Elwyn et al. (2011b) examined a number of popular decision making models with a view to identify their potential to contribute to the development of information and deliberation components and to inform the selection of outcome measures. They noted that most theories describe how people make decisions, but do not readily address how interventions might help people during this process (Elwyn et al. 2011b). Therefore, it may not be surprising that many interventions in the current study were found to be lacking a solid theory base.

6.5.2 Use of the CODE framework as an assessment tool

The CODE framework was developed to provide a framework that may be applied more readily to the development of decision support interventions (Witt et al. 2012). Informed by a number of theories and models relevant to decision

making and coping, it sets out a series of questions. Provision of answers or guidance relating to these questions within an intervention may support cognitive, as well as emotional, appraisal processes. Additionally, the framework includes questions exploring potential coping options, the answers to which might improve awareness of different coping options and ultimately lead to improved coping with the health threat, related decision making processes and associated emotions.

Assessment of interventions against the RRSO-adapted version of the CODE framework revealed that cognitive appraisal of the health threat and available options was usually adequately facilitated through the provision of information within interventions. While cognitive appraisal appeared well supported, emotional aspects of appraisal were rarely addressed. This could indicate that it is too ambitious to expect patient decision support interventions to support emotional appraisal, as this process is a very personal experience that will differ greatly from patient to patient. However, encouraging patients to consciously consider their feelings and to envisage how outcomes might affect their life could facilitate emotional appraisals and affective forecasting (Loewenstein et al. 2001, Wilson and Gilbert 2005).

There also appeared to be a substantial lack of coping advice provided within interventions included in the current study. As patients deliberate about healthcare decisions, they have to cope with a large quantity of new, and often threatening, information regarding their health and their options. Additionally, the mere existence of choice in healthcare and of being asked to play an active part in decision making may be a new concept for some patients that could result in heightened feelings of vulnerability and anxiety (Quill and Cassel 1995, Say et al. 2006). Coping advice may therefore be beneficial in aiding patients to cope more effectively and to regulate their emotions in response to the various threats they are facing during deliberation.

6.5.3 *Results in context*

Numerous decision support interventions have been published in recent years with numbers steadily increasing (Ottawa Hospital Research Institute 2013, Stacey et al. 2011). However, only a very small proportion of these interventions is aimed specifically at women at increased genetic risk of ovarian and/or breast cancer, such as carriers of mutations in BRCA. Of the interventions that are designed for this population, most are concerned with genetic testing or breast cancer risk reduction strategies (e.g. (Healthwise 2011a, McInerney-Leo et al. 2004, Wakefield et al. 2008a, Wakefield et al. 2008b). It has been noted previously that interventions to help women make decisions about risk management options, including RRSO, would be useful (Fang et al. 2005, Howard et al. 2009a). However, the current study only identified eleven interventions that included information about RRSO, only five of which specifically focused on reducing elevated ovarian cancer risk.

Furthermore, interventions included in the current study were mostly developed for non-UK populations. Most had been developed in the United States, two in the Netherlands, one in Canada and one in Australia. As guidelines and available options may differ between countries, these interventions cannot be readily adopted in the UK. For example, in the USA oral contraceptives are recommended for carriers of mutations in BRCA1/2 to achieve a reduction in the risk of ovarian cancer, whereas UK guidelines do not recommend the use of oral contraceptives solely for this purpose (ACOG Committee on Practice Bulletins-Gynecology 2006, National Guideline Clearinghouse 2011, National Institute for Health and Care Excellence 2013). This demonstrates the need for interventions tailored to the UK population that conform to UK policy and guidelines.

Only two interventions had been specifically developed for UK women and these interventions focused on decisions regarding RRSO as a concomitant procedure to a planned hysterectomy (Bhavnani et al. 2002, Pell et al. 2002). This shows the current lack of decision support available outside clinical consultations

tailored to the needs of women at increased risk of ovarian cancer in the UK. It was previously suggested that information regarding RRSO and other options, as well as additional forms of support, would be beneficial to address women's knowledge gaps regarding their management options and to support women during deliberation (Brain et al. 2004). Furthermore, women themselves noted that it was difficult to obtain reliable information and that they had received conflicting advice from different health professionals (Chapter 4). This suggests that additional materials and interventions would likely be well received by the target population.

However, while decision support in healthcare is a rapidly growing field and more and more decision support interventions are developed, a substantial number of new interventions are never implemented in clinical practice (Bhavnani and Fisher 2010, Elwyn et al. 2010b, Frosch et al. 2011). Four of the interventions included in this study were not implemented after initial evaluation, including the two interventions developed in the UK (Bhavnani et al. 2002, Pell et al. 2002, van Roosmalen et al. 2004a, van Roosmalen et al. 2004b). This shows that considerable effort goes into the design and primary evaluation of interventions, particularly in academia, but only a limited number actually become available to patients (Bhavnani and Fisher 2010, Elwyn et al. 2008, Elwyn et al. 2013c, Frosch et al. 2011, Harter et al. 2011). This may be due to a variety of factors. Three of these interventions were designed for delivery by trained facilitators, which is associated with substantial staff and time requirements that may not be feasible in clinical practice and could hamper implementation (Bhavnani et al. 2002, Pell et al. 2002, van Roosmalen et al. 2004a). Two of these were also outdated by the time primary evaluation was complete due to a change in clinical guidance (Bhavnani et al. 2002, Pell et al. 2002). Others might encounter problems due to a lack of willingness on the part of health professionals to use and recommend them (Coulter 2010).

This lack of implementation beyond the duration of research has led some authors to suggest that shorter tools, which require less time and money for development and slot more readily into the routine flow of consultations, may be

easier to implement (Elwyn et al. 2013b, Marrin et al. 2013). These tools might bypass some of the practical challenges that have stifled longer, self-administered interventions in the past, such as a lack of time to read through and digest information or a need for follow-on consultations post-intervention (Bhavnani and Fisher 2010). However, the amount of information that can be delivered in such short tools is limited and may not cater for the needs of those patients who wish to adopt a deliberative decision making approach (e.g. delayed deciders; see section 4.4.1vii, Chapter 4). Therefore, a 'one-size-fits-all' intervention may not be feasible. However, there may be scope to develop multipart decision support interventions that slot into clinical workflows and support different approaches to deliberations, such as fast and frugal, as well as deliberative, approaches. For instance, a two-part intervention consisting of a short and a more extensive tool could address these issues. The shorter component may be readily implemented into clinical workflows and could cater for the needs of immediate deciders who use fast and frugal heuristics to make decisions and do not wish to learn further details. The more extensive part could act as a complementary resource for those patients who wish to find out more, take a deliberative approach and/or take longer to decide (delayed deciders). Chapter 7 outlines the development of such a two-part decision support intervention.

6.5.4 Strengths and limitations

The current study aimed to identify a range of patient decision support interventions that included information about RRSO. Therefore, the search strategy was purposefully wide in order to include interventions with information about RRSO that were not aimed at women at increased risk (e.g. those which considered RRSO as a concomitant procedure to hysterectomy). Supplementary searches of patient decision support intervention databases and websites identified further interventions, which had been developed by commercial developers and would not have been identified through the literature search. Therefore, it is likely that the interventions analysed in this chapter represent the majority of interventions

developed so far that include the option of RRSO. However, due to the search strategy employed, interventions in languages other than English might have been missed, unless reports about these interventions were published in English. Although this may have resulted in the omission of some interventions, it is unlikely to have had a major effect, as most interventions to date have been published in English speaking countries (Stacey et al. 2011). Furthermore, non-English interventions with associated publications in English, such as the two Dutch interventions, were included in the study (van Roosmalen et al. 2004a, van Roosmalen et al. 2004b).

This is the first study to attempt using an adapted version of the CODE framework to assess the content of interventions (Witt et al. 2012). The framework's structure allows relatively easy application as an assessment tool, as reviewers simply need to examine whether the intervention content addresses questions outlined in the framework, without the need for extensive adaptation. Therefore, this study demonstrates that adapted versions of the CODE framework may be readily used to assess the content of decision support interventions relating to other preference-sensitive healthcare decisions. However, as this was the first time the framework was employed as an assessment tool, no inferences can be made about the quality of interventions using only the results from assessment with the adapted CODE framework. Therefore, interventions were further examined using a data extraction form based on the IPDAS checklist to supplement the results of assessment with the CODE framework and to additionally provide information about intervention design, theory-base and effectiveness.

Some limitations of this approach should be noted. The data extraction form used to assess information and other content of interventions was not validated prior to application. However, it was based on items in the IPDAS checklist, which was developed through a Delphi consensus process and has since been used to assess a number of patient decision support interventions (Elwyn et al. 2006, Elwyn et al. 2009b, Ottawa Hospital Research Institute 2013). Furthermore, during

development the extraction form was applied to two interventions that included the option of RRSO and subsequently amended to improve relevance of the items. Detailed validation of the form itself was not possible due to time constraints. In the future, the extraction form would benefit from further content, as well as construct, validation to ensure items are clear and assess chosen constructs satisfactorily.

During analysis, only two interventions were assessed by two independent raters. Ideally, all interventions should have been double rated; however, due to time and staff limitations this was not possible. Inter-rater consistency was high and any discrepancies were resolved through discussion. In order to increase consistency in the ratings of the remaining interventions, the two interventions were double rated and discussions were scheduled before the other interventions were assessed, so that rater 1 (Jana Witt) could then rate the remaining interventions with this in mind.

6.5.5 Conclusions

Out of eleven patient decision support interventions identified that included the option of RRSO, only five were focused on managing risk of ovarian cancer in women at increased genetic risk. None of these had been developed for the UK population nor conformed to UK guidelines. Therefore, none are readily adoptable in the UK. Furthermore, while interventions focused on information provision to support cognitive appraisal, most lacked guidance on emotional appraisal, coping and emotional well-being, thus insufficiently supporting these aspects of decision making.

Chapter 3 highlighted the complexity of decisions about RRSO and Chapter 4 emphasised that women in the UK struggle with this decision and would welcome additional support materials. Therefore, there is scope for a patient decision support intervention tailored to the needs of women at increased genetic risk of ovarian cancer in the UK that conforms to UK guidelines and includes coping advice.

Ideally, this intervention would consist of two complementary parts, a short tool, which can be readily embedded into clinical workflows, and a more extensive tool, which provides detailed information, coping advice and a value clarification exercise. Chapter 7 outlines the development of such an intervention.

7 Developing a decision support intervention for women at increased risk of ovarian cancer

7.1 Introduction

Decision making regarding risk-reducing surgery in the context of increased genetic risk of cancer is complex, as shown in Chapters 3 and 4. Women making decisions about risk-reducing bilateral salpingo-oophorectomy (RRSO) consider a number of medical, psychological, social and other factors during their deliberations and often struggle with making a final decision (Chapters 3 and 4; Fang et al. 2005, Howard et al. 2009a, Lifford et al. 2012). Currently, decision support for these women is limited to consultations in genetics services and few evidence-based additional support materials exist. Patient decision support interventions that include the option of RRSO often are not specifically aimed at women at increased genetic risk, and many have been developed outside the UK context (Chapter 6). These findings suggest that an intervention specifically aimed at women in the UK, who are at increased genetic risk of ovarian cancer and making decisions about RRSO, would be useful to complement clinical genetics services. It has previously been argued that deliberations and coping are linked, interdependent processes (Chapters 2 and 5; Witt et al. 2012). Hence, an intervention that facilitates emotional, as well as cognitive, processing and appraisal of coping options might benefit patients and provide a more holistic approach to decision support than educational materials alone. Therefore, this chapter aims to use the RRSO-adapted CODE framework (Chapter 5) as a theoretical basis to develop a decision support intervention for women at increased genetic risk of ovarian cancer in the UK that supports cognitive and emotional appraisal, and coping processes.

As discussed in Chapter 4, the preference construction pathway can vary depending on women's values, goals and past experiences as well as various barriers and facilitators that may influence deliberations (Howard et al. 2011). Women may be able to reach an immediate decision based on fast and frugal heuristics (immediate deciders) or may prefer a more deliberative approach and take longer to reach a decision (delayed deciders) (see section 4.4.1vii, Chapter 4). It is likely that these types of deciders differ in information processing styles, preferences for mode of delivery and amount of information, emotional appraisal and need for emotional support, as well as coping strategies. Therefore, a one-size-fits-all solution to decision support in this context is unlikely. Hence, the patient decision support intervention developed in this chapter is split into two parts: a brief Option Grid and a longer tool called the 'Oophorectomy Decision Explorer' (OvDex). It was envisaged that this separation would cater for the different deliberation styles (i.e. immediate deciders and delayed deciders, Chapter 4) and would additionally aid implementation. The stand-alone Option Grid was designed to be used during consultations to initiate discussions about options and to enable patients to highlight the issues most important to them (Elwyn et al. 2013b). It may, in conjunction with advice from a health professional, provide sufficient information for immediate deciders to base their decision on. The grid can be taken home after the consultation to act as a memory aid and to support discussion with important others. If women wish to learn more about their risk and available management options, the grid then refers patients to OvDex. This may be especially helpful for delayed deciders. The Option Grid may additionally enable faster implementation of the whole intervention, as a short grid may be more readily integrated into clinical workflows than more extensive tools (see section 6.5.3, Chapter 6; Elwyn et al. 2013b).

It has previously been recommended that goals of patient decision support interventions should be defined prior to development and that developers should

adhere to relevant theory throughout (Charles et al. 2005, Durand et al. 2008). The intervention developed in this chapter will have four main goals:

1. Provide evidence-based information to support appraisal processes, in order to improve knowledge and accuracy of risk perceptions and to encourage deliberation about choice/options (Janis and Mann 1977, Leventhal et al. 1980, Power et al. 2011, Svenson 1992, Witt et al. 2012);
2. Provide information in a way that facilitates affective forecasting, in order to improve predictions of reactions to and impacts of future states and to support emotional appraisals (Wilson and Gilbert 2003, 2005);
3. Provide coping advice to improve awareness of coping options and support coping efforts, in order to support deliberation and reduce emotional distress and anxiety (Balneaves and Long 1999, Lazarus et al. 1985, Witt et al. 2012);
4. Provide tools for value clarification to support preference construction, in order to reduce decisional conflict and improve value-adjustment of decisions (Gigerenzer 2004, Janis and Mann 1977, Svenson 1992).

The intervention developed in this chapter aims to achieve these goals by providing answers to the questions pertinent to the decision, as outlined in the RRSO-adapted CODE framework (Chapter 5). Cognitive appraisal may be supported by educational materials offering factual information in response to questions. In contrast, emotional appraisal may not be sufficiently supported by factual information alone, as it has been established that individuals often struggle to accurately forecast the duration and intensity of emotions resulting from future events (Wilson and Gilbert 2005). This is due to the fact that they do not consider other events demanding their attention at the time and tend to underestimate how quickly they adjust to unexpected outcomes, whether positive or negative (Wilson and Gilbert 2005). In particular, negative outcomes or losses are often predicted to

have a greater impact than they actually do, leading individuals to avoid losses when making decisions (Kermer et al. 2006). Individuals also tend to shy away from irreversible options, such as surgery, even though they may be more satisfied with an irreversible option (Gilbert and Ebert 2000). Furthermore, when making predictions about the future, emotions experienced in the present can contaminate imagined reactions to future events (Gilbert et al. 2002). Therefore, apart from providing factual information, answers should be written in such a way as to facilitate emotional appraisal processes, affective forecasting and consideration of coping options (Elwyn et al. 2011b, Wilson and Gilbert 2005). Option Grids support value clarification and fast and frugal decision making through direct comparison of options (Gigerenzer 2004, Marrin et al. 2013, Svenson 1992). Explicit value clarification exercises enable patients to weigh gains and losses associated with the option of RRSO, thereby eliciting their personal values and allowing them to visualise their preference (Elwyn et al. 2011b, Janis and Mann 1977, Svenson 1992).

7.2 Methods

7.2.1 Focus group and interview data

Recruitment, data collection and data analysis for focus groups and interviews were described in detail in Chapter 4 (see section 4.2). During the focus groups and interviews, participants were asked to share their views on: (i) the decision making process about RRSO (reported in Chapter 4) and (ii) the content and format of a possible decision support intervention. Any comments on the latter items, i.e. scope for decision support, topics to be covered in an intervention, preferred format, style of delivery and layout, were coded under the theme of 'decision support' (Coding Frame in Appendix 7.1). During these discussions participants were shown examples of a comparison table of options and a value clarification exercise. Details of the coding process and analysis are described in Chapter 4, along with data regarding the decision making process about RRSO. Data on ideas for format, layout and content are reported in section 7.3.1 of the present chapter. Quotes presented in this chapter were selected as examples and were

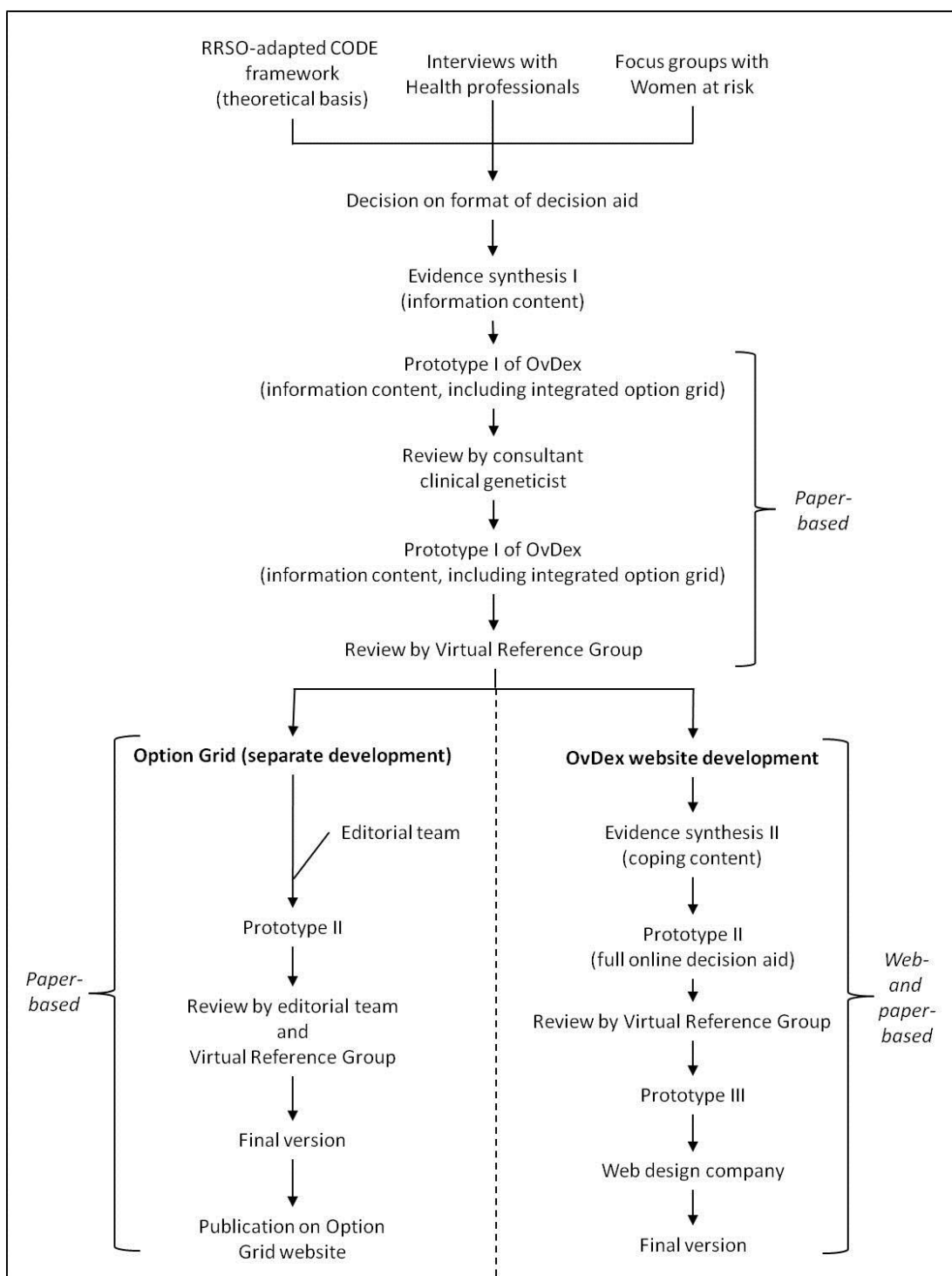
edited to remove word repetitions and irrelevant sections. Where content has been removed this is indicated by a [...]. Quotes from focus groups are denoted as P[1/2/3][S/C/L], where the number describes the participant and the final letter indicates the location (S=Swansea, C=Cardiff and L=London). Quotes from health professionals are denoted with HP.

7.2.2 Intervention development process

The development process is depicted in Figure 7.1 and followed the MRC recommendations for the development of complex interventions (Craig et al. 2008). The underlying theoretical basis, i.e. the CODE framework, was developed in previous chapters (Chapters 2 and 5) and best available evidence was identified through systematic literature searches (see sections 7.2.3 and 7.2.4). The final intervention comprises two parts: Option Grids for use in clinic (part 1) and a more extensive OvDex website / booklet (part 2). Feedback from women at increased genetic risk of ovarian cancer and health professionals guided decisions on format and design, as well as content (see section 7.3.1). The RRSO-adapted CODE framework acted as a theoretical and practical basis to guide development. The individual questions addressed in the intervention were based on questions proposed in the framework. Coping advice was included in the intervention in accordance with the CODE framework, which postulates that each phase of deliberation gives rise to an individual coping process.

The Option Grids were developed according to the Option Grid framework V0.5 (Option Grid Collaborative 2013) and versions of the grids were also integrated in OvDex. As implications of RRSO differ substantially for pre- and post-menopausal women, two separate Option Grids were developed, one for each population. The longer OvDex tool was developed following recommendations for development of decision support interventions outlined previously (Elwyn et al. 2011a, Volk and Llewellyn-Thomas 2012). The term 'intervention' in this chapter will refer to both parts of the intervention, encompassing the Option Grid as well as the longer OvDex tool.

Figure 7.1 - Intervention development process



Two literature reviews were undertaken to collate evidence about ovarian cancer risk, risk management options and coping interventions. Information content was developed using the evidence generated through evidence synthesis I (see section 7.2.3). Coping advice in OvDex was developed using evidence generated through evidence synthesis II (see section 7.2.4). Prototypes of the Option Grid and OvDex were reviewed at multiple time points by researchers, health professionals and patients (see section 7.2.5-7). To reduce the burden on reviewers, the Option Grid was first reviewed as an integral part of the information content of OvDex and later developed into a separate item.

Development of OvDex was split into two stages: in the first stage the information content was developed in written format; in the second stage coping advice and a value clarification exercise were developed, and the content was converted to an online format. Design of the website was completed in Adobe Dreamweaver CS5.5 (Adobe Systems Incorporated 2011) and supported by the Cardiff School of Medicine IT office. Development of the JavaScript for the value clarification exercise was supported by Cardiff University Information Services (INSVR). The website was validated using the Mark-up Validation Service W3C® (W3C 2013). An Innovation grant was awarded from the charity Tenovus, which allowed the Cardiff-based web design company DigitalMorphosis to contribute to the final stages of web development.

7.2.3 Evidence synthesis I – ovarian cancer risk and risk-reducing surgery

A systematic literature search focused on RRSO was conducted in April 2012. Databases (Embase, MEDLINE and PsycINFO) were searched via Ovid for studies published within the last ten years, between 2002 and July 2012. Search terms used in the key word search were: “risk reducing salpingo oophorectomy”, “risk reducing oophorectomy”, “prophylactic oophorectomy”, “prophylactic salpingo oophorectomy”, “risk reducing ovarian surgery” and “prophylactic ovarian surgery”. Duplicates were removed using the Ovid de-duplicate function prior to review of the abstracts. No grey literature was included in this review. Additional

papers were identified through a review of the references included in relevant papers. These included publications about the surgical menopause and hormone replacement following RRSO. Studies were critically appraised using appraisal checklists (Appendix 3.1 and 3.2) and results from studies were used as evidence to support the information content of the intervention.

Following feedback from the virtual reference group, this same search approach was deployed repeatedly when the material was updated in October 2012 and January 2013, in order to identify newly published studies relevant to the information content in the intervention.

A complementary search was also carried out using keywords “Lynch syndrome”, “HNPCC” or “hereditary non-polyposis colorectal cancer” combined with “oophorectomy” and/or “hysterectomy” in October 2012 and January 2013 to obtain further information about risk-reducing hysterectomy in conjunction with oophorectomy in the context of Lynch syndrome.

7.2.4 Evidence synthesis II – coping interventions

A systematic review of the literature concerned with coping interventions was conducted in July 2012 to identify coping interventions aimed at populations undergoing genetic testing, at individuals at risk of genetic diseases (including cancer) or at cancer patients. Databases (Embase, MEDLINE and PsycINFO) were searched via Ovid for studies published between 1996 and July 2012. Search terms used in the title search are detailed in Table 7.1. Duplicates were removed using the Ovid de-duplicate function prior to review of the abstracts. No grey literature was included in this review.

Additional papers were identified through a review of the references included in relevant papers. Any studies of interventions were critically appraised using the CASP appraisal checklists (Appendix 3.1 and 3.2) and contents of coping interventions were then summarised and used to inform the coping advice within the OvDex tool.

Table 7.1 - Search terms for title search in Ovid

| Term 1 | | Term 2 | | Term 3 |
|---------------|-----|--------------|-----|--------------|
| Coping | | Intervention | | Cancer |
| | AND | Tool | AND | |
| | | Aid | | Genetic risk |
| Self-efficacy | | Exercise | | |
| | | Advice | | Genetic test |
| | | Leaflet | | |
| | | Brochure | | |

7.2.5 *Virtual reference group recruitment*

Obtaining feedback from a virtual reference group made up of patients and health professionals is helpful in ensuring that stakeholders in the decision contribute throughout the development process. This follows the suggested best practice for the development of web-based patient decision support interventions, as proposed by Elwyn et al. (2011a).

The virtual reference group for this study consisted of patients and health professionals who had previously registered for focus groups or taken part in interviews (Chapter 4). They were recruited either directly after the focus group or interview, or were invited by email if they had registered for, but were unable to attend, the focus group. A study pack containing an information sheet and consent form was provided and individuals were given two weeks to consent.

7.2.6 *Virtual reference group data collection*

Feedback from the virtual reference group was obtained in August and November 2012. The first round of feedback focused exclusively on the information content of the intervention, which was provided to participants in written format. The second round requested feedback on all content (including a value clarification exercise and coping advice), as well as on design and layout. In the second round material was presented to participants online, with a paper version available upon request. Participants had three weeks to give feedback in each round. A feedback guide with open ended questions was also provided to support the review process;

however, participants were allowed to deviate from the suggested structure and questions in their feedback if they wished (Appendix 7.2).

7.2.7 Virtual reference group data analysis

Written feedback was obtained from participants and collated in one document. Comments were coded as 'positive', 'suggestion for improvement', 'emotional responses' and 'previously unknown facts'. Within the 'suggestions for improvements' theme several subthemes were identified according to the type of suggestion. These included 'spelling and grammar errors', 'not relevant', 'confusing or needs rewording', 'additions/expansions', 'repetition' and 'other'. For Round 2 an additional subtheme was added called 'website issues', which included any references to technical and display problems with the website. The codes were additionally analysed to review differences between patients' and health professionals' comments. Quotes presented from the virtual reference group are denoted with a VR, followed by a number. Each number corresponds to one participant. Whether the participant was a patient or a health professional is indicated in parentheses after the quote.

7.2.8 Adaptation

After each round of feedback from the virtual reference group, the collated suggestions for improvements were used to amend the content of the intervention. New and supplementary literature searches were conducted to obtain the latest research evidence to complement and update the information provided. Changes to the layout were made in response to feedback from Round 2 only.

7.3 Results

7.3.1 Focus group and interview recommendations

In focus groups and interviews, participants were asked about their views of and ideas for a decision support intervention to support women's decision making about RRSO in the context of increased genetic risk. The data highlighted how such

an intervention could be used in clinical practice and provided recommendations about the format and content of such a tool.

7.3.1i Scope for decision support: Health professionals remarked upon the current lack of appropriate materials to give to patients, commenting on the low quality and on the fact that educational materials without additional features, such as value clarification exercises, have failed to facilitate decision making.

HP6: I hate what we give. It's terrible. Our patient information leaflet. Packed full with information in very small size. [...] It's terrible.

HP7: There is information leaflets and DVDs and letters and we are still here today [...]. So I don't know whether information is the answer to the question. It will contribute to the answer, but it's not the whole answer. Certainly not.

Some acknowledged that other countries offered more information and advice in written and web-based formats, but remarked that this was not directly applicable to patients in the UK.

HP10: They [patients] can read about MD Anderson Cancer Centre [USA] but they are never going to go there. You know, what you want to know is what my local hospital can offer.

Patients spoke about the difficulty of finding information themselves, especially highlighting the problem of identifying reliable, trustworthy and relevant information via the internet.

P2S: I don't know what I haven't had, but I just feel like there is something I haven't had. There is something missing [...]. They refer you to this and they refer you to that [...]. I've gone into [the internet] [...] and you just get swamped. Depending on what website you look at it will tell you different things.

P3S: It does yeah.

P2S: And you just think: "Well I'll read that one and then I'll go into one a bit further down." And I'll think: "Well it's the same sort of thing but it's slightly different." And I just gave up then, I thought I can't be bothered.

P3S: Unless you're going on the internet and you're literally trawling through everything and trying to ignore all the crap and take in all the stuff that is relevant to you [...], unless you're a doctor really, you don't know really what's relevant to you, do you?

Professionals also emphasised the need for reliable information and could see a role for novel materials that they would refer patients to, knowing that the information within it was trustworthy and up to date.

HP2: People need to know that the decision aid is evidence-based, is reliant on strong research studies and therefore that they are able to use the important criteria when thinking about a decision...

HP5: ...if there was a decision aid that could give women clear information, I think that that would definitively be something that I could see a role for in my clinic [...]. I think it would be a very helpful resource...

Some spoke about how the intervention could be used by patients to facilitate deliberation and information processing in order to reach an informed, value-adjusted decision.

HP7: A woman could take that [home] and reflect on it as long as she wants to, return to it, read through it, think again and come back to it or whatever. And if it sort of flows logically then it's a beginning to end and she can work her way through it and probably by the end make a decision that she is happy about.

Overall, these comments suggest that an intervention to facilitate decisions about RRSO would be well received by patients and health professionals.

7.3.1ii General format: Stakeholders were consulted about their preferred format for the intervention. The majority favoured a website, pointing to the numerous advantages of online formats with regards to interactivity, ease of updating and opportunity for tailoring information.

HP5: I suppose my personal preference would be to have it online [...] I sort of feel that you can do more online. [...] I can see a lot of benefits of doing it online.

P1S: For me, I think I would rather have something on the web because I think that [...] I might go back a couple of times and put the information in and see if it comes out with the same thing, almost to test the system. [...] Maybe it's a peace of mind thing, I don't know...

P2S: I know that I would also like to think – from the point of view of the web – that it was up to date and [...] within two days there could be a change to it.

However, many pointed out that there may be issues with accessibility, if the material was only online, and therefore recommended that a printed version be available as well for those who do not have access to the internet.

HP5: [...] but I suppose for women who don't have internet access, it would be nice to have the backup of a paper-based resource as well.

P3S: ...my only other thing would be the people who [...] haven't got a computer in the house. And that's not the kind of thing you really want to be reading in your local library, is it? [...] So you would have to [have] both [web- and paper-based] options really, wouldn't you? To cover everybody.

Others emphasised the need for a printed version based on patient preferences, stating that some may prefer printed materials despite having access to the internet.

HP11: It's patient choice really, I think you should have both [paper- and web-based] because some people like to go on the computer, others just don't, so there should be a choice available.

SP2: What I tend to do is I usually [print] things off if I really need to concentrate on them, because I tend to take it in if I'm reading. [...] I would like to download it from the website or I'd print from the website and then sit down quietly and read it. [...] Because I would get a little bit frustrated with the screen sometimes, but I like the fact that it would be current and up to date.

P2C: I like it in writing, something that I can read and look at and that. I don't, I'm not very good on the computer so, for me to have a booklet in my hand would be beneficial.

These comments highlighted the need for different formats to cater for the varying needs of individual patients. It was therefore decided that the intervention material should be available in a web-based as well as paper-based format.

7.3.1iii Personalisation: Health professionals pointed out that cancer risk varies depending on family history and mutation status as well as age, that the consequences of RRSO differ substantially depending on patients' menopausal status and that prior breast cancer will prevent patients from taking HRT. Therefore, many recommended that a range of materials were needed so that the correct information could be given to each group of patients.

HP9: So I think you would [...] almost have to have different ones. Because otherwise you will confuse the women [...] who have got BRCA mutations from the women who have got a Lynch syndrome mutation because it's different.

HP6: You give me an aid, which I could use when I am 25, when I am 30, when I am 50 to make this decision [...]. I must have the appropriate pamphlet for my age and not have a broad one [...]. So, I must say personally, I very much individualise to age.

HP5: I think for post-menopausal women you might have to have a different decision aid, because the implications are very different.

In order to address these issues, it was decided to allow tailoring of the information within the intervention. Personalisation would be based on family history or mutation status, breast cancer status and age.

7.3.1iv Information content: According to the RRSO-adapted CODE framework, patients go through four phases before making a decision: appraising cancer risk, appraising choice, understanding their options and constructing preferences (Chapter 5; Witt et al. 2012). The three former phases may be

supported by the provision of relevant information and advice, while the latter may additionally require value clarification. Stakeholders made a number of recommendations regarding information content (Table 7.2). Together with the questions during primary appraisal (outlined in the RRSO-adapted CODE framework) and evidence synthesis I, these suggestions guided development of the information content to achieve goals 1 and 2 of the intervention, i.e. support appraisal processes and affective forecasting.

Health professionals and patients felt that information about *cancer risk* would be important to place the decision in context and to ensure that patients understood their own risk and how it might change depending on family history, genetic mutation status and age. Furthermore, patients were concerned about other family members, especially their children, and how their own risk might affect their offspring. In terms of introducing the idea of *choice*, professionals felt that the importance of patient values in this decision should be highlighted and that criteria for being offered and for making the choice (e.g. being at high risk, completing family) should be outlined. Additionally, they felt that patients should be encouraged to discuss their decision with important others.

Recommendations regarding information about the *options* were split into two sub-topics: 'risk-reducing surgery' and 'other options'. Apart from stating the reduction in ovarian cancer risk achieved by RRSO, professionals also highlighted that patients should be made aware of the reduction in breast cancer risk following surgery and of the physical, psychological and social effects of RRSO. Patients themselves were keen to obtain information about surgical procedures and risks, chances of finding cancer during surgery and the cancer risk following surgery. For pre-menopausal women, premature menopause and HRT were particularly important issues, as previously discussed in Chapter 4. Both groups expressed a need for information about menopausal symptoms, HRT and non-hormonal alternatives.

Table 7.2 - Recommendations regarding information content of the patient decision support intervention

| CODE Phase / Subtopic | Recommendations from patients and health professionals |
|-----------------------|---|
| Cancer Risk | HP9: The information that I think is important to them [patients] is to understand the risk, and not just to understand the lifetime risk, [...] but to get an idea at least of how the risk rises over time. |
| | P2L: I think you really want to know, well for me, it was important to know the statistics, you know, the percentages and the risks |
| | P2C: Well we have to think about our children, don't we? [...] it would be helpful to know what are the chances of me developing it so that they perhaps knew how it was going to affect them really, in later life. |
| The Choice | HP2: The other thing I think that is important [...] is to encourage people to think about what their preferences would be [...]. |
| | HP7: We generally standardly say that one of the criteria is that you know you have completed your family [...]. That you are high risk and that you have completed your family. |
| | HP4: There's the danger of going to the group that you think is the answer you want, rather than actually ask people who might challenge you a little bit more. And actually maybe your family, who you disagree with, [...] are actually challenging you because they [...] might be concerned that you might react differently. It's complicated sometimes. |
| Options | |
| Risk-reducing surgery | HP1: I think it might help people if they knew – and I hope this is still the case – that actually having their ovaries out might actually reduce your risk of having breast cancer. So[...] I think they might find that useful to know |
| | P2L: [Information about] what can go wrong. And probably also what exactly each operation involves, you know and how they do the laparoscopic [surgery]. |
| | HP4: [Surgery] can have a profound effect on somebody's relationship and to be blunt about it, somehow that has to be introduced in the conversation beforehand |
| | HP4: I tend to not go into too much detail, [...] but you introduce the concept of well: "look how much is it going to affect relationships, sexuality, your feelings of femininity, womanhood..." |
| | HP9: I think it would also be important [...] to know about the risks of primary peritoneal cancer after [surgery] and to know about the possible complications of surgery. You can't possibly make a decision unless you know what might go wrong really. |

Table 7.2 cont. – Recommendations regarding information content of the patient decision support intervention

| CODE Phase / Subtopic | Recommendations from patients and health professionals |
|-----------------------|---|
| Risk-reducing surgery | P1L: I think it would be interesting if you ever have any kind of statistics of people who are given like hysterectomy or whatever, ovaries removed, and then those ovaries are dissected and there is possibly pre-cancerous cells or something |
| | P3C: Again it's what's menopause after surgery like? That it's the symptoms may be more severe. |
| | P3C: ...there needs to be a lot more information about the menopause and different treatments whether they're conventional treatments, alternative treatments and combinations of them. |
| | P1C: And like for the negative parts [of the menopause], is there anything that can be done to counteract that? Like, you know, the HRT or the loss of libido, all that kind of stuff. |
| | HP3: So it isn't a necessary thing, but they may want to know all about natural HRT [...], they love that sort of thing |
| Other options | P1L: Just having information about the screening, people that it did help in a positive way, whether it would be worth your while to pay and to go ahead with the screening now. |
| | HP9: Then they need information about ovarian cancer I guess, symptoms, but that's very controversial. |
| | P3S: Like [for] how many people with ovarian cancer do the CA125 [...] actually go up [...]? Are there any other things [indicators of ovarian cancer] because all the other things [symptoms] that you get with ovarian cancer are quite vague, aren't they? The symptoms themselves [...]. So those are the things that I would find interesting. |

In terms of alternative management options to RRSO, women asked for information about screening, which is not surprising as the majority of participants had been taking part in the UK Familial Ovarian Cancer Screening (UKFOCS) study (Rosenthal et al. 2013a). Many suggested that information about trans-vaginal ultrasounds and CA125 blood tests would be helpful, while health professionals were reluctant to recommend including such information due to the lack of established clinical efficacy of ovarian screening. Stakeholders also mentioned that information about ovarian cancer symptoms may be useful, although some professionals and patients voiced concerns about the vague and non-specific nature of early ovarian cancer symptoms.

7.3.1v Coping content: Coping is an integral component of the CODE framework, which postulates that each phase of the deliberation gives rise to a coping process (Chapter 2, Chapter 5; Witt et al. 2012). Together with the questions during secondary appraisal (outlined in the RRSO-adapted CODE framework) and evidence synthesis II, the suggestions women and health professionals made regarding additional support were used to guide development of the coping advice. This content aimed to achieve goal 3 of the intervention; i.e. improve awareness of coping options and support coping. Some stakeholders discussed the need for content other than information, which may be labelled ‘coping content’ or ‘coping advice’, designed to facilitate coping processes and support the woman as a whole.

HP7:...looking at the woman holistically, as part of your decision aid, might be the new thing that hasn't been done before.

One professional emphasised the need to normalise emotional reactions to threats, such as an increased risk of cancer, in order to validate patient's feelings and reduce anxieties experienced in response to the threat.

HP2:...if you go over expected normal psychological reactions, that can be a huge relief to somebody, because they realise that they understand their emotional reaction as being more part of the process as opposed to being fearful that they are going mad or anything else that is more extremist and catastrophic...

Encouraging patients to explore and express their feelings and share their thoughts with close family and friends might be one way to support coping during decision making about ovarian cancer risk management. According to the CODE framework, other coping content might include advice to seek information and professional help, encouragement of positive re-appraisal and guidance on relaxation techniques to regulate emotions and intrusive thoughts. Women and health professionals also mentioned the advantages of patient testimonies and peer support to facilitate coping and decision making.

P3L: I think [we need] a tool which includes some facts [and] some anecdotal evidence from women who have been through a similar situation and made different decisions and had different experiences and with the honesty for some women to say, 'This was hell, and I made the wrong decision'.

HP3: The other thing I suppose is what no woman has and would like at any point in making a decision is to talk to somebody who has already had it done.

Evidence regarding the effectiveness of patient stories in decision support interventions is controversial, as balanced portrayal of outcomes is difficult and such stories might bias patients' decision making (Winterbottom and Bekker 2009, Winterbottom et al. 2008). However, women were keen to read about and speak to others who had already made the decision. Therefore, it was decided to refer users of the intervention to other websites that included patient blogs and videos, as well as signposting them to peer support groups that could cater for those needs.

7.3.1vi Value clarification: To achieve goal 4 of the intervention; i.e. support preference construction, an explicit value clarification exercise may be beneficial. Therefore, stakeholders' views of the format and content of such exercises were sought. Focus groups and interview participants were shown an example value clarification exercise and were asked to comment and discuss (Figure 7.2)

Figure 7.2 - Example of value clarification exercise shown in focus groups and interviews

OPTION 1a WORKSHEET

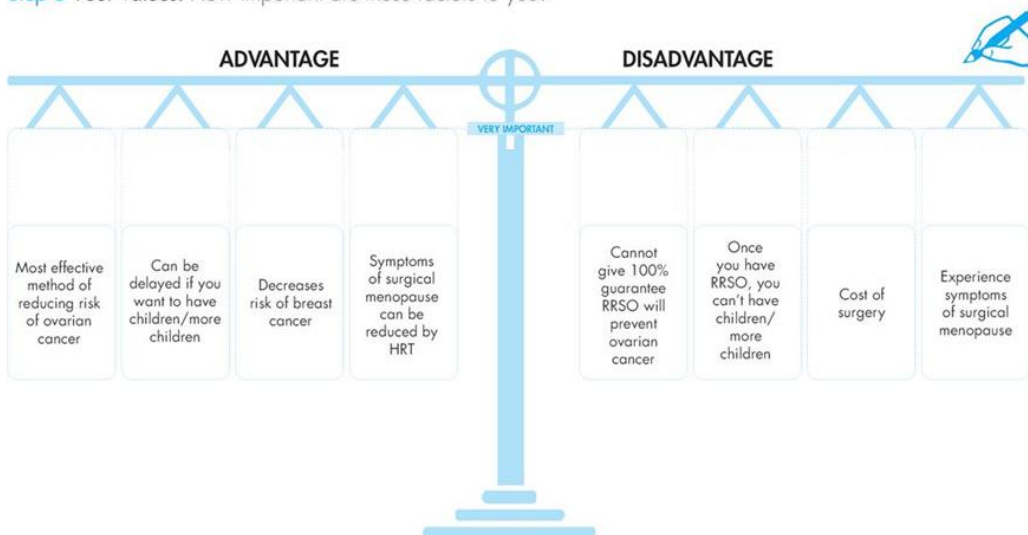
Personal Decision Aid Worksheet
BILATERAL RISK-REDUCING SALPINGO-OOPHORECTOMY (RRSO)

Step 1 Briefly review the information in this booklet about managing ovarian cancer risk with RRSO and HRT.

Step 2 Possible advantages and disadvantages of RRSO

| Advantages | Disadvantages |
|---|---|
| Most effective method of reducing ovarian cancer risk | Not possible to give a 100% guarantee that ovarian/peritoneal cancer will be prevented |
| Can be delayed if you wish to have children/more children | Once a woman has RRSO, she can't have children/more children – this may produce feelings of guilt or grief |
| Decreases risk of breast cancer | Cost of surgery (While these procedures are covered by Medicare rebates, insured patients using private hospital facilities may have additional charges). |
| Symptoms of surgical menopause can be reduced using HRT | Pre-menopausal women having RRSO will experience symptoms of surgical menopause |

Step 3 Your values: How important are these factors to you?



Step 4 Your leaning

| | | | | | | |
|------|--|--|--------|--|--|------|
| Yes | | | Unsure | | | No |
| RRSO | | | | | | RRSO |

On balance, you decided

From: Decision aid "Risk Management Options for Women at Increased Risk of Developing Ovarian Cancer" (New South Wales Centre for Genetics Education 2008, Tiller et al. 2003)

(Llewellyn-Thomas 2009, Peate et al. 2013). Stakeholders could see the advantages of such an exercise in terms of clarifying values and supporting the process of decision making.

P1S: I think it [the value clarification exercise] focuses your mind. Whether it would make you anymore definite about your decision I don't know, if that makes sense.

P2S: No, it is helpful [...] going through it.

HP7: Well I think that this [value clarification exercise] probably would take someone forward, the decision making. And it may result in a consent for surgery or it might not.

The format used in the sample exercise weighed advantages for surgery on one side and disadvantages on the other, using a set of scales. Stakeholders found this type of visualisation appealing and easy to understand. One professional noted that he was already using this type of visualisations in his consultations.

HP8: It's interesting you draw the scales there, [...] because that is exactly what I use to explain that. It's a balancing of the risks and benefits. And these go up and down and [...] when you see that the risks of surgery outweigh the benefits, then surgery may not be ethically justifiable. And the other way around.

However, while stakeholders liked the visualisation using the scales and the possibility of attributing different weights to different factors, they noted that the sample exercise did not allow users to input any factors of their own, which limited the usefulness of the exercise.

HP9: That sort of thing itself would be quite good, [...] but there might be other things that are important for them personally and how do you put those things in?

P3L: Would there be the option to put personal factors in? [...] So [...] would there be a possibility of putting in, or you being able to add factors that mean a lot to you? [...]

P1L: Kind of an ad-hoc, yeah.

P3L: Yea, an 'any other'

P1L: Yea, 'any other' that's, yea, like an empty box.

Based on this feedback, it was decided that a value clarification exercise using a set of scales was appropriate, on the condition that users were able to attribute different weights to different factors and to add their own personal factors.

7.3.2 *Prototype I*

An initial prototype of OvDex was developed informed by the RRSO-adapted CODE framework (Chapter 5; Witt et al. 2012). OvDex was structured into four main and four supplementary sections, all of which were informed by the deliberation phases proposed in the CODE framework (see Appendix 7.3 for OvDex prototype I). The main sections corresponded to the four phases of deliberation, which include 'threat', 'choice', 'options' and 'preference construction'. Within OvDex these were entitled 'cancer risk', 'the choice', 'options at a glance' and 'your decision'. The 'cancer risk' and 'the choice' sections provided information about ovarian cancer risk and explained the choice, respectively. The 'options at a glance' section consisted of an Option Grid to provide an overview of the options (Table 7.3). The questions within the Option Grid were informed by questions outlined in the 'presentation of options' phase in the RRSO-adapted CODE framework. The 'your decision' section would later encompass the value clarification exercise; however, this was not included in prototype I. In line with the content of the CODE framework, each of the four main sections would later also include coping advice. However, the initial prototype only consisted of the information content. This was informed by the results of evidence synthesis I.

The supplementary sections gave additional and more detailed information about the options and were entitled: 'risk-reducing surgery', 'the menopause', 'hormone replacement' and 'other options'. These were created in response to requests from participants, who felt detailed information about these topics would be particularly important. Each section contained a number of questions guided by the RRSO-adapted CODE framework (Table 7.4).

Table 7.3 - The 'Options at a Glance' section in prototype I (general version)

| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|---|---|---|
| Will this reduce my ovarian cancer risk? | Yes, removing the ovaries will greatly reduce your lifetime risk of ovarian cancer | No, your risk of ovarian cancer will remain increased |
| Will I be able to become pregnant? | No. | Yes. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. | You may worry about developing ovarian cancer. |
| Will I go into early menopause? | Yes, your oestrogen levels fall and the menopause will start immediately. | No. |
| What is menopause after surgery like? | Menopause after surgery is similar to natural menopause, but because it happens suddenly, the symptoms may be more severe. | Not applicable. |
| Will this change how I feel about myself as a woman? | Most women do not notice any change in how they feel about themselves as a woman. Your desire for and/or enjoyment of sex may decrease a lot. | Not applicable. |
| Are there any risks linked to early menopause? | Yes, there is a risk of bone thinning (osteoporosis). Some patients report memory changes as well. | Not applicable. |
| Will I need hormone replacement therapy (HRT)? | HRT can reduce some of the effects of sudden menopause. You will need to discuss this possibility with your doctor. | Not applicable |
| Is HRT safe for women at increased familial risk? | HRT is safe to take for the majority of women until they are 50. It is not recommended for women who have had breast cancer themselves already. | Not applicable |
| What are the risks of surgery? | There is a small risk of complications depending on the type of surgical procedure (key hole or open surgery) you have. Discuss this with your doctor. | Not applicable. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after key hole surgery and are back to normal in 4 weeks. For open surgery this is slightly longer. | Not applicable. |
| Can I get screened for ovarian cancer? | No, after surgery there is no need for any screening because your risk will be low | No, there is no routine screening available for ovarian cancer. |

Table 7.4 - Questions within the sections of the general (non-personalised) OvDex prototype I

| Main Section | Question |
|--------------------------------|---|
| Cancer risk | What is the ovarian cancer risk if I have a family history of ovarian cancer? |
| | How can I find out whether I have a faulty gene? |
| | What would my risk be if a faulty gene is found? |
| | What if I cannot get tested or my genetic test is unclear? |
| | Does my cancer risk change over time? |
| | How will my ovarian cancer risk affect my life? |
| | How will my ovarian cancer risk affect my children? |
| | What can I do to reduce my risk? |
| The choice | What can I do to reduce my risk? |
| | Why is there a choice? |
| | Who should decide? |
| | Who else should I involve in this decision? |
| | When should I decide? |
| | How can I decide? |
| Options at a glance | Options at a glance (Option grid) |
| <i>Supplementary Sections:</i> | |
| Risk-reducing surgery | What is risk-reducing bilateral salpingo-oophorectomy (RRSO)? |
| | What are the main advantages of this operation? |
| | What are the main disadvantages of this operation? |
| | What is my risk after surgery? |
| | What does the surgery involve? |
| | How long does it take to recover? |
| | Do I need to have my womb removed as well? |
| | What are the complications of RRSO? |
| | Can women die from RRSO? |
| | Could cancer be found during the surgery? |
| How would RRSO affect my life? | |
| Surgical menopause | What is surgical menopause? |
| | What is surgical menopause like? |
| | How could it affect my life? |
| | How long does surgical menopause last? |
| | Are there other problems linked to surgical menopause? |
| | Is there anything I can do to prevent the long term health effects of surgical menopause? |
| Hormone replacement | What is Hormone Replacement Therapy (HRT)? |
| | Why is HRT important? |
| | Who should take HRT? |
| | How long should I take HRT? |
| | Can I take HRT if I have a family history of breast cancer? |
| | But I have heard that HRT increases breast cancer risk? |
| | Will HRT reduce all the symptoms of menopause? |
| | Are there any alternatives to HRT? |
| Other options | Is there any screening available for ovarian cancer? |
| | What is the alternative to surgery? |
| | What would happen if I do not have surgery? |
| | Is there anything else I can do? |

As was suggested by health professionals during the interviews, information might differ depending on patient variables, such as age, mutation status and breast cancer history. Therefore, to give users the possibility to personalise content, different prototype versions were developed. Patients could request and view a tailored version by answering three questions (Figure 7.3).

Figure 7.3 - Personalisation questions in prototype I

| |
|---|
| <p>1. Do you have a faulty gene or a family history of Lynch Syndrome?</p> <p>a) Yes, I have a BRCA1 faulty gene</p> <p>b) Yes, I have a BRCA2 faulty gene</p> <p>c) Yes, I am from a Lynch Syndrome family</p> <p>d) Don't know, I have not been tested for a faulty gene or my genetic test was unclear</p> <p>e) No, I have been tested and no faulty gene was identified</p> <p>2. Have you ever had breast cancer?</p> <p>a) No, I have never had breast cancer</p> <p>b) Yes, I have had breast cancer</p> <p>3. How old are you?</p> <p>a) Under 35</p> <p>b) 35-39</p> <p>c) 40-49</p> <p>d) Over 50</p> |
|---|

The information within the Option Grids in the 'options at a glance' section was tailored depending on personalisation. Information within the personalised versions also differed in sections such as cancer risk, menopause and HRT. The versions for patients who carry mutations in BRCA1/2 and women from families with Lynch syndrome provided specific cancer risk information relevant to mutation status / family history. The version for patients with a history of breast cancer stated that HRT was not recommended for this population and therefore only included a shortened version of the HRT information section. The version for pre-menopausal women (under 50) included detailed information about the menopause and HRT. In contrast, the version for post-menopausal women (50 and over) did not include the sections on menopause and HRT, as these were not

relevant to this population. A version for individuals who tested negative for a genetic mutation was also created, which advised women that they might be at average risk and therefore did not need to make this decision. This version only consisted of a section on risk and choice (Appendix 7.4). The draft information content of all versions was reviewed by a consultant clinical geneticist and amended according to his comments, before being sent out to the virtual reference group.

7.3.3 Virtual Reference Group – Feedback Round 1

Ten patients and eight health professionals were invited to provide feedback on prototype I. Six patients and five professionals responded to the invitation. Most felt that the information was accessible and pitched at the right level.

VR1: One can expect the inclusion of a certain amount of facts and figures but the information is presented in a plain English style. (Patient)

VR8: In general I think the information is very good, and clearly explained, and probably aimed at the right level. I found it easy to understand, and I think it would not be unduly difficult for someone with less knowledge in the area to understand. (Professional)

One woman positively commented on the possibility to personalise information depending on mutation status / family history.

VR2: It is also very useful to have personalised information for different groups of women where there is differentiated risk of ovarian cancer. Separation of information for BRCA1 and Lynch Syndrome is definitely helpful. (Patient)

Some patients also commented on how they responded emotionally to the information presented in the intervention.

VR1: It was comforting to read that the studies linking HRT with an increased risk of breast cancer have been criticised, as on a personal

level this was a major factor in holding me back from considering an oophorectomy pre menopause. (Patient)

However, most comments from patients and health professionals regarded improvements that could be made to the content of the tool. The two groups did not differ significantly in the types of responses they provided in this round of feedback, most of which fell within the 'confusing/needs rewording' and 'additions/expansions' categories. Table 7.5 shows a selection of quotes for the categories under the 'suggestions for improvements' code.

Unsurprisingly, compared to the comments from patients, professionals' feedback was more often pointing to clinical evidence, for instance referring to the NICE guidance of ovarian cancer symptoms (National Institute for Health and Care Excellence 2011). However, patients also requested additional information that they had previously obtained from other sources, for example information regarding hysterectomy for Lynch women and the effects of premature menopause on cardiovascular disease. Overall, respondents made 86 actionable suggestions for improvements (31 from patients, 55 from professionals) that resulted in changes to the information content in prototype I. For a detailed list of all recommendations for improvements and actions taken in response see Appendix 7.5.

7.3.4 Prototype II

In response to feedback from the virtual reference group, prototype II was developed. This prototype included two stand-alone paper-based Option Grids (one for pre-menopausal and one for post-menopausal women) and the OvDex tool, as an online and paper-based version. Option Grids also remained embedded in OvDex in the 'Options at a glance' section. Content in the grids within the 'Options at a glance' section differed slightly depending on the personalisation. Content of the Option Grids was amended using feedback from the virtual reference group, as well as suggestions from the editorial team. Changes were generally non-substantial. In the non-personalised version for instance, a numerical estimate of

Table 7.5 - Examples of suggestions for improvements by virtual reference group members: Round 1

| Code | Example quotes |
|-----------------------------|---|
| Spelling / Grammar | VR9: "What are the complications of RRSO" Last sentence of the first paragraph: "the surgeon has to convert to an operation or to open surgery." (Professional) |
| Not relevant | VR8: Question on how can I find out whether I have a faulty BRCA1 gene is irrelevant at this point, they already have it confirmed. (Professional; comment on BRCA version) |
| Confusing / needs rewording | VR3: On the Risk Reducing Surgery section the part detailing amount of women having surgery and those that are converted from keyhole to open surgery when I first read the explanation to the right of the grid in bold I thought the grid showed amount that was converted to open surgery...which would put a lot of people off the idea. (Patient) |
| | VR8: Question: When should I decide? I am not certain about the phrasing in this context: "It is important you make sure you have completed your family before you make this choice." It sounds like an instruction. (Professional) |
| | VR10: The risk for ovarian cancer is still low at age 35 (your graph is rather misleading as quick glance by someone who doesn't understand it may suggest that it is higher than it is). For this reason I would suggest that you change the scale of the graph. (Professional; comment on: under 35 versions) |
| | VR8: The symptoms should be the ones used in the NICE Guidance CG122. (Professional) |
| Additions / expansions | VR4: It mentions genetic testing. [...] More information on this would be useful. (Patient) |
| | VR2: For Lynch Syndrome women the surgery would involve hysterectomy as well as oophorectomy. It, therefore, doesn't make complete sense to consider removal of the ovaries in isolation. It would be useful to either include more information on the hysterectomy or to link to it from another source at the relevant points in the text. (Patient; comment on: Lynch version) |
| | VR1: I would like to have seen Chapter numbers and individual paragraph / section numbers. A minor point, but this would be helpful as there are quite a lot of cross references to different sections throughout. (Patient) |
| | VR10: A picture of the anatomy might be helpful as many women do not know their anatomy or understand the difference between the ovary uterus and cervix! (Professional) |
| Repetition | VR4: There was some repetition (Patient) |
| | VR11: "For all other women the risk of primary peritoneal cancer after surgery is close to zero." – Repeated info in this paragraph. (Professional) |
| Other | VR5: I am clearly given the indication from the gynecologist during my yearly hysteroscopy that I should have my ovaries and womb removed as the risk of cancer outweighs my fears on being on HRT after surgery. (Patient) |

If comments were only applicable to a specific version, this is specified as (...comment on:...) after the quote

risk of ovarian-type cancer after surgery was added. The question on availability of screening for ovarian cancer was also re-worded; however, the content of Option Grids largely remained the same. The stand-alone versions also included a statement to define the target audience and the health decision (Figure 7.4a and Figure 7.4b).

The structure of the online and paper-based versions of OvDex remained the same as in prototype I, guided by the phases outlined in the CODE framework; however, the title for one of the supplementary sections changed from 'Surgical menopause' to 'The menopause'. In the paper version, page numbers were added and sections and questions were also numbered to aid navigation, as suggested by members of the virtual reference group. Table 7.6 depicts the questions addressed in each of the sections of the generic, non-personalised prototype II of OvDex (see Appendix 7.6 for paper-based version).

In response to comments from the virtual reference group, the answers to most questions were modified and some questions were added to address previously omitted topics or expand on existing ones. For example, a diagram outlining women's anatomy and specifying where ovarian-type cancer develops was added to the 'cancer risk' section. Furthermore, a statement about women who have been offered CA125 blood tests and ultrasounds was added to the 'other options' section to emphasise the lack of clinical evidence for screening, despite privately funded screening being available in some areas. The version for Lynch women underwent substantial amendments due to the inclusion of risk-reducing hysterectomy as an important option to reduce endometrial cancer risk in this population. Additionally, at the end of prototype II of OvDex, a list of research evidence was added to reference publications that had been used in the development of the intervention.

Figure 7.4a - The Option Grid / 'options at a glance' section in prototype II
(general version for pre-menopausal women)




Working on what matters to you most

Ovarian Cancer for Women at Increased Familial Risk

This grid is to help pre-menopausal women at increased risk of ovarian cancer and their clinicians decide whether to remove the ovaries to reduce the risk of ovarian cancer.

| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|--|---|--|
| Will this reduce my ovarian cancer risk? | Yes, removing the ovaries and fallopian tubes will greatly reduce your lifetime risk of ovarian-type cancer to less than 2 in 100. | No, your risk of ovarian cancer will remain high. |
| Will I be able to become pregnant? | No. | Yes. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. | You may worry about developing ovarian cancer. |
| Will I go into menopause? | Yes, your oestrogen levels fall and the menopause will start immediately. | No. |
| What is menopause after surgery like? | Menopause after surgery is similar to natural menopause, but because it happens suddenly, the symptoms may be more severe. | Not applicable. |
| Will this change how I feel about myself as a woman? | Many women do not notice any change in how they feel about themselves as a woman. However, your desire for and/or enjoyment of sex may decrease a lot. | Not applicable. |
| Are there any risks linked to menopause? | Yes, there is a risk of bone thinning and cardiovascular disease. Some patients report memory changes as well. | Not applicable. |
| Will I need hormone replacement therapy (HRT)? | HRT can reduce some of the effects of sudden menopause. You will need to discuss this possibility with your doctor. | Not applicable |
| Is HRT safe for women at increased familial risk? | HRT is safe to take for the majority of women until they are 50. It is not recommended for women who have had breast cancer themselves already. | Not applicable |
| What are the risks of surgery? | There is a small risk of complications depending on the type of surgical procedure (key hole or open surgery) you have. Discuss this with your doctor. | Not applicable. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after key hole surgery and are back to normal in 4 weeks. For open surgery this is slightly longer. | Not applicable. |
| Is there a routine screening programme to detect ovarian cancer? | No, and after surgery there is no need for any screening because your risk will be low. | No, there is no routine screening available on the NHS to detect ovarian cancer. |

Figure 7.4b – The Option Grid / ‘options at a glance’ section in prototype II
(general version for post-menopausal women)

option grid 

Working on what matters to you most

Ovarian Cancer for Women at Increased Familial Risk

This grid is to help post-menopausal women at increased risk of ovarian cancer and their clinicians decide whether to remove the ovaries to reduce the risk of ovarian cancer.

| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|--|---|--|
| Will this reduce my ovarian cancer risk? | Yes, removing the ovaries and fallopian tubes will greatly reduce your lifetime risk of ovarian-type cancer to less than 2 in 100. | No, your risk of ovarian cancer will remain high. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. | You may worry about developing ovarian cancer. |
| Will I get menopausal symptoms again? | No, if you have completed the menopause removing your ovaries should not cause any menopausal symptoms to recur. If you are peri-menopausal and have not quite completed the menopause some symptoms may occur. | No. |
| Will this change how I feel about myself as a woman? | Many women do not notice any change in how they feel about themselves as a woman. However, your desire for and/or enjoyment of sex may decrease a lot. | Not applicable. |
| What are the risks of surgery? | There is a small risk of complications depending on the type of surgical procedure (key hole or open surgery) you have. Discuss this with your doctor. | Not applicable. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after key hole surgery and are back to normal in 4 weeks. For open surgery this is slightly longer. | Not applicable. |
| Is there a routine screening programme to detect ovarian cancer? | No, and after surgery there is no need for any screening because your risk will be low. | No, there is no routine screening available on the NHS to detect ovarian cancer. |

Table 7.6 - Questions within the sections of the general (non-personalised) OvDex prototype II

| Main Section | Question |
|----------------------------------|--|
| 1 - Cancer risk | Q1 Where does ovarian cancer develop? |
| | Q2 What is the ovarian cancer risk if I have a family history of ovarian cancer? |
| | Q3 How can I find out whether I have a faulty gene? |
| | Q4 What would my risk be if a faulty gene is found? |
| | Q5 What if I cannot get tested or my genetic test is unclear? |
| | Q6 Does my cancer risk change over time? |
| | Q7 How will my ovarian cancer risk affect my life? |
| | Q8 How will my ovarian cancer risk affect my children? |
| | Q9 How can I deal with my cancer risk? (Coping advice) |
| 2 - The choice | Q1 What can I do to reduce my risk? |
| | Q2 Why is there a choice? |
| | Q3 Who should decide? |
| | Q4 Who else should I involve in this decision? |
| | Q5 When should I decide? |
| | Q6 How can I decide? |
| | Q7 How can I deal with the choice? (Coping advice) |
| 3 - Options at a glance | Options at a glance (Option grid) |
| | Q1 How can I deal with this decision? (Coping advice) |
| <i>Supplementary Sections:</i> | |
| 4 - Risk-reducing surgery | Q1 What is risk-reducing bilateral salpingo-oophorectomy (RRSO)? |
| | Q2 What are the main advantages of this operation? |
| | Q3 What are the main disadvantages of this operation? |
| | Q4 What is my risk after surgery? |
| | Q5 What does the surgery involve? |
| | Q6 How long does it take to recover? |
| | Q7 Do I need to have my womb removed as well? |
| | Q8 What are the complications of RRSO? |
| | Q9 Could cancer be found during the surgery? |
| | Q10 How would RRSO affect my life? |
| 5 - The menopause | Q1 What is surgical menopause? |
| | Q2 What is the menopause like? |
| | Q3 How could the menopause affect my life in the short term? |
| | Q4 How long does surgical menopause last? |
| | Q5 Are there any long term health effects linked to surgical menopause? |
| | Q6 Is there anything I can do to reduce the effects of surgical menopause? |
| 6 - Hormone replacement | Q1 What is Hormone Replacement Therapy (HRT)? |
| | Q2 Why is HRT important? |
| | Q3 Who should take HRT? |
| | Q4 How long should I take HRT? |
| | Q5 Can I take HRT if I have a family history of breast cancer? |
| | Q6 But I have heard that HRT increases breast cancer risk? |
| | Q7 Will HRT reduce all the symptoms of menopause? |
| | Q8 Will HRT deal with the long term health effects of the menopause? |
| | Q9 Are there any alternatives to HRT? |
| 7 - Other options | Q1 Is there any screening available for ovarian cancer? |
| | Q2 What is the alternative to surgery? |
| | Q3 But I have been offered CA125 blood tests and/or trans-vaginal scans |
| | Q4 What would happen if I do not have surgery? |
| | Q5 Is there anything else I can do? |

Table 7.6 cont. – Questions within the sections of the general (non-personalised) OvDex prototype II

| | |
|--------------------------|-------------------------------------|
| 8 - Your decision | Value clarification exercise |
| | Q1 How can I deal with my decision? |

Personalisation within the online version was enabled using the three questions outlined earlier (Figure 7.3). These questions were answered sequentially and subsequently took users to versions of the website that were tailored to the answers they had provided. Once personalised, users could download a pdf copy of the materials or navigate the website online.

Coping advice and a value clarification exercise were also added to prototype II. Development of the coping advice was guided by items included in the 'secondary appraisal' row of the RRSO-adapted CODE framework (Chapter 5) and evidence synthesis II (see section 7.2.4). Each of the four main sections of OvDex included a paragraph on coping advice. The coping advice pointed women towards sources of medical information and promoted problem solving and action planning approaches. These sections aimed to support problem-focused coping, i.e. increase awareness and use of problem-focused coping options and resources. Other sections within the coping advice encouraged women to seek social support and/or referred patients to external websites for further information on relaxation and mind-body therapies. These sections aimed to support emotion-focused coping, i.e. increase awareness and use of emotion-focused coping options and resources. Figure 7.5 shows a sample section of coping advice labelled to highlight the different coping options included.

Figure 7.5 - Example of coping advice in OvDex prototype II with labelled options

| How can I change how I feel about my cancer risk? | <i>Coping option / resource</i> |
|--|---|
| If you would like to find out more about your risk and what it means, you can use the resources provided in OvDex and talk to your doctor or genetic counsellor. | <i>Seeking information and informational support from experts</i> |
| Knowing that you are at increased risk of cancer might make you feel worried or angry or frustrated. This is completely normal and there is no need to bottle those feelings up. It can actually help to talk about them. | <i>Acknowledging and venting emotions</i> |
| You can try to find someone you trust to talk to or if you prefer to talk to someone you don't know, you can use the contact details for charities and patient support networks under Contacts & Resources. | <i>Seeking emotional support from family / friends / peers</i> |
| If you have a partner, sharing your thoughts and concerns with your partner can be useful in helping them understand what you are going through and in helping you cope together as a couple. | <i>Couple coping and seeking emotional support from partner</i> |
| It may also help to see a positive side to knowing your risk status. For example, this means that you will get access to genetics services and have the chance to do something about your risk. Other women have found it helpful to stay optimistic, have joined patient networks or have found strength in their faith. | <i>Positive re-appraisal and seeking emotional support</i> |
| Being at risk of cancer may sometimes lead to unwanted thoughts and worry, especially at times when there are no active steps you can take, for example when you are waiting for genetic test results. There are useful techniques you can use to reduce such thoughts. One such method is active distraction, which means actively thinking about something else whenever unwanted thoughts pop in your head. Watch the 'How do I cope' video on the Cancer Genetics storybank website for a guide to how to do this. | <i>Active distraction</i> |
| Relaxation techniques, meditation and guided imagery may also help you deal with any stress you might experience relating to your cancer risk. Examples include progressive muscle relaxation and certain breathing exercises. Visit the 'Mind-body therapy' section on the Macmillan website for more information on and guides to mind-body therapies. | <i>Relaxation and meditation</i> |

The value clarification exercise was developed based on items in the 'preference construction' column of the RRSO-adapted CODE framework (Chapter 5) and on two decision making models: the conflict model of decision making (Janis and Mann 1977) and differentiation and consolidation theory (Svenson 1992). The conflict model of decision making postulates that decisional conflict ensues when a decision needs to be made between options that are associated with potential positive outcomes (anticipated gains) and negative outcomes (anticipated losses) (Janis and Mann 1977). It suggests that decision makers should ideally take a vigilant approach to information seeking, i.e. obtain as much relevant information as possible about attributes associated with each option (Janis and Mann 1976). The model further proposes that options should then be distinguished based on anticipated gains and losses (Janis and Mann 1977). The best option is that which is associated with the most gains and least losses. Deliberation components based on this model could take the form of weighing or balancing exercises, enabling patients to identify anticipated gains and losses associated with options (Elwyn et al. 2011b). Differentiation and consolidation theory postulates that decision makers differentiate options according to the relative importance they ascribe to attributes associated with options (Svenson 1992, 2003). The best option is that which is associated with the most important positive attribute(s) and, ideally, the least important negative attributes (Svenson 1992, 2003). Elwyn et al. (2011b) suggested that deliberation components based on this theory should allow a direct comparison of attributes and a differentiation based on the sum of positive and negative attributes for each option. Ideally, these sums should take into account the relative importance of each attribute from the decision maker's perspective.

In accordance with these models, the value clarification exercise in OvDex was designed to elicit the importance patients ascribe to different attributes (anticipated gains and losses) associated with RRSO. In line with the conflict model of decision making, patients were asked to decide whether attributes made them want to *have* or want to *avoid* surgery, i.e. whether they were perceived as

potential gains or potential losses if one was to choose surgery. Pre-defined attributes covered in the exercise included statements such as 'surgery will reduce my ovarian cancer risk' and 'I will not be able to become pregnant after surgery'. Patients could also add their own attributes. In line with differentiation and consolidation theory, all attributes could additionally be weighed as to their relative importance on a scale of 1 (a little important) to 3 (very important). For example, a reduction in ovarian cancer risk may be perceived as a potential gain, making a patient want to have surgery, and be ascribed high importance (i.e. a weight of 3). In contrast, a change in body image may be perceived as a potential loss, making a patient want to avoid surgery, but be ascribed low importance (i.e. a weight of 1). The sum of weights ascribed to gains resulting from RRSO was then compared to the sum of weights ascribed to losses resulting from RRSO using a set of weighing scales. This allowed decision makers to visualise the overall importance they ascribed to gains and losses associated with RRSO.

The value clarification exercise was available in an interactive online format (Figure 7.6) and as a paper-based version (Figure 7.7 and 7.8). The online format allowed users to mark radio buttons to indicate whether a statement made them want to *have* or *avoid* surgery and the relative importance they attributed to that statement. The overall outcome was displayed automatically on a set of scales with weights that were sized according to the importance that had been attributed to them. In the paper-based version, women had to indicate the weight as a number in the correct column to indicate whether a statement made them want to have or avoid surgery. Women had to then calculate and transfer the total sum for each column into a picture of a set of scales. Three summary statements were displayed at the end. The statement most relevant depended on the individual outcome of the exercise and the tipping of the scales. Patients needed to identify the summary statement that applied to them. Interactive exercises, such as these, engage users and can enhance the decision making process (Coulter 2009a).

Figure 7.6 - Interactive online value clarification exercise

Weighing it up

This exercise might help you to weigh up the potential reasons that might make you want / not want to have surgery. Simply read the statements on the left and decide how much they make you want to have surgery or avoid surgery and then click the appropriate button. The scales will then show where you stand with your decision.

| Potential reasons for/against surgery: | Makes me <i>want to HAVE surgery</i> | | | Does not come into my decision | Makes me <i>want to AVOID surgery</i> | | |
|---|--------------------------------------|----------------------------------|----------------------------------|----------------------------------|---------------------------------------|----------------------------------|-----------------------|
| | a lot | quite a bit | a little | | a little | quite a bit | a lot |
| Surgery will reduce my ovarian cancer risk | <input checked="" type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| Surgery will reduce my breast cancer risk | <input type="radio"/> | <input checked="" type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| I will not be able to become pregnant after surgery | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input checked="" type="radio"/> | <input type="radio"/> |
| I will go into the menopause after surgery | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input checked="" type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| There is no effective screening for ovarian cancer | <input type="radio"/> | <input type="radio"/> | <input checked="" type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| The symptoms of ovarian cancer are very vague | <input type="radio"/> | <input type="radio"/> | <input checked="" type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| I might feel different about my body after surgery | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input checked="" type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| I might enjoy sex less after surgery | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input checked="" type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| Enter your own reasons: | | | | | | | |
| I'll have to take time off | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input checked="" type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |
| | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |

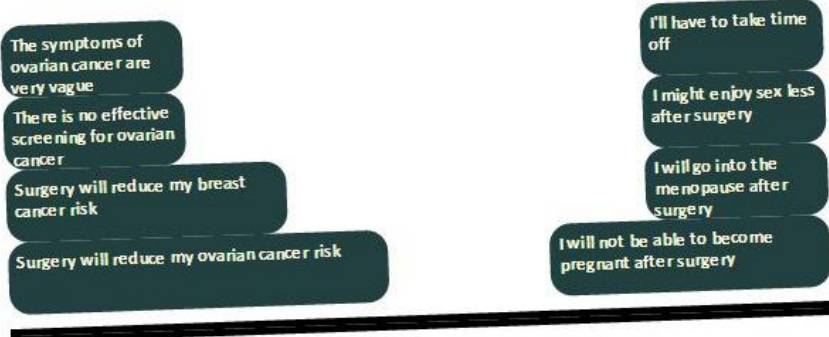


Figure 7.7 - Paper-based value clarification exercise – ranking

In the exercise on the next few pages you can rate some facts depending on how much they make you want **to have** or want **to avoid** surgery. Give each fact a ranking number (e.g. from 1 (a little) to 3 (a lot), then add the numbers up to see where you stand.

Make sure you give the score on the correct side of the table.

| The fact that... | Makes me want to have surgery | Does not come into my decision | Makes me want to avoid surgery |
|--|--------------------------------------|--------------------------------|---------------------------------------|
| ...surgery will reduce my ovarian cancer risk | | | |
| ...surgery will reduce my breast cancer risk | | | |
| ...I will not be able to become pregnant after surgery | | | |
| I will go into surgical menopause after surgery | | | |
| There is no effective screening for ovarian cancer | | | |
| The symptoms of ovarian cancer are very vague | | | |
| I might feel different about my body after surgery | | | |
| I might enjoy sex less after surgery | | | |
| Enter your own reasons: | | | |
| | | | |
| | | | |
| | | | |
| | | | |
| Add together the numbers in each column: | | | |

Figure 7.8 - Paper-based value clarification exercise – summary and outcome

Below you can enter the overall number into the scales. Then note down how you feel about the decision and where you stand.



More weight for than against surgery:

Overall your answers show that at this moment in time you are leaning towards having the surgery. Even though you may have some reasons why you would want to avoid surgery, they are not as important to you as the reasons that make you want surgery. Therefore you could now make an appointment to discuss this decision with your doctor and go through any concerns you might have.

Same weights both sides:

Overall your answers show that at this moment you are completely undecided about whether or not to have surgery. Your reasons for why you would want to have and why you would want to avoid surgery balance each other out. Therefore you may want to discuss your options further with your doctor/genetic counsellor.

More weight against than for surgery:

Overall your answers show that at this moment in time you are leaning towards not having surgery. Even though you may have some reasons why you would want to have surgery, they are not as important to you as the reasons that make you want to avoid surgery. Therefore at this moment in time surgery is probably not the right option. However, keep in mind that things may change over time and that eventually you may lean more towards surgery. You can come back to OvDex and do this exercise again in a years time to see whether anything has changed.

Overall decision: _____

Next steps: _____

7.3.5 *Virtual Reference Group – Feedback Round 2*

Ten patients and seven health professionals were invited to provide feedback on prototype II. One professional had opted out of the study and was therefore not invited in Round 2. Six patients and four professionals responded to the invitation. Overall, the majority of patients' comments in this round were positive (n=28) and patients made few suggestions for improvements (n=12). As in the first round of feedback from this group, many patients commented on the accessibility and readability of the content.

VR6: It was easy to understand because it was written in plain everyday English, but without being patronising. I know this isn't easy and it is rare to find the balance struck as well as it has been here! (Patient)

Patients also felt that the amount of information included in the intervention was appropriate.

