



# The Amniocentesis Dilemma

Needs Assessment, Development and Field-Testing  
of a Theory-Based Decision Support Intervention

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**Marie-Anne Durand**

Thesis submitted for the degree of  
Doctor of Philosophy  
at Cardiff University

July 2009

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**Supervisors: Professor Glyn Elwyn and Doctor Jacky Boivin**

# **Abstract**

## **The amniocentesis dilemma: needs assessment, development and field-testing of a theory-based decision support intervention**

### **Background:**

Amniocentesis is the most common prenatal diagnostic procedure undertaken in the United Kingdom, usually performed after 15 completed weeks of pregnancy. The procedure is reported to have a 1% risk of miscarriage and the results of the chromosome tests may require further decision making about whether to continue with the pregnancy. Deciding about amniocentesis is a complex and emotionally charged decision, often undertaken in a short period of time and, under current practice, with little systematic decision support. Decision Support Interventions, also known as Patient Decision Aids, have been developed to help individuals learn about the features and implications of their treatment or screening options while improving communication with their health professionals. Those interventions are specifically targeted at preference-sensitive decisions with significant harms, benefits and uncertainty, where no screening or treatment option is objectively better than the other.

This thesis proposed to assess information and decision support needs of pregnant women undertaking amniocentesis testing and to design and field-test, in collaboration with pregnant women and health professionals, a theory-based Decision Support Intervention for amniocentesis testing (amnioDex).

### **Methods:**

A multi-method approach was adopted that included a systematic review, theoretical review, and qualitative analysis to develop and pilot a theory-based intervention intended for pregnant women facing a decision to undertake amniocentesis testing. The content areas and themes to be covered in the intervention were determined by a literature review and needs assessment conducted with pregnant women and health professionals. The prototype development of amnioDex (amniocentesis decision explorer) was guided by theory and included heuristic-based deliberation tools. Incremental prototypes of amnioDex and embedded deliberation tools were field-tested with lay users, health professionals and pregnant women facing a decision to undertake amniocentesis, using the “think-aloud” technique.

### **Results:**

The amnioDex intervention was developed over a period of two years and field-tested for eight months.

### **Conclusion:**

Findings from this thesis showed that it was feasible to use theory to generate a Decision Support Intervention acceptable to women facing amniocentesis testing and to health professionals counselling them. Future research needs to evaluate the effectiveness of amnioDex in a randomised controlled trial and to examine methods for effectively transferring theory into practice.

**APPENDIX 1:  
Specimen Layout for Thesis Summary and Declaration/Statements page  
to be included in a Thesis**

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**STATEMENT 2**

This thesis is the result of my own independent work/investigation, except where otherwise stated.

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## **Acknowledgements**

I would like to thank all the patients and health professionals who kindly took part in the needs assessment and development of amnioDex. This research would not have been possible without the exemplary support of the midwives at the antenatal clinic of the University Hospital of Wales. I am very grateful to the members of the amnioDex steering group for their support, advice and expertise: Lynne Taylor, Doctor Annie Procter, Doctor Andrew Dawson and Jane Fisher to name a few.

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This thesis is dedicated to my parents, Solange and Jacques, and to my sister, Natacha.

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# Chapter 1

## Introduction and Thesis Overview

### 1.1 Involving Patients in Medical Decision Making: the Patient Centred Approach

Medical decision making has evolved from a traditionally paternalistic model of decision making to a patient centred approach where shared decision making is considered the gold standard (Deber 1994; Emanuel and Emanuel 1992). Historically, the paternalistic model was recognised as the dominant approach in western medicines (Kaba and Sooriakumaran 2007; Parsons 1951). It assumed full authority and autonomy of the doctor, who provided information and made decisions without consulting the patient. The latter endorsed a passive role in sharing the physician's values and complying with the decisions made (Benbassat et al. 1998).

Since the 1980's, the patient's right to be informed and to participate in medical care decisions has been increasingly advocated. The assumption that physicians were in the best position to make healthcare decisions, independently of the patient's values, has been challenged. Research suggested that the experience and the consequences of an illness highly depended on the patient's "biography", individual characteristics and values (Armstrong 1979, 1984; Mead and Bower 2000; Smith and Hoppe 1991). In the early 1990's, there was a paradigm shift in medical practice from the paternalistic approach to Evidence Based Medicine (EBM). EBM postulates that clinical decisions (treatments, screening or diagnostic tests) should be justified by external clinical evidence and not by the physician's authority or medical traditions. Individual expertise should be combined with best available clinical and scientific evidence to decide on the care of individual patients (Sackett et al. 1996).

The patient status progressively shifted from a client and passive recipient of care to an active partner in medical decisions and care. The doctor-patient relationship was undergoing changes worldwide (Kaplan et al. 1996; Roter et al. 1997) and pressures on the NHS to promote patient autonomy and involvement in healthcare were rising (Smith 1998). A reform promoting increased patients' rights and involvement in healthcare was published in 1991 in the Patient's Charter: "you have the right to have any proposed treatment (...) clearly explained to you before you decide whether to agree with it" (Department of Health 1991). In parallel, informed consent was progressively recognised as a legitimate patient's right, where all patients were entitled to consent prior to any medical treatments or screening procedures (Sutherland et al. 1989). The legal obligation to inform patients (informed consent) combined with the consumer rights movement (Haug and Lavin 1983) and evolving nature of the physician-patient encounter prepared the transition towards shared decision making (Charles et al. 1997; Coulter 1999; Richards 1998).

Shared decision making is a conceptual model of medical decision making (Emanuel and Emanuel 1992) increasingly recognised as an ideal consultation style, extensively discussed and advocated but rarely implemented (Holmes-Rovner et al. 2000). Shared decision making has been defined (Charles et al. 1997, p. 5) as involving at least two actors (physician and patient) who share information and are jointly engaged in the decision making process to choose a treatment/screening option which is consistent with the patient's values and preferences. The literature examining patients' willingness to share medical decisions has been widely criticised for the heterogeneity of the methods used and conflicting results, frequently imputed to the difficulty to define participation in decision making (Deber 1994). A review of patient participation in decision making concluded that (i) patients wanted to be informed of treatment alternatives (Deber et al. 1996; Strull et al. 1984) and (ii) depending on



the circumstances, wished to be involved in decision making (Guadagnoli and Ward 1998). Further evidence suggested that patients wanted to be informed and consulted during the medical encounter (Elwyn et al. 1999) and that patients' satisfaction increased with the physician's empathy and patient-centeredness (Williams et al. 1998). Despite endless attempts at defining the concept, debates remain as to which medical encounter falls within or outside shared decision making. There is a difficulty defining patient involvement in medical decision making and when to adopt a shared decision making approach.

## **1.2 The Amniocentesis Dilemma or How to Cope With Clinical Equipoise**

There is often uncertainty about the benefits of a treatment or screening option. Godlee (2005) established that 47% of medical treatments were associated with insufficient scientific evidence. Situations of clinical equipoise are defined by genuine balance between the scientific evidence of estimated harms, benefits and outcome probabilities associated with each option presented to the patient. In situations of equipoise, patients' values and preferences may therefore be determinant in choosing a particular course of action and warrant patients' participation in medical decision making (Elwyn et al. 2000).

Amniocentesis testing is an invasive diagnostic procedure, performed in the second trimester of pregnancy, to provide foetal cells for karyotyping, that presents both harms and benefits. The values and preferences of the pregnant woman and her partner are dominant in deciding whether to accept or decline the test. Prenatal screening tests for Down's syndrome (i.e., blood tests or ultrasound scan) are routinely offered to all pregnant women in the UK in order to determine their risk of foetal chromosomal abnormality. Women who receive a higher risk result will be offered to undergo amniocentesis testing (see Chapter 3). The amniocentesis procedure is associated with a 1% risk of miscarriage, although this rate may vary by operator (Gaudry et al. 2008; Tabor et al. 1986). There is a lack of scientific evidence

about the exact cause and predisposing factors of a miscarriage. Amniocentesis testing is also characterised by uncertain outcomes. The procedure may lead to detection of chromosomal abnormality, further decision making about whether to continue with the pregnancy or foetal loss. Approximately 4% of women undergoing amniocentesis testing will receive a diagnosis of foetal chromosomal abnormality (Caine et al. 2005; Han et al. 2008; Lewin et al. 2000) while an estimated 1% will miscarry after amniocentesis. The trade-off between the 1% miscarriage risk and the gain in information provided by the chromosome test results is not always clear for women considering amniocentesis. In addition, the chromosome tests performed on the amniotic fluid will identify most common chromosomal abnormalities (e.g., trisomy 13, 18, 21 and exchange of chromosomes) but will not diagnose small changes in chromosomes (e.g., microdeletions). Finally, the chromosome test results do not indicate the severity of the abnormality detected. The severity of common chromosomal problems, such as Down's syndrome, is highly variable, ranging from mild learning disabilities to severe impairments and associated medical problems (e.g., heart conditions, leukaemia, diabetes) (Cleves et al. 2007; Paladini et al. 2000).

With uncertain outcomes, far-reaching-consequences, and significant harms and benefits, deciding whether or not to undergo amniocentesis is one of the most difficult decisions for pregnant women and their partner to consider (Beeson and Golbus 1979). Current research suggests that women receiving a high chance result of chromosomal abnormality and facing the decision to undergo amniocentesis generally experience acute stress and anxiety (Ng et al. 2004; Sarkar et al. 2008; Statham et al. 1997; Tercyak et al. 2001). A study of pregnant women's responses to an increased risk of carrying a baby with Down's syndrome revealed that the risk information provided by the screening test results triggered intense reactions of anxiety and worry (Susanne et al. 2006). Further research

suggests that maternal stress may be associated with poor outcomes for the mother and the foetus (Talge et al. 2007).

Further, there are documented concerns that information and decision support available during the diagnostic phase of pregnancy is not sufficient (Marteau 1995; St-Jacques et al. 2008). Poor understanding of prenatal tests accepted or declined and unrealistic expectations have been reported (Marteau 1995; Marteau 2002). In the literature, unrealistic expectations have been associated with increased anxiety, pain and likelihood of litigation (Johnston and Vogele 1993; Petticrew et al. 2000). In addition, information provided pre-amniocentesis is not tailored to pregnant women's interests, values and goals (Hunt et al. 2005) and does not always appear to be comprehensive (Marteau et al. 1992; Marteau 1993). The content and accuracy of prenatal genetic testing counselling has been documented by Bernhardt et al. (Bernhardt et al. 1998), indicating that specific topics were not systematically discussed with pregnant women (e.g., implications of test results, elective termination of pregnancy) and that information was sometimes inaccurate. Finally, an additional complexity is the recent implementation of Quantitative-Fluorescent Polymerase Chain Reaction test (QF-PCR test), a rapid test which provides results for specific chromosomal abnormalities (i.e., trisomy 13, 18 and 21) in 3 working days (Onay et al. 2008). In all antenatal clinics in Wales and elsewhere in the UK, provided a woman's consent is obtained, both QF-PCR and a full karyotype tests will be carried out on all amniotic fluid samples. There are unresolved issues surrounding the introduction of this service and the information required by both health professionals and pregnant women prior to the collection and analysis of foetal material and after the results are available (see Chapter 3). There is therefore scope for investigating and addressing women's information, decision support and emotional needs associated with amniocentesis testing.

### **1.3 Decision Support Interventions**

Decision Support Interventions (DESI) for patients (Elwyn et al. 2009a), also known as Patient Decision Aids, are designed to help individuals learn about the features, issues and implications of their treatment or screening options while improving communication with their healthcare providers (Estabrooks et al. 2001; Molenaar et al. 2000; O'Connor et al. 1999a). According to the Cochrane systematic review of decision aids, "Decision aids differ from usual health education materials because of their detailed, specific, and personalised focus on options and outcomes for the purpose of preparing people for decision making" (O'Connor et al. 2006, p. 2). DESIs have been specifically developed for preference-sensitive decisions (O'Connor et al. 2003a; Wennberg 1991) with significant harms, benefits, and equipoise, where no screening or treatment option is objectively better than the other. DESIs do not aim to replace the physician/patient interaction but intend to supplement medical counselling. Pioneer decision aids appeared 30 years ago, in the form of consultations structured by decision analysis (Pauker and Pauker 1979), preference elicitation techniques (Llewellyn-Thomas et al. 1982; O'Connor et al. 1985) and shared decision making programmes (Kasper et al. 1992).