VR1: I felt it has a good balance of info. It is inevitably difficult to try and balance the amount of information presented but I thought this gave sufficient detail and would prompt users to consult their GP if more in depth info was required. (Patient)

VR6: Generally I thought there was the right amount of information. There were no obvious gaps, but not an information overload. (Patient)

Some specifically commented on the website's layout and design as well. They felt that the structure of the site was 'easy to navigate' and 'flows well'.

VR5: I was impressed with the website as it was user friendly and clear with headings and easy to find your way back. Overall the layout was excellent and I also think the branding is well done. (Patient)

One respondent also noted that the online format would enable patients to return to the website whenever they choose, check for updates and explore whether their preferences had changed.

VR5: I believe this will definitely help women in thinking about the right factors for their overall decision, and something they can always come back to and read again and see if they feel differently about the issue. (Patient)

The value clarification exercise section of OvDex, which had not been reviewed before, also received positive feedback from patients.

VR6: I thought that this is one of the best weighing up tools I have ever seen. I really did like the option of [giving] different weights to the factors and the option to include one's own factors. I like having the visual representation. Again this was achieved in a way that is easy to understand without being patronising. (Patient)

VR1: It is helpful to see your own thoughts set out. It focuses your mind and almost feels as if the pressure of making a decision is removed slightly as the tool comes to the conclusion albeit based on your answers. (Patient)

VR3: I thought that the action plan part on 'how can I deal with my decision' [is] a very good idea and one that I will personally use. (Patient)

In contrast to patients, health professionals made fewer positive comments in this round and appeared to concentrate on making suggestions for improvements. The majority of these comments once again fell within the 'confusing / needs rewording' and 'additions / expansions' categories. Table 7.7 shows a selection of quotes for the categories under the 'suggestions for improvements' code in round 2. Overall, respondents made 64 actionable suggestions for improvements (12 from patients, 52 from professionals). For a detailed list of all recommendations for improvements and actions taken in response, see Appendix 7.7.

Table 7.7 - Examples of suggestions for improvements by virtual reference group members: Round 2

| Code | Example quotes |
|-----------------------------|---|
| Spelling / Grammar | VR10: I thought some of the grammar could have been improved. Some of it was repetitive and some of the sentences were a bit long and clumsy. (Professional) |
| | VR12: "Yes HRT will reeducde the risk" – Reduce (Professional) |
| Not relevant | VR7: I indicated that I had had breast cancer, I found a lot of the information on HRT irrelevant to me. (Patient; comment on: BRCA2 version) |
| Confusing / needs rewording | VR12: "You can come back to OvDex and do this exercise again in a year's time" - why specify a year's time. Surely we would be happy for someone to revisit their decision as often as they wish?? (Professional) |
| | VR6: The other place I did not find it helpful was the assumption that everyone has a partner. Not only was this unhelpful, it is also extremely irritating. (Patient) |
| | VR10: I would have major concerns about this bit [information about oral contraceptives in 'Other Options']. If a woman carries a BRCA1/2 mutation it would not be recommended for her to go on the pill to reduce her risk of ovarian cancer and she could be significantly increasing her risk of breast cancer. (Professional) |
| | VR12: You refer to "navigation menu". Nothing is called that, and although I guessed what is meant by it I wonder if it needs to be identified as such? (Professional) |
| Additions / expansions | VR8: On "How will my risk affect children...." it says boys who inherit the gene are not at risk of ovarian cancer (but does not say that they may however pass on the gene to their children) (Professional) |
| | VR9: Add an explanation of what the dotted lines are for Lynch women cancer risk in the general version (Professional; comment on: General version) |
| | VR8: Under main disadvantages of the operation - under sex can be painful - is it worth including the disclaimer on the menopause page, which gives some reassurance that help is at hand. (Professional) |
| | VR6: I thought that the initial surgery/no surgery section was rather weighted for surgery. There could have been a mention of the possibility of privately funded screening at this point. (Patient) |
| Repetition | VR10: "So for example, your risk of getting ovarian cancer in the next year when you are 35 is quite low, whereas your risk of getting ovarian cancer in the next year when you are 50 is higher." Repeats 'in the next year' (Professional) |
| Other | VR6: I'm still not totally convinced by the name, but I do appreciate the reasons for it! (Patient) |
| | VR10: "You should also be aware that the yearly smear test you receive is designed to detect cervical cancer and will not detect ovarian cancer." - I would give this a separate paragraph and make the 'not' bold! (Professional) |
| Website issues | VR3: The start of the text on the left hand side was missing all the way through (Patient) |
| | VR12: I assume/hope the balance will become an actual balance that tilts!?? (Professional) |

If comments were only applicable to a specific version, this is specified as (...comment on:...)

7.3.6 *The stand-alone Option Grids*

Following comments from the virtual reference group and the editorial team, the content of the stand-alone Option Grids was amended. Figure 7.9a and b show the stand-alone versions of the Option Grids for pre- and post-menopausal women as published in February 2013. The introductory paragraph was amended to include a definition of what is meant by 'increased risk'. The order of the questions in the grids was also changed and items were re-worded to facilitate understanding. The grids were then formatted to conform to the guidelines recommended in the Option Grid Development Framework V0.5. In line with these guidelines, an evidence document providing the studies and publications used to inform the answers to frequently asked questions was also created as a reference document (Appendix 7.8). Ultimately, the grids may also include the web address for the OvDex online tool to encourage patients to visit the full intervention if they require more information.

7.3.7 *OvDex: the Oophorectomy Decision Explorer*

The final version of OvDex was created from prototype II in response to feedback from the virtual reference group. Main sections and questions within each section remained the same; however, some content was re-worded and added. The research evidence section was extended to include evidence on coping advice and further references on the long-term health effects of premature menopause. Funders and collaborators were disclosed in a new section called 'About OvDex'. A copy of the final paper-based version of OvDex is included in Appendix 7.9.

The OvDex website was finalised by DigitalMorphosis (a Cardiff-based web design company). Personalisation was simplified, so that users could answer all three questions on one page (Figure 7.10). The online value clarification exercise layout was amended to include sliders instead of radio buttons, enabling users to weigh attributes on a scale of 1 to 10 instead of 1 to 3 (Figure 7.11). This allowed even further differentiation of the relative importance of different attributes.

Figure 7.9a - Final versions of the Option Grid (pre-menopausal women)



Oophorectomy for risk of ovarian cancer - before the menopause

This grid is to help **pre-menopausal** women at increased risk of ovarian cancer and their clinicians decide whether to remove the ovaries to reduce the risk of ovarian cancer. For women at increased risk, lifetime risk of ovarian cancer varies from 4 in 100 to more than 40 in 100 in some cases: because of this wide range in individual risk it is important to check your own risk with your clinician.

| Frequently asked questions | Surgical removal of the ovaries and fallopian tubes | No surgery |
|--|---|--|
| Will this reduce my risk of ovarian cancer? | Yes, removing the ovaries and fallopian tubes will reduce your lifetime risk of ovarian-type cancer to that of the general population, which is about 2 in 100. | No, your risk of ovarian cancer will remain increased. |
| Will I be able to become pregnant? | No, therefore it is important that you feel you have completed your family before having surgery. | Yes, your ability to become pregnant will be unaffected. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. | You may worry about developing ovarian cancer. |
| Will surgery change how I feel about myself as a woman? | Many women do not notice any change in how they feel about themselves as a woman. However, your desire for and/or enjoyment of sex may decrease. | Does not apply. |
| Will I go into menopause? | Yes, after surgery your oestrogen levels fall and the menopause will start immediately. | Yes, you will go through a natural menopause when you are around 50 years. |
| Are there any long-term health risks linked to surgery? | Yes, if surgery is done before age 45 there is a higher risk of bone thinning and cardiovascular disease. Some patients report memory changes as well. | Does not apply. |
| Will I need hormone replacement therapy (HRT) and is that safe? | HRT can reduce some of the effects of surgery. HRT is safe to take for the majority of women until they are 50. Discuss this with your clinician. It is not usually recommended for women who have had breast cancer. | No, if you go through a natural menopause you would not be expected to need HRT, unless your symptoms are very severe. |
| What are the risks of surgery? | About 4 in every 100 patients experience a complication during or after surgery. These can be minor infections or major complications. Discuss this with your clinician. | Does not apply. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after keyhole surgery and are back to normal in 4 weeks. For open surgery, time in hospital and recovery will be longer. | Does not apply. |
| Is there a routine screening programme to detect ovarian cancer? | No, after surgery there is no need for screening because your risk will be similar to that of the general population. | No, there is no evidence that screening for ovarian cancer is effective. |

Editors: Jana Witt, Kate Brain, Elissa Ozanne, Kate Lifford, Fiona Wood, Mark Rogers, Glyn Elwyn.

More information: <http://www.optiongrid.org/about.php>

Evidence document: http://www.optiongrid.org/resources/oophorectomyBefore_grid.pdf

Last update: 05-Feb-2013 Next update: 01-Jul-2013 ISBN: 978-0-9575461-1-0

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Figure 7.9b – Final versions of the Option Grid (post-menopausal women)



Oophorectomy for risk of ovarian cancer - after the menopause

This grid is to help **post-menopausal** women at increased risk of ovarian cancer and their clinicians decide whether to remove the ovaries to reduce the risk of ovarian cancer. For women at increased risk, lifetime risk of ovarian cancer varies from 4 in 100 to more than 40 in 100 in some cases: because of this wide range in individual risk it is important to check your own risk with your clinician.

| Frequently asked questions | Surgical removal of the ovaries and fallopian tubes | No surgery |
|--|--|--|
| Will this reduce my risk of ovarian cancer? | Yes, removing the ovaries and fallopian tubes will reduce your lifetime risk of ovarian-type cancer to that of the general population, which is about 2 in 100. | No, your risk of ovarian cancer will remain increased. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. | You may worry about developing ovarian cancer. |
| Will surgery change how I feel about myself as a woman? | Many women do not notice any change in how they feel about themselves as a woman. However, your desire for and/or enjoyment of sex may decrease. | Does not apply. |
| Will I get menopausal symptoms again? | No, if you have completed the menopause removing your ovaries should not cause any menopausal symptoms to recur. If you are peri-menopausal and have not quite completed the menopause, some symptoms may occur. | No, everything will remain the same. If you have completed the menopause you should not experience any more menopausal symptoms. If you are peri-menopausal and have not quite completed the menopause, some symptoms may occur. |
| What are the risks of surgery? | About 4 in every 100 patients experience a complication during or after surgery. These can be minor infections or major complications. Discuss this with your clinician. | Does not apply. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after keyhole surgery and are back to normal in 4 weeks. For open surgery, time in hospital and recovery will be longer. | Does not apply. |
| Is there a routine screening programme to detect ovarian cancer? | No, after surgery there is no need for ovarian screening because your risk will be similar to that of the general population. | No, there is no evidence that screening for ovarian cancer is effective. |

Editors: Jana Witt, Kate Brain, Elissa Ozanne, Kate Lifford, Fiona Wood, Mark Rogers, Glyn Elwyn.

Evidence document: http://www.optiongrid.org/resources/oophorectomyAfter_evidence.pdf

More information: <http://www.optiongrid.org/about.php>

Last update: 05-Feb-2013 Next update: 01-Jul-2013 ISBN: 978-0-9575461-2-7

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Figure 7.10 - Personalisation page on OvDex website

Helping you make the decision about risk-reducing ovarian surgery

Personalise OvDex

Please answer the following questions to the best of your knowledge. The information in OvDex will then be more relevant to your situation.

When you personalise the site we will set a cookie on your computer to allow us to personalise the site for you. By using this tool you give us permission to do this. If you would not like to personalise the information, go to [General](#).

Question 1 - Do you have a faulty gene or a family history of Lynch Syndrome?

- Yes, I have a faulty BRCA1 gene
- Yes, I have a faulty BRCA2 gene
- Yes, I am from a Lynch Syndrome family
- Don't know, I have not been tested for a faulty gene or my genetic test was uninformative
- No, I have been tested and no faulty gene was identified

Question 2 - Have you ever had breast cancer?

- No, I have never had breast cancer
- Yes, I have had breast cancer

Question 3 - How old are you?

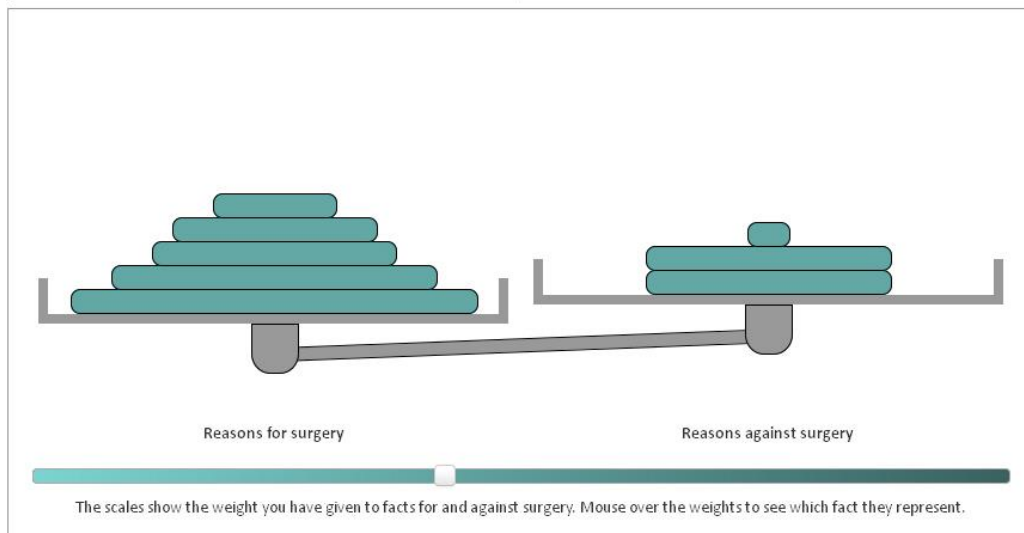
- I am under 35 years old
- I am between 35 and 39 years old
- I am between 40 and 49 years old
- I am 50 years old or older

Figure 7.11 - Example of interactive online value clarification exercise in final version

Weighing it up

This exercise might help you to weigh up the facts that make you want / not want to have surgery. Simply read the statements and decide how much they make you want to have surgery or avoid surgery and then move the slider. The scales will show where you stand with your decision.

| Makes me want to HAVE surgery | Does not come into my decision | Makes me want to AVOID surgery |
|---|--------------------------------|--------------------------------|
| Surgery will reduce my ovarian cancer risk | | |
| Surgery will reduce my breast cancer risk | | |
| I will not be able to become pregnant after surgery | | |
| I will go into the menopause after surgery | | |
| There is a risk of complications linked to surgery | | |
| There is no effective screening for ovarian cancer | | |
| The symptoms of ovarian cancer are very vague | | |
| I might feel different about my body after surgery | | |
| I might enjoy sex less after surgery | | |
| I will have to worry less about cancer | | |
| Enter your own reason | | |
| Enter your own reason | | |



7.4 Discussion

7.4.1 *The oophorectomy patient decision support intervention*

The current chapter outlines the development of a decision support intervention for women at increased genetic risk of ovarian cancer. Development was guided by the RRSO-adapted CODE framework, and other relevant theories, and was supported by a group of stakeholders, who provided feedback throughout. Figure 7.12 outlines which items within the RRSO-adapted CODE framework are covered by the content of the intervention. Questions relating to cognitive appraisal were primarily addressed by the information content within the Option Grids and OvDex. Emotional appraisal items were more difficult to address, as the questions relating to emotional appraisal are more abstract and depend on individuals' past experiences and personal values. Chapter 6 showed that previously developed patient decision support interventions did not support emotional appraisal. In OvDex, these questions were indirectly addressed by some of the coping advice, by components designed to facilitate affective forecasting and within contacts and resources. For example, the questions 'What did others decide? How did they fare?' may be addressed by patient testimonials. Although OvDex did not include patient testimonials itself, it does refer to other websites that include patient blogs and to patient networks. Affective forecasting may also be supported by patient testimonials. Additionally, information in OvDex was worded in such a way as to facilitate affective forecasting by describing possible physical, psychological and social outcomes that may be experienced following surgery. Items within the secondary appraisal section, which focus on possible coping options, were covered by coping advice. Finally, weighing of attributes and alignment of options with values and goals, a part of the preference construction phase, was supported by the value clarification exercise.

Figure 7.12 - RRSO-adapted CODE framework and items covered by Option Grid and OvDex content (indicated in colour)

| Deliberation | | | | | |
|----------------------------|--|---|--|--|--|
| | Disclosure of ovarian cancer risk status | Presentation of choice | Presentation of options (RRSO or symptom awareness) | Preference construction | |
| Coping | Cognitive | What is ovarian cancer (OC)? What is OC risk? How high is my risk? What caused me to have an increased risk of OC? How does my family history influence OC risk? Do I have a faulty gene? Does my risk change over time? How could my risk affect my children? How will my risk affect my life? What can be done about my OC risk? | What does 'choice' mean in this context? Why is there a choice for women at risk of OC? When should I decide? What is at stake in this choice? Who can decide? Who else should I involve? Can I change my mind? How can I decide? | What is risk-reducing bilateral salpingo oophorectomy (RRSO) / symptom awareness? Why is symptom awareness the only alternative? What does RRSO involve? How long would it take to recover from RRSO? What would happen if I do not have surgery? What are the risks and benefits of RRSO / symptom awareness? What are the complications of RRSO? Would I go into an early menopause? Would I have to take hormone replacement? How would RRSO affect my life? Do I need to have my womb removed as well? Is there a chance cancer might be found during RRSO? Is there any screening available for OC? Is there anything else I can do to avoid OC? What did others decide? How did they fare? Can I make sure I get a good surgeon in a good hospital? | Is this the right time to decide? Is RRSO / not having RRSO congruent with my/my family's/partner's/doctor's beliefs goals and values? Have I completed my family? Can I afford to take time off? Could I get screening or some other additional care? Do I think I could detect the symptoms of OC? Do I feel ready to decide? How likely is it that I will regret having / not having RRSO? |
| | Primary Appraisal | What are my past experiences of OC? How do I feel about OC and my risk? | How do I feel about choosing? Can I deal with choosing myself? | Do I have experiences that could help me imagine what it would be/feel like if I had / didn't have RRSO? How do I feel about having / not having RRSO? | |
| | Emotional | | | | |
| | Problem-focused | Can I find out more? Can my doctor help? Can I trust my doctor? Can I seek instrumental / informational support from my family / friends / a peer or support group? | Can I create an action plan to approach this decision? Can I seek instrumental / informational support from my family / friends / a peer or support group? | Is there someone or something that could help me to compare and weigh my options? Can I speak to someone who has already made this decision and is living with the consequences? Can I seek instrumental / informational support from my family / friends / a peer or support group? | |
| Secondary Appraisal | Can I seek emotional support from my family / friends / a peer or support group? Can I turn to my faith? Can I change how I feel about knowing my OC risk? Can I express my fear of cancer? Can I do something to stop thinking about my OC risk? Can I do something to reduce the stress my OC risk is causing me? Could I ignore my OC risk? | Can I seek emotional support from my family / friends / a peer or support group? Can I let someone else decide for me? Could I refuse to choose and just not make this decision? | Can I seek emotional support from my family / friends / a peer or support group? Can I do something to stop thinking about this decision? Can I do something to reduce the stress this decision is causing me? | | |
| Emotion-focused | | | | | |

Colour Code:

- Blue** – Covered by information content in OvDex
- Purple** – Covered by information content in Option Grids and OvDex
- Red** – Covered by coping advice
- Pink** – Indirectly covered by coping advice or contacts & resources
- Yellow** – Covered by value clarification exercise
- Black** – Not covered

The intervention was split into two parts: an Option Grid, designed for use in clinic, and a more extensive online intervention called OvDex, designed for use by patients at home. It was anticipated that this separation would accommodate individual differences in information processing and decision making style, for example, catering for immediate and delayed deciders as identified in Chapter 4. The former are able to make a decision relatively quickly once presented with the options, while the latter tend to take a more deliberative approach. Hence, immediate deciders may appraise the health threat and their options comparatively briefly and are unlikely to go through all the questions proposed in the RRSO-adapted CODE framework before being able to make a decision. Instead, they may use fast and frugal heuristics to identify the best option by only considering a limited number of questions. Heuristics have been shown to be useful in decision making, especially when decisions are time sensitive and only limited information is available (Gigerenzer 2004). Gigerenzer (2004) outlined two classes of heuristics based on reasoning: 'one reason' / 'take-the-best' decision making and 'tallying'. The former relies on a single attribute that allows individuals to choose one option over another. 'Take-the-best' decision making requires that only the attribute with the highest validity is considered before a decision is made. The best option is that which has a positive value on the attribute with the highest validity. If options were identical on this attribute, the attribute with the second highest validity is considered and so on, until options differ. In contrast, 'tallying' assesses options based on two or more attributes. Values are assigned for each attribute and then added up for each available option. The best option is that which has the most positive values on attributes considered.

Option Grids are designed to facilitate heuristics based decision making, as they allow both 'take-the-best' and 'tallying' to be used to discern the best option (Elwyn et al. 2013b, Marrin et al. 2013). The questions within the Option Grid act as cues, allowing a direct comparison of options with regards to specific attributes. Immediate decision makers may select one question and base their decision on the

answer to that single question, a process that may be described as a 'take-the-best' approach. For example, an individual may consider a reduction of their cancer risk as the attribute with the highest validity due to their past experience of cancer. They may therefore only look at the question "Will this reduce my ovarian cancer risk?" and base their decision solely on the answer to this question. In this case, the individual would elect to have surgery, as RRSO reduces cancer risk, whereas avoiding surgery does not. Alternatively, decision makers may opt to consider several or all questions listed in the Option Grid and make a decision based on the answers to those questions, a process that may be described as a 'tallying' approach. For instance, they may consider the reduction in ovarian cancer risk to be a positive attribute of surgery; however, retaining their ability to become pregnant and avoiding a premature menopause may be considered positive attributes of the no surgery option. If no further attributes were considered, this individual would decide against surgery in this instance.

The information contained within the Option Grid may thus be sufficient for immediate deciders to make their choice, without the need for more elaborate decision support. However, for others decisions about how to manage their increased risk of ovarian cancer may be more complex and distributed over time (Rapley 2008). Therefore, fast and frugal heuristics, may not be the preferred decision making approach for delayed deciders. These individuals may favour a more deliberative process and could benefit from additional support. In agreement with this, a randomised trial of a CD-Rom decision support intervention for carriers of mutations in BRCA1/2 by Schwartz et al. (2009) found that women who struggled with the decision benefited from the intervention, whereas women who were able to make an immediate decision, did not.

Deliberative decision making approaches may be described by theories such as Janis and Mann's (1977) conflict model of decision making and Svenson's (1992) differentiation and consolidation theory. They also map more fully onto the CODE framework (Witt et al. 2012), as delayed deciders are likely to consider more of the

questions proposed in the RRSO-adapted CODE framework than immediate deciders, as they seek out additional information and appraise coping options. Deliberative decision making includes detailed cognitive and emotional appraisal of information relevant to the health threat, as well as the choice and available options. Delayed deciders may also employ a number of problem- or emotion-focused coping strategies in order to deal with the decision. These strategies may change and be adapted over time. Supporting delayed deciders therefore requires more extensive decision support than that needed for immediate deciders.

The intervention presented in this chapter therefore attempts to cater for this population with its two complementary parts. During clinical consultations, when options are first discussed, patients could receive an Option Grid. In line with Svenson's (1992) differentiation and consolidation theory, Option Grids facilitate direct comparison of options (Elwyn et al. 2011b). They also enable patients to initiate a discussion about options with their clinician (Elwyn et al. 2013b). However, the information provided in the grid may not be perceived as sufficient by delayed deciders, who experience decisional conflict and report several barriers to decision making (Janis and Mann 1977). While they may ask their clinician for further information during the consultation, delayed deciders may then return home without having made a decision. These individuals may appreciate access to additional information, which they can review any time and in consultation with close others. For this reason, the Option Grid is complemented by a more extensive online tool that delayed deciders can choose to visit in order to obtain more information to base their decision on (Janis and Mann 1976, 1977). It should of course be noted that immediate deciders are also free to visit the OvDex website; however, they may choose not to do so.

The structure and content of OvDex was guided by the phases and questions outlined in the RRSO-adapted CODE framework (Chapter 5). Answers to questions were worded to facilitate the cognitive and emotional appraisal of each phase of the deliberation process (Power et al. 2011, Wilson and Gilbert 2003). To support

affective forecasting, answers encouraged patients to imagine the physical, social and/or psychological effects of different outcomes and to discuss these with health professionals and/or important others (Joseph-Williams et al. 2013).

Affective forecasting and emotional appraisals may also be facilitated by patient stories; however, the evidence for this effect is controversial (Bekker et al. 2012, Khangura et al. 2008, Winterbottom and Bekker 2009, Winterbottom et al. 2008). While patients generally like personal accounts, as these help them to imagine possible futures, there is concern that stories may bias patients' forecasting and decision making, as a balanced portrayal of all possible outcomes is basically impossible and patients often identify with a particular person's story (Bekker et al. 2012). Therefore, OvDex does not include patient stories directly; however, within the contacts and resources list it does refer users to other sites that include patient stories, blogs and networks.

Coping advice was also provided within OvDex to support coping efforts during decision making (Balneaves and Long 1999, Folkman 2011, Lazarus et al. 1985, Witt et al. 2012). Patients were encouraged to seek information, to actively plan their decision making and to explore their options with professionals and important others, all of which reflect problem-focused coping options (Lazarus et al. 1985). Patients were also advised that emotional reactions to the situation were normal and were encouraged to seek emotional support from family, friends and/or peers and to positively reappraise the situation, which reflect emotion-focused coping strategies (Lazarus et al. 1985). Active distraction, mind-body techniques and relaxation exercises were also referred to, as these may be useful for patients to regulate intrusive thoughts. This is especially relevant in periods when no action can be taken, such as when waiting for results of genetic tests (Phelps et al. 2013, Phelps et al. 2006).

Finally, the value clarification component was developed to facilitate preference construction during deliberative decision making. In contrast to immediate deciders, who may prefer fast and frugal heuristics, deliberative

deciders may want to consider most, or all, of the information relevant to their decision and prefer to decide using what they have learnt about different attributes and how important those attributes are to them (Janis and Mann 1976, 1977, Svenson 1992, 2003). The exercise within OvDex was designed to facilitate such deliberative approaches by allowing users to identify attributes as either gains (reasons to have surgery) or losses (reasons to avoid surgery) and simultaneously assign a value to their relative importance. While this exercise might at first glance resemble a tallying approach, and therefore heuristics based decision making, it is more complex than that. It assumes the user has done research and knows and understands the attributes of options and it additionally allows a weighing of relative importance, not usually part of tallying (Gigerenzer 2004, Janis and Mann 1976, Svenson 1992).

7.4.2 Practice implications and future research

Currently, there is a lack of decision support materials that can be used outside the context of clinical consultations and is specifically designed for women at increased genetic risk of ovarian cancer in the UK (Chapter 6). Many women in this situation report that they struggle to identify relevant and trustworthy information and that additional, non-educational materials would also be useful (Chapter 3 and 4; Babb et al. 2002, Hallowell 2000). Combined with face-to-face counselling in consultations, such materials may improve patient experience and satisfaction (Coulter and Ellins 2007). Therefore, a patient decision support intervention that integrates cognitive and emotional aspects of deliberation and coping processes could complement genetic services in the UK and support patients more holistically (Chapter 2 and 5; Witt et al. 2012). Health professionals agree that such materials would be helpful as complementary adjuncts to consultations. Hence, the intervention developed here addresses a need that has been voiced by patients and health professionals alike.

However, patient decision support interventions have faced substantial implementation issues in the past, partially owing to the fact that many were

developed for independent use by patients and were therefore not readily integrated into clinical pathways (Elwyn et al. 2008, Frosch et al. 2011, Harter et al. 2011). To counteract these implementation issues, the current intervention is split into two parts. Implementation of the Option Grids may be more straightforward than implementation of more extensive self-administered interventions has been in the past. Immediate deciders may be able to make a decision based on information delivered in the grid and clinical discussion alone; however, delayed deciders may want more information. Therefore, the Option Grids will include a reference to OvDex, which patients can choose to visit following the consultation.

The full OvDex tool may then help those patients who struggle with the decision to obtain evidence-based, up to date information, to share their thoughts with important others and to weigh their values and preferences. It allows patients to explore issues over longer periods of time, to take breaks and return to the information whenever they wish and to examine their options in the comfort of their own home, before returning to clinic for more detailed discussion. As preferences change and develop over time, they can re-read the information and repeat the exercise until they are ready to make a final decision. This is in accordance with Rapley (2008), who put forward a model of *distributed* decision making, proposing that many decisions are not made in a single clinical consultation, but are distributed across time, spanning numerous encounters with others, various situations and interactions with technologies (Montori et al. 2006).

OvDex may also help those who make an immediate decision against surgery due to family planning issues, but who may wish to return to the decision and reconsider surgery at a later date after their family is complete. For these individuals the intervention is a resource they can come back to when they are finally ready to decide, as it allows them to obtain up to date information and clarify their values before then making an appointment to discuss further details.

Both the Option Grids and online tool developed here require further evaluation in order to assess whether they are acceptable to patients and health

professionals, whether they truly support decision making and coping, and whether they can be implemented as proposed.

7.4.3 Strengths and Limitations

The development process outlined in this chapter has several strengths. Firstly, it was guided by recommendations and guidelines written by experts in relevant fields. The development of the Option Grids adhered to the protocol produced by the Option Grid Collaborative (2013). The development of OvDex followed recommendations set out by the International Patient Decision Aid Standards (IPDAS) collaboration (Volk and Llewellyn-Thomas 2012), as well as those specifically proposed for web-based decision support tools by Elwyn et al. (2011a). In line with these recommendations, the development process was also supported by a group of stakeholders, who were consulted throughout, to ensure that content was easy to understand and relevant to this medical decision.

The CODE framework was used as the practical and theoretical basis for the development of this patient decision support intervention (Chapter 2; Witt et al. 2012). The structure and content of the intervention was guided by the phases and questions proposed in the RRSO-adapted CODE framework, which had been created using data from a literature review, as well as a focus group and interview study (Chapter 5). In addition to the CODE framework, some supplementary theories and models were also used to inform content, coping advice and the design of the value clarification exercise (Gigerenzer 2004, Janis and Mann 1976, Lazarus et al. 1985, Svenson 1992, Wilson and Gilbert 2003). Therefore, all parts of the intervention are firmly grounded in theory.

The development process demonstrated that an adapted version of the CODE framework can be used readily as the basis for the development of patient decision support. It clearly sets out questions, issues and coping options patients might explore during deliberations about specific medical decisions and therefore provides developers with a clear guide of what issues they should address in an

intervention. As this intervention is the first to use an adapted version of the CODE framework as theoretical basis, it is not possible to determine at this point whether the intervention will be able to achieve the goals set out in the introduction. Furthermore, there is as yet no empirical evidence to show that Option Grids have the same positive effects on decision making as longer tools (Stacey et al. 2011), although they have been shown to facilitate clinical consultations and enhance patient involvement in SDM (Elwyn et al. 2013b). Therefore, the effectiveness of the individual components of the intervention is currently unknown and will require detailed prospective evaluation.

Some further limitations should be noted. The intervention is only relevant to the UK context, as it conforms to NICE guidelines with respect to ovarian cancer screening, symptoms and chemoprevention using the oral contraceptive pill (National Institute for Health and Care Excellence 2011, 2013). Hence, it may need to be adapted if used outside the UK. Furthermore, the outcomes of the UKFOCS study are expected to be published soon, therefore the content of the Option Grids and OvDex, particularly the 'other options' section, will need to be updated regularly to guarantee their accuracy.

One further limitation concerns accessibility. The Option Grids are available for download from the Option Grid Collaborative website. Ideally, they should be printed by professionals and then handed to patients as a paper copy. The OvDex website, however, requires patients themselves to access the internet, which may limit its use, as those without internet access might struggle to obtain a copy. A paper-based version was therefore made available, which may be printed by professionals directly from the website for those patients who do not have ready access to the internet.

7.4.4 Conclusions

This chapter reports on the development of a patient decision support intervention using an adapted version of the CODE framework (Witt et al. 2012).

The intervention is split into two parts that are designed to facilitate immediate and delayed decision making by providing an Option Grid as well as a more extensive tool. Although this intervention was developed in response to an identified need reported by stakeholders (Chapters 3 and 4) and due to a lack of appropriate decision support for patients in the UK (Chapter 6), it needs to undergo usability testing before it can be made fully available to patients. The next chapter reports on the results of a usability testing study.

8 Usability testing of a decision support intervention for patients considering risk-reducing bilateral salpingo-oophorectomy

8.1 Introduction

Decision support interventions are designed to support decision making in situations when decisions are preference-sensitive and depend on the goals, values and preferences of patients (O'Connor et al. 2004, Stacey et al. 2011, Thistlethwaite et al. 2006). Developers of extensive self-administered interventions, such as websites, DVDs and booklets, have experienced substantial implementation issues, which has led some researchers to suggest that shorter tools might fit better into clinical pathways (Elwyn et al. 2008, Elwyn et al. 2013b, Elwyn et al. 2013c). In the previous chapter a two part decision support intervention for women at increased genetic risk of ovarian cancer was developed consisting of an Option Grid and a longer tool, called 'OvDex'.

Before publication and dissemination, developers need to ensure that patient decision support interventions are user-friendly, easy to understand and deliver information relevant to the decision in a balanced and unbiased way (Elwyn et al. 2006, Elwyn et al. 2009b, Joseph-Williams et al. 2013). This may be achieved by involving stakeholders throughout the development process and by carrying out additional usability assessments (Elwyn et al. 2011a). Testing usability is especially important for web-based tools, such as OvDex, to ensure that websites are accessible, easy to use and navigate, and that functionality works as expected (Elwyn et al. 2011a, Hinchliffe and Mummery 2008, Hoffman et al. 2012, Reisman 1996, Rubin and Chisnell 2008). This chapter aims to (i) assess usability of the patient decision support intervention developed in Chapter 7, (ii) evaluate perceived helpfulness of the intervention for facilitating decisions about RRSO and (iii) report user suggestions for implementation.

8.2 Methods

This study received approval from the Multi-Centre Ethics Committee for Wales (Ref: WA-11-0094, Amendment 3; Appendix 8.1).

8.2.1 Recruitment of reviewers

Due to the problems experienced when recruiting through the clinical genetics services for previous studies (Chapter 4 and 5), it was decided that recruiting volunteers from a relevant charity's database was a suitable alternative for usability testing. While it was hoped that this would lead to a higher response rate, it could also result in responses from women who were not at increased genetic risk of ovarian cancer and who were therefore not the intended target audience for the intervention. However, lay reviewers from various backgrounds may be able to make a valuable contribution regarding usability (Elling et al. 2012, Sawka et al. 2011). The only eligibility criteria for this study, therefore, were that participants were female and aged between 30 and 80 years. Only women who could not give informed consent were excluded.

Volunteer reviewers were recruited through the charity Target Ovarian Cancer, who sent out an email, Facebook and Twitter invite to their registered supporters. Recruitment was open for two days (1st until 2nd of November 2012) and was stopped once over 50 responses had been received. The study aimed to conduct a total of 15 telephone interviews, as previous research suggests that small numbers are sufficient during usability testing (Kushniruk et al. 1997, Nielsen 1994). Volunteers responded to the invite by registering their interest in the study. The first 25 respondents were sent a study pack, which contained an information sheet and a consent form. Contact details of all other respondents were kept on file. In case less than 15 of the first 25 individuals consented to take part, respondents on file would be sent a study pack, until at least 15 had been consented. Once consent had been received, participants were registered on the study and a date and time for interview was arranged when the OvDex website became available online.

8.2.2 Usability testing

Usability testing requires users to provide feedback about a website, or other product, often in interviews or focus groups, or via questionnaires (Battleson et al. 2001, Elwyn et al. 2011a, Hom 1998). Techniques such as cognitive interviews, which are usually used to validate questionnaires, may also be useful in usability testing (Willis 2005). The current study employed a cognitive interviewing technique termed 'verbal probing' to conduct usability testing (Willis 1999, 2005). During verbal probing, the interviewer asks targeted questions, which the participant answers. The interviewer then probes for further information, asking the respondent to expand upon, explain and/or qualify their answer (Boyce and Neale 2006). In the current study, participants were provided with the decision support intervention (Option Grids and access to the OvDex website/booklet) before the interview and could look at the materials in their own time. Participants could select to access the OvDex website online or request a printed copy of the OvDex booklet. Participants were asked to make notes on their thoughts whilst reviewing the materials and to have the Option Grids and OvDex tool, as well as any notes they had made, to hand during the telephone interview. Interviews were conducted in March 2013. The interview guide is included in Appendix 8.2.

8.2.3 Data analysis

Interviews were audio recorded and transcribed prior to being analysed using thematic analysis (Boyatzis 1998). The coding frame is included in Appendix 8.3. Four interviews (30%) were double coded by two independent coders and any discrepancies were discussed in meetings until agreement was reached. Coded extracts were managed and analysed using the data analysis software NVivo8 (QSR International Pty Ltd 2008). Analysis was specifically focused on (i) how information content was understood and processed by participants and (ii) identifying any problems with content, accessibility and functionality. Additionally, perceived helpfulness and effects were discussed and possible implementation strategies were explored. A sample of a coded transcript is included in Appendix 8.4.

Quotes included in this chapter act as examples of themes and codes that emerged from the data. Sections where quotes were edited to remove repetitions and irrelevant portions are denoted by [...]. Participant characteristics are indicated in parenthesis after each quote. Characteristics are given in the following order: Age group (under35, 35to39, 40to49, over50), previous ovarian cancer diagnosis (yes/noOC), family history of breast cancer (BC) or ovarian cancer (OC) or both (B&OC) (noFH, BCFH, OCFH, B&OCFH) and gene status (unknown, BRCA1/2).

8.2.4 Editing of decision support intervention

Following analysis of the interview data, reported problems and suggestions for improvements were collated and discussed within the supervisory team. In response, changes to the structure and content of the Option Grids and OvDex tool were made to improve usability and understanding.

8.3 Results

8.3.1 Response rates

The first 25 respondents to the invitation were sent an information pack and consent form. Of these, 19 consented to be interviewed. Due to a time delay of two months between consent and invitation to interview, dates were agreed with 17 of 19 consented participants (10% attrition). Thirteen interviews were conducted in total. One woman cancelled due to poor health, another due to bereavement and two women did not answer their phone on the day of the interview (24% attrition). Characteristics of the 13 interviewees are outlined in Table 8.1.

Interviews lasted between 21 and 49 minutes, with a mean interview time of 35 minutes. The majority of participants were over the age of 50, had been directly affected by ovarian cancer and had not been tested for a genetic mutation. One respondent (R2) had only viewed the Option Grids, but not the OvDex tool, at the time of the interview. Therefore, the analysis includes feedback on Option Grids from 13 reviewers and comments on the OvDex tool from 12 reviewers.

Table 8.1 - Characteristics of interviewees

| Characteristic: | | No of women (n=13) |
|-----------------------------|---------------------------------|-----------------------|
| Age group | Under 35 | 2 |
| | 35 to 39 | 2 |
| | 40 to 49 | 2 |
| | Over 50 | 7 |
| UK Region | North England | 3 |
| | South England | 7 |
| | Scotland | 2 |
| | Wales | 1 |
| Prior cancer | No | 5 |
| | Yes | 8 |
| Family history | Ovarian cancer only | 2 |
| | Breast cancer only | 4 |
| | Both | 3 |
| | None | 4 |
| Gene mutation status | Not tested | 9 |
| | BRCA 1 | 1 |
| | BRCA 2 | 3 |
| | Lynch mutation / family history | 0 |

8.3.2 Option Grids

8.3.2i Layout and structure: The majority of participants were satisfied with the layout and structure of the Option Grids. They described them as ‘easy to follow’ and ‘clear’ and most felt that options were portrayed in a balanced way.

R9: I could see exactly what you were doing. You had your [...] frequently asked questions and then you were giving the answers [...]. I thought it was very good for being able to balance things up. (Over50, yesOC, noFH, unknown)

R2: I find the Grid a lot easier to read as well than if it was just written [...] questions [...]. If it was just questions on a paper with answers I don't think it's as easy to read as a Grid is. (Under35, noOC, B&OCFH, BRCA2)

One respondent felt that the condensed nature of the grid allowed her to focus on the most salient questions first, rather than having to read through the detailed information included in the full online tool.

R12: ...if I [...] sat down with just your whole web page, I'd probably be a bit overwhelmed, so I think that the table does a really good job of just distilling the [...] obvious things that I would expect to have concerns about. (Under35, noOC, OCFH, unknown)

Overall, respondents seemed to like the layout and structure of the Option Grid and found it useful as a first summary of options, benefits and harms. One respondent made the suggestion that the column with the frequently asked questions should be more distinct from the columns with the answers, to help interpretation.

R1: I thought they were quite good. [...] from an ease to read point of view, there just needs to be a line to separate the frequently asked questions and then [...] what the options were, because I read it first off and then was a little bit confused. (35to39, noOC, B&OCFH, BRCA2)

8.3.2ii Order: When asked about the order of the questions within the grid, participants' responses were divided. Some felt that the current order was logical and did not need to be changed.

R12: I thought it made perfect sense, because it starts out with the things that I'm most concerned about. [...] I think it really made sense, it was very easy to read. (Under35, noOC, OCFH, unknown)

R9: I think the order of them was very clear and it was quite a logical sort of progression. [...] I didn't think that anything there needed to be re-jigged or anything. (Over50, yesOC, noFH, unknown)

Others felt that the very last question concerned with screening for ovarian cancer should be moved towards the top of the grid.

R8: I think the routine screening question should probably be closer to the top. Although it's a bit of a negative then, but it's still I think one of the first things you think about. (35to39, noOC, OCFH, unknown)

R5: I think possibly I would say the routine screening programme should be higher, because that's a major thing, because if there's some screening programme you wouldn't want to go any further. (Over50, yesOC, noFH, unknown)

Other than moving the screening question further to the top of the grid, most respondents were satisfied with the order of questions. Only one woman commented on further possible restructuring; however, this respondent also added that the order did not 'make much difference'.

R2: My concerns were the menopause, the HRT and the health risks if I had surgery. For me they are the most important. For me they would want to be at the top. That's my feeling anyway. (Under35, noOC, B&OCFH, BRCA2)

8.3.2iii Understanding and questions: Respondents were asked whether they found the information within the Option Grids easy to understand, whether they felt there were any problems with terminology and whether any information was missing. Most respondents were satisfied with the content of the grids and felt that the selected questions reflected the most salient issues they would explore when making a decision about RRSO.

R5: I thought the questions were the sort of questions that you would be asking I think, definitely, and I couldn't think of anything other. (Over50, yesOC, noFH, unknown)

R9: There weren't a lot of questions or queries I had about anything. There wasn't a lot of things that jumped out at me and I thought: 'Oh, I don't understand that!' [...] I thought it was really very clear. (Over50, yesOC, noFH, unknown)

R3: I thought it was quite helpful, quite clear. [...] It answered all the questions that anybody would want to know I think. (Over50, yesOC, BCFH, BRCA2)

A few women provided detailed accounts of how they cognitively processed the information presented in the Option Grids and asked questions that had arisen whilst reading the grid. Some of these responses highlighted specific problems with terminology and wording that might result in misunderstandings or misinterpretations. For instance, Respondent 6 queried the wording of the opening paragraph, which read 'This grid is to help pre-menopausal women at increased risk

of ovarian cancer and their clinicians decide whether to remove the ovaries to reduce the risk of ovarian cancer’.

R6: Most people will read this and think: ‘Hang on, I’m getting my ovaries taken out so surely that’s going to completely remove the risk of ovarian cancer.’ But you do [...] later on explain that peritoneal cancer is part of the ovarian cancer class [...]. I wonder if that’s really clear to your average person on the street. (40to49, yesOC, BCFH, BRCA1)

This respondent felt that within the Option Grid it was not clear why RRSO only ‘reduces’ the risk of ovarian cancer, rather than eliminating it. She acknowledged that explanations within the OvDex tool clarified that primary peritoneal cancer was considered an ovarian-type cancer. However, she felt that other women may struggle to understand the current Option Grid opening paragraph.

Regarding risk estimates, one respondent felt that it was important that users of the Option Grid should be informed about their personal risk level before viewing the grid to avoid causing misunderstandings and anxiety due to the wide range of risk described in the opening paragraph.

R12: In the little explanation where you talk about [...] ‘life time risk varies from four in a hundred to forty in a hundred’ [...], as a patient, will I have been told what, where I am on that scale before I see this grid? [...] Otherwise that’s quite scary information, you think: ‘Oh god, which one am I?’ (Under35, noOC, OCFH, unknown)

Another respondent struggled with the meaning of ‘increased’ or ‘high’ risk, which to her, as a cancer patient, meant the likelihood of cancer spread and relapse, whereas within the Option Grid and the OvDex website, this term referred to the likelihood of cancer developing. However, she acknowledged that ultimately she understood how the term was used in the intervention and that women who had not been directly affected by cancer may not struggle with the interpretation of this term as she did.

R11: Quite a lot of the time I was trying to define what was high risk. So I now understand what you’ve defined as high risk [...]and that’s not

quite the same as how my [cancer care] team would define high risk [...]. Your high risk is predicting occurrence; their high risk was predicting reoccurrence. (Over50, yesOC, noFH, unknown)

One of the youngest respondents commented that the fourth question 'Will surgery change how I feel about myself as a woman?' was the least helpful question, as it did not provide any statistics or factual information.

R12: The 'how I feel' question [...], I can see why it's in there, but I wasn't entirely sure that [...] the answers actually added anything. [...] It doesn't give you any facts, [...] it's quite bland. [...] Have women who have had to have this operation at thirty-three, or chosen to have this operation at forty-three [...] come back and said: 'Oh god, I really regret it, I wish I hadn't had it!' I think that sort of information would be more useful. (Under35, noOC, OCFH, unknown)

The same respondent also reported that other questions in the Option Grid were helpful in highlighting some of the important issues she would consider and in leading to further, more detailed, questions about these issues.

R12: If this was something I was having to go through I had quite a lot of questions around the menopause [...] and this box [...] starts to tease some of those out, which I thought was really useful. [...] What are the likely side effects of having [...] this operation in terms of the menopause, emotionally speaking? If you have the menopause and you're much younger, what have other people's experiences been? (Under35, noOC, OCFH, unknown)

She then acknowledged that many of the questions that had arisen whilst reading the Option Grid had later been addressed and resolved within the OvDex tool.

R12: Having gone on to read the rest of the information pretty much all of that stuff is covered [...] in the main documents, so that was really helpful. (Under35, noOC, OCFH, unknown)

Respondent 6 described her reaction to some of the wording within the grid, particularly referring to the final question about the availability of screening for ovarian cancer. She felt that, by stating that the remaining cancer risk following

surgery was 'similar' to that of the general population, the perceived impact of surgery was reduced.

R6: It says: 'No after surgery there is no need for screening because your risk will be similar to that of the general population' and I thought: 'Well surely it's going to be lower actually?' [...] So I just thought well maybe using the word similar there isn't really showing that there's much of a difference between you having gone through the operation and somebody out of the general population who has not. So it's almost like making you think: 'Oh hang on a minute there's really no benefit then of having the operation.' (40to49, yesOC, BCFH, BRCA1)

Some respondents had knowledge of screening or diagnostic tests that were being trialled or used and therefore queried the answer to the final question in the Option Grid, which stated that no medically effective screening was currently available.

R1: [The grid] was very black and white, [either] you have surgery or you don't. And it's almost like if you don't have surgery you take your life in your own hands, whereas there is the possibility of having ultrasounds for early detection [...]. I'm lucky that I can get that done but I don't know if other [NHS] Trusts [...] do that, so I suppose that's quite a tricky thing. (35to39, noOC, B&OCFH, BRCA2)

Overall, many respondents were positive about the content of the grids and felt that the information was clear, easy to understand and addressed the salient questions they had thought of. However, some described their cognitive processing of wording and terminology in detail, and, in doing so, highlighted potential areas for improvements. Some also reported further questions that had been inspired by the information in the grid. Many felt that those issues were later addressed in the OvDex tool.

R1: Your website bit explains it a lot better than the grid and gives you all the [...] ups and downs and [...] the time scales and everything like that. (35to39, noOC, B&OCFH, BRCA2)

8.3.2iv Improvements: In addition to discussing issues with layout, terminology and content, which highlighted areas where amendments may be required, respondents also made a number of direct suggestions for improvements to the Option Grids. For example, one woman felt that, from her own experience, the information about recovery was insufficient and should be supplemented with further details.

R13: It might be a good idea just to mention that it can be quite traumatic afterwards for a few weeks. (Over50, yesOC, BCFH, unknown)

Another respondent felt that the answer to the last question about screening for ovarian cancer in the surgery column was inadequate.

R10: If you were having surgery [the answer] didn't answer the question. [...] Whereas if you were not having surgery it did answer the question, because there isn't [a screening programme]. It should have said: 'No there isn't a screening programme and after surgery anyway...' I feel it needs a little addition. (Over50, yesOC, BCFH, unknown)

One respondent commented specifically on the Option Grid for post-menopausal women and felt that this should include some information about hormone treatment options for this population.

R4: On the post menopausal women thing I think that there could be something added about taking HRT [...] afterwards, because I am post menopausal obviously, but I was on HRT before I was diagnosed and have had to go back onto some oestrogen only HRT after. (Over50, yesOC, noFH, unknown)

8.3.2v Amendments to Option Grids: Overall, participants' comments highlighted 16 areas where changes to the Option Grid content may be required. A full list of these, including the decisions of the supervisory team, can be found in Appendix 8.5. In total 10 changes were actioned (Table 8.2) and the Option Grids were reviewed and amended accordingly to improve layout and enhance understanding (Figure 8.1). Option Grids are not designed to be comprehensive;

Table 8.2 - Changes made to Option Grid in response to lay reviewer feedback

| Option Grid section | Action / Change |
|------------------------|---|
| Introductory paragraph | Wording of opening paragraph was changed to "...surgery reduces risk of ovarian-type cancer..." |
| Layout | Frequently Asked Questions column was coloured in light orange |
| Question order | Screening question was moved up to be the second question in the grid |
| Question 3* | Factual information about satisfaction with RRSO was added |
| Question 4* | Wording changed to "...is likely to decrease" |
| Question 8* | Added statement about post-surgical effects during recovery |
| Question 9* | Reworded answer to screening question in the surgery column to not include 'similar to general population' |
| Question 9* | Reworded answer to screening question in the surgery column to be more relevant to the question |
| Question 9* | Reworded answer to screening question to be more relevant to emphasise that there is no medically proven screening |
| Question 9* | Reworded answer to screening question to explain why there is no evidence that screening is effective (i.e. no proven survival benefit) |

*Questions are numbered according to their order in original grid viewed by lay reviewers

instead, they are intended to provide a scaffold for better conversations about options between patients and health professionals in clinic. It should, therefore, be noted that the Option Grid was being used outside its intended context in this study. Hence, requests made by respondents for further information to be included in the grid may have been due to the unavailability of a health professional at the time of viewing the grid. Furthermore, limitations on the size of Option Grids, which should fit on one A4 page, mean that information in grids has to be selected on a priority basis. For these reasons, the remaining potential changes proposed by respondents in the current study were not actioned. Most of these were requests for additional information, which was available and covered in the full OvDex tool and/or may be discussed with health professionals whilst viewing the grid.

Figure 8.1- Amended version of the Option Grid for pre-menopausal women



Oophorectomy for risk of ovarian cancer - before the menopause

Use this Grid to help you and your healthcare provider talk about whether to remove the ovaries before the menopause to reduce the risk of ovarian cancer. For women at increased risk, lifetime risk of ovarian cancer varies from 4 in 100 (4%) to more than 40 in 100 (40%) in some cases: because of this wide range in individual risk it is important to check your own risk with your clinician.

| Frequently asked questions | Surgery to remove the ovaries and fallopian tubes | No surgery |
|--|---|--|
| Will this reduce my risk of ovarian cancer? | Yes, removing the ovaries and fallopian tubes will reduce your lifetime risk of ovarian-type cancer to that of the general population, which is about 2 in 100 (2%). | No, your risk of ovarian cancer will remain increased. |
| Is there a routine screening programme to detect ovarian cancer? | No, there is no evidence that screening for ovarian cancer is effective and after surgery your risk is sufficiently low that, even if screening was available, it would not be necessary. | No, there is no evidence that screening for ovarian cancer is effective in saving lives. |
| Will I be able to become pregnant? | No, therefore it is important that you are sure you have completed your family before having surgery. | Yes, your ability to become pregnant will be unaffected. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. Most women report feeling satisfied with their decision after surgery. | You may worry about developing ovarian cancer. |
| Will surgery change how I feel about myself as a woman? | Many women do not notice any change in how they feel about themselves as a woman. However, your desire for and/or enjoyment of sex is likely to decrease. | Not applicable. |
| Will I go into menopause? | Yes, after surgery your oestrogen levels fall and the menopause will start immediately. | Yes, most women will go through a natural menopause around 50. |
| Are there any long-term health risks linked to this surgery? | Yes, if surgery is done before age 45 there is a higher risk of bone thinning and cardiovascular disease. Some patients report memory changes as well. | Not applicable. |
| Will I need hormone replacement therapy (HRT) and is that safe? | HRT can reduce some of the effects of surgery. HRT is safe to take for the majority of women until they are 50. Discuss this with your clinician. It is not usually recommended for women who have had breast cancer. | No, if you go through a natural menopause you would not be expected to need HRT, unless your symptoms are very severe. |
| What are the risks of surgery? | About 4 in every 100 patients (4%) experience a complication during or after surgery. These can be minor infections or major complications. Discuss this with your clinician. | Not applicable. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after keyhole surgery and are back to normal in 4 weeks. For open surgery, time in hospital and recovery will be longer. During recovery you may feel pain / tiredness and may need help with everyday tasks. | Not applicable. |

For more information about this decision and your options, speak to your clinician and visit: www.OvDex.co.uk

Editors: Jan a Witt, Kate Brain, Elissa Ozanne, Kate Lifford, Fiona Wood, Mark Rogers, Glyn Elwyn.

More information: <http://www.optiongrid.org/about.php>

Evidence document: http://www.optiongrid.org/resources/oophorectomyBefore_evidence.pdf

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8.3.3 *OvDex usage, accessibility, functionality and navigation*

8.3.3i Usage: Four participants requested printed versions of OvDex, six used the website and two accessed the website, but then chose to view a PDF version. Respondents reported having spent between 20 minutes and 2 hours reading through the materials, with the majority taking between 30 minutes to 1 hour. Most viewed the OvDex website/booklet alone, but one respondent shared it with her mother and another used it together with her husband.

R11: I then got my husband to sit down with me and [...] just go through this with me and I bounced questions off him. And that's how I went through it. (Over50, yesOC, noFH, unknown)

This respondent later also noted that the material was useful, not only to her, but also to her husband, indicating that shared use with important others may be beneficial.

R11: I hadn't shared anything with him prior to my having my op. He didn't know what I was having done and I think had this been around at that time it certainly would have made things clearer for both of us. (Over50, yesOC, noFH, unknown)

8.3.3ii Accessibility: Most respondents who chose to access the OvDex website (n=8) reported no problems with access. Only two women could not get access initially; however both were able to resolve the problem later.

R12: The only problem I encountered, and this is potentially my own fault, is I couldn't actually get onto the web link originally [...]. I typed it in and it didn't work, I had to actually cut and paste it. (Under35, noOC, OCFH, unknown)

This respondent was accessing the website via an iPad and reported no further problems once the website was loaded. These issues with access may have been due to the complex path (<http://medic.cf.ac.uk/OvDex/index.html>) at which the online tool was available during usability testing. Ultimately, the online tool will be

accessible at a direct web-address (www.OvDex.co.uk), which should help to avoid such issues.

8.3.3iii Functionality: The OvDex website has two major interactive functions: the personalisation of website content and the value clarification exercise. As the majority of lay reviewers in this study were not at increased risk of ovarian cancer, most chose not to use the personalise function on the website and viewed the general version of the tool. However, they acknowledged that tailoring the information is a useful function for the target population of the intervention.

R12: If I was one of the people who had been told that I was at high risk [...] I would absolutely use [...] the one that's restricted to you, because it's giving you the information that's particularly relevant to you. (Under35, noOC, OCFH, unknown)

Some users, especially those who carried a confirmed BRCA mutation, did attempt to personalise the site. Most of those who used the personalise function found the interactive question format easy to use and valued the tailored information it resulted in.

R1: I think it was really good, the fact that you can personalise it from my point of view was really interesting. (35to39, noOC, B&OCFH, BRCA2)

However, one user reported problems with this functionality and could not access the personalised site.

R6: I actually tried to use the online thing because I thought: 'Well I'll personalise it to myself obviously, because I've got a BRCA 1 mutation.' [...] And it didn't seem to want to work, so it [...] just came back to the original question. (40to49, yesOC, BCFH, BRCA1)

Furthermore, some users reported not seeing the option of personalising the site, and others felt that the personalisation option should be more prominent.

R5: The thing that I thought was a bit odd at first is the 'it can be personalised'. I would have thought you would want it to be

personalised really. [...] I don't know [under] what circumstances you wouldn't want to personalise it. (Over50, yesOC, noFH, unknown)

R9: I would definitely try and sort it round so that you personalise it and then, if after you've read your personalised information you would like to just look at the general version you can do so, but try and get people not to waste time on the general version. (Over50, yesOC, noFH, unknown)

The second interactive function of the website is the value clarification exercise, which requires users to move sliders to weigh the importance of various factors. Most users found this exercise easy to use and had no problems with functionality.

R8: It was easy to move [the sliders] either way [...]. I don't think there's anything that I would say to improve on. It [...] seemed easy to access and you could [...] move it back and forward and make your decision. (35to39, noOC, OCFH, unknown)

The respondent who had used an iPad to view the site reported that the function differed from that of a site viewed via a PC or laptop.

R12: It took me a couple of goes to figure out how to move the little circles around [...] that's because I was on an iPad rather than a laptop. [...] I was expecting to be able to drag it across and I couldn't [...] You just had to click on the circle [...] and then put your finger just on the line where you want the circle to go and it jumps there (Under35, noOC, OCFH, unknown)

One user also commented that she occasionally moved the sliders in the wrong direction. Although the functions worked correctly, this may indicate a layout issue.

R1: I did, a couple of times, get confused which way I was going with it. [...] You're going one way and then you do something and then you think: 'Oh no, I didn't mean it [like] that!' and you have to go back and change it. (35to39, noOC, B&OCFH, BRCA2)

Respondents' comments about the content, rather than functionality, of the value clarification exercise are outlined in section 8.3.4 below.

8.3.3iv Navigation: Initial navigation to the index page of the website was found to be a problem for some women. One respondent described her confusion as she entered the website and viewed a page that assumed she had already seen the personalisation questions.

R5: Right at the beginning it confused me, once I got into it, it didn't at all, but the very beginning confused me. [...] The way it comes in at the moment it just says: 'Welcome to OvDex, please note it can be personalised you should have seen and answered these questions before viewing these pages.' [...] You think 'Oh hang on what questions then?'. (Over50, yesOC, noFH, unknown)

This respondent had opened the website on a different page to the index page and therefore had not seen the personalisation questions. However, this may again have been due to the complex path (<http://medic.cf.ac.uk/OvDex/index.html>) at which the online tool was available during usability testing.

When navigating the main part of the OvDex website, i.e. either the general version or a personalised version, or when reading through the OvDex booklet, most respondents read content in a sequential manner following the order suggested by the layout.

R9: I just went through all the sections [...]: the choice, the options format and then down through the risk reducing surgery and so on. I just worked my way through it. (Over50, yesOC, noFH, unknown)

Some respondents followed links to specific questions or sections within the website, instead of reading it sequentially. These women felt that the website allowed them to pick and choose the information they accessed.

R12: I like that because you can do it both ways can't you? [...] you're going to come across the information anyway, but equally if that's all you want to know about, then you can just go straight to it. (Under35, noOC, OCFH, unknown)

R4: What is good about it is, you can take from it what you want. So you can click onto things that are perhaps key in your mind at the time and

find out information to answer [...] [what] you're fretting about and come back to it again, when you're ready to look at some more. (Over50, yesOC, noFH, unknown)

These respondents also felt that jumping between sections was facilitated by the website's layout and links within the site.

R4: Within every section you have 'Go back to top' or 'Go to this' and [...] I just used the back button as well. [...] If I clicked onto different tabs on the left it would still let me go back to [...] an earlier one. It was just fantastic. It is fantastic the way it works. (Over50, yesOC, noFH, unknown)

8.3.4 *OvDex layout and content*

8.3.4i Layout and structure: Respondents felt that the possibility to choose between browsing the website and viewing, or printing, a PDF was advantageous and appreciated the possibility of choosing between the two.

R5: There's an awful lot of information to take in so I think the fact you have it as a PDF so people can download it and read it at leisure is a good idea. (Over50, yesOC, noFH, unknown)

One respondent also commented positively about the colour scheme.

R5: I know that's only a little small thing but it links in quite nicely, because lots of the things to do with [...] ovarian cancer and stuff obviously are that colour. And it kind of links it all in your mind. (Over50, yesOC, noFH, unknown)

The majority of participants were satisfied with the layout and structure of the OvDex website and booklet. In particular, women felt that the natural progression of the sections and the list of questions at the top of each section were helpful, as these gave them an overview of the section's content.

R12: It's a really great looking website as well. It's really nice looking, so it's really easy to surf through [...] Really easy to use. (Under35, noOC, OCFH, unknown)

R9: Underneath each chapter heading [...] it listed down the questions. And I liked that, because I could glance down through and think: 'Oh right, yep, that's what this is gonna cover.' And then you go through each question and I just liked the clarity of it all and the structure of it all. (Over50, yesOC, noFH, unknown)

R5: I thought [the layout was] quite good, because you've got the risk first, which is the major question, and then the choices that you've got and then you break it down [...] and it sort of defines it clearly. [...] So I think that, and the fact you've got other contacts and resources and stuff as well, yeah I think it's extremely well laid out. (Over50, yesOC, noFH, unknown)

8.3.4ii Information quantity: Although many respondents acknowledged that the tool included a lot of information, which initially could be seen as overwhelming, most felt that the quantity of information included in the intervention was appropriate and useful.

R4: I mean I could sit here and say: 'Well it's really wordy!', you know. I mean there's loads of information in it, but then you know: you want loads of information. (Over50, yesOC, noFH, unknown)

R7: I've got more information from this than what I've actually had from reading books that have been written by doctors [...] I've got so much information from this little leaflet to what I've had from books that I've sat and read for hours and hours on end. (40to49, noOC, B&OCFH, unknown)

Many noted that the information did not need to be read in one session and that users could come back to it when they wanted to learn more.

R1: There's certainly a lot of information, but I don't think that's a bad thing. And I think it's certainly something that you don't have to do in one hit. (35to39, noOC, B&OCFH, BRCA2)

R5: I thought it was very good, because it's all broken down into sections. So you don't have to read the whole thing through in one minute [...] You can keep going back to it, you can go back to sections [...], you can print it out, you can read it again and again and keep going back as

much as you like until you've got it clear in your mind. (Over50, yesOC, noFH, unknown)

R11: I thought the amount was quite a lot and [...] the first time I looked at it I thought: 'No I can't, there's too much here, I can't take it all in at once.' [...] [but] I thought it was quite a reliable source to go back to [...] I can pick and choose what I want. And there's a lot of it where I now think that makes sense and I'm looking for next sections and everything else. (Over50, yesOC, noFH, unknown)

Only one respondent strongly felt that there was too much information and that content should be condensed. This respondent may have preferred to view just an Option Grid rather than the more extensive website.

R13: It was helpful, but far too much. It could have been condensed and [...] probably been a lot clearer [...]. Just far too much reading. (Over50, yesOC, BCFH, unknown)

8.3.4iii Understanding and questions: When respondents were asked about the information content within OvDex, most noted that the wording was clear and easy to understand.

R7: You don't need to be a rocket scientist to read it. It's self-explanatory and even for somebody who doesn't have much common sense it would be very easy to follow. (40to49, noOC, B&OCFH, unknown)

R12: Really not patronising. It gave you the information that you required and it gave you more detail if you wanted it, so it was good. (Under35, noOC, OCFH, unknown)

Respondent 7 also added that OvDex was very easy to understand compared to other information materials she had viewed previously.

R7: I took a lot more in with this, because [...] it's so well written. I've read a lot of material since my mother has had ovarian cancer and they come out with big words that only doctors and nurses would possibly understand and [...] it sounds like gobbledegook, but this is just [...] so simple to understand. (40to49, noOC, B&OCFH, unknown)

Similar to feedback received about the Option Grids, some respondents gave detailed accounts of their cognitive processing of information and of questions that arose whilst reading OvDex. These highlighted areas where potential changes need to be made to facilitate understanding. For instance, one woman wondered why prevention of non-cancerous problems with the ovaries / fallopian tubes (an additional benefit of RRSO) might be advantageous.

R6: It says 'having the surgery will also prevent other non-cancer related problems happening with the ovaries such as cysts' and [...] maybe you should sort of expand on that a bit and maybe explain why it's bad to have cysts. (40to49, yesOC, BCFH, BRCA1)

Another respondent noted that when reading the section on alternatives to HRT, she wondered how and why the examples given in OvDex had been selected.

R9: There was one bit there, [...] 'the alternatives to HRT' [...] I didn't know whether the ones that were listed were the most commonly used [...] I didn't know why, in particular, you had listed those. (Over50, yesOC, noFH, unknown)

This respondent later also reported struggling with the use of the word 'alternative' in the question 'Are there any alternatives to surgery?', as this term had previously been used to describe medical and herbal remedies to HRT and was now being used in a different context.

R9: I think it's that term 'alternative'. [...] I was thinking of something very different to surgery, so I think maybe what [...] would be clearer for me would have been to say, 'Are there other treatments apart from surgery'? [...] Just on the previous page you're reading about alternatives to HRT and you're talking about herbal things and everything, so I've just gone off on that track of thinking and then I was distracted by the use of the word 'alternative' again. (Over50, yesOC, noFH, unknown)

Women also commented on the figures, diagrams, graphs and pictograms included in OvDex to facilitate understanding. One woman specifically felt that the picture in the cancer risk section showing women's anatomy was helpful.

R4: There's a fantastic diagram. I loved the diagram you've got of the physiology of a person. (Over50, yesOC, noFH, unknown)

Most respondents were positive about the pictograms showing cancer risk and risk of complications during surgery, and felt that these facilitated visualisation. Additionally, respondents appreciated that both positive and negative framing was used.

R9: I thought the little pictorial things [...] were very good. [...] That just really made it clearer to you what that sort of number was looking like, so I liked that. It was a very simple thing. (Over50, yesOC, noFH, unknown)

R11: I found them [the pictograms] really good actually [...] I particularly liked that it was balanced. [...] It is good to have that positive reaffirmation that just because you've been told you've got a predisposition doesn't mean you're going to get cancer. So I found that useful, I also just found it useful to break up all the text anyway, just give your eyes a bit of a rest. (Over50, yesOC, noFH, unknown)

R5: It reminded me of a theatre seating plan. It looks exactly like an online booking service doesn't it? [...] But no, I think that's really good because it's a good clear display [...]. The dots indicate the risk that you're talking about. (Over50, yesOC, noFH, unknown)

One respondent felt that the pictograms with dots may not be the most effective way of portraying risk, as she struggled to understand their meaning. She suggested using small female signs instead. However this respondent also noted that she struggles with interpreting frequencies correctly and therefore sought help from her husband, who understood and liked the pictograms.

R11: You know the little bits with all the circles on? [...] I would change those to little people. [...] I didn't understand the circles. My husband thought they were good, but I said well no I don't like those, because I don't understand what's good and what's bad. [...] It says 'The pictures below', but they're not pictures and they're sort of blobs. I like more sort of pictury things. (Over50, yesOC, noFH, unknown)

Two respondents suggested removing the pictograms completely, as they felt numerical information was sufficient. However, Respondent 6 later also acknowledged that some users may benefit from visual aids, such as these.

R6: What I wasn't really sure about was your pictures showing the chances of getting cancer [...]. You seem to do this quite a lot all the way through. [...] I suppose it's quite a nice indication, but it's taking up quite a lot of room. I don't know if it's totally necessary, because people can just as easily look at numbers. (40to49, yesOC, BCFH, BRCA1)

R3: I think actually reading [...] the writing besides [the pictograms] makes it clearer than the dots. I think [...] you could possibly do it without using the dots maybe. (Over50, yesOC, BCFH, BRCA2)

8.3.4iv Coping: The OvDex tool includes four sections concerned with providing coping advice and links to external sites with further information about coping. Three respondents provided feedback on these sections specifically. Respondent 10 commented on the coping advice within OvDex.

R10: I thought this was really good. [Reads from coping advice] 'How can I change how I feel about my cancer risk?' [...] Find someone to talk to: partners, your contacts or resources list. Yeah, I thought that was very good. (Over50, yesOC, BCFH, unknown)

R10: [Reads heading] 'How can I deal with the choice?' I thought this was really good [...] I thought that was excellent. [Reads from coping advice]: 'I will decide by myself using everything I have learnt. I will decide but will seriously consider my doctor's opinion. The doctor and I should decide together. The doctor should decide but seriously consider my opinion and the doctor should decide'. I thought they were really good choices. (Over50, yesOC, BCFH, unknown)

Although she found several parts of the coping advice useful, Respondent 10 also felt that some of the sections were repetitive and needed to be reviewed.

R10: The thing about feelings and anxiety [...] gets reproduced a lot in this book. But then again [...] you have to do that don't you? In a way. [...] I think it could possibly be a little less. (Over50, yesOC, BCFH, unknown)

Respondent 9 described her thoughts about the terminology used in the question introducing the second section on coping advice within the chapter about 'Choice' and made a suggestion for changing the wording of this question.

R9: I wasn't sure if that was quite the right wording. Because I wasn't sure what that meant [...]. I did wonder whether it [should be] something about: 'What will help me make the best decision for me? Or 'What will help me make this decision? [...] I wasn't sure about the word 'deal'. (Over50, yesOC, noFH, unknown)

Respondent 4 followed the links within OvDex's coping advice to external sites. She visited the Cancer Genetics Story Bank, watched the coping video and other content, and commented on the additional material (Iredale 2010, Phelps et al. 2006).

R4: There was a fantastic video regarding the waiting time. [...] It's very reassuring and useful tips on how to cope and ideas on how to relax etc I thought that was brilliant. [...] And I liked all the patient stories. [...] I thought the testimonies were really powerful and they made me feel less alone as a patient watching them. (Over50, yesOC, noFH, unknown)

Overall, few respondents provided feedback on the coping advice specifically. However, those who did felt that these sections, including external sites, were useful, although some parts may need to be reviewed to reduce repetition.

8.3.4v Value clarification exercise: Six women used the interactive online value clarification exercise. Four women had requested a printed booklet of OvDex and two women chose to review the pdf version online, therefore six women used the paper-based value clarification exercise. Many felt that weighing the options was a natural step in the decision making process and that this was facilitated by the exercise.

R5: I thought it was great fun and it's a nice interactive little tool. [...] We all do the list don't we of good and bad, for and against but to actually see it like that and to see the decision the effect it has what your

choices are I thought it was brilliant. (Over50, yesOC, noFH, unknown) [online version]

R9: I really like this section eight, 'Your Decision', because, I just thought that this was really helping you after all the information and everything and it was almost like testing you on what you'd read (Over50, yesOC, noFH, unknown) [paper version]

R12: I thought it was really useful, really interactive and I was at that point, at the end, [...]and I was almost sitting there thinking would I have to write a pros and cons list and then you come to that and you think: 'Oh, it's there!' (Under35, noOC, OCFH, unknown) [online version]

R6: Then I got onto chapter 8 which is 'Your decision' kind of questions and how much weighting you give the questions and everything, and I actually thought that was really helpful. (40to49, yesOC, BCFH, BRCA1) [paper version]

For those using the paper-based version, the example included gave clear instructions on how to complete the exercise and enabled all participants to fill in the written version without difficulty.

R9: [The example] made a lot of sense and actually, I think it's a good idea to have an example, because that just tunes you in again to the sort of things that you might be considering, so I thought that was good having the example and then having the blank [version] on there to have a go at yourself. (Over50, yesOC, noFH, unknown) [paper version]

The scales summarising a user's leaning at the end of the exercise were also perceived as helpful visual aids by most respondents.

R1: I thought that was really good. [...] I dare say it gives you confidence in what you're thinking. (35to39, noOC, B&OCFH, BRCA2) [online version]

R8: It was basic, but I think it's basic and then it covers everyone. Everyone can work with it, so I don't think you want anything complicated, because it's complicated decisions that people are making. (35to39, noOC, OCFH, unknown) [online version]

Following the value clarification exercise, the possibility of formulating an action plan and taking a printed or written version of the exercise and plan to the GP or consultant was an option users valued.

R4: I thought that was really clever and the 'My Plan' [...] that's really useful for taking and discussing things with your GP. I thought that was great. [...] I could have printed it off with my action plan and all those questions I might have wanted to ask. I think that's a brilliant section. (Over50, yesOC, noFH, unknown) [online version]

Overall, users of the interactive online exercise understood the instructions and could do the exercise without major problems. Users of the paper-based version reported that they understood the example and were able to complete the written exercise without difficulty. All respondents found the exercise useful.

8.3.4vi Contacts, resources and research evidence: Respondents appreciated the list of contacts and resources towards the end of the OvDex tool. They felt that it provided them with reassurance that further help could be obtained from other sources and that the intervention materials were not the only advice available.

R5: I think that that's really good, to say to people these are other places you can go. [...] You're saying: 'Well look if you want to have more you've got your GP, you've got your specialist'. You've got all these people and you're giving them lots of places they can go to get more information, help, whatever they need. [...] So I think that, at the end, is excellent. (Over50, yesOC, noFH, unknown)

R4: The other thing that you've done very well is [...] put down: 'For further information you can check out contacts and resources' and the list of all those other agencies [...]. In my situation it would have thrown up further issues and questions but I'd have then felt comfortable going to one of those other agencies to ask those things. (Over50, yesOC, noFH, unknown)

Women also valued the list of research evidence at the end of the decision aid. Although many had not gone on to look at any of the specialist publications, they

felt that the references provided reassurance that factual information had come from reliable sources.

R5: To actually say that this is where the evidence is from, this is where the information is for anybody who wants to go and look at that, that's a nice touch, because very often you don't get that. (Over50, yesOC, noFH, unknown)

R4: I thought the research evidence was really good [...] for further reading before deciding on what treatments you were going to have [...]. I really like looking at research myself. (Over50, yesOC, noFH, unknown)

R9: It gives it more credibility, doesn't it? When you see lots of references at the end and you see all the research evidence and everything. So, yeah, I thought that was well worth putting in. (Over50, yesOC, noFH, unknown)

8.3.4vii Improvements: In addition to discussing questions and issues with functionality, terminology and some of the content, which highlighted areas where amendments to OvDex may be required, respondents also made a number of direct suggestions for improvements. For instance, one respondent felt that there was insufficient information about BRCA testing in the general version.

R11: I wanted to know a little bit more about the BRCA test and whether or not it was available if I just walked in [...] And also probably another question about how quickly results would come back and about what that would involve. (Over50, yesOC, noFH, unknown)

As many respondents had had ovarian cancer and therefore had undergone oophorectomies, a number of women commented on the section describing recovery and recommended changes to this section based on their experiences.

R13: I wasn't too bad, but a lot of women can experience extreme tiredness [...]. They need to know all this, especially if they're looking after somebody or they need to work. (Over50, yesOC, BCFH, unknown)

Many women also felt that more emphasis should be put on the symptoms of ovarian cancer and on recommending that women who experience symptoms should seek professional help immediately. This may, in part, have been due to many of the women in this group having experienced ovarian cancer themselves.

R8: I just think the symptoms, that didn't really stand out on the website. Although everything was green and grey and black, everything was kept uniform and the same. I just think something like that needs to stand out a wee bit. (35to39, noOC, OCFH, unknown)

8.3.4viii Amendments to OvDex: Overall, participants' comments highlighted 39 areas where changes to the content of OvDex may be required. A full list of these, including the decisions of the supervisory team, can be found in Appendix 8.6. In total 31 changes were actioned and led to a review of functions, additions to the instructions included on various pages and alterations to information content (Table 8.3). The final version of OvDex is included in Appendix 8.7.

Eight potential changes were not implemented. The main reason for non-implementation was a lack of support for a given change from several other lay reviewers, who had commented positively about the same section. Other reasons included the supervisory team concurring that issues were already addressed satisfactorily or that additions were not sufficiently relevant to the target population of the OvDex tool. For instance, one respondent had suggested the addition of a new section on ovarian cancer, including pathology, treatment options and statistics; however this was rejected by the supervisory team, as it was not directly relevant to the target audience: women at increased risk, who have not been diagnosed with ovarian cancer. Additionally, the team was concerned about causing unnecessary anxiety among this group by including such information.

Table 8.3 - Changes made to OvDex in response to lay reviewer feedback

| OvDex section | Action / Change |
|--|---|
| Index page (Personalisation) | Index page was made default home page for URL www.OvDex.co.uk |
| Index page (Personalisation) | Personalise functionality was reviewed in available browsers |
| Index page (Personalisation) | Section on Index page was reworded to emphasise benefit of personalisation |
| Home pages | Added statement on introductory home page to clarify that intervention had been entered |
| Home pages | Added statement that announces that Value Clarification Exercise exists and added link |
| Overall (Information content) | OvDex tool was checked for repetitions and those were removed |
| Overall (Pictograms) | Changed dots to small female signs in pictograms |
| Cancer Risk (Q1)* | Added links to CRUK statistics and Contacts & Resources for information about ovarian cancer |
| Cancer Risk (Q3; General/Uninformative)* | Added information about possibility of private testing; Emphasised requirements for testing on the NHS |
| Cancer Risk (Q3; General/Uninformative)* | Added statement about gene test, timeline and possible outcomes. |
| Cancer Risk (Q6; General/Uninformative)* | Shortened answer and added graph showing risk curve |
| Cancer Risk (Q8)* | Added information that if a gene mutation is confirmed in the family, children can choose to get tested |
| The Choice (Q5)* | Added statement about why surgery is most effective at 40 years of age |
| Options at a Glance (Table) | Screening question was moved up to be the second question in the grid |
| Options at a Glance (Q1)* | Reworded question title |
| Cancer Risk (Q9) and Options at a Glance (Q1)* | Reworded answers to make them more distinct |
| Options at a Glance (Q1)* | Added link to Genetics Story Bank and Macmillan website to reference in text |
| Risk-reducing surgery (Q2)* | Added statement to advantages of surgery about ovarian cysts |
| Risk-reducing surgery (Q3)* | Wording changed to "...your desire for sex is likely to decrease" |
| Risk-reducing surgery (Q4)* | Added statement that peritoneum cannot be removed |
| Risk-reducing surgery (Q9)* | Added information about next steps / procedures if cancer is found during the surgery |
| Risk-reducing surgery (Q9)* | Added information about HRT if ovarian cancer is found |
| Risk-reducing surgery (Q10)* | Added statement about post-surgical effects during recovery |
| HRT (Q9)* | Added statement about how examples of alternatives were selected |

Table 8.3 cont. - Changes made to OvDex in response to lay reviewer feedback

| OvDex section | Action / Change |
|-----------------------------|---|
| Other Options (Q1&3)* | Reworded answers to emphasise that although screening might be bought privately, it is not effective at detecting cancer early and might provide false reassurance and to emphasise the difference between screening and diagnostic tests |
| Other Options (Q2)* | Reworded question title |
| Other Options (Q2)* | Added statement to emphasise similarity to IBS and encourage people to consult their GP if they notice anything unusual and link to symptom list |
| Other Options (Q2)* | Made symptoms box stand out more by adding colour and bold writing |
| Your Decision (Web version) | Added instructions for users of iPads |
| Your Decision (S5&6)* | Expanded statements in value clarification exercise to clarify their relevance to the decision |
| Your Decision (Q1) | Reworded question title and included information about what to do once decision is made |

*Questions(Q)/Statements(S) are numbered according to their order in original OvDex tool (General Version) viewed by lay reviewers. If changes only apply to a certain version this is indicated behind the question number.

8.3.5 Perceived helpfulness

The majority of respondents felt that the intervention would benefit women at increased risk of ovarian cancer making decisions about RRSO.

R6: I think it's really helpful, because it does give a lot of information and it really makes you sort of sit down and think what your priorities are as well. (40to49, yesOC, BCFH, BRCA1)

In particular, respondents remarked upon the emotions associated with learning about and living with an increased risk of ovarian cancer and how the content of the intervention might help women to appraise and understand their situation and regulate these emotions.

R10: I think it's very clear and if I was faced with what these poor women are faced with, it would certainly help. When you've found out something that's really possibly life changing [...], everything goes to pot really. Logic goes out the window, so you need an aid to help you. [...] I think they'll be glad to have it. (Over50, yesOC, BCFH, unknown)

R1: You get a lot of information and you can't always get your head round it and there's a lot of emotion involved as well [...], so [...] to have something [...] to take away, that is useful. (35to39, noOC, B&OCFH, BRCA2)

R12: If they wake up in the middle of the night and they're a bit worried or they think: 'Really I need to get to grips with this!', then they can refer to it. I think that that's really, really useful. (Under35, noOC, OCFH, unknown)

Respondents also felt that the ability to personalise OvDex and the opportunity to weigh different factors using the value clarification exercise were helpful. Women felt that the decision support intervention provided relevant information in a balanced, unbiased way and encouraged women to consider their values and preferences.

R5: I think this sets it out very clearly but doesn't [...] push in any direction. It gives the information for the person themselves to make their decision without any sort of pushing, which is critical I think, because at the end of the day they are the ones who have got to make the decision and live with it. (Over50, yesOC, noFH, unknown)

Respondents could imagine a number of ways in which patients with different preferences for information processing might use the intervention. Many stated that individuals with a monitoring coping style, who like to obtain as much information as possible, would benefit most from OvDex. Some thought that certain patients would not want to or need information at this level of detail, while others again might selectively read parts of the information that interested them.

R12: I think if you're the sort of person that is reflective [...] [and] wants to sit down somewhere quietly and read through the information and think about it, completely to yourself, I think it works absolutely perfectly for those kinds of people. (Under35, noOC, OCFH, unknown)

R9: I think it would be very, very helpful [...] obviously not for all, because [...] it's not everybody's cup of tea to read information and think about it themselves. [...] But I think for anybody who wants to research, read up and get the information themselves [...] this is a really clear simple

way of getting the personalised information that you need. [...] I think most people would find this extremely useful. (Over50, yesOC, noFH, unknown)

R11: I think some of them will completely dismiss it and say: 'No I don't need to read that.' and I think some of them [...] would look at it, and I think the other group would probably do it on a need-to-know basis. (Over50, yesOC, noFH, unknown)

Respondents also felt that partners, family members and friends might benefit from the information included in the intervention.

R9: I think this would be really good to be able to go back and discuss with family members and look through it with your partner or with your children. (Over50, yesOC, noFH, unknown)

Beyond its intended use as a decision support intervention for women at increased risk of ovarian cancer, women in this study felt that OvDex could also benefit those directly affected by ovarian cancer and their relatives.

R4: I think it's brilliant for what you've intended it for. I just think it could be used for a wider audience really. [...] Not just for [those] having [risk-reducing] ovarian surgery, but for [those] having ovarian cancer [...], having treatment for it, [those] who have come through it and for their carers. (Over50, yesOC, noFH, unknown)

R7: I would have liked something like this available when my mother had ovarian cancer, because there was nothing at the time. [...] If something like this was available at the beginning of when she found out she had cancer it would have been quite good for us to have, [...] not just for her, but also for the family as well (40to49, noOC, B&OCFH, unknown)

8.3.6 *Implementation of OvDex*

Respondents were asked what they thought would be the most appropriate way of introducing the decision support intervention to patients and at what point in time they felt the intervention would be most useful to patients. Most respondents felt that a health professional, particularly a genetic counsellor, would

be the most suitable professional to introduce the Option Grids and refer patients to the OvDex tool.

R1: I mean it might be something that's useful for either a doctor or a genetic counsellor to introduce. (35to39, noOC, B&OCFH, BRCA2)

R6: I suppose if they are going for genetic testing then normally they'll go and see a genetic counsellor, so maybe that would be the most appropriate person to put this in front of the patient. (40to49, yesOC, BCFH, BRCA1)

Some respondents also suggested that provision in GP surgeries might be appropriate. These were the respondents who had also felt that cancer patients, rather than women at increased risk, could benefit from the intervention and had recommended a widening of the target audience.

Most respondents agreed that early access to the intervention would be beneficial for patients, preferably at the point of finding out about an increased risk and being introduced to clinical management options and the possibility of choice.

R5: I would say the earlier the better for this. [...] Because the more information they've got from the beginning the clearer their thought processes are going to become. (Over50, yesOC, noFH, unknown)

R3: I suppose as soon as you know the problem that you've got and you're trying to weigh up in your mind [whether] to have anything done about it or not, then this would help you[...]. So I think yes probably early on, as soon as you know what the problem is. (Over50, yesOC, BCFH, BRCA2)

Respondent 7 felt that even earlier access would be beneficial; however, Respondent 13 opposed such early access before high risk had been confirmed.

R7: Something like this I would have liked to have read in the earlier stages when I was making the enquiries, when I started looking into it at first. (40to49, noOC, B&OCFH, unknown)

R13: If I was tested and my daughter was tested and she had this gene, then yes [she should be given the intervention], but if she hadn't had the gene, then I suppose [...] it can be a quite frightening thing to give to a young woman. (Over50, yesOC, BCFH, unknown)

One respondent felt that the Option Grid should be introduced at the end of a consultation and that the health professional could then decide on a case-by-case basis whether a patient would need instant discussion or whether it could simply be an aide-memoire to be taken home for further deliberation.

R12: I don't know that it needs somebody to go through with you because it's quite self-explanatory [...]. It would probably be better that it comes at the end of the conversation. [...] It's up to the doctor then, if they think that person looks like they're going to need talking through it [...], then they could take the time to talk them through it. (Under35, noOC, OCFH, unknown)

8.4 Discussion

8.4.1 Discussion of findings

The current study assessed usability of the patient decision support intervention developed in Chapter 7. Overall, lay reviewers liked the layout and structure of the Option Grids and felt that these were easy to understand and useful in providing answers to the most pressing questions. Many described how reading the information in the grid raised further questions that were later answered in the full OvDex tool. Reviewers reported few problems with accessibility and functionality of the OvDex website and were satisfied with the layout and structure of this more extensive part of the intervention. They found the information within OvDex easy to understand and especially valued the interactive features.

High satisfaction with layout and structure of OvDex indicates that the CODE framework is a good underlying framework for use in the structuring of extensive patient decision support interventions (Chapter 2; Witt et al. 2012). The four pre-

decisional deliberation phases distinguished in the framework lend themselves to informing the content of different sections within decision support interventions. Questions within the first phase outline appraisal of the health threat and informed the content of a background section about the medical problem at hand, i.e. increased ovarian cancer risk, and possible coping options. Most respondents in the current study felt that this section was useful in setting the scene and providing context. Questions within the second phase describe appraisal of choice and guided the content of a section explaining medical uncertainty and decision making in healthcare contexts. Respondents in this study liked the guidance provided within this section regarding decision making. Questions within the third phase, undoubtedly the most important with regards to decision making, outline appraisal of options and informed the content of several sections describing properties, benefits and risks of the available options. The current study demonstrated that women liked the unbiased and comprehensive information about options covered in this section, and felt that it would help patients make an informed decision. Finally, the fourth phase, preference construction, inspired a section designed to actively facilitate decision making. This section includes an interactive value clarification exercise. Respondents particularly appreciated the exercise in OvDex and felt that it was a natural activity that they might have intuitively done themselves in the form of a pros and cons list, if it had not been provided. These results indicate that the structure of OvDex, which corresponds to the phases in the CODE framework, was perceived as logical and easy to follow by participants, guiding them through the decision making process.

Furthermore, participants in the current study felt that the majority, or all, of the questions they had thought of were addressed within the content of the OvDex tool. This content had been guided by the questions included in the RRSO-adapted version of the CODE framework (Chapters 5 and 7). The framework does not aim to be comprehensive. However, as respondents had few additional

questions, this study shows that the framework seems to cover the most common and salient questions and coping options explored by women.

Despite the general satisfaction with the intervention materials, many reviewers highlighted one or more areas where improvements were necessary. The variety of comments received in the current study shows the range of individual preferences for structure, layout and types of information. For example, while most women felt the amount of information presented in OvDex was appropriate and helpful, one lady remarked that it was 'far too much' (see section 8.3.4ii). While some respondents were satisfied with the order of questions in the Option Grids, others preferred to have the screening question further towards the top of the grid (see section 8.3.2ii). While some stated that they liked the pictograms included in OvDex, others felt that numerical information would be sufficient (see section 8.3.4iii). This shows that, due to the range of individual preferences, creating an intervention that satisfies every potential user is not feasible. However, using feedback from lay reviewers, a number of improvements were made to the intervention developed in Chapter 7 to increase usability and acceptability.

Amendments made to the Option Grid in response to reviewers' comments focused mainly on the item addressing availability of screening, which was reworded and moved towards the top of the grid. This finding was unsurprising, as many women struggle to understand why there is currently a lack of evidence for ovarian cancer screening. This is especially true for those who took part in the recently concluded screening trial and reported having gained a lot of reassurance from the screening (Lifford et al. 2013). In addition, many women in this study had heard of CA125 and trans-vaginal ultrasound tests, some even had had those procedures as diagnostic tests, and therefore associated such tests with the detection of ovarian cancer, not discerning between screening and diagnostic tests. The feedback received as part of this study led to amendments in the wording of the answers to the screening question and a change in the order of questions within the grid. Other alterations included additions of statements about women's

satisfaction post-RRSO and possible pain and tiredness experienced during recovery. Amendments to the content of OvDex made in response to reviewer's comments included putting more emphasis on the personalisation function, conversion of dots to female signs in all pictograms and expansion and rewording of a number of questions for clarification purposes. Again, the variety of the issues highlighted shows the differences in individual preferences for structure, layout and information presentation. A number of minor functionality problems within the OvDex website were also fixed following lay reviewer's feedback. Hence, usability testing was able to enhance overall acceptability and usability of the patient decision support intervention (Durand et al. 2012).

Other recommendations by lay reviewers were not acted upon. As noted earlier, preferences for delivery, and amount, of information differ between different individuals, depending on various factors. These include prior knowledge, health literacy and preferred information processing style. Satisfying every potential user is not a feasible aim of support materials. Rather, decisions about what information to include, and what to omit, should be made based on careful consideration of the needs of the majority of persons among the target audience and, ideally, a review by several experts. As the majority of respondents in this study had been directly affected by ovarian cancer, many of the comments regarding additions and changes to the content in the OvDex tool focused on information particularly relevant to cancer patients. These suggestions were carefully reviewed by the supervisory team in order to decide whether the changes requested would also be beneficial to the target audience, i.e. women at increased risk of ovarian cancer. Hence, some suggestions, for instance the addition of a section about ovarian cancer or expansion of the section explaining diagnostic CA125 blood tests, were not fully implemented. Instead of adding further information about ovarian cancer, such as survival statistics, additional links to external sites where such information could be found were added. This was deemed sufficient by the team, because information about ovarian cancer statistics

may not be directly relevant to the intended target population. Additionally, such information may cause unnecessary worry and anxiety among women at increased risk, which should be avoided. Further information about diagnostic CA125 blood tests was not included in the intervention, as the supervisory team felt that this might add to the confusion between screening and diagnostic testing and would therefore be unhelpful for women at increased risk.

8.4.2 Implementation

Currently, available decision support for the decision of whether or not to undergo RRSO is limited (Chapter 6) and women making this decision report a number of unmet information and support needs (Chapters 3 and 4). By producing an intervention consisting of two complementary parts, the developers acknowledge that there is no 'one-size-fits-all' approach to decision support in this context. The feedback received in the current study indicates that women appreciate the individual components of the intervention and the possibility of choosing whether or not to view OvDex / certain components of OvDex according to individual preferences. Once available to health professionals and patients, the Option Grids and OvDex tool, in conjunction with genetics services, may contribute to better, more holistic support for women at increased genetic risk of ovarian cancer. The results presented here show that both parts of the decision support intervention were perceived as informative and helpful by a diverse group of lay reviewers. This indicates that they could, ultimately, support patients in reaching informed, value-adjusted decisions.

Women in this study additionally made recommendations about the future implementation of the intervention in clinical practice. In line with the recommendations for Option Grids, which are tools designed for use in consultations (Elwyn et al. 2013b, Marrin et al. 2013), lay reviewers recommended that health professional would be the most appropriate persons to introduce this tool to patients. Women who were familiar with the genetics services referral pathway felt that genetic counsellors, who may introduce the option of RRSO

initially, would be the most appropriate health professionals to introduce the Option Grid. Some women felt that the intervention should be available in GP surgeries. However, the information in the intervention was specifically designed for women at increased risk of ovarian cancer. Therefore, provision in GP surgeries may not be appropriate, as it may raise anxiety in members of the public for whom OvDex may not be relevant.

Most respondents agreed that access to the intervention should be granted early on in the decisional pathway, arguing that the information in the grid and OvDex tool would help women who had just found out they are at increased risk to explore their options.

Finally, several women felt that the information within the Option Grids and OvDex was useful not only to women at increased risk of ovarian cancer, but also those who had been diagnosed with the disease. This finding may imply that there is scope for a future adaptation of some of the content within OvDex to cater for the information needs of ovarian cancer patients and their relatives.

8.4.3 Strengths and limitations

Usability testing of interventions, particularly web-based interventions, is an important step during development to assess acceptability and usability of materials to users (Elwyn et al. 2011a, Hoffman et al. 2012). Incorporating usability testing as a final step, before field testing, evaluation and/or publication, ensures that many potential problems with accessibility, functionality and terminology are corrected at an early stage. Therefore, this study and the amendments in which it resulted have made an important contribution to increasing the intervention's acceptability and usability. Although subsequent studies may highlight more areas for improvements, a number of important changes were made to ensure that users can achieve the highest level of functionality, understanding and satisfaction possible at this point.

The current study included participants from a range of backgrounds and with a range of experiences of ovarian cancer. This population was familiar with terminology relevant to ovarian cancer and was eager to contribute to the study, which resulted in a wide range of comments and suggestions for improvements. However, due to the recruitment method, many respondents were women directly affected by ovarian cancer. While most did understand the hypothetical nature of the task (of reviewing the intervention 'as if' one was at increased risk of ovarian cancer) some initially misunderstood the target audience to be individuals directly affected by cancer. This indicates that the information material provided beforehand was not clear enough and should be reviewed. Careful consideration was given to these misinterpretations during data analysis in order to identify and, if appropriate, exclude any comments that exclusively referred to cancer patients' use of the intervention.

For this study a phone interview approach was selected, which allowed interviews to be conducted UK-wide, permitted women with ill health (e.g. those currently undergoing treatment for ovarian cancer) to take part and might have made respondents feel more relaxed, contributing to more honesty and open discussion of potentially sensitive subjects, such as sex life post-RRSO (Novick 2008, Opendakker 2006). As the interviews were conducted over the phone, important non-verbal cues may have been missed (Aquilino 1994, Groves 1990). Such cues can be helpful in the interpretation of data and can give vital information about emotions associated with certain statements. However, previous research suggests that the lack of such cues does not substantially hinder or compromise the effective interpretation of data (Novick 2008). Hence, the advantages of using phone interviews for this study outweighed the importance of non-verbal cues and it was therefore judged a suitable method.

This study shows that interviews using verbal probing are able to reveal cognitive processing issues and retrospectively explore users' navigation of the website. Some may argue that other qualitative methods, such as concurrent think-

aloud techniques, might be more appropriate to test web-based interventions (Davison et al. 1997). However, the current study shows that, despite being retrospective, users' accounts of how they used the intervention and of any issues they encountered were specific and detailed. Furthermore, this technique allows the interviewer to probe and question any statements and thereby obtain further detail, which is not possible when using think-aloud techniques. Previous research has also noted that retrospective accounts of usage reveal comparable numbers and types of problems to concurrent feedback and that having to provide simultaneous feedback can be detrimental to task performance (van den Haak et al. 2003). Therefore, retrospective exploration of women's use of the materials, as conducted in this study, may have been advantageous, as it gave women an opportunity to use the intervention, particularly the OvDex website, as intended (i.e. independently in their own home). Many participants in the current study also checked and referred to the intervention materials briefly whilst speaking over the phone, which further improved accuracy of their retrospective accounts.

Although the number of interviews conducted in this study was below the target of 15, it yielded a rich data set and was considered sufficient for a usability testing study (Kushniruk et al. 1997, Nielsen 1994). Some have noted that much larger numbers of cognitive interviews are required to uncover all potential issues (Conrad 2006); however, this was beyond the scope of the current project. Further issues may be uncovered in future evaluation studies and, as the intervention will be updated regularly, may be corrected at that time.

Ideally, usability should be tested with the target user group. Lay reviewers recruited to this study were not necessarily at increased risk of ovarian cancer and therefore were not in the target audience. This may have reduced the relevance of some of the comments and suggestions made. However, this approach was chosen due to previous issues with recruitment of the target population. Lay reviewers recruited through the charity Target Ovarian Cancer were deemed an appropriate alternative to the actual target audience, as this group would be familiar with

terminology specific to ovarian cancer. Furthermore, it has been suggested that lay audiences can make a valuable contribution to usability testing (Elling et al. 2012, Sawka et al. 2011). It is also important to note that three respondents, who had not had ovarian cancer or RRSO themselves, may, in fact, belong to the intended audience for the intervention, as two were carriers of mutations in BRCA2 (R1 and R2) and one had a family history of breast and ovarian cancer (R7).

Most participants in this study were over the age of 50, which may have resulted in higher than expected use of the paper-based version of OvDex, as older generations may be less familiar with the internet and/or comfortable using interventions online (Millward 2003, Sourbati 2009). However, as much of the information and coping content in the online and paper-based versions is identical, many comments applied to both versions. Furthermore, the current study achieved a balanced use of the online and paper-based versions of OvDex, with six patients using and commenting on each. Therefore, this issue may be seen as an advantage, as it resulted in feedback from several participants on both versions. This was particularly beneficial with regards to the value clarification exercises, which differ substantially, as it allowed assessment of the usability of each version with a number of participants.

Finally, Option Grids are designed to be used within clinical consultations to provide a scaffold for a better conversation about a given healthcare decision (Elwyn et al. 2013b, Marrin et al. 2013). By asking participants to review the Option Grid outside clinical consultations, the current study tested usability of the grid outside its intended context. While lay reviewers found the grid easy to use and understand when looking at it in their own home, the study did not provide information about how it might be used in clinic and during consultations. Therefore, it will now be necessary to conduct a pilot trial of the intervention, including a trial of the Option Grid being used in its intended context, in order to assess its acceptability and usability in clinic.

8.4.4 *Conclusions*

The usability testing study reported in this chapter shows that a group of lay reviewers positively appraised the decision support intervention developed in Chapter 7. The Option Grids were deemed useful and respondents felt they provided clear introductory materials for women newly identified as being at increased risk of ovarian cancer. The full OvDex tool was appreciated for its comprehensive content, tailored information output and value clarification exercise. Usability and understanding was further improved by various suggestions made by lay reviewers. Additionally, women were able to make a number of recommendations regarding implementation of the intervention in clinical practice.

This study showed that a decision support intervention structured according to the deliberation phases outlined in the CODE framework follows a logical flow, which users found easy to grasp and navigate. Information as well as coping components, guided by the questions within the RRSO-adapted CODE framework were appraised positively and most reviewers felt that the questions covered the majority, or all, of the issues they wanted to explore. Finally, this study demonstrates that the cognitive interviewing technique of verbal probing may be a feasible technique to use, not only in the validation of questionnaires, but also during usability testing of information materials. A pilot study is now required to test acceptability, usability and effectiveness of the intervention in clinical settings with the intended target audience.

9 General Discussion

9.1 Introduction

The studies in this thesis outline the development and operationalisation of a novel framework that integrates the processes of deliberations and coping during preference-sensitive medical decisions. In this chapter, the principal findings of the thesis are discussed and placed within the wider context of patient centred care (PCC) and shared decision making (SDM). Possible future applications of the generic CODE framework are explored and suggestions for further evaluation, and ultimately implementation, of the decision support intervention are provided. Finally, methodological strengths and limitations are discussed.

9.2 The need for holistic decision support

Previous assessment has shown that, compared to other countries, the UK has performed relatively poorly on measures of PCC, such as communication and engagement with patients (Coulter 2009b, Davis et al. 2010). For this reason, recent policy developments have pushed for more patient-centredness within the NHS (Department of Health 2011, National Health Service 2013, Secretary of State for Health 2010). An important element of such developments has been a call for SDM to become established as an integral part of clinical practice (van der Weijden et al. 2013). To achieve this, a number of requirements should be met, including an increase in the types of decision support interventions available to patients in various health contexts (Coulter et al. 2011).

While information leaflets undoubtedly have their part to play in supporting patient decision making, more complex patient decision support interventions might be required in many settings. Such interventions have the potential to achieve a number of important healthcare quality goals. These include improving health literacy, promoting the use of SDM and enhancing the overall care experience (Coulter and Ellins 2006). Interventions that allow tailoring of information to individual patients' circumstances may avoid overloading patients

with information and provide facts and figures that are more relevant to the patient, thereby helping them to understand their individual circumstances and options (Edwards and Elwyn 1999, Thistlethwaite et al. 2006). Interactive components are designed to keep patients engaged and can help in teasing out personal values and preferences that can later be discussed with a health professional (Coulter 2009a, Stacey et al. 2011).

While these are important and worthwhile goals for decision support interventions, as an additional dimension, such interventions could also support affective forecasting and coping efforts, thus providing more holistic decision support and PCC (Wilson and Gilbert 2003). This dimension could encompass the provision of information in such a way as to facilitate affective forecasting, and the provision of coping advice to enhance emotional well-being during and after decision making. As has previously been noted, emotions and coping efforts play an important role during deliberations (Balneaves and Long 1999, Power et al. 2011, Witt et al. 2012). However, advocates and developers of patient decision support have largely neglected the role of emotions and coping in decision making; and the concepts of decision making and coping have traditionally been treated as separate entities. The theoretical work presented in Chapter 2 of this thesis advances the field by merging these concepts and by suggesting a novel framework that describes deliberations in preference-sensitive healthcare contexts as a multi-step appraisal and coping process (Witt et al. 2012). This highlights the need for the integration of coping support within approaches to PCC and SDM, including the need to embed coping elements in patient decision support interventions.

The integration of coping advice into such tools will facilitate information processing and SDM. It will also empower patients to identify and use various coping options, enhance emotional well-being and, ultimately, enable them to make better quality decisions. A patient is not a strictly rational decision maker. Decisions are made within the context of, and influenced by, social environment and emotional states. Patients are often sick, vulnerable and dependent on the

health professional. Additionally, decision making environments are 'messy' and often emotionally charged (Olthuis et al. 2013). Coping advice given during consultations and within interventions may mitigate these factors in several ways. It can help patients to understand and regulate their emotional responses. It can help them identify, and make use of, their social support network. Finally, it can outline problem-focused courses of action to deal with the decision. To realise these benefits and to support the patient holistically, coping advice should be an integral part of PCC and SDM.

This thesis demonstrates that women at increased genetic risk of ovarian cancer, and who are making decisions about risk-reducing surgery, have a range of support needs, and that these needs are not fully met by currently available decision support (Chapter 6). Chapters 3 and 4 showed that such women's needs not only relate to medical information and care, but also include psychological support and effective communication. Women are keen to speak with others, who have either already made a decision or are in the same situation as them. They seek emotional support from partners, family and friends, as well as independent bodies, such as peer support groups and charitable organisations. They want help in finding reliable information, guidance on how to make a decision and, where relevant, strategies to deal with stress, anxiety and worry related to their cancer risk. Coping advice and signposting within interventions may cater to their needs by facilitating the use of various coping resources. Responses from women who reviewed the decision support intervention developed in this thesis showed that the coping advice included in the tool was well received (Chapter 8). This indicates that embedding coping components in decision support interventions is feasible and that these sections are valued by those using the tool. It also demonstrates that there is a need for more holistic decision support that may be guided by the CODE framework.

9.3 The future of the Coping in Deliberation (CODE) framework

The novel framework developed as part of this thesis (Chapter 2) describes appraisal and coping in the context of medical decision making, showing the transactional and interlinked nature of these processes (Witt et al. 2012). The CODE framework has the potential to act as the practical basis for the development of holistic decision support interventions, which aim to facilitate both coping and deliberation processes. Chapter 5 demonstrated that the CODE framework is readily adaptable to specific preference-sensitive healthcare decisions. Chapters 6 and 7 showed how an adapted version of the framework might be used to assess and/or develop patient decision support interventions.

9.3.1 Further validation of the CODE framework

Currently, the CODE framework is a theory-based model of deliberation and coping processes, that has undergone face validation with expert groups before being adapted to a specific preference-sensitive healthcare decision. Validity is generally defined as the extent to which a given instrument, such as a questionnaire, “measures what it is intended to measure” (p. 328, Lynn 1986). It has mainly been used in the context of survey design (McGartland Rubio et al. 2003, Streiner and Norman 2008). However, validation may also be important in the context of assessing the validity of constructs and items within theoretical frameworks. In this context, content validity may be defined as the extent to which items within a framework are relevant to, and representative of, the construct(s) of interest (Haynes et al. 1995). In this definition, ‘item’ refers to the descriptive content of a framework, e.g. statements, questions or examples, and ‘construct’ refers to the overarching domain(s) under which items are grouped within a framework. For example, content validity assessment could determine whether questions within the CODE framework are relevant to, and truly representative of, the constructs they are intended to illustrate.

The main *a priori* constructs of interest within the CODE framework are defined as the five deliberation phases (Health Threat, Choice, Options, Preference Construction, Consolidation) and two coping and appraisal phases (Primary and Secondary). Sub-constructs include the types of primary/secondary appraisal (i.e. cognitive and emotional or problem- and emotion-focused). Assessment by experts of whether the questions within the framework reflect those constructs could lead to refinement and strengthening of the framework as a valid model of deliberation and coping processes.

Validation of the CODE framework could be conducted using a sort task approach, as recently described by Cane et al. (2012). Ideally, expert participants should possess a good understanding of decision making theories and/or coping theories and should be unaware of the original framework developed by Witt et al. (2012). They may then be asked to perform a closed sort task, in which they are presented with an unpopulated framework that only shows the main constructs and sub-constructs. Participants could then be asked to sort the questions into the constructs, i.e. they can decide which deliberation phase a question represents, which appraisal phase a question represents and which type of appraisal (cognitive/problem-focused or emotional/emotion-focused) it reflects. Analysis of these responses would reveal how experts distribute the questions within the framework. A high level of overlap of sorting a certain question into a construct/sub-construct would indicate that this question is relevant to and representative of those constructs. In contrast, a low level of agreement among experts would indicate that a question may not be relevant to, or clearly representative of, a single construct/sub-construct and may apply to more than one. Therefore, results of such a task would provide information about the extent to which questions represent certain constructs and sub-constructs within the framework.

9.3.2 *Testing of the CODE framework*

The CODE framework could also be tested in clinical scenarios and/or real clinical settings to explore whether it genuinely acts as a model of patients' questions and deliberations about preference-sensitive healthcare choices (Norman 2004). For example, a testing study may look at consultations addressing the questions outlined within the framework and assess whether they lead to higher satisfaction with consultations and improved awareness of coping resources. It may be hypothesised that the more questions are addressed during a consultation, the higher satisfaction will be and vice versa. Results confirming this hypothesis would indicate that the framework is a good model of deliberation processes that encompasses the majority of questions patients may have. Testing of the generic CODE framework may be carried out using a variety of simulated or real world healthcare decisions to assess the extent to which the framework applies to differing healthcare contexts.

Such a study may be designed as a simulation study in which volunteers attend a hypothetical consultation with a clinician. The hypothetical consultation may follow a predefined guide in which certain questions or groups of questions outlined in the CODE framework are not discussed. It may be possible to develop a number of scenarios that cover different sets of questions. Following the consultation, volunteers may be asked to rate their satisfaction with the consultation and/or to complete a questionnaire to assess their awareness of different coping options. Analysis of data collected from a number of consultations may then provide information about whether consultations that address all, or most, of the questions within the framework lead to higher satisfaction and better awareness of coping options. Additionally, volunteers may be asked to suggest questions they feel had not been answered during the consultation. This could explore whether these questions map onto the questions that had been purposefully omitted from the consultation or whether volunteers have further questions, not yet included in the framework.

Alternatively, testing could be conducted in real world settings through observations and assessment of real clinical consultations. Similarly to the approach suggested for the simulation study above, following the consultation, patients may be asked to rate their satisfaction with the consultation and/or to complete a questionnaire to assess their awareness of different coping options. Such an approach to testing the framework would highlight (i) which questions in the framework are commonly discussed / omitted during clinical consultations and (ii) whether patients are more satisfied and more aware of coping options following consultations that covered most, or all, of the questions within the framework. Although this approach would undoubtedly require more time and runs the risk that some possible scenarios would not be explored, it may produce more valid results than a simulation study, as it involves actual (rather than simulated) patients, consultations and healthcare decisions.

9.3.3 Operationalisation of the CODE framework

Chapter 5 described the adaptation of the generic CODE framework to a specific preference-sensitive healthcare decision. Adaptation allows the content of the framework to become more focused and relevant to a specific healthcare context. This is intended to result in a more accurate depiction of cognitive and emotional appraisal processes. This is a useful exercise when planning to use the framework to highlight the information and support needs of patients facing a specific decision. The same approach could be used to describe the support needs of particular sub-populations, such as elderly patients with a specific disease. For example, a study that is currently being conducted at the Universities of Sheffield, Sheffield Hallam and Cardiff specifically explores the information and support needs of older women with breast cancer (Brain et al. 2013, Collins 2012, Reed and Wyld 2012). Data collected during this study may be used to adapt the CODE framework to specifically describe older women's decision making about breast cancer treatment options.

Ultimately, the generic version of the CODE framework may be adapted to a whole range of preference-sensitive healthcare decisions, including decisions outside the cancer context. Examples of non-cancer related preference-sensitive healthcare decisions that the framework may be adapted for include treatment options for coronary artery disease (CAD), treatment options for osteoarthritis of the knee, genetic testing for familial conditions, such as Huntington's disease, and prenatal testing using amniocentesis. The approach to adaptation outlined in Chapter 5 may guide future adaptations of the generic framework.

Adapted versions of the framework could then be operationalised, either as assessment tools for already existing interventions (Chapter 6) or as a guide to structure and populate novel interventions (Chapter 7), as suggested in this thesis. The study exploring older women's decision making about breast cancer treatment (see above; Brain et al. 2013, Collins 2012, Reed and Wyld 2012), for instance, ultimately aims to create a decision support intervention for older women who are making breast cancer treatment decisions. An adapted version of the CODE framework, that specifically describes older women's decision making and coping processes in this context, could be useful to guide the development of such a tool and to ensure that its content caters to the needs of this sub-population.

There are numerous decisions that could potentially be depicted using an adapted version of the CODE framework and that may benefit from decision support developed using these adapted versions. However, as this thesis is the first to suggest how the framework might be adapted and operationalised, there is also scope for further studies investigating other approaches to adaptation and operationalisation and potential additional applications of the framework. For instance, it may be possible to use a quantitative approach to framework adaptation, rather than the qualitative approach employed in this thesis. It may also be feasible to operationalise the CODE framework as an assessment tool for clinical consultations, rather than just written decision support materials. Furthermore, the framework could not only be used as a guide for the development

of decision support materials and interventions, but also act as a model of patients' deliberation and coping pathways that enables health professionals to better understand these processes in a given healthcare context and to tailor the content of their consultations accordingly, thus enhancing PCC. Future studies may propose a number of changes and amendments to the framework and may suggest a variety of other applications in the healthcare field or elsewhere.

9.4 The future of the Oophorectomy Option Grids and OvDex

9.4.1 Feasibility and evaluation

Before the development of the Option Grids and OvDex, there were no decision support materials available specifically tailored to the needs of women at increased risk of ovarian cancer who are making decisions about RRSO in the UK (Chapter 6). By developing this decision support intervention, the thesis makes an important contribution to decision support in the context of ovarian cancer risk in this country. It proposes a novel, theory-based, holistic intervention that may support women and their clinicians whilst making decisions about ovarian cancer risk management. Chapters 3, 4 and 6 outlined the need for a tailored decision support intervention due to the complexity of the ovarian cancer risk management decision, the range of barriers and facilitators that might influence women's deliberations about RRSO, the variety of support needs and the lack of tailored, theory-based support materials aimed at this population.

The decision support intervention developed in this thesis was the first tool to be based on an adapted version of the CODE framework (Chapter 7) and to be delivered in two parts: an Option Grid and associated online tool. The intervention received positive feedback in an initial usability testing study with lay reviewers (Chapter 8). However, studies investigating the feasibility and effectiveness of this intervention should now be conducted (Craig et al. 2008). This could not only show whether the intervention improves outcomes relating to decision making and

coping, but would also further validate the CODE framework as an appropriate basis for the development of decision support materials.

Many interventions are evaluated in small pilots or larger trials before implementation (see section 6.3.6, Chapter 6). Evaluation of novel interventions in such trials can establish feasibility and effectiveness, which can, ultimately, aid implementation. A study recently published by Schackmann et al. (2013) reported on the outcomes of a feasibility study testing an online decision support intervention previously developed by Kurian et al. (2012; Chapter 6). This study used the Systems Usability Scale and the Center for Healthcare Evaluation Provider Satisfaction Questionnaire to assess patients' and health professionals' attitudes towards the intervention (Brooke 1996, Ely and Plomp 1996). It showed that all stakeholders felt that the tool was easy to use and could support deliberations and SDM in practice. The Kurian tool was developed for carriers of mutations in BRCA1/2 in the US and mainly focused on breast cancer management options. Therefore, its target audience differed from the intended audience for the intervention developed in this thesis. Nevertheless, this recent study indicates that decision support interventions for women at increased risk of ovarian cancer, particularly those carrying mutations in BRCA1/2, are likely to be well received by stakeholder groups.

Field testing of the decision support intervention developed in this thesis with women at increased risk of ovarian cancer, and health professionals involved in their care, would now be beneficial to assess feasibility. This could establish whether the Option Grid and OvDex website are acceptable to stakeholders and fit into clinical workflows (Craig et al. 2008). A small pilot trial to estimate rates of recruitment and retention and to test intervention fidelity and possible measurement issues in larger trials may also be advantageous (Craig et al. 2008). However, many of the commonly used outcome measures for SDM interventions, such as knowledge, generic psychological constructs, decision effectiveness/quality and satisfaction, do not truly capture the range of outcomes that may be affected

by interventions used in clinical genetics (McAllister et al. 2011a). Recently McAllister et al. (2011b) proposed the Genetic Counselling Outcome Scale (GCOS-24; Appendix 9.1), which is a 24 item patient reported outcome measure (PROM) that focuses on the construct of empowerment and covers five dimensions (Cognitive control, Decisional control, Behavioural control, Hope and Emotional regulation) (McAllister et al. 2011a). The development of empowerment as a PROM was guided by patients and clinicians and is therefore likely to cover the range of outcomes targeted by genetic counselling interventions. Hence, use of such a measure may be useful in evaluating the impact of the decision support intervention proposed in this thesis, as this intervention was designed as a complementary tool for genetic counselling.

The GCOS captures many of the intended outcomes of the intervention, as stated in Chapter 7 (McAllister et al. 2011b). It allows assessment of perceived knowledge of the health threat, awareness of management options and risk perceptions by including items on cognitive control (e.g. 'I can explain what the condition means to people in my family who may need to know'; 'I don't know what could be gained from each of the options available to me'). Aspects of coping and emotional appraisals are assessed in the dimensions of 'Behavioural control' (e.g. 'I know how to get the non-medical help I / my family needs'), 'Hope' (e.g. 'I feel positive about the future') and 'Emotional regulation' (e.g. 'Having this condition in my family makes me feel anxious'). Reduction of decisional conflict and ability to make decisions is covered by items in the 'Decisional control' dimension (e.g. 'I can make decisions about the condition that may change my child(ren)'s future / the future of any child(ren) I may have').

However, some of these items may require adaptation for the specific context of increased risk of ovarian cancer. The focus on inheritance and children in the GCOS, for example, is less relevant in the ovarian cancer risk context than in many other genetic conditions; therefore, item 24 may be in need of rewording. Alternatively, the original GCOS-24 may be used in combination with another scale,

such as the DelibeRATE scale (Appendix 9.2), as used previously by Sivell et al. (2012a). This would additionally measure affective forecasting (e.g. 'I can imagine what it would feel like to live with each option') and the deliberation process about RRSO (e.g. 'I feel I've given the options available to me enough thought'). Furthermore, although a number of items relevant to appraisal and coping are included in the GCOS-24, use of a measure that specifically assesses impact on appraisal and coping processes may also be advantageous. For example, the brief COPE may be an appropriate measure to use alongside GCOS-24 in a field test and/or small pilot of the intervention (Brain et al. 2008, Carver 1997).

In addition to assessing the decision support intervention's impact on patients, field testing should include exploration of health professionals' attitudes. The Healthcare Evaluation Provider Satisfaction Questionnaire (Appendix 9.3), as recently used by Schackmann et al. (2013) in a similar study, may be an appropriate measure to use in this context. This could provide information about professionals' views of the content (e.g. 'How useful was the information provided?'), perceptions of the usefulness of the intervention (e.g. 'This tool could improve patient-doctor encounters') and intentions to use and recommend the tool (e.g. 'I would use it regularly in practice'). Additionally, basic information about consultation length, with and without Option Grids, may also be gathered during field testing.

Quantitative data obtained by the means discussed above may be supplemented by qualitative data from a sub-sample of participants to explore in depth any issues highlighted by the data. This could establish how the Option Grids affect consultations and fit into clinical workflows and how the OvDex tool is used by patients independently (Craig et al. 2008). Results from the field test might then lead to changes to the decision support intervention.

A similar approach, using the GCOS-24, DelibeRATE and/or brief COPE with patients and the Healthcare Evaluation Provider Satisfaction Questionnaire with health professionals, may subsequently also be used during a small pilot trial of the intervention. Such a pilot trial may use a before and after design or an intervention

versus control group design. The most appropriate study design will depend on expected recruitment figures. Such a trial could assess the impact of the intervention as a whole and could additionally explore the effects of individual components of the intervention (e.g. Option Grids, value clarification exercise, coping advice). The pilot trial should also collect data on consultation length with/without the Option Grid and on the use of the OvDex website by patients in the study, e.g. number of visits, lengths of visits, sites viewed. Pilot trial results may then help to estimate rates of recruitment and retention and to calculate sample sizes for further, larger trials (Craig et al. 2008).

Ultimately, a randomised controlled trial (RCT) may be conducted to confirm the cause-effect relationship between using the decision support intervention in conjunction with genetic counselling and any significant changes in outcome measures observed. However, the funding, time and number of participants required for such a large scale trial may not be feasible. For instance, the current project faced a number of recruitment difficulties (Chapter 4), indicating that the target population may not be large enough to recruit the numbers needed for an RCT. Additionally, it has now been widely accepted that patient decision support interventions have positive effects on knowledge, decrease decisional conflict and enhance value-adjustment of decisions (Stacey et al. 2011). Therefore, a small pilot trial using the GCOS-24, and potentially other measures as discussed above, may be sufficient to drive implementation of the intervention, without the need for a large scale RCT.

9.4.2 Implementation

In the past, patient decision support interventions have faced substantial implementation issues, despite their endorsement by governments and other public health entities (Elwyn et al. 2013a, Elwyn et al. 2013c). This may in part be due to a disinterest in change and new technologies that Alvesson and Spicer (2012) termed 'functional stupidity'. Although this terminology may seem severe, it describes an organisation's and/or individual's inability or unwillingness to mobilise

cognitive capacities, such as questioning norms, seeking justifications and engaging in substantive reasoning (Alvesson and Sköldbberg 2009). Breaking through the routine flow of clinical practice and changing the way consultations are done can be difficult, even if it only involves use of a short tool, such as an Option Grid (Elwyn et al. 2013b).

To encourage uptake and integration into routine workflows, new materials and interventions should ideally have a champion within the organisation itself in order to overcome the first hurdle (Coulter et al. 2011). A clinical champion approach may be useful for implementing the intervention proposed in this thesis, as women felt that a health professional, ideally a genetic counsellor, would be the most appropriate person to introduce the first part of the intervention, i.e. the Option Grids, to patients (Chapter 8). Hence, promotion among colleagues through a clinical champion might increase the number of patients who are given the grids during consultations. As the grids bear the web-address of the full online tool, this in turn will also promote the OvDex tool. Ideally, the champion should be someone with a high profile, who is willing to support and push for implementation within the professional community and motivate teams to use the intervention (Elwyn and Thomson 2013). Throughout the development of the intervention presented in this thesis, a number of health professionals were consulted, two of whom (Prof Usha Menon, Institute for Women's Health UCL, and Dr Mark T. Rogers, Cancer Genetics Service for Wales) may in future be recruited as champions to support implementation of the Option Grids and OvDex tool. Champions and other health professionals, however, often want evidence of effectiveness of an intervention before promoting its use in clinical practice. Therefore, it will be important to present the results of any trials, such as evidence of the effects of the intervention on consultation length and PROMs and health professionals' satisfaction, to these individuals and the wider professional community.

Another approach to implementation may be to create demand for the decision support intervention among the patients who are its potential users. This

bottom-up approach is commonly used by pharmaceutical companies advertising prescription drugs in the US and may be useful when exploring alternative avenues to implementation (Frosch et al. 2007, Hollon 1999). The 'Ask Three Questions' campaigns in the UK and Australia (Elwyn and Thomson 2013, Shepherd et al. 2011), for example, aimed to encourage patients to ask three key questions to prompt a discussion of options and to stimulate SDM in a bid to overcome the implementation problems facing SDM (Légaré et al. 2008, Légaré and Witteman 2013). Similarly, encouraging patients to ask for decision support materials during consultations might result in a shift of attitude of health professionals towards such tools and may eventually result in integration of more interventions into clinical workflows. However, at a time when healthcare resources are limited, and in the absence of funding for marketing campaigns that can make patients aware of the existence of these tools, the potential to implement this method is limited. Patient networks and charities may be informed of the availability of relevant tools and encourage patients to ask health professionals for them, but without larger scale funding a bottom-up approach may not be feasible.

Following initial introduction of a patient decision support intervention into clinical practice, the next phase could be described as 'normalisation', which involves use of the intervention becoming part of routine care (Elwyn and Thomson 2013, May and Finch 2009). Achieving normalisation is a long and complex process that is described by Normalisation Process Theory (NPT) (May and Finch 2009). Coherence (clarity about what the work is), cognitive participation (clarity about who does the work), collective action (agreement about how the work gets done) and reflexive monitoring (appraisal of the work) are the four constructs outlined in NPT that lead to embedding of an intervention into routine workflows (Elwyn et al. 2013a). Normalisation of use of the RRSO Option Grids in genetic counselling sessions and referral of patients to the OvDex tool may ultimately be achieved through a combination of the implementation strategies discussed above.

9.4.3 *Additional decision support*

The patient decision support intervention developed in this thesis is designed as an adjunct to support discussions with genetic counsellors and/or gynaecologists and to facilitate decision making and coping. Both parts of the intervention emphasise the option of, and need for, further discussion with health professionals. The Option Grids are designed for use in clinical consultations as a scaffold to stimulate discussions of options and preferences during clinic visits. Although OvDex is designed for independent use by patients, it too encourages users to consult relevant health professionals and discuss their personal values and preferences before reaching a final decision. This is essential to ensure that any additional questions are answered and that any misunderstandings or misinterpretations are rectified. Furthermore, individual differences in information processing needs and preferences for delivery of information, which have been highlighted at various points in this thesis (Chapters 4, 7 and 8), demonstrate the need for the availability of additional decision support, particularly support delivered by health professionals. Option Grids and OvDex cannot cater for every possible need of every possible patient and, as has been noted previously, there is no one-size-fits-all intervention. Therefore, concurrent support from health professionals is essential and this intervention should not be seen as an isolated tool for women to use. Rather it forms part of a care pathway for women at increased risk of ovarian cancer and complements information and support provided by health professionals. Hence women may seek additional decision support from professionals at any time and are encouraged to do so.

In addition to the help women may receive during consultations, there are a number of other decision support interventions that are, or may in the future become, available to women at increased risk of ovarian cancer in the UK. These include the website by Healthwise (2011b) and the online tool by Kurian et al. (2012), which has recently undergone further testing and was shown to be well received by patients and health professionals (Schackmann et al. 2013). Patients

may also access the booklet previously developed by Tiller et al. (2003). While these interventions are available online and target women at increased risk of ovarian cancer, there are significant drawbacks to patients accessing and using them in the UK. Importantly, none of these interventions was developed for the UK context, therefore information may not be relevant to, or even contradict, UK guidelines. Furthermore, the Healthwise intervention (2011b) is a commercial product that has not been evaluated and does not clearly state a theory base. The intervention by Kurian et al. (2012) was developed for use with a health professional and is specifically designed for women carrying BRCA mutations, therefore it does not provide relevant information for women who do not know their gene mutation status. Finally, the Australian booklet (New South Wales Centre for Genetics Education 2008, Tiller et al. 2003) was last updated in 2008 and therefore, additionally to the fact that it contains information in line with Australian guidelines, may be outdated. Hence, the intervention developed in this thesis, if updated regularly, will provide the most relevant and tailored information for women living with an increased risk of ovarian cancer in the UK, despite other materials being available. Therefore it would be preferable if patients were guided to this intervention and encouraged to seek additional support from health professionals, rather than from other available tools they may find and access online.

9.4.4 New evidence, changes in recommendations and update policy

RRSO remains the only clinically proven management option for women at increased risk, two large scale trials of ovarian cancer screening for the general population and high risk groups have recently been conducted in the UK. Data obtained from the UK Familial Ovarian Cancer Screening (UKFOCS) study is currently being analysed and may lead to changes in recommendations regarding CA125 testing and trans-vaginal ultrasound scans in high risk women (Rosenthal et al. 2013b). Depending on the results, such changes may include endorsement of screening in women who decline surgery and therefore lead to changes in how

screening is discussed with women at increased risk. This would then also have to be reflected in an updated version of the decision support intervention, which currently does not endorse screening in any context due to a lack of evidence.

However, it is unlikely that the UK National Screening Committee will issue a statement or amend the guidelines regarding ovarian cancer screening before the results of the UK Collaborative Trial of Ovarian Cancer Screening (UKCTOCS) are available. This trial screened women from the general population and collected evidence on mortality benefit of screening. This evidence will be crucial in determining whether a national screening programme should be introduced. If results of the UKFOCS study and UKCTOCS support the introduction of routine screening, then the guidelines will be reviewed and may be changed. However, this process could take years and also depends on cost benefit analyses and other non-medical influences. In light of these developments, regular checks of evidence regarding ovarian cancer screening and early detection should be carried out, and any changes to recommendations should result in updates of the information included in the decision support intervention to ensure that the content reflects the latest developments.

Other possible developments in the field may include publication of more accurate ovarian cancer risk estimates for women with mutations in BRCA, women from Lynch families or women with a strong family history. Additionally, gene variants that are currently categorised as being of “unknown significance” may in future be shown to be benign or associated with an increased risk of ovarian cancer, meaning that additional at-risk groups may be differentiated. Furthermore, as surgical techniques become safer and more sophisticated, data on the risks of complications and rates of laparoscopic versus open surgery may change. This should be reflected in the decision support intervention. Therefore, the Option Grids and the OvDex tool will be reviewed and updated on a yearly basis. Additionally an emergency update policy is in place regarding evidence for or against ovarian cancer screening, which will be monitored more frequently and

updated as new evidence becomes available. Finally, as the decision support intervention will be available freely online, users should be asked whether they would be willing to complete an online questionnaire at a later stage to report their personal outcomes. This could provide information about individual outcomes post-intervention that may be incorporated into updates of the website (Coulter 2009a).

9.5 Methodological strengths and weaknesses

The research methods used in this thesis were selected to provide in-depth information about topics of interest to the research and were therefore of a qualitative nature (Black 1994, Kvale 1996, Pope and Mays 1995). Three searches of the literature were conducted: one narrative and two systematic searches. The narrative search was used to identify decision making and coping theories (Chapter 2). Systematic searches were employed to select studies which assessed women's decision making (Chapter 3) and to identify decision support interventions that included the option of RRSO (Chapter 6). Systematic searches were also combined with supplementary searches to identify additional publications that the initial search may have missed. The strengths of these search strategies included coverage of a wide range of literature and identification of a variety of relevant publications, all of which were subsequently critically appraised to ensure they were of an appropriate quality and standard.

Qualitative research methods were used to examine women's and health professionals' views of deliberations about RRSO and the scope for decision support (Chapter 4), to gather feedback on prototypes of the decision support intervention (Chapter 7) and to assess usability of this intervention (Chapter 8). Focus groups were deemed most suitable to explore women's questions, concerns, emotions and decision support needs during decision making about RRSO (Bloor et al. 2000, Krueger 1994, Pope and Mays 1995). This is because they allow mutual stimulation to occur and enable a collective exercise to rank different factors, providing an overall indication of relative importance (Slaughter et al. 1999, Kitzinger 1994). This study may, however, be criticised for the small number of women recruited to the

focus groups. Women invited via three centres in Wales and England frequently declined to participate in the study, which may be due to a number of different factors. These may include distress caused by the study invitation and/or their cancer risk, dislike of the focus group approach or unwillingness to travel to a group. Unfortunately, no data for non-responders was collected. Therefore any explanation as to why these women declined to participate is speculative. However, it has been noted that recruitment to focus groups is difficult and previous studies have experienced similar recruitment issues when using this method (Bloor et al. 2000, Phelps et al. 2006). The rich data collected in the three focus groups did, however, provide a good indication of the salient issues that women explore while making decisions about RRSO. Furthermore, these data were complemented by the results of semi-structured interviews with health professionals as well as data from the literature review in order to inform the adaptation of the CODE framework and development of the decision support intervention.

The intervention in this thesis was developed using an iterative approach with regular input from a variety of stakeholders. This approach allowed the researcher to tailor information to the needs of patients and to ensure that statements were in line with clinical guidelines and recommendations discussed in consultations. Additionally, this method ensured that a number of health professionals and patients contributed throughout the development process, potentially resulting in a feeling of ownership. Such feelings among stakeholder groups (particularly health professionals) may lead to higher uptake, as it has been suggested that feelings of ownership positively affect attitudes towards materials, products and guidelines, and may aid implementation (Orem et al. 2012, Reb and Connolly 2007).

Finally, a usability study assessed acceptability and usability of the intervention among a group of lay reviewers. Again, qualitative methodology was selected for this part of the study. This allowed an in-depth analysis of how the intervention was used by individuals and of any problems they encountered, including issues with functions on the website and cognitive problems (Adams and

Cox 2008, Boyce and Neale 2006). Some may criticise this research for the lack of quantitative data, especially with regards to the usability testing phase. While a quantitative evaluation of the effects of the decision support intervention would have been advantageous, it was beyond the scope of the thesis. However, the qualitative data obtained during these studies helped to highlight, and subsequently rectify, various issues with the content of the Option Grids and OvDex tool and has resulted in a more user-friendly intervention that may benefit from further testing and evaluation.

9.6 Conclusions

The research presented in this thesis outlines the development and operationalisation of a novel framework that merges decision making and coping theories. It demonstrates that integration of these two processes is a worthwhile exercise, as it results in a more holistic model of patients' appraisal and coping processes. The research further shows that the structure and content of a decision support intervention based on an adapted version of the novel framework was easily understood and well received by lay reviewers. Emotional aspects of deliberations, and coping efforts initiated to deal with such aspects, are an integral part of decision making that should be addressed within decision support interventions. A more holistic approach to developing and providing decision support, based on an understanding that emotions and coping play important roles in deliberations, may ultimately improve patient-centred care and promote the use of SDM in clinical practice.

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Appendix 1

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Appendix 1.1 - Referral Guidelines for Individuals with a Family History of Cancer

Referral Guidelines for Individuals With a Family History of Cancer

Cancer Genetics Service for Wales



Contacts

| North Wales | South West / Mid Wales | South East Wales |
|--|--|--|
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We will accept referral where the following criteria are met

Breast Cancer

- 1 first degree relative diagnosed at 40 years or less
 - 2 first degree relatives or 1 first degree and 1 second degree relative on the same side of the family, diagnosed at 60 years or less
 - 3 first or second degree relatives on the same side of the family diagnosed at any age
 - 1 first degree male breast cancer
 - A first degree relative with bilateral breast cancer
- N.B. breast cancer can also be inherited through the **paternal** side of the family

Breast / Ovarian Cancer

- Minimum: 1 of each cancer in first and second degree relatives (If only one of each cancer, the breast cancer diagnosed under 50 years)
- A first degree relative who has **both** breast and ovarian cancer

Ovarian Cancer

- 2 or more ovarian cancers on the same side of the family, at least one of whom is a first degree relative

Colorectal Cancer

- 1 first degree relative diagnosed at age 45 or less
 - 2 first degree relatives (incl. both parents) or 1 first degree and 1 second degree relative on same side of the family, diagnosed at any age
 - 3 relatives, all on the same side of the family (at least 1 should be a first degree relative)
 - Familial Adenomatous Polyposis (FAP) or other polyposis syndromes
 - Hereditary non polyposis colorectal cancer
- N.B. Families with cases of colorectal and endometrial cancer may also be referred

Other Cancers

- A family history of a known single gene cancer syndrome (e.g. von Hippel-Lindau disease, multiple endocrine neoplasia, retinoblastoma)
- "Related Cancers": There are some rare cancer syndromes (e.g. Li Fraumeni syndrome and Cowden syndromes) where a variety of different cancers occur within a family.
- A family history of 2 or more cases of the same cancer in first or second degree relatives (e.g. gastric cancer, prostate cancer, renal cancer, pancreatic cancer, melanoma)
- In any family where there is suspicion of a hereditary aetiology, the possibility of referral should be discussed on an individual basis

These guidelines are the result of an All-Wales multidisciplinary consensus group meeting and feedback from GP focus groups. They are subject to ongoing validation.

The Cancer Genetics Service

5-10% of cancers of the breast, ovary and colon are due to an inherited predisposition. The **Cancer Genetic Service for Wales (CGSW)**, part of the All-Wales Genetics Service, has been established to provide a service for individuals who meet the criteria for referral. Our remit is to assess the risk of cancer based on the reported family history, to work with relevant specialist clinicians and recommend screening strategies where appropriate. The referral criteria suggest who may be at significantly increased risk of an inherited form of cancer.

Process: On receipt of a referral

- Questionnaire sent to patient, to identify family history and baseline information. (We only take the referral further if the questionnaire is returned.)
- Where necessary, family history clarified and confirmed.
- Risk assessment process to categorise patient as 'High', 'Moderate' or 'Average' risk.
- Patient, referring doctor and GP informed of risk category.
- All high-risk and some moderate-risk patients offered an appointment in Cancer Genetics clinic.
- All patients given contact details for Cancer Genetics Service and invited to recontact us in the event of a change in family history.

Average risk

We aim to reassure this group that, on the information given, their risk is not significantly raised above that of the general population, so extra surveillance is not suggested. They should continue standard health awareness and screening as for the general population.

Moderate risk

We aim to facilitate or coordinate ongoing management between primary care and appropriate specialist clinicians (e.g. local breast or colorectal surgeons) or screening organisations (e.g. Breast Test Wales). Patients may be referred directly by the Cancer Genetics Service or we will suggest an appropriate referral route for the GP. In some cases laboratory investigations may be recommended to help clarify a patient's risk status.

High risk

In addition to suggesting the involvement of other specialist clinicians and recommending appropriate surveillance, this group will be seen in the Cancer Genetics clinic and genetic testing may be offered in some families.

An appointment in the Cancer Genetics clinic offers

- The time to discuss concerns about a family history of cancer and implications for both the patient and other family members.
- Where appropriate:
 - Referral or recommendation for appropriate screening/surveillance.
 - Storage of DNA and organisation of genetic testing, where appropriate.
 - Opportunity to participate in research studies.

All Wales Queries:

Cancer Genetics Service for Wales,
Institute of Medical Genetics,
University Hospital of Wales,
Heath Park, Cardiff CF14 4XN
Tel: 029 2074 6620

Copies of these guidelines in Welsh are also available from this address.

Appendix 1.2 - Ovarian cancer risk management letter (UCL Hospital)

University College London Hospitals 
NHS Trust

THE
NORTH
LONDON
CANCER
NETWORK 

Familial Cancer Clinic
Department of Gynaecological Oncology
UCLH Gynaecological Cancer Centre
Elizabeth Garrett Anderson Hospital
Huntley Street
LONDON
WC1E 6DH

Telephone: 020 7380 9193
Fax: 020 7380 6929
Email: michelle.johnson@ucl.ac.uk

Clinic Date:
Hospital No:

Dear Ms

I was pleased to see you in the familial clinic and I am writing to summarise the outcome of the consultation. Your family history is as follows:

| Relative | Cancer | Age at diagnosis |
|----------|--------|------------------|
| | | |
| | | |
| | | |
| | | |
| | | |
| | | |
| | | |

You have Jewish ancestry. Regarding personal risk factors, currently you are not using the oral contraceptive pill. In the past, you have used it for a total of less than year(s). You have had ...children. You have fertility treatment.

The first subject that we discussed was the level of your chance of developing ovarian cancer based on your family history.

I explained that with regard to ovarian cancer, there are broadly 3 groups of women:

1. *Women at low risk:* Most women do not have a close relative with ovarian/tubal cancer and have a low risk of developing ovarian/tubal cancer themselves. The risk of developing ovarian cancer over a life time in this group is approximately 2% (i.e. 1 in 50). For comparison, the risk to woman of developing breast cancer is higher (approximately 11% or 1 in 9).
2. *Women at intermediate risk:* Some women have a mother or sister or daughter (one relative only) who has had ovarian/tubal cancer. The risk of developing ovarian/tubal cancer by the age of 70 in this group is approximately 4% (i.e. 1 in 25), which is still, less than the risk of developing breast cancer. Others have a slightly increased risk as they have a combination of relatives with breast and ovarian cancer in the family but their risk is still below that of the women described below.
3. *Women at increased risk:* A small number of women belong to families with 2 or more close relatives who have had ovarian/tubal cancer or developed breast cancer at young age. The risk of developing ovarian cancer by the age of 70 in this group is 10% or more (1 in 10 or more).

Included in this group, are a very small group of women who are found to have alterations



UCL Hospitals is an NHS Trust incorporating the Eastman Dental Hospital, Elizabeth Garrett Anderson Hospital, Hospital for Tropical Diseases, The Heart Hospital, The Middlesex Hospital, National Hospital for Neurology & Neurosurgery, Royal London Homoeopathic Hospital and University College Hospital.

University College London Hospitals

NHS Trust

(mutations) in the two genes, BRCA1 and BRCA2. The risk of developing ovarian cancer by the age of 70 in a woman with a BRCA1 mutation is about 30-45% (up to 1 in 2) and in a woman with a BRCA2 mutation is 11-25 % (up to 1 in 4).

On the basis of your family history I believe that you are at an high risk of developing ovarian cancer. This risk is related to age. Based on available data the ovarian cancer risk begins to rise from the age of 40 years in BRCA1 carriers and from the age of 45 years in those with BRCA2 gene mutations. There may be some exceptions in families where it is known that women have had ovarian cancer at ages below 40 years.

The second area we discussed was the possibility of genetic testing. In some families a strong history of cancer is due to inheritance of an abnormal gene, which is passed down through a family. Two of the genes involved (BRCA1 and BRCA2) have been identified but testing for them is not without problems. It is sometimes possible to arrange testing for these genes for women in the increased risk group if a blood sample can be obtained from a relative who has had cancer. Abnormalities in these genes are more common in people of Ashkenazi Jewish descent. **Your risk of cancer is sufficiently high to justify genetic testing.**

There is good evidence that the overall risk of ovarian cancer is reduced by the use of the combined oral contraceptive pill. The benefits persist for up to 30 years after stopping the pill though the level of benefit decreases with time. The protection is probably because the pill stops ovulation (the monthly release of an egg from the ovaries). A reduction in risk of approximately 50% has been reported in women with a BRCA alteration following 5 years use. The pill also reduces the risk of endometrial cancer.

There is a very small increased risk of developing breast cancer with the combined pill. This lasts during the period of use and for up to 10 years after stopping the pill. Our advice in this situation is that it would be probably best to take the pill for three to five years and ideally to take it after the age of 30. Women who have been diagnosed with breast cancer should NOT take the pill.

The most effective method of reducing risk is to remove the ovaries and tubes. This removes the risk of a cancer arising in the ovary or tube. In premenopausal women, surgery also decreases the risk of breast cancer by about 50%. There is however, a small (about 2-4%) persistent risk of cancer of the lining of the abdomen called peritoneal cancer, which behaves like ovarian cancer. There is a small risk of complications (1-3%) associated with any surgical procedure. The other main considerations are that you will not be able to have children after this procedure and surgery will lead to early menopause for which we usually (there may be some exceptions) recommend hormone replacement therapy till the age of 50. Surgery is usually undertaken about 5 years ahead of the age when the youngest person in your family was diagnosed with ovarian cancer or in the early to mid forties after you have completed your family. **Your risk of ovarian cancer is sufficiently high to justify primary surgery to remove the ovaries and tubes.**

Finally we discussed other options available

Screening

The current situation is that we do not know if screening works. There are two tests (ultrasound scanning and a blood test for a tumour marker called CA125) that are being evaluated in research trials. These tests can detect some cases of ovarian cancer before they cause any symptoms. However, neither test has yet been shown to save lives when used to screen for ovarian cancer. In women over 50 years at low and intermediate risk, screening is being tested in a national trial called UK Collaborative Trial of Ovarian Cancer Screening (UKTOCS). In the high-risk population, a national trial called the UK Familial Ovarian Cancer Screening Study (UKFOCSS). UKFOCSS ended recruitment on 31st March 2010 and will screen participants till 2011. It is scheduled to report in 2013. **A national screening programme for high-risk women will only be available if the results of these trials show definite benefit.**

Management of key symptoms

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The Department of Health has published information about the key symptoms of ovarian cancer to increase the likelihood of earlier diagnosis. The key symptoms include, persistent pelvic and abdominal pain, increased abdominal size/persistent bloating (not bloating that comes and goes) and difficulty eating and feeling full quickly.

We discussed these symptoms and you were given printed information about them. **Should you regularly experience these symptoms which are not normal for you, it is important you see your GP, who can make a referral to the Familial Cancer Clinic requesting an ultrasound and CA125. While it is unlikely these symptoms are due to cancer, it is important to be checked.**

When women undergo ultrasound and CA125 testing, there is a definite risk of false positive results leading to anxiety and unnecessary surgery. This is higher in younger women who are subject to cyclical variations leading to functional cysts and may suffer from benign conditions like endometriosis. Also it is important to note that these tests can sometimes miss cancer. Having understood the risks and limitations, you opted to undergo a baseline screen.

Appendix 2

2.1 *Patient Education and Counselling Paper*

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Appendix 2.1 – Patient Education and Counselling Paper

Patient Education and Counseling 88 (2012) 256–261



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Medical Decision Making

Decision making and coping in healthcare: The Coping in Deliberation (CODE) framework

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ABSTRACT

Objective: To develop a framework of decision making and coping in healthcare that describes the two processes of appraisal and coping faced by patients making preference-sensitive healthcare decisions. **Methods:** We briefly review the literature for decision making theories and coping theories applicable to preference-sensitive decisions in healthcare settings. We describe first decision making, then coping and finally attempt to integrate these processes by building on current theory.

Results: Deliberation in healthcare may be described as a six step process, comprised of the presentation of a health threat, choice, options, preference construction, the decision itself and consolidation post-decision. Coping can be depicted in three stages, beginning with a threat, followed by primary and secondary appraisal and ultimately resulting in a coping effort.

Conclusions: Drawing together concepts from prominent decision making theories and coping theories, we propose a multidimensional, interactive framework which integrates both processes and describes coping in deliberation.

Practice implications: The proposed framework offers an insight into the complexity of decision making in preference-sensitive healthcare contexts from a patient perspective and may act as theoretical basis for decision support.

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1. Introduction

In the face of medical uncertainty, a number of healthcare decisions are dependent on personal circumstances, values and preferences of the patient [1]. These 'preference-sensitive' decisions may be complex, unfamiliar and difficult for patients and deliberation and coping processes in these situations are not well understood and rarely described in detail [2]. For preference-sensitive medical decisions, a detailed description of the deliberation and coping processes undergone by patients may be helpful to understand the different issues and concerns involved in these decisions and to improve the support available to patients in those situations.

There are a number of theories addressing decision making and coping processes in general and in healthcare settings in particular [3–11]. However, many of the more traditional theories do not consider the relationships between both processes, despite their interdependency in real life. More recently, researchers have begun to integrate coping and decision making theories and thereby describe more fully the processes individuals go through when making medical decisions [12,13].

Building on this body of work, we review decision making and coping theories with a view to develop a multidimensional, interactive framework of patient's coping responses in preference-sensitive medical decisions. We begin by examining the process of deliberation in healthcare settings, followed by a description of coping in response to health threats. Finally, we attempt to integrate both processes in one framework.

2. Deliberation in preference-sensitive medical contexts

Particularly in situations of equipoise when no clear medical guidance is available or when decisions are preference-sensitive, decision making in healthcare requires patient input [1]. The decision making process in such situations may be described in several stages, including pre-decisional deliberation, decision determination and consolidation ([14]; Fig. 1).

First, the health threat is presented in the form of a diagnosis, test result or risk assessment. This is followed by the introduction of choice, which is often a new and unfamiliar concept for patients. Next, options are described to, and interpreted by, patients, while preferences are constructed. Once preference construction has been completed to identify the preferred option, a decision can be made. At any point in this process, a patient may decide that s/he does not wish to make this decision for her/himself and prefers to transfer responsibility to her/his physician. A decision may be final

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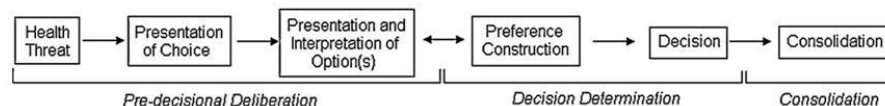


Fig. 1. The deliberation process in healthcare.

or may be revised at a later stage, deferred or avoided [15]. Consolidation of a decision will protect the decision maker from regret regarding her/his decision in the future [11,16].

Patients' reactions to a health threat and associated treatment or prevention options depend on a large number of contextual influences, such as personal beliefs, values and goals, past experiences, and social support [17]. Interpretations of information regarding the health threat and treatment or prevention options also depend on the current emotional and health state of the patient [7]. Emotions such as distress and anxiety can have detrimental effects on decision making and may lead to poor decisions [6].

When faced with decisions in healthcare, patients are often given a substantial amount of information that is new, relatively complex and threatening. Both, the quantity and content of this information may cause distress. It is likely that patients struggle to remember all the information they are given during a consultation [10]. Decisions are therefore based on the parts of the information that were understood and remembered, rather than exact numerical values and associations. Affect heuristics and intuition have also been considered important in medical decisions, especially those which do not allow the patient much time to consider the options, for instance when parents need to decide about amniocentesis [4,18].

Power et al. [13] developed a framework of medical decision making which considers the importance of emotion as a motivational aspect in decisions. "Cognitive-emotional decision making" distinguishes cognitive processes in response to the health threat and the decision from emotional processes in response to the health threat and the decision. This distinction allows consideration of each aspect of decision making in turn and of the motivational influence emotions have on cognitive processes relating to the health threat and the decision. For instance, feelings often influence risk estimates, as patients struggle to understand the concept of risk and therefore use their emotional responses to the threat to interpret their risk [19]. This can in turn influence decisions relating to that risk.

As distinguished by Loewenstein et al. [19], decision makers are experiencing *anticipatory* and *anticipated* emotions. Each phase of the decision making process in healthcare is influenced by both types of emotions. Anticipatory emotions arise in response to the immediate issue of being faced with a health threat and with the need to make a decision. Anticipated emotions are predicted emotions which the patient expects s/he will experience when one or more of the possible outcomes associated with the decision occur. These will form at the same time as anticipatory emotions. For instance a patient may imagine what a health threat might mean for their future and how they would feel about this as soon as they become aware of the threat. Furthermore, each option and associated outcomes will give rise to anticipated emotions.

As discussed by Wilson and Gilbert [20], individuals struggle to accurately predict both the duration and intensity of anticipated emotions. Therefore, emotions can lead to biases in the decision making process, as patients may base their decision partly on inaccurate predictions of their future emotions.

Regulating both anticipatory and anticipated emotions is an important motivational target in decision making [13]. Strong negative anticipatory emotions may lead to avoidant behaviour, which allows patients to temporarily regulate these emotions [15].

However, it has been noted that unless the situation is out of their control, non-avoidant behaviours lead to better long-term outcomes [21].

3. Coping with healthcare decisions

Patients who are required to play an active role in decisions about their healthcare have to cope with a number of different challenges and emotions. They need to cope with the fact that they are either ill or at risk of disease. Additionally, they need to come to terms with the fact that there are several options, and that they are being asked to take an active role in decision making. Further, they have to consider the different options and all their potential outcomes in order to make that decision.

Researchers have explored coping processes in healthcare, for example using Leventhal's Self-regulatory Model (SRM) of illness representations [9] and Lazarus and Folkman's Transactional Model of Stress, Appraisal and Coping [8]. An outline of the coping process as described by Leventhal and Lazarus and Folkman is presented in Fig. 2.

According to Lazarus and Folkman, once a threat has been presented, it is appraised in two steps [8]. During *primary* appraisal, the patient cognitively and emotionally assesses the health threat, its severity and its relevance to her/his life [8,9]. As proposed by Leventhal [9], cognitive appraisal of the health threat is based on the patient's understanding and interpretation of the threat's identity, cause, timeline, consequences and possibilities for control. During *secondary* appraisal, the patient assesses the coping resources at her/his disposal [8]. Coping resources may be problem-focused, designed to deal with the threat directly, or emotion-focused, designed to regulate the emotions experienced in response to the threat [22]. Some coping resources may provide

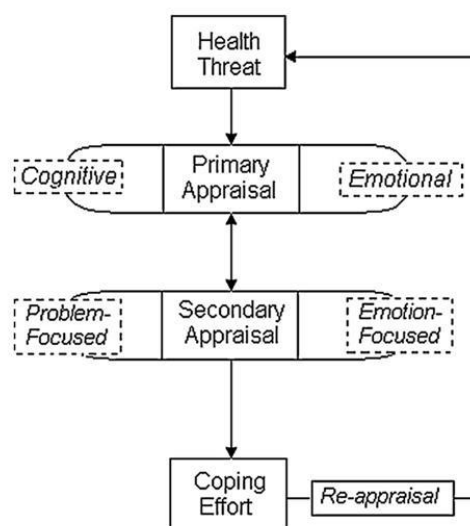


Fig. 2. An outline of the coping process as proposed by Leventhal and Lazarus and Folkman [8].

a source for problem-focused and emotion-focused coping. For instance social contacts may provide valuable help and information and at the same time emotional support.

Primary and secondary appraisal do not necessarily occur sequentially, but rather form a feedback loop, in which a quick primary appraisal gives rise to an initial assessment of the most important coping resources, which may be followed by a more detailed appraisal [17].

Following appraisal, the patient selects one or more coping strategies to deal with the health threat. These may include a mixture of problem- and emotion-focused strategies. For instance an individual may both positively re-appraise the situation and seek information about the threat and available options. The preferred coping strategy may be influenced by personal factors and characteristics, such as self-efficacy beliefs, past experiences and social settings [17,23]. It may also depend on the patient's preferred coping style [24,25]. Once a coping strategy has been selected the threat may be reappraised and strategies may be adapted if the desired outcome was not achieved. The effectiveness of a coping strategy depends on its appropriateness or 'goodness of fit' for the situation [8,22]. Problem-focused strategies may be adaptive for modifiable situations, for instance when there is a choice between treatments for a medical condition, whereas emotion-focused strategies appear more effective in situations when nothing can be done, such as periods of waiting for the results of medical tests [23]. Coping flexibility describes the ability of individuals to adapt their coping strategy to cater for the coping demands of a specific situation [26]. The embedded decisional model of stress and coping [12], describes the decision making process by outlining a series of questions based on coping theory. The authors combined Janis and Mann's conflict theory [6] with Lazarus and Folkman's transactional model and their illustration of system variables for stress [8,27]. The resulting model explores causal antecedents, mediating processes and short- and long-term effects of a decision through a series of five questions: "what are the antecedent circumstances in making this treatment decision?", "what is at stake in making this treatment decision?", "what are my options in making this treatment decision?", "what are the possible ways of coping with this decision?" and "what are the adaptational outcomes of this decision?". These questions address different considerations important in the decision making process, including potential coping resources. This was one of the first papers to integrate decision making and coping in an attempt to provide a more holistic vision of this process than previous work.

Building on the models by Balneaves and Long [12] and Powers [13], the framework presented in this paper adopts Leventhal's [9] distinction between cognitive and emotional appraisal of the health threat and Lazarus and Folkman's [8] discrimination of problem- versus emotion-focused coping. This allows a similar breakdown of processes during primary and secondary appraisal, into cognitive or problem-focused approaches on the one hand and emotional processes on the other (Fig. 2). However, others have published alternative ways to understand coping distinctions, such as engagement versus disengagement and accommodative and meaning-focused coping [28–31], which should be acknowledged. Rather than being mutually exclusive, these categories are overlapping and many of the coping strategies explored in the framework presented in the next section may fit with more than one conceptual approach.

4. The Coping in Deliberation (CODE) framework

Coping with a health threat and its potential short- and long-term consequences is an integral part of the deliberation process in preference-sensitive healthcare decisions. Without appraisal and coping strategies, individuals may not be able to make informed,

value-adjusted decisions, as their emotional state would hinder an effective analysis of the situation and their options. The transactional functions of coping aid the decision maker in regulating their emotional state whilst going through the decision making process and may also allow her/him to devise active strategies to deal with a given threat. Therefore integrating decision making theory and coping theory is a useful exercise, describing a more holistic vision of human decision making in preference-sensitive health contexts.