Over the past 10 years, an increasing number of DESIs have been developed, in a variety of formats (paper, audio, video, web-based), addressing over 23 clinical decisions (O'Connor et al. 2006). In 2006, it was estimated that DESIs were accessed about 9 million times, principally online (O'Connor et al. 2007a). The Cochrane systematic review of decision aids for patients facing health treatments and screening decisions (O'Connor et al. 2001; O'Connor et al. 2006) examined the results of 55 randomised controlled trials of DESIs (2009 update) addressing 23 screening or treatment decisions. The findings revealed that DESIs increased knowledge, realistic expectations, participation in decision making and

reduced decisional conflict and indecision post-intervention compared to usual practice. However, further research suggested that while knowledge may be increased, DESIs did not significantly influence treatment or screening decisions (Estabrooks et al. 2001). Molenaar et al. established that the DESI's effect on satisfaction with the decision, decision uncertainty and health outcomes was limited and rarely evaluated (Molenaar et al. 2000). Furthermore, the heterogeneity of formats, decisions addressed and methods used to develop and evaluate those interventions (e.g., controlled versus non-controlled studies) may bias the assessment of DESIs' effectiveness (Molenaar et al. 2000).

Multiple measures of DESIs' effectiveness have been developed but no consensus exists as to which measures should be used to optimally assess their effect (Elwyn et al. 2009b). Difficulties measuring DESIs' effectiveness are related to the lack of consensus on the aims of those interventions and criteria on which their effectiveness should be assessed (Kennedy 2003). There have been multiple attempts at defining a good decision and several definitions of decision quality have been put forward (Ratliff et al. 1999; Sepucha et al. 2007). Sepucha et al. defined decision quality as "the extent to which the implemented decision reflects the considered preferences of a well-informed patient" (Sepucha et al. 2007, p. 262). While definitions exist, there is currently no well-validated measure of decision quality, able to determine the match between patients' values and treatment (or screening) decisions (Sepucha et al. 2008; Kennedy 2003). Elwyn et al. established that existing measures of decision quality were post-hoc measurements which focussed on two key constructs: knowledge (making an informed choice) and preferences (the decision must be consistent with the patient's preferences). They exposed several limitations to current definitions and post-hoc measures of decision quality (Elwyn et al. 2009b). The duration of decision making is indeterminate. The difficulty therefore arises as to when to measure

decision quality and for how long. In addition, measuring the quality of decisions according to the outcomes (i.e., post-hoc) is not a relevant standpoint. Current research directions in this area are therefore moving towards a process measure of decision making: the measure of deliberation (Elwyn et al. 2009b). Finally, measures of DESIs' effectiveness have been criticised for their inability to measure the DESI's impact on health status and well-being (Entwistle et al. 1998).

DESIs are complex interventions which can potentially influence patients' treatment or screening decisions. It is therefore imperative to ensure that those interventions are unbiased and safe for patients to use. A set of internationally accepted standards has been developed by the International Patient Decision Aids Standards (IPDAS) collaboration to assess the quality and potential biases of existing DESIs (Elwyn et al. 2006). The IPDAS collaboration developed a checklist for the assessment of DESIs using an online Delphi process where 122 stakeholders from 14 countries rated 80 criteria divided into 12 quality domains. The IPDAS checklist assessed whether the intervention featured relevant components and had been rigorously developed and evaluated. The checklist did not provide quantitative assessments about the DESI's quality. The IPDAS instrument (IPDASi) was subsequently developed and validated to achieve a detailed quality assessment of existing DESIs (Elwyn et al. 2009c). The validated version of IPDASi comprised 47 items divided in 10 domains (see Chapter 4). IPDASi was validated using dual rater assessment of 30 DESIs developed by five major producers. The findings showed that IPDASi could effectively measure DESIs' quality provided adequate rater calibration training was undertaken pre-assessment (Elwyn et al. 2009c).

As noted previously, in the absence of conclusive scientific evidence towards one treatment option or the other, patients' values, preferences and biography play a crucial role in deciding on a specific course of action. Most recent accounts of research directions in DESI development define DESIs as "interventions that describe and justify the conditions where equipoise exists for both clinicians and patients (dual equipoise), provide information about options which help people to deliberate about counterfactuals, construct and forecast preferences about short, intermediate and long-term outcomes which have relevant consequences" (Elwyn et al. 2009a, in submission). DESIs can therefore be conceptualised as including a core component of information about options, associated harms and benefits, and a deliberation component, namely strategies or methods designed to facilitate the expression and clarification of patients' values (Elwyn et al. 2009d). Such methods or strategies have been developed to assist patients in structuring their preferences to achieve decision making, and have been described in a number of ways: value/attribute/preference elicitation (Feldman-Stewart et al. 2006), value-clarification exercises or preference-clarification exercises (O'Connor et al. 1999b) etc. Llewellyn-Thomas (2009) exposed four prerequisites to the development and use of value-clarification or deliberation strategies. First, deliberation strategies should be made available to patients who have not yet chosen a specific course of action and/or wish to be involved in their healthcare decisions. Second, the provider's views and values should not be imposed upon the patient/user's deliberation. Third, deliberation strategies should ideally avoid framing biases. Finally, those methods may be used in iteration, to account for the fact that preferences may not remain constant over time.

This thesis' aims were to (i) understand the information and decision support needs of women who have been offered an amniocentesis, and (ii) design and pilot, in collaboration with pregnant women and their health professionals, a theory-based DESI capable of

addressing these needs. The following section presents an overview of the issues discussed in this thesis.

## **1.4 Thesis Overview**

### **1.4.1 Identifying Evidence and Theoretical Basis (Chapters 2, 3 and 4)**

The conceptual and practical development of the amnioDex intervention was informed by the Medical Research Council (MRC) framework for design and evaluation of complex interventions (Campbell et al. 2000; Craig et al. 2008). According to this framework, best available evidence should be identified through high quality systematic reviews. The theoretical basis underlying the intervention development should also be examined in the pre-clinical phase of development. Best available evidence and relevant theory should then be combined to define the intervention's components (phase 1: modelling). For best practice, qualitative testing (e.g., focus groups, surveys) may be used to determine and develop intervention components. The intervention should be developed in phase 2, where acceptability and feasibility issues will be concurrently examined. Pilot studies should be conducted before evaluating the intervention to verify its acceptability, usability and feasibility (phase 3).

While theories or models of behaviour change have been commonly applied to the development of behavioural interventions (Abraham and Michie 2008; Serlachius and Sutton 2009), little attention has been given to the theoretical underpinnings of interventions designed to support decision making. Decision making theories exist and their applications have improved our understanding of how individuals make decisions. They attempt to explain and predict how individuals make complex decisions and describe the factors or situations likely to impair the decision-making process and lead to poor decision outcomes and decisional regret. It would therefore seem appropriate to integrate theoretical constructs



into the development of information and deliberation components embedded in DESIs, in the anticipation of significant impact on the decision quality and outcomes. The aim of Chapter 2 was to undertake a theoretical review of models and theories underlying the development of DESIs included in the Cochrane systematic review of patient decision aids (O'Connor et al. 2006). This chapter examined the extent to which theory guided the conception, prototype development and evaluation of selected DESIs, with a view to embed theory-based information and deliberation components in the development of amnioDex.

As noted previously, DESIs have been specifically developed to help individuals make preference-sensitive decisions, characterised by uncertain outcomes and limited scientific evidence. The decision to undertake amniocentesis involves complex information, uncertain outcomes and far-reaching consequences, at a time of emotional upheaval. Chapter 3 described the complexity, clinical characteristics, and equipoise associated with amniocentesis testing while examining existing arrangements for informing and supporting pregnant women (and their partners) facing a decision to undertake amniocentesis. The aim of Chapter 4 was to conduct a systematic review of existing DESIs for amniocentesis testing, using the IPDAS instrument to critically appraise the quality of interventions included for review.

#### **1.4.2 Needs Assessment and Prototype Development (Chapters 5, 6 and 7)**

To comply with the MRC framework and IPDAS quality criteria, a qualitative methodological approach was adopted to determine the content of the intervention and to assess potential users' and health professionals' needs. The aim of Chapter 5 was to assess information and decision support needs of pregnant women facing a decision to undertake amniocentesis testing using semi-structured interviews. The direct assessment of pregnant women's needs related to amniocentesis testing was completed by the professionals'

evaluation of information and decision support needed prior to deciding about amniocentesis (Chapter 6). Based on the literature reviews and needs assessment conducted with relevant stakeholders, the intervention was developed. Chapter 7 described the conceptual, theoretical and practical development of amnioDex: a web-based decision explorer for women considering amniocentesis testing. Normative decision making theories, such as expected utility theory, do not normally account for the individual's emotional, cognitive, environmental and/or time constraints. Normative theories are derived from mathematical models and assume unbounded rationality: unlimited computational capacities, knowledge and time (Todd and Gigerenzer 2000). By contrast, descriptive theories or models of decision making recognise that decision makers have limited reasoning and computational abilities and examine ways of overcoming these difficulties. Interventions designed to facilitate decision making processes, such as amnioDex, may benefit from building on key concepts of how individuals actually make decisions under risk. In the clinical context of amniocentesis testing, where high emotional demands and complex information limit people's capacity to quantify utilities of options, the assumption was made that descriptive models of decision making would fit this specific context better than normative theories. Two descriptive theories of decision making were therefore chosen to guide the development of amnioDex: prospect theory and differentiation and consolidation theory (Chapter 7).

### **1.4.3 Qualitative Field-Testing of AmnioDex and Deliberation Components (Chapters 8 and 9)**

In order to investigate potential usability and acceptability issues associated with the evaluation and implementation of amnioDex, the intervention was piloted with relevant stakeholders. The aim of Chapter 8 was to develop deliberation tools based on models of bounded rationality (i.e., heuristics) and to field-test these tools with relevant stakeholders

while Chapter 9 examined the overall usability and acceptability of the amnioDex intervention piloted with lay users, pregnant women and health professionals.

Finally, the main findings, clinical implications of this thesis and future research directions will be discussed in Chapter 10.

# Chapter 2

## Theoretical Review

### 2.1 Introduction

The purpose of this chapter was to review and critically appraise the theoretical underpinnings of DESIs included in a Cochrane systematic review. Decision making theories exist and their applications have improved our understanding of how individuals make decisions. However, the use of decision making theories for the development of DESIs seems relatively rare in practice. Over the past decade, the conceptual and theoretical basis of DESIs has remained largely unexplored (O'Connor et al. 2007b). In spite of criteria for the design and evaluations of DESIs being developed (Elwyn et al. 2006), the necessity to adhere to conceptual or theoretical frameworks relevant to decision making has not yet been recognised. Most interventions in this field appear to have been developed in a practical manner, using a wide range of media, timeframes and purposes (Entwistle et al. 1998).

A literature review of 547 studies of health technologies ranging from the comparison of information mediums to the use of DESIs revealed that 82 % of the interventions did not make use of any theory or model of decision making (Bekker et al. 1999). Among interventions that explicitly referred to theory, there was little account of how a chosen theory was subsequently applied to the practical design of health technologies. Similarly, Bowen et al. (2006) investigated the theoretical basis of interventions promoting patient's informed decision making in the clinical context of cancer screening. The findings showed that 5 out of 14 interventions referred to a theoretical framework but did not specify how selected theories had shaped the design of the intervention. None of the articles reporting the development and evaluation of the interventions commented on the utility of the chosen theoretical foundation.

There is no clear description of a deliberate avoidance of theory nor is there detailed attention to how some, albeit a minority, used a specific theory for design, development and evaluation of DESIs. Furthermore, the impact of theory on the DESI's efficacy has not been formally assessed. Interventions that are based on theory may be more efficient and reliable than interventions developed without relevant theoretical framework. However, for the time being, empirical evidence is missing in this area. The aim of the research presented in this chapter was to describe and analyse rigorously developed DESIs in order to determine the contribution of theories or models of decision making to their conception, design, development and evaluation. As a sample frame, 55 published randomised controlled trials of DESIs included in the Cochrane systematic review were selected for review (O'Connor et al. 2006).

## **2.2 Methods**

The sample frame included 55 trials of patient decision aids for people facing health treatment or screening decisions, included in the Cochrane systematic review. The assumption was made that DESIs evaluated by randomised controlled trials included in a Cochrane review would have been among those most rigorously developed. In the Cochrane review, 22,778 citations were identified and 55 randomised controlled trials of DESIs were selected for review. The interventions focussed on 23 screening and treatment decisions in various clinical contexts. The DESIs were evaluated in randomised controlled trials and compared to usual care (usual verbal information or routine information leaflet) or to simpler decision tools.

All interventions to be considered received independent dual rating (M-A D and MS). All full text articles reporting the development and evaluation of the DESIs in a randomised

controlled trial were reviewed. All relevant articles were rigorously analysed to assess the degree to which each chosen decision making theory or model had informed the conception, prototype development, field-testing (if applicable) and evaluation of the intervention. For the purpose of this review, field-testing was defined as the process whereby the prototype DESI is shown to potential users who comment on its content and usability prior to evaluation (Evans et al. 2007). Any mention of a theoretical framework in the text or in the reference list was independently noted by each reviewer. The nature and category of the identified theoretical framework were then discussed between raters, who met on a fortnightly basis until all citations had been reviewed. The agreement between raters regarding the theoretical review of articles was examined. After a theoretical framework was identified and named, the authors of the article were contacted to investigate how theory had guided the design and evaluation of their interventions. They were informed that the theoretical review would be based on their published work if they did not provide a reply within two months.