By proposing a detailed framework of coping processes during deliberation, we attempt to follow on from the ideas of Balneaves and Long [12] as well as Power [13]. The Coping in Deliberation (CODE) framework postulates that each phase in deliberation gives rise to an appraisal and coping process, the result of which can influence a patient's attitude and behaviour in other stages of deliberation. Deliberation is therefore described as a multidimensional coping process, depicted in Fig. 3 as the *x*- and *y*-axis. The framework applies to preference-sensitive decisions in healthcare contexts, meaning those which require the exploration of patient values and allow patient input as no clear medically recommended option exists.

The patient may move through the process from top to bottom and left to right. At the start of the deliberation process the patient is presented with a health threat. S/he appraises this threat cognitively and emotionally during primary appraisal by exploring questions relating to the threat itself and her/his feelings about it. S/he also appraises the coping resources that could be helpful in dealing with this threat. These include her/his ability to find out more, the availability of social support and the possibility of seeing a positive side to the situation. S/he selects one or more coping strategies that may be helpful in dealing with this health threat. For instance s/he may decide to find out as much as possible about the diagnosis and her/his options. The coping strategies selected will determine her/his reaction to, and interest in, the idea of choice and the different treatment or prevention options, which will be presented in the next stages of the deliberation process.

While this description depicts a relatively linear approach to decision making and coping, one has to consider that coping is interactive, multidimensional and transactional. Primary and secondary appraisal can occur simultaneously and individuals may revisit certain threats, reappraise their situation and readjust their coping efforts. They may also skip certain steps in the process, or first appraise a threat briefly to then return later for a more comprehensive appraisal. The framework aims to portray this possibility of moving through the process in a non-linear fashion.

In the framework, a series of questions for each appraisal process outlines possible aspects a patient might explore. The questions were designed to be adaptable to different decisions. Statements at the end of each column represent some of the coping strategies the patient might select, which may again be adapted to different contexts. The framework was designed to accommodate a wide range of preference-sensitive medical decisions, ranging from decisions about genetic testing and cancer prevention to those about choices between different medications or types of surgery.

To give an example of the adaptability of the framework, consider how the questions might be adapted if the health threat was increased genetic risk for ovarian cancer. The woman may initially appraise her risk and ask what ovarian cancer is, what genetic risk means, how threatening this risk is to her health and how she feels about being at increased risk. She may then appraise the resources for dealing with that risk, such as talking to her clinician or family. Similarly, during the deliberation about options, the woman may ask specific questions about the risks and benefits of options such as risk-reducing salpingo-oophorectomy (i.e. surgery to remove health ovarian tissue) and the effects of surgery on her life, and she may consider follow-on decisions

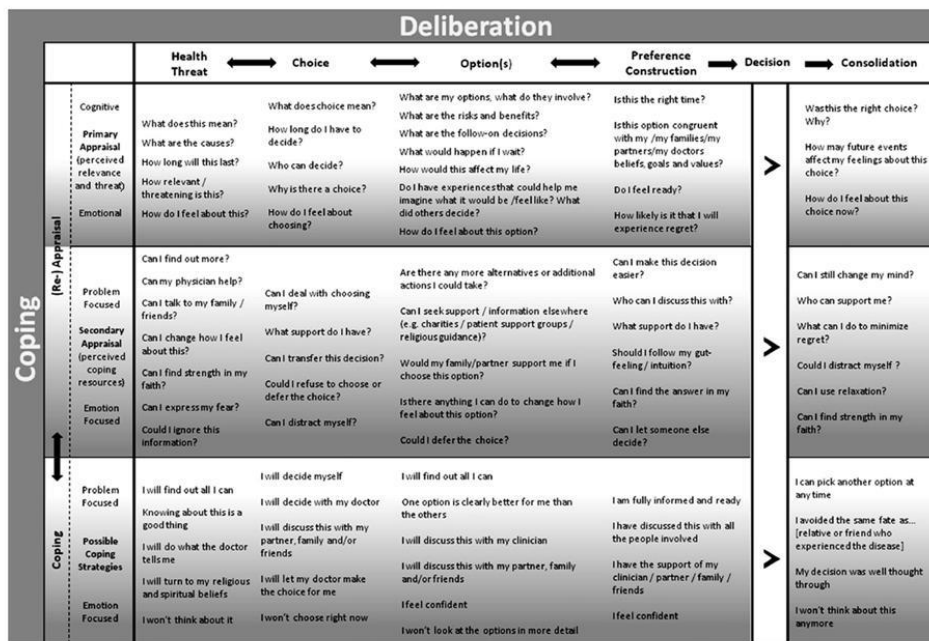


Fig. 3. The Coping in Deliberation (CODE) framework.

such as hormone replacement therapy (HRT) and alternatives such as ovarian screening [32]. The questions within the framework can be adapted accordingly to reflect these cognitive and emotional processes. It should be noted that questions and coping strategies will vary not only depending on the decision at hand, but also on the characteristics of the individual patient. Additionally to preferred coping style, other influences such as life stage, experience and values will play a role. In this example, a patient aged 55 may not consider HRT, whereas a woman in her early 40 s might explore that option in detail. Similarly, a single mother would not look for her partner for coping support, but might turn to her clinician or friends, whereas someone in a stable relationship might value input from a partner and explore how options would affect their relationship. A religious person might explore whether s/he might find strength in their faith as a coping resource.

To demonstrate the flexibility of the CODE framework, consider a completely different medical decision, such as amniocentesis. Parents are required to cope with the fact that a routine test has indicated a potential problem and are now faced with the decision of whether or not to explore this further. They may ask what the current result means, how it might affect their pregnancy and who they could rely on for support. During deliberation they may consider the risks and benefits of amniocentesis, asking themselves what they would do if the test confirmed an abnormality, consider termination of pregnancy and explore their coping resources. During preference construction they may ask whether they both agree and how likely they are to experience regret.

Another example may be selected from the coronary artery disease (CAD) area, where patients may be asked whether they would like to have a catheter investigation to check for artery disease or whether they would rather have medical therapy. Patients in this situation need to appraise and cope with the fact that they are at risk of CAD and what this means to their life. They are also required to appraise their options, such as catheter

investigation, asking about the risks and benefits, potential outcomes and exploring whether they know someone who went through this procedure. During secondary appraisal they may ask whether they can turn to others for support or simply avoid dealing with the situation.

These three examples depict preference-sensitive medical decisions and demonstrate the flexibility of the framework by adding, removing and adapting questions depending to the decision at hand. Further questions can be added, removed or adapted depending on the individual patient's coping style, life-stage and experiences.

Incorporated in the CODE framework are many of the concepts found within both coping and decision making theories. The questions posed during the appraisal process were selected to represent a wide range of considerations and influences. For instance, personal values and goals which can affect the interpretation of different options are considered when the patient assesses the relevance of the health threat and again when s/he evaluates the potential effects of options on her/his life.

The coping resources and strategies were selected to represent different coping strategies that patients may use, such as vigilance, avoidance and seeking social support [5,6]. Coping strategies may change over time or vary from one stage to another.

The contents of the CODE framework are not aiming to be comprehensive, they rather reflect a variety of different questions and coping strategies a patient may consider whilst going through the deliberation process, maintaining the multidimensional and transactional functions of coping [12]. The framework does not propose that one coping strategy is more effective than another, as different strategies may be appropriate in different situations [8]. It therefore includes a range of strategies and allows decision makers to select any number and adapt their approach at a later stage, if necessary. The number and types of questions asked, the answers to these questions, the choice of coping strategies and ultimately the decision itself will depend on the type of health threat, the

options, the patient's coping style, her/his preferences, past experiences, personal values and ability to forecast feelings accurately.

The framework includes a consolidation phase following the actual decision. Consolidation is in essence a coping mechanism designed to defend a decision from regret in the future [11,16]. The inclusion of this phase allows the framework to describe deliberation beyond the actual decision and acknowledges that the consequences of a decision should be an integral part of any decision making model, as patients ultimately have to live with their decision and its consequences.

5. The CODE framework in context

Leventhal et al. [9] were the first to consider the importance of emotional representations in addition to cognitive representations in response to threats. More recently, Power [13] distinguished cognitive and emotional representations formed in response to a health threat from those formed in response to the need for a decision. She also argues that emotions play an important motivational role during decision making. Following on from this idea, the CODE framework proposes a further distinction between the different stages within the decision and postulates that each of these stages gives rise to its own individual cognitive and emotional appraisal process. This additional distinction allows a more detailed analysis of the representations formed during the different deliberation stages. In agreement with Power [13] and other theorists such as Lazarus and Folkman [8], the CODE framework considers emotional appraisal and coping processes to be of equal importance to cognitive or problem-focused processes. Both occur in parallel, influence each other and are essential for good decision making.

The embedded decisional model of stress and coping describes decision making as a series of quite general questions [12]. The CODE framework similarly identifies questions relating to deliberation and coping processes, but does so in a more detailed fashion. For instance, what Balneaves and Long [12] cover with the question "what are my options in making this decision", we explore with a series of more detailed questions about benefits and risks, effects on life and possible related decisions. Similarly, Balneaves' question "what are the possible ways of coping with this decision", is split into a series of twenty questions during secondary appraisal, describing different coping strategies at different stages in the deliberation process, such as "can I talk to my family/friends", "can I change how I feel about this" and "can I transfer this decision". Additionally the framework includes a consolidation phase following the decision, as proposed by Svenson [11]. Furthermore, the CODE framework explores these issues from the patient perspective, which may allow practitioners to develop an understanding of their patients' deliberation processes.

6. Practice implications and future research

Shared decision making and patient centred care (PCC) are becoming increasingly important in healthcare, especially in the context of preference-sensitive decisions which require patient input [1,11,33–35]. In order to provide PCC, practitioners are expected to understand their patients' needs and concerns and to address them accordingly. The preliminary CODE framework provides a description of preference-sensitive decision making from a patient perspective, which may ultimately – when adapted to specific decisions – help practitioners understand their patients' questions and concerns, by shedding light on underlying cognitive and emotional processes. Additionally, the framework may provide a theoretical underpinning for the development of decision aids

which are designed to support patient decision making, as currently many of these tools lack a theoretical basis [36].

The framework now needs to be adapted to actual decisions in healthcare and tested with patients to explore its applicability and provide feedback on its validity and potential amendments that may be needed. The adaptation of the preliminary framework to specific decisions should be closely monitored by individuals involved in those decisions, including patients, patient organisations and health professionals, to allow for an accurate representation of questions and coping strategies patients may explore when facing the selected decision. Currently, research is under way to adapt the CODE framework to describe decisions about risk-reducing surgery in the context of familial ovarian cancer risk. Adaptations to other decisions may follow. Empirical evidence about the use of adapted versions of the framework as a guide for practitioners in consultations and for decision aid development will also be gathered once adapted versions are available.

7. Limitations

Currently the CODE framework is a theoretical description of deliberation and coping processes, designed to be applicable to a range of preference-sensitive decisions. The CODE framework now needs to be applied to actual decisions and tested with patients to evaluate its validity.

Few theories to date have specifically considered emotional and information support needs of patients when they are making decisions about their health. The questions included in the CODE framework may provide health professionals with a basis for looking at the variety of patients' questions and concerns. It also draws attention to the fact that deliberation and coping processes are closely linked and therefore may help practitioners in exploring coping resources together with their patients, potentially adding a new element to PCC. In the future, the framework may benefit from an extension which allows the inclusion of patient information and coping needs. This would additionally strengthen its value as a support tool for PCC and a guide for the development of patient decision aids.

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Appendix 3

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| 3.2 | <i>Critical Appraisal Checklist (Qualitative)</i> | 418 |

Appendix 3.1 - Critical Appraisal Checklist (Quantitative)

Quantitative research papers on women's needs, concerns and characteristics

| Paper Title | Year | First Author | |
|--|------|-----------------|-----------|
| Screening Questions | | | |
| | | Yes | No |
| Was there a clear statement of the aims of the research? -was the goal stated? -why is this important? -how relevant is it? | | | |
| Is a quantitative methodology appropriate? -does the research aim to illuminate the characteristics/demographics of participants or assesses their opinions/attitudes? | | | |
| Is it worth continuing? | | | |
| Detailed Questions | | Comments | |
| Was the research design appropriate to address the aim of the research? -has the research design been justified? | | | |
| Was the recruitment strategy appropriate to the aims of the research study? -has the researcher explained how participants were selected? -has the researcher explained why the participants were appropriate to provide answers to the research question? -has the researcher discussed non-participants and their reasons? -has the researcher discussed response rate? -has the researcher discussed how the control group was selected and why (if appropriate)? -were demographic variables of respondents discussed? | | | |
| Were the data collected in a way that addressed the research issue? -was mode of data collection justified (medical records / questionnaires etc.)? -is it clear how data was collected? -has the researcher made methods explicit? -if a survey was used, was the survey piloted? - " " , were there open questions? How were they analysed? | | | |
| Has the relationship between researcher and participants been adequately considered? -has the researcher considered their own role, potential bias and influence? 1. during formulation of research questions 2. during recruitment and data collection (if data collected from participants through face-to-face or telephone survey) -has the researcher considered the implications of any changes in the research design and events during the study? | | | |

| | |
|---|--|
| <p>Have ethical issues been taken into consideration? <i>-were ethical standards maintained (if enough information provided)?</i> <i>-has approval been sought from ethics committee?</i></p> | |
| <p>Was the data analysis sufficiently rigorous? <i>-has an in-depth description of data analysis been provided?</i> <i>-is it clear which statistical tests were used / were the tests appropriate to answer the research question?</i> <i>-is sufficient data presented to support the findings?</i> <i>-to what extent was contradictory data taken into account?</i></p> | |
| <p>Is there a clear statement of findings? <i>-are the findings explicit?</i> <i>-is there adequate discussion of the evidence for and against the researcher's arguments?</i> <i>-has the researcher discussed the credibility of their findings?</i> <i>-are findings discussed in relation to other research?</i></p> | |
| <p>How valuable is the research? <i>-has the researcher discussed the contribution his study makes to existing knowledge?</i> <i>-does the researcher identify areas for further research?</i> <i>-has the researcher discussed whether findings can be transferred to other populations or considered ways the research can be used?</i></p> | |
| <p>Further comments</p> | |

Adapted from the national Critical Appraisal Skills Programme (CASP) collaboration Critical Appraisal Checklist

Appendix 3.2 - Critical Appraisal Checklist (Qualitative)

Qualitative research papers on women's needs, concerns and characteristics

| Paper Title | Year | First Author | |
|--|------|-----------------|----|
| Screening Questions | | | |
| | | Yes | No |
| Was there a clear statement of the aims of the research? -was the goal stated? -why is this important? -how relevant is it? | | | |
| Is a qualitative methodology appropriate? -does the research aim to illuminate the actions/experiences/opinions and subjective views of participants? | | | |
| Is it worth continuing? | | | |
| Detailed Questions | | Comments | |
| Was the research design appropriate to address the aim of the research? -has the research design been justified? | | | |
| Was the recruitment strategy appropriate to the aims of the research study? -has the researcher explained how participants were selected? -has the researcher explained why the participants were appropriate to provide answers to the research question? -has the researcher discussed non-participants and their reasons? | | | |
| Were the data collected in a way that addressed the research issue? -was the setting for data collection justified? -is it clear how data was collected? -has the researcher made methods explicit? -is the form of data clear? -has the researcher discussed saturation of data? | | | |
| Has the relationship between researcher and participants been adequately considered? -has the researcher considered their own role, potential bias and influence? 1. during formulation of research questions 2. during recruitment and data collection -has the researcher considered the implications of any changes in the research design and events during the study? | | | |
| Have ethical issues been taken into consideration? -were ethical standards maintained (if enough information provided)? -has approval been sought from ethics committee? | | | |
| Was the data analysis sufficiently rigorous? -has an in-depth description of data analysis been provided? | | | |

| | |
|--|--|
| <ul style="list-style-type: none"> -is it clear how categories/themes were derived (if used)? -has the researcher explained how data extracts were selected? -is sufficient data presented to support the findings? -to what extent was contradictory data taken into account? | |
| <p>Is there a clear statement of findings?</p> <ul style="list-style-type: none"> -are the findings explicit? -is there adequate discussion of the evidence for and against the researcher's arguments? -has the researcher discussed the credibility of their findings? -are findings discussed in relation to other research? | |
| <p>How valuable is the research?</p> <ul style="list-style-type: none"> -has the researcher discussed the contribution his study makes to existing knowledge? -does the researcher identify areas for further research? -has the researcher discussed whether findings can be transferred to other populations or considered ways the research can be used? | |
| <p>Further comments</p> | |

Adapted from the national Critical Appraisal Skills Programme (CASP) collaboration Critical Appraisal Checklist

Appendix 4

| | | |
|-----|---|-----|
| 4.1 | <i>Multi-Centre Ethics Committee for Wales Favourite Opinion Letter</i> | 421 |
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Appendix 4.1 - Multi-Centre Ethics Committee for Wales Favourite Opinion Letter

Part of the research infrastructure for Wales funded by the National Institute for Social Care and Health Research, Welsh Assembly Government
Rhan o seilwaith ymchwil Cymru a ariannir gan y Sefydliad Cenedlaethol ar gyfer Ymchwil Gofal Cymdeithasol ac Iechyd, Llywodraeth Cynulliad Cymru



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Website : www.nres.nhs.uk

03 May 2011

Miss Jana Witt
PhD Student
Cardiff University
2nd Floor, Neuadd Meirionydd
Heath Park
Cardiff CF14 4YS

Dear Miss Witt

Study title: OvDex - Development and preliminary evaluation of a decision support intervention for risk management options in familial ovarian/breast cancer
REC reference: 11/WA/0094
Protocol number: SPON 945-11

Thank you for your letter of 21 April 2011, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chairman of the REC for Wales, Dr. Gordon Taylor.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

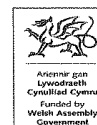
Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rforum.nhs.uk>.



Cynhelir Cydweithrediad Gwyddor Iechyd Academaidd y Sefydliad Cenedlaethol ar gyfer Ymchwil Gofal Cymdeithasol ac Iechyd gan Fwrdd Addysgu Iechyd Powys

The National Institute for Social Care and Health Research Academic Health Science Collaboration is hosted by Powys Teaching Health Board



Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

Other conditions specified by the REC

- The information sheets refer to NHS Cardiff and Vale Ethics Committee and this should be replaced with the Research Ethics Committee for Wales.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

You should notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. Confirmation should also be provided to host organisations together with relevant documentation.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

| Document | Version | Date |
|---|--|---------------|
| Protocol | version VIII | |
| Response to Request for Further Information | Letter from Miss Witt | 21 April 2011 |
| Participant Information Sheet: Observations and interviews - clinicians | version VIII | 18 March 2011 |
| Participant Information Sheet: Cardiff - Focus Groups - Women who opt for surgery | IX | 14 April 2011 |
| Participant Information Sheet: London - Virtual reference group - Patients | IX | 14 April 2011 |
| REC application | signed in ink by Miss Witt; electronically by Matthew Harris, sponsor's representative; electronically by Professor Glyn Elwyn, Dr Kate Brain and Dr Fiona Wood (academic supervisors) | 30 March 2011 |
| Participant Consent Form: Interviews | version VIII | 18 March 2011 |
| Participant Consent Form: Virtual reference group | version VIII | 18 March 2011 |
| Participant Information Sheet: Cardiff - Focus Groups - Women who are undecided | IX | 14 April 2011 |
| Participant Consent Form: Observation and interview - patient | version VIII | 18 March 2011 |
| Participant Information Sheet: London - Observation and interview - Patients | IX | 14 April 2011 |
| Interview Schedules/Topic Guides | Focus groups (1-4) guidelines - women who are undecided; version VIII | 18 March 2011 |
| Interview Schedules/Topic Guides | Focus group 95) guidelines - women who opted for surgery | 18 March 2011 |
| Interview Schedules/Topic Guides | Semi structured interview schedule - health professionals (clinical); version VIII | 18 March 2011 |

| | | |
|---|---|-------------------|
| Interview Schedules/Topic Guides | Semi structured interview schedule - health professionals (charity); version VIII | 18 March 2011 |
| Evidence of insurance or indemnity | Zurich Municipal certificate of insurance - Cardiff University - expires 31 July 2011 | 27 July 2010 |
| Referees or other scientific critique report | Clinical epidemiology interdisciplinary research group peer review | 04 September 2010 |
| Interview Schedules/Topic Guides | Interview scheduled after use of the tool (patients); version VIII | 18 March 2011 |
| Interview Schedules/Topic Guides | Interview schedule after use of the tool (professionals); version VIII | 18 March 2011 |
| Covering Letter | signed Miss Witt | 29 March 2011 |
| Summary/Synopsis | version VIII (Layman) | 18 March 2011 |
| Letter from Sponsor | signed Dr KJ Pittard Davies, Cardiff University | 22 March 2011 |
| Investigator CV | Jana Witt | 24 March 2011 |
| Investigator CV | Glyn Elwyn | 23 March 2011 |
| Investigator CV | Katherine Brain | 29 March 2011 |
| Participant Information Sheet: Interviews with health professionals | version VIII | 18 March 2011 |
| Investigator CV | Fiona Wood | |
| Participant Information Sheet: Cardiff - Observation and interviews - Patients | IX | 14 April 2011 |
| Participant Information Sheet: Cardiff - Virtual reference group - Patients | IX | 14 April 2011 |
| Participant Information Sheet: Swansea - Focus group - Women who are undecided | IX | 14 April 2011 |
| Participant Information Sheet: Virtual reference group (professionals) | version VIII | 18 March 2011 |
| Participant Consent Form: Focus group | version VIII | 18 March 2011 |
| Participant Consent Form: Future contact | version VIII | 18 March 2011 |
| Participant Consent Form: Observation and interview (professional) | version VIII | 18 March 2011 |
| Participant Information Sheet: London - Focus group - Women who are undecided | IX | 14 April 2011 |
| Participant Information Sheet: Swansea - Observation and interview - Participants | IX | 14 April 2011 |
| Participant Information Sheet: Swansea - Virtual reference group - Patients | IX | 14 April 2011 |

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Now that you have completed the application process please visit the National Research Ethics Service website > After Review

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

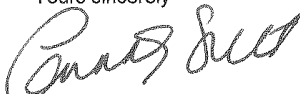
We would also like to inform you that we consult regularly with stakeholders to improve our service. If you would like to join our Reference Group please email referencegroup@nres.npsa.nhs.uk.

11/WA/0094

Please quote this number on all correspondence

With the Committee's best wishes for the success of this project

Yours sincerely


PP Dr Gordon Taylor
Chairman

Email: corinne.scott@wales.nhs.uk

Enclosures: "After ethical review – guidance for researchers"

Copy to: Kathy Pittard-Davies, Cardiff University, RACD

Appendix 4.2 - Topic Guide for Focus Groups (Needs Assessment)

Focus Groups introduction (approx 15 minutes)

Researcher introduces the team and states the aims of the research. Discussion of rules of conduct and confidentiality before proceeding to the discussion.

PART I - explore needs, concerns and questions (approx 45 minutes)

Participants are given and read vignettes about fictional woman 'Emma' [Vignette reads: Emma is 38 years old. Her sister was recently diagnosed with ovarian cancer aged 40. Their mother developed breast cancer when she was 45 and their aunt had breast cancer when she was 56. Emma does not know whether there is a faulty gene in the family at the moment. Her genetic counsellor has said she could have surgery to remove her ovaries to reduce her cancer risk. Emma is struggling to make a decision about whether or not she wants to have surgery.]

What do you think this woman's questions would be? What kinds of things do you think she would consider when making a decision about whether or not to have an oophorectomy? What are the possible reasons why she might struggle with the decision?

Prompts (if needed):

- she is very young, so maybe she wants children
- she could be quite frightened about the surgical menopause
- she is approaching the age at which her mother was diagnosed
- she may have children and feel some kind of obligation towards them
- she may be scared of surgery

And in your view, what are the really important questions that should be answered when women such as Emma are asked to make a decision about oophorectomy?

Prompts (if needed):

- These could be things like 'how much will my risk be reduced?'
- or 'how will surgery affect my life?' and so on...
- anything you can think of that is really important and should be discussed

And is there something, some reason or some event that could sway that decision one way or the other in your opinion? So I guess what I am asking is whether one of the considerations we have just talked about (such as... NAME TWO CONSIDERATIONS AS EXAMPLE) might outweigh others and then tip the scale... Like with other important decisions, such as buying a house, if you had to choose would you buy the bigger house or buy the smaller house with a bigger garden? So in this case as an example, should Emma accept the risk of having surgery now to reduce her cancer risk in the future?

Is there something you can think of that might help to compare the options and to bring out the differences? Maybe a certain way of presenting the information that might make it easier to understand and compare?

Ranking exercise:

What I would like to do next is to do a ranking exercise and see which reasons are most and which are least important. So I have got a sheet here with 'most important' and one with 'least important' and if we put these on opposite ends of the table and say that we create a scale, then anything that is very important goes here and anything that's just a bit important goes somewhere there. And what I would like you to do is take these reasons that we just talked about and place them somewhere along this line. And this should be a collective exercise, so you should all discuss and decide together where each reason goes. You may not all agree, but that's absolutely fine, I really want you all to speak up and discuss what you think should go where and if you don't think something is in the right place, just say so. Please try not to speak on top of each other though.

PART II - discuss expectations of a potential tool (approx 20-30 minutes)

I would like to move on now, for the last (MINUTES LEFT) to chat a little bit about this idea for developing a decision aid, so something that can help women who are struggling to make up their mind (one way or the other). Are we all okay for time or does anyone need to leave early?

So, first of all, what do you think such a decision aid could look like?

Prompts (if needed):

What kind of format do you think would work best? Something on paper, like a Brochure or maybe something online, like a website... what would be best to use?
What would you feel most comfortable using?

And, from your own experience, how do you think women in this situation, who really struggle to make a decision, could be supported through a (WEBSITE or BOOKLET)?

Prompts (if needed):

- For example, how should information be presented?
- What do you think about this (show option grid / value clarification ex.)?

Researcher sums up main points discussed in Part II and closes discussion.

Appendix 4.3 - Topic Guide for Interviews (Needs Assessment)

Introduction and reiteration of the study's purpose

Could you just explain what your actual role is, what do you do on a day-to-day basis?

And for women who have been found to be at risk of cancer, specifically ovarian or breast cancer, what is your contact experience with them? And how do you get involved with them?

So, when women talk to you about risk-reducing surgery, what do they usually want to talk about? What are their main questions / concerns?

- I'm particularly interested in women asking about oophorectomy: what is important for them?
- And what do you think are the most important / essential points that need to be discussed in this context?
- For example, are they interested in the different options to reduce their risk, the risk of family members, genetic testing, etc.?
- When it comes to women with mainly or only breast cancer family history, do they know about oophorectomy and its reduction in breast cancer risk?

And could you just explain to me from your own experience, what you think the **most important needs** (information wise and emotional) are of women who are told they are at increased risk of ovarian / breast cancer?

- Are they looking for facts and information or do you feel they just want to get some emotional support? Or both?
- Are there differences between women who have just been told and women who have known for a while and maybe return to the service?
- A lot of the time women who are at risk for ovarian cancer are also at risk for other cancers, especially breast, do you see one taking a priority over the other?

Do you have any experience of women's decision making, regarding oophorectomy? [If yes: Could you maybe tell me a little bit about your experience of the decision making process when it comes to managing ovarian cancer risk?]

- Do women take a long time?
- Do they involve you in the decision; do they involve their family or others?
- Do they try to get lots of information? Where do you think they get this from?
- What is their attitude towards this option when they come to see you?

What do you think are the **main challenges/barriers and catalysts** for women who are trying to make a decision about prophylactic oophorectomy or maybe other strategies to address their cancer risk?

- For example do they find it hard to find reliable information? Or do they just want to talk to someone to talk it through?

Could you describe for me what you think would be the most important points (information or otherwise) that should be covered in a decision aid that would be designed to help women make this decision? So just a little bit in terms of content...

- For example, what kinds of information should be in it?
- Could you think of anything apart from information about options that could be covered? Maybe something that would help the women with their emotional turmoil? Or you mentioned earlier [... maybe family issues, worry for relatives...]

Option grid example – participant view the printed option grid

Value Clarification example – participant views a printed value clarification exercise

What do you think of these? Do you think they might be helpful?

Thinking about a decision aid for those women, what do you think might be the best format for this?

- Web-based / paper / video?

Final run through issues mentioned in the literature [Select issues that have NOT been mentioned in previous conversation]

Age

Ovarian cancer family history and personal experiences

BRCA1/2 mutation

Previous cancer diagnosis

Loss of fertility / Wish to have more children

Perception of femininity and fertility

Sexuality and sexual relationships

Perceived cancer risk/cancer-related distress/worry/anxiety

Availability of screening / Beliefs in screening / Detection effectiveness

Family obligations

Issues with surgical menopause and HRT

Surgical complications / Recovery time from surgery

Residual cancer risk

Is there anything else that we have not covered today, that you think might be relevant or important and that you would like to talk about?

| Code | Theme | Definition |
|---|---|--|
| Background Information | Participant information | Any information pertaining to the participant; age; workplace; marital status; medical history (other than ovarian cancer), job responsibilities |
| | Organisation information (professionals ONLY¹) | Any information regarding the workplace (cancer genetics service; charity; network), main purpose / mission, what the organisation offers |
| | Experience of working with / supporting women at risk (professionals ONLY¹) | Any information regarding facts about experience of working with / supporting women at risk; length in role; average number of women seen / spoken to; mode of contact (face-to-face, telephone, email) |
| | Experience of being at risk (patients ONLY²) | Family history of cancer, when realised at increased risk; feelings about risk; others' reaction to risk; past experiences of ovarian cancer / cancer risk; coping with risk |
| Women's needs, questions and concerns | Initial diagnosis | Process of finding out about risk, how and when this came about, any remarks on the process of genetic testing to confirm mutation status, comments about genetics service / risk assessment process |
| | Information requests re Ovarian cancer / genetics | Women's questions and concerns <u>about ovarian cancer, risk, genetic testing</u> (NOT management options and their consequences – see L; NOT information requests sparked by media/research hypes – see H) |
| | Emotional needs | Any needs relating to non-information, requests for support groups, need to talk to someone, consideration of counselling for emotional support, contact with charities / families / friends / support groups; coping needs |
| | Responses to media /research Group and time differences | Reactions to media and research hype, concerns specifically raised by such hypes What is important to which group (pre- / post-menopausal; high / medium risk; family history only / gene mutation; ovarian-only / breast-only / ovarian/breast family); when do certain issues become important (time-point in process / in relation to each other); women who have had breast cancer (special needs regarding decision / possibility of HRT); women who have been on UKFOCS screening |
| ¹ This code is specific for professional interviews ² This code is specific for focus groups | | |

| | | |
|---|---|---|
| Decision making about ovarian cancer risk management | Facts about management options | Factual information / comments about available options, their properties, availability, usefulness, what is involved, (EXCEPT where speaking of consequences of option as a specific consideration during decision making); please specify: 1. <u>Surgery</u> – anything <u>factual</u> relating to the option of surgery, who can do it, what is done, HRT 2. <u>Other options</u> – anything <u>factual</u> relating to options other than surgery, including screening, oral contraceptives, watchful waiting, symptom awareness |
| | Process of decision making | Information about the timescale and logistics of decision making; who is involved in decision; how it came/comes about, process of consultation(s); pressures on decision making |
| | Considerations during decision making | Factors that were / are being considered during decision making; factors that influence(d) the decision; questions regarding those factors; including specific catalysts / facilitators / barriers or anything that contributes to remaining undecided; please specify: 1. <u>Medical and physical</u> – anything relating to medical history (e.g. previous cancer diagnosis, benign findings, false positives in screening), age, genetic mutation status, family history (regarding types of cancer, frequency, age at which diagnosed, outcomes), menopausal status, loss of oestrogen, issues with HRT, risk category, risk reduction (objective), effects of surgery on physical condition, surgical procedures, scars, co-morbidities 2. <u>Psychological and emotional</u> – anything relating to perceived risk, perceived effectiveness of options, distress, worry, anxiety (due to cancer risk, screening scare etc.), perceptions of femininity / sexuality / body image, goals and values (e.g. wish to have more children), self-efficacy, coping ability, indecisiveness, health beliefs, fear of hospitals / doctors / surgery 3. <u>Social Context</u> – anything relating to family obligations (e.g. bringing the kids to school, being a carer), family history (regarding closeness to affected family members, role in their care), family communication, sexual relationships with partner, work commitments 4. <u>Other considerations</u> – anything relating to physician recommendation, healthcare system, demographics, recovery time, time in hospital, other logistics, alternative therapies, social support, availability of screening and other alternatives to surgery, pressure from others, distrust of medical profession, relationship with clinic staff, vagueness of symptoms of ovarian cancer |
| | Catalyst / tipping point Facilitators to decision making Barriers to decision making | Any remarks on <u>salient</u> events that sway(ed) the decision one way or the other Remarks specifically referring to anything that made/makes the decision easier Remarks specifically referring to anything that made/makes the decision harder or more difficult, anything that results in <u>unnecessary</u> delay of the decision or confusion; Remarks specifically referring |

| | | |
|-------------------------|---|--|
| | | to anything that contributes to being undecided |
| | Coping with decision making and decision outcome | Coping strategies employed whilst making the decision / after the decision |
| | | |
| Decision support | Scope for support | Where and when support is needed (timing of decision aid, setting for decision aid, i.e. inside or outside clinic), who needs most support; comments about target audience; change of scope for support due to change in what is offered (e.g. no screening available) |
| | Decision aid content (information) | Any comments about what should be included in a decision aid regarding information (on cancer, risk, options etc.), contact details, references; level of detail for information; evidence base, format for information (e.g. option grid / table) |
| | Decision aid content (coping and value clarification) | Any comments regarding content for value clarification exercise, preference elicitation, coping advice and emotional support |
| | Decision aid format | Remarks about the best / worst format (e.g. web-based vs. paper) for the decision aid, reasons why format is good / bad; remarks about colour, font type / size etc.; sub-formatting, e.g. blank spaces, interactivity |
| | Decision aid purpose | Purpose of the decision aid, any comments about the importance of a decision aid for this context, main/minor purpose |
| | Other areas for decision support | Any comments about other areas for decision support, changes in counselling content, timing, opportunities for peer-support etc. |
| | Problem areas | Any comments regarding the current lack of support / information / trust; the way information / support is given; awareness of the need for a decision |
| | Potential effects of a decision aid | Remarks about the potential effects of the decision aid on patients, clinicians, individually or together; on interactions in- or outside the clinic |
| | Recommendations for decision aid development | Any comments regarding the development process; who should be involved; why; how it could be supported / designed |
| | Clinician attitude to options (professionals ONLY¹) | Any remarks on what women should do, what the medically preferred option is, whether options work |

| | | |
|----------------------|---------------------------------------|---|
| Miscellaneous | Participant questions | Any questions asked by participants |
| | Misunderstandings | Any misunderstandings / clarifications due to miss-understandings |
| | References / things to look at | References / websites / tools mentioned by participants which might be helpful for the decision aid development process |
| | GPs / healthcare system | Any comments regarding difficulties with or criticism of GPs and/or the healthcare system when dealing with patients, ideas for improvements of the healthcare system / service; comments regarding referral procedures between services, clinical guidelines / protocols / recommendations |

Appendix 4.5 - Extract of Coded Transcript (Focus Group)

- P1: Well I guess, you know, if you have your ovaries out what are the knock – what are the im-, first of all, what are the implications of that. You know, because I know from having discussed it, that it throws you into an early menopause, which for me is what is making me stick my head in the sand a bit at the moment. You know, but I think that’s really what my primary concern would be.
- P2: Yea, I think it’s the effects of it really, isn’t it? ...
- P1: Yeah.
- P2: And some of the questions that I have had answered have been, well “You are just as likely to be knocked down by a bus, so do you want to take that course of action?” That’s what my GP told me. Even through, you know, my mum and my gran both had it and that’s what he said. You are just as likely to be knocked over by a bus so that choice is entirely yours and when you get that off your GP you think: “Oh I don’t know if I’ll consider that any further now.” You know I had gone with a serious conversation of considering it.
- M: Yeah, and that kind of put you off, did it?
- P2: Yeah, because I, I mean I, I have just been feeling very vulnerable since the end of the programme. Very vulnerable.
- M: So surgical menopause, kind of these kinds of effects of surgery?
- P1: Yeah and because I think my, you know it is primarily breast cancer where my family tree goes, but there was, my mother’s cousin had ovarian cancer and so that’s why I think that the, and I forget the name the doctor who sort of heads up the genetics unit, who got in touch with me initially.
- M: In [Place 1] is it?
- P1: No, in [Place 2], I can’t remember her name. But she invited me anyway. But I know that you know when I discussed it with, with her and she said you know you could have your ovaries out and she said you know you could go onto HRT but of course HRT then brings up the increased risk of breast cancer. And you know, where to do you, where do you pitch it?
- P3: Yea, where is the risk?
- P1: Yeah, you know.
- M: Yeah. Do you agree with that or?
- P3: I haven’t actually discussed it with anybody at all, haven’t had any conversations with anybody apart from my mother’s doctor, who told - a

Decision Making Considerations (Medical & Physical)
+
Barriers

Decision Making Considerations (Other)
+
Misc GP & Healthcare system
+
Decision Support Clinician attitude

Background Participant Info
+
Experience of risk

Women’s Needs Initial diagnosis

Decision Making Considerations (Medical & Physical)

Women’s Needs Initial diagnosis
+
Decision Support Clinician attitude

doctor here, a cancer doctor - who told me that basically I should have the surgery straight away as soon as I can.

M: So that's very dissimilar to what you [P2] were just saying about your GP telling you.

P3: Well I wouldn't discuss it with my GP I don't think. But I haven't got that kind of relationship with my GP and I always find that they are a bit general. You know, they are, aren't they?

P2: Yea well, it might have been, that was my position, you know, I mean you never see the same GP twice anyway so.

P3: No you don't.

P2: They don't, you know your notes are on, your history is on your notes but I don't think any of them would have thought "Right, let's have a look what she's got in her family" and before they sort of gave me that advice.

M: So you didn't really talk to anyone about the surgery itself, did you?

P3: No.

M: Okay, right. Okay but if you, if you think about it now what are your questions? What would you want to know?

P3: My main thing would be the menopause, exactly the same as you [P1]. Would be that starting because every time I read about it on the internet everybody, because it happens gradually, your body sort of gets used to what is going on but where you have your ovaries out it literally starts straight away doesn't it? And I think that would be, for me anyway, the last couple of years hasn't really been an option because there has been other things to deal with and I haven't wanted to concentrate on myself.

P2: Come to that, yeah.

M: Mmm, yeah, yeah. So are you, so now that the screening programme has finished, it has finished for all of you has it?

P3: Yeah. / P2: Yes.

M: Right, okay. Do you find yourself starting to think about this decision more again?

P2: Yes. It's the first time I have ever given it any serious thought. Because wrongly or rightly I always felt that it was a little bit of a safety net...

P1: Yea.

*Decision Making
Considerations
(Other)
+
Misc
GP & Healthcare
system*

*Decision Making
Considerations
(Medical &
Physical)
+
Barriers*

*Decision Making
Considerations
(Social)*

*Decision Making
Considerations
(Other)*

Appendix 4.6 – Extract of Coded Transcript (Interview with Health Professional)

J: That's no problem, I was asking how women react when you tell them that...

R: ...that surgery is the option. Well, mixed reactions, some of them come – especially if they come through the genetics clinics – some of them, uhm, sort of have, you know, they are very clear there's the usual bell-shaped curve involved, yes? So this is the end of women who come from the genetics team and they yes, they already come knowing, they want surgery, so they hear the whole story, screening and surgery and everything, symptoms and everything, they say: "Yes, I think surgery is it." And that's pretty straight forward, it's easy. And then there are those that are in the sort of average, in the, the larger proportion that are: "Yes", you know, some are "Yes, yes...", but they are not sure, some are sort of really indecisive, some of them are just sort of well... And then there are those on this end where it's just not even something they are going to consider it's quite extreme, in fact there are one or two that I know who get upset when you... and then one of them has written to say "Please don't ask me this question again and could you let the hysteroscopy people know not to ask me when I come for my hysteroscopy, not, when I come for my hysteroscopy next year, don't raise the issue of surgery with me." So, you get the extreme reactions. Uhm, and then you got, have kind of the majority in this middle bit, where just a little bit of perhaps some peer-support or another visit would sort of tip them over into having surgery. And then there are some that come for years and years, have lots of chats and they are, they're pretty resolute and I in a way kind of respect that because you have, you know, it's a mature person, you have a sense of their, they have weighed it up and for them, you know what, they are not that worried about cancer, even though they know they are high risk and they are coming for their screens, they are comfortable with that and feel that perhaps, you know: "As long as I am healthy, why should I have a general anaesthetic, just happy to have a scan when I can." And so there is a mixture, quite a broad spectrum of reactions really and responses to this idea of prophylactic surgery.

J: And what are generally the questions or kind of concerns women raise when you start talking about this kind of issue?

R: Uhm, well, for the younger ones it has to do with the menopause, uhm, and what it would mean for them, what the implications are, what the menopausal symptoms are like to be, they want to be reassured that HRT will make the symptoms, you know, control the symptoms, that they won't be living in discomfort or distress as a result of having their ovaries out. Uhm, for that younger group the sort of most difficult will be if you like, are those who have had breast cancer in the past, that are hormone recep-, are hormone receptive, uhm, because obviously they can't have HRT. And some of them, or

Decision Making Process

Women's Needs Emotional + Decision Making Facilitator

Decision Making Barrier

Decision Making Considerations (Medical & Physical)

Women's Needs Group and Time Differences

all of them are really dis-, can be distressed and anxious, you know, because they've, they've had breast cancer young, they don't want to have ovarian cancer, but what are they going to do at 41 or 42, if they can't have HRT? Uhm, and so what we would do in that scenario we will talk a lot with the breast team and just kind of support the person as far as we can really. Uhm, and so then the – sorry, and so the question was? [laughs]

J: What women's questions were, so...

R: Yea, so their questions.

[Talk over each other - inaudible]

Menopause, HRT for the younger ones. And for the sort of over 45s, again menopause is an issue obviously, but we reassure them that, you know, HRT is kind of optional for them, because the mortality impact is in the younger women if they are not on HRT. Uhm, and they kind of want to know, uhm, you know, I suppose they all really in their 40s just want to be reassured from the breast cancer side of things. So, as soon as you say "HRT" then the next question that kind of comes up is: "Well, I thought HRT increases your risk of breast cancer" or "Isn't it associated with breast cancer?" And so you have to kind of unravel and undo a lot of the stuff the stuff they have heard in the media, whatever...

J: Yea.

R: ... and contextualise it. Uhm, and just explain, you know that data is really in post-menopausal women on HRT. And even sort of explain that even in post-menopausal women on HRT uhm, you know, the number – the extra cases of breast cancer – is actually, the risk is just a marginally increased risk, it's not a huge increased risk. Uhm, so those are the main questions for the younger women. But the older women, one or two will still ask: "Will I start, will my hot flushed come back?" or something like that. [Laughs]

J: [Laughs] They wouldn't, would they?

R: No, they wouldn't. [Laughs] No, they wouldn't.

J: [Laughs]

R: No, they wouldn't. And so you just say: "No, actually you have been menopausal for the last three years – x number of years – you probably will feel exactly the way you feel today. We wouldn't expect any changes in that direction." But those are the main questions. It's usually around menopause obviously and the use of HRT and the control of menopausal symptoms.

Women's Needs
Group and Time
Differences

Decision Making
Considerations
(Medical &
Physical)

Women's
Needs
Media Hype

Women's Needs
Group and Time
Differences

Appendix 5

| | | |
|-----|--|-----|
| 5.1 | <i>Topic Guide for Interview (Retrospective account)</i> | 438 |
| 5.2 | <i>Coding Frame based on CODE Framework</i> | 439 |
| 5.3 | <i>Extract of Coded Transcript (Interview)</i> | 443 |

Appendix 5.1 - Topic Guide for Interview (Retrospective account)

Explain purpose of interview and of wider study

Ask whether participant has any questions before we start the interview

How did you make the decision to have the surgery? Could you walk me through it?

Prompts (if needed)

- What were the questions you were asking?
- What support did you have? (Family, friends, peers, professionals)
- How did you tell family / friends?
- What was your experience of the surgery itself?
- What is your life like now after the surgery? (worry, menopause, HRT, adjustment)
- Is there anything you wished you had known beforehand?

What are the kinds of support that you used and that you think women need when making this decision?

Prompts (if needed):

- What kind of support did you receive from genetics services?
- What do you think about peer support?
- Would extra materials be helpful?

What do you think of the idea of decision support materials for women making these decisions?

Prompts (if needed):

- What kind of format do you think would work best?

What should be covered by such materials? What would you have found useful?

Prompts (if needed):

- These could be things like 'how much will my risk be reduced?'
- or 'how will surgery affect my life?' and so on...
- anything you can think of that is really important and should be discussed

| Overarching theme | Code | Included in Code |
|-------------------------|---------------------|---|
| Participant information | Medical history | Anything about the patient’s medical history; including family history of cancer |
| | Other | Anything else about the patient (occupation, parity, age etc.) |
| Primary appraisal | Ovarian cancer risk | Anything about how the patient appraised / appraises ovarian cancer risk Anything relating to exploring / finding out about or interpreting: What is ovarian cancer (OC)? What is OC risk? How high is my risk? What caused me to have an increased risk of OC? How does my family history influence OC risk? Do I have a faulty gene? Does my risk change over time? How could my risk affect my children? How will my risk affect my life? What can be done about my OC risk? |
| | | Anything about how she felt /feels about ovarian cancer risk; Anything relating to exploring / finding out about, interpreting or appraising: What are my past experiences of OC? How do I feel about OC and my risk? |
| | Choice | Anything about how the patient appraised / appraises choice; Anything relating to exploring / finding out about or interpreting: What does ‘choice’ mean in this context? Why is there a choice for women at risk of OC? When should I decide? What is at stake in this choice? Who can decide? Who else should I involve? Can I change my mind? How can I decide? |
| | | Anything about how she felt /feels about choice; Anything relating to exploring / finding out about or interpreting: How do I feel about choosing? Can I deal with choosing myself? |
| | Options | Anything about how the patient appraised / appraises RRSO or symptoms awareness or screening; Anything relating to exploring / finding out about or interpreting: What is risk-reducing bilateral salpingo oophorectomy (RRSO) What does RRSO involve? How long would it take to recover from RRSO? What are the risks and benefits of RRSO / symptom awareness? What are the complications of RRSO? Would I go into an early menopause? Would I have to take hormone replacement? How would RRSO affect my life? Do I need to have my womb removed as well? Is there a chance cancer might be found during RRSO? What did others decide? How did they fare? Can I make sure I get a good surgeon in a good hospital? What is symptoms awareness? Why is symptom awareness the only alternative? What would happen if I do not |

| | | |
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| | | have surgery? Is there any screening available for OC? Is there anything else I can do to avoid OC? |
| | | Anything about how she felt /feels about RRSO or symptoms awareness or screening; Anything relating to exploring / finding out about or interpreting: Do I have experiences that could help me imagine what it would be/feel if I had / didn't have RRSO? How do I feel about having / not having RRSO? |
| | Preference construction | Anything about how the patient differentiated / differentiates one option from the others, forming her preference; Anything relating to exploring / finding out about or interpreting: Is this the right time to decide? Is RRSO / not having RRSO congruent with my/my family's/partner's/doctor's beliefs goals and values? Have I completed my family? Can I afford to take time off? Could I get screening or some other additional care? Do I think I could detect the symptoms of OC? |
| | | Anything about how she felt when she was differentiating one option from the other, forming her preference; Anything relating to exploring / finding out about or interpreting: Do I feel ready to decide? How likely is it that I will regret having / not having RRSO? |
| Secondary appraisal and coping resources used | Ovarian cancer risk | Anything about problem-focused coping resources she explored / used when she appraised / appraises ovarian cancer risk; Can I find out more? Can my doctor help and can I trust my doctor? Can I seek instrumental / informational support from my family / friends / a peer or support group? |
| | | Anything about emotion-focused coping resources she explored OR used when heard about cancer risk; Can I seek emotional support from my family / friends / a peer or support group? Can I turn to my faith? Can I change how I feel about knowing my OC risk? Can I express my fear of cancer? Can I do something to stop thinking about my OC risk? Can I do something to reduce the stress my OC risk is causing me? Could I ignore my OC risk? |
| | Choice | Anything about problem-focused coping resources she explored / used when she appraised / appraises choice; Can I create an action plan to approach this decision? Can I seek instrumental / informational |

| | | |
|-------------------------|---------------------------------------|---|
| | | support from my family / friends / a peer or support group? |
| | | Anything about emotion-focused coping resources she explored OR used when faced with the choice; Can I seek emotional support from my family / friends / a peer or support group? Can I let someone else decide for me? Could I refuse to choose and just not make this decision? |
| | Options and preference construction | Anything about problem-focused coping resources she explored / used when she appraised / appraises options and when opted for surgery; Is there someone or something that could help me to compare and weigh my options? Can I speak to someone who has already made this decision and is living with the consequences? Can I seek instrumental / informational support from my family / friends? |
| | | Anything about emotion-focused coping resources she explored OR used when considering options and when opted for surgery; Can I seek emotional from my family / friends / a peer or support group? Can I do something to stop thinking about this decision? Can I do something to reduce the stress this decision is causing me? |
| Post-surgery | Experience | Anything relating to her surgery and post-surgery experience (e.g. HRT experience, complications) or what she did after surgery |
| | Coping behaviours | Anything about problem/emotion-focused coping resources she used AFTER having the surgery; anything about things that helped her cope; anything about why she still thinks surgery was the right option |
| Service | | Anything about the service she received from genetics and other healthcare facilities |
| Decision support | Scope for support | Where and when support is needed (timing of decision aid, setting for decision aid, i.e. inside or outside clinic), who needs most support; comments about target audience; change of scope for support due to change in what is offered (e.g. no screening available) |
| | Decision aid content (information) | Any comments about what should be included in a decision aid regarding information (on cancer, risk, options etc.), contact details, references; level of detail for information; evidence base, format for information (e.g. option grid / table) |
| | Decision aid content (coping and VCE) | Any comments regarding content for value clarification exercise, preference elicitation, coping advice and emotional support |

| | | |
|-------------------------|-----------------------|--|
| | Decision aid format | Remarks about the best / worst format (e.g. web-based vs. paper) for the decision aid, reasons why format is good / bad; remarks about colour, font type / size etc.; sub-formatting, e.g. blank spaces, interactivity |
| Misc | Participant questions | Any questions asked by participants |
| | Misunderstandings | Any misunderstandings / clarifications due to miss-understandings |
| General decision making | | Anything describing the complete process of decision making rather than individual aspects of it |
| Related decisions | Mastectomy | Anything about decision making regarding mastectomy and how this related to / differs from decision making about RRSO |

Appendix 5.3 - Extract of Coded Transcript (Interview)

- R: ...yes yeah, talking about the statistics of it we talked a lot about em, you know, statistically em, how likely it is that I would em, if I was BRCA 1 or not BRCA 1 you know, we talked about all the statistics of that em and then once we'd sort of decided, that you know I've got the result, I was BRCA 1, we'd, we'd sort of done all the talking almost you know and we were just at the stage of like well, decision making now really, Oh I know what I was going to say, yeah, the other thing that I did talk to them a lot about in the lead up to the operation was em, how to address it to other family members because that I, I've, I still find that quite hard because em, my parents weren't together, so my dad em, didn't see the whole thing as it unfolded with regards to my mum, you know although he was around, he didn't you know live with it and he didn't see how awful it was and stuff and so I think it's more difficult for him to understand where I'm coming from because, and I think that for anybody who hasn't actually been through it because I know I've got friends who are, just look at me and they just think well this all seems a bit kind of crazy bit kind of radical em, you know because they all sort of say, well, you know, you don't know what's going to happen to you in the future but then if you are given the statistics that you know, say, say somebody gave you the statistic that, statistic, you're statistically likely, eighty five per cent likely to be run over by a bus or something today you might think, I'll just stay in ((laughter)) you know and that's the way I'm sort of thinking about it but it's hard for, it's hard for people when they haven't been through it, I think, you know, to em understand, you know, understand where I'm coming from so I talked to them a lot about that and they gave me a really helpful letter em, which I gave out to family members and I know that they, that my family could have contacted them if they'd wanted to and, and I'm, sometimes I still wonder if my dad perhaps did contact them 'cos I don't know, he did have a bit of a change of heart whether he did phone them and talk to them or whether he just did you know sort of do a bit of research himself and then thought you know, but I think that is quite difficult, to try and, 'cos you need the support of your family and friends em and you don't want people to just think, 'oh, god, is she still banging on about this' d'you know what I mean, you know, you do need the support, and you don't want people to think that you're just trying to make something out of nothing you know em and, you know because some people do sort of say, yeah, but everybody's got something that they to have to deal with and I think that's absolutely right you know some people have heart conditions, some people have diabetes but you wouldn't not do something if you...
- J: Knew it beforehand
- Prim. Appraisal*
OC risk
- Prim. Appraisal*
Choice
- Prim. Appraisal*
OC risk
- Service*
- Second. Appraisal*
OC risk &
Choice

R: ...had the option, yeah yeah, and that's what I think is the difference you know em so yeah, we did talk a lot about that and I did find them very helpful in the sort of advice that they were give, giving and things

} Service

J: And did, did you tell your family er before you had the operations yeah

R: Yes, they all knew about it before and, and I think, I think I'd decided to have, like the you know we'd decided on the date and stuff in about July time and then I didn't have it until October so everybody had a lot of time to sort of, sort of em, well not to really do anything but just to know that I was going to have it done and that was the decision made and I think the same with the mastectomy you know em er, I think again, that's more radical so it's harder em and we haven't actually told all our family about it yet because I, we just felt, actually we need to make the decision ourselves without, sort of, any other influences and then sort of say what the situation is.

} Second. Appraisal Options and Preference Construction

J: So, 'we', is that you and your husband?

R: Me and my husband yeah me and my husband I mean I have talked to my brothers about it em and I, I do speak to [Name cousin] my cousin about it so obviously we're all totally on the same page em and my dad and you know our outside family are very supportive it's not that they're not, they are, I mean they're very kind and they, you know they want the best for, you know, for, for us em, but I think you know it is, it's harder for people who haven't directly seen the effects of ovarian and breast cancer on like, an individual person and then how it affects the whole family it's difficult for them to understand perhaps where we're coming from you know and I think there is the sort of, the, the mind set of like, why don't you just enjoy your life and see what happens you know and I can understand that because I've sort of you know thought that myself, let's just kind of, let's just, sort of, put our heads down and just enjoy life you know ((coughs)) er, you know and just you know I, it's just whether you really can do that or whether it would just always be at the back of your mind you know so

} Prim. Appraisal OC risk

} Second. Appraisal OC risk & Choice

J: Yes, as you said earlier and then you, you, you would have been worrying and also you did er say that you found that lump which at which point you weren't actually going to do anything but then you found the lump and that kind of changed changed your attitude

R: Yeah, it did, and, and again, with the mastectomy thing em, I mean I know we're not, you know that's not what the, the thing is about but with the, you know talking about making decision for the mastectomy now em, I think I thought after the hysterectomy, no, I'm not going to have a mastectomy I'll just em, you know just have a, an MRI ...

} Participant Info Medical history

Appendix 6

| | | |
|-----|--|-----|
| 6.1 | <i>Data Extraction form based on IPDAS checklist</i> | 446 |
| 6.2 | <i>Data Extraction form based on RRSO-adapted CODE framework</i> | 450 |

Appendix 6.1 - Data Extraction form based on IPDAS checklist

Developer & associated publications:

Intervention title:

Target audience:

Decision:

| Aspect | | Yes | No |
|----------|---|-----|----|
| 1 | Format | | |
| a | Paper-based | | |
| b | Web-based | | |
| c | DVD-based | | |
| d | Other | | |
| e | Used independently (outside clinical encounter) | | |
| f | Used together or by professional (inside clinical encounter) | | |
| 2 | Accessibility | | |
| a | Available freely online (as website or to download) | | |
| b | Available freely as leaflet | | |
| c | Available through practitioners | | |
| d | Currently in trial | | |
| e | Other: | | |
| 3 | Information about decision making | | |
| a | Explanation of preference-sensitive decisions and why choice exists | | |
| b | Information on making 'good' decisions (e.g. how values affect decisions) | | |
| c | Information on options for involvement in decisions | | |
| d | Information on making choices under stress or anxiety | | |
| e | Information on coping strategies | | |
| f | Explanation / list of options available | | |
| g | Other: | | |
| 4 | Information about the health condition [ovarian cancer] | | |
| a | Information about the function of ovaries | | |
| b | Information about ovarian cancer (general information about the disease) | | |
| c | Information about ovarian cancer (symptoms) | | |
| 5 | Information about the health condition [increased ovarian cancer risk] | | |
| a | Risk explained (in words) | | |
| b | Risk explained (in numbers/probabilities) | | |
| c | Risk explained (in pictures/diagrams) | | |
| d | Explanation of familial/hereditary cancer | | |

| | | | |
|---|---|--|--|
| e | Information about genetic testing | | |
| 6 Information about option 1 [risk-reducing bilateral salpingo-oophorectomy] | | | |
| a | Information about what option entails (e.g. surgical procedures) | | |
| b | Information about recovery time and post-operative period | | |
| c | Information about surgical menopause | | |
| d | Information about hormone replacement therapy (HRT) | | |
| e | Information about infertility | | |
| f | Information about psychosocial issues (e.g. femininity, sexuality) | | |
| g | Information about residual risk after surgery (in words) | | |
| h | Information about residual risk after surgery (in numbers/probabilities) | | |
| i | Information about residual risk after surgery (in pictures/diagrams) | | |
| j | Risks / Negative features explained (in words) | | |
| k | Risks / Negative features explained (in numbers/probabilities) | | |
| l | Risks / Negative features explained (in pictures/diagrams) | | |
| m | Benefits / Positive features explained (in words) | | |
| n | Benefits / Positive features explained (in numbers/probabilities) | | |
| o | Benefits / Positive features explained (in pictures/diagrams) | | |
| 7 Information about option 2 [watchful waiting / symptom awareness] | | | |
| a | Information about what option entails (e.g. do nothing, knowing symptoms) | | |
| b | Information about symptoms (types, frequency, when to consult GP) | | |
| c | Risks / Negative features explained (in words) | | |
| d | Risks / Negative features explained (in numbers/probabilities) | | |
| e | Risks / Negative features explained (in pictures/diagrams) | | |
| f | Benefits / Positive features explained (in words) | | |
| g | Benefits / Positive features explained (in numbers/probabilities) | | |
| h | Benefits / Positive features explained (in pictures/diagrams) | | |
| 8 Information about option 3 [ultrasound and CA125 screening] | | | |
| a | Information about what option entails (e.g. scans, blood tests) | | |
| b | Risks / Negative features explained (in words) | | |
| c | Risks / Negative features explained (in numbers/probabilities) | | |
| d | Risks / Negative features explained (in pictures/diagrams) | | |
| e | Benefits / Positive features explained (in words) | | |
| f | Benefits / Positive features explained (in numbers/probabilities) | | |
| g | Benefits / Positive features explained (in pictures/diagrams) | | |
| 9 Information about option 4 [oral contraceptive pill] | | | |
| a | Information about what option entails (e.g. taking oral contraceptives) | | |
| b | Risks / Negative features explained (in words) | | |
| c | Risks / Negative features explained (in numbers/probabilities) | | |
| d | Risks / Negative features explained (in pictures/diagrams) | | |
| e | Benefits / Positive features explained (in words) | | |

| | | | |
|-----------|--|--|--|
| f | Benefits / Positive features explained (in numbers/probabilities) | | |
| g | Benefits / Positive features explained (in pictures/diagrams) | | |
| 10 | Information about option 5 [other:] | | |
| a | Information about what option entails | | |
| b | Risks / Negative features explained (in words) | | |
| c | Risks / Negative features explained (in numbers/probabilities) | | |
| d | Risks / Negative features explained (in pictures/diagrams) | | |
| e | Benefits / Positive features explained (in words) | | |
| f | Benefits / Positive features explained (in numbers/probabilities) | | |
| g | Benefits / Positive features explained (in pictures/diagrams) | | |
| 11 | General assessment of information presentation | | |
| a | Presentation of positive <u>and</u> negative frames for probabilities | | |
| b | Probabilities placed in the context of other events / risks | | |
| c | Acknowledgement of uncertainty around probabilities | | |
| d | Procedures and outcomes described in a way to help imagine the effects | | |
| e | Overall, are probabilities presented in a balanced, unbiased way? | | |
| f | Overall, is information provided within sufficient detail? | | |
| 12 | Value clarification | | |
| a | Information / step-by-step guidance to making the decision | | |
| b | Are patients asked to consider which positive/negative features matter most? | | |
| c | Explicit value clarification exercise included | | |
| d | i. Tallying (weighing of options) | | |
| e | ii. Ranking (in order of importance, "take the best") | | |
| f | iii. Scoring different attributes (on scales) | | |
| g | iv. Other: | | |
| h | Instructions on how to use the value clarification exercise | | |
| 13 | Communication aid | | |
| a | Recommends / helps with communication with practitioners | | |
| b | Recommends / helps with communication with family/friends | | |
| 14 | Personalisation | | |
| a | Personalisation of probabilities possible (e.g. by age, other risk factors etc.) | | |
| b | Personalisation of value clarification possible (e.g. enter own reasons) | | |
| 15 | Additional outputs | | |
| a | Space for personal notes | | |
| b | Printable version | | |
| c | Pictures and/or audiovisual material | | |
| d | Personal stories (written) | | |
| e | Personal stories (audio/visual) | | |

| | | | |
|-----------|---|--|--|
| f | Links and/or references to research evidence | | |
| g | Target audience clearly stated | | |
| h | Purpose of tool clearly stated | | |
| i | Other: | | |
| | | | |
| 16 | Usability | | |
| a | Use of plain language (i.e. minimal use of jargon / scientific terms) | | |
| b | Glossary included | | |
| c | Contents page | | |
| d | Knowledge check / exercise | | |
| | | | |
| 17 | Design and Evaluation | | |
| a | Theory base stated | | |
| b | Evaluation before publication | | |
| c | Version / Date last updated | | |
| d | Date for next update | | |
| | | | |
| 18 | Effectiveness | | |
| a | Helps patients to recognise a decision needs to be made | | |
| b | Helps patients to know their options and their features | | |
| c | Helps patients to understand that values affect decisions | | |
| d | Helps patients to be clear about option features that matter most to them | | |
| e | Helps patients to discuss values with their practitioners | | |
| f | Helps patients to become involved in preferred ways | | |
| | | | |
| 19 | Additional comments | | |
| | | | |

This assessment framework was adapted from the International Patient Decision Aid Standards Checklist, available at:

<http://www.decisionlaboratory.com/resources/IPDAS%20Criteria%20Checklist.pdf>

| RRSO-adapted CODE framework assessment form – tick box if question is addressed within the decision aid | | | | DA name: _____ |
|---|--|---|--|--|
| | Disclosure of ovarian cancer risk status | Presentation of choice | Presentation of options (RRSO or symptom awareness) | Preference construction |
| Cognitive | What is ovarian cancer (OC)? <input type="checkbox"/> What is OC risk? <input type="checkbox"/> How high is my risk? <input type="checkbox"/> What cause me to have an increased risk of OC? <input type="checkbox"/> How does my family history influence OC risk? <input type="checkbox"/> Do I have a faulty gene? <input type="checkbox"/> Does my risk change over time? <input type="checkbox"/> How could my risk affect my children? <input type="checkbox"/> How will my risk affect my life? <input type="checkbox"/> What can be done about my OC risk? <input type="checkbox"/> | What does 'choice' mean in this context? <input type="checkbox"/> Why is there a choice for women at risk of OC? <input type="checkbox"/> When should I decide? <input type="checkbox"/> What is at stake in this choice? <input type="checkbox"/> Who can decide? <input type="checkbox"/> Who else should I involve? <input type="checkbox"/> Can I change my mind? <input type="checkbox"/> How can I decide? <input type="checkbox"/> How do I feel about choosing? <input type="checkbox"/> Can I deal with choosing myself? <input type="checkbox"/> | What is risk-reducing bilateral salpingo oophorectomy (RRSO) / symptoms awareness? <input type="checkbox"/> Why is symptom awareness the only alternative? <input type="checkbox"/> What does RRSO involve? <input type="checkbox"/> How long would it take to recover from RRSO? <input type="checkbox"/> What would happen if I do not have surgery? <input type="checkbox"/> What are the risks and benefits of RRSO / symptom awareness? <input type="checkbox"/> What are the complications of RRSO? <input type="checkbox"/> Would I go into an early menopause? <input type="checkbox"/> Would I have to take hormone replacement? <input type="checkbox"/> How would RRSO affect my life? <input type="checkbox"/> Do I need to have my womb removed as well? <input type="checkbox"/> Is there a chance cancer might be found during RRSO? <input type="checkbox"/> Is there any screening available for OC? <input type="checkbox"/> Is there anything else I can do to avoid OC? <input type="checkbox"/> What did others decide? How did they fare? <input type="checkbox"/> Can I make sure I get a good surgeon in a good hospital? <input type="checkbox"/> Do I have experiences that could help me imagine what it would be/feel like if I had / didn't have RRSO? <input type="checkbox"/> How do I feel about having / not having RRSO? <input type="checkbox"/> | Is this the right time to decide? <input type="checkbox"/> Is RRSO / not having RRSO congruent with my/my family's/partner's/doctor's beliefs goals and values? <input type="checkbox"/> Have I completed my family? <input type="checkbox"/> Can I afford to take time off? <input type="checkbox"/> Could I get screening or some other additional care? <input type="checkbox"/> Do I think I could detect the symptoms of OC? <input type="checkbox"/> Do I feel ready to decide? <input type="checkbox"/> How likely is it that I will regret having / not having RRSO? <input type="checkbox"/> |
| Primary Appraisal | What are my past experiences of OC? <input type="checkbox"/> How do I feel about OC and my risk? <input type="checkbox"/> | | | |
| Emotional | | | | |
| Problem-focused | Can I find out more? <input type="checkbox"/> Can my doctor help and can I trust my doctor? <input type="checkbox"/> Can I seek instrumental / informational support from my family / friends / a peer or support group? <input type="checkbox"/> Can I seek emotional support from my family / friends / a peer or support group? <input type="checkbox"/> Can I turn to my faith? <input type="checkbox"/> Can I change how I feel about knowing my OC risk? <input type="checkbox"/> Can I express my fear of cancer? <input type="checkbox"/> Can I do something to stop thinking about my OC risk? <input type="checkbox"/> Can I do something to reduce the stress my OC risk is causing me? <input type="checkbox"/> Could I ignore my OC risk? <input type="checkbox"/> | Can I create an action plan to approach this decision? <input type="checkbox"/> Can I seek instrumental / informational support from my family / friends / a peer or support group? <input type="checkbox"/> Can I seek emotional support from my family / friends / a peer or support group? <input type="checkbox"/> Can I let someone else decide for me? <input type="checkbox"/> Could I refuse to choose and just not make this decision? <input type="checkbox"/> | Is there someone or something that could help me to compare and weigh my options? <input type="checkbox"/> Can I speak to someone who has already made this decision and is living with the consequences? <input type="checkbox"/> Can I seek instrumental / informational support from my family / friends? <input type="checkbox"/> Can I seek emotional from my family / friends / a peer or support group? <input type="checkbox"/> Can I do something to stop thinking about this decision? <input type="checkbox"/> Can I do something to reduce the stress this decision is causing me? <input type="checkbox"/> | |
| Secondary Appraisal | | | | |
| Emotion-focused | | | | |

Appendix 7

| | | |
|-----|---|-----|
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Appendix 7.1 - Coding scheme Focus Groups / Interviews
[Decision support sub-theme]

| | | |
|-------------------------|---|--|
| Decision support | Scope for support | Where and when support is needed (timing of decision aid, setting for decision aid, i.e. inside or outside clinic), who needs most support; comments about target audience; change of scope for support due to change in what is offered (e.g. no screening available) |
| | Decision aid content (information) | Any comments about what should be included in a decision aid regarding information (on cancer, risk, options etc.), contact details, references; level of detail for information; evidence base, format for information (e.g. option grid / table) |
| | Decision aid content (coping and value clarification) | Any comments regarding content for value clarification exercise, preference elicitation, coping advice and emotional support |
| | Decision aid format | Remarks about the best / worst format (e.g. web-based vs. paper) for the decision aid, reasons why format is good / bad; remarks about colour, font type / size etc.; sub-formatting, e.g. blank spaces, interactivity |
| | Decision aid purpose | Purpose of the decision aid, any comments about the importance of a decision aid for this context, main/minor purpose |
| | Other areas for decision support | Any comments about other areas for decision support, changes in counselling content, timing, opportunities for peer-support etc. |
| | Problem areas | Any comments regarding the current lack of support / information / trust; the way information / support is given; awareness of the need for a decision |
| | Potential effects of a decision aid | Remarks about the potential effects of the decision aid on patients, clinicians, individually or together; on interactions in- or outside the clinic |
| | Recommendations for decision aid development | Any comments regarding the development process; who should be involved; why; how it could be supported / designed |
| | Clinician attitude to options (professionals ONLY¹) | Any remarks on what women should do, what the medically preferred option is, whether options work |

Appendix 7.2 - Reviewer guide for Virtual Reference Group

**Below you can find a few questions that might be helpful when you give us feedback.
Please make sure you explain why you answered yes or no to any of the questions:**

Please let us know if you chose to personalise OvDex: Yes / No

If yes, which version did you view (please tick):

- BRCA1
- BRCA2
- Lynch syndrome
- Not tested / Unclear genetic test
- Negative genetic test

Was the website easy to use? Why / why not? (Please ignore if you did not view the decision aid online) Did you have any problems using it?

Did the information cover the right topics in the right amount of detail? Why / why not? Please give examples.

Was the information easy to understand? Why / why not? Please give examples.

Was the information helpful? Why / why not? Please give examples.

Was there any information that you did not know about before reading OvDex? What information was this?

Was any important information missing? What information was this?

Was any irrelevant information included? What information was this?

What did you think of the exercise to weigh up different factors of the decision? Was this useful? Why / Why not?

Any other comments and suggestions:

Appendix 7.3 - OvDex booklet prototype I (General version)



Welcome to OvDex

OvDex (The Oophorectomy Decision Explorer) has been developed to help you find out more about your options for reducing your ovarian cancer risk. If you are viewing OvDex you should have been referred to it by a doctor or geneticist as you are at increased risk of ovarian cancer.

Please note that OvDex can be personalised by answering three questions . You should have seen these questions before you opened these pages. If you do wish to personalise the information you get, please go back and answer the questions .

The information on the following pages has not been personalised. If you do not wish to personalise OvDex then please view the general information on the following pages.

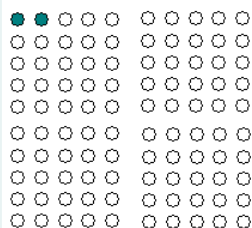
Questions in this chapter:

- What is the ovarian cancer risk if I have a family history of ovarian cancer?
- How can I find out whether I have a faulty gene?
- What would my risk be if a faulty gene is found?
- What if I cannot get tested or my genetic test is unclear?
- Does my cancer risk change over time?
- How will my ovarian cancer risk affect my life?
- How will my ovarian cancer risk affect my children?
- What can I do to reduce my risk?

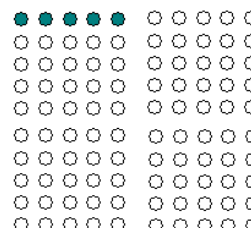
What is the ovarian cancer risk if I have a family history of ovarian cancer?

Ovarian cancer occurs by chance in less than 2 of every 100 women. For women who have a family history and/or a faulty gene, this number can be much higher. Generally the risk is low in young women and rises with age.

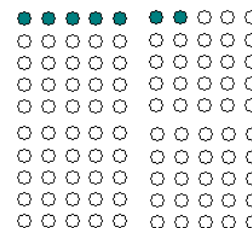
Studies have shown that out of 100 women with one first degree relative (mother, sister or daughter) with ovarian cancer, 5 will develop ovarian cancer themselves at some point in their life. Women who have more than one relative with ovarian cancer have an even higher chance with about 7 out of 100 women with 2 or more relatives developing ovarian cancer themselves in their lifetime.



Less than 2 out of 100 women in the general population get ovarian cancer in their lifetime



About 5 out of 100 women with a first degree relative with ovarian cancer get ovarian cancer in their lifetime



About 7 out of 100 women with two or more relatives with ovarian cancer get ovarian cancer in their lifetime

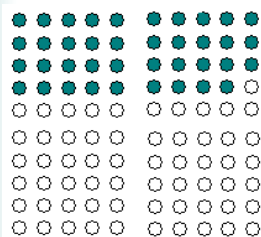
How can I find out whether I have a faulty gene?

Some women might come from families where cancer is even more common and these women might have a faulty gene. Testing for a faulty gene in the family usually begins with a test on a blood sample from a person who has had cancer. If a faulty gene is found, other members of the family can have a genetic test to see if they also carry the same gene.

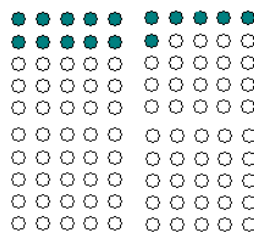
What would my risk be if a faulty gene is found?

The most common faulty genes linked to ovarian cancer are called Breast Cancer (BRCA) genes 1 and 2 and genes common in Lynch Syndrome (formerly known as Hereditary Non-Polyposis Colorectal Cancer or HNPCC). The lifetime risk of developing ovarian cancer can be much higher for women who have a faulty gene compared to the general population.

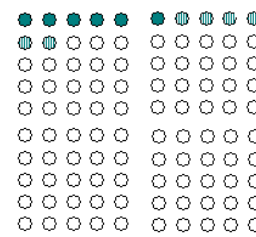
Cancer risk



About 39 out of 100 women with a BRCA1 faulty gene get ovarian cancer by the time they are 70



About 16 out of 100 women with a BRCA2 faulty gene get ovarian cancer by the time they are 70



About 6 to 12 out of 100 women with a HNPCC faulty gene get ovarian cancer by the time they are 70

What if I cannot get tested or my genetic test is unclear?

For some women it might not be possible to get tested because they have no living relative with cancer or their relative does not want to be tested. In other cases a genetic test may come back as 'unclear' which means the result is not certain. In this case the genetics service will estimate the woman's risk based on her family history.

If you have been told that your family history looks like you might have a faulty gene and you would like more specific information for people with that gene, you can look at the information by personalising this aid and selecting the gene that most closely resembles your family history as you have been told by your genetic counsellor.

Does my cancer risk change over time?

Your so-called 'lifetime' risk (which is the risk that you will get ovarian cancer at some point in your life) stays the same over time. However, your 'age-related' ovarian cancer risk (which is the risk that you will get ovarian cancer in a specified time frame, for example one year or five years) increases with age. This means when you are young your age-related risk is relatively low and rises when you get older. So for example, your risk of getting ovarian cancer in the next year when you are 35 is quite low, whereas your risk of getting ovarian cancer in the next year when you are 50 is higher.

How will my ovarian cancer risk affect my life?

Being at risk for ovarian cancer does not necessarily mean that you will develop ovarian cancer. However, knowing your risk will give you access to additional medical services such as genetic counselling and means that you might be offered risk-reducing surgery. Some people may feel worried or anxious after finding out about their risk and their quality of life may be affected.

How will my ovarian cancer risk affect my children?

There is a chance that your children will also have an increased risk of ovarian cancer. You can discuss your children's risk with your doctor to find out more. Your doctor will also be able to advise you about how best to discuss this risk with your children.

What can I do to reduce my risk?

Your doctor has probably discussed with you the option of having risk-reducing ovarian surgery to reduce your ovarian cancer risk. This decision aid is designed to help you look at your options. Please refer to the sections 'Options at a glance', 'Risk-reducing surgery' and 'Other options' for more information.



The Choice

Questions in this chapter:

What can I do to reduce my risk?

Why is there a choice?

Who should decide?

Who else should I involve in this decision?

When should I decide?

How can I decide?

What can I do to reduce my risk?

As you are at increased risk of ovarian cancer, you have the option of undergoing risk-reducing surgery to remove your healthy ovaries (see 'options at a glance' and 'risk-reducing surgery'). You will need to decide if and when to have this surgery. Unfortunately, there is no screening for ovarian cancer at the moment (see 'Other options'), therefore the only other choice is 'watchful waiting'.

Why is there a choice?

Often when you go to your doctor, there is one clearly recommended treatment. However, some situations are more complicated. These are situations in which your personal preferences play an important role. In the case of cancer risk reduction, your options have very different effects on your life. This means that you need to be clear about what might happen if you choose one option over the other.

Who should decide?

As the best choice for you is based on your preferences, you should be closely involved in the decision. You can either make the decision on your own or if you do not wish to make this decision yourself, your doctor can help you. They will encourage you to think about the options and your preferences, so that the final choice is right for you. If you then don't want to choose for yourself, just say so and your doctor may make a recommendation.

Who else should I involve in this decision?

Whether or not you would like to bring anyone else into this decision is your choice. Often it is helpful to speak to your partner, other members of your family or some friends who could work through this decision with you. Especially with a decision about risk-reducing ovarian surgery, the views of your partner can be important, so it is recommended that you speak to your partner and try to reach a decision together. Your doctor will also be happy for you to bring your partner along to appointments and to answer any questions they might have.



The Choice

When should I decide?

The decision you are facing is not an easy one and you should not feel under any pressure to decide quickly. Risk-reducing surgery has benefits and risks that need to be weighed carefully, so take your time and make sure you are ready before making a decision. It is important that you make sure that you have completed your family before you make this choice.

How can I decide?

When it comes to important decisions everyone is different. Some people like to find out as much as they can about their options, while others prefer to just know what is absolutely necessary. Some might find it helpful to talk to their family and friends. Some might like to speak to people who have made a similar decision. It really depends on you. Have a think about other important decisions in your life and how you managed to make those. That could give you an idea of how you like to decide about things.

Options at a Glance

| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|---|---|---|
| Will this reduce my ovarian cancer risk? | Yes, removing the ovaries will greatly reduce your lifetime risk of ovarian cancer | No, your risk of ovarian cancer will remain increased |
| Will I be able to become pregnant? | No. | Yes. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. | You may worry about developing ovarian cancer. |
| Will I go into early menopause? | Yes, your oestrogen levels fall and the menopause will start immediately. | No. |
| What is menopause after surgery like? | Menopause after surgery is similar to natural menopause, but because it happens suddenly, the symptoms may be more severe. | Not applicable. |
| Will this change how I feel about myself as a woman? | Most women do not notice any change in how they feel about themselves as a woman. Your desire for and/or enjoyment of sex may decrease a lot. | Not applicable. |
| Are there any risks linked to early menopause? | Yes, there is a risk of bone thinning (osteoporosis). Some patients report memory changes as well. | Not applicable. |
| Will I need hormone replacement therapy (HRT)? | HRT can reduce some of the effects of sudden menopause. You will need to discuss this possibility with your doctor. | Not applicable |
| Is HRT safe for women at increased familial risk? | HRT is safe to take for the majority of women until they are 50. It is not recommended for women who have had breast cancer themselves already. | Not applicable |
| What are the risks of surgery? | There is a small risk of complications depending on the type of surgical procedure (key hole or open surgery) you have. Discuss this with your doctor. | Not applicable. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after key hole surgery and are back to normal in 4 weeks. For open surgery this is slightly longer. | Not applicable. |
| Can I get screened for ovarian cancer? | No, after surgery there is no need for any screening because your risk will be low | No, there is no routine screening available for ovarian cancer. |

Risk-reducing Surgery

Questions in this Chapter:

- What is risk-reducing bilateral salpingo-oophorectomy (RRSO)?
- What are the main advantages of this operation?
- What are the main disadvantages of this operation?
- What is my risk after surgery?
- What does the surgery involve?
- How long does it take to recover?
- Do I need to have my womb removed as well?
- What are the complications of RRSO?
- Can women die from RRSO?
- Could cancer be found during the surgery?
- How would RRSO affect my life?

What is risk-reducing bilateral salpingo-oophorectomy (RRSO)?

Risk-reducing bilateral salpingo-oophorectomy (RRSO for short) is surgery to remove healthy ovaries and fallopian tubes to reduce the cancer risk. The term 'bilateral' means that the ovaries and fallopian tubes on both sides of the body are removed. The word 'salpingo-oophorectomy' means removal of the fallopian tubes and ovaries. The aim of the surgery is to remove these tissues before cancer develops.

What are the main advantages of this operation?

- 1) This operation will reduce your ovarian and fallopian tube cancer risk greatly, as these are completely removed during surgery. For women with a faulty BRCA1 gene cancer might still develop in the peritoneum, which is the lining of the pelvis and abdomen. This means RRSO will not prevent cancer in about 2 in every 100 women. This risk is about the same as the ovarian cancer risk in the general population.
- 2) For some women, not having to worry about ovarian cancer anymore comes as a great relief.
- 3) For the vast majority of women having RRSO could also reduce the risk of breast cancer.
- 4) Having the surgery will also prevent other, non-cancer related problems happening with the ovaries, such as cysts.

What are the main disadvantages of this operation?

- 1) You will no longer be able to become pregnant once you have had surgery, therefore it is important that you have completed your family before going for surgery.
- 2) After the operation you will immediately enter the menopause and may start to experience the typical menopausal symptoms, including hot flushes, mood swings, night sweats and vaginal dryness (Please see 'Surgical Menopause' for more details). Furthermore, you may be at increased risk of bone thinning and you may experience some memory changes. These effects might be reduced by hormone replacement therapy (Please see 'Hormone Replacement Therapy' for more details).
- 3) There is a small risk of complications during and after surgery.

Risk-reducing Surgery

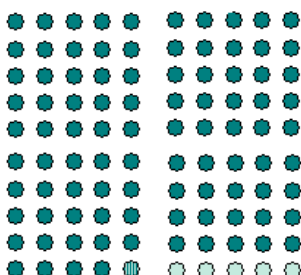
What is my risk after surgery?

After surgery your risk of ovarian and fallopian tube cancer is effectively eliminated. However, there is a rare type of cancer which may still develop after surgery that is very similar to ovarian cancer. This is called primary peritoneal cancer. Your risk of primary peritoneal cancer depends on whether or not you have a faulty gene. The highest risk for primary peritoneal cancer is in women with a BRCA1 faulty gene. Their risk is about 2 in every 100 women, which is similar to the risk of ovarian cancer in the general population. For all other women the risk of primary peritoneal cancer after surgery is close to zero.

What does the surgery involve?

Most often the operation is done as keyhole surgery (laparoscopy). This involves 3 or 4 small (about 1cm) cuts, usually one cut is close to your tummy button and 2-3 just below the bikini line, so they are not visible. Keyhole surgery itself usually takes less than 2 hours.

In some cases it may not be possible to do keyhole surgery, because of previous surgery on your tummy or because of your weight. Then surgeons use the more traditional open surgery. This means a longer cut, usually along the bikini line. Sometimes a surgeon might have to convert to an open surgery when doing keyhole surgery because of complications or old scar tissue. This happens in about 1 of every 100 keyhole operations.



Type of procedure in 100 women who have the operation

The figure on the left shows **how many of 100 women will have keyhole surgery, open surgery and planned keyhole which is converted to open surgery.**

94 of 100 women will have planned keyhole surgery (dark teal dots). Five of 100 women will have planned open surgery (light teal dots). One of 100 women will have planned keyhole surgery which is converted to open surgery (striped dot).

How long does it take to recover?

75 of 100 patients who have keyhole surgery leave the hospital the day after surgery. They are usually back to normal activity about 4 weeks after surgery. If you have open surgery you are likely to stay in hospital a bit longer than with keyhole surgery. Usually patients leave the hospital about 5 days after open surgery and are back to normal in about 6 weeks. After surgery you are not allowed to do any heavy lifting for a few weeks. You may also have to refrain from driving until you can comfortably wear a seatbelt and make an emergency stop without pain.

Do I need to have my womb removed as well?

Normally it is not recommended that you have your womb removed at the same time as having RRSO, unless you know that you have Lynch Syndrome or you have other problems with your womb and have been told by a doctor that removing the womb would help you with those problems.

Risk-reducing Surgery

What are the complications of RRSO?

There is a small risk of complications linked to RRSO. About 4 in every 100 people will experience some complication. Minor complications can include wound or urinary tract infections and usually have no long-term effects on your health. More serious complications might happen during surgery and can include damage to blood vessels, the bowel or the bladder. If you are having keyhole surgery this might mean that the surgeon has to convert to an open surgery to repair the damage.

There are a number of other rare complications that might happen and your surgeon will go through those with you if you wish before you go in for surgery.

Please note that about 96 of every 100 women do not experience any complications at all.



Complications in 100 women who have the operation

The figure on the left shows **how many of 100 women will experience complications** during or after surgery.

4 of 100 women will experience some kind of complication either during or after surgery. 96 of 100 women will not experience any complications.

Can women die from RRSO?

As with any surgery, there is a very small risk of death. However, this is highly unlikely. The risk may be greater in women with health problems before surgery.

Could cancer be found during the surgery?

Once your ovaries and fallopian tubes have been removed they will be checked thoroughly for any signs of cancer. There is a chance that cancer may be discovered during this check. This happens in up to 4 out of every 100 operations in the highest risk patients. For others the chance of finding cancer during the surgery are much lower.

How would RRSO affect my life?

In the short-term, if you decide to go for surgery, you will have to take time off work and will not be able to do some of the things you might usually do, such as driving or heavy lifting. However most women recover fully within 4 to 6 weeks.

In the longer term there are a number of things you should consider. After surgery you may feel less worried about ovarian cancer. You will no longer be able to become pregnant and you will enter the menopause if you have not gone through it yet (Please look at the 'Surgical menopause' section). You may opt to take hormone replacement (Please look at 'Hormone replacement therapy' section). These factors may affect your life after surgery and should be considered carefully before making a decision.



Surgical Menopause

Questions in this Chapter:

What is surgical menopause?

What is surgical menopause like?

How could it affect my life?

How long does surgical menopause last?

Are there other problems linked to surgical menopause?

Is there anything I can do to prevent the long term health effects of surgical menopause?

What is surgical menopause?

Surgical menopause is the menopause you enter when your ovaries are removed by surgery and your body no longer produces oestrogen. Similar to the natural menopause, which is when the ovaries naturally stop producing oestrogen, the surgical menopause can have a range of symptoms.

What is surgical menopause like?

It is very difficult to describe what surgical menopause is like, because it is different for every person, just like the natural menopause. Some women have no or very few symptoms and cope very well. Other women can have very severe symptoms and their quality of life can be affected.

Symptoms you may experience are: Hot flushes, night sweats, difficulty sleeping, fatigue, mood swings, weight gain, vaginal dryness, loss of interest in sex and changes in memory.

The most commonly reported symptoms of surgical menopause are hot flushes, night sweats, mood swings, vaginal dryness and loss of interest in sex. The great majority of women who had surgery report that they have experienced one or more of these symptoms. However, any data available about surgical menopausal symptoms is from small studies and it is difficult to predict what surgical menopause will be like for you personally.

How could it affect my life?

While none of the menopausal symptoms mentioned above are dangerous for your health, they may affect you in different ways. Hot flushes can be very uncomfortable and may happen at inconvenient times, for example when you are giving a presentation at work. In one study two out of every three women reported having hot flushes after surgery. Night sweats can lead to problems with sleep and insomnia. Vaginal dryness can lead to pain during sex and therefore you may enjoy sex less. Loss of interest in sex, which is also experienced by many women after surgery, may affect your relationship with your partner. In one study just over half of women reported that they were not satisfied with their sexual functioning after having surgery and in another study one in every three patients felt that vaginal dryness was bothersome and reported pain with sex. The way that these symptoms might affect your life depends on your personal situation.



Surgical Menopause

How long does surgical menopause last?

Like the natural menopause it is impossible to say how long symptoms will last. Some women have hardly any problems and symptoms disappear quickly, while others have symptoms for many years. It really is different for everyone.

Are there other problems linked to surgical menopause?

The loss of oestrogen before natural menopause can cause long term health problems, particularly bone thinning. This can lead to fractures in older age. Bone thinning is mainly a problem in women who have surgery when they are under 45 years old. Some women also report changes in memory following this surgery.

Is there anything I can do to prevent the long term health effects of surgical menopause?

Hormone replacement therapy (HRT) is usually recommended for women who have RRSO before the age of 45 and have not had breast cancer themselves (Please see 'Hormone replacement therapy' for more details).

If you do not wish to or cannot take HRT then you can take dietary supplements to reduce bone thinning. You should speak to your doctor about this. Patients are also recommended to have a bone density scan about 18 months after their operation. An active lifestyle and healthy diet is also recommended.



Hormone Replacement

Questions in this Chapter:

- What is Hormone Replacement Therapy (HRT)?
- Why is HRT important?
- Who should take HRT?
- How long should I take HRT?
- Can I take HRT if I have a family history of breast cancer?
- But I have heard that HRT increases breast cancer risk?
- Will HRT reduce all the symptoms of menopause?
- Are there any alternatives to HRT?

What is Hormone Replacement Therapy (HRT)?

Hormone Replacement Therapy, or HRT for short, is a medical treatment that puts back the female hormones that are lost when the ovaries are removed or when they stop functioning. There are a number of ways that HRT can be used:

- It can be taken as oral tablets usually once a day
- It can be applied as patches that are put on the tummy or bottom about once or twice a week
- It can be applied as a gel directly to the skin once a day (for example the lower abdomen or inner thigh)

Why is HRT important?

There are two main reasons why HRT is recommended after surgery:

1. To reduce the risk of bone thinning
2. To reduce symptoms of surgical menopause

Who should take HRT?

HRT is strongly recommended for women who have surgery before they are 45 years of age, as this group may experience bone thinning later in life as a result of the lack of hormones after surgery. This can lead to a risk of fractures in older age.

Women who have surgery when they are over 45 years of age can choose to have HRT to reduce the symptoms of surgical menopause. However these women do not have to have HRT if they do not want to, as the risk of bone thinning in later life in this group is reduced.

How long should I take HRT?

If you use HRT, it is recommended that you take it until the age of the natural menopause, which is 50 years in the UK.

Can I take HRT if I have a family history of breast cancer?

Yes, you should be able to take HRT even if you have a family history of breast cancer. Several studies have shown that HRT is safe to take for the vast majority of women with a family history of breast cancer as long as they have not had breast cancer themselves and they only use HRT until the age of the natural menopause.

But I have heard that HRT increases breast cancer risk?

The study that showed that HRT increases breast cancer risk was done with older women who had already gone through the natural menopause and were taking HRT beyond 50 years of age. These women did not have surgery to have their ovaries removed. Therefore this group of women is very different to women with a family history who choose to have their ovaries out before they are 50 years. Furthermore you should be aware that the findings of this study have now been widely criticized.

Having your ovaries out removes all the natural hormone that your ovaries would have produced until the age of the natural menopause. The amount of hormone that is added back by taking HRT is less than the amount your ovaries would have produced naturally. If you stop taking HRT at the age of the natural menopause (so when you are 50) then there is no evidence that the breast cancer risk is increased.

Will HRT reduce all the symptoms of menopause?

Taking HRT is not the same as having your own hormones. HRT cannot reduce all symptoms of the menopause and women who have had surgery and are on HRT do report more symptoms than women who have not had surgery. However, HRT can relieve some of the symptoms of the menopause. For example, women who took HRT reduced the average number of hot flushes from about 4 to about 1 per day when compared to women who did not take HRT. Women on HRT also had fewer night sweats.

Are there any alternatives to HRT?

There are numerous non-hormonal alternatives to HRT, none of which have been shown to be as effective in relieving menopausal symptoms. For people who cannot take HRT these alternatives may be a good option but for those who can, HRT is the best option.

Alternatives include:

- Antidepressants, such as Selective Serotonin Reuptake Inhibitors (SSRIs)
- Gabapentin
- Red Clover
- Ginseng
- Evening primrose oil
- Agnus Castus
- And many more

Some of these options may be prescribed by your doctor, while others can be bought in health shops or over the internet. However, there is no medical proof that freely available herbal options work. These alternatives to HRT may have side-effects and you should always consult your doctor before deciding to use any of these options.



Other Options

Questions in this Chapter:

Is there any screening available for ovarian cancer?

What is the alternative to surgery?

What would happen if I do not have surgery?

Is there anything else I can do?

Is there any screening available for ovarian cancer?

No, unfortunately there is no medically proven screening available for ovarian cancer. A large trial of ovarian cancer screening finished in December 2011 and the results will not be available until 2013. Until then the NHS will not offer routine screening. Some areas may offer private, self-funded screening, however there is still no proof that this screening is effective.

What is the alternative to surgery?

At the moment the only alternative to surgery is to do nothing and simply be aware of the symptoms of ovarian cancer should they develop. However it is important to realise that these symptoms can be very vague and are not specific to ovarian cancer. The symptoms include:

Constant bloating (big or swollen tummy)
 Feeling less hungry or feeling full quickly
 Constant pain in your tummy or below
 Needing the toilet more than usual

If any of these symptoms are prolonged, so last longer than usual, you should contact your GP.

What would happen if I do not have surgery?

If you decide not to have surgery, then nothing will really change. You will need to look out for any symptoms and if you think anything is wrong you need to go to your GP to get it checked. Make sure you tell your GP that you have a family history of ovarian cancer and are at increased risk when you talk to them.

Remember you can reconsider surgery at any time.

Is there anything else I can do?

As there is no screening available there is very little else you can do apart from keeping an eye on any symptoms. Studies have found that a healthy diet with plenty of fruit and vegetables, keeping a healthy weight and an active lifestyle can improve overall well-being and might reduce your chances of getting cancer.



Contacts and Resources

Here are some links and contact details that you might find helpful (even though we have taken great care in choosing websites from reliable sources please note we are not responsible for the content of these websites):

The Cancer Genetics Story Bank – An online collection of stories told by patients and professionals about cancer genetics, developed by the Cancer Genetics Service for Wales (CGSW)

www.cancergeneticsstorybank.co.uk

Target Ovarian Cancer – A UK charity which supports research into ovarian cancer and provides useful information including an 'Ask the Expert' feature. Available in several languages.

www.targetovariancancer.org.uk

Ovacome – A UK charity providing information and support for everyone affected by ovarian cancer. Includes a symptom awareness tool and links to a number of patient blogs.

www.ovacome.org.uk or call the helpline on 08453710554

Your GP and/or Genetics Service – There to help you with any questions or concerns.

Appendix 7.4 - OvDex prototype I (Negative test version)



Welcome to OvDex

OvDex (The Oophorectomy Decision Explorer) has been developed to help you find out more about your options for reducing your ovarian cancer risk. If you are viewing OvDex you should have been referred to it by a doctor or geneticist as you are at increased risk of ovarian cancer.

Please note that OvDex can be personalised by answering three questions. You should have seen and answered these questions before viewing these pages, as the information on the following pages has been personalised and is only relevant to women who have had a negative genetic test result (no faulty gene was identified).

(Information on the following pages relates to the answers indicated in bold writing)

1. Do you have a faulty gene or a family history of Lynch Syndrome?
 - a) Yes, I have a BRCA1 faulty gene
 - b) Yes, I have a BRCA2 faulty gene
 - c) Yes, I am from a Lynch Syndrome family
 - d) Don't know, I have not been tested for a faulty gene or my genetic test was unclear
 - e) No, I have been tested and no faulty gene was identified**

2. Have you ever had breast cancer?
 - a) No, I have never had breast cancer**
 - b) Yes, I have had breast cancer**

3. How old are you?
 - a) Under 35**
 - b) 35-39**
 - c) 40-49**
 - d) Over 50**

If the answers shown in bold do not apply to you then the information on the following pages may not be relevant to your situation. Please go back and answer the questions to see information relevant to you or go back and view the general information if you do not wish to personalise OvDex.

Questions in this Chapter:

How can I find out whether I have a faulty gene?

What is my ovarian cancer risk if no faulty gene is found?

How can I find out whether I have a faulty gene?

Ovarian cancer occurs by chance in less than 2 of every 100 women. For women who have a family history and/or a faulty gene, this can be much higher.

Testing for a faulty gene in the family usually begins with a test on a blood sample from a person who has had cancer. If a faulty gene is found, other members of the family can have a genetic test to see if they also carry the same gene.

What is my ovarian cancer risk if no faulty gene is found?

If no faulty gene is found your genetic test is said to be 'negative' and there are two possibilities:

1. Someone else in your family has already been tested and has been found to have the faulty gene. Now you have been found not to have this gene.

In this case your risk of ovarian cancer is likely to be low, therefore you do not need to consider the option of risk-reducing surgery any further at this point.

2. You were the first person to be tested in your family and were not found to have a faulty gene.

In this case your risk of ovarian cancer may still be higher than the general population, as science is not yet able to find all the faulty genes that might be linked to ovarian cancer. Therefore you may still want to consider risk-reducing surgery and should discuss this with your doctor. They will estimate your risk based on your family history. You can also go back to the main page and have a look at the general information if you wish to find out more about surgery.

Questions in this Chapter:

What are my choices?

What if my genetic test is negative and I am from a family where a faulty gene has been found before?

What if my genetic test is negative and I am the first person to be tested?

What are my choices?

As you have not been found to carry a faulty gene, there are different situations that might apply to you. Please find the situation below that applies to you.

What if my genetic test is negative and I am from a family where a faulty gene has been found before?

If there is a faulty gene in your family and your genetic test is negative for that gene (i.e. you do not have the faulty gene that other members of your family have), then you are at low risk and will probably not have to make a choice about risk reduction.

What if my genetic test is negative and I am the first person to be tested?

If you are the first person to be tested in your family and no faulty gene is identified, there might still be a gene that science cannot find yet. In this case you will need to discuss your risk level and options with your genetic counsellor. You can come back to this website once you have found out more about your situation and whether or not risk-reducing ovarian surgery is an option for you.



Contacts and Resources

Here are some links and contact details that you might find helpful (even though we have taken great care in choosing websites from reliable sources please note we are not responsible for the content of these websites):

The Cancer Genetics Story Bank – An online collection of stories told by patients and professionals about cancer genetics, developed by the Cancer Genetics Service for Wales (CGSW)

www.cancergeneticsstorybank.co.uk

Target Ovarian Cancer – A UK charity which supports research into ovarian cancer and provides useful information including an 'Ask the Expert' feature. Available in several languages.

www.targetovariancancer.org.uk

Ovacom – A UK charity providing information and support for everyone affected by ovarian cancer. Includes a symptom awareness tool and links to a number of patient blogs.

www.ovacome.org.uk or call the helpline on 08453710554

Your GP and/or Genetics Service – There to help you with any questions or concerns.

Appendix 7.5 - Actions in response to suggestions for improvements – Round 1

| Suggestions for improvements | Exact action taken / Decision |
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| PATIENTS | |
| <p>"I have had a read over the documents, they are very comprehensive and I wondered whether some sections could be condensed for example: I felt that the section entitled 'Does my cancer risk change over time' a simple yes may have sufficed?"</p> | <ul style="list-style-type: none"> - For closed questions added Yes/No responses to have a short / clear answer (but left longer responses as extra info) - Removed repetitions to condense info |
| <p>"On the Risk Reducing Surgery section the part detailing amount of women having surgery and those that are converted from keyhole to open surgery when I first read the explanation to the right of the grid in bold I thought the grid showed amount that was converted to open surgery...which would put a lot of people off the idea."</p> | <ul style="list-style-type: none"> - Changed colour coding of dots - Moved dot representing "conversion from keyhole to open" to end, so dot order corresponds to order in text - Un-bolded "planned keyhole which is converted to open surgery..." to remove emphasis |
| <p><i>Section on Why is there a choice? Participant stated: "I am clearly given the indication from the gynecologist during my yearly hysteroscopy that I should have my ovaries and womb removed as the risk of cancer outweighs my fears on being on HRT after surgery."</i></p> | <ul style="list-style-type: none"> - Added a sentence referring to potential health professional attitudes/recommendations to question "Why is there a choice" - Also added statement 'should not feel pressured' |
| <p><i>In option grid participant highlighted "No, your risk of ovarian cancer will remain increased." And asked: "why??...is this because I am getting older?"</i></p> | <ul style="list-style-type: none"> - Replaced the word 'increased' in option grid with the word 'high' to avoid confusion with increasing risk with age etc.. |
| <p><i>In option grid participant highlighted "Are there any risks linked to early menopause?" and said: "+ heart disease + memory failure + depression (I think it is very important to mention these risks)"</i></p> | <ul style="list-style-type: none"> - Added references to possible increased risk of CVD in option grid, surgery disadvantages, menopause and HRT section - Added information about risk of CVD in menopause section under question 'Are there any long term health effects linked to the menopause?' - Removed refs to 'surgical menopause' from entire menopause section to emphasize that all effects and symptoms are equal to natural menopause and alleviate the fear of 'surgical' menopause a little - In HRT section added info on oestrogen + progesterone HRT and oestrogen only HRT and added that evidence regarding HRT against CVD risk is controversial - In menopause and HRT section mentioned that bone thinning & CVD risk depends on medical history and lifestyle and should be discussed with doctor - Left 'memory changes' instead of 'failure' in the option grid |

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| | <ul style="list-style-type: none"> - Added depression as menopausal symptom in Surgical Menopause chapter |
| <p><i>In option grid participant highlighted "HRT is safe to take for the majority of women until they are 50." And stated "I know someone who insists on continuing taking oestrogen only HRT, and has been doing so since her removal of ovaries at the age of 28 (she is now 54 - she had ovaries removed due to cysts not risk of cancer) as she will otherwise have masculine features such as becoming very hairy including facial hair, and a deep masculine voice."</i></p> | <ul style="list-style-type: none"> - Added some explanation of why HRT should be stopped at 50 in HRT chapter - Added sentence to state that memory changes are also experienced by women going through the natural menopause and are a normal part of ageing |
| <p><i>Participant highlighted "The most commonly reported symptoms of surgical menopause are hot flushes, night sweats, mood swings, vaginal dryness and loss of interest in sex." And stated "Osteoporosis, heart disease, memory failure, are the most worrying."</i></p> | <ul style="list-style-type: none"> - Changed title of first question to include term 'short-term' - Changed title for problem question to include term 'long-term' - Added references to possible increased risk of CVD in option grid, surgery disadvantages, menopause and HRT section (indicated in blue until confirmed) - Added sentence to state that memory changes are also experienced by women going through the natural menopause and are a normal part of ageing |
| <p><i>Participant highlighted "Are there other problems linked to surgical menopause?" and asked "Why is heart disease not mentioned?"</i></p> | <ul style="list-style-type: none"> - Added references to possible increased risk of CVD in option grid, surgery disadvantages, menopause and HRT section - Added information about risk of CVD in menopause section under question 'Are there any long term health effects linked to the menopause?' - Removed refs to 'surgical menopause' from entire menopause section to emphasize that all effects and symptoms are equal to natural menopause and alleviate the fear of 'surgical' menopause a little - In HRT section added info on oestrogen + progesterone HRT and oestrogen only HRT and added that evidence regarding HRT against CVD risk is controversial - In menopause and HRT section mentioned that bone thinning & CVD risk depends on medical history and lifestyle and should be discussed with doctor |
| <p><i>Participant highlighted "Hormone replacement therapy (HRT) is usually recommended for women who have RRSO before the age of 45" and stated "I thought this was before the age of 52"</i></p> | <ul style="list-style-type: none"> - No action taken – after review of literature and clinical recommendations (left age at 45 and wording as 'strongly recommended...' and 'chose to...') |
| <p><i>Participant highlighted "Women who have surgery when they are over 45 years of age can"</i></p> | <ul style="list-style-type: none"> - No action taken – after review of literature and clinical recommendations (left age at 45 and |

| | |
|---|--|
| <p><i>choose to have HRT to reduce the symptoms of surgical menopause. However these women do not have to have HRT if they do not want to..." and stated "Again, I thought the risks of bone thinning etc was for women up to 52 years (not 45 years)"</i></p> | <p>wording as 'strongly recommended...' and 'chose to...'</p> |
| <p><i>Participant highlighted "If you use HRT, it is recommended that you take it until the age of the natural menopause, which is 50 years in the UK" and asked "What will happen if I chose to continue taking HRT for many more years past the age of 52yrs?"</i></p> | <p>- Added some explanation of why HRT should be stopped at 50 in HRT chapter</p> |
| <p><i>Participant highlighted "If you stop taking HRT at the age of the natural menopause (so when you are 50) then there is no evidence that the breast cancer risk is increased" and asked "Sorry but I have a problem of understanding HRT and I get very confused when reading information on this subject. Can you please simply clarify - "too much oestrogen in HRT taken over the age of 52years / or too much oestrogen in the body causes breast cancer"??"</i></p> | <p>- Clarified that high oestrogen levels increase BC risk in HRT chapter Question: "But I have heard..." - Added graph to explain oestrogen levels in surgical versus natural menopause in menopause chapter - In HRT chapter added graph to explain how HRT adds back oestrogen and increases oestrogen levels after pre-menopausal surgery and how it then is reduced gradually</p> |
| <p><i>Participant highlighted "However, HRT can relieve some of the symptoms of the menopause" and asked "Will HRT also help keep at bay heart disease and thinning of bones?"</i></p> | <p>- Added new question in HRT chapter "Will HRT deal with the long-term effects of menopause?" with short explanatory answer - Also added in Menopause chapter question "Is there anything I can do" statement 'HRT can may effectively reduce some of the short- and long-term effects of surgical menopause'</p> |
| <p><i>Participant highlighted "No, unfortunately there is no medically proven screening available for ovarian cancer" and "At the moment the only alternative to surgery is to do nothing and simply be aware of the symptoms of ovarian cancer should they develop." And "If you decide not to have surgery, then nothing will really change. You will need to look out for any symptoms and if you think anything is wrong you need to go to your GP to get it checked." And "As there is no screening available there is very little else you can do apart from keeping an eye on any symptoms." and stated "Again, I need to reiterate internal vagina scan + CA125 blood test"</i></p> | <p>- Changed wording of 'Other options' chapter question about screening to explain CA125 and TV scan policy and emphasize lack of proof - Added new question in 'Other options' asking: 'But I have been offered CA125/TV scans...'</p> |
| <p>" There was some repetition"</p> | <p>- Removed repeated question "What can I do..." from Cancer Risk section (now only in Choice section) - Removed repetition in menopause symptoms</p> |
| <p>"It mentions genetic testing. I have personally not had this and have struggled to get it. More information on this would be useful." [General</p> | <p>- Added more detail in "How can I find out whether I have a faulty gene..." - Added link to Macmillan in Contacts &</p> |

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| version] | Resources with specific pointer to 'genetic testing' section |
| "Page numbers would be good." | - Added page numbers on top right hand corner of pdf docs |
| "I would like to have seen Chapter numbers and individual paragraph / section numbers. A minor point, but this would be helpful as there are quite a lot of cross references to different sections throughout." | - Numbered all Chapters in pdf (1, 2, 3... etc.) - Numbered all questions (Q1, Q2... etc.) in each chapter - Added table of contents of first page of pdf - Changed all cross refs within pdfs to point to chapter number and page number |
| "During the round table discussion session I attended, one of the participants mentioned that there would be an increased risk of heart disease following an oophorectomy. Whether this is correct I don't know but if there are any other potential significant health risks associated with this surgery (other than cancer of the peritoneum) it would be helpful to refer to these." | - Added references to possible increased risk of CVD in option grid, surgery disadvantages, menopause and HRT section - Added information about risk of CVD in menopause section under question 'Are there any long term health effects linked to the menopause?' - Removed refs to 'surgical menopause' from entire menopause section to emphasize that all effects and symptoms are equal to natural menopause and alleviate the fear of 'surgical' menopause a little - In HRT section added info on oestrogen + progesterone HRT and oestrogen only HRT and added that evidence regarding HRT against CVD risk is controversial - In menopause and HRT section mentioned that bone thinning & CVD risk depends on medical history and lifestyle and should be discussed with doctor |
| "For Lynch Syndrome women the surgery would involve hysterectomy as well as oophorectomy. It, therefore, doesn't make complete sense to consider removal of the ovaries in isolation. It would be useful to either include more information on the hysterectomy or to link to it from another source at the relevant points in the text. In addition, your links page could include links to more information about hysterectomy for Lynch Syndrome women." [Lynch version] | - Added hysterectomy as integral part of the RRSO decision for Lynch women by: - Changing information in 'Cancer risk chapter' to mention hysterectomy and link to 'risk-reducing surgery' section - Changing wording in 'The choice' to include hysterectomy - Changing 'The surgery' chapter to include information about hysterectomy throughout |
| "It is stated that the alternative to surgery is 'watchful waiting.' This doesn't ring true for women with Lynch Syndrome. It may be correct to state that there is no effective screening. In practice however, Lynch Syndrome women receive (via the NHS) an annual transvaginal ultrasound scan for womb cancer screening. The ovaries are visible in this scan and so ovarian cysts are monitored and followed up with further scans and CA125 blood tests. This is my experience- it might not serve a useful purpose | - Rewrote section about screening in 'other options chapter' to specifically address screening that Lynch women might receive and to emphasize again that it is not effective for ovarian cancer |

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| <p>but it is a kind of screening/monitoring. Maybe this experience could be reflected in the text as something that does happen with Lynch Syndrome.” [Lynch version]</p> | |
| <p>“The information in this file was designed for all women over 50. However, in the text it states that ‘most ovarian cancers in women with Lynch Syndrome are diagnosed between the ages of 40 and 55.’ Further down on the same page it is suggested that most ovarian cancers in Lynch women are diagnosed before the age of 50. Given these facts, I am not sure that it is coherent having all women over 50 looking at the same information when there are vastly differing rates of risk. Would it not make more sense to add an additional group?” [Lynch version]</p> | <ul style="list-style-type: none"> - Removed sentence that said: ‘most OCs in lynch occur between 40 and 55’ and left sentence that read ‘before age 60’ to be more consistent - Included info on endometrial cancer in ‘Cancer Risk’ section; endometrial cancer risk is much higher for Lynch and combined with hysterectomy decision, so ovarian risk no longer stand-alone issue - Did not split into extra age group |
| <p><i>Participant highlighted sentence: “ but it is now understood that people from Lynch families do not necessarily need to have a confirmed faulty gene to be at risk, as many genes that might be linked to Lynch Syndrome are not yet known” and asked “My sisters who were also tested for MSH2 faulty gene are clear and have not inherited this faulty gene - are they ok?”</i> [Lynch version]</p> | <ul style="list-style-type: none"> - Yes, if a faulty gene is found in a member of the family, any member who is not found to have that gene is then at low risk. - Added sentence to Cancer Risk chapter question about faulty genes: ‘However if a faulty gene is found in a member of the family and other members of the family do not have this gene, then those members are at low risk.’ |
| <p><i>Participant highlighted: “Lynch Syndrome (for example: MLH1, MSH2, MSH6 and PMS2).” And asked “please give ovarian cancer risk in percentage for each Lynch Syndrome)”</i> [Lynch version]</p> | <ul style="list-style-type: none"> - Added exact risk estimates for MSH1, MSH2 and MSH6, which were available form 2009 paper; no further exact ovarian cancer risk estimates available for other mutations |
| <p><i>Participant highlighted: “However, knowing your risk will give you access to additional medical services such as...” and said “need to mention surveillance for internal vagina scan, hysteroscopy, CA125 blood test.”</i></p> | <ul style="list-style-type: none"> - Added ‘...and screening’ to the paragraph about ‘how will this affect my life’, but did not specify types of screening etc., as this is discussed later in the decision aid |
| <p><i>Participant highlighted: “To reduce your risk of endometrial cancer you may consider a risk-reducing hysterectomy (removal of healthy uterus). This you will need to discuss with your doctor.” And said “1. Need to mention the higher risk of endometrial cancer (i think this is about 60% for MSH2 carriers) compared to ovarian cancer risk of 6-12%. Therefore if you are considering having your ovaries removed, it is probably best to have a hysterectomy at the same time. 2. MSH2 carriers are also at risk of skin cancer I believe.”</i> [Lynch version]</p> | <ul style="list-style-type: none"> - Have now mentioned endometrial cancer risk estimates for MSH1, 2 and 6 in cancer risk chapter - Did mention ‘other cancers’ for Lynch, so that I won’t have to specify all cancers they may be at risk for and recommended discussion with doctor - Added hysterectomy as integral part of the RRSO decision for Lynch women by: - Changing information in ‘Cancer risk chapter’ to mention hysterectomy and link to ‘risk-reducing surgery’ section - Changing wording in ‘The choice’ to include hysterectomy - Changing ‘The surgery’ chapter to include information about hysterectomy throughout |

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| <p>Participant highlighted: “Unfortunately, there is no screening for ovarian cancer at the moment” and said: “Internal vagina scan, and CA125 blood test is available.”</p> | <ul style="list-style-type: none"> - Rewrote section about screening in ‘other options chapter’ to specifically address screening that Lynch women might receive and to emphasize again that it is not effective for ovarian cancer |
| <p>In option grid participant highlighted “Yes, you can have your ovaries taken out at the same time as having your hysterectomy.” And asked “Will this then be ‘open’ surgery or ‘key hole’ surgery?” [Lynch version]</p> | <ul style="list-style-type: none"> - In Surgery chapter now have detailed description of hysterectomy surgical techniques (open, vaginal and laparoscopic) |
| <p>In option grid participant highlighted “No, there is no routine screening available for ovarian cancer” and stated “There is internal vagina scan and also CA125 blood test”</p> | <ul style="list-style-type: none"> - Rewrote section about screening in ‘other options chapter’ to specifically address screening that Lynch women might receive and to emphasize again that it is not effective for ovarian cancer |
| <p>Participant highlighted entire paragraph on “Do I need to have my womb removed as well?” and asked “More onus on this point to stress the riskier considerably higher percentage of womb cancer for MSH2 carriers. Therefore if going through surgery and removing ovaries - the uterus should be removed at the same time rather than go through two surgery/operations.” [Lynch version]</p> | <ul style="list-style-type: none"> - Added hysterectomy as integral part of the RRSO decision for Lynch women by: - Changing information in ‘Cancer risk chapter’ to mention hysterectomy and link to ‘risk-reducing surgery’ section - Changing wording in ‘The choice’ to include hysterectomy - Changing ‘The surgery’ chapter to include information about hysterectomy throughout |
| <p>“I strongly believe the two procedures [RRSO and hysterectomy] are linked and information on hysterectomy should certainly be included.” [Lynch version]</p> | <ul style="list-style-type: none"> - Added hysterectomy as integral part of the RRSO decision for Lynch women by: - Changing information in ‘Cancer risk chapter’ to mention hysterectomy and link to ‘risk-reducing surgery’ section - Changing wording in ‘The choice’ to include hysterectomy - Changing ‘The surgery’ chapter to include information about hysterectomy throughout |
| Suggestions for improvements | Exact action taken / Decision |
| PROFESSIONALS | |
| <p>“The Ovacom symptom awareness tool is not NICE compliant, in that it does not highlight urinary symptoms.”</p> | <ul style="list-style-type: none"> - Removed reference to Ovacom symptom awareness tool in Contacts and Resources section - Added reference to symptoms appearing more than 12 times a month in ‘Other options’ section on symptoms in agreement with NICE guidance - Changed ‘needing the toilet’ to ‘needing to urinate’ in symptoms list |
| <p>“Question: When should I decide? I am not certain about the phrasing in this context – It is important you make sure you have completed your family before you make this choice. It sounds like an instruction. I think at this point it is important for women to understand that undergoing surgery will mean they cannot get</p> | <ul style="list-style-type: none"> - Rewrote the second part of the question to refer to ‘family planning’ and potential of meeting someone / remarrying to state: ‘Therefore it is essential to consider your plans for a family and any potential future changes to these plans (for example if there is a chance you might meet a new partner / re-marry) before |

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| <p>pregnant, therefore it is important that they discuss implications and timing of any possible surgery on their plans to complete a family.”</p> | <p>making a final decision’ - Also reworded in RRSO chapter disadvantages section on family planning into: “...have considered your plans for a family before making a final decision.”</p> |
| <p>“Question: How will my ovarian cancer risk affect my children? It would be useful to say what sort of chance that your children may have of developing ovarian cancer rather than just ‘there is a chance’ which would worry most parents.”</p> | <p>- Rewrote section on children’s risk in Cancer Risk chapter to include risk of inheriting a faulty gene and resulting risk of cancer and age relationship - Added link to NHS Choices website info in inheritance / genetics in Cancer Risk chapter and in Contacts and Resources</p> |
| <p>“Options at a glance: Will this change how I feel about myself as a woman? I would suggest that if your desire for and/or enjoyment of sex may decrease a lot, then there is a pretty good chance it will change how you feel as a woman.”</p> | <p>- Replaced the word ‘Most’ with ‘Many’ in option grid - Added “However, your desire....” So women can decide themselves whether they think their feeling of womanhood might be affected</p> |
| <p>“Options at a glance – can I get screen for ovarian cancer – the answer is no on both counts, If however you are thinking that this may change soon because of the UKFOCSS trial, then the answer would surely be yes, as it has been stated that not all genes relating to lynch syndrome have been found?”</p> | <p>- Changed wording in option grid to ask “Is there a routine screening programme to detect ovarian cancer?” - Changed answer to this question in option grid to read: ‘No, and....’ and ‘...no routine screening is available on the NHS to detect OC’</p> |
| <p>“Risk Reducing Surgery – What are the main advantages of this operation – again the word greatly needs quantifying (as above).”</p> | <p>- Removed the word ‘greatly’ - Added information on BRCA1 PPC risk and non-BRCA1 PPC risk (i.e. very low risk for all others) to emphasize this point</p> |
| <p>“Risk Reducing Surgery – What are the main disadvantages of this operation Need to include potential loss of desire and/or enjoyment of sex.”</p> | <p>- Added new point to disadvantage list to state: ‘Your desire for sex will decrease and due to some menopausal symptoms, such as vaginal dryness, you may enjoy sex less as intercourse can be painful.’</p> |
| <p>“What is surgical menopause like? Need consistency when talking about the range of symptoms, and there is repetition. Could say the most commonly experienced symptoms are x, y, z. Other symptoms which may be experienced are”</p> | <p>- Deleted repetition and split sentence to distinguish ‘most common’ and ‘other’ symptoms.</p> |
| <p>“How could it affect my life? Would it be appropriate here to include some information about help that can be sought?”</p> | <p>- Added paragraph to ‘How will it affect my life’ question in Menopause chapter to refer to lubricant and other options such as CBT - Added ref to NHS choices website</p> |
| <p>“What is the alternative to surgery? The symptoms should be the ones used in the NICE Guidance CG122. In particular the reference to needing the toilet more than usual could be construed as bowel rather than bladder related. There should always be a reference to frequency of symptoms as well (particularly more than 12 times a month) otherwise there is the</p> | <p>- Added reference to symptoms appearing more than 12 times a month in ‘Other options’ section on symptoms in agreement with NICE guidance - Changed ‘needing the toilet’ to ‘needing to urinate’ in symptoms list</p> |

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| potential to cause undue concern over transient symptoms.” | |
| “Does my cancer risk change over time – a little bit difficult to understand” | - Added simple one word answer ‘Yes’ to have a short / clear answer (but left longer responses as extra info) |
| “What can I do to reduce my risk? Does there need to be some explanation about screening as some people will have heard about blood tests and scans and although the evidence is not proven they may not understand- or refer to the bit in Other Options section.” | - Added ‘no medically proven screening’ sentence to Choice Chapter “What can I do?” question - Added reference to ‘Other options’ in “What can I do?” question - Changed wording of ‘Other options’ chapter question about screening to explain CA125 and TV scan policy and emphasize lack of proof |
| “Page 4 Does there need to be explanation as to why there is no screening available for ovarian cancer at present given that many people will have heard about scans Ca125 blood tests and trial?” | - Changed wording of ‘Other options’ chapter question about screening to explain CA125 and TV scan policy and emphasize lack of proof and did specify that there is no evidence that screening detects ovarian cancer early - Added new question in ‘Other options’ asking: ‘But I have been offered CA125/TV scans...’ |
| “Page 7 Risk Reducing Surgery Although most people considering this surgery will be pre menopausal there will be some who have already gone through the menopause (and having spoken to several they are really concerned about the effects of surgery when most of what they read applies to premenopausal women- strangely they seem to be even more worried than premenopausal women)” | - Wrote ‘Surgical menopause’ chapter for post-menopausal docs to specify how surgical menopause might affect peri- and post-menopausal women - Added new graph with oestrogen levels in surgical & natural menopause and with surgery during natural menopause to demonstrate difference in drop |
| “Page 8 The striped dot in the dot matrix – conversion to open surgery is not very clear” | - Changed colour coding of dots - Moved dot representing “conversion from keyhole to open” to end, so dot order corresponds to order in text - Un-bolded “planned keyhole which is converted to open surgery...” to remove emphasis |
| “What are the complications of RRSO Last sentence of the first paragraphthe surgeon has to convert to an operation or to open surgery.” | - Removed the word “an” |
| “Can women die? Is it possible to make this less prominent?” | - Removed individual question “Can women die...?” and instead integrated risk of death at the end of previous question about complications |
| “Page 9 What is surgical menopause? I think a lot of people who have had a surgically induced menopause will find that they experience symptoms much more suddenly than those who experience the menopause naturally | - Added statement to explain that surgical menopause is more sudden and severe in Chapter Menopause under what is it like - Also added paragraph explaining that surgical menopause is essentially the same as natural menopause, only more sudden |

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| particularly amongst younger women" | <ul style="list-style-type: none"> - Added graph to explain oestrogen levels in surgical versus natural menopause in menopause chapter - Added highlighted statement at beginning of menopause chapter to alert readers that symptoms are described for women who are not taking HRT |
| <p>"Page 12 What is the alternative to surgery? 'If any of these symptoms are prolonged' it might be helpful to give an indication of how long it should be before some one goes to the doctors – 3 weeks is usually the suggested length of time."</p> | <ul style="list-style-type: none"> - Added reference to symptoms appearing more than 12 times a month in 'Other options' section on symptoms in agreement with NICE guidance - Changed 'needing the toilet' to 'needing to urinate' in symptoms list |
| <p>"Page 13 Ovacomecall the support line rather than helpline."</p> | <ul style="list-style-type: none"> - Changed to 'support-line' |
| <p><i>Participant highlighted "and/or enjoyment of sex may decrease..." and said "think may need qualification"</i></p> | <ul style="list-style-type: none"> - Did not change content of option grid, but: in chapter 'RRSO' added new point to disadvantage list to state: 'Your desire for sex will decrease and due to some menopausal symptoms, such as vaginal dryness, you may enjoy sex less as intercourse can be painful.' |
| <p><i>Participant highlighted "For the vast majority of women having RRSO..." and added "before the menopause"</i></p> | <ul style="list-style-type: none"> - Added 'before the natural menopause' to RRSO chapter advantages section about BC risk |
| <p><i>Participant highlighted "...it is important that you have completed your family before going for surgery." And added "or not in a time of your life where you may remarry etc"</i></p> | <ul style="list-style-type: none"> - Rewrote the second part of the question to refer to 'family planning' and potential of meeting someone / remarrying to state: 'Therefore it is essential to consider your plans for a family and any potential future changes to these plans (for example if there is a chance you might meet a new partner / re-marry) before making a final decision' |
| <p><i>Participant highlighted "These effects might be reduced by hormone replacement therapy" and said "can be effectively treated...not all women can take hrt however. This should be discussed before surgery"</i></p> | <ul style="list-style-type: none"> - Reworded answer to say 'effectively treated' in RRSO chapter disadvantages, as suggested by respondent - Added statement that HRT should be discussed before surgery in RRSO chapter, menopause chapter and HRT chapter where appropriate |
| <p><i>Participant highlighted "For all other women the risk of primary peritoneal cancer after surgery is close to zero." And said "Repeated info in this paragraph"</i></p> | <ul style="list-style-type: none"> - Simply underlined types of cancer, as this was not a repetition: first sentence refers to ovarian cancer & fallopian tube cancer risk and last sentence to PPC |
| <p><i>Participant highlighted "The most commonly reported symptoms of surgical menopause are hot flushes, night sweats, mood swings, vaginal dryness and loss of interest in sex." And said "repeated"</i></p> | <ul style="list-style-type: none"> - Deleted repetition and split sentence to distinguish 'most common' and 'other' symptoms |
| <p><i>Participant highlighted "The way that these symptoms might affect your life depends on your personal situation." And asked "was this study</i></p> | <ul style="list-style-type: none"> - Added highlighted statement at beginning of menopause chapter to alert readers that symptoms are described for women who are |

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| with or without hrt...as it seems very off-putting??" | not taking HRT |
| <i>Participant highlighted "...about 18 months after their operation..." and stated "curious about this one as to where you got this info...not sure our patients get this ..is it from london?"</i> | - Changed paragraph in 'menopause' chapter to say that some regions recommend BDS and that women should ask their doctor |
| "How will my cancer risk affect my life? – this seems to be aimed at women at an earlier stage than the OVDEX tool - ...means you might be offered risk reducing surgery.... Over the page you say = you have the option of undergoing risk reducing surgery..." | - Changed wording in Cancer risk chapter "How will my risk affect my life" to read 'you are offered...' (changed tense) |
| "[Options at a glance] There is no mention of the increased cardiovascular risk of early menopause." | <ul style="list-style-type: none"> - Added references to possible increased risk of CVD in option grid, surgery disadvantages, menopause and HRT section - Added information about risk of CVD in menopause section under question 'Are there any long term health effects linked to the menopause?' - Removed refs to 'surgical menopause' from entire menopause section to emphasize that all effects and symptoms are equal to natural menopause and alleviate the fear of 'surgical' menopause a little - In HRT section added info on oestrogen + progesterone HRT and oestrogen only HRT and added that evidence regarding HRT against CVD risk is controversial <p>In menopause and HRT section mentioned that bone thinning & CVD risk depends on medical history and lifestyle and should be discussed with doctor</p> |
| "A picture of the anatomy might be helpful as many women do not know their anatomy or understand the difference between the ovary uterus and cervix! It would also be sensible to explain somewhere where the cervix is and that cervical cancer is not raised in LS and that cervical screening will not detect ovarian or endometrial cancer." | <ul style="list-style-type: none"> - Added question 'where does Ovarian cancer develop' in Cancer Risk chapter and included sketch of anatomy with labels and legend explaining difference between OC/fallopian tube cancer and cervical / uterine cancer - Added emphasis that surgical smear does not detect ovarian cancer in other options chapter (screening paragraph) |
| "If they do have a family history of breast cancer they should be advised to seek advice about this from the genetics service. This is referred to in the last question about HRT." | <ul style="list-style-type: none"> - Added statement that HRT should be discussed before surgery in RRSO chapter, menopause chapter and HRT chapter where appropriate - Added in HRT chapter in question "Is there anything I can do..." statement: 'However, not all women can take hormone replacement and this should be discussed before having surgery.' |
| "You have not mentioned the protective effect of the oral contraceptive Pill for ovarian cancer." | <ul style="list-style-type: none"> - No official guidelines regarding use of OCP in UK - Added paragraph in 'other options' section under 'is there anything else...' to mention that some countries recommend OCP use and that it |

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| | decreases OC risk but increases BC risk and should be discussed with doctor |
| “The information about ovarian symptom awareness is vague. You've said if the symptoms last longer than usual. I suggest you use the same terminology as the NHS choices website ie 'if the symptoms occur on most days'. You could also direct women to the NHS choices link for ovarian symptom awareness.” | - Added reference to symptoms appearing more than 12 times a month in 'Other options' section on symptoms in agreement with NICE guidance - Changed 'needing the toilet' to 'needing to urinate' in symptoms list |
| “Although perhaps for this age group you do not need to give so much information about surgery as I would suggest that they really should be counselled against RRBSO until their risk is higher.” | - Changed info for under 35s to say: “therefore you do not need to make a decision about risk-reducing surgery right now. The information in the rest of this document describes the different options and focuses on surgery, therefore you may wish to come back to this at a later point when it is more relevant to you” |
| “There is mention of the ovarian cancer risk if there is a family history of ovarian cancer but some families have a mixed history of breast and ovarian cancer – should the information for this group be different? [General version] | - Reworded section on family history to include ovarian, breast and bowel - Added paragraph in RRSO chapter question about family history and risk to emphasise other family history 'The number of relatives with cancers other than ovarian cancer, such as breast and bowel cancer, can also affect...' |
| 'Women who have more than one relative with ovarian cancer.....' should there be mention of whether this is first or second degree relationships" [General version] | - Reworded sentence in RRSO chapter question about family history and risk to state: 'Women who have 2 or more first (mother, sister or daughter) and/or second degree (aunt, grandmother etc.) relatives...' |
| “Page 2 'How can I find out whether I have a faulty gene.... Some women might come from familiesis even more common' –than ? the general population or than those with a first degree or second relatives. Should there be an explanation somewhere as to what 1st and 2nd degree means? (there may already be)” [General version] | - Reworded sentence in RRSO chapter question about faulty gene to state: '... [where cancers] have affected more than just one or two relatives and these women...' - Reworded sentence in RRSO chapter question about family history and risk to state: 'Women who have 2 or more first (mother, sister or daughter) and/or second degree (aunt, grandmother etc.) relatives...' to give indication of what 1 st and 2 nd degree mean |
| <i>Participant highlighted “Some women might come from families where cancer is...” and stated “i would say certain types of cancer such as breast/ovary or ovary/womb/bowel”</i> [General version] | - Reworded RRSO chapter question about faulty gene and risk to state: 'Some women might come from families where certain types of cancer (e.g. ovarian, breast, bowel) have affected...' |
| “Question on how can I find out whether I have a faulty BRCA1 gene is irrelevant at this point, they already have it confirmed.” [BRCA version] | - Removed question |
| “Graph on risk of ovarian cancer – legend on graph says ovarian cancer, but the text on the right is all about breast cancer. And the graph matches the next one for breast cancer.” [BRCA | - Removed wrong graph and legend and inserted ovarian cancer risk for 35-39 year old BRCA1 carrier graph |

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| version] | |
| “Will my cancer risk affect my children. Should quantify the risk here.” [BRCA version] | - Copied new ‘children risk’ paragraph from General file and adapted for BRCA1 risk (i.e. risk of inheritance of faulty BRCA1/2 gene and resulting cancer risk) |
| “Options at a glance – will this affect my ovarian and breast cancer risk? You have a better explanation here of the reduction in risk (see comments above) but needs qualifying somewhere as to why it is not 100%. Other comments are as per previously. [BRCA version] | - No action taken here, there is explanation about PPC later-on in RRSO chapter and OG needs to stay compact. |
| “Options at a glance: Will this reduce my ovarian cancer risk? The answer Yes, removing the ovaries will greatly reduce your lifetime risk of ovarian cancer is not clear enough. What do you mean by greatly? Reference is made later on that there is a small chance ovarian cancer is found when they operate, but it does not address the risk of Primary Peritoneal Cancer.” | - Removed the word ‘greatly from surgery section questions about advantages, but left in the option grid, as it is not possible to quantify - No action taken in OG, there is explanation about PPC later-on in Surgery chapter and OG needs to stay compact. |
| “What is my risk after surgery? – Here you say the risk is effectively eliminated. Again this is not so, and even at odds with greatly reduced. Note point about Primary Peritoneal Cancer which is classified as a type of ovarian cancer.” [Lynch version] | - Added explanation about PPC in Surgery chapter ‘advantages’ to quantify PPC risk after RRSO for Lynch as below 2/100 - Reworded answer to question “What is my risk after...” to say specifically: ‘risk of <u>endometrial, ovarian and fallopian tube</u> cancer is effectively eliminated’ and to then include description of what PPC is and specify risk of PPC after surgery |
| “Could cancer be found during the surgery – this is a good explanation (especially if you quantify the risk) that should be used elsewhere. However it does not explain that even so there is still a residual risk from PPC, which often has a genetic connection.” [Lynch version] | - Added explanation about PPC in Surgery chapter ‘advantages’ to quantify PPC risk after RRSO for Lynch as below 2/100 - Reworded answer to question “What is my risk after...” to say specifically: ‘risk of <u>endometrial, ovarian and fallopian tube</u> cancer is effectively eliminated’ and to then include description of what PPC is and specify risk of PPC after surgery - Because the incidence of cancer during surgery or of PPC after surgery is so low there are no stats available with exact numbers for Lynch |
| “I appreciate that you wish to focus on surgery for ovarian cancer but it seems odd to have such a lot of information focusing on surgery for one type of cancer when others are more relevant to these patients and may make the other cancers seem less relevant. Although the risk of ovarian cancer is raised above the population risk in LS, it is in fact significantly lower than the risk of other cancers. Given that the management of colorectal cancer risk in Lynch Syndrome tends to be screening rather than surgery, perhaps this is less of a problem for colorectal cancer but I do | - Added hysterectomy as integral part of the RRSO decision for Lynch women by: - Changing information in ‘Cancer risk chapter’ to mention hysterectomy and link to ‘risk-reducing surgery’ section - Changing wording in ‘The choice’ to include hysterectomy - Changing ‘The surgery’ chapter to include information about hysterectomy throughout |

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| <p>think that equal weight should be given in this document to decision-making about a hysterectomy.” [Lynch version]</p> | |
| <p>“The risk for ovarian cancer is still low at age 35 (your graph is rather misleading as quick glance by someone who doesn't understand it may suggest that it is higher than it is). For this reason I would suggest that you change the scale of the graph. You talk about women having an option to have surgery at age 35 but I am concerned that the risk at this age is not sufficiently raised. This tool may lead some young women to go ahead with surgery at a younger age than the guidance recommends. In addition, BSO at age 35, even if a woman has completed her family, means a lot of years of HRT which would carry its own risks (eg cardiovascular disease).” [Lynch version]</p> | <ul style="list-style-type: none"> - Replaced graph in Lynch syndrome version completely with pictogram for different mutations instead - Changed info for under 35s to say: “therefore you do not need to make a decision about risk-reducing surgery right now. The information in the rest of this document describes the different options and focuses on surgery, therefore you may wish to come back to this at a later point when it is more relevant to you” |
| <p>“You only refer to the uterus and endometrial cancer. I think you may need to explain that this is often called the ‘womb’ so that women know what you are talking about.”</p> | <ul style="list-style-type: none"> - Added ‘womb’ to picture of female anatomy in cancer risk chapter - Added the words uterus/womb throughout text |
| <p>“The greatest risk for these women is endometrial cancer (particularly for those with MSH6 mutations) yet this is only mentioned in passing ie you could think about having your uterus removed as well. If women read this they may decide to go ahead with RRBSO and not have a hysterectomy and then have to go back for that. The tool really needs to focus on gynaecological risk reducing surgery for these women rather than just BSO.” [Lynch version]</p> | <ul style="list-style-type: none"> - Added hysterectomy as integral part of the RRSO decision for Lynch women by: - Changing information in ‘Cancer risk chapter’ to mention hysterectomy and link to ‘risk-reducing surgery’ section - Changing wording in ‘The choice’ to include hysterectomy - Changing ‘The surgery’ chapter to include information about hysterectomy throughout |
| <p>“You need to be more clear about the fact that, although the BC risk is reduced by RRBSO, this is not actually increased in Lynch Syndrome.” [Lynch version]</p> | <ul style="list-style-type: none"> - Changed sentence structure in Surgery section advantages to read: “Even though Lynch women are not thought to be at higher risk for breast cancer, RRSO before the natural menopause could also reduce the risk of breast cancer” to clarify Lynch does not predispose to BC |
| <p>“You have not mentioned the symptoms of endometrial cancer. This is very important for these women.” [Lynch version]</p> | <ul style="list-style-type: none"> - Added endometrial cancer symptoms to Lynch document ‘other options’ |
| <p>“Questions in this chapter: about faulty genes- the first question and answer is irrelevant at this point (How can I find out whether I have a faulty gene). They have already been tested.”</p> | <ul style="list-style-type: none"> - Removed question |
| <p>“The second question is relevant but needs the tense changing (What is my ovarian cancer risk if no faulty gene WAS found). In the answer it says ‘they will estimate your risk’ – but at this point it will have already been assessed unless I am mistaken.”</p> | <ul style="list-style-type: none"> - Changed wording of question in Cancer Risk chapter to past tense: ‘What is my ovarian cancer risk if no faulty gene was found’ - Removed sentence ‘They will estimate your risk...’ |

“Questions in this chapter on choice: What if my genetic test is negative and I am from a family where a faulty gene has been found before – the answer here is not as clear as on the previous page.... Saying you will probably not have to make a choice about risk reduction is very unclear.”

- Reworded sections in Chapter on Cancer risk and Choice (Cancer Risk chapter now only about risk, i.e. for point 1 risk is low, for point 2 risk might still be high; and Choice is about whether they still need to make a decision about RRSO, i.e. for point 1 no choice needed, for point 2 might need to make choice)

Appendix 7.6 - OvDex booklet prototype II (General version)

The logo for OvDex, featuring the letters 'OvDex' in a stylized font. The 'O' and 'v' are teal, and 'Dex' is grey. The 'v' has a unique shape with a loop at the top.Oophorectomy
Decision ExplorerA large version of the OvDex logo, with the 'O' and 'v' in teal and 'Dex' in grey. The 'v' has a unique shape with a loop at the top.

Oophorectomy Decision Explorer

A decision aid designed to help you
and your doctor make a decision
about risk-reducing ovarian surgery

For women at increased risk of ovarian cancer

OvDex (The Oophorectomy Decision Explorer) has been developed to help you find out more about your options for reducing your ovarian cancer risk. If you are viewing OvDex you should have been referred to it by a doctor or geneticist as you are at increased risk of ovarian cancer.

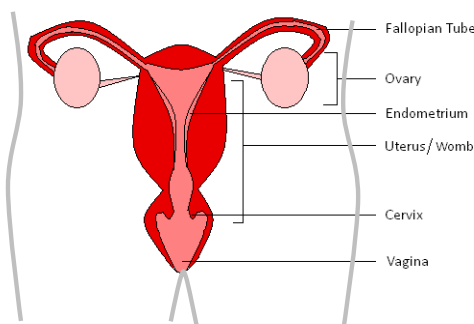
Please note that OvDex can be personalised by answering three questions . You should have seen these questions before you opened these pages. If you do wish to personalise the information you get, please go back and answer the questions .

The information on the following pages has not been personalised. If you do not wish to personalise OvDex then please view the general information on the following pages.

| Chapter | Page |
|--------------------------------|-------------|
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Questions in chapter 1:

- Q1 Where does ovarian cancer develop?
 Q2 What is the ovarian cancer risk if I have a family history of ovarian cancer?
 Q3 How can I find out whether I have a faulty gene?
 Q4 What would my risk be if a faulty gene is found?
 Q5 What if I cannot get tested or my genetic test is unclear?
 Q6 Does my cancer risk change over time?
 Q7 How will my ovarian cancer risk affect my life?
 Q8 How will my ovarian cancer risk affect my children?
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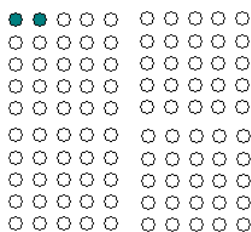
Q1 - Where does ovarian cancer develop?

The picture on the left shows the female reproductive system. The ovaries are connected to the uterus by the fallopian tubes. Ovarian-type cancer can develop in the ovaries, the fallopian tubes or the lining of the abdomen (called the 'peritoneum'). Other female cancers, such as cancer of the endometrium, uterus/womb or cervix, are very different and should not be confused with ovarian cancer.

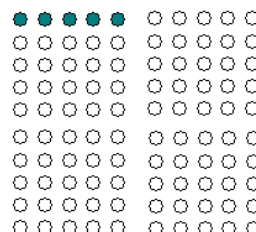
Q2 - What is the ovarian cancer risk if I have a family history of ovarian cancer?

Ovarian cancer occurs by chance in less than 2 of every 100 women. For women who have a family history of ovarian, breast and/or bowel cancer and/or a faulty gene, the risk can be much higher than 2 in 100. Generally the risk is quite low in young women and rises with age.

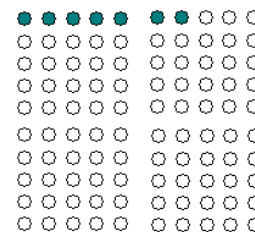
Studies have shown that out of 100 women with one first degree relative (mother, sister or daughter) with ovarian cancer, 5 will develop ovarian cancer themselves at some point in their life. Women who have 2 or more first (mother, sister or daughter) and/or second degree (aunt, grandmother etc.) relatives with ovarian cancer have an even higher chance of developing ovarian cancer themselves of about 7 out of 100.



Less than 2 out of 100 women in the general population get ovarian cancer in their lifetime



About 5 out of 100 women with a first degree relative with ovarian cancer get ovarian cancer in their lifetime



About 7 out of 100 women with two or more relatives with ovarian cancer get ovarian cancer in their lifetime

1. Cancer risk

3

The number of relatives with cancers other than ovarian cancer, such as breast and bowel cancer, can also affect a woman's chances of developing ovarian cancer. A woman's risk is estimated by genetics services once the full family history is available.

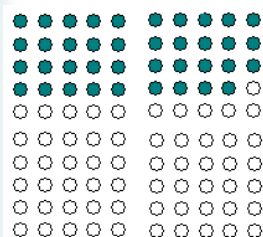
Q3 - How can I find out whether I have a faulty gene?

Some women might come from families where certain types of cancer (e.g. ovarian, breast, bowel) have affected more than just one or two relatives and these women might have a faulty gene. To get tested for a faulty gene you will need to be referred to genetics services (usually by your GP). There your family history of cancer will be assessed in detail and you will be told whether there might be a faulty gene involved.

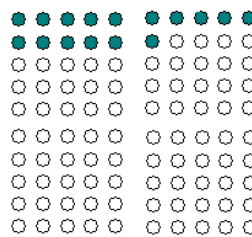
Testing for a faulty gene in the family then usually begins with a test on a blood sample from a person who has had cancer. If you have had cancer yourself you may be offered a genetic test as the first person in your family. If you have not had cancer yourself, then a relative who has had cancer will be offered a genetic test first. If a faulty gene is found, other members of the family can then have a genetic test to see whether they also have the same gene. If there is no living relative with cancer, or the relative doesn't want to get tested, then it may not be possible to do a genetic test for you. For sources of more information see chapter 9 (Contacts and Resources), page 27.

Q4 - What would my risk be if a faulty gene is found?

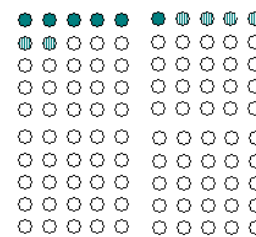
The most common faulty genes linked to ovarian cancer are called Breast Cancer (BRCA) genes 1 and 2 and genes common in Lynch Syndrome (formerly known as Hereditary Non-Polyposis Colorectal Cancer or HNPCC). The lifetime risk of developing ovarian cancer can be much higher for women who have a faulty gene compared to the general population.



About 39 out of 100 women with a BRCA1 faulty gene get ovarian cancer by the time they are 70



About 16 out of 100 women with a BRCA2 faulty gene get ovarian cancer by the time they are 70



About 6 to 12 out of 100 women with Lynch Syndrome get ovarian cancer by the time they are 70

Q5 - What if I cannot get tested or my genetic test is unclear?

For some women it might not be possible to get tested because they have no living relative with cancer or their relative does not want to be tested. In other cases a genetic test may come back as 'unclear' which means the result is not certain. In this case the genetics service will estimate the woman's risk based on her family history.

If you have been told that your family history looks like you might have a faulty gene and you would like more specific information for people with that gene, you can look at the information by personalising this aid and selecting the gene that most closely resembles your family history as you have been told by your genetic counsellor

Q6 - Does my cancer risk change over time?

Yes. Your so-called 'lifetime' risk (which is the risk that you will get ovarian cancer at some point in your life) stays the same over time. However, your 'age-related' ovarian cancer risk (which is the risk that you will get ovarian cancer in a specified time frame, for example one year or five years) increases with age. This means when you are young your age-related risk is relatively low and rises when you get older. So for example, your risk of getting ovarian cancer in the next year when you are 35 is quite low, whereas your risk of getting ovarian cancer in the next year when you are 50 is higher.

Q7 - How will my ovarian cancer risk affect my life?

Being at risk for ovarian cancer does not necessarily mean that you will develop ovarian cancer. However, knowing your risk will give you access to additional medical services such as genetic counselling and means that you are offered risk-reducing surgery. Some people may feel worried or anxious after finding out about their risk and their quality of life may be affected.

Q8 - How will my ovarian cancer risk affect my children?

There is a chance that your children will also have an increased risk of ovarian cancer. Their risk depends on how high your own risk is and on whether or not you have a faulty gene. If you have a faulty gene, then your children have a 50/50 chance to inherit this gene. If they inherited the gene, then their cancer risk would be the same as yours. If they did not inherit the gene, then their cancer risk would be low. Boys who inherit the gene are not at risk of ovarian cancer. Please also note that cancer risk increases with age and therefore children's risk only starts to rise in later life. More information about genetics can be found on the NHS choices website (see Chapter 9 Contacts and Resources, page 27).

You can discuss your children's risk with your doctor to find out more. Your doctor will also be able to advise you about how best to discuss this risk with your children.

Q9 - How can I deal with my cancer risk?

If you would like to find out more about your risk and what it means, you can use the resources provided in OvDex and talk to your doctor or genetic counsellor.

Knowing that you are at increased risk of cancer might make you feel worried or angry or frustrated. This is completely normal and there is no need to bottle those feelings up. It can actually help to talk about them. You can try to find someone you trust to talk to or if you prefer to talk to someone you don't know, you can use the contact details for charities and patient support networks under Contacts & Resources.

If you have a partner, sharing your thoughts and concerns with your partner can be useful in helping them understand what you are going through and in helping you cope together as a couple. It may also help to see a positive side to knowing your risk status. For example, this means that you will get access to genetics services and have the chance to do something about your risk. Other women have found it helpful to stay optimistic, have joined patient networks or have found strength in their faith.

Being at risk of cancer may sometimes lead to unwanted thoughts and worry, especially at times when there are no active steps you can take, for example when you are waiting for genetic test results. There are useful techniques you can use to reduce such thoughts. One such method is active distraction, which means actively thinking about something else whenever unwanted thoughts pop in your head. Watch the 'How do I cope' video on the Cancer Genetics Storybank website for a guide of how to do this.

Relaxation techniques, meditation and guided imagery may also help you deal with any stress you might experience relating to your cancer risk. Examples include progressive muscle relaxation and certain breathing exercises. Visit the 'Mind-body therapy' section on the Macmillan website for more information on and guides to mind-body therapies.

Questions in chapter 2:

- Q1 What can I do to reduce my risk?
- Q2 Why is there a choice?
- Q3 Who should decide?
- Q4 Who else should I involve in this decision?
- Q5 When should I decide?
- Q6 How can I decide?
- Q7 How can I deal with the choice?

Q1 - What can I do to reduce my risk?

Your doctor has probably discussed with you the option of having risk-reducing ovarian surgery to reduce your ovarian cancer risk. You will need to decide if and when to have this surgery. Unfortunately, there is no medically proven screening available on the NHS for ovarian cancer at the moment.

This decision aid is designed to help you look at your options. Please refer to chapter 3 (Options at a glance) page 8, chapter 4 (Risk-reducing surgery) page 10 and chapter 7 (Other options) page 20 for more information about surgery, screening and other alternatives.

Q2 - Why is there a choice?

Often when you go to your doctor, there is one clearly recommended treatment. However, some situations are more complicated. These are situations in which your personal preferences play an important role. In the case of cancer risk reduction, your options have very different effects on your life. This means that you need to be clear about what might happen if you choose one option over the other.

Some health professionals may recommend surgery quite strongly; however you need to make the final decision and should be aware of the possible benefits and risks before you do. You should certainly not feel pressured to opt for surgery.

Q3 - Who should decide?

As the best choice for you is based on your preferences, you should be closely involved in the decision. You can either make the decision on your own or if you do not wish to make this decision yourself, your doctor can help you. They will encourage you to think about the options and your preferences, so that the final choice is right for you. If you then don't want to choose for yourself, just say so and your doctor may make a recommendation.

Q4 - Who else should I involve in this decision?

Whether or not you would like to bring anyone else into this decision is your choice. Often it is helpful to speak to your partner, other members of your family or some friends who could work through this decision with you. Especially with a decision about risk-reducing ovarian surgery, the views of your partner can be important, so it is recommended that you speak to your partner and try to reach a decision together. Your doctor will also be happy for you to bring your partner along to appointments and to answer any questions they might have.

Q5 - When should I decide?

The decision you are facing is not an easy one and you should not feel under any pressure to decide quickly. Risk-reducing surgery has benefits and risks that need to be weighed carefully, so take your time and make sure you are ready before making a decision. It is important that you understand that undergoing surgery will mean you can no longer get pregnant. Therefore it is essential to consider your plans for a family and any potential future changes to these plans (for example if there is a chance you might meet a new partner / re-marry) before making a final decision.

Q6 - How can I decide?

When it comes to important decisions everyone is different. Some people like to find out as much as they can about their options, while others prefer to just know what is absolutely necessary. Some might find it helpful to talk to their family and friends. Some might like to speak to people who have made a similar decision. It really depends on you. Have a think about other important decisions in your life and how you managed to make those. That could give you an idea of how you like to decide about things.

Q7 - How can I deal with the choice?

It can be helpful to create a plan of how and when you will make this choice. If you are not ready to decide right now, it might be useful to set yourself a deadline of when you will revisit this decision. For example: "Just after my 40th birthday I will look at this information again." or "Once I have completed my family I will revisit this decision." Once you are ready you can decide how you want to make this choice:

1. I will decide by myself using everything I have learnt
2. I will decide but seriously consider my doctor's opinion
3. The doctor and I should decide together
4. The doctor should decide but seriously consider my opinion
5. The doctor should decide for me

One constructive way to deal with a difficult decision is to empower yourself with information. OvDex is designed to help you to learn more about ovarian cancer risk and your options. With the wealth of information that is available on the internet, it can be difficult to find reliable and trustworthy information. The information in OvDex is supported by recent scientific findings and has been carefully reviewed by health professionals to make sure it is accurate. You should at least understand your options and their benefits and risks before making a decision. Find out more about the most important questions to ask at: www.ask3questions.co.uk. Once you have read the information in OvDex, it could help to make a note of any remaining questions and take those to your doctor or genetic counsellor for a more detailed discussion.

You may feel that you are not comfortable making decisions about your health. This is okay. You do not need to make the choice alone if you don't want to. You can decide together with your doctor or ask them to make the choice for you. But you need to remember that you are the expert when it comes to your own life and that only you know what is important to you. So even if you decide to let the doctor make the decision for you, make sure they know about your goals and values. Tell them what is important to you.

3. Options at a Glance

| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|---|---|--|
| Will this reduce my ovarian cancer risk? | Yes, removing the ovaries and fallopian tubes will greatly reduce your lifetime risk of ovarian-type cancer to less than 2 in 100. | No, your risk of ovarian cancer will remain high. |
| Will I be able to become pregnant? | No. | Yes. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. | You may worry about developing ovarian cancer. |
| Will I go into menopause? | Yes, your oestrogen levels fall and the menopause will start immediately. | No. |
| What is menopause after surgery like? | Menopause after surgery is similar to natural menopause, but because it happens suddenly, the symptoms may be more severe. | Not applicable. |
| Will this change how I feel about myself as a woman? | Many women do not notice any change in how they feel about themselves as a woman. However, your desire for and/or enjoyment of sex may decrease a lot. | Not applicable. |
| Are there any risks linked to menopause? | Yes, there is a risk of bone thinning and cardiovascular disease. Some patients report memory changes as well. | Not applicable. |
| Will I need hormone replacement therapy (HRT)? | HRT can reduce some of the effects of sudden menopause. You will need to discuss this possibility with your doctor. | Not applicable |
| Is HRT safe for women at increased familial risk? | HRT is safe to take for the majority of women until they are 50. It is not recommended for women who have had breast cancer themselves already. | Not applicable |
| What are the risks of surgery? | There is a small risk of complications depending on the type of surgical procedure (key hole or open surgery) you have. Discuss this with your doctor. | Not applicable. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after key hole surgery and are back to normal in 4 weeks. For open surgery this is slightly longer. | Not applicable. |
| Is there a routine screening programme to detect ovarian cancer? | No, and after surgery there is no need for any screening because your risk will be low. | No, there is no routine screening available on the NHS to detect ovarian cancer. |

Q1 – How can I deal with this decision?

Some women may find it useful to discuss surgery with others, who have already made this decision. In some areas your genetic counsellor may be able to put you in touch with someone who has already gone through the decision, otherwise you can use the contact details for charities and patient support networks under Contacts & Resources to read about or get in touch with others in a similar situation. Going over the decision with someone who knows you well, such as your partner or a member of your family can also help you clarify your decision. If you decide to opt for surgery and you are in a relationship, preparing yourself together with your partner for the time after surgery can help you both deal better with any consequences of the operation.

Being clear about the reasons why surgery could be the right or wrong thing for you can help you make a decision and avoid regretting it later. The information in OvDex is designed to help you identify the benefits and the risks of surgery and any other options, so you can decide for yourself. The exercise called 'Your Decision' (page 22) can help you clarify what is most important to you.

Being at risk of cancer may sometimes lead to unwanted thoughts and worry, especially at times when there are no active steps you can take, for example if you have decided to defer surgery or while you are waiting for your surgery appointment. There are useful techniques you can use to reduce such thoughts. One such method is active distraction, which means actively thinking about something else whenever unwanted thoughts pop in your head. Watch the 'How do I cope' video on the Cancer Genetics Storybank website for a guide of how to do this.

Relaxation techniques, meditation and guided imagery may also help you deal with any stress you might experience relating to your cancer risk. Examples include progressive muscle relaxation and certain breathing exercises. Visit the 'Mind-body therapy' section on the Macmillan website for more information on and guides to mind-body therapies.