### **2.3 Results**

In total, 78 full text articles reporting the development and evaluation of DESIs in a randomised controlled trial were reviewed. The 55 trials of patient decision aids for people facing health treatment or screening decisions were included in the Cochrane systematic review. However, the authors noted that three interventions (Gatellari and Ward 2003; Green et al. 2004a; O'Connor et al. 1998a) had been evaluated in two or more trials (Dodin et al. 2001; Gatellari and Ward 2005; Green et al. 2005; Legare et al. 2003; O'Connor et al. 1998b). There may have been small changes between versions but the assumption was made that the theoretical framework would remain the same. The present chapter was therefore based on the analysis of 50 DESIs and their associated publications. The consistency between raters on the theoretical review of all 78 citations was high (96%).

### **2.3.1 Prevalence of “Atheoretical” Interventions**

The analysis revealed that 17 out of 50 interventions referred to a theory or model of decision making: the majority of which could be categorised as normative theories (see Figure 2.1). Ten theories or models of decision making were identified. As far as could be determined, the rigorous analysis of all 78 citations revealed that the conception, prototype development and evaluation of 33 DESIs were not based on any theoretical foundation. All 17 authors of the theory-based interventions were contacted and asked to provide additional information regarding the use of theory in conceiving, developing and evaluating the intervention. Seven authors answered and provided additional information on the use of their chosen theoretical framework.

### **2.3.2 Normative Theories of Decision Making**

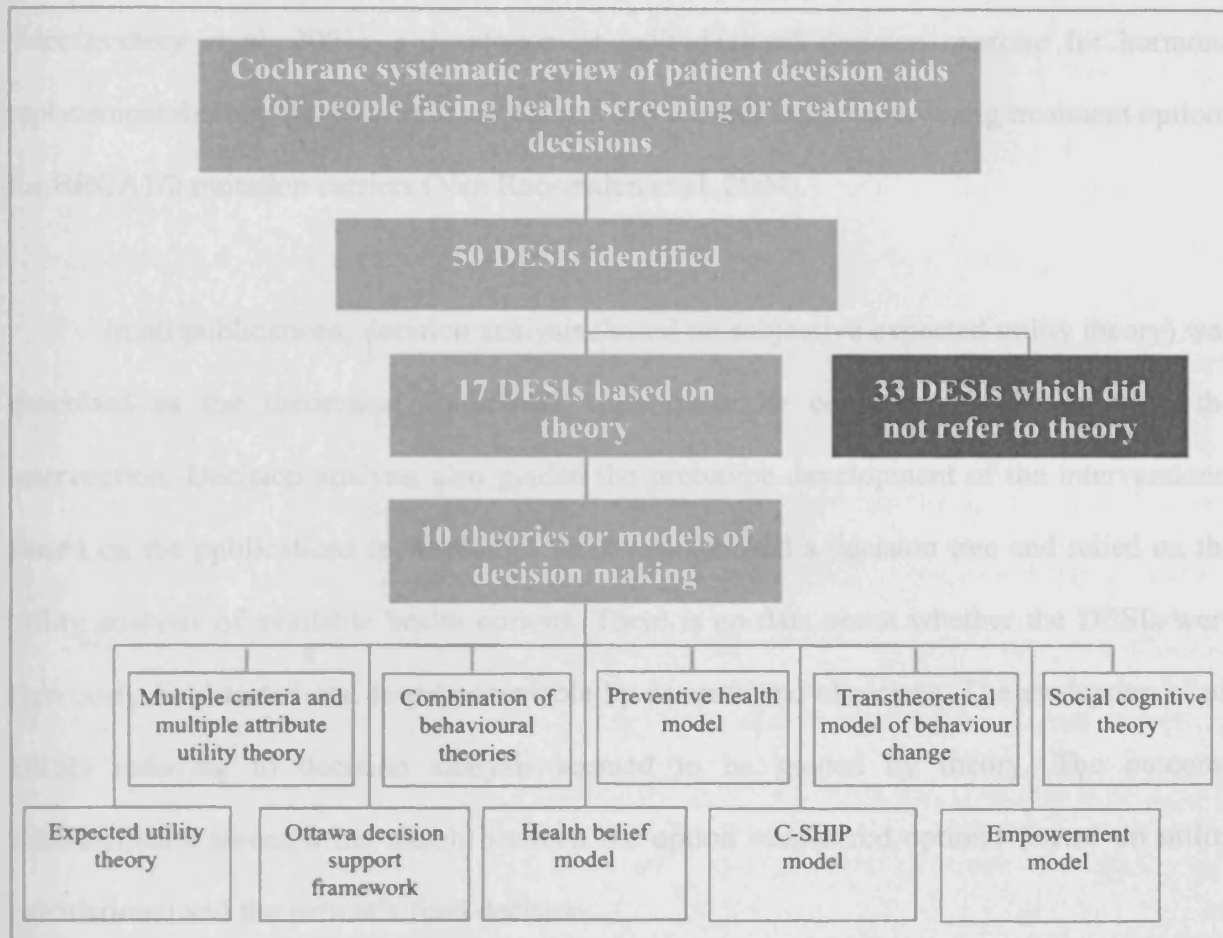
Table 2.1 shows the theory-driven DESIs. Five interventions referred to decision analysis (i.e., expected utility theory) (Bekker et al. 2003; Clancy et al. 1988; Holmes-Rovner et al. 1999; Montgomery et al. 2003; Rothert et al. 1997; Van Roosmalen et al. 2004). Decision analysis is an operationalisation of the expected utility theory (Howard and Matheson 1984), derived from the expected utility hypothesis. Expected utility theory is a normative theory of decision making, originally formulated in 1738 by Bernoulli and later developed by Von Neumann and Morgenstern (Kahneman and Tversky 1979; Pratt et al. 1964; Von Neumann and Morgenstern 1944). Normative theories of decision making specify how individuals should process information and make a decision under what are presumed ideal conditions. The decision analytic method and decision tree have been widely used in designing DESIs for the past 10 years (Magee 1964). Decision analysis was first applied to patient counselling in 1979 (Pauker and Pauker 1979).

Table 2.1 *Characteristics of Theory-Driven Decision Support Interventions Included for Review*

<b>Theoretical foundation</b>	<b>DESI component informed by theory</b>	<b>First author, Year</b>	<b>Health decision addressed in DESI</b>	<b>Format</b>
Decision analytic method based on the expected utility theory	Conception Prototype development Evaluation	Bekker 2004 (Bekker et al. 2004)	Prenatal diagnosis for Down's syndrome	Decision analysis plus consultation
Decision analytic method based on the expected utility theory	Conception Prototype development Evaluation	Clancy 1988 (Clancy et al. 1988)	Hepatitis B vaccine	Leaflet & decision analysis
Decision analytic method based on the expected utility theory	Conception Prototype development Evaluation	Montgomery 2003 (Montgomery et al. 2003)	Hypertension treatment	Decision analysis
Decision analytic method based on the expected utility theory	Conception Prototype development Evaluation	Rothert 1997 (Rothert et al. 1997)	Hormone replacement therapy	Discussion or personalised decision exercise
Decision analytic method based on the expected utility theory	Conception Prototype development Evaluation	Van Roosmalen 2004 (Van Roosmalen et al. 2004)	Treatment options for BRCA1/2 mutation carriers	Video & leaflet with decision analysis
Multiple attribute and multiple criteria decision making theories	Conception Prototype development Evaluation	Dolan 2002 (Dolan and Frisina 2002)	Colon cancer screening	Standardised interview (using analytic hierarchy process) & leaflet
Ottawa decision support framework	Conception Evaluation (partial use of the theories)	Drake 1999 (Drake 1999)	Prenatal diagnostic testing	Audiotape & booklet
Ottawa decision support framework	Conception Prototype development Evaluation (partial use)	Lalonde 2004 (Lalonde et al. 2004)	Cardiovascular health treatment	Video & booklet
Ottawa decision support framework	Conception Prototype development Evaluation (partial use)	O'Connor 1998 (O'Connor et al. 1998)	Hormone replacement therapy	Audiotape & booklet
Ottawa decision support framework	Conception Prototype development Evaluation (partial use)	Shorten 2005 (Shorten et al. 2005)	Birth options after previous caesarean	Booklet
Combination of behavioural models of decision making	Poor use of theory	Lerman 1997 (Lerman et al. 1997)	Breast cancer genetic testing	Discussion & counselling
Cognitive-social health information processing model (C-SHIP)	Conception Prototype development Evaluation	Miller 1996 (Miller et al. 1996)	Breast cancer genetic testing	Discussion & leaflet
The preventive health Model	Conception Prototype development	Myers 2005 (Myers et al. 2005)	Prostate Specific Antigen (PSA) testing	Discussion & leaflet
Social cognitive theory	Conception (partial use) Prototype development (partial use) Evaluation	Partin 2004 (Partin et al. 2004)	PSA testing	Video
Health belief model	Early conception Evaluation	Schapira 2000 (Shapira and VanRuiswyk 2000)	Prostate cancer screening	Booklet
The transtheoretical model	Poor use of theory	Pignone 2000 (Pignone et al. 2000)	Colon cancer screening	Video
Empowerment model	Early conception	Davison 1997 (Davison and Degner 1997)	Prostate cancer treatment	Written information package & audiotape of medical consultation & discussion



Figure 2.1 *Theories and Models of Decision Making Identified in the Review*



In the context of DESI development, decision makers are expected to specify the utility of available options on a numerical scale and multiply this number by their outcome probabilities to identify the option with the highest expected utility. In contrast to other theoretical frameworks, the theory and mechanisms of decision analysis (decision trees and subjective expected utility calculations) provided a recognisable architecture for the DESI development.

Based on the analysis of all related publications and contact with authors, decision analysis appeared to have informed the early conception of all DESIs citing this theory: a decision analytic consultation for prenatal diagnostic testing for Down's syndrome (Bekker et al. 2004), a leaflet coupled with individualised decision analysis for hepatitis B vaccine

(Clancy et al. 1988), a computerised self-completed interview for hypertension treatment (Montgomery et al. 2003), a discussion or individualised decision exercise for hormone replacement therapy (Rothert et al. 1997) and a video and leaflet addressing treatment options for BRCA1/2 mutation carriers (Van Roosmalen et al. 2004).

In all publications, decision analysis (based on subjective expected utility theory) was described as the theoretical framework supporting the conception and design of the intervention. Decision analysis also guided the prototype development of the interventions. Based on the publications reviewed, all interventions used a decision tree and relied on the utility analysis of available health options. There is no data about whether the DESIs were rigorously field-tested and found acceptable by patients and clinicians. The evaluation of all DESIs referring to decision analysis seemed to be guided by theory. The outcome measurements assessed the match between the option considered optimal (based on utility calculations) and the patient's final decision.

One intervention for colon cancer screening, combining an interview with a leaflet (Dolan and Frisina 2002) explicitly referred to the multiple criteria decision making (Zeleny 1982) and multiple attribute utility theory (Hwang and Yoon 1981). Multiple criteria decision making and multiple attribute utility theories are normative theories of decision making derived from mathematical theories of multiple criteria or multiple attributes problem solving (Dyer et al. 1992). Contrary to the subjective expected utility theory, they do not elicit patients' values using decision analytic trees but frequently resort to an analytic hierarchy process (Saaty 1990). The analytic hierarchy process places options in a comparison matrix of paired attributes, thus allowing the decision maker to compare the consistency of preferences. The multiple criteria decision making and multiple attribute utility theories did

provide the conceptual framework for designing and developing the intervention. However, there was no evidence of field-testing prior to evaluation. The evaluation of the DESI seemed to be informed by theory since the outcome measures (decision process and decision outcomes assessing the match between the screening plans and patients' final screening choice) matched the key constructs of the multiple criteria decision making and multiple attribute utility theories.

### **2.3.3 The Ottawa Decision Support Framework**

Four DESIs referred to the Ottawa decision support framework: an audiotape and booklet DESI for prenatal diagnostic testing (Drake et al. 1999), a video and booklet DESI for cardiovascular health treatment (Lalonde et al. 2004), an audiotape and booklet intervention for hormone replacement therapy (O'Connor et al. 1998a) and a booklet providing information about birthing options after previous caesarean (Shorten et al. 2005). The Ottawa decision support framework is a combination of several decision making theories including the expectancy value model, decision analysis (described earlier), prospect theory, the conflict theory model of decision making and social support theories (Keeney and Raiffa 1976; Norbeck 1988). The expectancy value model assumes that individuals who have to make a choice between two or more options with significant harms and benefits are more likely to opt for the option with the highest expected values and success (Fishbein 1975). Prospect theory postulates that most decision makers do not normally behave in accordance with the axioms of expected utility theory (Kahneman and Tversky 1979). Prospect theory distinguishes two phases in the choice process: editing and evaluating. The editing phase consists of analysing the offered prospects before evaluating them and choosing the prospect of highest value (evaluating phase). This theory also assumes that the choice between two courses of actions is biased by the way in which the choices are described or framed. For instance, prospect theory demonstrated that losses loom larger than gains (Tversky and

Kahneman 1992). This has led to significant attention being given to risk communication formats. The conflict theory model of decision making assumes that making a decision generates stress, uncertainty and conflict within the choice situation (Janis and Mann 1977). The decision maker would therefore cope with stress and uncertainty through the search for, and evaluation of information and alternatives.

Based on the analysis of all related publications and contact with authors, the Ottawa decision support framework appeared to have informed the early conception and design of all the interventions naming this theoretical framework. The prototype development of three out of four DESIs (Lalonde et al. 2004; O'Connor et al. 1998a; Shorten et al. 2005) was guided by some of the theories included in the Ottawa decision support framework and appeared primarily informed by the conflict theory model of decision making and the expectancy value model. The prototype development of all interventions naming this framework did not appear to be based on all theories included in the Ottawa decision support framework. The transfer of the theoretical constructs of the expectancy value model and the conflict theory model of decision making into the design of the intervention was mainly identifiable as a value-clarification exercise. None of the related publications specified whether field-testing had been carried out prior to evaluation. Finally, the DESIs' evaluation seemed to be guided by some but not all of the theories included in the Ottawa decision support framework, namely the expectancy value model and the conflict theory model of decision making. Decisional conflict, measured using the decisional conflict scale (O'Connor 1995), was the main outcome measurement that appeared to be informed by the Ottawa decision support framework.

### **2.3.4 Combination of Decision Making Theories**

An education intervention for breast cancer genetic testing (Lerman et al. 1997) used a combination of behavioural models of decision making: the theory of reasoned action (Fishbein 1980), the consumer behaviour model (Engel et al. 1978) and the conflict theory model of decision making (Janis and Mann 1977). The theory of reasoned action assumes that the intention to engage in behaviour is determined by the decision maker's attitudes as well as the subjective norms of significant others regarding this behaviour. The consumer behaviour model is a model of consumer decision making process that identifies a set of variables that shape decision making such as individual differences, environmental influences or psychological processes. The analysis of all articles reporting the DESI's development and evaluation did not provide any evidence that behavioural models of decision making guided the design and prototype development of the intervention. The publications did not mention whether the intervention had been rigorously field-tested and found acceptable by patients and clinicians. The evaluation was not explicitly informed by behavioural models of decision making. Apart from knowledge, a very common if not systematic outcome measure in DESI evaluation, the outcome variables did not relate to the chosen theoretical framework.

One intervention combining a leaflet with a discussion about the pros and cons of breast cancer genetic testing referred to the cognitive-social health information processing model (C-SHIP) (Miller et al. 1996). It postulates that decision makers generally favour a systematic processing of information where both cognitive and emotional components (i.e., individual perception of risk, knowledge, beliefs, and expectancies) are integrated into the decision making process. In their publications, the authors thoroughly described the C-SHIP model as the theoretical framework supporting the DESI's design (Miller et al. 2005; Miller et al. 1996). The educational intervention they developed appeared to be strongly anchored in

the C-SHIP model. Most theoretical constructs were addressed and integrated into the concrete development of the intervention. There was a lack of information as to whether the intervention had been rigorously field-tested and found acceptable by patients and clinicians. In the evaluation, the outcome measurements were related to all major components of the C-SHIP model and therefore matched theory.

One intervention combining a consultation and a leaflet for Prostate Specific Antigen (PSA) testing referred to the preventive health model (Myers et al. 2005; Myers and Wolf 1990). This model was developed by the DESI's developers and took constructs from the health belief model, the theory of reasoned actions and the social cognitive theory. This model identifies a series of internal and external factors (e.g., socio-cultural background, cognitive and affective representations associated with the disease or condition) that strongly influence people's intention to act on their health. The preventive health model was comprehensively described in the publications reporting the development and evaluation of the intervention and seemed to have informed its early conception and prototype development. The decision education session designed to elicit patients' values was based on the key constructs of the preventive health model: preference clarification, cognitive evaluation, affective evaluation and social evaluation. There was no evidence of field-testing. The evaluation of the tool was partially informed by the preventive health model. Only one key principle of the model (personal preference) was related to the primary outcome measure: a screening decision preference score.

### **2.3.5 Cognitive and Social Theories of Decision Making**

A video-based intervention for PSA testing (Partin et al. 2004) referred to social cognitive theory (Bandura 1986). This model describes the developmental changes that individuals undergo over the course of their existence and is structured around the concept of

agency. The DESI's early conception and prototype development were partly informed by social cognitive theory. The pamphlet they designed explicitly addressed one key construct of social cognitive theory: cognitive processes (e.g., knowledge and attitudes). However, the intervention did not seem to integrate other key dimensions of social cognitive theory (e.g., environmental factors). The publications did not mention whether the intervention had been rigorously field-tested and found acceptable by patients and clinicians. The evaluation was predominantly driven by theory. The outcome measurements integrated three dimensions of the social cognitive theory: knowledge (of screening), patient characteristics (demographics and health status) and behaviour (screening uptake). As far as could be determined, there was no explicit measure of environmental factors, which is considered a key construct of social cognitive theory.

One intervention offering a booklet for prostate cancer screening (Shapira and VanRuiswyk 2000) mentioned the health belief model (Rosenstock 1974). This model describes the factors that influence and determine preventive healthcare behaviours. The perception of susceptibility, seriousness, benefits, and barriers associated with each health option is assumed to influence decision making. The health belief model appeared to have informed the DESI's early conception. The developers conducted focus groups where the health belief model was used to probe participants about their knowledge and feelings regarding prostate cancer screening. However, analysis of the associated publications did not provide any evidence of the transfer of the key theoretical constructs (i.e., susceptibility, seriousness, benefits, and barriers associated with each health option) into the development of the intervention. None of the related publications specified whether field-testing had been conducted. The evaluation seemed to be informed by the health belief model. The outcome measurements (i.e., knowledge, natural history of prostate cancer, perceptions of available

screening tests and intended screening behaviour) were related to all major dimensions of the health belief model.

### **2.3.6 Behavioural Theories**

An intervention combining a video and brochures for colon cancer screening (Pignone et al. 2000) referred to the transtheoretical model of behaviour change (Prochaska and Velicer 1997). This model of intentional change has taken constructs from 18 major theories of psychotherapy and behaviour change. Prochaska's model describes how people acquire or modify a behaviour using emotional, cognitive and behavioural components. It is organised in stages (i.e., the five stages of change): precontemplation, contemplation, preparation, action and maintenance. The early conception of the intervention was partially informed by the model. The colon cancer brochures were explicitly based on the transtheoretical model of behaviour change. However, the 11-minute educational video did not explicitly rely on theory. There was a lack of data as to whether the DESI had been field-tested. Based on the publications, there was no evidence that the evaluation had been explicitly informed by the transtheoretical model of behaviour change.

One DESI for prostate cancer treatment (Davison and Degner 1997) referred to the empowerment model (Conger 1989), derived from management and psychology theories. Empowerment is the process of enhancing individuals' belief in their self-efficacy and includes five stages (Conger and Kanungo 1988). When given sufficient decisional power, individuals are more likely to assume an active role in decision making and to achieve their desired outcomes. The empowerment model, thoroughly described by the DESI's authors, seemed to have informed the early conception of the intervention. However, the five stages of empowerment described in the model did not explicitly guide the prototype development. There was no evidence whether field-testing had been carried out. The evaluation of the



intervention was not explicitly informed by theory since none of the outcome measurements (i.e., sociodemographic variables, preferred roles, levels of anxiety and levels of depression) directly related to the process of empowerment.

To summarise, the early conception and preliminary design of 17 out of 50 DESIs were explicitly informed by theory. Further analysis revealed significant variations in the extent to which theory guided the prototype development, field-testing and evaluation of the interventions. The prototype development of 13 out of 17 interventions appeared to be informed by theory. However, the data analysis suggested important variations in the degree to which theoretical constructs were applied to the practical development of the intervention. The evaluation of 14 out of 17 DESIs was partially informed by theory and most likely reflected the difficulty to use and apply key theoretical constructs in a substantive manner. None of the 17 DESIs reviewed explicitly reported field-testing the intervention prior to evaluation. The transfer of key theoretical constructs into the design of DESIs is a subject that requires further attention (Elwyn et al. 2009d).

## **2.4 Discussion**

The analysis of 50 DESIs evaluated by randomised controlled trial revealed that only a third had described the contribution of decision making theories or models to their design, development and evaluation. All reviewed citations held little evidence that DESI developers were basing the intervention design, construction and evaluation on their chosen theoretical framework. Further analysis revealed that all theory-based interventions were evaluated without prior field-testing, therefore raising the issue of the validity and usability of interventions that have not been tested with patients or health professionals (Evans et al. 2007). It was also uncommon for outcome measurements to be based on theoretically derived hypotheses. The exceptions were DESIs based on subjective expected utility theory since this

theoretical framework provided an explicit architecture (i.e., decision tree) for the development and, to an extent, the evaluation of the intervention. However, the validity and appropriateness of decision analysis based interventions have been questioned. While decision analysis has been widely applied to treatment or screening decisions, substantial obstacles need overcoming for decision analytic interventions to be useful and adapted to clinical settings and patients' needs. The lack of theory-based outcome measurements needs to be seen in the wider context of DESI development and evaluation. It is worth noting that difficulties developing and validating widely accepted outcome measurements are inherent to this field. There is a lack of consensus on the criteria on which the efficacy of DESIs should be judged, and no measure is yet able to accurately assess decision quality (Kennedy 2003). Difficulties developing and validating accepted measures of DESIs' effectiveness may explain the aforementioned tendency to develop atheoretical outcome measurements. Further research is needed into exploring how relevant theoretical frameworks can guide the development of outcome measurements.

The analysis of all published material indicated that 66% of interventions included in this review did not explicitly rely on theories or models of decision making and could therefore be described as atheoretical interventions. Other conceptual frameworks might have informed the DESI's early conception but decision making theories were not mentioned in any publications reviewed. The assumption was therefore made that 33 out of 50 interventions (66%) were not informed by theory. The findings are consistent with previous research and confirm the tendency to develop and implement DESIs without solid theoretical underpinnings (Bekker et al. 1999; Bowen et al. 2006).

Strengths of this theoretical review were the quality of the sampling frame and inclusion criteria. All interventions were evaluated in a randomised controlled trial and included in a Cochrane review. It was therefore presumed that all interventions selected for review had been rigorously developed. All 78 citations were rated by 2 independent reviewers. Two limitations need to be considered. The sampling frame only included interventions evaluated in randomised controlled trials. Independent or commercial DESI developers were not included in this sample since it was assumed that highest quality interventions would have been submitted to evaluation. The bias generated by the specificity of the sampling frame, which could also be described as a methodological strength, could be addressed by including a wider range of interventions, produced by smaller developers and not evaluated in randomised controlled trials. A further limitation lies in the assumption that some DESI developers might have used theory in conceptualising the intervention but did not mention it in publications. However, the assumption was made that if theory had played a crucial role in developing the intervention, related publications would have specified how theory had informed its conception.

## **2.5 Conclusion**

This chapter draws attention to the difficulty to integrate theories or models of decision making into the prototype development and evaluation of DESIs. With the exception of subjective expected utility theory, the initial effort to use theoretical frameworks in the early stages of the DESI conception became impoverished when developing and evaluating prototypes. The lack of theoretical basis underpinning the development and evaluation of DESIs points to a paradox. Technologies intended to facilitate decision making processes do not build on key concepts of how individuals make decisions. However, theories or models of decision making do attempt to explain and predict how individuals make complex decisions. They describe the factors or situations likely to impair the decision making process and/or

lead to poor decision outcomes and decisional regret. It would therefore seem legitimate to integrate theoretical constructs into the development and evaluation of DESIs in the anticipation of significant impact on decision quality and outcomes. Using theories to develop DESIs may prove beneficial to decision makers provided appropriate theories are used and are correctly transferred into practice.