Questions in chapter 4:

- Q1 What is risk-reducing bilateral salpingo-oophorectomy (RRSO)?
- Q2 What are the main advantages of this operation?
- Q3 What are the main disadvantages of this operation?
- Q4 What is my risk after surgery?
- Q5 What does the surgery involve?
- Q6 How long does it take to recover?
- Q7 Do I need to have my womb removed as well?
- Q8 What are the complications of RRSO?
- Q9 Could cancer be found during the surgery?
- Q10 How would RRSO affect my life?
- Q11 How could I deal with RRSO?

Q1 - What is risk-reducing bilateral salpingo-oophorectomy (RRSO)?

Risk-reducing bilateral salpingo-oophorectomy (RRSO for short) is surgery to remove healthy ovaries and fallopian tubes to reduce the cancer risk. The term 'bilateral' means that the ovaries and fallopian tubes on both sides of the body are removed. The word 'salpingo-oophorectomy' means removal of the fallopian tubes and ovaries. The aim of the surgery is to remove these tissues before cancer develops.

Q2 - What are the main advantages of this operation?

- 1) This operation will reduce your ovarian and fallopian tube cancer risk, as these are completely removed during surgery. For women with a faulty BRCA1 gene cancer might still develop in the peritoneum, which is the lining of the pelvis and abdomen. This means RRSO will not prevent cancer in about 2 in every 100 women with a BRCA1 faulty gene. This risk is about the same as the ovarian cancer risk in the general population. Even fewer cancers of the peritoneum have been found in women without a faulty BRCA1 gene who have had this surgery, therefore their risk is likely to be much lower than 2 in 100 after surgery.
- 2) For some women, not having to worry about ovarian cancer anymore comes as a great relief.
- 3) For the vast majority of women having RRSO before the natural menopause could also reduce the risk of breast cancer.
- 4) Having the surgery will also prevent other, non-cancer related problems happening with the ovaries, such as cysts.

4. Risk-reducing Surgery

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Q3 - What are the main disadvantages of this operation?

- 1) You will no longer be able to become pregnant once you have had surgery, therefore it is important that you have considered your plans for a family before making a final decision.
- 2) After the operation you will immediately enter the menopause and may start to experience the typical menopausal symptoms, including hot flushes, mood swings, night sweats and vaginal dryness (Please see Chapter 5 (The Menopause), page 14 for more details). Furthermore, you may be at increased risk of bone thinning, cardiovascular (heart & blood vessel) disease and memory changes. These effects may be effectively treated with hormone replacement therapy (Please see chapter 6 (Hormone Replacement), page 17 for more details), however not all women can take hormone replacement and this should be discussed before having surgery.
- 3) Your desire for sex will decrease and due to some menopausal symptoms, such as vaginal dryness, you may enjoy sex less as intercourse can be painful.
- 4) There is a small risk of complications during and after surgery.

Q4 - What is my risk after surgery?

After surgery your risk of ovarian and fallopian tube cancer is greatly reduced, however, there is a rare type of cancer which may still develop after surgery that is very similar to ovarian cancer. This is called primary peritoneal cancer and develops in the lining of the abdomen and pelvis. Your risk of primary peritoneal cancer depends on whether or not you have a faulty gene. The highest risk for primary peritoneal cancer is in women with a BRCA1 faulty gene. Their risk is about 2 in every 100 women, which is similar to the risk of ovarian cancer in the general population. For women who do not have a BRCA1 faulty gene the risk of primary peritoneal cancer after surgery is much lower than 2 in 100.

Q5 - What does the surgery involve?

Most often the operation is done as keyhole surgery (laparoscopy). This involves 3 or 4 small (about 1cm) cuts, usually one cut is close to your tummy button and 2-3 just below the bikini line, so they are not visible. Keyhole surgery itself usually takes less than 2 hours.

In some cases it may not be possible to do keyhole surgery, because of previous surgery on your tummy or because of your weight. Then surgeons use the more traditional open surgery. This means a longer cut, usually along the bikini line. Sometimes a surgeon might have to convert to an open surgery when doing keyhole surgery because of complications or old scar tissue. This happens in about 1 of every 100 keyhole operations.

**Type of procedure in 100 women who have the operation**

The figure on the left shows **how many of 100 women will have keyhole surgery, open surgery** and planned keyhole which is converted to open surgery.

94 of 100 women will have planned keyhole surgery (white dots). Five of 100 women will have planned open surgery (light teal dots). One of 100 women will have planned keyhole surgery which is converted to open surgery (dark dot).

4. Risk-reducing Surgery

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Q6 - How long does it take to recover?

75 of 100 patients who have keyhole surgery leave the hospital the day after surgery. They are usually back to normal activity about 4 weeks after surgery. If you have open surgery you are likely to stay in hospital a bit longer than with keyhole surgery. Usually patients leave the hospital about 5 days after open surgery and are back to normal in about 6 weeks. After surgery you are not allowed to do any heavy lifting for a few weeks. You may also have to refrain from driving until you can comfortably wear a seatbelt and make an emergency stop without pain.

Q7 - Do I need to have my womb removed as well?

No, normally it is not necessary that you have your womb removed at the same time as having RRSO, unless you know that you have Lynch Syndrome or you have other problems with your womb and have been told by a doctor that removing the womb would help you with those problems. However, you may chose to have your womb removed at the time of RRSO, as it may affect the type of hormone replacement you would be given (see Chapter 6 (Hormone replacement), page 17). You should discuss this with your doctor.

Q8 - What are the complications of RRSO?

There is a small risk of complications linked to RRSO. About 4 in every 100 people will experience some complication. Minor complications can include wound or urinary tract infections and usually have no long-term effects on your health. More serious complications might happen during surgery and can include damage to blood vessels, the bowel or the bladder. If you are having keyhole surgery this might mean that the surgeon has to convert to open surgery to repair the damage. There are a number of other rare complications that might happen and your surgeon will go through those with you if you wish before you go in for surgery. Please note that about 96 of every 100 women do not experience any complications at all.

Complications in 100 women who have the operation

The figure on the left shows **how many of 100 women will experience complications** during or after surgery.

4 of 100 women will experience some kind of complication either during or after surgery. 96 of 100 women will not experience any complications.

As with any surgery, there is a very small risk of death. However, this is highly unlikely. The risk may be greater in women with health problems before surgery.

Q9 - Could cancer be found during the surgery?

Yes, although this is rare. Once your ovaries and fallopian tubes have been removed they will be checked thoroughly for any signs of cancer. There is a chance that cancer may be discovered during this check. This happens in up to 4 out of every 100 operations in the highest risk patients. For others the chance of finding cancer during the surgery are much lower.

Q10 - How would RRSO affect my life?

In the short-term, if you decide to go for surgery, you will have to take time off work and will not be able to do some of the things you might usually do, such as driving or heavy lifting. However most women recover fully within 4 to 6 weeks.

In the longer term there are a number of things you should consider. After surgery you may feel less worried about ovarian cancer. You will no longer be able to become pregnant and you will enter the menopause if you have not gone through it yet (Please see chapter 5 (Surgical menopause), page 14). You may opt to take hormone replacement (Please see chapter 6 (Hormone replacement), page 17). These factors may affect your life after surgery and should be considered carefully before making a decision.

Questions in chapter 5:

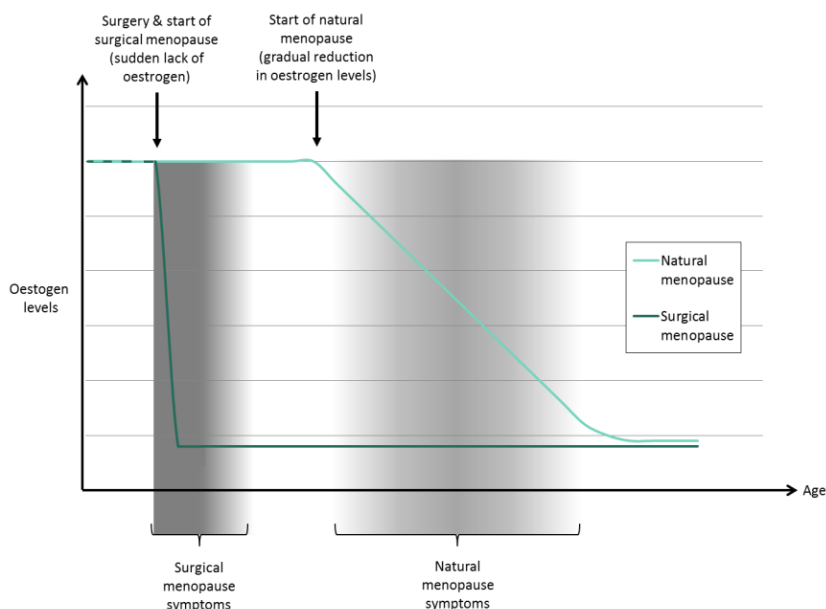
- Q1 What is surgical menopause?
 Q2 What is the menopause like?
 Q3 How could the menopause affect my life in the short term?
 Q4 How long does surgical menopause last?
 Q5 Are there any long term health effects linked to surgical menopause?
 Q6 Is there anything I can do to reduce the effects of surgical menopause?



Please note that the information about symptoms of the menopause in this chapter describes symptoms experienced by women who do not take hormone replacement therapy (HRT). Most symptoms of the menopause are reduced by hormone replacement therapy (for more information see Chapter 6 (Hormone Replacement) on page 17).

Q1 - What is surgical menopause?

Surgical menopause is the menopause you enter when your ovaries are removed by surgery and your body no longer produces the female hormone oestrogen. The surgical menopause is essentially the same as the natural menopause, it only happens earlier and more suddenly. Therefore surgical menopause results in the same symptoms as the natural menopause. These symptoms are caused by the lack of oestrogen. As oestrogen levels drop suddenly after surgery, the symptoms of surgical menopause start instantly and can be more severe than those of the natural menopause, in which oestrogen levels fall gradually.



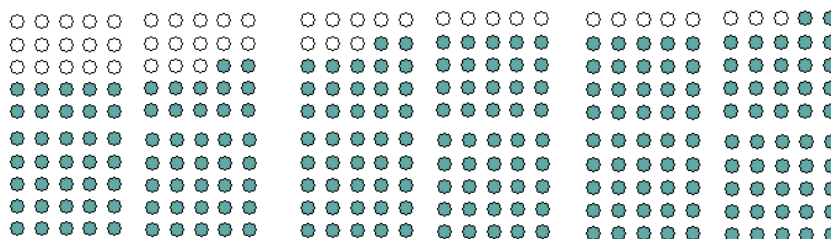
The graph above shows the drop in the levels of the female hormone oestrogen during surgical and natural menopause. Potential menopausal symptom duration and severity are indicated in shades of grey.

Q2 - What is the menopause like?

It is very difficult to describe what the menopause is like, because it is different for every person. Some women have no or very few symptoms and cope very well. Other women can have very severe symptoms and their quality of life can be affected.

Common menopausal symptoms you may experience are: Hot flushes, night sweats, mood swings, vaginal dryness and loss of interest in sex. Other symptoms include difficulty sleeping, fatigue, weight gain, changes in memory and depression.

The great majority of women who had surgery report that they experience one or more of the common symptoms. However, any data available about menopausal symptoms is from small studies and each study uses different ways to assess what women experienced, so it is difficult to predict what the menopause will be like for you personally. The pictures below show how common different types of symptoms are according to one study of women with a family history of ovarian cancer.



About 72 of 100 women experience vasomotor symptoms (hot flushes and night sweats)

About 87 of 100 women experience physical and sexual symptoms (aching, vaginal dryness, loss of libido)

About 92 of 100 women experience psychosocial symptoms (mood swings, feeling depressed or down)

Q3 - How could menopause affect my life in the short term?

While none of the menopausal symptoms mentioned above are dangerous for your health, they may affect you in different ways. Hot flushes can be very uncomfortable and may happen at inconvenient times, for example when you are giving a presentation at work. In one study two out of every three women reported having hot flushes after surgery. Night sweats can lead to problems with sleep and insomnia. Vaginal dryness can lead to pain during sex and therefore you may enjoy sex less. Loss of interest in sex, which is also experienced by many women after surgery, may affect your relationship with your partner. In one study just over half of women reported that they were not satisfied with their sexual functioning after having surgery and in another study one in every three patients felt that vaginal dryness was bothersome and reported pain with sex. The way that these symptoms might affect your life depends on your personal situation.

There are several options available to help you deal with the short-term symptoms of the menopause. These include physical options, such as lubricant to counteract vaginal dryness, or psychological options, such as sex counselling and cognitive behavioural therapy to improve emotional and physical functioning. You can get more information about these options from your genetics services. The NHS choices website also has information about dealing with the menopause (See Chapter 9 (Contacts & Resources), page 27).

Q4 - How long does the menopause last?

It is impossible to say how long symptoms will last. Some women have hardly any problems and symptoms disappear quickly, while others have symptoms for many years. It really is different for everyone.

Q5 - Are there any long term health effects linked to the menopause?

Yes. The loss of oestrogen is thought to affect long term health, particularly the bones and the cardiovascular system (heart and blood vessels). In older age this can lead to a higher risk of fractures due to bone thinning (osteoporosis) and a higher risk of cardiovascular disease (heart disease, stroke). It should be noted that these effects are seen in all women after the menopause, whether it was a surgical or natural menopause. Your personal risk depends on your lifestyle and personal and family history of osteoporosis and cardiovascular disease and should be discussed with your doctor.

Due to the sudden lack of oestrogen after surgery, the risk of bone thinning and cardiovascular disease may start to rise immediately after surgery. This could be before the woman has reached the age of the natural menopause. Therefore, women who undergo surgery before the age of 45 are recommended to take hormone replacement to reduce these effects until they reach the age of the natural menopause (See Chapter 6 (Hormone replacement) page 17).

Some women also report changes in memory following the menopause, which is again due to the lack of oestrogen.

Q6 - Is there anything I can do to reduce the effects of the menopause?

Yes. Hormone replacement therapy (HRT) is usually recommended for women who have RRSO before the age of 45 and have not had breast cancer themselves (See Chapter 6 (Hormone replacement) page 17). HRT may effectively reduce some of the short- and long-term effects of surgical menopause. However, not all women can take hormone replacement and this should be discussed before having surgery.

If you do not wish to or cannot take HRT then you can take dietary supplements to reduce bone thinning. You should speak to your doctor about this. In some regions in the UK patients are also recommended to have a bone density scan about 18 months after their operation. You can discuss this with your doctor.

An active lifestyle and healthy diet is also recommended.

6. Hormone Replacement

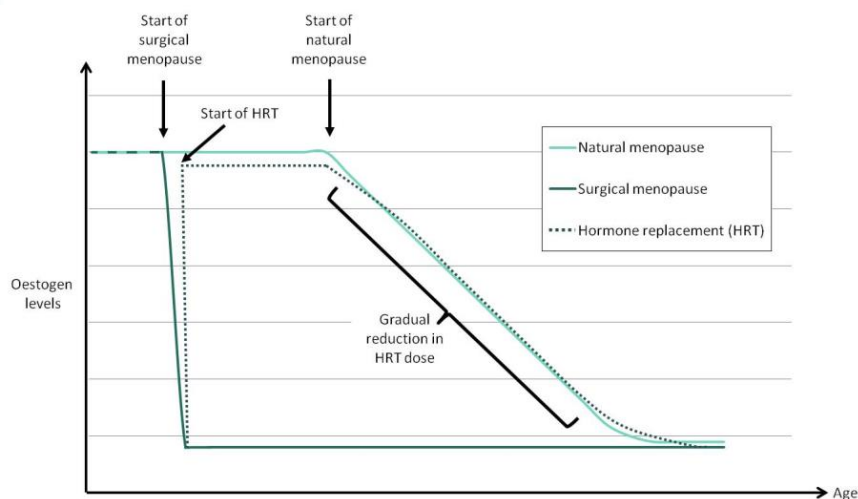
17

Questions in chapter 6:

- Q1 What is Hormone Replacement Therapy (HRT)?
 Q2 Why is HRT important?
 Q3 Who should take HRT?
 Q4 How long should I take HRT?
 Q5 Can I take HRT if I have a family history of breast cancer?
 Q6 But I have heard that HRT increases breast cancer risk?
 Q7 Will HRT reduce all the symptoms of menopause?
 Q8 Will HRT deal with the long term health effects of the menopause?
 Q9 Are there any alternatives to HRT?

Q1 - What is Hormone Replacement Therapy (HRT)?

Hormone Replacement Therapy, or HRT for short, is a medical treatment that puts back the female hormones that are lost when the ovaries are removed or when they stop functioning.



The graph above shows the oestrogen levels after surgical and natural menopause. It also shows the effect of HRT on the oestrogen levels after the operation if surgery was performed before the natural menopause.

There are a number of ways that HRT can be used:

- It can be taken as oral tablets usually once a day
- It can be applied as patches that are put on the tummy or bottom about once or twice a week
- It can be applied as a gel directly to the skin once a day (for example the lower abdomen or inner thigh)

There are two types of hormone replacement, one includes oestrogen and progesterone and the other includes oestrogen only. Women who only have their ovaries removed will usually be given oestrogen and progesterone HRT, while women who have their womb/uterus and ovaries removed (RRSO plus hysterectomy) are usually given oestrogen only HRT.

Q2 - Why is HRT important?

There are two main reasons why HRT is recommended after surgery:

1. To reduce the risk of bone thinning
2. To reduce symptoms of the menopause

Q3 - Who should take HRT?

HRT is strongly recommended for women who have surgery before they are 45 years of age, as this group is most affected by the long term health issues that are linked to loss of oestrogen, especially bone thinning (osteoporosis).

Women who have surgery when they are over 45 years of age can choose to have HRT to reduce the symptoms of surgical menopause. However these women do not have to have HRT if they do not want to, as the long term health effects of early loss of oestrogen do not affect them, as their oestrogen levels would have started to fall anyway.

It is important that the possibility of HRT is discussed before you decide about surgery.

Q4 - How long should I take HRT?

If you use HRT, it is recommended that you take it until the age of the natural menopause, which is 50 years in the UK. From the age of 50 your oestrogen levels would have declined naturally, so taking HRT for many years beyond 50 is not recommended. This is because there is controversial evidence about the benefits and risks of HRT if it is taken beyond the age of natural menopause. Therefore the health effects are unclear and HRT may cause more harm than good if taken beyond 50.

Q5 - Can I take HRT if I have a family history of breast cancer?

Yes. You should be able to take HRT even if you have a family history of breast cancer. Several studies have shown that HRT is safe to take for the vast majority of women with a family history of breast cancer as long as they have not had breast cancer themselves and they only use HRT until the age of the natural menopause. You should discuss your family history of breast cancer with your genetics service before deciding to have surgery.

Q6 - But I have heard that HRT increases breast cancer risk?

Yes, HRT contains oestrogen and there is some evidence that higher amounts of oestrogen in the body can increase the risk of breast cancer. However, the study that showed that HRT increases breast cancer risk was done with older women who had already gone through the natural menopause and were taking HRT beyond 50 years of age. Therefore these women were taking oestrogen when their body has naturally stopped producing this hormone. Furthermore, these women did not have surgery to have their ovaries removed. Therefore this group of women is very different to women with a family history who choose to have their ovaries out before they are 50 years. Furthermore you should be aware that the findings of this study have now been widely criticized.

Having your ovaries out removes all the natural oestrogen that your ovaries would be producing until the age of the natural menopause. The amount of oestrogen that is added back by taking HRT is less than the amount your ovaries would have produced naturally. If you stop taking HRT at the age of the natural menopause (so when you are 50) then there is no evidence that the breast cancer risk is increased.

Q7 - Will HRT reduce all the symptoms of menopause?

No. Taking HRT is not the same as having your own hormones. HRT cannot reduce all symptoms of the menopause and women who have had surgery and are on HRT do report more symptoms than women who have not had surgery. However, HRT can relieve some of the symptoms of the menopause. For example, women who took HRT reduced the average number of hot flushes from about 4 to about 1 per day when compared to women who did not take HRT. Women on HRT also had fewer night sweats.

Q8 - Will HRT deal with the long term health effects of the menopause?

Yes. HRT will reduce the risk of bone thinning and changes in memory functioning linked to the loss of oestrogen. However, the evidence of HRT's effects on cardiovascular disease (heart disease and stroke) is more controversial and should be discussed with your doctor in terms of your personal and family history of cardiovascular disease as well as your lifestyle, which can also affect your risk of cardiovascular disease.

Q9 - Are there any alternatives to HRT?

Yes. There are numerous non-hormonal alternatives to HRT, none of which have been shown to be as effective in relieving menopausal symptoms. For people who cannot take HRT these alternatives may be a good option but for those who can, HRT is the best option.

Alternatives include:

- Antidepressants, such as Selective Serotonin Reuptake Inhibitors (SSRIs)
- Gabapentin
- Red Clover
- Ginseng
- Evening primrose oil
- Agnus Castus
- And many more

Some of these options may be prescribed by your doctor, while others can be bought in health shops or over the internet. However, there is no medical proof that freely available herbal options work. These alternatives to HRT may have side-effects and you should always consult your doctor before deciding to use any of these options.

Questions in chapter 7:

- Q1 Is there any screening available for ovarian cancer?
 Q2 What is the alternative to surgery?
 Q3 But I have been offered CA125 blood tests and/or trans-vaginal scans?
 Q4 What would happen if I do not have surgery?
 Q5 Is there anything else I can do?

Q1 - Is there any screening available for ovarian cancer?

No. Unfortunately there is no medically proven screening available for ovarian cancer. A large trial of ovarian cancer screening finished in December 2011 and the results will not be available until 2013, so we do not yet know whether this screening is effective and can detect ovarian cancer early. Until the results of the trial are available the NHS will not offer routine screening. The trial offered women CA125 blood tests every four months and one yearly trans-vaginal ultrasound scan. Some areas or GPs may offer private, self-funded CA125 and/or trans-vaginal screening to high risk women, however women need to be aware that there is still no proof that this screening is effective in detecting ovarian cancer early. You should also be aware that the yearly smear test you receive is designed to detect cervical cancer and will not detect ovarian cancer.

Women from families with Lynch syndrome may receive trans-vaginal ultrasound screening to look for womb cancer. The ovaries may be visible on these scans and if something is found the GP may order a CA125 blood test. In this case the CA125 blood test is a diagnostic test and not a screening test.

Any woman with symptoms of ovarian cancer will be offered a diagnostic CA125 test and trans-vaginal ultrasound scan, however this is not part of a screening programme. Women with no symptoms will not be offered routine screening on the NHS until the screening has been shown to be effective.

Q2 - What is the alternative to surgery?

At the moment the official alternative to surgery is to do nothing and simply be aware of the symptoms of ovarian cancer should they develop. However it is important to realise that these symptoms can be very vague and are not specific to ovarian cancer. If any of these symptoms happen more than 12 times a month you should contact your GP.

The symptoms include:

Persistent bloating (big or swollen tummy)
 Feeling less hungry or feeling full quickly
 Persistent pain in your tummy or below
 Needing to urinate more than usual

Q3 – But I have been offered CA125 blood tests and/or trans-vaginal scans?

Some centres and/or GPs may offer private CA125 tests or trans-vaginal scans to women at high risk of ovarian cancer. As these are not offered as screening tests by the NHS you may have to pay for these yourself. You can choose to have these tests done, but you should be aware that there is no evidence that these screening tests are effective at detecting ovarian cancer early.

Women from Lynch syndrome families, may be offered hysteroscopies or trans-vaginal ultrasound scans to detect uterus/womb cancer, which these women are at higher risk for. However usually these tests are designed to detect womb cancer only and not ovarian cancer.

Q4 - What would happen if I do not have surgery?

If you decide not to have surgery, then nothing will really change. You will need to look out for any symptoms and if you think anything is wrong you need to go to your GP to get it checked. Make sure you tell your GP that you have a family history of ovarian cancer and are at increased risk when you talk to them.

Remember you can reconsider surgery at any time.

Q5 - Is there anything else I can do?

As there is no routine screening available, you can choose to have screening privately, but you should be aware that this has not yet been shown to be effective. You should also keep an eye on any symptoms. Studies have found that a healthy diet with plenty of fruit and vegetables, keeping a healthy weight and an active lifestyle can improve overall well-being and might reduce your chances of getting cancer.

In some countries it is recommended that women at high risk of ovarian cancer take the oral contraceptive pill when they are pre-menopausal. The oral contraceptive pill reduces ovarian cancer risk by up to half, but it has also been found to increase breast cancer risk slightly. For women at high risk the reduction in ovarian cancer risk is thought to outweigh the slight increase in breast cancer risk. Whether the oral contraceptive pill might be an option for you depends on your risk and family history of breast cancer and should be discussed with your doctor.

8. Your Decision (Example)

22

In the exercise on the next few pages you can rate some facts depending on how much they make you want **to have** or want **to avoid** surgery. Give each fact a ranking number (e.g. from 1 (a little) to 3 (a lot), then add the numbers up to see where you stand.

The table below is already filled in to give you an example. On page 24 you will find an empty table for you to fill in.

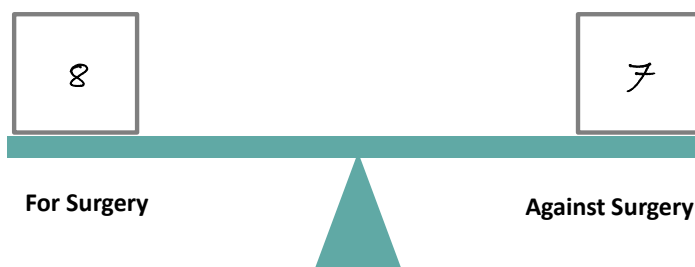
| The fact that... | Makes me want to have surgery | Does not come into my decision | Makes me want to avoid surgery |
|--|--------------------------------------|--------------------------------|---------------------------------------|
| ...surgery will reduce my ovarian cancer risk | 3 | | |
| ...surgery will reduce my breast cancer risk | 2 | | |
| ...I will not be able to become pregnant after surgery | | X | |
| I will go into surgical menopause after surgery | | | 3 |
| There is no effective screening for ovarian cancer | 2 | | |
| The symptoms of ovarian cancer are very vague | 1 | | |
| I might feel different about my body after surgery | | | 2 |
| I might enjoy sex less after surgery | | | 1 |
| Enter you own reasons: | | | |
| Have to take time off work | | | 1 |
| | | | |
| | | | |
| | | | |
| Add together the numbers in each column: | 8 | | 7 |

8. Your Decision (Example)

23

Below you can enter the overall number into the scales. Then note down how you feel about the decision and where you stand.

The scales below are already filled in to give you an example. On page 25 you will find an empty scale for you to fill in.

**More weight for than against surgery:**

Overall your answers show that at this moment in time you are leaning towards having the surgery. Even though you may have some reasons why you would want to avoid surgery, they are not as important to you as the reasons that make you want surgery. Therefore you could now make an appointment to discuss this decision with your doctor and go through any concerns you might have.

Same weights both sides:

Overall your answers show that at this moment you are completely undecided about whether or not to have surgery. Your reasons for why you would want to have and why you would want to avoid surgery balance each other out. Therefore you may want to discuss your options further with your doctor/genetic counsellor.

More weight against than for surgery:

Overall your answers show that at this moment in time you are leaning towards not having surgery. Even though you may have some reasons why you would want to have surgery, they are not as important to you as the reasons that make you want to avoid surgery. Therefore at this moment in time surgery is probably not the right option. However, keep in mind that things may change over time and that eventually you may lean more towards surgery. You can come back to OvDex and do this exercise again in a years time to see whether anything has changed.

Overall decision: My reasons for surgery weigh more than those
against surgery, but I am still not sure. The
numbers are very similar.

Next steps: I will make an appointment with my genetic
counsellor to discuss the menopause and find out
whether I can take HRT.
Will talk to my boss about taking time off and
whether it would cause any problems.

8. Your Decision

In the exercise on the next few pages you can rate some facts depending on how much they make you want **to have** or want **to avoid** surgery. Give each fact a ranking number (e.g. from 1 (a little) to 3 (a lot), then add the numbers up to see where you stand.

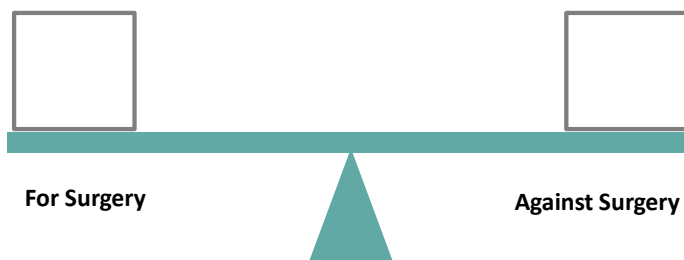
Make sure you give the score on the correct side of the table.

| The fact that... | Makes me want to have surgery | Does not come into my decision | Makes me want to avoid surgery |
|--|--------------------------------------|--------------------------------|---------------------------------------|
| ...surgery will reduce my ovarian cancer risk | | | |
| ...surgery will reduce my breast cancer risk | | | |
| ...I will not be able to become pregnant after surgery | | | |
| I will go into surgical menopause after surgery | | | |
| There is no effective screening for ovarian cancer | | | |
| The symptoms of ovarian cancer are very vague | | | |
| I might feel different about my body after surgery | | | |
| I might enjoy sex less after surgery | | | |
| Enter you own reasons: | | | |
| | | | |
| | | | |
| | | | |
| | | | |
| Add together the numbers in each column: | | | |

8. Your Decision

25

Below you can enter the overall number into the scales. Then note down how you feel about the decision and where you stand.



More weight for than against surgery:

Overall your answers show that at this moment in time you are leaning towards having the surgery. Even though you may have some reasons why you would want to avoid surgery, they are not as important to you as the reasons that make you want surgery. Therefore you could now make an appointment to discuss this decision with your doctor and go through any concerns you might have.

Same weights both sides:

Overall your answers show that at this moment you are completely undecided about whether or not to have surgery. Your reasons for why you would want to have and why you would want to avoid surgery balance each other out. Therefore you may want to discuss your options further with your doctor/genetic counsellor.

More weight against than for surgery:

Overall your answers show that at this moment in time you are leaning towards not having surgery. Even though you may have some reasons why you would want to have surgery, they are not as important to you as the reasons that make you want to avoid surgery. Therefore at this moment in time surgery is probably not the right option. However, keep in mind that things may change over time and that eventually you may lean more towards surgery. You can come back to OvDex and do this exercise again in a years time to see whether anything has changed.

Overall decision: _____

Next steps: _____

Q1 – How can I deal with my decision?

If you are considering surgery, it can help to think about the support you do have and how different people might be able to help you. This can include simple things such as driving you to and from the hospital, helping you out with household chores or childcare issues right after surgery.

Before committing fully to surgery you can discuss any further questions with your doctor or genetic counsellor and you can also seek a second professional opinion at any time if you wish.

If you decide not to have surgery it might be helpful speak to your GP and let them know about your risk. Some women also find it reassuring to have an action plan in case they feel like any symptoms develop. This can be a simple reminder, such as a plan to contact your doctor if symptoms last 2 weeks or longer.

Here are some links and contact details that you might find helpful (even though we have taken great care in choosing websites from reliable sources please note we are not responsible for the content of these websites):

The Cancer Genetics Story Bank – An online collection of stories told by patients and professionals about cancer genetics, developed by the Cancer Genetics Service for Wales (CGSW)

www.cancergeneticsstorybank.co.uk

Macmillan – A UK charity for anyone affected by cancer with good information about genetic testing for cancer and mind-body therapies

www.macmillan.org.uk or call the support line on 0808 808 00 00

NHS Choices – A website created by the NHS to provide information to patients. Includes information on ovarian cancer and genetics.

www.nhs.uk

Target Ovarian Cancer – A UK charity which supports research into ovarian cancer and provides useful information including an 'Ask the Expert' feature. Available in several languages.

www.targetovariancancer.org.uk

Ovacom – A UK charity providing information and support for everyone affected by ovarian cancer. Includes links to a number of patient blogs.

www.ovacom.org.uk or call the supportline on 08453710554

Your GP and/or Genetics Service – There to help you with any questions or concerns.

Evidence for cancer risk

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Chen S and Parmigiani (2007) Meta-analysis of BRCA1 and BRCA2 penetrance. *J Clin Oncol* 25(11): 1329–1333

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Evidence for cancer risk after surgery

Finch A, Beiner M, Lubinski J, Lynch HT, Moller P, Rosen B, Murphy J, Ghadirian P, Friedman E, Foulkes WD, Kim-Sing C, Wagner T, Tung N, Couch F, Stoppa-Lyonnet D, Ainsworth P, Daly M, Pasini B, Gershoni-Baruch R, Eng C, Olopade OI, McLennan J, Karlan B, Weitzel J, Sun P, Narod SA; Hereditary Ovarian Cancer Clinical Study Group (2006) Salpingo-oophorectomy and the risk of ovarian, fallopian tube, and peritoneal cancers in women with a BRCA1 or BRCA2 Mutation. *JAMA*. 296(2):185-92

Evans DG, Clayton R, Donnai P, Shenton A, Lalloo F. (2009) Risk-reducing surgery for ovarian cancer: outcomes in 300 surgeries suggest a low peritoneal primary risk. *Eur J Hum Genet*. 17(11):1381-5

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Evidence for surgical procedure and complication rate

Kenkhuis MJ, de Bock GH, Elferink PO, Arts HJ, Oosterwijk JC, Jansen L, Mourits MJ. (2010) Short-term surgical outcome and safety of risk reducing salpingo-oophorectomy in BRCA1/2 mutation carriers. *Maturitas*. 2010 Jul;66(3):310-4

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Finch A, Metcalfe KA, Chiang JK, Elit L, McLaughlin J, Springate C, Demsky R, Murphy J, Rosen B, Narod SA. (2011) The impact of prophylactic salpingo-oophorectomy on menopausal symptoms and sexual function in women who carry a BRCA mutation. *Gynecol Oncol.* 121(1):163-8

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Elit L, Esplen MJ, Butler K, Narod S. (2001) Quality of life and psychosexual adjustment after prophylactic oophorectomy for a family history of ovarian cancer. *Fam Cancer.* 1(3-4):149-56

Robson M, Hensley M, Barakat R, Brown C, Chi D, Poynor E. (2003) Quality of life in women at risk for ovarian cancer who have undergone risk-reducing oophorectomy. *Gynecol Oncol* 89(2):281-7.

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Appendix 7.7 - Actions in response to suggestions for improvements – Round 2

| Suggestions for improvements | Exact action taken / Decision |
|--|---|
| PATIENTS | |
| <p>I thought it was not quite so helpful in the 'negative genetic test, but the first one in the family to be tested' section (which is my personal situation). I thought that the comment about coming back when you'd got more information about the risk from your doctor might be unrealistic as you don't always get much more information that could be brought back to the tool. In practice, it might be good to refer to a discussion with a genetic counsellor as I found mine to be far more knowledgeable and helpful than any of the doctors!</p> | <p>- Changed wording to: "You can come back to this website once you have had a discussion with your genetic counsellor about your risk and about whether risk-reducing surgery is still something you might want to consider."</p> |
| <p>I thought that the initial surgery/no surgery section was rather weighted for surgery. There could have been a mention of the possibility of privately funded screening at this point.</p> | <p>- Added statements in the 'no surgery' column of the Option Grid to visually balance it more - Decided not to add information about screening as it is not medically proven nor recommended</p> |
| <p>The other place I did not find it helpful was the assumption that everyone has a partner. Not only was this unhelpful, it is also extremely irritating. Many women on their own find the 'Noah's Ark Mentality' tedious to say the least.</p> | <p>- Checked all docs that mentioned the word 'partner' and ensured that there is never an assumption that there is a partner (always 'if' statement) - In menopause docs reworded entire section: "Vaginal dryness can lead to pain during sex and therefore you may enjoy sex less. Additionally, many women also experience a loss of interest in sex after surgery. In one study just over half of women reported that they were not satisfied with their sexual functioning after having surgery and in another study one in every three patients felt that vaginal dryness was bothersome and reported pain with sex. <u>If you are in a relationship</u>, these factors may affect your relationship with your partner." Moved ref to partner to the end and added an 'if' statement. - In the choice changed wording to: "Whether or not you would like to bring anyone else into this decision is your choice. Often it is helpful to <u>speak to someone who knows you well, such as a partner</u>, other members of your family or a friend, who could work through this decision with you. <u>If you are in a relationship the views of your partner can</u> be important, especially in the context of risk-reducing ovarian surgery, so it is recommended that you speak to your partner and try to reach a</p> |

| | |
|---|--|
| | decision together.” To reduce the emphasis on partner |
| I'm not sure if this is the place for it, but in practice it will be somewhere people might want to find it, but what about including some information about genetic testing? This is likely to be an issue for many women using this tool so information might be useful. I know that you have signed to Macmillan for this, but sadly they do take a much more patronising tone than this tool takes. | - Genetic testing really is beyond the scope of this decision aid, however I re-arranged the contacts&resouces and added that CGSW story bank and NHS choices also have info about genetic testing, so that people do not only refer to Macmillan |
| I'm still not totally convinced by the name, but I do appreciate the reasons for it! | - No action taken |
| the start of the text on the left hand side was missing all the way through | - Browser issue fixed |
| will the final format have scales, thus making the weighing up very evident? | - Yes balance fixed |
| I indicated that I had had breast cancer, I found a lot of the information on HRT irrelevant to me (I was ER+) | - Shortened section about 'What is HRT' to remove types of HRT and modes of delivery info - As personalisation question 2 only asks about breast cancer (and not breast cancer type) a little information about HRT should be included, in case that breast cancer was ER negative... - Even ER+ women might be able to take HRT if their quality of life is seriously affected after surgery, so this needs to be mentioned |
| Mentions Lynch Syndrome a lot, which does not apply to me | - Removed all references to Lynch in 'Other options' websites for BRCA women (Other_options 2-13) |
| Graph on page 10 was conflicting in that it talked about BRCA2 gene but had BRCA1 gene notated on the side (Possibly printing error?) | - Checked and correct in online version - Corrected in print version |
| Just wondered whether there was any information on the benefits of Zoladex injections to stop the ovaries working? I had them during my chemo in 2005 and again more recently due to 'irratc ovaries' picked up on several scans. | - Discussed with expert – Zoladex is not offered to women at risk as a standard treatment. No information included |
| As a lot of women would naturally go through the menopause at approx 45 anyway, I just wanted to know if the menopausal symptoms were the same? Since I would go through the menopause anyway it makes the decision to have my ovaries removed much easier. | - Checked and stated clearly menopause is happening at 50, however it also says levels start to fall around 45 (without symptoms), but as this is true it should be included - Checked and it is clearly stated in 'What is a surgical menopause' that the symptoms are the same |

| Suggestions for improvements | Exact action taken / Decision |
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| Lynch <u>Syndrome</u> (in section on cancer risk in general / unknown) – have a look whether ‘syndrome’ is spelt wrong anywhere | - Checked but haven’t found miss-spelling |
| Add an explanation of what the dotted lines are for Lynch women cancer risk in the general version | - Added (striped dots indicate the range of cancer risk from 6 to 12 per 100) below pictogram - In General and all unclear websites |
| If you have been told that your family history looks like you might have a faulty gene and you would like more specific information for people with that gene, you can look at the information by personalising this aid and selecting the gene that most closely resembles your family history as you have been told by your genetic counsellor. Add sth here to make it clearer what is meant e.g. example of BRCA | - Added “For example, if you have been told your family history suggests you might have a BRCA1 gene, personalise OvDex by answering the first question with "Yes, I have a faulty BRCA1 gene". to this section - If you have been told that your family history suggests you might have a faulty gene and you would like more specific information, you can personalise this aid by selecting the gene that most closely resembles your family history according to your genetic counsellor. - In General and all unclear websites |
| How can I deal with my cancer risk? Change question to how can I change how I feel about my cancer risk. | - Changed title of question to “How can I change how I feel about my cancer risk?” - Changed question in question index list - In all cancer risk websites |
| When should I decide? Is there an age limit, what’s the best point in time with the best impact | - Added statement “The operation is most effective if it is done at 40 years of age, but this may not be ideal for everyone. So, even if you decide not to have it at 40, the operation will still decrease your ovarian cancer risk if performed after that age.” Based on evidence from Kurian paper - In all websites (used past tense for postmeno sites) |
| Complications in 100 women who have the operation pictogram Put dots at the top | - Changed pictograms for “type of surgery” and “surgical complications”, so coloured dots are at the top |
| Menopause warning change colour | - Changed colour to light teal in CSS file |
| On the generalise page it says you should have seen these questions (ie personalised) before you opened this page (but I hadn't) | - Changed wording to “You should be given the option to personalise the decision aid before viewing this page. If you wish to personalise OvDex now, please Personalise OvDex.” - In General_home only |
| On the Personalised page - the fourth option - appears to be missing something/error - says....faulty gene or my genetic test | - Added “my genetic test was uninformative” |
| On How will my risk affect children.... it says boys who inherit the gene are not at risk of ovarian caner (but does not say that they may | - Added statement to say that they can carry the gene and pass in on to their children and may be at risk for other cancer: “Boys who inherit |

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| <p>however pass on the gene to their children)</p> | <p>the faulty gene are not at risk of ovarian cancer, but may be at risk for other cancers. Boys who inherit the faulty gene may also pass it on to their children.”</p> <p>- In all websites</p> |
| <p>Under main disadvantages of the operation - under sex can be painful - is it worth including the disclaimer on the menopause page, which gives some reassurance that help is at hand. Otherwise it is a very negative list!</p> | <p>- Added additional short statement that hormone replacement can counteract this: “Your desire for sex will decrease and due to some menopausal symptoms, such as vaginal dryness, you may enjoy sex less as intercourse can be painful. Again, hormone replacement may counteract these effects.”</p> <p>- In all pre-meno websites</p> |
| <p>Under alternatives to surgery, I suggest the symptoms are those used by NICE www.nice.org.uk/cg122</p> | <p>- Added NICE guidance to references in Research evidence</p> |
| <p>The Choice Section: When should I decide? ‘It is important that you understand that undergoing surgery will mean you can no longer get pregnant.’ I think it would be helpful to remind here that this is in relation to having your ovaries removed. ‘It is important that you understand that undergoing surgery to remove your ovaries will mean you can no longer get pregnant.’</p> | <p>- Changes wording to resemble suggestion: “It is important that you understand that undergoing surgery will mean you can no longer get pregnant.”</p> <p>- In all premeno documents</p> |
| <p>You refer to “navigation menu”. Nothing is called that, and although I guessed what is meant by it I wonder if it needs to be identified as such? The counter argument is that anybody using a computer might be assumed to have a pretty good idea of what is meant by it anyway?</p> | <p>- Changed by deleting “navigation” and just leaving ‘menu’</p> <p>- In all websites</p> |
| <p>I have to admit one of the things I don’t like is the “gynae” diagram – it may be because I am medical, or because I am male, but I don’t think so!! The shape and size is all wrong, it is dominated by the uterus and the cervix is wide open, and the vagina ridiculously short. You are just NOT shaped like that!?! But I am prepared to admit that exact anatomical detail is not the main point, and if it is MORE helpful for women then disregard my comment. Although I have just asked one of the secretaries to look at it and she said it looks like a “cows head”!! But she also wasn’t bothered about anatomical detail as I would expect.</p> | <p>- Drew new diagram with larger and longer vagina, smaller uterus and closed cervix</p> |
| <p>“How will my cancer risk affect my life? Being at risk for ovarian cancer does not necessarily mean that you will develop ovarian cancer. However, knowing your risk will give you access to additional medical services such as genetic counselling and means that you <u>are</u> offered risk-</p> | <p>- Changed wording to “may be offered”</p> <p>- In all websites</p> |

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| <p>reducing surgery. Some people may feel worried or anxious after finding out about their risk and their quality of life may be affected” I think it preferable to say you may or might be offered risk-reducing surgery than that you are. Because they might not be offered it, and then wonder why they weren’t</p> | |
| <p>How will my cancer risk affect my children? There is a chance that your children will also have an increased risk of <u>ovarian cancer</u> [I would favour being less specific, and using BRCA1 related cancers, because boys are not at risk of ovarian cancer]. As you have a faulty BRCA1 gene, your children have a 50/50 chance to inherit this gene</p> | <ul style="list-style-type: none"> - Changed wording to just “cancer” in first sentence - In all websites - Took out “ovarian” in the question title and the question list at top of page - In General, Unclear and Lynch sites (BRCA already had only ‘cancer’) |
| <p>helpful to stay optimistic, have joined patient networks <u>space</u> or have found strength in their</p> | <ul style="list-style-type: none"> - Checked and added space in all BRCA websites (other sites were fine) |
| <p>Why is there a choice? Often when you go to your doctor, there is one clearly recommended treatment. However, some situations are more complicated. These are situations in which your personal preferences play an important role. In the case of cancer risk reduction, your options have very different effects on your life. This means that you need to be clear about what might happen if you choose one option over the other. Some health professionals may recommend surgery quite strongly; however you need to make the final decision and should be aware of the possible benefits and risks before you do. You should certainly not feel pressured to opt for surgery. This paragraph seems to imply that women’s choices are going to be based mainly on factually balancing the risks. But a lot of their decisions will be based on personal experience, personal preference, and personal feelings.</p> | <ul style="list-style-type: none"> - Changed section to read: “Often when you go to your doctor, there is one clearly recommended treatment. However, some situations are more complicated. These are situations in which your personal preferences <u>and feelings</u> play an important role. In the case of cancer risk reduction, your options have very different effects on your life. This means that you need to be clear about what might happen if you choose one option over the <u>other and how that would impact on your life</u>. Some health professionals may recommend surgery quite strongly; however you need to make the final decision and before you do, you should consider the possible benefits and risks, <u>how these might affect your life and how you feel about them</u>.” - In all websites |
| <p>In some cases it may not be possible to do keyhole surgery, because of previous surgery on your tummy or because of your weight. Then surgeons use the more traditional open surgery. This means a longer cut, usually along the bikini line. Sometimes a surgeon might have to convert to an open <u>surgery (operation)</u> when doing keyhole surgery because of complications or old scar tissue. This happens in about 1 of every 100 keyhole operations</p> | <ul style="list-style-type: none"> - Reviewed the use of the words ‘surgery’ and ‘operation’ and made changes: - In General, Unclear and BRCA websites: “Sometimes a surgeon might have to convert to an open <u>operation</u> when doing keyhole surgery because of complications or old scar tissue.” - In Lynch websites: “RRH plus RRSO can be done as laparoscopic (keyhole), vaginal or abdominal surgery. The type of surgery you will be offered depends on your personal medical history. Most often the operation is done as keyhole surgery (laparoscopic hysterectomy plus RRSO). This involves 3 or 4 |

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| | <p>small (about 1cm) cuts. Usually one cut is close to your belly button and 2-3 just below the bikini line, so they are not visible. This <u>operation</u> can take about an hour and a half. During vaginal hysterectomy plus RRSO, surgical instruments are inserted through the vagina, so that no cut is needed for the instruments. The uterus and cervix are then removed through a cut just above the vagina. This <u>operation</u> takes less than an hour. Abdominal hysterectomy plus RRSO is an 'open' <u>operation</u> and involves a cut (several inches) in the tummy, either horizontally just above the bikini line or vertically from your belly button down to the bikini line. This <u>operation</u> takes less than an hour."</p> |
| <p>What is <u>a</u> surgical menopause? <u>A</u> surgical menopause is the <u>a</u> menopause you enter when your ovaries are removed by surgery and your body no longer produces the female hormone oestrogen. The <u>A</u> surgical menopause is essentially the same as the natural menopause, it only happens earlier and more suddenly. Therefore <u>a</u> surgical menopause results in the same symptoms as the natural menopause. These symptoms are caused by the lack of oestrogen. As oestrogen levels drop suddenly after surgery, the symptoms of <u>a</u> surgical menopause start instantly and can be more severe than those of the natural menopause, in which oestrogen levels fall gradually</p> | <p>- Made changes in all menopause websites: "What is a surgical menopause? A surgical menopause is a menopause you enter when your ovaries are removed by surgery and your body no longer produces the female hormone oestrogen. A surgical menopause is essentially the same as a natural menopause, it only happens earlier and more suddenly. Therefore a surgical menopause results in the same symptoms as a natural menopause. These symptoms are caused by the lack of oestrogen. As oestrogen levels drop suddenly after surgery, the symptoms of a surgical menopause start instantly and can be more severe than those of a natural menopause, in which oestrogen levels fall gradually."</p> |
| <p>The great majority of women who had surgery report that they - "had surgery <u>reported</u>" or "<u>have</u> surgery report"</p> | <p>- Changed tense to: "The great majority of women who had surgery <u>reported</u> that they <u>experienced</u> one or more..." - In all pre-meno websites</p> |
| <p>There are several options available to help you deal with the short-term symptoms of the menopause. These include physical options, such as lubricant to counteract vaginal dryness, or psychological options, such as sex counselling and cognitive behavioural therapy to improve emotional and physical functioning. You can get more information about these options from your <u>genetics services</u>. I think it would be better to state from your doctor or gynaecologist. This did raise an eyebrow from the GCs.</p> | <p>- Made change and referred to doctor or gynaecologist. - In all pre-meno websites</p> |
| <p>Are there any long term health effects linked to the menopause?Therefore, women who undergo surgery before the age of 45 <u>are usually</u> recommended to take hormone replacement to</p> | <p>- Added the word 'usually' to sentence as proposed - In all pre-meno websites</p> |

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| reduce these effects | |
| Women who have surgery when they are over 45 years of age can choose to have HRT to reduce the symptoms of surgical menopause. However these women do not have to have HRT if they do not want to, as the long term health effects of early loss of oestrogen do not affect them <u>as much as a women having surgery before 45</u> , as their oestrogen levels would have started to fall. It is important that the possibility of HRT is discussed before you decide <u>making a decision</u> about surgery | <ul style="list-style-type: none"> - Made changes: "Women who have surgery when they are over 45 years of age can choose to have HRT to reduce the symptoms of surgical menopause. However these women do not have to have HRT if they do not want to, as the long term health effects of early loss of oestrogen do not affect them <u>as much as a women having surgery before 45</u>, because their oestrogen levels would have started to fall anyway. It is important that the possibility of HRT is discussed before <u>making a decision</u> about surgery." - In all hormone replacement (noBC) |
| Can I take HRT if I have a family history of breast cancer? – please can we discuss. I'm not disagreeing but just trying to think of our advice outside of BRCA1/2 families. | <ul style="list-style-type: none"> - Discussed with expert and decided that section should remain as it is |
| Yes. HRT will recude <u>reduce</u> the risk | <ul style="list-style-type: none"> - Corrected spelling mistake - In all hormone replacement (noBC) |
| Is there any screening available for ovarian cancer? Any woman with symptoms of ovarian cancer will is likely to be offered a diagnostic... | <ul style="list-style-type: none"> - Made change to 'is likely to' - In all websites |
| If any of these symptoms happen more than 12 times - where did the "12 times" come from – surely not the NHS website? | <ul style="list-style-type: none"> - No action |
| What would happen if I do not have surgery? Make sure you tell your GP that you have a family history of ovarian cancer and are at increased risk - this paragraph came up when I was in as a patient with a personal hx of BRCA1. I might not have a FHx of ovarian cancer. So may be <u>Make sure you tell your GP that you have a family history of cancer and are at increased risk of ovarian cancer.</u> | <ul style="list-style-type: none"> - Changed wording to "If you decide not to have surgery, then nothing will really change. You will need to look out for any symptoms and if you think anything is wrong you need to go to your GP to get it checked. Make sure you tell your GP that you have a family history of cancer and are at increased risk of ovarian cancer." - In all websites |
| You can come back to OvDex and do this exercise again in a years time - why specify a years time. Surely we would be happy for someone to revisit their decision as often as they wish?? | <ul style="list-style-type: none"> - Removed time frame and replace with "you can come back to OvDerx anytime and do this exercise again." |
| Having got this far I tried the decision aid – I assume/hope the balance will become an actual balance that tilts!?? | <ul style="list-style-type: none"> - Yes balance will be fixed – Digital Morphopsis |
| Finally having seen "my choice" it all seemed to end a bit abruptly – a bit like the criticism of when running out of time the student writing an essay then says "and then he fell off a cliff. The end..." So is there a way of letting people come to the end more gently... even if it is only to return them to the menu or direct them | <ul style="list-style-type: none"> - Added link to Contacts and Resources at the end of each 'Your Decision' page - Added "My plan" section - Web developer will add free text fields and print results option to enable users to keep a summary of their decision |

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| <p>straight to the list of alternative sources of information?</p> | |
| <p>I thought some of the grammar could have been improved. Some of it was repetitive and some of the sentences were a bit long and clumsy. I also wasn't sure about using words like 'tummy'. I assume you have taken this to a focus group of patients or something but I think you have to be careful not to patronise.</p> | <ul style="list-style-type: none"> - Removed some repetitions (e.g. about primary peritoneal risk) and checked grammar and spelling again (e.g. uses of terms 'operation' and 'surgery'; missing spaces etc.) |
| <p>'Studies have shown that out of 100 women with one first degree relative (mother, sister or daughter) with ovarian cancer, 5 will develop ovarian cancer themselves at some point in their life. Women who have 2 or more first (mother, sister or daughter) and/or second degree (aunt, grandmother etc.) relatives with ovarian cancer have an even higher chance of developing ovarian cancer themselves of about 7 out of 100.' I think this paragraph is confusing and as its right at the beginning may it some people off using the tool. Perhaps you could explain what a first or second-degree relative is elsewhere or earlier in the paragraph so that you don't have to use the brackets in the middle of the sentence. I also think you could put the figures showing the circles in the text rather than all together which would make the numbers clearer. Later on you just show the diagram which is clearer so you may not need this text.</p> | <ul style="list-style-type: none"> - Deleted section that reiterates cancer risks out of 100 and changed paragraph to just explain first/second degree relatives: "Your risk depends on how many of your relatives had cancer and on how closely related they are to you. 'First degree relatives' are those directly related to you, for example, mother, sister or daughter. 'Second degree relatives' are those who are less closely related, for example your aunt or your grandmother. The diagrams below show how risk changes depending on the number of relatives with ovarian cancer." - Also changed wording under third pictogram: "About 7 out of 100 women with two or more first and/or second degree relatives with ovarian cancer get ovarian cancer in their lifetime." |
| <p>This sentence is also confusing 'The number of relatives with cancers other than ovarian cancer, such as breast and bowel cancer, can also affect a woman's chances of developing ovarian cancer.' I would turn the sentence round ie 'a woman's chances of developing ovarian cancer can also be increased by</p> | <ul style="list-style-type: none"> - Changed sentence structure to "A woman's chances of developing ovarian cancer can also be increased if she has relatives with cancers other than ovarian cancer, such as breast and bowel cancer." - In General and Unclear websites |
| <p>I found the section below repetitive and a bit clumsy. It might be best to talk about you rather than 'some women' sometimes and 'you' at others. And it had lots of 'thens' How can I find out whether (or not) I have a faulty gene? Some women might come from families where certain types of cancer (e.g. ovarian, breast, bowel) have affected more than just one or two relatives and these women might have a faulty gene. To get tested for a faulty gene you will need to be referred to genetics services (usually by your GP). There your family history of cancer will be assessed in detail and you will be told whether there might be a faulty gene involved. Testing for a faulty gene in the family then</p> | <ul style="list-style-type: none"> - Changed wording to add "or not" to question title - Removed 'might' and 'then' in most sections - Reworded some parts to make it less clumsy - Made consistent by using "you" throughout: "If you come from a family where certain types of cancer (e.g. ovarian, breast, bowel) have affected more than just one or two relatives, you might have a faulty gene. To get tested for a faulty gene you will need to be referred to genetics services (usually by your GP). The genetics service will assess your family history in detail and you will be told whether there might be a faulty gene involved. Testing for a faulty gene in the family usually begins with a |

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| <p>usually begins with a test on a blood sample from a person who has had cancer. If you have had cancer yourself you may be offered a genetic test as the first person in your family. If you have not had cancer yourself, then a relative who has had cancer will be offered a genetic test first. If a faulty gene is found, other members of the family can then have a genetic test to see whether they also have the same gene. If there is no living relative with cancer, or the relative doesn't want to get tested, then it may not be possible to do a genetic test for you. For sources of more information see Contacts and Resources.</p> | <p>test on a blood sample from a person who has had cancer. If you have had cancer yourself you may be offered a genetic test as the first person in your family. If you have not had cancer yourself a relative who has had cancer will be offered a genetic test first. If a faulty gene is found, other members of the family can then have a genetic test to see whether they also have the same faulty gene. If there is no living relative with cancer, or the relative doesn't want to get tested, it may not be possible to do a genetic test for you.”</p> <p>- In General and Unclear websites</p> |
| <p>A genetic test result does not come back as 'unclear'. Do you mean a variant of uncertain significance? If so I think its best to say this and then explain that this is an unclear result. If no mutation is detected this would be uninformative rather than unclear.</p> | <p>- Changed first personalisation question to read “my genetic test was uninformative”</p> <p>- In Cancer Risk section changed question 5 to: “What if I cannot get tested or my genetic test is uninformative? For some women it might not be possible to get tested because they have no living relative with cancer or their relative does not want to be tested. In other cases a genetic test may come back as 'uninformative' which means a gene variant of uncertain significance has been found, but it is not clear whether this variant increases the risk of cancer or not. In these cases the genetics service will estimate the woman's risk based on her family history.”</p> <p>- In General and Unclear websites</p> |
| <p>Suggestions to improve the sense of this section 'If you have been told that your family history looks like (suggests?) you might have a faulty gene and you would like more specific information (for people with that gene - ?delete), you can (delete? look at the information by) personalize (ing) this aid and select (ing) the gene that most closely resembles your family history (????as you have been told by your genetic counsellor.)</p> | <p>- Changed wording of this section to “If you have been told that your family history suggests you might have a faulty gene and you would like more specific information, you can personalise this aid by selecting the gene that most closely resembles your family history according to your genetic counsellor.”</p> <p>- Added “For example, if you have been told your family history suggests you might have a BRCA1 gene, personalise OvDex by answering the first question with "Yes, I have a faulty BRCA1 gene". to this section</p> <p>- In General and Unclear websites</p> |
| <p>So for example, your risk of getting ovarian cancer in the next year when you are 35 is quite low, whereas your risk of getting ovarian cancer in the next year when you are 50 is higher. Repeats 'in the next year'</p> | <p>- Reviewed section and reworded to make it clearer: “This means when you are young your age-related risk is relatively low and rises when you get older. So for example, if you are 35, your risk of getting ovarian cancer in the next year (i.e. by the time you are 36) is quite low, whereas if you are 50, your risk of getting ovarian cancer in the next year (i.e. by the time</p> |

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| | you are 51) is higher.” |
| Which Dr? GPs will have no idea – suggest advise to get back in touch with genetics ‘You can discuss your children’s risk with your doctor to find out more. Your doctor will also be able to advise you about how best to discuss this risk with your children’. | - Changed to say “genetic counsellor” - In all websites |
| It doesn't make sense to talk about ‘the gene’. It should be ‘the gene fault’ – ‘If they inherited the gene, then their cancer risk would be the same as yours. If they did not inherit the gene, then their cancer risk would be low. Boys who inherit the gene are not at risk of ovarian cancer. ‘ | - Changed to “faulty gene” - In all websites |
| Who should decide? As the best choice for you is based on your preferences, you should be closely involved in the decision. You can either make the decision on your own or if you do not wish to make this decision yourself, your doctor can help you. (They) Your doctor will encourage you to think about the options and your preferences, so that the final choice is right for you. If you then don’t want to choose for yourself, just say so and your doctor may make a recommendation. Better to you ‘your doctpr’ or he or she than they? Also which dr is this? The surgeon, gp, genetics clinician??? | - Specified type of doctor: “You can either make the decision on your own or if you do not wish to make this decision yourself, your genetic counsellor or gynaecologist can help you.” - Left the “they” - In all websites |
| Option grid: HRT is safe to take for the majority of women until they are 50. It is not recommended for women who have had breast cancer themselves already Careful with this statement as women who have had ER-ve BC (more commonly seen in BRCA1) may be able to take HRT | - Changed wording to “HRT is safe to take for the majority of women until they are 50. It is not recommended for women who have had oestrogen receptor positive breast cancer themselves already.” |
| The term ‘bilateral’ means both (otherwise you are saying the same thing twice!) | - Reviewed use of the term ‘both’ to describe what bilateral means - Reworded section so it is clearer, but left the word ‘both’ in: “Risk-reducing bilateral salpingo-oophorectomy (RRSO for short) is an operation to remove healthy ovaries and fallopian tubes to reduce the cancer risk. <u>The word ‘salpingo-oophorectomy’ means surgical removal of the fallopian tubes (salpinges) and ovaries. The term ‘bilateral’ in this context describes the fact that the ovaries and fallopian tubes on both sides of the body are removed.</u> The aim of the operation is to remove these tissues before cancer develops.” |
| RRSO section: ‘Even fewer cancers of the peritoneum have been found in women without | - Completely removed this section as it was a repetition |

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| <p>a faulty BRCA1 gene who have had this surgery, therefore their risk is likely to be much lower than 2 in 100 after surgery.’ Confusing</p> | |
| <p>The highest risk for primary peritoneal cancer is in women with a BRCA1 faulty gene. Their risk is about 2 in every 100 women, which is similar to the risk of ovarian cancer in the general population. For women who do not have a BRCA1 faulty gene the risk of primary peritoneal cancer after surgery is much lower than 2 in 100. You say this twice ie also in:What are the main advantages of this operation? [...]For women with a faulty BRCA1 gene cancer might still develop in the peritoneum, which is the lining of the pelvis and abdomen. This means RRSO will not prevent cancer 6 in about 2 in every 100 women with a BRCA1 faulty gene. This risk is about the same as the ovarian cancer risk in the general population. Even fewer cancers of the peritoneum have been found in women without a faulty BRCA1 gene who have had this surgery, therefore their risk is likely to be much lower than 2 in 100 after surgery.</p> | <ul style="list-style-type: none"> - Completely removed section on primary peritoneal cancer under ‘disadvantages’ and only left detailed explanation in ‘What is my risk after surgery’. |
| <p>Other options: I wouldn't put in dates as some of this data is now published! It also still doesn't mean it will be introduced as a screening test even once the results of the trial are available – that could take years even if it detects cancer early. ‘A large trial of ovarian cancer screening finished in December 2011 and the results will not be available until 2013, so we do not yet know whether this screening is effective and can detect ovarian cancer early. Until the results of the trial are available the NHS will not offer routine screening. The trial offered women CA125 blood tests every four months and one yearly trans-vaginal ultrasound scan.’</p> | <ul style="list-style-type: none"> - Removed reference to the exact year: “A large trial of ovarian cancer screening finished in December 2011 and <u>the results are not yet available</u>, so we do not yet know whether this screening is effective and can detect ovarian cancer early.” - In all websites |
| <p>You should also be aware that the yearly smear test you receive is designed to detect cervical cancer and will not detect ovarian cancer. - I would give this a separate paragraph and make the not bold!</p> | <ul style="list-style-type: none"> - Made separate paragraph - Added em for emphasis to the ‘not’ (bold is not used for emphasis on websites) |
| <p>Women from families with Lynch syndrome may receive trans-vaginal ultrasound screening to look for womb cancer. And Women from Lynch syndrome families, may be offered hysteroscopies or trans-vaginal ultrasound scans to detect uterus/womb cancer, which these women are at higher risk for. However usually these tests are designed to detect womb cancer only and not ovarian cancer. - should be</p> | <ul style="list-style-type: none"> - Added statement: “Additionally, the value of these tests in detecting womb cancer is also not fully proven.” - In Lynch, general and unclear websites - Deleted any sections on Lynch in BRCA linked other options sites |

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| emphasized that this is not proven to be an effective screening test | |
| <p>In some countries it is recommended that women at high risk of ovarian cancer take the oral contraceptive pill when they are pre-menopausal. The oral contraceptive pill reduces ovarian cancer risk by up to half, but it has also been found to increase breast cancer risk slightly. For women at high risk the reduction in ovarian cancer risk is thought to outweigh the slight increase in breast cancer risk. I would have major concerns about this bit. If a woman carries a BRCA1/2 mutation it would not be recommended for her to go on the pill to reduce her risk of ovarian cancer and she could be significantly increasing her risk of breast cancer. Certainly for BRCA1/2 carriers this should be discussed with the genetics clinician not just the GP.</p> | <ul style="list-style-type: none"> - Reviewed section and removed reference to safety in line with feedback: “In some countries it is recommended that women at high risk of ovarian cancer take the oral contraceptive pill when they are pre-menopausal. The oral contraceptive pill reduces ovarian cancer risk by up to half, but it has also been found to increase breast cancer <u>risk</u>.” - Added that women should consult their genetics clinician as well: “Whether the oral contraceptive pill might be an option for you depends on your risk and family history of breast cancer and should be discussed with your <u>genetic counsellor and GP</u>.” - In all pre-meno websites |

Appendix 7.8 - Option Grid Evidence Document

Evidence Document



Oophorectomy Option Grid - pre-menopausal

Cancer risk statement

- Stratton JF, Pharoah P, Smith SK, Easton D, Ponder BA. A systematic review and meta-analysis of family history and risk of ovarian cancer. *Br J Obstet Gynaecol*, 1998; 105(5):493-9.
- Antoniou A, Pharoah PD, Narod S, Risch HA, Eyfjord JE, Hopper JL, et al. Average risks of breast and ovarian cancer associated with BRCA1 or BRCA2 mutations detected in case Series unselected for family history: a combined analysis of 22 studies. *Am J Hum Genet*, 2003; 72(5):1117-30.
- Chen S, Parmigiani G. Meta-analysis of BRCA1 and BRCA2 penetrance. *J Clin Oncol*, 2007; 25(11):1329-33.

1) Will this reduce my risk of ovarian cancer?

- Rebbeck T, Kauff ND and Domchek SM. Meta-analysis of risk-reduction estimates associated with risk-reducing salpingo oophorectomy in BRCA1 or BRCA2 mutation carriers. *JNCI*, 2009; 101 (2): 80-86.
- Schmeler KM, Lynch HT, Chen LM, Munsell MF, Soliman PT, Clark MB, et al. Prophylactic Surgery to Reduce the Risk of Gynecologic Cancers in the Lynch Syndrome. *N Engl J Med*, 2006 Jan 19;354(3):261-9.
- Schmeler KM, Daniels MS, Soliman PT, Broaddus RR, Deavers MT, Vu TM, et al. Primary peritoneal cancer after bilateral salpingo-oophorectomy in two patients with Lynch syndrome. *Obstet Gynecol*, 2010; 115(2 Pt 2):432-4.

2) Will I be able to become pregnant?

- Editorial consensus.

3) Will this change how I feel about my risk of ovarian cancer?

- Finch A, Metcalfe K, Lui J, Springate C, Demsky R, Armel S, et al. Breast and ovarian cancer risk perception after prophylactic salpingo-oophorectomy due to an inherited mutation in the BRCA1 or BRCA2 gene. *Clinical Genetics*, 2009; 75: 220-224.

4) Will surgery change how I feel about myself as a woman?

- Finch A, Metcalfe KA, Chiang JK, Elit L, McLaughlin J, Springate C, et al. The impact of prophylactic salpingo-oophorectomy on menopausal symptoms and sexual function in women who carry a BRCA mutation. *Gynecologic Oncology*, 2011; 121(1): 163-168.
- Finch A, Narod SA. Quality of life and health status after prophylactic salpingo-oophorectomy in women who carry a BRCA mutation: A review. *Maturitas*, 2011; 70:261-265
- Additional Information obtained through discussion with medical professionals.

5) Will I go into menopause?

- Finch A, Metcalfe KA, Chiang JK, Elit L, McLaughlin J, Springate C, et al. The impact of prophylactic salpingo-oophorectomy on menopausal symptoms and sexual function in women who carry a BRCA mutation. *Gynecologic Oncology*, 2011; 121(1): 163-168.

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Evidence Document



- Finch A, Narod SA. Quality of life and health status after prophylactic salpingo-oophorectomy in women who carry a BRCA mutation: A review. *Maturitas*, 2011; 70:261-265.
- Hollowell N, Baylock B, Heiniger L, Butow PN, Patel D, Meiser B, et al. Looking different, feeling different: Women's reactions to risk-reducing breast and ovarian surgery. *Familial Cancer*, 2012 Jun; 11(2):215-224.
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- Additional Information obtained through discussion with medical professionals

6) Are there any long-term health risks linked to surgery?

- Challberg J, Ashcroft L, Lallo F, Eckersley B, Clayton R, Hopwood P, et al. Menopausal symptoms and bone health in women undertaking risk reducing bilateral salpingo-oophorectomy: significant bone health issues in those not taking HRT. *British Journal of Cancer*, 2011; 105: 22-27.
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- Jacoby VL, Grady D, Sawaya GF. Oophorectomy as a risk factor for coronary heart disease. *Am J Obstet Gynecol*, 2009; 200(2):140.e1-9.
- Parker WH, Broder MS, Chang E, Feskanich D, Farquhar C, Liu Z, et al. Ovarian conservation at the time of hysterectomy and long-term health outcomes in the nurses' health study. *Obstet Gynecol*, 2009; 113(5):1027-37.

7) Will I need hormone replacement therapy (HRT) and is that safe?

- Challberg J, Ashcroft L, Lallo F, Eckersley B, Clayton R, Hopwood P, et al. Menopausal symptoms and bone health in women undertaking risk reducing bilateral salpingo-oophorectomy: significant bone health issues in those not taking HRT. *British Journal of Cancer*, 2011; 105: 22-27.
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8) What are the risks of surgery?

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Evidence Document



- Kenkhuis MJA, de Bock GH, Oude Elferink P, Artsa HJG, Oosterwijkb JC, Jansenc L, et al. Short-term surgical outcome and safety of risk reducing salpingo-oophorectomy in BRCA1/2 mutation carriers. *Maturitas*, 2010; 66: 310-314.
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- Additional Information obtained through discussion with medical professionals.

9) How long will it take me to recover from surgery?

- Kenkhuis MJA, de Bock GH, Oude Elferink P., Artsa H.J.G., Oosterwijk J.C., Jansenc L., et al. Short-term surgical outcome and safety of risk reducing salpingo-oophorectomy in BRCA1/2 mutation carriers. *Maturitas*, 2010; 66: 310-314.
- Additional Information obtained through discussion with medical professionals.

10) Is there a routine screening programme to detect ovarian cancer?

- Olivier RI, Lubsen-Brandsma MA, Verhoef S, van Beurden M. CA125 and transvaginal ultrasound monitoring in high-risk women cannot prevent the diagnosis of advanced ovarian cancer. *Gynecol Oncol*, 2006; 100:20–26.
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- Rosenthal AN, Fraser L, Manchanda R, Badman P, Philpott S, Mozersky J, et al. Results of Annual Screening in Phase I of the United Kingdom Familial Ovarian Cancer Screening Study Highlight the Need for Strict Adherence to Screening Schedule. *J Clin Oncol*, 2013; 31(1):49-57.

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Appendix 7.9 - OvDex booklet Final (General version)

The logo for OvDex, featuring the letters 'OvDex' in a stylized font. The 'O' and 'V' are teal, and 'Dex' is grey. The 'V' has a unique shape with a loop at the top.Oophorectomy
Decision ExplorerA large version of the OvDex logo, with 'OvDex' in teal and grey. A horizontal line is positioned below the logo.

Oophorectomy Decision Explorer

A decision aid designed to help you
and your doctor make a decision
about risk-reducing ovarian surgery

For women at increased risk of ovarian cancer

OvDexD4 Feb 2013
©Cardiff University

Next update:
July 2013

OvDex (The Oophorectomy Decision Explorer) has been developed to help you find out more about your options for reducing your ovarian cancer risk. If you are viewing OvDex you should have been referred to it by a doctor or geneticist as you are at increased risk of ovarian cancer.

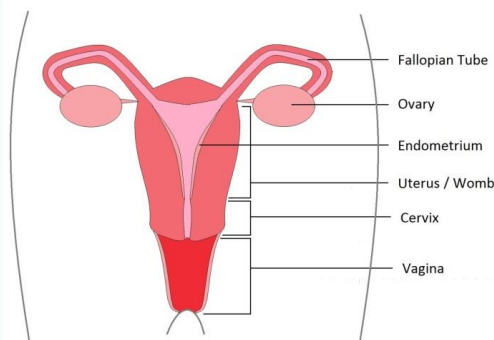
Please note that OvDex can be personalised by answering three questions . You should have seen these questions before you opened this document. If you do wish to personalise the information you get, please go to the OvDex website and answer the questions .

The information the following pages has not been personalised. If you do not wish to personalise OvDex then please view the general information on the following pages.

| Chapter | Page |
|--------------------------------|-------------|
| 1. Cancer risk..... | 3 |
| 2. The choice..... | 7 |
| 3. Options at a glance..... | 10 |
| 4. Risk-reducing surgery..... | 12 |
| 5. The menopause..... | 16 |
| 6. Hormone replacement..... | 19 |
| 7. Other options..... | 22 |
| 8. Your decision..... | 24 |
| 9. Contacts and Resources..... | 29 |
| 10. References..... | 30 |

Questions in chapter 1:

- Q1 Where does ovarian cancer develop?
 Q2 What is the ovarian cancer risk if I have a family history of ovarian cancer?
 Q3 How can I find out whether I have a faulty gene?
 Q4 What would my risk be if a faulty gene is found?
 Q5 What if I cannot get tested or my genetic test is uninformative?
 Q6 Does my cancer risk change over time?
 Q7 How will my ovarian cancer risk affect my life?
 Q8 How will my ovarian cancer risk affect my children?
 Q9 How can I change how I feel about my cancer risk?

Q1 - Where does ovarian cancer develop?

The picture shows the female reproductive system. The ovaries are connected to the uterus by the fallopian tubes. Ovarian-type cancer can develop in the ovaries, the fallopian tubes or the lining of the abdomen (called the 'peritoneum'). Other female cancers, such as cancer of the endometrium, uterus/womb or cervix, are very different and should not be confused with ovarian cancer.

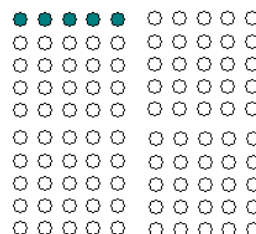
Q2 - What is the ovarian cancer risk if I have a family history of ovarian cancer?

Ovarian cancer occurs by chance in less than 2 of every 100 women. For women who have a family history of ovarian, breast and/or bowel cancer and/or a faulty gene, the risk can be much higher than 2 in 100. Generally the risk is quite low in young women and rises with age.

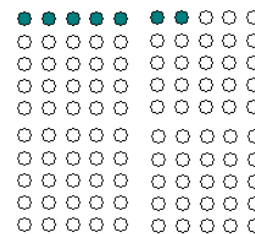
Your risk depends on how many of your relatives had cancer and on how closely related they are to you. 'First degree relatives' are those directly related to you, for example, mother, sister or daughter. 'Second degree relatives' are those who are less closely related, for example your aunt or your grandmother. The diagrams below show how risk changes depending on the number of relatives with ovarian cancer.



Less than 2 out of 100 women in the general population get ovarian cancer in their lifetime. 98 women will not.



About 5 out of 100 women with a first degree relative with ovarian cancer get ovarian cancer in their lifetime. 95 women will not.



About 7 out of 100 women with two or more relatives with ovarian cancer get ovarian cancer in their lifetime. 93 women will not.

1. Cancer risk

4

A woman's chances of developing ovarian cancer can also be increased if she has relatives with cancers other than ovarian cancer, such as breast and bowel cancer. A woman's risk is estimated by genetics services once the full family history is available.

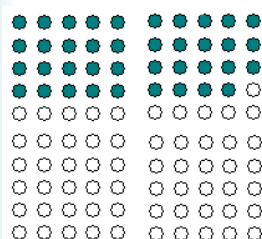
Q3 - How can I find out whether I have a faulty gene?

If you come from a family where certain types of cancer (e.g. ovarian, breast, bowel) have affected more than just one or two relatives, you might have a faulty gene. To get tested for a faulty gene you will need to be referred to genetics services (usually by your GP). The genetics service will assess your family history in detail and you will be told whether there might be a faulty gene involved.

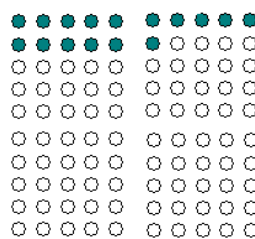
Testing for a faulty gene in the family usually begins with a test on a blood sample from a person who has had cancer. If you have had cancer yourself you may be offered a genetic test as the first person in your family. If you have not had cancer yourself a relative who has had cancer will be offered a genetic test first. If a faulty gene is found, other members of the family can then have a genetic test to see whether they also have the same faulty gene. If there is no living relative with cancer, or the relative doesn't want to get tested, it may not be possible to do a genetic test for you. For sources of more information see chapter 9 (Contacts and Resources), page 29.

Q4 - What would my risk be if a faulty gene is found?

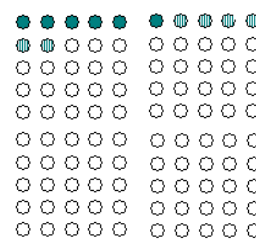
The most common faulty genes linked to ovarian cancer are called Breast Cancer (BRCA) genes 1 and 2 and genes common in Lynch Syndrome (formerly known as Hereditary Non-Polyposis Colorectal Cancer or HNPCC). The lifetime risk of developing ovarian cancer can be much higher for women who have a faulty gene compared to the general population.