# **Chapter 3**

## **Amniocentesis Testing**

### **3.1 Introduction**

Amniocentesis is an invasive diagnostic procedure, involving complex information, potential harms, benefits and far-reaching consequences. Facing amniocentesis testing generally triggers heightened stress and anxiety, at a time of increased sensitivity (Ng et al. 2004; Robinson et al. 1984; Sarkar et al. 2006; Sarkar et al. 2008). There is no single best decision. Pregnant women and their partners are sole decision makers as to what is best for them and their baby. The decision to undertake amniocentesis testing is highly dependent on values and preferences and prone to high decisional conflict. As discussed in Chapter 1, DESIs designed to support decision making and facilitate the trade-off between options have been specifically developed for preference-sensitive decisions such as amniocentesis testing. Deciding about amniocentesis should be the result of an informed choice, determined by the expectant parents' preferences and attitudes to the risks involved and how possible harms and benefits are valued and evaluated (Marteau 1995). In addition, achieving informed choice is a cornerstone of the British National Health Service (NHS) and becomes especially relevant in decisions involving ethical considerations such as amniocentesis testing. Expectant parents ought to be informed about the benefits, potential harms and implications of amniocentesis before deciding whether or not to have the test. The aim of this chapter was to address the complexity and clinical characteristics of amniocentesis testing and to demonstrate why decision making could be facilitated by DESIs.

### 3.2 Screening for Down's Syndrome

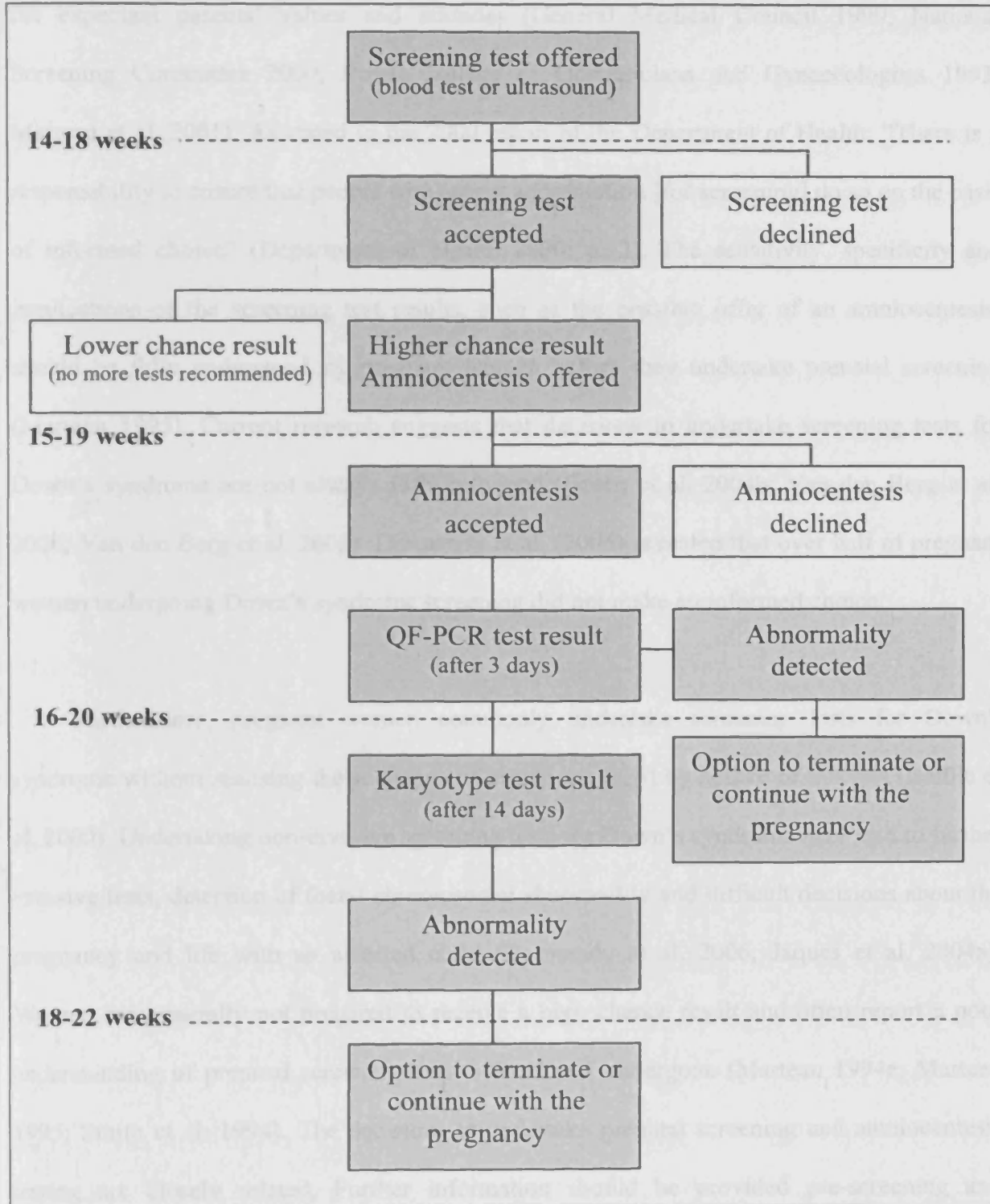
Over the past decades, advanced maternal age (e.g., >35) has been the most common indication for amniocentesis testing (Palo et al. 1994). In the United Kingdom, screening tests for Down's syndrome are currently offered as part of routine clinical practice to all pregnant women (Saller and Canick 2008), in order to determine their chance of foetal chromosomal abnormality. Prenatal screening tests for Down's syndrome will identify women with a higher risk of having a foetus with a chromosomal abnormality but cannot provide a diagnosis of chromosomal abnormality. Only invasive prenatal diagnostic tests (i.e., amniocentesis testing or chorionic villus sampling) are able to detect and diagnose the most common foetal chromosomal abnormalities. Prenatal screening tests for Down's syndrome include ultrasound scans (i.e., nuchal translucency scan) and maternal serum screening tests. Maternal serum screening tests (generally undertaken between 10 and 18 weeks of pregnancy) measure up to four biochemical markers in the blood (i.e.,  $\alpha$ -fetoprotein, oestriol, inhibin a, beta-hCG, human chorionic gonadotropin) and combine the blood test measures with maternal age, gestational age and weight in order to determine an adjusted chance of foetal chromosomal abnormality (Cate and Ball 1999). The nuchal translucency scan is a non invasive test based on ultrasound examination, usually performed between 10 and 13 weeks of pregnancy, to measure the thickness of fluid at the back of the baby's neck. Increased foetal nuchal translucency (i.e., thickness of fluid in the skin of the baby's neck) is associated with a wide range of chromosomal abnormalities including Down's syndrome (Nicolaidis 2004).

The NHS routinely offers second trimester maternal serum screening tests for Down's syndrome to all pregnant women between 14 and 18 weeks of pregnancy (Weisz and Rodeck 2006), with variations between England, Scotland, Wales and Northern Ireland. The nuchal

translucency scan is not routinely offered as part of the NHS screening programme but may be undertaken privately. The detection rates (i.e., detection of foetus with Down's syndrome) of maternal serum screening tests vary between 60% and 75% depending on the number of biochemical markers measured in the blood and there is a 5% false positive rate (Wald et al. 2003). The detection rate of the nuchal translucency scan is 72% with a 5% false positive rate (MacRae et al. 2008). The National Institute for Clinical Excellence (NICE) recommends that by April 2007, screening tests for Down's syndrome should provide a detection rate of 75% with a false positive rate lower than 3%. In Wales, screening tests for Down's syndrome are currently performed using the triple test, a maternal serum screening test measuring three biochemical markers in the blood (60% detection rate for a 5% false positive rate). In the NHS, to distinguish between high risk and low risk maternal serum screening test results, the cut-off of 1 in 250 is used (e.g., a result of 1 in 251 would be considered a low risk result and amniocentesis would not be offered). All women whose screening test result falls between 1 in 2 (50%) and 1 in 250 chance (0.4%) of having a baby with Down's syndrome will be offered to undergo amniocentesis testing (NHS Antenatal and Newborn Screening Programmes 2009). About 5% to 10% of women who undertake screening tests for Down's syndrome receive a high risk result and are offered to undergo amniocentesis testing to confirm the presence of abnormality (see Figure 3.1) (Benn et al. 2006; Gidiri et al. 2007).

The low predictive value of prenatal screening tests implies that a considerable number of women will be offered an amniocentesis while carrying a baby which does not have a chromosomal abnormality (i.e., false positive result). Conversely, women who receive a normal screening test result may carry a baby with a chromosomal abnormality (i.e., false negative result). It is generally accepted that undergoing screening tests (for Down's syndrome or any other condition) should be the result of an informed choice

Figure 3.1 Schematic Representation of the Prenatal Testing Process





commonly described as a decision based on comprehensive information and consistent with the expectant parents' values and attitudes (General Medical Council 1999; National Screening Committee 2000; Royal College of Obstetricians and Gynaecologists 1993; Marteau et al. 2001). As stated in the 2000 report of the Department of Health: "There is a responsibility to ensure that people who accept an invitation [for screening] do so on the basis of informed choice" (Department of Health 2000, p. 2). The sensitivity, specificity and implications of the screening test results, such as the possible offer of an amniocentesis, should be fully understood by pregnant women before they undertake prenatal screening (Marteau 1995). Current research suggests that decisions to undertake screening tests for Down's syndrome are not always fully informed (Green et al. 2004b; Van den Berg et al. 2006; Van den Berg et al. 2005). Dormandy et al. (2006) revealed that over half of pregnant women undergoing Down's syndrome screening did not make an informed choice.

Furthermore, pregnant women commonly undertake screening tests for Down's syndrome without realising the sequence of events triggered by uptake of this test (Baillie et al. 2000). Undertaking non-invasive screening tests for Down's syndrome may lead to further invasive tests, detection of foetal chromosomal abnormality and difficult decisions about the pregnancy and life with an affected child (Dormandy et al. 2006; Jaques et al. 2004a). Women are generally not prepared to receive a high chance result and often report a poor understanding of prenatal screening tests offered and undergone (Marteau 1994a; Marteau 1995; Smith et al. 1994). The decisions to undertake prenatal screening and amniocentesis testing are closely related. Further information should be provided pre-screening and informed choice achieved, to avoid heightened stress and anxiety when facing a decision to undertake amniocentesis (Green et al. 2004b).

### **3.3 The Amniocentesis Procedure and Chromosome Tests**

Amniocentesis is the most common prenatal diagnostic procedure undertaken in the United Kingdom. The procedure is generally performed after 15 completed weeks of pregnancy, to provide foetal cells for karyotyping (Abbott and Benn 2002; Evans and Wapner 2005). It is estimated that 5 to 10% of pregnant women in the UK are offered prenatal diagnostic procedures (i.e., amniocentesis or chorionic villus sampling). Chorionic villus sampling (CVS) is another method of obtaining foetal genetic material, performed in the first trimester of pregnancy (10-14 weeks), by withdrawing placental tissue rather than foetal cells from amniotic fluid (Brun et al. 2003). The CVS procedure is not routinely offered in the NHS and involves different risks (Caughey et al. 2006). For the purpose of this thesis, the focus is on amniocentesis testing.

The amniocentesis procedure consists of withdrawing 15 millilitres of amniotic fluid from the amniotic sac, in the uterus, under continuous ultrasound guidance. The procedure is reported to have a 1% risk of miscarriage although this may vary by operator (Gaudry et al. 2008; Odibo et al. 2008; Papantoniou et al. 2001). The best estimate of the rate of miscarriage following an amniocentesis is based on a randomised controlled trial conducted in 1986, among 4606 low risk women (Tabor et al. 1986). The miscarriage rate in the amniocentesis group exceeded the control group by 1%, which is the national figure normally quoted in counselling (Royal College of Obstetricians and Gynaecologists 2005). Amniocentesis may also be associated with a risk, albeit low and not quantified, of foetal trauma, rupture of membranes, foetal cutaneous lesions and maternal infections (Borrelli et al. 2006; Palo et al. 1994; Vilar Coromina et al. 2007).

It is estimated that between 40% and 80% of women who are offered an amniocentesis will undertake the test (Lesser and Rabinowitz 2001; Sharda and Phadke 2007; Sjogren and Uddenberg 1988). Following the procedure, chromosomal assessment will be performed on the amniotic sample. Traditionally, a full karyotype analysis is systematically performed on foetal cells after cell culture (Ogilvie 2003). It involves carefully examining the structure and number of all chromosome pairs, and usually takes a minimum of 10 working days (Warburton 1991). A karyotype analysis can identify aneuploidy such as trisomy 21, 18 or 13 but may also detect other abnormalities such as exchange of material between chromosomes. The classic karyotype analysis will not detect changes in single genes, microdeletions and other small changes in chromosomes. Since 1980, the karyotyping procedure has been the gold standard of prenatal diagnosis worldwide. Where minor chromosomal abnormalities are detected after karyotype analysis, the question arises as to what threshold termination of pregnancy should be considered. While the karyotype test identifies most chromosomal abnormalities, it cannot provide information about the phenotypic consequences or severity of the abnormality diagnosed. The severity of a particular chromosomal abnormality such as Down's syndrome for example, is extremely variable and cannot be predicted. Women should be informed about the range of chromosomal abnormalities tested for and uncertainty accompanying the diagnosis. Given the number and complexity of chromosomal abnormalities potentially detected, the question arises as to how much information is too much information? In other words, when deciding about amniocentesis, is there a need for extensive information about all abnormalities tested for, or would this lead to information overload?