About 39 out of 100 women with a BRCA1 faulty gene get ovarian cancer by the time they are 70. About 61 women with a BRCA1 faulty gene will not.



About 16 out of 100 women with a BRCA2 faulty gene get ovarian cancer by the time they are 70. About 84 women with a BRCA2 faulty gene will not.



About 6 to 12 out of 100 women with Lynch Syndrome get ovarian cancer by the time they are 70. About 88-94 women with Lynch Syndrome will not.

Q5 - What if I cannot get tested or my genetic test is uninformative?

For some women it might not be possible to get tested because they have no living relative with cancer or their relative does not want to be tested. In other cases a genetic test may come back as 'uninformative' which means a gene variant of uncertain significance has been found, but it is not clear whether this variant increases the risk of cancer or not. In these cases the genetics service will estimate the woman's risk based on her family history.

If you have been told that your family history suggests you might have a faulty gene and you would like more specific information, you can personalise this aid by selecting the gene that most closely resembles your family history according to your genetic counsellor. For example, if you have been told your family history suggests you might have a BRCA1 gene, personalise OvDex by going to the website and answering the first question with "Yes, I have a faulty BRCA1 gene".

Q6 - Does my cancer risk change over time?

Yes. While your so-called 'lifetime' risk (which is the risk that you will get ovarian cancer at some point in your life) will stay the same over time, your 'age-related' ovarian cancer risk (which is the risk that you will get ovarian cancer in a specified time frame, for example one year or five years) increases with age. This means when you are young your age-related risk is relatively low and rises when you get older. So for example, if you are 35, your risk of getting ovarian cancer in the next year (i.e. by the time you are 36) is quite low, whereas if you are 50, your risk of getting ovarian cancer in the next year (i.e. by the time you are 51) is higher.

Q7 - How will my ovarian cancer risk affect my life?

Being at risk for ovarian cancer does not necessarily mean that you will develop ovarian cancer. However, knowing your risk will give you access to additional medical services such as genetic counselling and means that you may be offered risk-reducing surgery. Some people may feel worried or anxious after finding out about their risk and their quality of life may be affected.

Q8 - How will my ovarian cancer risk affect my children?

There is a chance that your children will also have an increased risk of cancer. Their risk depends on how high your own risk is and on whether or not you have a faulty gene. If you have a faulty gene, then your children have a 50/50 chance to inherit this. If they inherited the faulty gene, then their cancer risk would be the same as yours. If they did not inherit the faulty gene, then their cancer risk would be low. Boys who inherit the faulty gene are not at risk of ovarian cancer, but may be at risk for other cancers. Boys who inherit the faulty gene may also pass it on to their children.

Please also note that cancer risk increases with age and therefore children's risk only starts to rise in later life. More information about genetics can be found on the NHS choices website (see Chapter 9 Contacts and Resources, page 29).

You can discuss your children's risk with your genetic counsellor to find out more. Your genetic counsellor will also be able to advise you about how best to discuss this risk with your children.

Q9 - How can I change how I feel about my cancer risk?

If you would like to find out more about your risk and what it means, you can use the resources provided in OvDex and talk to your doctor or genetic counsellor.

Knowing that you are at increased risk of cancer might make you feel worried or angry or frustrated. This is completely normal and there is no need to bottle those feelings up. It can actually help to talk about them.

You can try to find someone you trust to talk to or if you prefer to talk to someone you don't know, you can use the contact details for charities and patient support networks under Contacts & Resources. If you have a partner, sharing your thoughts and concerns with your partner can be useful in helping them understand what you are going through and in helping you cope together as a couple.

It may also help to see a positive side to knowing your risk status. For example, this means that you will get access to genetics services and have the chance to do something about your risk. Other women have found it helpful to stay optimistic, have joined patient networks or have found strength in their faith.

Being at risk of cancer may sometimes lead to unwanted thoughts and worry, especially at times when there are no active steps you can take, for example when you are waiting for genetic test results. There are useful techniques you can use to reduce such thoughts. One such method is active distraction, which means actively thinking about something else whenever unwanted thoughts pop in your head. Watch the 'How do I cope' video on the Cancer Genetics storybank website for a guide of how to do this (see Contacts & Resources).

Relaxation techniques, meditation and guided imagery may also help you deal with any stress you might experience relating to your cancer risk. Examples include progressive muscle relaxation and certain breathing exercises. Visit the 'Mind-body therapy' section on the Macmillan website for more information on and guides to mind-body therapies (see Contacts & Resources).

Questions in chapter 2:

- Q1 What can I do to reduce my risk?
- Q2 Why is there a choice?
- Q3 Who should decide?
- Q4 Who else should I involve in this decision?
- Q5 When should I decide?
- Q6 How can I decide?
- Q7 How can I deal with the choice?

Q1 - What can I do to reduce my risk?

Your doctor has probably discussed with you the option of having risk-reducing ovarian surgery to reduce your ovarian cancer risk. You will need to decide whether and when to have this surgery. Unfortunately, there is no medically proven screening available on the NHS for ovarian cancer at the moment.

This decision aid is designed to help you look at your options. Please refer to chapter 3 (Options at a glance) page 10, chapter 4 (Risk-reducing surgery) page 12 and chapter 7 (Other options) page 22 for more information about surgery, screening and other alternatives.

Q2 - Why is there a choice?

Often when you go to your doctor, there is one clearly recommended treatment. However, some situations are more complicated. These are situations in which your personal preferences and feelings play an important role. In the case of cancer risk reduction, your options have very different effects on your life. This means that you need to be clear about what might happen if you choose one option over the other and how that would impact on your life.

Some health professionals may recommend surgery quite strongly; however you need to make the final decision and before you do, you should consider the possible benefits and risks, how these might affect your life and how you feel about them.

Q3 - Who should decide?

As the best choice for you is based on your preferences, you should be closely involved in the decision. You can either make the decision on your own or if you do not wish to make this decision yourself, your genetic counsellor or gynaecologist can help you. They will encourage you to think about the options and your preferences, so that the final choice is right for you. If you then don't want to choose for yourself, just say so and they may make a recommendation.

Q4 - Who else should I involve in this decision?

Whether or not you would like to bring anyone else into this decision is your choice. Often it is helpful to speak to someone who knows you well, such as a partner, other members of your family or a friend, who could work through this decision with you. If you are in a relationship the views of your partner can be important, especially in the context of risk-reducing ovarian surgery, so it is recommended that you speak to your partner and try to reach a decision together. Your doctor will also be happy for you to bring your partner along to appointments and to answer any questions they might have.

Q5 - When should I decide?

The operation is most effective if it is done at 40 years of age, but this may not be ideal for everyone. So, even if you decide not to have it at 40, the operation will still decrease your ovarian cancer risk if performed after that age.

The decision you are facing is not an easy one and you should not feel under any pressure to decide quickly. Risk-reducing surgery has benefits and risks that need to be weighed carefully, so take your time and make sure you are ready before making a decision.

It is important that you understand that undergoing surgery to remove your ovaries will mean you can no longer get pregnant. Therefore it is essential to consider your plans for a family and any potential future changes to these plans (for example if there is a chance you might meet a new partner / re-marry) before making a final decision.

Q6 - How can I decide?

When it comes to important decisions everyone is different. Some people like to find out as much as they can about their options, while others prefer to just know what is absolutely necessary. Some might find it helpful to talk to their family and friends. Some might like to speak to people who have made a similar decision. It really depends on you. Have a think about other important decisions in your life and how you managed to make those. That could give you an idea of how you like to decide about things.

Q7 - How can I deal with the choice?

It can be helpful to create a plan of how and when you will make this choice. If you are not ready to decide right now, it might be useful to set yourself a deadline of when you will revisit this decision. For example: "Just after my 40th birthday I will look at this information again." or "Once I have completed my family I will revisit this decision."

Once you are ready you can decide how you want to make this choice:

1. I will decide by myself using everything I have learnt
2. I will decide but seriously consider my doctor's opinion
3. The doctor and I should decide together
4. The doctor should decide but seriously consider my opinion
5. The doctor should decide for me

One constructive way to deal with a difficult decision is to empower yourself with information. OvDex is designed to help you to learn more about ovarian cancer risk and your options. With the wealth of information that is available on the internet, it can be difficult to find reliable and trustworthy information. The information in OvDex is supported by recent scientific findings and has been carefully reviewed by health professionals to make sure it is accurate. You should at least understand your options and their benefits and risks before making a decision. Find out more about the most important questions to ask at: www.Ask3Questions.co.uk. Once you have read the information in OvDex, it could help to make a note of any remaining questions and take those to your doctor or genetic counsellor for a more detailed discussion.

2. The Choice

You may feel that you are not comfortable making decisions about your health. This is okay. You do not need to make the choice alone if you don't want to. You can decide together with your doctor or ask them to make the choice for you. But you need to remember that you are the expert when it comes to your own life and that only you know what is important to you. So even if you decide to let the doctor make the decision for you, make sure they know about your goals and values. Tell them what is important to you.

3. Options at a Glance

| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|---|---|--|
| Will this reduce my risk of ovarian cancer? | Yes, removing the ovaries and fallopian tubes will greatly reduce your lifetime risk of ovarian-type cancer to that of the general population, which is about 2 in 100. | No, your risk of ovarian cancer will remain increased. |
| Will I be able to become pregnant? | No, therefore it is important that you feel you have completed your family before having surgery. | Yes, your ability to become pregnant will be unaffected. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. | You may worry about developing ovarian cancer. |
| Will this change how I feel about myself as a woman? | Many women do not notice any change in how they feel about themselves as a woman. However, your desire for and/or enjoyment of sex may decrease. | Not applicable. |
| Will I go into menopause? | Yes, after surgery your oestrogen levels fall and the menopause will start immediately. | Yes, you will go through a natural menopause when you are around 50 years. |
| What is menopause like? | Menopause after surgery is similar to natural menopause, but because it happens suddenly, the symptoms may be more severe. | Natural menopause is a gradual decline of oestrogen levels. Typical symptoms include hot flushes, night sweats, mood swings and vaginal dryness. |
| Are there any long-term health risks linked to surgery? | Yes, if surgery is done before age 45 there is a higher risk of bone thinning and cardiovascular disease. Some patients report memory changes as well. | Not applicable. |
| Will I need hormone replacement therapy (HRT)? | HRT can reduce some of the effects of surgery and is recommended for women who have surgery before age 45. | No, if you go through a natural menopause you would not be expected to need HRT, unless your symptoms are very severe. |
| Is HRT safe for women at increased familial risk? | HRT is safe to take for the majority of women until they are 50. Discuss this with your clinician. It is not usually recommended for women who have had breast cancer. | HRT is safe to take for the majority of women until they are 50. Discuss this with your clinician. It is not usually recommended for women who have had breast cancer. |

3. Options at a Glance

| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|--|--|--|
| What are the risks of surgery? | About 4 in every 100 patients experience a complication during or after surgery. These can be minor infections or major complications. Discuss this with your clinician. | Not applicable. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after key hole surgery and are back to normal in 4 weeks. For open surgery time in hospital and recovery will be longer. | Not applicable. |
| Is there a routine screening programme to detect ovarian cancer? | No, after surgery there is no need for screening because your risk will be similar to that of the general population. | No, there is no evidence that screening for ovarian cancer is effective. |

Q1 – How can I deal with this decision?

Some women may find it useful to discuss surgery with others, who have already made this decision. In some areas your genetic counsellor may be able to put you in touch with someone who has already gone through the decision, otherwise you can use the contact details for charities and patient support networks under Contacts & Resources to read about or get in touch with others in a similar situation. Going over the decision with someone who knows you well, such as your partner or a member of your family can also help you clarify your decision. If you decide to opt for surgery and you are in a relationship, preparing yourself together with your partner for the time after surgery can help you both deal better with any consequences of the operation.

Being clear about the reasons why surgery could be the right or wrong thing for you can help you make a decision and avoid regretting it later. The information in OvDex is designed to help you identify the benefits and the risks of surgery and any other options, so you can decide for yourself. The exercise called 'Your Decision' can help you clarify what is most important to you.

Being at risk of cancer may sometimes lead to unwanted thoughts and worry, especially at times when there are no active steps you can take, for example if you have decided to defer surgery or while you are waiting for your surgery appointment. There are useful techniques you can use to reduce such thoughts. One such method is active distraction, which means actively thinking about something else whenever unwanted thoughts pop in your head. Watch the 'How do I cope' video on the Cancer Genetics storybank website for a guide of how to do this.

Relaxation techniques, meditation and guided imagery may also help you deal with any stress you might experience relating to your cancer risk. Examples include progressive muscle relaxation and certain breathing exercises. Visit the 'Mind-body therapy' section on the Macmillan website for more information on and guides to mind-body therapies.

Questions in chapter 4:

- Q1 What is risk-reducing bilateral salpingo-oophorectomy (RRSO)?
- Q2 What are the main advantages of this operation?
- Q3 What are the main disadvantages of this operation?
- Q4 What is my risk after surgery?
- Q5 What does the surgery involve?
- Q6 How long does it take to recover?
- Q7 Do I need to have my womb removed as well?
- Q8 What are the complications of RRSO?
- Q9 Could cancer be found during the surgery?
- Q10 How would RRSO affect my life?

Q1 - What is risk-reducing bilateral salpingo-oophorectomy (RRSO)?

Risk-reducing bilateral salpingo-oophorectomy (RRSO for short) is an operation to remove healthy ovaries and fallopian tubes to reduce the cancer risk. The word 'salpingo-oophorectomy' means surgical removal of the fallopian tubes (salpinges) and ovaries. The term 'bilateral' in this context describes the fact that the ovaries and fallopian tubes on both sides of the body are removed. The aim of the operation is to remove these tissues before cancer develops.

Q2 - What are the main advantages of this operation?

- 1) This operation will reduce your ovarian and fallopian tube cancer risk, as these are completely removed during surgery.
- 2) For some women, not having to worry about ovarian cancer anymore comes as a great relief.
- 3) For the vast majority of women having RRSO before the natural menopause could also reduce the risk of breast cancer.
- 4) Having the surgery will also prevent other, non-cancer related problems happening with the ovaries, such as cysts.

4. Risk-reducing Surgery

Q3 - What are the main disadvantages of this operation?

- 1) You will no longer be able to become pregnant once you have had surgery, therefore it is important that you have considered your plans for a family before making a final decision.
- 2) After the operation you will immediately enter the menopause and may start to experience the typical menopausal symptoms, including hot flushes, mood swings, night sweats and vaginal dryness (Please see Chapter 5 (The Menopause), page 16 for more details). Furthermore, you may be at increased risk of bone thinning, cardiovascular (heart and blood vessel) disease and memory changes. These effects may be effectively treated with hormone replacement therapy (Please see chapter 6 (Hormone Replacement), page 19 for more details), however not all women can take hormone replacement and this should be discussed before having surgery.
- 3) Your desire for sex will decrease and due to some menopausal symptoms, such as vaginal dryness, you may enjoy sex less as intercourse can be painful. Again, hormone replacement may counteract these effects.
- 4) There is a small risk of complications during and after surgery.

Q4 - What is my risk after surgery?

After surgery your risk of ovarian and fallopian tube cancer is greatly reduced, however, there is a rare type of cancer which may still develop after surgery that is very similar to ovarian cancer. This is called primary peritoneal cancer and develops in the lining of the abdomen and pelvis. Your risk of primary peritoneal cancer depends on whether or not you have a faulty gene. The highest risk for primary peritoneal cancer is in women with a BRCA1 faulty gene. Their risk is about 2 in every 100 women, which is similar to the risk of ovarian cancer in the general population. For women who do not have a BRCA1 faulty gene the risk of primary peritoneal cancer after surgery is much lower than 2 in 100.

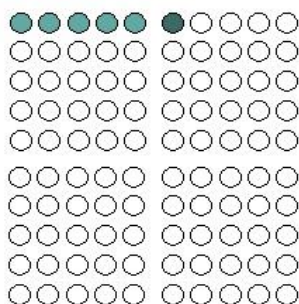
Q5 - What does the surgery involve?

Most often the operation is done as keyhole surgery (laparoscopy). This involves 3 or 4 small (about 1cm) cuts, usually one cut is close to your belly button and 2-3 just below the bikini line, so they are not visible. Keyhole surgery itself usually takes less than 2 hours.

In some cases it may not be possible to do keyhole surgery, because of previous surgery on your tummy or because of your weight. Then surgeons use the more traditional open surgery. This means a longer cut, usually along the bikini line. Sometimes a surgeon might have to convert to an open operation when doing keyhole surgery because of complications or old scar tissue. This happens in about 1 of every 100 keyhole operations.

4. Risk-reducing Surgery

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Type of procedure in 100 women who have the operation

The figure on the left shows **how many of 100 women will have keyhole surgery , open surgery** and planned keyhole which is converted to open surgery.

94 of 100 women will have planned keyhole surgery (white dots). Five of 100 women will have planned open surgery (light teal dots). One of 100 women will have planned keyhole surgery which is converted to open surgery (dark dot).

Q6 - How long does it take to recover?

75 of 100 patients who have keyhole surgery leave the hospital the day after surgery. They are usually back to normal activity about 4 weeks after surgery. If you have open surgery you are likely to stay in hospital a bit longer. Usually patients leave the hospital about 5 days after open surgery and are back to normal in about 6 weeks. After surgery you are not allowed to do any heavy lifting for a few weeks. You may also have to refrain from driving until you can comfortably wear a seatbelt and make an emergency stop without pain.

Q7 - Do I need to have my womb removed as well?

No, normally it is not necessary that you have your womb removed at the same time as having RRSO, unless you know that you have Lynch Syndrome or you have other problems with your womb and have been told by a doctor that removing the womb would help you with those problems. However, you may choose to have your womb removed at the time of RRSO, as it may affect the type of hormone replacement you would be given. You should discuss this with your doctor.

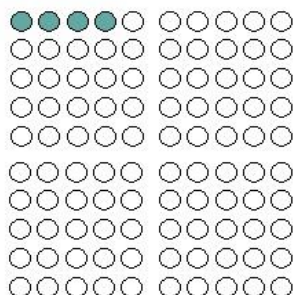
Q8 - What are the complications of RRSO?

There is a small risk of complications linked to RRSO. About 4 in every 100 people will experience some complication. Minor complications can include wound or urinary tract infections and usually have no long-term effects on your health. More serious complications might happen during surgery and can include damage to blood vessels, the bowel or the bladder. If you are having keyhole surgery this might mean that the surgeon has to convert to open surgery to repair the damage. There are a number of other rare complications that might happen and your surgeon will go through those with you if you wish before you go in for surgery.

Please note that about 96 of every 100 women do not experience any complications at all.

4. Risk-reducing Surgery

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**Complications in 100 women who have the operation**

The figure on the left shows **how many of 100 women will experience complications** during or after surgery.

4 of 100 women will experience some kind of complication either during or after surgery. 96 of 100 women will not experience any complications.

As with any surgery, there is a very small risk of death. However, this is highly unlikely. The risk may be greater in women with health problems before surgery.

Q9 - Could cancer be found during the surgery?

Yes, although this is rare. Once your ovaries and fallopian tubes have been removed they will be checked thoroughly for any signs of cancer. There is a chance that cancer may be discovered during this check. This happens in up to 4 out of every 100 operations in the highest risk patients. For others the chances of finding cancer during the surgery are much lower.

Q10 - How would RRSO affect my life?

In the short-term, if you decide to go for surgery, you will have to take time off work and will not be able to do some of the things you might usually do, such as driving or heavy lifting. However most women recover fully within 4 to 6 weeks.

In the longer term there are a number of things you should consider. After surgery you may feel less worried about ovarian cancer. However you may feel differently about your body and you may lose interest in sex. You will also no longer be able to become pregnant and you will enter the menopause if you have not gone through it yet (Please see chapter 5 (Surgical menopause), page 16). You may opt to take hormone replacement (Please see chapter 6 (Hormone replacement), page 19). These factors may affect your life after surgery and should be considered carefully before making a decision.

5. The Menopause

16

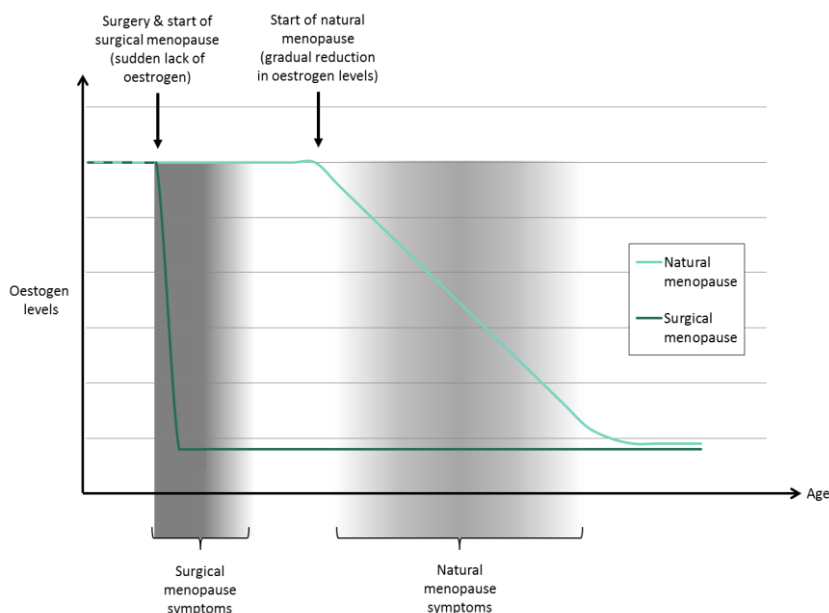
Note that the information about symptoms of the menopause in this chapter describes symptoms experienced by women who do not take hormone replacement therapy (HRT). Most symptoms of the menopause are reduced by hormone replacement therapy (for more information see Chapter 6 (Hormone Replacement) on page 19).

Questions in chapter 5:

- Q1 What is surgical menopause?
 Q2 What is the menopause like?
 Q3 How could the menopause affect my life in the short term?
 Q4 How long does surgical menopause last?
 Q5 Are there any long term health effects linked to surgical menopause?
 Q6 Is there anything I can do to reduce the effects of surgical menopause?

Q1 - What is surgical menopause?

A surgical menopause is a menopause you enter when your ovaries are removed by surgery and your body no longer produces the female hormone oestrogen. A surgical menopause is essentially the same as a natural menopause, it only happens earlier and more suddenly. Therefore a surgical menopause results in the same symptoms as a natural menopause. These symptoms are caused by the lack of oestrogen. As oestrogen levels drop suddenly after surgery, the symptoms of a surgical menopause start instantly and can be more severe than those of a natural menopause, in which oestrogen levels fall gradually.



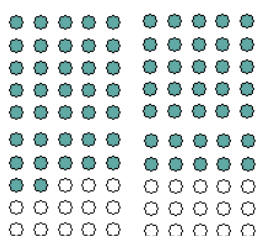
The graph above shows the drop in the levels of the female hormone oestrogen during surgical and natural menopause. Potential menopausal symptom duration and severity are indicated in shades of grey.

Q2 - What is the menopause like?

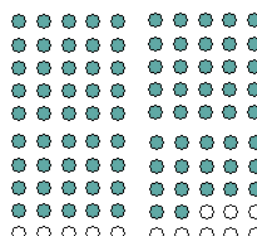
It is very difficult to describe what the menopause is like, because it is different for every person. Some women have no or very few symptoms and cope very well. Other women can have very severe symptoms and their quality of life can be affected.

Common menopausal symptoms you may experience are: Hot flushes, night sweats, mood swings, vaginal dryness and loss of interest in sex. Other symptoms include difficulty sleeping, fatigue, weight gain, changes in memory and depression.

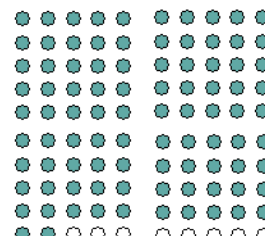
The great majority of women who had surgery reported that they experienced one or more of the common symptoms. However, any data available about menopausal symptoms is from small studies and each study uses different ways to assess what women experienced, so it is difficult to predict what the menopause will be like for you personally. The pictures below show how common different types of menopausal symptoms are according to one study of women after RRSO.



About 72 of 100 women experience vasomotor symptoms (hot flushes and night sweats). 28 women do not.



About 87 of 100 women experience physical and sexual symptoms (aching, vaginal dryness, loss of libido). 13 women do not.



About 92 of 100 women experience psychosocial symptoms (mood swings, feeling depressed or down). 8 women do not.

Q3 - How could menopause affect my life in the short term?

While none of the menopausal symptoms mentioned above are dangerous for your health, they may affect you in different ways. Hot flushes can be very uncomfortable and may happen at inconvenient times, for example when you are giving a presentation at work. In one study two out of every three women reported having hot flushes after surgery. Night sweats can lead to problems with sleep and insomnia.

Vaginal dryness can lead to pain during sex and therefore you may enjoy sex less. Additionally, many women also experience a loss of interest in sex after surgery. In one study just over half of women reported that they were not satisfied with their sexual functioning after having surgery and in another study one in every three patients felt that vaginal dryness was bothersome and reported pain with sex. If you are in a relationship, these factors may affect your relationship with your partner. The way that these symptoms might affect your life depends on your personal situation.

There are several options available to help you deal with the short-term symptoms of the menopause. These include physical options, such as lubricant to counteract vaginal dryness, or psychological options, such as sex counselling and cognitive behavioural therapy to improve emotional and physical functioning. You can get more information about these options from your doctor or gynaecologist. The NHS choices website also has information about dealing with the menopause (See Chapter 9 (Contacts & Resources), page 29.

Q4 - How long does the menopause last?

It is impossible to say how long symptoms will last. Some women have hardly any problems and symptoms disappear quickly, while others have symptoms for many years. It really is different for everyone.

Q5 - Are there any long term health effects linked to the menopause?

Yes. The loss of oestrogen is thought to affect long term health, particularly the bones and the cardiovascular system (heart and blood vessels). In older age this can lead to a higher risk of fractures due to bone thinning (osteoporosis) and a higher risk of cardiovascular disease (heart disease, stroke).

It should be noted that these effects are seen in all women after the menopause, whether it was a surgical or natural menopause. Your personal risk depends on your lifestyle and personal and family history of osteoporosis and cardiovascular disease and should be discussed with your doctor.

Due to the sudden lack of oestrogen after surgery, the risk of bone thinning and cardiovascular disease may start to rise immediately after surgery. This could be before the woman has reached the age of the natural menopause. Therefore, women who undergo surgery before the age of 45 are usually recommended to take hormone replacement to reduce these effects until they reach the age of the natural menopause (See Chapter 6 (Hormone replacement) page 19).

Some women also report changes in memory following the menopause, which is again due to the lack of oestrogen.

Q6 - Is there anything I can do to reduce the effects of the menopause?

Yes. Hormone replacement therapy (HRT) is usually recommended for women who have RRSO before the age of 45 and have not had breast cancer themselves (See Chapter 6 (Hormone replacement) page 19). HRT may effectively reduce some of the short- and long-term effects of surgical menopause. However, not all women can take hormone replacement and this should be discussed before having surgery.

If you do not wish to or cannot take HRT then you can take dietary supplements to reduce bone thinning. You should speak to your doctor about this. In some regions in the UK patients are also recommended to have a bone density scan about 18 months after their operation. You can discuss this with your doctor.

An active lifestyle and healthy diet is also recommended.

6. Hormone Replacement

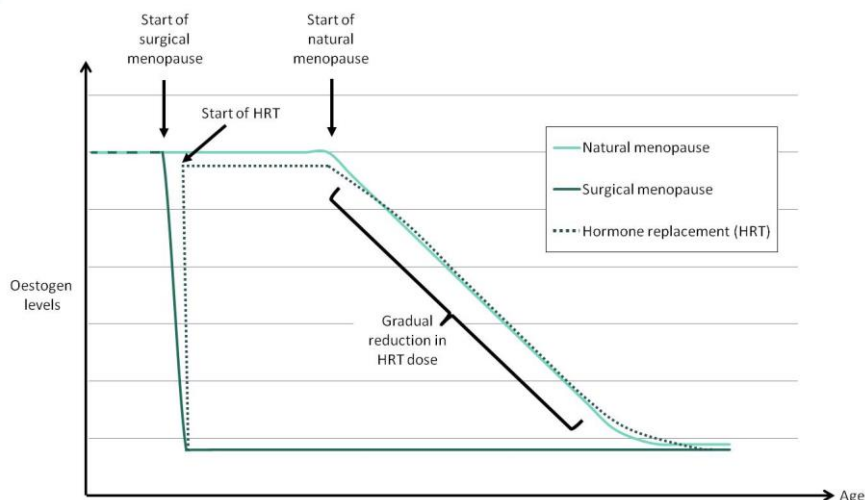
19

Questions in chapter 6:

- Q1 What is Hormone Replacement Therapy (HRT)?
 Q2 Why is HRT important?
 Q3 Who should take HRT?
 Q4 How long should I take HRT?
 Q5 Can I take HRT if I have a family history of breast cancer?
 Q6 But I have heard that HRT increases breast cancer risk
 Q7 Will HRT reduce all the symptoms of menopause?
 Q8 Will HRT deal with the long term health effects of the menopause?
 Q9 Are there any alternatives to HRT?

Q1 - What is Hormone Replacement Therapy (HRT)?

Hormone Replacement Therapy, or HRT for short, is a medical treatment that puts back the female hormones that are lost when the ovaries are removed or when they stop functioning.



The graph above shows the oestrogen levels after surgical and natural menopause. It also shows the effect of HRT on the oestrogen levels after the operation if surgery was performed before the natural menopause.

There are a number of ways that HRT can be used:

- It can be taken as oral tablets usually once a day
- It can be applied as patches that are put on the tummy or bottom about once or twice a week
- It can be applied as a gel directly to the skin once a day (for example the lower abdomen or inner thigh)

There are two types of hormone replacement, one includes oestrogen and progesterone and the other includes oestrogen only. Women who only have their ovaries removed will usually be given oestrogen and progesterone HRT, while women who have their womb/uterus and ovaries removed (RRSO plus hysterectomy) are usually given oestrogen only HRT.

Q2 - Why is HRT important?

There are two main reasons why HRT is recommended after surgery:

1. To reduce the risk of bone thinning
2. To reduce symptoms of the menopause

Q3 - Who should take HRT?

HRT is strongly recommended for women who have surgery before they are 45 years of age, as this group is most affected by the long term health issues that are linked to loss of oestrogen, especially bone thinning (osteoporosis).

Women who have surgery when they are over 45 years of age can choose to have HRT to reduce the symptoms of surgical menopause. However these women do not have to have HRT if they do not want to, as the long term health effects of early loss of oestrogen do not affect them as much as a women having surgery before 45, because their oestrogen levels would have started to fall anyway.

It is important that the possibility of HRT is discussed before making a decision about surgery.

Q4 - How long should I take HRT?

If you use HRT, it is recommended that you take it until the age of the natural menopause, which is 50 years in the UK. From the age of 50 your oestrogen levels would have declined naturally, so taking HRT for many years beyond 50 is not recommended. This is because there is controversial evidence about the benefits and risks of HRT if it is taken beyond the age of natural menopause. Therefore the health effects are unclear and HRT may cause more harm than good if taken beyond 50.

Q5 - Can I take HRT if I have a family history of breast cancer?

Yes. You should be able to take HRT even if you have a family history of breast cancer. Several studies have shown that HRT is safe to take for the vast majority of women with a family history of breast cancer as long as they have not had breast cancer themselves and they only use HRT until the age of the natural menopause. You should discuss your family history of breast cancer with your genetics service before deciding to have surgery.

Q6 - But I have heard that HRT increases breast cancer risk?

Yes, HRT contains oestrogen and there is some evidence that higher amounts of oestrogen in the body can increase the risk of breast cancer.

However, the study that showed that HRT increases breast cancer risk was done with older women who had already gone through the natural menopause and were taking HRT beyond 50 years of age. Therefore these women were taking oestrogen when their body has naturally stopped producing this hormone.

Furthermore, these women did not have surgery to have their ovaries removed. Therefore this group of women is very different to women with a family history who choose to have their ovaries out before they are 50 years. Furthermore you should be aware that the findings of this study have now been widely criticised.

Having your ovaries out removes all the natural oestrogen that your ovaries would be producing until the age of the natural menopause. The amount of oestrogen that is added back by taking HRT is less than the amount your ovaries would have produced naturally. If you stop taking HRT at the age of the natural menopause (so when you are 50) then there is no evidence that the breast cancer risk is increased.

Q7 - Will HRT reduce all the symptoms of menopause?

No. Taking HRT is not the same as having your own hormones. HRT cannot reduce all symptoms of the menopause and women who have had surgery and are on HRT do report more symptoms than women who have not had surgery. However, HRT can relieve some of the symptoms of the menopause. For example, women who took HRT reduced the average number of hot flushes from about 4 to about 1 per day when compared to women who did not take HRT. Women on HRT also had fewer night sweats.

Q8 - Will HRT deal with the long term health effects of the menopause?

Yes. HRT will reduce the risk of bone thinning and changes in memory functioning linked to the loss of oestrogen. However, the evidence of HRT's effects on cardiovascular disease (heart disease and stroke) is more controversial and should be discussed with your doctor in terms of your personal and family history of cardiovascular disease as well as your lifestyle, which can also affect your risk of cardiovascular disease.

Q9 - Are there any alternatives to HRT?

Yes. There are numerous non-hormonal alternatives to HRT, none of which have been shown to be as effective in relieving menopausal symptoms. For people who cannot take HRT these alternatives may be a good option but for those who can, HRT is the best option.

Alternatives include:

- Antidepressants, such as Selective Serotonin Reuptake Inhibitors (SSRIs)
- Gabapentin
- Red Clover
- Ginseng
- Evening primrose oil
- Agnus Castus
- And many more

Some of these options may be prescribed by your doctor, while others can be bought in health shops or over the internet. However, there is no medical proof that freely available herbal options work. These alternatives to HRT may have side-effects and you should always consult your doctor before deciding to use any of these options.

Questions in chapter 7:

- Q1 Is there any screening available for ovarian cancer?
 Q2 What is the alternative to surgery?
 Q3 But I have been offered CA125 blood tests and/or trans-vaginal scans?
 Q4 What would happen if I do not have surgery?
 Q5 Is there anything else I can do?

Q1 - Is there any screening available for ovarian cancer?

No. Unfortunately there is no medically proven screening available for ovarian cancer.

A large trial of ovarian cancer screening finished in December 2011 and the results are not yet available, so we do not yet know whether this screening is effective and can detect ovarian cancer early. Until the results of the trial are available the NHS will not offer routine screening. The trial offered women CA125 blood tests every four months and one yearly trans-vaginal ultrasound scan.

Some areas or GPs may offer private, self-funded CA125 and/or trans-vaginal screening to high risk women, however women need to be aware that there is still no proof that this screening is effective in detecting ovarian cancer early.

You should also be aware that the yearly smear test you receive is designed to detect cervical cancer and will not detect ovarian cancer.

Women from families with Lynch syndrome may receive trans-vaginal ultrasound screening to look for womb cancer. The ovaries may be visible on these scans and if something is found the GP may order a CA125 blood test. In this case the CA125 blood test is a diagnostic test and not a screening test.

Any woman with symptoms of ovarian cancer is likely to be offered a diagnostic CA125 test and trans-vaginal ultrasound scan, however this is not part of a screening programme. Women with no symptoms will not be offered routine screening on the NHS until the screening has been shown to be effective.

Q2 - What is the alternative to surgery?

At the moment the official alternative to surgery is to do nothing and simply be aware of the symptoms of ovarian cancer should they develop. However it is important to realise that these symptoms can be very vague and are not specific to ovarian cancer. If any of these symptoms happen more than 12 times a month you should contact your GP.

The symptoms include:

Persistent bloating (big or swollen tummy)
 Feeling less hungry or feeling full quickly
 Persistent pain in your tummy or below
 Needing to urinate more than usual

Q3 – But I have been offered CA125 blood tests and/or trans-vaginal scans?

Some centres and/or GPs may offer private CA125 tests or trans-vaginal scans to women at high risk of ovarian cancer. As these are not offered as screening tests by the NHS you may have to pay for these yourself. You can choose to have these tests done, but you should be aware that there is no evidence that these screening tests are effective at detecting ovarian cancer early.

Women from Lynch syndrome families may be offered hysteroscopies or trans-vaginal ultrasound scans to detect uterus/womb cancer, which these women are at higher risk for. However usually these tests are designed to detect womb cancer only and not ovarian cancer.

Q4 - What would happen if I do not have surgery?

If you decide not to have surgery, then nothing will really change. You will need to look out for any symptoms and if you think anything is wrong you need to go to your GP to get it checked. Make sure you tell your GP that you have a family history of ovarian cancer and are at increased risk of ovarian cancer when you talk to them.

Remember you can reconsider surgery at any time. If screening for ovarian cancer is shown to be effective in the future, you may then be offered screening as an alternative to surgery.

Q5 - Is there anything else I can do?

As there is no routine screening available, you can choose to have screening privately, but you should be aware that this has not yet been shown to be effective. You should also keep an eye on any symptoms.

Studies have found that a healthy diet with plenty of fruit and vegetables, keeping a healthy weight and an active lifestyle can improve overall well-being and might reduce your chances of getting cancer.

In some countries it is recommended that women at high risk of ovarian cancer take the oral contraceptive pill when they are pre-menopausal. The oral contraceptive pill reduces ovarian cancer risk by up to half, but it has also been found to increase breast cancer risk. The guidelines in the UK do not recommend taking the oral contraceptive pill solely for the prevention of ovarian cancer at the moment, although in some situations reduction in ovarian cancer risk may outweigh any increase in risk of breast cancer. Whether the oral contraceptive pill might be an option for you depends on your risk and family history of breast cancer and should be discussed with your genetic counsellor and GP.

8. Your Decision (Example)

In the exercise on the next few pages you can rate some facts depending on how much they make you want **to have** or want **to avoid** surgery. Give each fact a ranking number (e.g. from 1 (a little) to 3 (a lot), then add the numbers up to see where you stand.

The table below is already filled in to give you an example. On page 26 you will find an empty table for you to fill in.

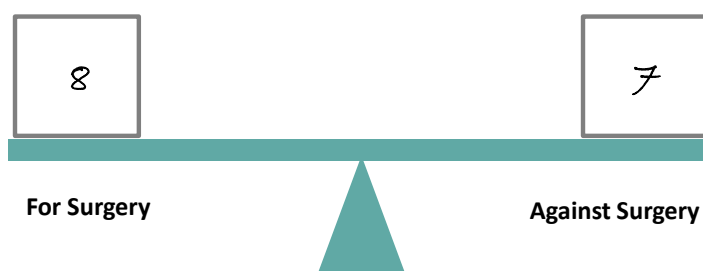
| The fact that... | Makes me want to have surgery | Does not come into my decision | Makes me want to avoid surgery |
|--|--------------------------------------|--------------------------------|---------------------------------------|
| ...surgery will reduce my ovarian cancer risk | 3 | | |
| ...surgery will reduce my breast cancer risk | 2 | | |
| ...I will not be able to become pregnant after surgery | | X | |
| I will go into surgical menopause after surgery | | | 3 |
| There is no effective screening for ovarian cancer | 2 | | |
| The symptoms of ovarian cancer are very vague | 1 | | |
| I might feel different about my body after surgery | | | 2 |
| I might enjoy sex less after surgery | | | 1 |
| Enter your own reasons: | | | |
| Have to take time off work | | | 1 |
| | | | |
| | | | |
| | | | |
| Add together the numbers in each column: | 8 | | 7 |

8. Your Decision (Example)

25

Below you can enter the overall number into the scales. Then note down how you feel about the decision and where you stand.

The scales below are already filled in to give you an example. On page 27 you will find an empty scale for you to fill in.

**More weight for than against surgery:**

Overall your answers show that at this moment in time you are leaning towards having the surgery. Even though you may have some reasons why you would want to avoid surgery, they are not as important to you as the reasons that make you want surgery. Therefore you could now make an appointment to discuss this decision with your doctor and go through any concerns you might have.

Same weights both sides:

Overall your answers show that at this moment you are completely undecided about whether or not to have surgery. Your reasons for why you would want to have and why you would want to avoid surgery balance each other out. Therefore you may want to discuss your options further with your doctor/genetic counsellor.

More weight against than for surgery:

Overall your answers show that at this moment in time you are leaning towards not having surgery. Even though you may have some reasons why you would want to have surgery, they are not as important to you as the reasons that make you want to avoid surgery. Therefore at this moment in time surgery is probably not the right option. However, keep in mind that things may change over time and that eventually you may lean more towards surgery. You can come back to OvDex and do this exercise again anytime to see whether anything has changed.

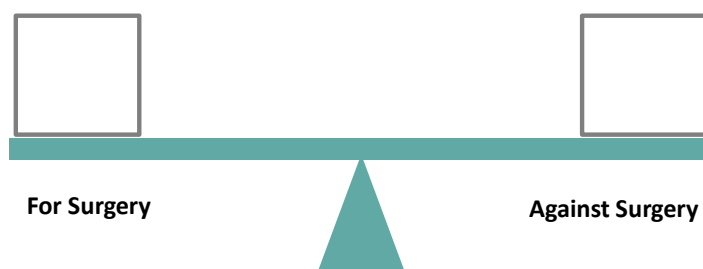
Overall decision: My reasons for surgery weigh more than those
against surgery, but I am still not sure. The
numbers are very similar.

My action plan: I will make an appointment with my genetic
counsellor to discuss the menopause and find out
whether I can take HRT.
Will talk to my boss about taking time off and
whether it would cause any problems.

8. Your Decision

27

Below you can enter the overall number into the scales. Then note down how you feel about the decision and where you stand.

**More weight for than against surgery:**

Overall your answers show that at this moment in time you are leaning towards having the surgery. Even though you may have some reasons why you would want to avoid surgery, they are not as important to you as the reasons that make you want surgery. Therefore you could now make an appointment to discuss this decision with your doctor and go through any concerns you might have.

Same weights both sides:

Overall your answers show that at this moment you are completely undecided about whether or not to have surgery. Your reasons for why you would want to have and why you would want to avoid surgery balance each other out. Therefore you may want to discuss your options further with your doctor/genetic counsellor.

More weight against than for surgery:

Overall your answers show that at this moment in time you are leaning towards not having surgery. Even though you may have some reasons why you would want to have surgery, they are not as important to you as the reasons that make you want to avoid surgery. Therefore at this moment in time surgery is probably not the right option. However, keep in mind that things may change over time and that eventually you may lean more towards surgery. You can come back to OvDex and do this exercise again anytime to see whether anything has changed.

Overall decision: _____

8. Your Decision

Q1 – How can I deal with my decision?

If you are considering surgery, it can help to think about the support you do have and how different people might be able to help you. This can include simple things such as driving you to and from the hospital, helping you out with household chores or childcare issues right after surgery.

Before committing fully to surgery you can discuss any further questions with your doctor or genetic counsellor and you can also seek a second professional opinion at any time if you wish.

If you decide not to have surgery it might be helpful to speak to your GP and let them know about your risk. Some women also find it reassuring to have an action plan in case they feel like any symptoms develop. This can be a simple reminder, such as a plan to contact your doctor if symptoms last 2 weeks or longer.

My action plan:

9. Contacts and Resources

Here are some links and contact details that you might find helpful (even though we have taken great care in choosing websites from reliable sources please note we are not responsible for the content of these websites):

The Cancer Genetics Story Bank – An online collection of stories told by patients and professionals about cancer genetics, genetic testing and risk-reduction decisions. Developed by the Cancer Genetics Service for Wales (CGSW)

www.cancergeneticsstorybank.co.uk

NHS Choices – A website created by the NHS to provide information to patients. Includes information on ovarian cancer, genetics and genetic testing.

www.nhs.uk

Macmillan – A UK charity for anyone affected by cancer with good information about genetic testing for cancer and mind-body therapies

www.macmillan.org.uk or call the support line on 0808 808 00 00

Target Ovarian Cancer – A UK charity which supports research into ovarian cancer and provides useful information including an 'Ask the Expert' feature. Available in several languages.

www.targetovariancancer.org.uk

Ovacom – A UK charity providing information and support for everyone affected by ovarian cancer. Includes links to a number of patient blogs.

www.ovacom.org.uk or call the supportline on 08453710554

Your GP and/or Genetics Service – There to help you with any questions or concerns.

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OvDex (The Oophorectomy Decision Explorer) was developed as part of a PhD project with Cardiff University between October 2010 and September 2013.

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Additional funding was provided by Tenovus - Your Cancer Charity



We also thank our collaborators from the All Wales Medical Genetics Service, the Elizabeth Garrett Anderson Institute for Women's Health and the Charity Target Ovarian Cancer for their support throughout this project.



Appendix 8

| | | |
|-----|--|-----|
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Appendix 8.1 - Multi-Centre Ethics Committee for Wales Favourite Opinion Letter (Amd. 3)

Part of the research infrastructure for Wales funded by the National Institute for Social Care and Health Research, Welsh Government.
Yn rhan o seilwaith ymchwil Cymru a ariannir gan y Sefydliad Cenedlaethol ar gyfer Ymchwil Gofal Cymdeithasol ac Iechyd, Llywodraeth Cymru



Research Ethics Committee (REC) for Wales
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Website : www.nres.nhs.uk

22 October 2012

Miss Jana Witt
PhD Student
Cardiff University
2nd Floor, Neuadd Meirionydd
Heath Park
Cardiff CF14 4YS

Dear Miss Witt

Study title: OvDex - Development and preliminary evaluation of a decision support intervention for risk management options in familial ovarian/breast cancer
REC reference: 11/WA/0094
Protocol number: SPON 945-11
Amendment number: 3
Amendment date: 04 October 2012

The above amendment was reviewed at the meeting of the Sub-Committee held on 19 October 2012.

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

| Document | Version | Date |
|--|---------|-----------------|
| Guidance for lay reviewers | I | 05 October 2012 |
| Advertisement material in newsletter / email | I | 05 October 2012 |
| Changes to REC form | III | 05 October 2012 |
| Participant Consent Form: Consent form for lay reviewers | I | 05 October 2012 |
| Participant Information Sheet: Information sheet for lay reviewers | I | 05 October 2012 |
| Protocol | XI | 05 October 2012 |
| Notice of Substantial Amendment (non-CTIMPs) | 3 | 04 October 2012 |
| Interview Schedules/Topic Guides | I | 05 October 2012 |

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

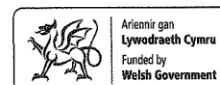
R&D approval

All investigators and research collaborators in the NHS should notify the R&D office for the relevant NHS care organisation of this amendment and check whether it affects R&D approval of the research.



Cynheir Cydweithrediad Gwyddor Iechyd Academaidd y Sefydliad Cenedlaethol ar gyfer Ymchwil Gofal Cymdeithasol ac Iechyd gan Fwrdd Addysgu Iechyd Powys

The National Institute for Social Care and Health Research Academic Health Science Collaboration is hosted by Powys Teaching Health Board



Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

| | |
|--------------------|---|
| 11/WA/0094: | Please quote this number on all correspondence |
|--------------------|---|

Yours sincerely



Dr Gordon Taylor
Chairman

E-mail: corinne.scott@wales.nhs.uk

Enclosures: List of names and professions of members who took part in the review

Copy to: Professor J I Bisson, Cardiff and Vale Research Review Service
Dr K J Pittard Davies, Cardiff University

REC for Wales**Attendance at Sub-Committee of the REC meeting on 19 October 2012**

| Name | Profession | Capacity |
|----------------------|--|----------|
| Dr Maurice Buchalter | Alternate Vice Chairman / Hospital Consultant (Cardiologist) | Expert |
| Ms Nicola Heales | Solicitor | Lay Plus |
| Dr Gordon Taylor | Chairman / Statistician | Expert |

Written comments received from:

| Name | Position |
|--------------|---------------------------------------|
| Dr Pete Wall | Vice Chairman / Clinical Physiologist |

Appendix 8.2 – Topic Guide for Interviews (Usability testing)

First of all I would like to talk about the Option Grid, so that's the A4 sheet of paper with the table of options, is that okay?

- What did you think of the 'Options Grid' table?
 - Probes:
 - What did you expect when you first saw that?
 - Why do you think you expected that?
 - What were the questions that came to mind when you read the grid?
 - Were there any questions that you think should have been included but weren't covered?
 - What did you think about the order of questions in the Option Grid?
- } Questions regarding Option Grid

Now I would like to move on and talk about the longer OvDex decision aid, okay?

- First of all, did you look at the online or printed version?
 - Did you find it easy to access the online decision aid?
 - Probe:
 - If not: What problems did you encounter?
 - How long did you look at it approximately?
 - Probe:
 - Did you look at it more than once, if yes, how often?
 - Can you walk me through how you used the website?
 - Probes:
 - Did you personalise it?
 - What did you expect from that part?
 - What did you think when you read that?
 - Why do you think you thought that?
 - How did that make you feel?
 - Was this what you expected?
 - What did you expect [name of section / question heading] to include?
 - Probes:
 - What do you think ... means?
 - Why do you think that might be important to include?
 - Did you use the weighing up exercise in the tool?
 - Probes:
 - What did you think of it?
 - If you were a woman at risk of ovarian cancer, would you trust the output? Why / Why not?
- } Questions regarding OvDex

For further prompting if needed:

Did you find the tool easy to navigate? Why / why not?

What did you think of the design / format? (i.e. layout, colour etc.)

Was the content easy to understand? Can you give examples?

Was the language straightforward enough, or perhaps too simplistic or too complex?

Was there the right balance between text and diagrams/graphs?

Which parts did you find most interesting? Why?

Where there any parts you struggled with or didn't understand?

Which parts?

Did you have a look at any of the additional resources? What did you think of them?

To finish off this part of the interview, could I just ask...

- Overall, what did you think about the amount of information given (in the grid and/or decision aid)?
Probes:
If too much info, what would you have liked less on?
If not enough info, what would you have liked more on?
- Do you have any suggestions for improvement of the option grid or the website?

Do you think the tool would be helpful/not helpful for patients at risk of ovarian cancer and if so how/why?

- What do you think is especially helpful / least helpful?
Probe:
Why?
- How do you think patients might react to the tool?
Probes:
Why do you think that?
Feel free to use your own feelings as examples.

And the final part of the interview is about how we should use this tool in clinical practice. How do you think this tool should be used in practice, how should it be introduced to patients?

- Who should give it to patients?
- When should it be given to patients?
- Do you think the tool is going to help patients in making a decision?
Why?

| Theme | Code | Details of code |
|--------------------------|--------------------|--|
| Option Grid (OG) content | Layout / Structure | Any comments about what they thought about the layout / structure of the Option Grids; comments about table format, possibility for direct comparison, what they expected when they first saw the format, whether they understood how to read the grid etc. |
| | Understanding | Any comments about whether the information in the Option Grid(s) was clear / unclear, easy to understand etc., any details about what was easy or difficult to understand, any comments on whether something had to be read multiple times to be understood, any comments about cognitive processing of questions / individual terms (i.e. what they thought when they read question / term etc.). |
| | Questions | Any comments about questions that came to mind when reading the information in the grid, questions which they wanted to explore further in response to reading the information in the grid; (NOT questions they think need to be added to the grid [see 'improvements']), |
| | Improvements | Any comments about possible improvements to grids (e.g. ideas for adding content, expanding content, restructuring content and deleting confusing / unnecessary content); comments about questions that might be missing in the grid (incl. if participant feels <u>no</u> questions are missing) (EXCEPT recommendations about changing the order of questions, see code 'Question order/') |
| | Question order | Any comments relating to the order of the questions in the option grid, including any ideas for changing / improving the order |

| | | |
|---|-------------------------------|---|
| OvDex content (OvDex) (web- and paper-based version) | Layout / Structure | Any comments about what they thought about the layout / structure of OvDex; comments about what they thought of the format (paper / website), what they expected to find under each section heading; any comments about section structure (i.e. questions at the top, then answers below etc.); any comments about colour scheme and branding; any comments about graphs / pictograms |
| | Usage | Any comments about time spent using the decision aid, how long they looked at the Option Grids and/or website, how many times they looked at it, whether they came back to it more than once etc.; any comments about using it <u>with</u> someone else |
| | Understanding | Any comments about whether the information in OvDex was clear / unclear, easy to understand etc., any details about what was easy or difficult to understand, any comments on whether something had to be read multiple times to be understood, any comments about cognitive processing of sections / individual terms (i.e. what they thought when they read section / term etc.) |
| | Value clarification (website) | Any comments specifically relating to the content (NOT functionality) of the online value clarification exercise, whether they understood the instructions and how to use it, whether they tried it out, what they thought when they did, whether they found it helpful or not, why they found it helpful / not helpful (any comments about problems with functionality must be coded 'OvDex website, functionality') |

| | | |
|---|-----------------------------|---|
| OvDex content (OvDex (web- and paper-based version) cont. | Value clarification (paper) | Any comments specifically relating to the content of the paper value clarification exercise, whether they understood the instructions and how to use it, whether they tried it out, what they thought when they did, whether they found it helpful or not, why they found it helpful / not helpful |
| | Other sections | Any comments on sections other than the information content and value clarification exercise; e.g. comments on contacts and resources, comments on external links they followed, comments on research evidence section, comments on 'about OvDex' section with disclosure of funding sources and collaborators. |
| | Improvements | Any comments about possible improvements to OvDex (e.g. ideas for adding content, expanding content, restructuring content and deleting confusing / unnecessary content). |
| | | |
| OvDex website | Accessibility | Any positive / negative comments relating to getting access to the online decision aid on a PC, iPad, mobile phone |
| | Functionality | Any comments on interacting with the decision aid's interactive functions (particularly the personalisation and value clarification sections); did user try to personalise OvDex, did user do the value clarification exercise and found it easy to interact with; any comments on things that worked / did not work (e.g. broken links, limited interactivity) |

| | | |
|-------------|--|---|
| | Navigation | Any comments on navigating the decision aid; any descriptions of how participant read through the decision aid and individual pages (did they read through sequentially, did they skip sections, did they follow links, did they go backward and forward between pages or jump to questions) |
| | | |
| OvDex paper | Navigation | Any comments on navigating the decision aid booklet; any descriptions of how participant read through the decision aid and individual pages (did they read through sequentially, did they skip sections, did they go backward and forward between pages or jump to questions) |
| | | |
| Overall | Impact / Effects (general, encompassing the whole decision aid) | Any comments about whether participant thinks the tool would be helpful / useful for themselves and/or women at risk of ovarian cancer and why they think this, any comments about how they or patients reacted / might react to the tool, any comments about feelings experienced whilst using the tool; any comments regarding how or whether the decision aid would facilitate decision making; any comments about wanting to or having recommended the decision aid to others |
| | Information amount | Any comments regarding the amount of information on the decision aid, whether it is too little, too much (specific ideas for improvements must be coded as such, i.e. under 'Improvements' for Option Grid or website) |

| | | |
|----------------|---|--|
| | Most helpful (specific to certain sections) | Any comments that are particularly positive, refer to sections / items they liked and found particularly helpful (can be both Option Grids and OvDex) |
| | Least helpful (specific to certain sections) | Any comments that are particularly negative, refer to sections they did not like and found particularly unhelpful (can be both Option Grids and OvDex) (specific ideas for improvements must be coded as 'Improvements') |
| | | |
| Implementation | Provider | Any comments about who should give the decision aid to patients, e.g. doctor, nurse, genetics specialist, oncologist etc. and where it should be given to patients, e.g. surgery, genetics service consultation etc. |
| | Timescale | Any comments about when the decision aid should be provided during the decision making pathway and why they think this time point is suitable |
| | | |
| Other | Medical History | Comments relating to personal or family history of diseases (incl. cancer and other diseases) |
| | Decision | Any comments about what the participant would decide for herself before / after using the tool |

| | | |
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| | Question | Any questions asked by participants and answers given by interviewer |
| | Reference | Any comments about things the interviewer, researcher should look up |
| | Misc | Any comments that do NOT readily fit with the suggested codes |

Appendix 8.4 - Extract of Coded Transcript (Usability testing)

| | | | | |
|---|---|---|---|------------------------------------|
| R: Yeah, it's a really great looking website as well, it's really nice looking so it's really easy to surf through | } | <u>OvDex</u> Layout / Structure + | | |
| I: Oh great, OK so you em, you found it quite em, easy to access it and then navigate around it did you? | | | } | <u>OvDex (web)</u> Navigation |
| R: Yeah, really easy to use, yeah and really not patronising, it gave you the information that you required and it gave you more detail if you wanted it, so it was good | | | | |
| I: Oh good, OK and how long, approximately, if you remember did it take you to go through it? | } | <u>OvDex</u> Usage | | |
| R: It took me about an hour I'd say | | | } | <u>Other</u> Medical History |
| I: OK | | | | |
| R: But I had to go through the general 'cos obviously I don't have the symptoms, I, I'm not one of these people, I couldn't say, I couldn't specify it for me. And I think it probably takes less time if you knew which faulty gene you had. | | | | |
| I: Yeah, yes, yeah, and I mean, you did see the personalised option em on the website did you? | | | } | <u>OvDex (web)</u> Function |
| R: Yeah | | | | |
| I: OK, great, yeah | | | | |
| R: Yes, I tried, I tried to do that first, I thought oh, maybe I'll pretend I've got Lynch Syndrome. | | | | |
| I: Yeah, you could have, yeah | | | | |
| R: That's right, then I thought well I wont, I wont, I don't know what that's going to miss out and I'll probably end up having to go through it a few times so I just did it generally but if I had, if I was one of the people who had been told... | | | | |
| (I: Yeah) | | | | |
| R: ...that I was at high risk and knew what it is, I would absolutely use the em the one that's restricted to you because it's giving you the information that's particularly relevant to you and I think you can get a bit scared or overly concerned if you're reading about something that doesn't even actually apply to you. | } | <u>Overall</u> Impact / Effect | | |

I: Yes, yes I mean that's partially why we did the personalisation option em, for those women, yeah, you're absolutely right and if you had had the information then you probably would have done that, that's good to know ((laughs)) and it all seemed to work

R: Yes

I: As well which is also good to know

R: Yeah, yes

I: Em, could you just er, walk me through how you used the website? Did you look at everything in, in sequence, these kind of things

R: Yeah, I mainly looked at things, I tried to look at things in sequence, so I went into the first section with the hyperlink that's used y'know, if this does not apply to you then you can browse the information using our menu or view the first section on cancer risk so I clicked on the cancer risk hyperlink there. Em, and I worked through most of it in the logical order, so I'd get to the end of the page and I'd click on the, y'know I'd click on the next page tab at the end, but in a few occasions there was a hyperlink, though I definitely remember I went to the menopause one early so I think in the, when I was looking at something and it had a hyperlink to the menopause that I, or the HRT bit, the hormone replacement bit, I clicked on that and then I realised later on, oh I would have come to that page anyway

I: Oh, OK yes, I see now, yeah OK so you, you em realised later that you would have come to it naturally but you

R: Yes

I: Skipped it in the first instance

R: Yeah, and I guess because it was one of the areas I knew the least about

I: Yeah

R: And, and it was mentioned early on, and I thought, ooh, whilst that's fresh in my mind I'll look at that, so I like that because you can do it both ways can't you, if you miss the hyperlink or whatever you're gonna come across the information anyway but equally if that's all you want to know about then you can just go straight to it

I: Yeah, yes, I mean that's er, the idea behind the hyperlinks and you can go backwards as well if you feel like you might have missed something em

R: Ah yeah, cool, yeah

OvDex (web)
Function

OvDex (web)
Navigation

| Comments from lay reviewers | Potential amendment(s) | Management team decision |
|---|---|---|
| <p>R1: I thought they were quite good [...]. I think just from an ease to read point of view, there just needs to be a line to separate the frequently asked questions and then [...] what the options were because I read it first off and then was a little bit confused. (35to39, noOC, B&OCFH, BRCA2)</p> | <p>Make the Frequently Asked Questions column more distinct</p> | <p>Frequently Asked Questions column was coloured in light orange</p> |
| <p>R6: Most people will read this and think: ‘Hang on, I’m getting my ovaries taken out so surely that’s going to completely remove the risk of ovarian cancer.’ But you do [...] later on explain that peritoneal cancer is part of the ovarian cancer class [...]. I wonder if that’s really clear to your average person on the street. (40to49, yesOC, BCFH, BRCA1)</p> <p>R6: I suppose again in the first part of the Grid when you’re saying: ‘Yes removing the ovaries and fallopian tubes will reduce your lifetime risk of ovarian type cancer.’ Again you’ve got ‘ovarian type cancer’ and I’ve just kind of put again primary peritoneal question mark there. So I guess if you kind of explain a little bit more in the first paragraph then that’s fine. (40to49, yesOC, BCFH, BRCA1)</p> | <p>Change wording in opening paragraph to clarify that surgery reduces risk of ovarian-<u>type</u> cancer; Explain what is defined as ‘ovarian-type’ cancer</p> | <p>Wording of opening paragraph was changed to “...surgery reduces risk of ovarian-<u>type</u> cancer...” No explanation was added to describe ‘ovarian-type’ cancer, as this is included in the full OvDex tool and as the Option Grid would be used together with a health professionals who could explain the term if required</p> |
| <p>R4: I think that flows quite well too really, except maybe the very last question should go first. (Over50, yesOC, noFH, unknown)</p> <p>R8: I think the routine screening question should probably be closer to the top. Although it’s a bit of a negative then but it’s still I think one of the first things you think about. (35to39, noOC, OCFH, unknown)</p> <p>R1: [The grids] are fairly logical [...]. It sort of works its way through the different questions, I think the order is okay. [...] You could put the screening question up at the front, [...] because I suppose really that might be an early question. (35to39, noOC, B&OCFH, BRCA2)</p> <p>R5: I think possibly I would say the routine screening programme should</p> | <p>Move screening question further towards the top of the grid</p> | <p>Screening question was moved up to be the second question in the grid</p> |

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| be higher, because that's a major thing, because if there's some screening programme you wouldn't want to go any further. (Over50, yesOC, noFH, unknown) | | |
| R2: My concerns were the menopause, the HRT and the health risks if I had surgery. For me they are the most important. For me they would want to be at the top. That's my feeling anyway. (Under35, noOC, B&OCFH, BRCA2) | Move 'How I feel about my cancer risk' and 'How I feel about myself' questions further down the grid | Not actioned, as all other respondents were satisfied with the order of questions in the Option Grids and the respondent herself admitted later that the order did not make a difference |
| R12: The 'how I feel' question [...], I can see why it's in there, but I wasn't entirely sure that [...] the answers actually added anything. [...] It doesn't give you any facts, [...] it's quite bland. [...] Have women who have had to have this operation at thirty-three, or chosen to have this operation at forty-three or whatever, have they come back and said: Oh god, I really regret it, I wish I hadn't had it? I think that sort of information would be more useful (Under35, noOC, OCFH, unknown) | Add factual information to 'Will this change how I feel about my risk...' question ; Move 'How I feel' question further down the grid | Factual information about satisfaction with RRSO was added (Finch, 2011; Tiller 2002; Westin 2010); The question was not moved further down the grid, as all other respondents were satisfied with the order of questions in the Option Grids |
| R12: The only thing that I didn't know in much detail when I was looking at the option grid, is what the actual operation was. But I'm assuming that these people would have been talked through the operation before they see the grid. (Under35, noOC, OCFH, unknown) | Provide clearer definition of what the surgery is in Option Grid. | Not actioned, as this is included in the full OvDex tool and as the Option Grid would be used together with a health professionals who could explain what surgery is |
| R6: The question 'Will surgery change how I feel about myself as a woman?' And you've said 'However your desire for and or enjoyment of sex may decrease' and I've put yes exclamation mark, because sorry I don't know you but I am totally not interested now. [...] And I just thought well maybe you should be a little bit stronger about that. (40to49, yesOC, BCFH, BRCA1) R3: I think yes it does give your sexuality a bit of a knock. That's all I'd say | Change wording of 'How I feel' question to be stronger regarding sexual functioning | Wording changed to "...is likely to decrease" |

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| really there. (Over50, yesOC, BCFH, BRCA2) | | |
| R10: Generally speaking I've had a very normal sex life and although we don't shall we say have conventional sex now but we do have intimate moments shall we say [...]. There are ways round things. [...] We're all different and 'Your desire for sex' it says 'will decrease'. It might not, might it? That's on page 13, number 3. (Over50, yesOC, BCFH, unknown) | Reduce emphasis on reduced sexual functioning | Wording changed to "...is likely to decrease" |
| R12: In what percentage of cases I guess does it go from keyhole to full surgery? (Under35, noOC, OCFH, unknown) | Add statistical info about number of keyhole versus open surgeries | Not actioned, as this is included in the full OvDex tool and as the Option Grid would be used together with a health professionals who could explain, if required |
| R13: It might be a good idea just to mention that it can be quite traumatic afterwards for a few weeks. I wasn't too bad, but a lot of women can experience extreme tiredness [...]. They need to know all this, especially if they're looking after somebody or they need to work. (Over50, yesOC, BCFH, unknown) | Add information about post-surgical pain, tiredness and other issues affecting functioning | Added statement about post-surgical effects during recovery saying "...during which time you may feel pain / tiredness and need help with everyday tasks." |
| R6: It says: 'No after surgery there is no need for screening because your risk will be similar to that of the general population' and I thought: 'Well surely it's going to be lower actually?' [...] So I just thought well maybe using the word similar there isn't really showing that there's much of a difference between you having gone through the operation and somebody out of the general population who has not. So it's almost like making you think: 'Oh hang on a minute there's really no benefit then of having the operation.' (40to49, yesOC, BCFH, BRCA1) | Reword / clarify answer to screening question in surgery column to avoid confusion, e.g. remove/reword "similar to that of the general population" | Reworded answer to screening question to "No, there is no evidence that screening for ovarian cancer is effective and after surgery there is no need for screening as your risk will be low." |
| R1: [The grid] was very black and white, [either] you have surgery or you don't. And it's almost like if you don't have surgery you take your life in your own hands, whereas there is the possibility of having ultrasounds | Clarify <u>why</u> there is no evidence that screening is effective (i.e. no proven survival benefit due to early detection) | Reworded answers to screening question to "No, there is no evidence that screening for ovarian cancer is |

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| <p>for early detection [...]. I'm lucky that I can get that done but I don't know if other [NHS] Trusts [...] do that, so I suppose that's quite a tricky thing. (35to39, noOC, B&OCFH, BRCA2)</p> <p>R13: I know the CA125 isn't a screening [test], it's more of a diagnostic [test], but [...] why can it not be a screening [test]? (Over50, yesOC, BCFH, unknown)</p> | | <p>effective in saving lives." and "No, there is no evidence that screening for ovarian cancer is effective and after surgery there is no need for screening as your risk will be low."</p> |
| <p>R10: If you were having surgery [the answer] didn't answer the question. [...] Whereas if you were not having surgery it did answer the question, because there isn't [a screening programme]. It should have said: 'No there isn't a screening programme and after surgery anyway...' I feel it needs a little addition. (Over50, yesOC, BCFH, unknown)</p> | <p>Reword answer to screening question in surgery column</p> | <p>Reworded answer to screening question to "No, there is no evidence that screening for ovarian cancer is effective and after surgery there is no need for screening as your risk will be low."</p> |
| <p>R5: Lots of women seem to think that if you have your smear test you're being checked for ovarian cancer at the same time so I think to make it very clear that that is not the case and there is no screening. There is no way of telling from any sort of check that is done. I think that needs to be something that would then concentrate their minds. (Over50, yesOC, noFH, unknown)</p> | <p>Make it even clearer that there is no screening for ovarian cancer; Emphasise that the smear test does not test for ovarian cancer</p> | <p>Reworded answers to screening question to "No, there is no evidence that screening for ovarian cancer is effective in saving lives." and "No, there is no evidence that screening for ovarian cancer is effective and after surgery there is no need for screening as your risk will be low."; Information about smear test was not added, as this is included in the full OvDex tool and as the Option Grid would be used together with a health professionals who could explain, if required</p> |
| <p>R4: On the post menopausal women thing I think that there could be</p> | <p>For post-menopausal grid, add</p> | <p>Not actioned, as current guidelines do</p> |

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| <p>something added about taking HRT [...] afterwards, because I am post menopausal obviously, but I was on HRT before I was diagnosed and have had to go back onto some oestrogen only HRT after. (Over50, yesOC, noFH, unknown)</p> | <p>information on hormone treatment options</p> | <p>not recommend any HRT for women who are over 50 / postmenopausal</p> |
| <p>R5: [In the pre-menopausal grid] it talks about the risk of bone thinning and cardiovascular disease, but that I think would be [...] reassuring to me if I only looked at the one for after the menopause to be told that actually the risk is t[...]to women before age 45. [...] Because that would actually be quite reassuring and would quite possibly help to make me make a decision. (Over50, yesOC, noFH, unknown)</p> | <p>For post-menopausal grid, add section on long-term health effects due to hormone deprivation, which are <u>not</u> relevant to this population</p> | <p>Not actioned, as Option Grids are designed to be uncomplicated and inclusion of irrelevant data for the target population may confuse users. Postmenopausal women will only receive the postmenopausal grid, hence they would not be alerted to the long term health risks affecting premenopausal women, as were participants in this study who viewed both grids.</p> |

| Comments from lay reviewers | Suggested amendment | Management team decision / action |
|---|--|---|
| Personalisation | | |
| <p>R5: Right at the beginning it confused me, once I got into it, it didn't at all, but the very beginning confused me. [...] The way it comes in at the moment it just says: 'Welcome to OvDex, please note it can be personalised you should have seen and answered these questions before viewing these pages.' [...] You think 'Oh hang on what questions then?'. (Over50, yesOC, noFH, unknown)</p> | <p>Ensure that <u>Index page</u> appears first when www.OvDex.co.uk is opened</p> | <p>Index page was made default home page for URL www.OvDex.co.uk</p> |
| <p>R6: I actually tried to use the online thing because I thought: 'Well I'll personalise it to myself obviously, because I've got a BRCA 1 mutation.' [...] And it didn't seem to want to work, so it [...] just came back to the original question. (40to49, yesOC, BCFH, BRCA1)</p> | <p>Review personalise functionality and ensure it is working correctly (if Return button does not work, add instructions that users need to use the 'Submit' button at the bottom of the screen)</p> | <p>Personalise functionality was reviewed in available browsers</p> |
| <p>R5: The thing that I thought was a bit odd at first is the 'it can be personalised'. I would have thought you would want it to be personalised really. [...] I don't know [under] what circumstances you wouldn't want to personalise it. (Over50, yesOC, noFH, unknown)</p> <p>R5: It says you can personalise or visit the general version if you don't want to personalise. [...] There's nothing there to tell people what that means. (Over50, yesOC, noFH, unknown)</p> <p>R9: I kind of wished that I'd been directed to personalise it first. (Over50, yesOC, noFH, unknown)</p> <p>R9: I wasn't quite sure, because it doesn't really give you very much information at all as to whether it's gonna be more helpful to personalise</p> | <p>Emphasise that personalisation is advantageous in the instructions and guide people further towards using the personalise function rather than viewing the general version.</p> | <p>Section on Index page was reworded to: "The information in OvDex can be tailored to your personal situation by answering three simple questions. We recommend that you use this function, so that the information displayed is more relevant to you. You can Personalise OvDex by clicking here. If you do not wish to personalise OvDex, you can visit a General Version here, but please be aware that some of</p> |

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| <p>it or visit the general version. (Over50, yesOC, noFH, unknown)</p> <p>R9: I kind of wonder whether it might be [useful] as well to guide people a bit more there and say that by personalising it, [it] is gonna be exactly that, it's gonna be personalised more, the information will be most relevant to what you need. (Over50, yesOC, noFH, unknown)</p> <p>R9: it might have been better to say: There is general information, but it will probably be most helpful to you to personalise it to your situation and read that first'. (Over50, yesOC, noFH, unknown)</p> <p>R9: I would definitely try and sort it round so that you personalise it and then, if after you've read your personalised information you would like to just look at the general version you can do so, but try and get people not to waste time on the general version. (Over50, yesOC, noFH, unknown)</p> <p>R9: There's nothing on that open page that tells you how simple it's gonna be to personalise it. (Over50, yesOC, noFH, unknown)</p> <p>R13: I didn't realise, I'm not very good with the internet, I don't know why, why I didn't see the personalised one. (Over50, yesOC, BCFH, unknown)</p> | | <p>the information in this version may not apply to you.”;</p> <p>Size of text was changed to be bigger</p> |
| <p>R5: 'The information on the following pages has not been personalised. If you do not wish to personalise OvDex then you can view the general information using the menu or see the first section on cancer risk.' That doesn't tell you you're actually going to go in to it. [...] It's not clear that you're actually going to go into, at this point now. (Over50, yesOC, noFH, unknown)</p> | <p>On home pages once personalisation has been done / general version selected, make it clearer that the users are now accessing the decision aid content.</p> | <p>Added statement on introductory home page after personalisation / in General version saying: “You have now entered the decision aid.”</p> |
| <p>OvDex content</p> | | |
| <p>R13: It was helpful, but far too much. It could have been condensed and</p> | <p>Remove repetitions;</p> | <p>OvDex tool was checked for repetitions</p> |

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| <p>[...] probably been a lot clearer [...]. Just far too much reading. (Over50, yesOC, BCFH, unknown)</p> <p>R13: I thought that was [...] [a] bit ambiguous, I thought it was too much, too much in there actually. [...] Sometimes less is more. [...] Sometimes people give up because it's just too much information. (Over50, yesOC, BCFH, unknown)</p> | <p>Shorten content</p> | <p>and those were removed; Content was not shortened, as all other respondents were satisfied with the information amount</p> |
| <p>R6: Personally I think a section about the disease would be really useful, You know and statistics of how much treatment people have to have and all this, that and the other. (40to49, yesOC, BCFH, BRCA1)</p> <p>R6: I thought I wonder if I don't know you should really include somewhere about the cancer and you know basically if you don't catch it early then what can potentially happen to you as well. [...] I mean I'm not saying you should sort of frighten women into having the surgery but I think they need all the facts in front of them. (40to49, yesOC, BCFH, BRCA1)</p> | <p>Add information / section about ovarian cancer or add links to this information</p> | <p>Added statement "For more information about ovarian cancer, check out the Cancer Research UK statistics and other websites under Contacts and Resources" with links to CRUK statistics and Contacts & Resources (e.g. NHS, Ovacome, Target Ovarian Cancer) for information about ovarian cancer</p> |
| <p>R11: You know the little bits with all the circles on? [...] I would change those to little people. [...] It says the pictures below, but they're not pictures and they're sort of blobs. I like more sort of pictury things. (Over50, yesOC, noFH, unknown)</p> <p>R5: It reminded me of a theatre seating plan yeah. It looks exactly like an online booking service doesn't it? [...] But no, I think that's really good because it's a good clear display [...]. The dots indicate the risk that you're talking about. (Over50, yesOC, noFH, unknown)</p> | <p>Change dots in pictograms to small female signs</p> | <p>Changed dots to small female signs in pictograms</p> |
| <p>R6: What I wasn't really sure about was your pictures showing the chances of getting cancer [...]. You seem to do this quite a lot all the way through. [...] I suppose it's quite a nice indication, but it's taking up quite a lot of room. I don't know if it's totally necessary, because people can</p> | <p>Remove pictograms</p> | <p>Not actioned, as positive feedback from a number of other respondents indicated that the pictograms are valued by many women</p> |

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| just as easily look at numbers. (40to49, yesOC, BCFH, BRCA1) | | |
| R12: The only thing I was surprised about was that it went in at cancer risk. [...] It was appropriate for me as somebody who hadn't been told that I'd got a high risk. [...] surely everyone who's using this website has already gone through that process so is the explanation at the beginning necessary? (Under35, noOC, OCFH, unknown) | In general / untested version, add that some questions are not relevant to people who have already been tested; Add intro sentence to each section before question list to explain what section is about (so users can decide whether they want to read on) | Not actioned, as section layout already lists questions included in the section at the start and allows users to jump to questions, ignoring those that they feel are irrelevant |
| R11: How could I get tested you know without having a family history and that kind of thing. (Over50, yesOC, noFH, unknown) R6: I know I'm an unusual case but basically I don't have any family. [...] There might be other people in my situation who are not being offered the genetic test and in fact they could probably benefit from it. [...] [Say] if there's some sort of private test thing that people could go and have done if the NHS won't cover it. (40to49, yesOC, BCFH, BRCA1) | Add info about who can / can't get tested for BRCA and why. | Added information about possibility of private testing "For anyone who cannot get tested on the NHS, there are some companies offering private testing, however this can be expensive and without a family history, there may be no need for testing."; Emphasised requirements for testing on the NHS |
| R11: I wanted to know a little bit more about the BRCA test and whether or not it was available if I just walked in [...] And also probably another question about how quickly results would come back and about what that would involve. (Over50, yesOC, noFH, unknown) | Add information about what gene test involves and how long results take to come back. | Added statement about gene test, timeline and possible outcomes. |
| R12: Adding a sentence in to say, if you definitely have the gene then your child can be tested in the future or something. (Under35, noOC, OCFH, unknown) | Add information that if a gene mutation is confirmed in the family, children can choose to get tested | Added information that if a gene mutation is confirmed in the family, children can choose to get tested |
| R12: [Speaking about 'Does my risk change with age?' question] it's the only thing I had to read twice. [...] I would say that and if there's any way you could show it in [...]diagram form, like you have with the circles we | Shorten answer to 'Does my risk change with age?' question and replace with diagram / graph. | Shortened answer to "Does my risk change with age?" question to: "Yes, cancer risk increases with age"; |