A recent development in Wales, and elsewhere in the UK, is the implementation of a rapid genetic test; Quantitative-Fluorescent Polymerase Chain Reaction test (QF-PCR test),

which provides results for the three most common chromosomal abnormalities: Down's, Edwards' and Patau's syndromes in 3 working days (Levett et al. 2001; Mann et al. 2008). The National Screening Committee recommends that the QF-PCR test alone is performed on all amniotic samples following a higher screening test result, as happens in England. In Wales, both QF-PCR and karyotype tests are systematically performed. Women who undertake amniocentesis testing in Wales will receive a result for Down's, Patau's and Edwards' syndromes after 3 working days and the full karyotype result after two weeks. Pregnant women considering an amniocentesis should be aware that after a normal QF-PCR result, the karyotype analysis may detect further abnormalities. The implementation of the QF-PCR test and detection of a wider range of chromosomal abnormalities in different timeframes requires an updated approach to information provision.

When an abnormality is found, expectant parents will have to decide between continuing the pregnancy and preparing for the birth of a baby diagnosed with a genetic abnormality or ending the pregnancy (Asch 1999; Pryde et al. 1993; Verp et al. 1988; Yilmaz et al. 2008). Termination rates following an amniocentesis vary. A study examining the determinants of parental decisions revealed that 93% of couples with severe foetal prognosis (e.g., trisomy 21, 18, 13) terminated the pregnancy while 27% of couples with questionable prognosis (e.g., mosaic 45, x, sex chromosome trisomy) opted for a termination (Drugan et al. 1990). The presumed or perceived severity of the chromosomal abnormalities was a determining factor of parental decision to terminate the pregnancy. Before deciding to undertake amniocentesis, women, and their partners, should be informed about the range of abnormalities tested for, and option to terminate the pregnancy as this may have a significant impact on their decision and post-decision outcomes (i.e., regret, cognitive dissonance and decisional conflict) (Priest et al. 1998).

### **3.4 Existing Information and Decision Support Arrangements**

In the literature, the information, decisional needs and psychological impact of invasive diagnostic procedures such as amniocentesis have rarely been documented (Marteau 1995; Rostant et al. 2003; St-Jacques et al. 2008). Further, the few studies examining decisional and informational issues surrounding prenatal testing have not specifically addressed the decision to undertake amniocentesis (Potter et al. 2008). The decision to undertake screening and diagnostic tests have commonly been confounded. There is limited research on how best to provide information on amniocentesis testing. A qualitative study on the perspectives of physicians and pregnant women with regards to amniocentesis testing revealed that the information provided to women did not address their own interests (Hunt et al. 2005). Pregnant women and physicians reported divergent concerns and approaches to the amniocentesis decision. Physicians were concerned with following pre-established communication strategies or protocols and provided explanations about the relative risks of amniocentesis and characteristics of the tests. Pregnant women were primarily concerned about coping with stress and anxiety and protecting the pregnancy.

In 1995, a review of informed decision about prenatal testing highlighted the lack of understanding of women undergoing or declining prenatal tests (screening and diagnostic tests) (Marteau 1995). This finding is consistent with a recent systematic review of women's decisional needs in the diagnostic phase of pregnancy, which confirmed that pregnant women considering prenatal testing generally lacked information (St-Jacques et al. 2008). Further research suggested that the risks associated with prenatal testing and the range of abnormalities tested for were particularly misunderstood (Cederholm et al. 1999). A study of women's knowledge about prenatal testing revealed that 35% of women undertaking diagnostic tests did not mention (when asked in a questionnaire) that amniocentesis was

testing for Down's syndrome (Jaques et al. 2004). In addition, expectations regarding amniocentesis or prenatal testing are often unrealistic (Marteau 2002). A study of amniocentesis related pains showed that expected pain and anxiety levels before the test were significantly higher than pain and anxiety levels reported post-procedure (Ferber et al. 2002).

Furthermore, the information women receive when offered amniocentesis testing is complex, specialised and potentially overwhelming. The amniocentesis decision generally involves heightened stress and anxiety (Sun et al. 2008; Susanne et al. 2006). Research suggests that maternal stress in women facing amniocentesis testing is higher than the norms of psychiatric and female surgical patients (Johnston 1980; Robinson et al. 1984) and could be associated with poor outcomes (i.e., gestational complication, foetal growth retardation) for the mother and foetus (Glover et al. 2008; Reading 1983; Talge et al. 2007). Understanding the risks (e.g., risk of miscarriage or chromosomal abnormality) and processing complex probabilistic information and numerical data at a time of increased sensitivity is difficult. To achieve decision making, most women will need to balance the risk of miscarriage (1 %) against their individual risk of foetal chromosomal abnormality, based on the screening test results (1 in 250 or more) (Gidiri et al. 2007; Saller and Canick 2008). The risk of miscarriage is expressed out of a 100 and the screening test result may be anything between 1 in 2 to 1 in 250 chance of having a baby with Down's syndrome. For instance, for a screening test result of 1 in 250 (0.4%), the risk of miscarriage (1%) is higher than the risk of having a baby with Down's syndrome. Women facing a decision to undertake amniocentesis should be able to accurately balance the risk of miscarriage against the screening test result. Comparing probabilities with multiple denominators at a time of heightened stress may be difficult and overwhelming (Quagliarini et al. 1998). Difficulties comprehending probabilistic information in the diagnostic phase of pregnancy have been

documented previously (Kuppermann et al. 2006; Pilnick et al. 2004). In addition, research shows that individual differences in processing numerical information exist and significantly affect performance (Booth and Siegler 2006). The ability to process complex numerical information significantly decreases under high-pressure or stress conditions (Beilock and DeCaro 2007). Finally, discussing the implications of amniocentesis testing and abnormalities potentially detected requires expert genetic knowledge. There is documented evidence that health professionals who counsel women about amniocentesis testing do not always have sufficient specialised knowledge to provide specific but simple information about genetic testing and chromosomal abnormalities tested for (Hunt et al. 2005; Williams et al. 2002a).

### **3.5 Summary**

This chapter discussed the complexity of the screening process, information provided, risks and implications associated with amniocentesis testing. Over 30,000 pregnant women in the UK every year face a decision to accept or decline amniocentesis testing (Benn et al. 2006; Gidiri et al. 2007). This decision involves uncertainty, far reaching consequences and complex probabilistic information that has to be weighed against the couple's values and attitudes, at a time of heightened stress and anxiety. Considering amniocentesis testing also involves understanding the possible consequences and limitations of the chromosome tests.

Current research highlights the lack of information and understanding of women undertaking or declining prenatal testing for Down's syndrome. The associated risks and range of chromosomal abnormalities tested for are particularly misunderstood. Informed choice is not systematically achieved and existing information and decision support is not always responsive to women's needs and concerns. There is scope for developing interventions capable of addressing their information needs, providing decision support and alleviating emotional strain experienced in the diagnostic phase of pregnancy.

# Chapter 4

## Review of Decision Support Interventions for Amniocentesis

### 4.1 Introduction

The decision to undertake amniocentesis testing is often associated with a lack of information and understanding of the test purposes and consequences. Deciding about amniocentesis should be determined by the expectant parents' awareness of the purposes of the test, their attitudes to the risks involved and how harms and benefits are valued (Hunt et al. 2005; St-Jacques et al. 2008). Difficult decisions such as these should involve parents in choosing the option that is consistent with their knowledge, values and preferences. To achieve these goals, DESIs intended to support individuals who face difficult health decisions for them or others in their families have been developed.

As highlighted in Chapter 1, DESIs' characteristics and effectiveness have been examined in several systematic and interpretative reviews, therefore highlighting the variable effect and outcome measurements used to assess their efficacy (O'Connor et al. 2001; Molenaar et al. 2000; Estabrooks et al. 2001). DESIs have been developed using a variety of formats, purposes and timeframes to address a wide array of preference-sensitive decisions: breast cancer surgery, PSA testing, hormone replacement therapy etc. To date, DESIs for amniocentesis have not been formally reviewed nor evaluated. The aim of this chapter was to identify, describe and assess the quality and effectiveness of DESIs for amniocentesis and examine their use and implementation in clinical practice. To meet the stated aims, the review was organised around three questions:

- 1. How many DESIs for amniocentesis exist and what are their aims?**
- 2. Do DESIs for amniocentesis meet published quality standards?**



### **3. What is the effectiveness of DESIs for amniocentesis?**

## **4.2 Methods**

### **4.2.1 Definitions**

For the purpose of this chapter, DESIs were defined as: “Interventions designed to help people make specific and deliberative choices among options by providing information on the options and outcomes relevant to a person’s health status” (O'Connor et al. 2006, p. 2). Based on the Cochrane review (O'Connor et al. 2006) and International Patient Decision Aids Standards instrument (IPDASi), four essential criteria allowing the distinction between information leaflets and DESIs were identified. Accordingly, a DESI should:

1. State the decision to be addressed and deliberated upon;
2. Provide information about the options, their harms, benefits and the associated probabilities of the decision outcomes;
3. Enable patients to express and clarify their values, attitudes, preferences in regard to the decision;
4. Provide structured guidance in achieving decision making (step-by-step way to make a decision).

Two out of four criteria (criteria 1 and 2) were used in selecting the DESIs for review. Therefore, interventions that explicitly addressed the decision to have amniocentesis and provided information about the harms, benefits and outcomes probabilities associated with each option were included in the review.

### **4.2.2 Literature Search Strategy**

Ten electronic databases were searched until June 2009: Medline (1966-2009), Medline In-Process, PubMed (2009), Embase (1980-2009), British Nursing Index (1994-Jun 2009), CINAHL (1982-Jun 2009), all EBM Reviews (2009), PsycINFO (1806-2009), Science

Citation Index Expanded (1970-2009), Social Sciences Citation Index (1970-2009). A list of key words and subject headings (MeSH words in PubMed) was written in Ovid and run in each database (see Table 4.1). DESIs for amniocentesis where reports had not been published in peer-reviewed journals or had not been evaluated in a trial, were identified through manual check of reference lists from published papers, internet search and manual check of the A-Z list of decision aids developed by the Ottawa Health Decision Centre (Ottawa Hospital Research Institute 1996). All major DESI developers such as the Ottawa Health Decision Centre, Healthwise, Mayo Clinic, Midwives Information and Resource Service (MIDIRS), Foundation for Informed Medical Decision Making (FIMDM) and Intelihealth (Harvard Medical School) were contacted.

#### **4.2.3 Study Inclusion and Exclusion Criteria**

Studies were included if they (i) considered DESIs that focussed on the decision to undertake amniocentesis (regardless of age and pathway of entry); or (ii) considered the decision to undertake amniocentesis as well as other prenatal screening tests or other available diagnostic tests such as CVS. Only interventions that could be classified as DESIs, as opposed to information leaflets, were included in this review (see Definitions section). Studies were excluded if they (i) considered DESIs that exclusively focussed on prenatal screening tests or CVS without addressing the decision to undertake amniocentesis; (ii) addressed a choice between amniocentesis and CVS (see Figure 4.1).

#### **4.2.4 DESI Assessment**

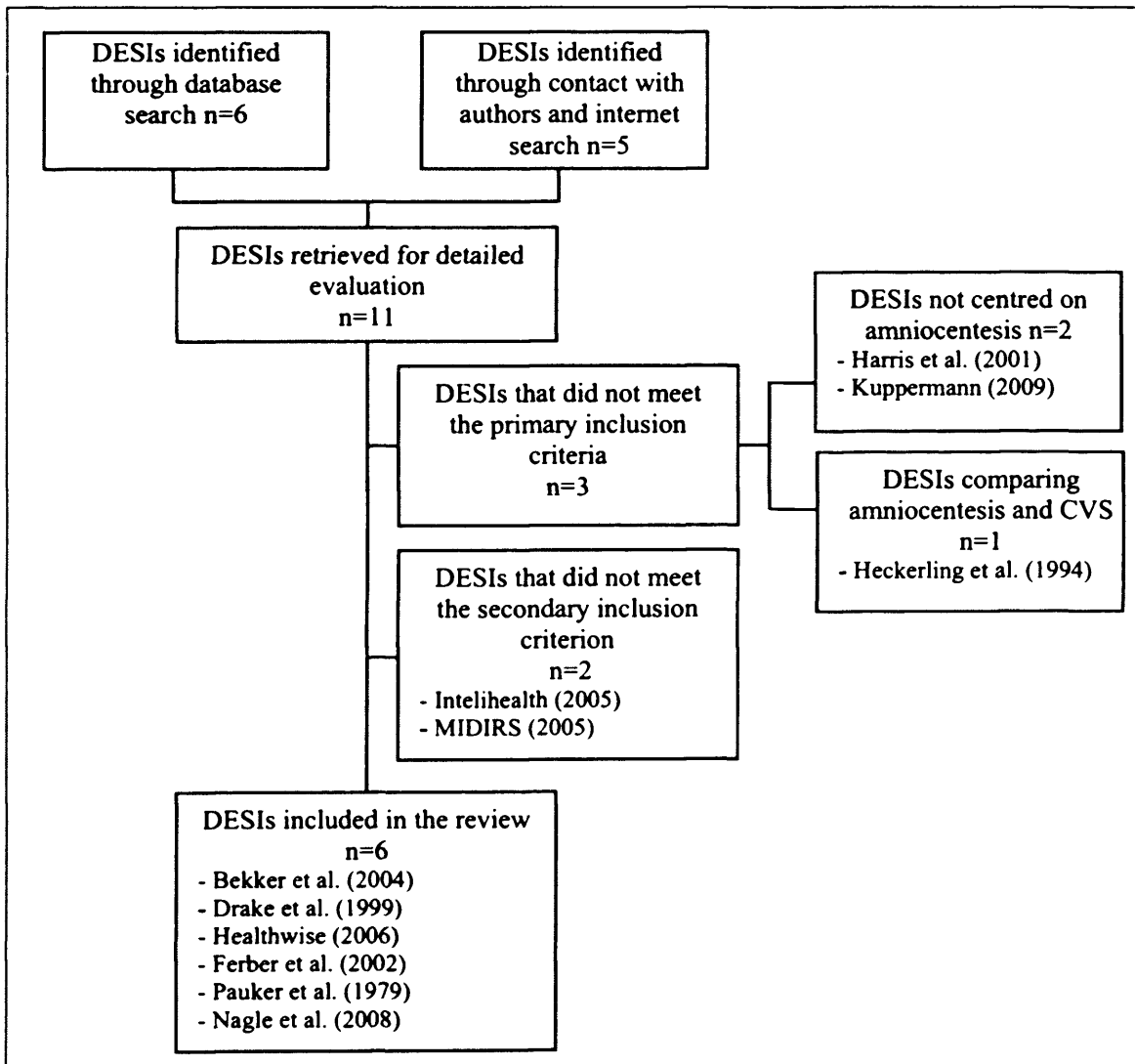
After having identified interventions that met the inclusion criteria, DESI developers were contacted by email to obtain a copy of the intervention and information on its current use and implementation in clinical settings. Information about (i) the DESI characteristics, aims, and current use, (ii) the DESI quality against published standards and (iii) efficacy, was

Table 4.1 Literature Search Strategy

Amniocentesis <sup>1</sup>	Decision Support Interventions
<b>Amniocentesis</b> <b>Prenatal diagnosis</b> <b>Antenatal diagnosis</b> <b>Prenatal testing</b> <b>Antenatal testing</b>	<b>Decision support technique</b> <b>Patient decision aid</b> <b>Decision aid</b> <b>Decision explorer</b> <b>Decision tool</b> <b>Decision support</b> <b>Decision\$</b> <b>Decision making</b> <b>Software</b> <b>Decision support systems</b> <b>Computer assisted</b> <b>Information systems</b> <b>Computer assisted decision support systems</b> <b>Genetic counselling</b>

<sup>1</sup> All terms in the first column were combined with terms in the second column

Figure 4.1 Selection Process of Decision Support Interventions Included for Review



collected to address the aforementioned research questions. First, a content analysis was performed to determine the specific features, the aims, the current use and implementation of each intervention. Second, the DESIs were rated against the IPDASi instrument (IPDASi) domains to assess the quality of essential components (see Appendix 1). The number of published DESIs has tripled since 1999 (O'Connor et al. 2007a) and there is a growing concern that development has been independent of relevant theoretical frameworks. However, the IPDASi domains do not include an item on the contribution of theory to the DESI development. Therefore, associated publications were independently examined and DESI developers were contacted to ascertain the theoretical underpinnings of each DESI. Third, the efficacy of the DESIs was determined by assessing evaluation methods and impact on decision outcomes.

The IPDASi ([www.ipdasi.org](http://www.ipdasi.org)) was developed and validated by an international group of researchers working to assess the quality of DESIs (Elwyn et al. 2009c; Elwyn et al. 2006). The author was trained to perform IPDASi ratings before assessment. IPDASi is a set of 47 quality criteria (or items) addressing 10 domains that should ideally be covered in a DESI: Information, Test, Probabilities, Values, Guidance, Development, Evidence, Disclosure, Plain Language, Evaluation. The Information domain assesses the quality of information provided on the decision at stake, the options available and the positive and negative features of each option (8 items). The Test domain assesses the extent to which specific features of the diagnostic or screening investigation (e.g., rate of false positive or false negative results) are described in the intervention (9 items). The Probabilities domain examines how probabilistic information is presented and framed (8 items). The Values domain assesses whether the intervention facilitates the expression and clarification of the expectant parents' values with regards to the decision (5 items). The Guidance domain examines the extent to which the

intervention provides structured guidance in helping expectant parents achieve decision making (2 items). The Development domain evaluates the quality of the DESI development process by specifically looking at the involvement of patients and professionals, use of field test and expert review (6 items). The Evidence domain assesses the quality of the research evidence used in developing the intervention (5 items). The Disclosure domain appraises the transparency of the funding and author disclosure (2 items). The Plain Language domain assesses the DESI's clarity and readability levels (1 item). Finally, the Evaluation domain assesses the impact of the intervention on decision outcomes (1 item). Each item was rated on a scale from 1 = strongly disagree to 4 = strongly agree. Domain and total IPDASi percentage scores were calculated, first, by summing relevant items and then dividing by the number of items per domain, in order to account for the unequal number of items per domain. The IPDASi is enclosed in Appendix 1.

## **4.3 Results**

### **4.3.1 Selection of DESIs**

The literature search and contact with authors identified 11 interventions. After assessment of their content and/or available publications, five interventions were excluded. Three interventions did not focus on the decision to undertake amniocentesis. The intervention by Heckerling et al. was excluded since it compared amniocentesis testing and CVS (Heckerling et al. 1999; Heckerling et al. 1994). The intervention by Harris et al. focussed on prenatal screening tests without specifically addressing the decision to undertake amniocentesis and was therefore excluded (Harris et al. 2001). The intervention by Kuppermann et al. was excluded as it offered a comparison between prenatal screening tests and diagnostic tests without specifically addressing the decision to undertake amniocentesis (Kuppermann et al. 2009). Two interventions were classified as information leaflets and

excluded from the review after content analysis revealed that the interventions did not meet the criteria one and two of a DESI (see Definitions, p. 52) (InteliHealth 2005; MIDIRS 2005).

#### **4.3.2 How Many DESIs for Amniocentesis Exist and What Are Their Aims?**

Six DESIs for amniocentesis were examined in the review: (1) a decision analytic consultation by Bekker et al. (Bekker et al. 2004), (2) an intervention developed by Drake et al. combining an audiotape and a booklet entitled: “Making choices: prenatal testing” (Drake et al. 1999), (3) “the amniocentesis report”, a booklet downloaded from the internet produced by Ferber et al. (Ferber and Sicherman 2001), (4) a web-based DESI for amniocentesis developed by the Healthwise group entitled: “Should I have an amniocentesis?” (Healthwise 2006), (5) a DESI for prenatal testing developed by Nagle et al. (Nagle et al. 2008), and (6) a decision analytic model developed by Pauker et al. (Pauker and Pauker 1979, Pauker and Pauker 1987). Three out of six interventions were developed in the USA (Ferber and Sicherman 2001; Healthwise 2006; Pauker and Pauker 1979), one in Canada (Drake et al. 1999), one in Australia (Nagle et al. 2008) and one in the United Kingdom (Bekker et al. 2004). Two out of six interventions were available on the internet (Ferber and Sicherman 2001; Healthwise 2006) although one of the DESIs’ availability was subject to payment (Ferber and Sicherman 2001). Based on the Ottawa A to Z inventory and contact with authors, it was ascertained that two of six DESIs were used (2008) in clinical settings: Pauker’s decision analysis consultation (Pauker and Pauker 1979) and the intervention for prenatal testing of foetal abnormalities developed by Nagle et al. (Nagle et al. 2008). At the time of assessment, Pauker’s decision analysis consultation was used in routine genetic counselling at Harvard Vanguard Medical Associates across eastern Massachusetts, USA. The DESI by Nagle et al. was used by maternity care clinicians as part of a state-wide education programme in Victoria, Australia. The interventions are listed in Table 4.2 and described according to their name, decision considered, format, use and location, theoretical

Table 4.2 *Characteristics of Decision Support Interventions for Amniocentesis Included for Review*

<b>Authors</b>	<b>Title</b>	<b>Decision</b>	<b>Format, use and location</b>	<b>Theoretical framework</b>	<b>Outcome measures</b>
Bekker et al. (2004)	Decision analysis consultation	Prenatal diagnostic testing (amniocentesis and CVS)	Routine consultation structured by decision analysis, used in an additional information consultation offered to women after a high chance maternal serum screening test result for Down's syndrome. DESI developed in the UK.	Expected utility theory	Consultation length Informed decision making Test choice
Drake et al. (1999) Hunter et al. (2005)	Making choices: prenatal testing	Prenatal testing: - Maternal serum screening - Ultrasound scan - Amniocentesis + CVS	Workbook and audiotape, used in a genetic counselling consultation, including a discussion with a genetic counsellor. DESI developed in Canada.	Ottawa decision support framework	Knowledge Anxiety Decisional conflict Intervention satisfaction
Ferber and Sicherman (2001)	The amniocentesis report, decision guide for expectant parents and healthcare professionals	Amniocentesis testing	Web-based DESI <a href="http://www.amniocentesis.org">www.amniocentesis.org</a> DESI can be downloaded in PDF format online or shipped worldwide. Minimal fee: \$5.05. DESI developed in the USA.	No theory	No evaluation
Healthwise (last update 2006)	Should I have an amniocentesis?	Amniocentesis testing	Web-based DESI <a href="http://www.webmd.com/baby/should-i-have-an-amniocentesis">www.webmd.com/baby/should-i-have-an-amniocentesis</a> Open Access (free of charge). DESI developed in the USA.	No theory	No evaluation
Nagle et al. (2006)	A decision aid for prenatal testing of foetal abnormalities	Prenatal testing: -Maternal serum screening -Second trimester ultrasound scan, - Amniocentesis + CVS	24-page booklet DESI given to women in early pregnancy by their GP. DESI developed in Australia.	Ottawa decision support framework	Informed choice Decisional conflict Anxiety Depression Attitudes to the foetus/pregnancy Satisfaction with the DESI
Pauker and Pauker (1979)	A decision analytic model to counsel parents about amniocentesis	Amniocentesis testing	Routine consultation structured by decision analysis. Method used in a routine genetic counselling session for prenatal diagnosis. DESI developed in the USA.	Expected utility theory	Assessed cost of elective abortion Assessed cost of spontaneous abortion Actual decision Decision suggested by decision analytic model

framework and outcome measurements (when applicable). The DESIs varied in their content and approach.

The decision analysis consultation developed by Bekker et al. was based on Pauker and Pauker's decision analytic model and designed to help parents clarify and express their values (Bekker et al. 2004). The consultation was articulated around the use of a decision tree displaying test options and consequences, and a lottery technique designed to facilitate the trade off between options by eliciting the maximum utility (the “goodness” of each option and consequences). The lottery technique consisted of asking questions to compare the options (accepting or declining amniocentesis) on each attribute (e.g., chance of having a baby with Down's syndrome) by varying the gamble figure: “If we told you the chance of the baby having Down's syndrome was fifty per cent, and the chance of the baby not having Down's syndrome was fifty per cent, would you choose to carry on with or terminate the pregnancy?” (Bekker et al. 2004, p. 267). Finally, a graph combining the expectant parents' best utility and the results of the screening test was used to identify the option with the highest expected utility. From a rational standpoint, the option with the highest expected value should correspond to the best possible option.

The DESI by Drake et al., making a choice: prenatal testing, provided information about maternal serum screening tests, ultrasound scans, CVS and amniocentesis testing. The aim of the intervention was to improve knowledge, decrease decisional conflict, and decrease anxiety levels associated with prenatal testing. It consisted of a 35-page illustrated workbook, a 45-minute audiotape and a worksheet. The worksheet gave expectant parents the opportunity to clarify the reasons for undertaking or declining prenatal tests and provided a concrete basis for discussing the options with health professionals.