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| <p>talked about, [...] that would be really useful. (Under35, noOC, OCFH, unknown)</p> <p>R12: It does get more risky the older you get, that's all you need to know really. (Under35, noOC, OCFH, unknown)</p> | | <p>Added graph showing risk curve</p> |
| <p>R6: Why is the operation most effective if it's done at 40 years of age? (40to49, yesOC, BCFH, BRCA1)</p> | <p>Add information about why surgery is most effective at that age</p> | <p>Added statement about why surgery is most effective at 40 years of age: "The operation is most effective if it is done at 40 years of age, because ovarian cancer risk starts to rise from age 40 onwards. Additionally, if done at 40, the surgery reduces the risk of breast cancer. However, age 40 may..."</p> |
| <p>R1: 'If you're unsure about it you could let a doctor make the decision' [...] I felt quite uncomfortable with that because, to my mind [...], if you can't make the decision whether to have surgery or not, then you shouldn't be having surgery. [...] Maybe it needs to be reworded or taken out or something. [...] I wasn't comfortable with that wording and I don't think it's that helpful for anybody. I think it's definitely a decision the person that's having the surgery has to make. (35to39, noOC, B&OCFH, BRCA2)</p> | <p>Review wording of this section and reword</p> | <p>Not actioned, as reviewed wording and only identified statement saying "If you then don't want to choose for yourself, just say so and they [doctor] may make a recommendation.";</p> <p>Did not change coping advice in "The Choice" which includes option 'The doctor should decide for me', as this is a valid option if a patient does not want to make the decision.</p> |
| <p>R6: You go sort of straight into risk reducing surgery. And I thought you know a lot of people might be quite scared of having surgery [...]. It's quite good for people to see at the beginning other things that are available or aren't available. (40to49, yesOC, BCFH, BRCA1)</p> | <p>Mention screening earlier on</p> | <p>Screening question was moved up to be the second question in the grid (and therefore "Options at a Glance" section)</p> |
| <p>R10: The thing about feelings and anxiety [...] gets reproduced a lot in</p> | <p>Shorten, remove repetition in coping</p> | <p>Reworded answers to make them more</p> |

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| <p>this book. [...] I think it could possibly be a little less. (Over50, yesOC, BCFH, unknown)</p> | <p>advice 'How can I deal with my cancer risk' / 'How can I deal with this decision'.</p> | <p>distinct</p> |
| <p>R9: My first thought was, hang on, 'How can I DEAL with this decision?' and I wasn't sure if that was quite the right wording. Because I wasn't sure what that meant, I wasn't sure whether I was thinking with question one 'How can I MAKE this decision?' [...], I did wonder whether it was something about: What will help me make the best decision for me? Or: What will help me make this decision? [...] I wasn't sure about the word 'deal'. (Over50, yesOC, noFH, unknown)</p> | <p>Reword Q1 in Options at a Glance section</p> | <p>Reworded question title to "How can I cope with this decision?"</p> |
| <p>R6: You're talking about watching the how do I cope video on the cancer genetics story bank website [...]. I just wondered if it was an idea to sort of put the link to that at the end of that paragraph. [...] And then also you are talking about the MacMillan website in the next paragraph and I thought I wonder if you'd put the link to that there. (40to49, yesOC, BCFH, BRCA1)</p> | <p>Add link to Genetics Story Bank and Macmillan website to reference in text</p> | <p>Added link to Genetics Story Bank and Macmillan website to reference in text</p> |
| <p>R13: It might be a good idea just to mention that it can be quite traumatic afterwards for a few weeks. I wasn't too bad, but a lot of women can experience extreme tiredness [...]. They need to know all this, especially if they're looking after somebody or they need to work. (Over50, yesOC, BCFH, unknown)</p> | <p>Add information about post-surgical pain, tiredness and other issues affecting functioning, which wouldn't be defined as complications but are all part of normal recovery.</p> | <p>Added statement to "How would RRSO affect my life?" question about post-surgical effects during recovery saying "... heavy lifting. Therefore you may need help with everyday tasks. During recovery, you may also feel some pain and discomfort and you may be more tired than usual. However most women..."</p> |
| <p>R6: It says 'having the surgery will also prevent other non-cancer related problems happening with the ovaries such as cysts' and I just thought</p> | <p>Add information about why it might be good to avoid other problems with</p> | <p>Added statement to advantages of surgery list: "...such as cysts, which can</p> |

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| again maybe you should sort of expand on that a bit and maybe explain why it's bad to have cysts. (40to49, yesOC, BCFH, BRCA1) | ovaries. | cause some discomfort and pain." |
| R6: some people might say oh well if you're removing the ovaries why don't you remove the peritoneum as well? [...] You and I know that you just don't, you can't do that. [...] So you might just want to explain why that can't be done as well. (40to49, yesOC, BCFH, BRCA1) | Add sentence about that peritoneum cannot be surgically removed. | Added statement that peritoneum cannot be removed to "What is my risk after surgery? " question: "...and pelvis. Unfortunately, the peritoneum cannot be routinely removed during RRSO." |
| R6: I just thought some people might be wondering, well they might even be scared that if they're opened up for this surgery and cancer is found what actually happens then? (40to49, yesOC, BCFH, BRCA1) | Add info about next steps / procedures if cancer is found during the surgery. | Added information to "Could cancer be found during the surgery?" question saying: "If cancer is found during this check, you will be informed and might have to undergo further tests and possibly treatment for ovarian cancer." |
| R6: I just wondered if you would get hormone replacement if cancer had been found during the surgery and I thought well probably not. [...] I just didn't know whether you should sort of maybe say look you don't get hormone replacement if you've actually got cancer. (40to49, yesOC, BCFH, BRCA1) | Add statement about HRT if ovarian cancer is found. | Added statement to "Could cancer be found during the surgery?" question saying: "If cancer is found you might be unable to take HRT." |
| R6: The question 'Will surgery change how I feel about myself as a woman?' And you've said 'However your desire for and or enjoyment of sex may decrease' and I've put yes exclamation mark, because sorry I don't know you but I am totally not interested now. [...] And I just thought well maybe you should be a little bit stronger about that. (40to49, yesOC, BCFH, BRCA1) R3: I think yes it does give your sexuality a bit of a knock. That's all I'd say really there. (Over50, yesOC, BCFH, BRCA2) | Increase emphasis on reduced sexual functioning | Wording changed to "...is likely to decrease" |
| R10: Generally speaking I've had a very normal sex life and although we | Reduce emphasis on reduced sexual | Wording changed to "...is likely to |

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| don't shall we say have conventional sex now but we do have intimate moments shall we say [...]. There are ways round things. [...] We're all different and 'Your desire for sex' it says 'will decrease'. It might not, might it? That's on page 13, number 3. (Over50, yesOC, BCFH, unknown) | functioning | decrease" |
| R6: I mean if you're restricted to length then I would definitely maybe reduce the menopause section a bit. (40to49, yesOC, BCFH, BRCA1) | Reduce menopause section content. | Not actioned, as results from the needs assessment indicated that information about the menopause is extremely important to many women and as other respondents during the usability testing indicated that they appreciated this section and detailed information. |
| R9: There was one bit there, [...] 'the alternatives to HRT' [...] I didn't know whether the ones that were listed were the most commonly used [...] I didn't know why, in particular, you'd listed those. (Over50, yesOC, noFH, unknown) | Add statement about how examples of alternatives were selected. (e.g. 'Alternatives frequently used include ... however list is not exhaustive) | Added statement to "Are there any alternatives to HRT?" question: "Alternatives that are frequently used include..." and "Please note that this list is not exhaustive and there may be other alternatives to HRT." |
| R9:[About 'Alternatives to surgery' question]: I think it's that term 'alternative', that I was thinking of something very different to surgery so I think maybe what I was thinking would be clearer for me would have been to say, 'Are there other treatments apart from surgery'? [...] Just on the previous page you're reading about alternatives to HRT and you're talking about herbal things and everything so I've just gone off on that track of thinking and then I was distracted by the use of the word alternative again. (Over50, yesOC, noFH, unknown) | Reword question 'What are the alternatives to surgery?' | Reworded question title to "Are there other options apart from surgery?" |
| R13: I think it gives women, if they don't know about the CA125 [...] looking at that it seems as though [...] there's just nothing there [...], | Reword section on no effective screening again to emphasise that | Reworded "Is there any screening available for ovarian cancer?" question |

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| <p>because in my mind if there's that CA125 then there is an effective screening. (Over50, yesOC, BCFH, unknown)</p> <p>R1: [The Option Grid] was very black and white, [...] it's almost like if you don't have surgery you take your life in your own hands, whereas there is the possibility of having ultrasounds for early detection. [...] I'm lucky that I can get that done, but I don't know if other [NHS] Trusts or anything do that so I suppose that's quite a tricky thing. (35to39, noOC, B&OCFH, BRCA2)</p> <p>R4: I didn't know that the screening tests were not offered by the NHS [...] and that you might have to pay for those because I had it, my GP just referred me for it. (Over50, yesOC, noFH, unknown)</p> <p>R5: I think that was good to say [...] the bit about screening, that [...] there isn't really. [...] This focuses your mind again after you've taken all that information in about all the different things. [...] I'm a clear indication of what you're saying here actually that the screening doesn't work necessarily. It's not you know, there isn't a screening that is medically proven to work. There isn't. (Over50, yesOC, noFH, unknown)</p> | <p>although screening might be bought privately, it is not effective at detecting cancer early and might provide false reassurance; emphasise the difference between screening and diagnostic tests</p> | <p>paragraph 2: "Two large trials of ovarian cancer screening were done recently and the results are not yet available, so we do not yet know whether this screening is effective and can detect ovarian cancer early. The UKFOCS trial offered high risk women CA125 blood tests every four months and one yearly trans-vaginal ultrasound scan. The UKCTOCS trial offered such screening to women over 50, who were not at high risk. Both trials have not reported their final outcomes. Routine screening will not be offered until the results of these trials are available and only if these trials show that screening is effective and can save lives."</p> <p>Added a link to UKFOCS and UKCTOCS study pages</p> <p>Added statement to "Is there any screening available for ovarian cancer?" question paragraph 3 "...ovarian cancer early. Therefore going to private screening may provide false reassurance. Additionally, screening may result in unnecessary</p> |
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| | | <p>worry and anxiety if any results show changes that turn out not to be cancer.”</p> <p>Reworded section in “Is there any screening available for ovarian cancer?” question paragraph 6 “...CA125 test and trans-vaginal ultrasound scan. Please note that these diagnostic tests are <u>not</u> screening tests. Rather, they are designed to detect ovarian cancer when symptoms have already developed. Women with no...”</p> <p>Added statement to “But I have been offered CA125 blood tests and/or trans-vaginal scans?” question paragraph 1 “...ovarian cancer early. Therefore using such screening services might provide you with false reassurance.”</p> |
| <p>R13: I think the most important thing is, is to push the symptoms because they are so vague and so near to what IBS or something could be. [...] It needs to push the symptoms into people’s faces. (Over50, yesOC, BCFH, unknown)</p> <p>R4: I think it’s quite important to mention that actually symptoms can be anything that you wouldn’t necessarily think were to do with your abdomen. [...] It’s important to realise these symptoms can be very vague and are not specific and yeah ‘something about be aware of</p> | <p>Emphasise symptoms and their similarity to IBS more and encourage people to consult their GP if they notice anything unusual. Make these sections stand out more.</p> | <p>Added statement “Note that often these symptoms are similar to symptoms of other diseases, such as Irritable Bowel Syndrome and might be easily confused. Therefore it is very important that you tell your GP that you have a family history cancer.”</p> <p>Added statement to “What would</p> |

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| <p>changes in your body'. (Over50, yesOC, noFH, unknown)</p> <p>R8: I know the symptoms are vague but maybe more emphasis can be put on for women to go and get checked out. (35to39, noOC, OCFH, unknown)</p> <p>R8:Until there's a screening there's not a lot that can be said. [...] It says you should contact your GP but I don't know if a lot of women would. It could maybe be highlighted a wee bit more. [...] Even if it was bold [...] or in italics or something [...] just to stand out a wee bit. (35to39, noOC, OCFH, unknown)</p> <p>R8: I just think the symptoms, that didn't really stand out on the website. Although everything was green and grey and black, everything was kept uniform and the same. I just think something like that needs to stand out a wee bit. (35to39, noOC, OCFH, unknown)</p> | | <p>happen if I do not have surgery?"</p> <p>question: "You know your body best, so don't be afraid to go to your GP if you notice anything unusual. Make sure..."</p> <p>Made symptoms box stand out more by adding colour and bold writing;</p> <p>Added link to "Key messages to the public" on ovarian cancer symptoms</p> |
| <p>R4: You could put something in there about how they [TV scans] are not as scary as they sound. [...] It's not as terrifying as you think it's going to be. The thought of it is worse than actually having it. (Over50, yesOC, noFH, unknown)</p> | <p>Add sentence about what diagnostic TV scans involve.</p> | <p>Not actioned, as we need to avoid confusion between diagnostic / screening tests and do not want to emphasise screening further. This is not directly relevant to the target population at the time of reading the decision aid. If they develop symptoms they will be consented for these diagnostic tests at which point these will be explained.</p> |
| <p>R11: I didn't know what a CA 125 was, so I could have done some [with] more questions and information about those tests. (Over50, yesOC, noFH, unknown)</p> <p>R11: The screening page is quite interesting and everything else is and</p> | <p>Add information about how the diagnostic CA125 test is done, what it stands for and what results mean.</p> | <p>Not actioned, as we need to avoid confusion between diagnostic / screening tests and do not want to emphasise screening further. This is</p> |

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| <p>the CA 125 blood tests [...] That might be quite nice to actually say what that is. (Over50, yesOC, noFH, unknown)</p> <p>R11: It would be nice to sort of say what the blood test - you don't need to go into scary figures - but it just might be quite nice to go into what it was. (Over50, yesOC, noFH, unknown)</p> | | <p>not directly relevant to the target population at the time of reading the decision aid. If they develop symptoms they will be consented for these diagnostic tests at which point these will be explained.</p> |
| <p>R11: I think the only question I've got is how soon could I have got listened to and got treatment? (Over50, yesOC, noFH, unknown)</p> | <p>Add timeline of CA125 / ultrasound diagnostic test to treatment if OC is found.</p> | <p>Not actioned, as we need to avoid confusion between diagnostic / screening tests and do not want to emphasise screening further. This is not directly relevant to the target population at the time of reading the decision aid. If they develop symptoms they will be consented for these diagnostic tests at which point these will be explained.</p> |
| <p>Value clarification exercise</p> | | |
| <p>R12: I wonder if it's worth saying somewhere at the beginning that you have that [VCE] to come at the end. [...] At the beginning of the website maybe, if there's somewhere you could almost just let people know that that's coming, they don't have to be writing this information down on a notepad [...] or equally if they don't want to read through it all because they've spoken to their GP about it in quite a lot of detail, [...] if they just wanted to skip through. (Under35, noOC, OCFH, unknown)</p> | <p>Add statement on home pages (after entering decision aid) that announces that VCE exists and add link</p> | <p>Added statement to home pages saying "This website includes pages with information about the decision, an exercise in which you can weigh different factors according to your preferences (Your Decision [LINK]) and a list of useful Contacts and Resources."</p> |
| <p>R12: It took me a couple of goes to figure out how to move the little</p> | <p>On website VCE, add instructions for</p> | <p>Added instructions saying "If you are</p> |

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| <p>circles around [...] that's because I was on an I-pad rather than a lap top. [...] I was expecting to be able to drag it across and I couldn't [...] You just had to click on the circle [...] and then put your finger just on the line where you want the circle to go and it jumps there (Under35, noOC, OCFH, unknown)</p> | <p>users of iPads that the slider function does not work and using the tool requires them to click on the slider and then click on the point in the scale they want the slider to jump to.</p> | <p>viewing this website using an iPad, simply touch the slider, then touch the point in the scale where you would like the slider to jump to."</p> |
| <p>R13: The vague one wasn't too bad, because I suppose somebody say may be able to answer that, but it's that one 'there's no effective screening for ovarian cancer' [...] I don't know how you could answer that with a points system, whether it would [make you] want [...] to have surgery. (Over50, yesOC, BCFH, unknown) R5: 'The symptoms of ovarian cancer are very vague'. I'm not quite sure why that would affect your decision, though I suppose it would in the fact that if there was a family history of it you're not that likely to pick it up from symptoms so therefore perhaps yeah maybe. (Over50, yesOC, noFH, unknown)</p> | <p>Reword / expand on statements in VCE to clarify their relevance to the decision</p> | <p>Expanded statements to clarify: "There is no effective screening for ovarian cancer, so I cannot get checked out regularly" and "The symptoms of ovarian cancer are very vague, so I may not notice them early."</p> |
| <p>R11: I think what's missing then would be, once you've made your decision what do you do? (Over50, yesOC, noFH, unknown)</p> | <p>Add information about what to do once decision is made.</p> | <p>Reworded question "How can I deal with my decision?" to "How can I take my decision forward and how can I cope?" Reworded and expanded answer to this question to "<u>If you are considering surgery</u>, the next step is to make an appointment with a health professional. This can be your GP, genetic counsellor or gynaecologic surgeon. With them, you can discuss surgery further and get answers to any</p> |

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| | | <p>additional questions, before booking an appointment with a hospital. You can also seek a second professional opinion at any time if you wish. Preparing yourself for having surgery, both mentally and physically, can help to cope with the operation and with recovery time. It can also help to... [...]. Before committing fully to surgery you can discuss any further questions with your doctor or genetic counsellor and</p> |
| <p>R4: I thought I would like to be able to kind of obtain a [...] booklet with all the information to pick up and put down as opposed to having to look at videos and PDF's which could be time consuming. I'd like to be able to I don't know click a button on things that I would want to have sent to me. (Over50, yesOC, noFH, unknown)</p> | <p>Create option for users to order printed copy of booklet.</p> | <p>Not actioned, as, unfortunately, this is not within the financial scope of the PhD and therefore we are unable to provide such a service at this time.</p> |

Appendix 8.7 - OvDex booklet (General version) after lay reviewer feedback and changesThe logo for OvDex, featuring the letters 'OvDex' in a stylized font. The 'O' and 'V' are teal, and 'Dex' is grey. The 'O' and 'V' are connected at the top.Oophorectomy
Decision ExplorerA large version of the OvDex logo, with 'OvDex' in teal and grey, separated by a horizontal line.

Oophorectomy Decision Explorer

A decision aid designed to help you
and your doctor make a decision
about risk-reducing ovarian surgery

For women at increased risk of ovarian cancer

OvDex V5 Jul 2013
©Cardiff University

Next update:
July 2014

OvDex (The Oophorectomy Decision Explorer) has been developed to help you find out more about your options for reducing your ovarian cancer risk. If you are viewing OvDex you should have been referred to it by a doctor or geneticist as you are at increased risk of ovarian cancer.

The information in OvDex can be tailored to your personal situation by answering three simple questions. We recommend that you use this function, so that the information displayed is more relevant to you. If you do wish to personalise the information you get, please go to the OvDex website at www.OvDex.co.uk and answer the questions .

The information the following pages has not been personalised. If you do not wish to personalise OvDex then please view the general information on the following pages.

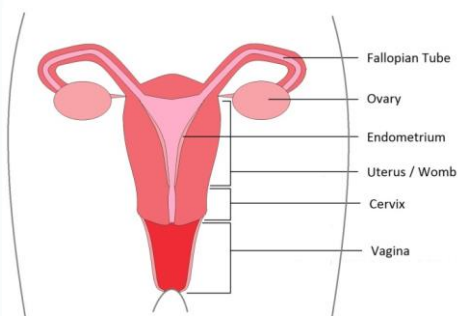
| Chapter | Page |
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1. Cancer risk

3

Questions in chapter 1:

- Q1 Where does ovarian cancer develop?
- Q2 What is the ovarian cancer risk if I have a family history of ovarian cancer?
- Q3 How can I find out whether I have a faulty gene?
- Q4 What would my risk be if a faulty gene was found?
- Q5 What if I cannot get tested or my genetic test is uninformative?
- Q6 Does my cancer risk change over time?
- Q7 How will my ovarian cancer risk affect my life?
- Q8 How will my ovarian cancer risk affect my children?
- Q9 How can I change how I feel about my cancer risk?

Q1 - Where does ovarian cancer develop?

This picture shows the female reproductive system. The ovaries are connected to the uterus (womb) by the fallopian tubes. Ovarian-type cancer can develop in the ovaries, the fallopian tubes or the lining of the abdomen (called the 'peritoneum'). Other female cancers, such as cancer of the endometrium (the lining of the uterus/womb) or cervix, are very different and should not be confused with ovarian cancer.

For more information about ovarian cancer check out the Cancer Research UK website at www.cancerresearchuk.org and other websites under Contacts & Resources.

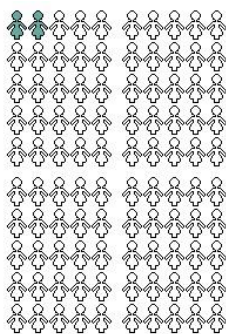
Q2 - What is the ovarian cancer risk if I have a family history of ovarian cancer?

Ovarian cancer occurs by chance in less than 2 of every 100 women. For women who have a family history of ovarian, breast and/or bowel cancer and/or a faulty gene, the risk can be much higher than 2 in 100. Generally the risk is quite low in young women and rises with age.

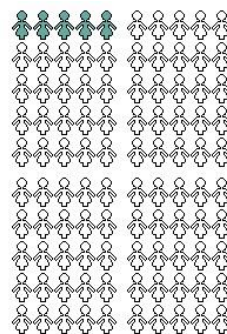
Your risk depends on how many of your relatives had cancer and on how closely related they are to you. 'First degree relatives' are those directly related to you, for example, mother, sister or daughter. 'Second degree relatives' are those who are less closely related, for example your aunt or your grandmother. The diagrams on the next page show how risk changes depending on the number of relatives with ovarian cancer.

1. Cancer risk

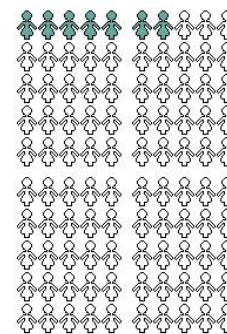
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Less than 2 out of 100 women in the general population get ovarian cancer in their lifetime. 98 women will not.



About 5 out of 100 women with a first degree relative with ovarian cancer get ovarian cancer in their lifetime. 95 women will not.



About 7 out of 100 women with two or more relatives with ovarian cancer get ovarian cancer in their lifetime. 93 women will not.

A woman's chances of developing ovarian cancer can also be increased if she has relatives with cancers other than ovarian cancer, such as breast and bowel cancer. A woman's risk is estimated by genetics services once the full family history is available.

Q3 - How can I find out whether I have a faulty gene?

If you come from a family where certain types of cancer (e.g. ovarian, breast, bowel) have affected more than just one or two relatives, you might have a faulty gene. To get tested for a faulty gene you will need to be referred to genetics services (usually by your GP). The genetics service will assess your family history in detail and you will be told whether there might be a faulty gene involved.

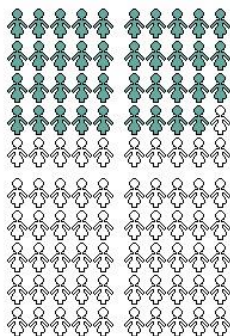
Within the NHS, testing for a faulty gene in the family usually begins with a test on a blood sample from a person who has had cancer. If you have had cancer yourself, you may be offered a genetic test as the first person in your family. If you have not had cancer yourself, a relative who has had cancer will be offered a genetic test first. For the test, a blood sample will be taken and sent to a laboratory for analysis. Test results then usually come back within 2 to 3 months. For more information about what the test involves see Contacts and Resources.

Results of this test may show that you either have a faulty gene that predisposes you to cancer (in this case, the gene test is said to be 'positive') or that you do not have a faulty gene that predisposes you to cancer (in this case, the gene test is said to be 'negative'). In some cases, test results may show that you have a 'variant of unknown significance', which means the test detected a change in your gene, but it is not known whether this change predisposes you to cancer or whether it is harmless. If a faulty gene is found, other members of the family can then have a genetic test to see whether they also have the same faulty gene.

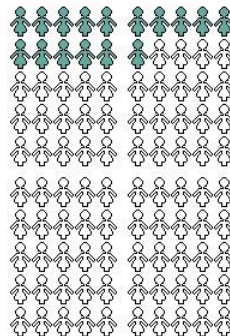
If there is no living relative with cancer, or the relative doesn't want to get tested, it may still be possible to do a genetic test for you on the NHS, if your genetic counsellor thinks that your chances of carrying a faulty gene are higher than 10 in 100 based on your family history. For anyone who cannot get tested on the NHS, there are some companies offering private testing. However, this can be very expensive and without a strong family history, there may be no need for testing.

Q4 - What would my risk be if a faulty gene was found?

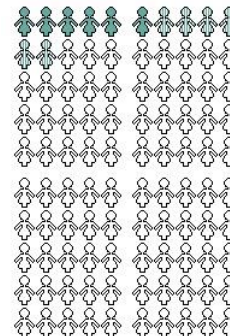
The most common faulty genes linked to ovarian cancer are called Breast Cancer (BRCA) genes 1 and 2 and genes common in Lynch Syndrome (formerly known as Hereditary Non-Polyposis Colorectal Cancer or HNPCC). The lifetime risk of developing ovarian cancer can be much higher for women who have a faulty gene compared to the general population.



About 39 out of 100 women with a BRCA1 faulty gene get ovarian cancer by the time they are 70. About 61 women with a BRCA1 faulty gene will not.



About 16 out of 100 women with a BRCA2 faulty gene get ovarian cancer by the time they are 70. About 84 women with a BRCA2 faulty gene will not.



About 6 to 12 out of 100 women with Lynch Syndrome get ovarian cancer by the time they are 70. About 88-94 women with Lynch Syndrome will not.

Q5 - What if I cannot get tested or my genetic test is uninformative?

For some women it might not be possible to get tested because they have no living relative with cancer or their relative does not want to be tested. In other cases a genetic test may come back as 'uninformative' which means a gene variant of uncertain significance has been found, but it is not clear whether this variant increases the risk of cancer or not. In these cases the genetics service will estimate the woman's risk based on her family history.

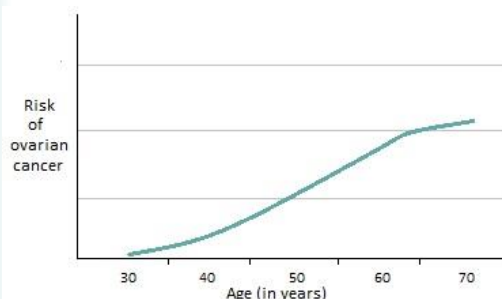
If you have been told that your family history suggests you might have a faulty gene and you would like more specific information, you can personalise this aid by selecting the gene that most closely resembles your family history according to your genetic counsellor. For example, if you have been told your family history suggests you might have a BRCA1 gene, personalise OvDex by going to the website and answering the first question with "Yes, I have a faulty BRCA1 gene".

Q6 - Does my cancer risk change over time?

Yes. Your cancer risk increases with age. While your so-called 'lifetime' risk (which is the risk that you will get ovarian cancer at some point in your life) will stay the same over time, your 'age-related' ovarian cancer risk (which is the risk that you will get ovarian cancer in a specified time frame, for example within the next 5 years) increases with age.

1. Cancer risk

6



This graph shows how cancer risk rises over time.

Q7 - How will my ovarian cancer risk affect my life?

Being at risk for ovarian cancer does not necessarily mean that you will develop ovarian cancer. However, knowing your risk will give you access to additional medical services such as genetic counselling and means that you may be offered risk-reducing surgery. Some people may feel worried or anxious after finding out about their risk and their quality of life may be affected.

Q8 - How will my ovarian cancer risk affect my children?

There is a chance that your children will also have an increased risk of cancer. Their risk depends on how high your own risk is and on whether or not you have a faulty gene. If you have a faulty gene, then your children have a 50/50 chance to inherit this. If a faulty gene has been confirmed in the family, children can choose to get tested for this gene at any time. The best time to get tested should be discussed with your genetic counsellor. If your child/children inherited the faulty gene, then their cancer risk would be the same as yours. If they did not inherit the faulty gene, then their cancer risk would be low. Boys who inherit the faulty gene are not at risk of ovarian cancer, but may be at risk for other cancers. Boys who inherit the faulty gene may also pass it on to their children.

Please also note that cancer risk increases with age and therefore children's risk only starts to rise in later life. More information about genetics can be found on the NHS choices website (see Contacts and Resources).

You can discuss your children's risk with your genetic counsellor to find out more. Your genetic counsellor will also be able to advise you about how best to discuss this risk with your children.

Q9 - How can I change how I feel about my cancer risk?

If you would like to find out more about your risk and what it means, you can use the resources provided in OvDex and talk to your doctor or genetic counsellor.

Knowing that you are at increased risk of cancer might make you feel worried or angry or frustrated. This is completely normal and there is no need to bottle those feelings up. It can actually help to talk about them.

You can try to find someone you trust to talk to or if you prefer to talk to someone you don't know, you can use the contact details for charities and patient support networks under Contacts & Resources. If you have a partner, sharing your thoughts and concerns with your partner can be useful in helping them understand what you are going through and in helping you cope together as a couple.

It may also help to see a positive side to knowing your risk status. For example, this means that you will get access to genetics services and have the chance to do something about your risk. Other women have found it helpful to stay optimistic, have joined patient networks or have found strength in their faith.

Being at risk of cancer may sometimes lead to unwanted thoughts and worry, especially at times when there are no active steps you can take, for example when you are waiting for genetic test results. There are useful techniques you can use to reduce such thoughts. One such method is active distraction, which means actively thinking about something else whenever unwanted thoughts pop in your head. Watch the 'How do I cope' video on the Cancer Genetics Storybank website for a guide of how to do this (see Contacts & Resources).

Relaxation techniques, meditation and guided imagery may also help you deal with any stress you might experience relating to your cancer risk. Examples include progressive muscle relaxation and certain breathing exercises. Visit the 'Mind-body therapy' section on the Macmillan website for more information on and guides to mind-body therapies (see Contacts & Resources).

Questions in chapter 2:

- Q1 What can I do to reduce my risk?
- Q2 Why is there a choice?
- Q3 Who should decide?
- Q4 Who else should I involve in this decision?
- Q5 When should I decide?
- Q6 How can I decide?
- Q7 How can I deal with the choice?

Q1 - What can I do to reduce my risk?

Your doctor has probably discussed with you the option of having risk-reducing ovarian surgery to reduce your ovarian cancer risk. You will need to decide whether and when to have this surgery. Unfortunately, there is no medically proven screening available on the NHS for ovarian cancer at the moment.

This decision aid is designed to help you look at your options. Please refer to chapter 3 (Options at a glance), chapter 4 (Risk-reducing surgery) and chapter 7 (Other options) for more information about surgery, screening and other alternatives.

Q2 - Why is there a choice?

Often when you go to your doctor, there is one clearly recommended treatment. However, some situations are more complicated. These are situations in which your personal preferences and feelings play an important role. In the case of cancer risk reduction, your options have very different effects on your life. This means that you need to be clear about what might happen if you choose one option over the other and how that would impact on your life.

Some health professionals may recommend surgery quite strongly; however you need to make the final decision and before you do, you should consider the possible benefits and risks, how these might affect your life and how you feel about them.

Q3 - Who should decide?

As the best choice for you is based on your preferences, you should be closely involved in the decision. You can either make the decision on your own or if you do not wish to make this decision yourself, your genetic counsellor or gynaecologist can help you. They will encourage you to think about the options and your preferences, so that the final choice is right for you. If you then don't want to choose for yourself, just say so and they may make a recommendation.

Q4 - Who else should I involve in this decision?

Whether or not you would like to bring anyone else into this decision is your choice. Often it is helpful to speak to someone who knows you well, such as a partner, other members of your family or a friend, who could work through this decision with you. If you are in a relationship the views of your partner can be important, especially in the context of risk-reducing ovarian surgery, so it is recommended that you speak to your partner and try to reach a decision together. Your doctor will also be happy for you to bring your partner along to appointments and to answer any questions they might have.

Q5 - When should I decide?

The operation is most effective if it is done at 40 years of age, because ovarian cancer risk starts to rise from age 40 onwards. Additionally, if done at 40, the surgery reduces the risk of breast cancer. However, age 40 may not be ideal for everyone. So, even if you decide not to have it at 40, the operation will still decrease your ovarian cancer risk if performed after that age.

The decision you are facing is not an easy one and you should not feel under any pressure to decide quickly. Risk-reducing surgery has benefits and risks that need to be weighed carefully, so take your time and make sure you are ready before making a decision.

It is important that you understand that undergoing surgery to remove your ovaries will mean you can no longer get pregnant. Therefore it is essential to consider your plans for a family and any potential future changes to these plans (for example if there is a chance you might meet a new partner / re-marry) before making a final decision.

Q6 - How can I decide?

When it comes to important decisions everyone is different. Some people like to find out as much as they can about their options, while others prefer to just know what is absolutely necessary. Some might find it helpful to talk to their family and friends. Some might like to speak to people who have made a similar decision. It really depends on you. Have a think about other important decisions in your life and how you managed to make those. That could give you an idea of how you like to decide about things.

Q7 - How can I deal with the choice?

It can be helpful to create a plan of how and when you will make this choice. If you are not ready to decide right now, it might be useful to set yourself a deadline of when you will revisit this decision. For example: "Just after my 40th birthday I will look at this information again." or "Once I have completed my family I will revisit this decision."

Once you are ready you can decide how you want to make this choice:

1. I will decide by myself using everything I have learnt
2. I will decide but seriously consider my doctor's opinion
3. The doctor and I should decide together
4. The doctor should decide but seriously consider my opinion
5. The doctor should decide for me

One constructive way to deal with a difficult decision is to empower yourself with information. OvDex is designed to help you to learn more about ovarian cancer risk and your options. With the wealth of information that is available on the internet, it can be difficult to find reliable and trustworthy information. The information in OvDex is supported by recent scientific findings and has been carefully reviewed by health professionals to make sure it is accurate. You should at least understand your options and their benefits and risks before making a decision. Find out more about the most important questions to ask at: www.Ask3Questions.co.uk. Once you have read the information in OvDex, it could help to make a note of any remaining questions and take those to your doctor or genetic counsellor for a more detailed discussion.

You may feel that you are not comfortable making decisions about your health. This is okay. You do not need to make the choice alone if you don't want to. You can decide together with your doctor or ask them to make the choice for you. But you need to remember that you are the expert when it comes to your own life and that only you know what is important to you. So even if you decide to let the doctor make the decision for you, make sure they know about your goals and values. Tell them what is important to you.

3. Options at a Glance

11

| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|---|---|--|
| Will this reduce my risk of ovarian cancer? | Yes, removing the ovaries and fallopian tubes will greatly reduce your lifetime risk of ovarian-type cancer to that of the general population, which is about 2 in 100. | No, your risk of ovarian cancer will remain increased. |
| Is there a routine screening programme to detect ovarian cancer? | No, there is no evidence that screening for ovarian cancer is effective and after surgery there is no need for screening as your risk will be low. | No, there is no evidence that screening for ovarian cancer is effective in saving lives. |
| Will I be able to become pregnant? | No, therefore it is important that you feel you have completed your family before having surgery. | Yes, your ability to become pregnant will be unaffected. |
| Will this change how I feel about my risk of ovarian cancer? | You may feel less worried about developing ovarian cancer. Most women report feeling satisfied with their decision after surgery. | You may worry about developing ovarian cancer. |
| Will this change how I feel about myself as a woman? | Many women do not notice any change in how they feel about themselves as a woman. However, your desire for and/or enjoyment of sex is likely to decrease. | Not applicable. |
| Will I go into menopause? | Yes, after surgery your oestrogen levels fall and the menopause will start immediately. | Yes, you will go through a natural menopause when you are around 50 years. |
| What is menopause like? | Menopause after surgery is similar to natural menopause, but because it happens suddenly, the symptoms may be more severe. | Natural menopause is a gradual decline of oestrogen levels. Typical symptoms include hot flushes, night sweats, mood swings and vaginal dryness. |
| Are there any long-term health risks linked to surgery? | Yes, if surgery is done before age 45 there is a higher risk of bone thinning and cardiovascular disease. Some patients report memory changes as well. | Not applicable. |
| Will I need hormone replacement therapy (HRT)? | HRT can reduce some of the effects of surgery and is recommended for women who have surgery before age 45. Discuss this with your clinician before your surgery. | No, if you go through a natural menopause you would not be expected to need HRT, unless your symptoms are very severe. |

3. Options at a Glance

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| Frequently asked questions | Surgery to remove ovaries and fallopian tubes | No surgery |
|---|---|--|
| Is HRT safe for women at increased familial risk? | HRT is safe to take for the majority of women until they are 50. Discuss this with your clinician before your surgery. It is not usually recommended for women who have had breast cancer. | HRT is safe to take for the majority of women until they are 50. Discuss this with your clinician. It is not usually recommended for women who have had breast cancer. |
| What are the risks of surgery? | About 4 in every 100 patients experience a complication during or after surgery. These can be minor infections or major complications. Discuss this with your clinician. | Not applicable. |
| How long will it take me to recover from surgery? | Most women leave the hospital the same day or the day after key hole surgery and are back to normal in 4 weeks. For open surgery time in hospital and recovery will be longer. During recovery you may feel pain / tiredness and may need help with everyday tasks. | Not applicable. |

Q1 – How can I cope with this decision?

Some women may find it useful to discuss surgery with others, who have already made this decision. In some areas your genetic counsellor may be able to put you in touch with someone who has already gone through the decision, otherwise you can use the contact details for charities and patient support networks under Contacts & Resources to read about or get in touch with others in a similar situation. Going over the decision with someone who knows you well, such as your partner or a member of your family can also help you clarify your decision. If you decide to opt for surgery and you are in a relationship, preparing yourself together with your partner for the time after surgery can help you both deal better with any consequences of the operation.

Being clear about the reasons why surgery could be the right or wrong thing for you can help you make a decision and avoid regretting it later. The information in OvDex is designed to help you identify the benefits and the risks of surgery and any other options, so you can decide for yourself. The exercise called 'Your Decision' can help you clarify what is most important to you.

There may be times when there are no active steps you can take, for example if you have decided to defer surgery or while you are waiting for your surgery appointment. During these times you may sometimes worry about your risk or experience unwanted thoughts. Useful techniques to reduce such worries and thoughts, especially at times when you cannot do anything but wait, include active distraction, relaxation techniques, meditation and guided imagery. Check out the 'How do I cope' video on the Cancer Genetics Storybank website and the Macmillan website for more information on these techniques (see Contacts & Resources).

Questions in chapter 4:

- Q1 What is risk-reducing bilateral salpingo-oophorectomy (RRSO)?
- Q2 What are the main advantages of this operation?
- Q3 What are the main disadvantages of this operation?
- Q4 What is my risk after surgery?
- Q5 What does the surgery involve?
- Q6 How long does it take to recover?
- Q7 Do I need to have my womb removed as well?
- Q8 What are the complications of RRSO?
- Q9 Could cancer be found during the surgery?
- Q10 How would RRSO affect my life?

Q1 - What is risk-reducing bilateral salpingo-oophorectomy (RRSO)?

Risk-reducing bilateral salpingo-oophorectomy (RRSO for short) is an operation to remove healthy ovaries and fallopian tubes to reduce the cancer risk. The word 'salpingo-oophorectomy' means surgical removal of the fallopian tubes (salpinges) and ovaries. The term 'bilateral' in this context describes the fact that the ovaries and fallopian tubes on both sides of the body are removed. The aim of the operation is to remove these tissues before cancer develops.

Q2 - What are the main advantages of this operation?

- 1) This operation will reduce your ovarian and fallopian tube cancer risk, as these are completely removed during surgery.
- 2) For some women, not having to worry about ovarian cancer anymore comes as a great relief.
- 3) For the vast majority of women having RRSO before the natural menopause could also reduce the risk of breast cancer.
- 4) Having the surgery will also prevent other, non-cancer related problems happening with the ovaries, such as cysts, which can cause some discomfort and pain.

Q3 - What are the main disadvantages of this operation?

- 1) You will no longer be able to become pregnant once you have had surgery, therefore it is important that you have considered your plans for a family before making a final decision.
- 2) After the operation you will immediately enter the menopause and may start to experience the typical menopausal symptoms, including hot flushes, mood swings, night sweats and vaginal dryness (Please see Chapter 5 “The Menopause” for more details). Furthermore, you may be at increased risk of bone thinning, cardiovascular (heart and blood vessel) disease and memory changes. These effects may be effectively treated with hormone replacement therapy (Please see chapter 6 “Hormone Replacement” for more details), however not all women can take hormone replacement and this should be discussed before having surgery.
- 3) Your desire for sex is likely to decrease and due to some menopausal symptoms, such as vaginal dryness, you may enjoy sex less as intercourse can be painful. Again, hormone replacement may counteract these effects.
- 4) There is a small risk of complications during and after surgery.

Q4 - What is my risk after surgery?

After surgery your risk of ovarian and fallopian tube cancer is greatly reduced, however, there is a rare type of cancer which may still develop after surgery that is very similar to ovarian cancer. This is called primary peritoneal cancer and develops in the lining of the abdomen and pelvis. Unfortunately, the peritoneum cannot routinely be removed during RRSO. Your risk of primary peritoneal cancer depends on whether or not you have a faulty gene. The highest risk for primary peritoneal cancer is in women with a BRCA1 faulty gene. Their risk is about 2 in every 100 women, which is similar to the risk of ovarian cancer in the general population. For women who do not have a BRCA1 faulty gene the risk of primary peritoneal cancer after surgery is much lower than 2 in 100.

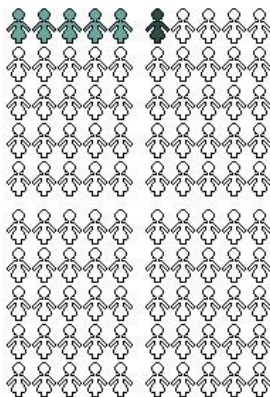
Q5 - What does the surgery involve?

Most often the operation is done as keyhole surgery (laparoscopy). This involves 3 or 4 small (about 1cm) cuts, usually one cut is close to your belly button and 2-3 just below the bikini line, so they are not visible. Keyhole surgery itself usually takes less than 2 hours.

In some cases it may not be possible to do keyhole surgery, because of previous surgery on your tummy or because of your weight. Then surgeons use the more traditional open surgery. This means a longer cut, usually along the bikini line. Sometimes a surgeon might have to convert to an open operation when doing keyhole surgery because of complications or old scar tissue. This happens in about 1 of every 100 keyhole operations.

4. Risk-reducing Surgery

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Type of procedure in 100 women who have the operation

The figure on the left shows **how many of 100 women will have keyhole surgery, open surgery** and planned keyhole which is converted to open surgery.

94 of 100 women will have planned keyhole surgery (white figures). Five of 100 women will have planned open surgery (light teal figures). One of 100 women will have planned keyhole surgery which is converted to open surgery (dark figure).

Q6 - How long does it take to recover?

75 of 100 patients who have keyhole surgery leave the hospital the day after surgery. They are usually back to normal activity about 4 weeks after surgery. If you have open surgery you are likely to stay in hospital a bit longer. Usually patients leave the hospital about 5 days after open surgery and are back to normal in about 6 weeks. After surgery you are not allowed to do any heavy lifting for a few weeks. You may also have to refrain from driving until you can comfortably wear a seatbelt and make an emergency stop without pain.

Q7 - Do I need to have my womb removed as well?

No, normally it is not necessary that you have your womb removed at the same time as having RRSO, unless you know that you have Lynch Syndrome or you have other problems with your womb and have been told by a doctor that removing the womb would help you with those problems. However, you may choose to have your womb removed at the time of RRSO, as it may affect the type of hormone replacement you would be given. You should discuss this with your doctor.

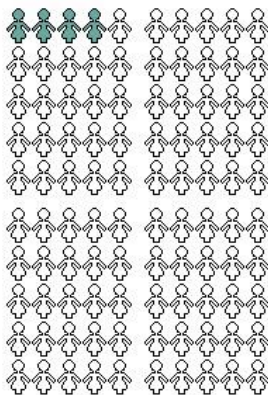
Q8 - What are the complications of RRSO?

There is a small risk of complications linked to RRSO. About 4 in every 100 people will experience some complication. Minor complications can include wound or urinary tract infections and usually have no long-term effects on your health. More serious complications might happen during surgery and can include damage to blood vessels, the bowel or the bladder. If you are having keyhole surgery this might mean that the surgeon has to convert to open surgery to repair the damage. There are a number of other rare complications that might happen and your surgeon will go through those with you if you wish before you go in for surgery.

Please note that about 96 of every 100 women do not experience any complications at all.

4. Risk-reducing Surgery

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**Complications in 100 women who have the operation**

The figure on the left shows **how many of 100 women will experience complications** during or after surgery.

4 of 100 women will experience some kind of complication either during or after surgery. 96 of 100 women will not experience any complications.

As with any surgery, there is a very small risk of death. However, this is highly unlikely. The risk may be greater in women with health problems before surgery.

Q9 - Could cancer be found during the surgery?

Yes, although this is rare. Once your ovaries and fallopian tubes have been removed they will be checked thoroughly for any signs of cancer. There is a chance that cancer may be discovered during this check. This happens in up to 4 out of every 100 operations in the highest risk patients. For others the chances of finding cancer during the surgery are much lower.

If cancer is found during the surgery, you will be informed as soon as possible and might have to undergo further tests, treatment and surveillance. If cancer is found you might be unable to take HRT after the operation.

Q10 - How would RRSO affect my life?

In the short-term, if you decide to go for surgery, you will have to take time off work and will not be able to do some of the things you might usually do, such as driving or heavy lifting. Therefore you may need help with everyday tasks. During recovery you may also feel some pain and discomfort and you may be more tired than usual. However most women recover fully within 4 to 6 weeks.

In the longer term there are a number of things you should consider. After surgery you may feel less worried about ovarian cancer. However you may feel differently about your body and you may lose interest in sex. You will also no longer be able to become pregnant and you will enter the menopause if you have not gone through it yet (Please see chapter 5 "The menopause"). You may opt to take hormone replacement (Please see chapter 6 "Hormone replacement"). These factors may affect your life after surgery and should be considered carefully before making a decision.

5. The Menopause

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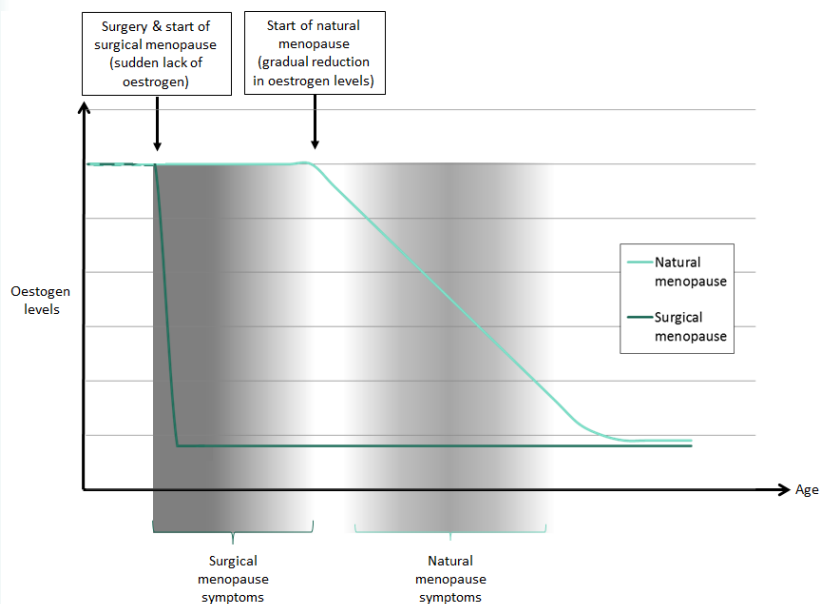
Note that the information about symptoms of the menopause in this chapter describes symptoms experienced by women who do not take hormone replacement therapy (HRT), unless stated otherwise. Most symptoms of the menopause are reduced by hormone replacement therapy (for more information see Chapter 6 “Hormone Replacement”.

Questions in chapter 5:

- Q1 What is a surgical menopause?
- Q2 What is the menopause like?
- Q3 How could the menopause affect my life in the short term?
- Q4 How long does surgical menopause last?
- Q5 Are there any long term health effects linked to surgical menopause?
- Q6 Is there anything I can do to reduce the effects of surgical menopause?

Q1 - What is a surgical menopause?

A surgical menopause is a menopause you enter when your ovaries are removed by surgery and your body no longer produces the female hormone oestrogen. A surgical menopause is essentially the same as a natural menopause, it only happens earlier and more suddenly. Therefore a surgical menopause results in the same symptoms as a natural menopause. These symptoms are caused by the lack of oestrogen. As oestrogen levels drop suddenly after surgery, the symptoms of a surgical menopause start instantly and can be more severe than those of a natural menopause, in which oestrogen levels fall gradually.



The graph above shows the drop in the levels of the female hormone oestrogen during surgical and natural menopause. Potential menopausal symptom duration and severity are indicated in shades of grey.

5. The Menopause

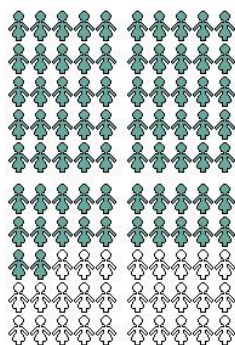
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Q2 - What is the menopause like?

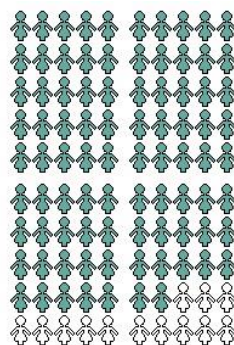
It is very difficult to describe what the menopause is like, because it is different for every person. Some women have no or very few symptoms and cope very well. Other women can have very severe symptoms and their quality of life can be affected.

Common menopausal symptoms you may experience are: Hot flushes, night sweats, mood swings, vaginal dryness and loss of interest in sex. Other symptoms include difficulty sleeping, fatigue, weight gain, changes in memory and depression.

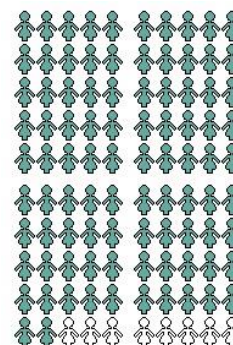
The great majority of women who had surgery reported that they experienced one or more of the common symptoms. However, any data available about menopausal symptoms is from small studies and each study uses different ways to assess what women experienced, so it is difficult to predict what the menopause will be like for you personally. The pictures below show how common different types of menopausal symptoms are according to one study of women after RRSO.



About 72 of 100 women experience vasomotor symptoms (hot flushes and night sweats). 28 women do not.



About 87 of 100 women experience physical and sexual symptoms (aching, vaginal dryness, loss of libido). 13 women do not.



About 92 of 100 women experience psychosocial symptoms (mood swings, feeling depressed or down). 8 women do not.

Q3 - How could menopause affect my life in the short term?

While none of the menopausal symptoms mentioned above are dangerous for your health, they may affect you in different ways. Hot flushes can be very uncomfortable and may happen at inconvenient times, for example when you are giving a presentation at work. In one study two out of every three women reported having hot flushes after surgery. Night sweats can lead to problems with sleep and insomnia.

Vaginal dryness can lead to pain during sex and therefore you may enjoy sex less. In one study, one in every three patients felt that vaginal dryness was bothersome and reported pain with sex. Additionally, many women also experience a loss of interest in sex after surgery. In one study just over half of women reported that they were not satisfied with their sexual functioning after having surgery. In that study, the decrease in sexual functioning seemed to persist even when women were taking hormone replacement. If you are in a relationship, these factors may affect your relationship with your partner. The way that these symptoms might affect your life depends on your personal situation.

There are several options available to help you deal with the short-term symptoms of the menopause. These include physical options, such as lubricant to counteract vaginal dryness, or psychological options, such as sex counselling and cognitive behavioural therapy to improve emotional and physical functioning. You can get more information about these options from your doctor or gynaecologist. The NHS choices website also has information about dealing with the menopause (See Chapter 9 “Contacts & Resources”).

Q4 - How long does the menopause last?

It is impossible to say how long symptoms will last. Some women have hardly any problems and symptoms disappear quickly, while others have symptoms for many years. It really is different for everyone.

Q5 - Are there any long term health effects linked to the menopause?

Yes. The loss of oestrogen is thought to affect long term health, particularly the bones and the cardiovascular system (heart and blood vessels). In older age this can lead to a higher risk of fractures due to bone thinning (osteoporosis) and a higher risk of cardiovascular disease (heart disease, stroke).

It should be noted that these effects are seen in all women after the menopause, whether it was a surgical or natural menopause. Your personal risk depends on your lifestyle and personal and family history of osteoporosis and cardiovascular disease and should be discussed with your doctor.

Due to the sudden lack of oestrogen after surgery, the risk of bone thinning and cardiovascular disease may start to rise immediately after surgery. This could be before the woman has reached the age of the natural menopause. Therefore, women who undergo surgery before the age of 45 are usually recommended to take hormone replacement to reduce these effects until they reach the age of the natural menopause (See Chapter 6 “Hormone replacement”).

Some women also report changes in memory following the menopause, which is again due to the lack of oestrogen.

Q6 - Is there anything I can do to reduce the effects of the menopause?

Yes. Hormone replacement therapy (HRT) is usually recommended for women who have RRSO before the age of 45 and have not had breast cancer themselves (See Chapter 6 (Hormone replacement) page 19). HRT may effectively reduce some of the short- and long-term effects of surgical menopause. However, not all women can take hormone replacement and this should be discussed before having surgery.

If you do not wish to or cannot take HRT then you can take dietary supplements to reduce bone thinning. You should speak to your doctor about this. In some regions in the UK patients are also recommended to have a bone density scan about 18 months after their operation. You can discuss this with your doctor.

An active lifestyle and healthy diet is also recommended.

6. Hormone Replacement

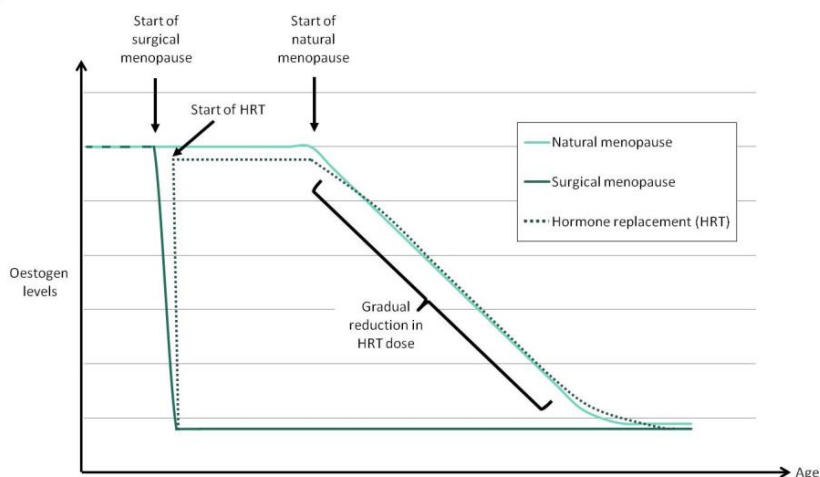
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Questions in chapter 6:

- Q1 What is Hormone Replacement Therapy (HRT)?
- Q2 Why is HRT important?
- Q3 Who should take HRT?
- Q4 How long should I take HRT?
- Q5 Can I take HRT if I have a family history of breast cancer?
- Q6 But I have heard that HRT increases breast cancer risk
- Q7 Will HRT reduce all the symptoms of menopause?
- Q8 Will HRT deal with the long term health effects of the menopause?
- Q9 Are there any alternatives to HRT?

Q1 - What is Hormone Replacement Therapy (HRT)?

Hormone Replacement Therapy, or HRT for short, is a medical treatment that puts back the female hormones that are lost when the ovaries are removed or when they stop functioning.



The graph above shows the oestrogen levels after surgical and natural menopause. It also shows the effect of HRT on the oestrogen levels after the operation if surgery was performed before the natural menopause.

There are a number of ways that HRT can be used:

- It can be taken as oral tablets usually once a day
- It can be applied as patches that are put on the tummy or bottom about once or twice a week
- It can be applied as a gel directly to the skin once a day (for example the lower abdomen or inner thigh)

There are two types of hormone replacement, one includes oestrogen and progesterone and the other includes oestrogen only. Women who only have their ovaries removed will usually be given oestrogen and progesterone HRT, while women who have their womb/uterus and ovaries removed (RRSO plus hysterectomy) are usually given oestrogen only HRT.

Q2 - Why is HRT important?

There are two main reasons why HRT is recommended after surgery:

1. To reduce the risk of bone thinning
2. To reduce symptoms of the menopause

Q3 - Who should take HRT?

HRT is strongly recommended for women who have surgery before they are 45 years of age, as this group is most affected by the long term health issues that are linked to loss of oestrogen, especially bone thinning (osteoporosis).

Women who have surgery when they are over 45 years of age can choose to have HRT to reduce the symptoms of surgical menopause. However these women do not have to have HRT if they do not want to, as the long term health effects of early loss of oestrogen do not affect them as much as a women having surgery before 45, because their oestrogen levels would have started to fall anyway.

It is important that the possibility of HRT is discussed before making a decision about surgery.

Q4 - How long should I take HRT?

If you use HRT, it is recommended that you take it until the age of the natural menopause, which is 50 years in the UK. From the age of 50 your oestrogen levels would have declined naturally, so taking HRT for many years beyond 50 is not recommended. This is because there is controversial evidence about the benefits and risks of HRT if it is taken beyond the age of natural menopause. Therefore the health effects are unclear and HRT may cause more harm than good if taken beyond 50.

Q5 - Can I take HRT if I have a family history of breast cancer?

Yes. You should be able to take HRT even if you have a family history of breast cancer. Several studies have shown that HRT is safe to take for the vast majority of women with a family history of breast cancer as long as they have not had breast cancer themselves and they only use HRT until the age of the natural menopause. You should discuss your family history of breast cancer with your genetics service before deciding to have surgery.

Q6 - But I have heard that HRT increases breast cancer risk?

Yes, HRT contains oestrogen and there is some evidence that higher amounts of oestrogen in the body can increase the risk of breast cancer.

However, the study that showed that HRT increases breast cancer risk was done with older women who had already gone through the natural menopause and were taking HRT beyond 50 years of age. Therefore these women were taking oestrogen when their body has naturally stopped producing this hormone.

Furthermore, these women did not have surgery to have their ovaries removed. Therefore this group of women is very different to women with a family history who choose to have their ovaries out before they are 50 years. Furthermore you should be aware that the findings of this study have now been widely criticised.

Having your ovaries out removes all the natural oestrogen that your ovaries would be producing until the age of the natural menopause. The amount of oestrogen that is added back by taking HRT is less than the amount your ovaries would have produced naturally. If you stop taking HRT at the age of the natural menopause (so when you are 50) then there is no evidence that the breast cancer risk is increased.

Q7 - Will HRT reduce all the symptoms of menopause?

No. Taking HRT is not the same as having your own hormones. HRT cannot reduce all symptoms of the menopause and women who have had surgery and are on HRT do report more symptoms than women who have not had surgery. However, HRT can relieve some of the symptoms of the menopause. For example, women who took HRT reduced the average number of hot flushes from about 4 to about 1 per day when compared to women who did not take HRT. Women on HRT also had fewer night sweats.

Q8 - Will HRT deal with the long term health effects of the menopause?

Yes. HRT will reduce the risk of bone thinning and changes in memory functioning linked to the loss of oestrogen. However, the evidence of HRT's effects on cardiovascular disease (heart disease and stroke) is more controversial and should be discussed with your doctor in terms of your personal and family history of cardiovascular disease as well as your lifestyle, which can also affect your risk of cardiovascular disease.

Q9 - Are there any alternatives to HRT?

Yes. There are numerous non-hormonal alternatives to HRT, none of which have been shown to be as effective in relieving menopausal symptoms. For people who cannot take HRT these alternatives may be a good option but for those who can, HRT is the best option.

Alternatives that are frequently used include:

- Antidepressants, such as Selective Serotonin Reuptake Inhibitors (SSRIs)
- Gabapentin
- Red Clover
- Ginseng
- Evening primrose oil
- Agnus Castus
- And many more (please note that this list is not exhaustive and there may be other alternatives to HRT)

Some of these options may be prescribed by your doctor, while others can be bought in health shops or over the internet. However, there is no medical proof that freely available herbal options work. These alternatives to HRT may have side-effects and you should always consult your doctor before deciding to use any of these options.

Questions in chapter 7:

- Q1 Is there any screening available for ovarian cancer?
 Q2 Are there any other options apart from surgery?
 Q3 But I have been offered CA125 blood tests and/or trans-vaginal scans?
 Q4 What would happen if I do not have surgery?
 Q5 Is there anything else I can do?

Q1 - Is there any screening available for ovarian cancer?

No. Unfortunately there is no medically proven screening available for ovarian cancer.

Two large trials of ovarian cancer screening were done recently and the results are not yet available, so we do not yet know whether this screening is effective and can detect ovarian cancer early. The UKFOCS trial offered high risk women CA125 blood tests every four months and one yearly trans-vaginal ultrasound scan. The UKCTOCS trial offered such screening to women over 50, who were not at high risk. Both trials have not reported their final outcomes. Routine screening will not be offered until the results of these trials are available and only if these trials show that screening is effective and can save lives.

Some areas or GPs may offer private, self-funded CA125 and/or trans-vaginal screening to high risk women, however women need to be aware that there is still no proof that this screening is effective in detecting ovarian cancer early. Therefore going to private screening may provide false reassurance. Additionally, screening may result in unnecessary worry and anxiety if any results show changes that turn out not to be cancer.

You should also be aware that the yearly smear test you receive is designed to detect cervical cancer and will not detect ovarian cancer.

Women from families with Lynch syndrome may receive trans-vaginal ultrasound screening to look for womb cancer. The ovaries may be visible on these scans and if something is found the GP may order a CA125 blood test. In this case the CA125 blood test is a diagnostic test and not a screening test.

Any woman with symptoms of ovarian cancer is likely to be offered a diagnostic CA125 test and trans-vaginal ultrasound scan. Please note that these diagnostic tests are not screening tests. Rather, they are designed to detect ovarian cancer when symptoms have already developed. Women with no symptoms will not be offered routine screening on the NHS until the screening has been shown to be effective.

Q2 – Are there any other options apart from surgery?

At the moment the official alternative to surgery is to do nothing and simply be aware of the symptoms of ovarian cancer should they develop. However it is important to realise that these symptoms can be very vague and are not specific to ovarian cancer. If any of these symptoms happen more than 12 times a month you should contact your GP.

7. Other Options

24

The symptoms of ovarian cancer include:

Persistent bloating (big or swollen tummy)
Feeling less hungry or feeling full quickly
Persistent pain in your tummy or below
Needing to urinate more than usual

Often these symptoms are similar to symptoms of other diseases, such as Irritable Bowel Syndrome and might be easily confused. Therefore it is very important that you tell your GP that you have a family history of cancer.

Q3 – But I have been offered CA125 blood tests and/or trans-vaginal scans?

Some centres and/or GPs may offer private CA125 tests or trans-vaginal scans to women at high risk of ovarian cancer. As these are not offered as screening tests by the NHS you may have to pay for these yourself. You can choose to have these tests done, but you should be aware that there is no evidence that these screening tests are effective at detecting ovarian cancer early. Therefore using such screening services might provide you with false reassurance.

Women from Lynch syndrome families may be offered hysteroscopies or trans-vaginal ultrasound scans to detect uterus/womb cancer, which these women are at higher risk for. However usually these tests are designed to detect womb cancer only and not ovarian cancer.

Q4 - What would happen if I do not have surgery?

If you decide not to have surgery, then nothing will really change. You will need to look out for any symptoms and if you think anything is wrong you need to go to your GP to get it checked. You know your body best, so don't be afraid to go to your GP if you notice anything unusual. Make sure you tell your GP that you have a family history of ovarian cancer and are at increased risk of ovarian cancer when you talk to them.

Remember you can reconsider surgery at any time. If screening for ovarian cancer is shown to be effective in the future, you may then be offered screening as an alternative to surgery.

Q5 - Is there anything else I can do?

As there is no routine screening available, you can choose to have screening privately, but you should be aware that this has not yet been shown to be effective. You should also keep an eye on any symptoms.

Studies have found that a healthy diet with plenty of fruit and vegetables, keeping a healthy weight and an active lifestyle can improve overall well-being and might reduce your chances of getting cancer.

In some countries it is recommended that women at high risk of ovarian cancer take the oral contraceptive pill when they are pre-menopausal. The oral contraceptive pill reduces ovarian cancer risk by up to half, but it has also been found to increase breast cancer risk. The guidelines in the UK do not recommend taking the oral contraceptive pill solely for the prevention of ovarian cancer at the moment, although in some situations reduction in ovarian cancer risk may outweigh any increase in risk of breast cancer. Whether the oral contraceptive pill might be an option for you depends on your risk and family history of breast cancer and should be discussed with your genetic counsellor and GP.

8. Your Decision (Example)

26

In the exercise on the next few pages you can rate some facts depending on how much they make you want **to have** or want **to avoid** surgery. Give each fact a ranking number (e.g. from 1 (a little) to 3 (a lot), then add the numbers up to see where you stand.

The table below is already filled in to give you an example. On page 28 you will find an empty table for you to fill in.

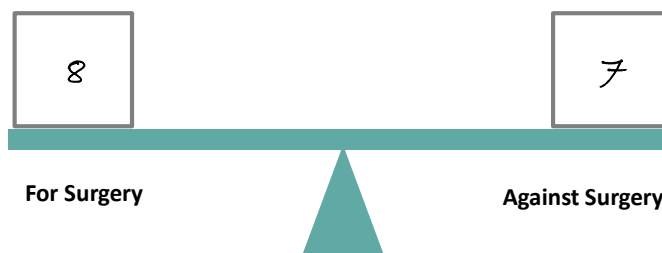
| The fact that... | Makes me want to have surgery | Does not come into my decision | Makes me want to avoid surgery |
|--|--------------------------------------|--------------------------------|---------------------------------------|
| ...surgery will reduce my ovarian cancer risk | 3 | | |
| ...there is a risk of complications linked to surgery | | | 1 |
| ...I will not be able to become pregnant after surgery | | X | |
| ...I will go into surgical menopause after surgery | | | 2 |
| ...there is no effective screening for ovarian cancer, so I cannot get checked out regularly | 3 | | |
| ...the symptoms of ovarian cancer are very vague, so I may not notice them early | 2 | | |
| ...I might feel different about my body after surgery | | | 2 |
| ...I might enjoy sex less after surgery | | | 1 |
| Enter your own reasons: | | | |
| Have to take time off work | | | 1 |
| | | | |
| | | | |
| | | | |
| Add together the numbers in each column: | 8 | | 7 |

8. Your Decision (Example)

27

Below you can enter the overall number into the scales. Then note down how you feel about the decision and where you stand.

The scales below are already filled in to give you an example. On page 29 you will find an empty scale for you to fill in.



More weight for than against surgery:

Overall your answers show that at this moment in time you are leaning towards having the surgery. Even though you may have some reasons why you would want to avoid surgery, they are not as important to you as the reasons that make you want surgery. Therefore you could now make an appointment to discuss this decision with your doctor and go through any concerns you might have.

Same weights both sides:

Overall your answers show that at this moment you are completely undecided about whether or not to have surgery. Your reasons for why you would want to have and why you would want to avoid surgery balance each other out. Therefore you may want to discuss your options further with your doctor/genetic counsellor.

More weight against than for surgery:

Overall your answers show that at this moment in time you are leaning towards not having surgery. Even though you may have some reasons why you would want to have surgery, they are not as important to you as the reasons that make you want to avoid surgery. Therefore at this moment in time surgery is probably not the right option. However, keep in mind that things may change over time and that eventually you may lean more towards surgery. You can come back to OvDex and do this exercise again anytime to see whether anything has changed.

Overall decision: My reasons for surgery weigh more than those
against surgery, but I am still not sure. The
numbers are very similar.

My action plan: I will make an appointment with my genetic
counsellor to discuss the menopause and find out
whether I can take HRT.
Will talk to my boss about taking time off and
whether it would cause any problems.

8. Your Decision

28

In the exercise on the next few pages you can rate some facts depending on how much they make you want **to have** or want **to avoid** surgery. Give each fact a ranking number (e.g. from 1 (a little) to 3 (a lot), then add the numbers up to see where you stand.

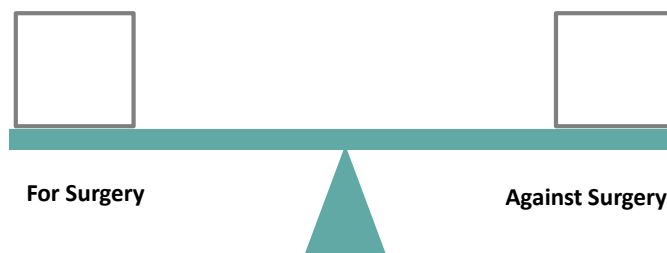
Make sure you give the score on the correct side of the table.

| The fact that... | Makes me want to have surgery | Does not come into my decision | Makes me want to avoid surgery |
|--|--------------------------------------|--------------------------------|---------------------------------------|
| ...surgery will reduce my ovarian cancer risk | | | |
| ...surgery will reduce my breast cancer risk | | | |
| ...I will not be able to become pregnant after surgery | | | |
| ...I will go into surgical menopause after surgery | | | |
| ...there is no effective screening for ovarian cancer, so I cannot get checked out regularly | | | |
| ...the symptoms of ovarian cancer are very vague, so I may not notice them early | | | |
| ...I might feel different about my body after surgery | | | |
| ...I might enjoy sex less after surgery | | | |
| Enter your own reasons: | | | |
| | | | |
| | | | |
| | | | |
| | | | |
| | | | |
| Add together the numbers in each column: | | | |

8. Your Decision

29

Below you can enter the overall number into the scales. Then note down how you feel about the decision and where you stand.



More weight for than against surgery:

Overall your answers show that at this moment in time you are leaning towards having the surgery. Even though you may have some reasons why you would want to avoid surgery, they are not as important to you as the reasons that make you want surgery. Therefore you could now make an appointment to discuss this decision with your doctor and go through any concerns you might have.

Same weights both sides:

Overall your answers show that at this moment you are completely undecided about whether or not to have surgery. Your reasons for why you would want to have and why you would want to avoid surgery balance each other out. Therefore you may want to discuss your options further with your doctor/genetic counsellor.

More weight against than for surgery:

Overall your answers show that at this moment in time you are leaning towards not having surgery. Even though you may have some reasons why you would want to have surgery, they are not as important to you as the reasons that make you want to avoid surgery. Therefore at this moment in time surgery is probably not the right option. However, keep in mind that things may change over time and that eventually you may lean more towards surgery. You can come back to OvDex and do this exercise again anytime to see whether anything has changed.

Overall decision: _____

9. Contacts and Resources

Here are some links and contact details that you might find helpful (even though we have taken great care in choosing websites from reliable sources please note we are not responsible for the content of these websites):

The Cancer Genetics Story Bank – An online collection of stories told by patients and professionals about cancer genetics, genetic testing and risk-reduction decisions. Developed by the Cancer Genetics Service for Wales (CGSW)

www.cancergeneticsstorybank.co.uk

NHS Choices – A website created by the NHS to provide information to patients. Includes information on ovarian cancer, genetics and genetic testing.

www.nhs.uk

Macmillan – A UK charity for anyone affected by cancer with good information about genetic testing for cancer and mind-body therapies

www.macmillan.org.uk or call the support line on 0808 808 00 00

Target Ovarian Cancer – A UK charity which supports research into ovarian cancer and provides useful information including an 'Ask the Expert' feature. Available in several languages.

www.targetovariancancer.org.uk

Ovacome – A UK charity providing information and support for everyone affected by ovarian cancer. Includes links to a number of patient blogs.

www.ovacome.org.uk or call the supportline on 08453710554

Your GP and/or Genetics Service – There to help you with any questions or concerns.

Evidence for cancer risk

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Appendix 9

| | | |
|-----|--|-----|
| 9.1 | <i>Genetic Counselling Outcome Scale (GCOS-24)</i> | 641 |
| 9.2 | <i>DelibeRATE scale</i> | 642 |
| 9.3 | <i>Healthcare Evaluation Provider Satisfaction Questionnaire</i> | 643 |

Appendix 9.1 - Genetic Counselling Outcome Scale (GCOS-24)

The Genetic Counselling Outcome Scale (GCOS-24)

Using the scale below, circle a number next to each statement to indicate how much you agree with the statement. Please answer all the questions. For questions that are not applicable to you, please choose option 4 (neither agree nor disagree).

| | | strongly disagree | disagree | slightly disagree | neither agree nor disagree | slightly agree | agree | strongly agree |
|---|---|-------------------|----------|-------------------|----------------------------|----------------|-------|----------------|
| 1 = strongly disagree 2 = disagree 3 = slightly disagree 4 = neither disagree nor agree 5 = slightly agree 6 = agree 7 = strongly agree | | | | | | | | |
| 1 | I am clear in my own mind why I am attending the clinical genetics service. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 2 | I can explain what the condition means to people in my family who may need to know. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 3 | I understand the impact of the condition on my child(ren)/any child I may have. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 4 | When I think about the condition in my family, I get upset. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 5 | I don't know where to go to get the medical help I / my family need(s). | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 6 | I can see that good things have come from having this condition in my family. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 7 | I can control how this condition affects my family. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 8 | I feel positive about the future. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 9 | I am able to cope with having this condition in my family. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 10 | I don't know what could be gained from each of the options available to me. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 11 | Having this condition in my family makes me feel anxious. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 12 | I don't know if this condition could affect my other relatives (brothers, sisters, aunts, uncles, cousins). | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 13 | In relation to the condition in my family, nothing I decide will change the future for my children / any children I might have. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 14 | I understand the reasons why my doctor referred me to the clinical genetics service. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 15 | I know how to get the non-medical help I / my family needs (e.g. educational, financial, social support). | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 16 | I can explain what the condition means to people outside my family who may need to know (e.g. teachers, social workers). | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 17 | I don't know what I can do to change how this condition affects me / my children. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 18 | I don't know who else in my family might be at risk for this condition. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 19 | I am hopeful that my children can look forward to a rewarding family life. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 20 | I am able to make plans for the future. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 21 | I feel guilty because I (might have) passed this condition on to my children. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 22 | I am powerless to do anything about this condition in my family. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 23 | I understand what concerns brought me to the clinical genetics service. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
| 24 | I can make decisions about the condition that may change my child(ren)'s future / the future of any child(ren) I may have. | 1 | 2 | 3 | 4 | 5 | 6 | 7 |

Appendix 9.2 - DelibeRATE scale

DelibeRATE (Pre-and Post-BresDex)

Please click on the number that matches with your agreement or disagreement with the statements below.

| | | | | | | | | | | |
|----|--|-------------------|---|---|---|---|---|---|---|----------------|
| D1 | I understand the options available to me | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |
| D2 | I know enough about each option to help me decide | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |
| D3 | I know about the advantages and disadvantages of each option | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |
| D4 | I know how I feel about each option | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |
| D5 | I can imagine what it would feel like to live with each option | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |
| D6 | I can judge which option is better for me | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |
| D7 | I can judge which option is worse for me | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |
| D8 | I feel I've given the options available to me enough thought | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |
| D9 | Even though it may be difficult, I am able to make a choice | Strongly disagree | 1 | 2 | 3 | 4 | 5 | 6 | 7 | Strongly agree |

From: Sivell S, Edwards A, Manstead A S, Reed M W R, Caldon L, Collins K, Clements A and Elwyn G (2012) Increasing readiness to decide and strengthening behavioral intentions: Evaluating the impact of a web-based patient decision aid for breast cancer treatment options (BresDex: www.bresdex.com). Patient Education and Counseling 88 (2) 209-217

Appendix 9.3 - Healthcare Evaluation Provider Satisfaction Questionnaire

Please answer the following questions on a scale of 1 (poor) to 5 (excellent)

| Item: | Poor | | | Excellent | |
|---|------|---|---|-----------|---|
| | 1 | 2 | 3 | 4 | 5 |
| How useful was the information provided? | | | | | |
| Did you find the information easy to understand? | | | | | |
| How effective are the graphics? | | | | | |
| What is your general satisfaction with this tool? | | | | | |

Please rate the following statements on a scale of 1 (strongly disagree) to 5 (strongly agree)

| Item: | Strongly disagree | | | Strongly agree | |
|---|-------------------|---|---|----------------|---|
| | 1 | 2 | 3 | 4 | 5 |
| This tool could improve patient-doctor encounters | | | | | |
| This tool could save me time | | | | | |
| I would use it regularly in practice | | | | | |
| I would recommend that patients use this tool | | | | | |

Adapted from Schackmann E A, Munoz D F, Mills M A, Plevritis S K and Kurian A W (2013)
Feasibility evaluation of an online tool to guide decisions for BRCA1/2 mutation carriers.
Familial Cancer 12 (1) 65-73