The amniocentesis report by Ferber et al., a decision guide for expectant parents and healthcare professionals, was a 16-page booklet providing structured information on amniocentesis testing, its potential risks and implications (Ferber and Sicherman 2001). The amniocentesis report was designed to provide unbiased information on amniocentesis testing in order to help expectant parents make an informed decision. The intervention was divided into six sections: (1) What is amniocentesis and how is it done, (2) Nature and accuracy of the amniocentesis results, (3) What are the benefits of amniocentesis, (4) What are the costs of amniocentesis (relevant to the United States), (5) Making the amniocentesis decision and (6) Alternative procedures.

The web-based DESI produced by Healthwise was an interactive website entitled: Should I have an amniocentesis? (Healthwise 2006). The intervention was designed to help expectant parents understand their choices regarding amniocentesis testing. The intervention was divided into four sections: (1) an introduction to amniocentesis testing, (2) medical information about the amniocentesis procedure, harms and benefits, (3) a section comparing the reasons to accept or decline an amniocentesis and (4) a worksheet for patients to clarify their ideas and values about amniocentesis testing.

The DESI for prenatal testing by Nagle et al. was a 24-page booklet containing graphic design elements (i.e., diagram, images, charts and dot points) and information about maternal serum screening, second trimester ultrasound scan, CVS and amniocentesis testing (Nagle et al. 2008). The intervention was designed to assist women in making an informed choice about amniocentesis and to reduce decisional conflict. It provided information on the reasons for being offered prenatal testing, the range of prenatal tests available and the results

and implications of each test. The intervention included scenarios of pregnant women's experiences, a worksheet to weigh up the pros and cons of each option and a list of additional information resources available. Finally, it included a risk report sheet presenting the risks estimates of having a baby affected with Down's syndrome, based on the expectant mother's age and gestation.

The decision analytic DESI developed by Pauker and Pauker was used to counsel parents about amniocentesis testing during the consultation and required the assistance of a physician (Pauker and Pauker 1979). The intervention was designed to help parents assess their values and attitudes about the outcomes of options and make a logical decision about amniocentesis (guided by decision analysis). The decision analytic model was used during the consultation and involved a lottery technique where the following outcomes were considered: miscarriage, detection of chromosomal abnormality and being faced with diagnostic errors. Prospective parents were asked to assign a utility (on a scale from 0 to 100) to the potential outcomes of both available options: undertaking or declining amniocentesis.

#### **4.3.3 Do DESIs for Amniocentesis Meet Published Quality Standards?**

Five out of six DESIs were rated against all IPDASi domains to assess their quality (see Table 4.3). The evaluation of the prenatal testing booklet developed by Nagle et al. was exclusively based on the analysis of published papers since the developers declined to provide a copy of the intervention.

First, the quality of the information provided and scores on the IPDAS instrument varied according to the type of information assessed (Information domain, Test domain, Probabilities domain, and Plain Language domain). Scores on the Information domain (i.e., information about the index decision and options available) reached 73.7% on average. Most

Table 4.3 Average IPDASi Ratings of Decision Support Interventions for Amniocentesis

<b>IPDASi Domains</b>	<b>Decision analysis consultation</b> Bekker et al. (2004)	<b>Making choices: prenatal testing</b> Drake et al. (1999) Hunter et al. (2005)	<b>The amniocentesis report</b> Ferber and Sicherman (2001)	<b>Should I have an amniocentesis?</b> Healthwise (last update 2006)	<b>A decision analytic model</b> Pauker and Pauker (1979)	<b>Average scores per domain</b>
<b>Information</b>	68.7 <sup>2</sup>	75.0	65.6	84.4	75.0	<b>73.7</b>
<b>Test</b>	55.5	63.9	55.5	50.0	83.3	<b>61.6</b>
<b>Probabilities</b>	65.6	56.2	43.7	40.6	68.7	<b>55.0</b>
<b>Values</b>	55.0	75.0	35.0	55.0	45.0	<b>53.0</b>
<b>Guidance</b>	100.0	100.0	50.0	100.0	50.0	<b>80.0</b>
<b>Development</b>	58.3	50.0	45.8	33.3	66.7	<b>50.8</b>
<b>Evidence</b>	80.0	60.0	40.0	95.0	55.0	<b>66.0</b>
<b>Disclosure</b>	75.0	75.0	62.5	87.5	62.5	<b>72.5</b>
<b>Plain language</b>	50.0	50.0	25.0	50.0	25.0	<b>40.0</b>
<b>Evaluation</b>	100.0	100.0	25.0	25.0	50.0	<b>60.0</b>
<b>Average score per DESI</b>	<b>70.8</b>	<b>70.5</b>	<b>44.8</b>	<b>62.1</b>	<b>58.1</b>	

<sup>2</sup> Scores on the IPDAS instrument in percents

DESI for amniocentesis showed very little variation regarding the provision of standard information and positive and negative features of options. Scores on the Test domain (evaluating the specific features of a diagnostic test) were on average lower (61.6%) than scores on the information domain. Three DESIs did not include full information on the false positive and false negative results associated with the tests and scored significantly lower on this domain than other interventions evaluated (Bekker et al. 2004; Ferber and Sicherman 2001; Healthwise 2006). Scores on the Probabilities domain reached 55% on average. Most DESIs provided little information about the outcome probabilities associated with the options. Scores on the Plain Language domain reached 40% on average. Most interventions did not use plain language throughout and did not report readability levels.

Second, the IPDASi evaluation revealed score differences in domains assessing the guidance provided to clarify values and achieve decision making. The average score on the Values domain was 53%. Three interventions only explicitly enabled expectant parents to express and clarify their values (Bekker et al. 2004; Drake et al. 1999; Healthwise 2006). In contrast, average scores on the Guidance domain reached 80%. Three DESIs reached the highest score by providing a step-by step-way to make a decision and worksheets designed to structure the expectant parents' decision making process (Bekker et al. 2004; Drake et al. 1999; Healthwise 2006).

Third, IPDASi scores concerning the process of DESI development and evaluation were variable. On the Development domain, scores reached 50.8% on average. The development process rarely involved the target population: pregnant women (and their partners) who had been offered an amniocentesis. According to the IPDAS standards, the development process should ideally involve (i) a needs assessment with expectant parents and

relevant professionals, (ii) a patient and experts review of the intervention, and (iii) field-testing before evaluation. Most DESIs were based on literature reviews, expert reviews by researchers or occasionally health professionals, and omitted expectant parents' needs assessment or review. Finally, only two out of six DESIs were explicitly field-tested with pregnant women who had been offered an amniocentesis (Drake et al. 1999; Nagle et al. 2008). Scores on the Evidence domain (assessing the scientific validity of the intervention) reached 66% on average. The development of three DESIs was explicitly based on research evidence (i.e., citations to studies used) (Bekker et al. 2004; Drake et al. 1999; Healthwise 2006). Scores on the Evaluation domain reached 60% on average. Four interventions were evaluated in a trial, including the DESI by Nagle et al., not otherwise rated on the IPDAS instrument (Bekker et al. 2004; Drake et al. 1999; Nagle et al. 2008; Pauker and Pauker 1979). Two DESIs were reported to help patients make a decision that was ideally consistent with their values and preferences (Bekker et al. 2004; Drake et al. 1999). Finally, most interventions acknowledged funders/sponsors and contributors. The average score on the Disclosure domain was 72.5%.

Since the IPDAS instrument does not assess the contribution of theory to the DESI design and development, the theoretical underpinnings of all DESIs selected for review were independently assessed. Four out of six DESIs mentioned the contribution of a theoretical framework in developing the intervention (Bekker et al. 2004; Drake et al. 1999; Nagle et al. 2008; Pauker and Pauker 1987). Operationalised as decision analysis, expected utility theory guided the design and use of two DESIs for amniocentesis (Bekker et al. 2004; Pauker and Pauker 1987). Two interventions relied on the Ottawa decision support framework (Drake et al. 1999; Nagle et al. 2008). As described in Chapter 3, the Ottawa decision support framework combines social support and cognitive psychology theories such as the

expectancy value model (Fishbein and Ajzen 1975), prospect theory (Kahneman 2003) and the conflict theory model of decision making (Janis and Mann 1977). There is as yet no evidence about the influence that theory-based design has on outcomes. However, two out of three theory-based DESIs had higher IPDASi scores (70.8% and 70.5%) than interventions that did not rely on theories or models of decision making (Bekker et al. 2004; Drake et al. 1999).

#### **4.3.4 What is the Effectiveness of Existing DESIs?**

Three out of six DESIs were evaluated using randomised controlled trials (Bekker et al. 2004; Hunter et al. 2005; Nagle et al. 2006). One intervention was evaluated in a pilot study (Pauker and Pauker 1979). The decision analysis consultation by Bekker et al. was evaluated in a randomised controlled trial of 117 pregnant women offered amniocentesis testing and randomised to a routine consultation or a decision analysis consultation. The risk perception of the screening test result, subjective expected utility (generated by the decision analytic method), knowledge of prenatal tests for Down's syndrome, consultation quality, decisional conflict (O'Connor 1995) and anxiety (Marteau and Bekker 1992) were measured. Informed decision making was also measured by applying a coding frame to the interview transcripts (Bekker 2003). The intervention reduced decisional conflict, improved informed decision making, and led to a more realistic evaluation of information. However, the decision analysis consultation did not significantly impact on consultation satisfaction, knowledge or anxiety when compared to the control group. The latter finding is consistent with the results of similar evaluations (O'Connor et al. 2001).

The DESI by Drake et al. was evaluated in a randomised controlled trial of three counselling methods for prenatal diagnostic testing (Hunter et al. 2005) and one before and after study (Drake et al. 1999). Hunter's randomised controlled trial of three counselling

methods for prenatal diagnosis (i.e., individual counselling, group counselling and use of a DESI) assessed the following outcome measures: knowledge (Goel et al. 1996), decisional conflict (O'Connor 1995), anxiety (Spielberger et al. 1970) and satisfaction with intervention (Shiloh et al. 1990) in a sample of 350 women (and 225 partners) who had been offered prenatal diagnosis testing because of advanced maternal age (Hunter et al. 2005). Compared to other counselling methods, the DESI was least efficient at improving knowledge, although knowledge increased compared to the pre-counselling phase. The DESI did not significantly diminish state anxiety. However, scores on the Decisional Conflict Scale were significantly reduced. The satisfaction with the DESI was high but not higher than alternative counselling methods (e.g., group counselling). Second, Drake's before and after study evaluated the DESI's impact on knowledge (Goel et al. 1996), decisional conflict (O'Connor 1995), anxiety (Spielberger et al. 1970) and intervention acceptability (Barry et al. 1995) in a sample of 21 women (and 17 partners) (Drake et al. 1999). The DESI was reported to have significantly increased knowledge and decreased decisional conflict scores but did not modify state anxiety level. The findings were not compared with a control group.

The DESI by Nagle et al. was evaluated in a cluster randomised controlled trial where 55 general practitioners were randomised to provide women (n=338) with the DESI (i.e., intervention group) or a pamphlet (i.e., control group) (Nagle et al. 2008). The following outcome measures were assessed: informed choice (Marteau et al. 2001), decisional conflict (O'Connor 1995), anxiety (Marteau and Bekker 1992), depression (Cox et al. 1996), attitudes to the foetus/pregnancy (Reading et al. 1984) and satisfaction with the DESI or pamphlet. The results showed that more women made an informed choice when given the DESI than when given the pamphlet. The satisfaction with the intervention was significantly higher in the intervention group (i.e., use of DESI). Decisional conflict scores were low in both

intervention and control groups. There were no significant differences on the secondary outcomes: anxiety, depression or attitudes to the pregnancy/foetus.

The decision analysis intervention by Pauker et al. was evaluated in a pilot study with 90 women (and 35 partners) who were offered an amniocentesis (Pauker and Pauker 1979). The evaluation consisted of assessing women's or couples' attitudes towards elective abortion and spontaneous abortion (i.e., miscarriage after an amniocentesis). The expected subjective utility (generated by the decision analytic method, based on their assessed attitudes), and actual decision to accept or decline amniocentesis testing were examined. Most pregnant women made a decision that was consistent with their attitudes towards miscarriage and elective pregnancy termination. However, choices made by some couples conflicted with their stated values. The findings showed that the final decision was, in general, not consistent with the choice suggested by decision analysis. The reliability of the findings could be questioned by the absence of a control group.

Finally, two DESIs were not evaluated in a trial but low scores on IPDASi suggest domains that could be improved (Ferber and Sicherman 2001; Healthwise 2006) (see Table 4.3). The amniocentesis report scored the lowest on IPDASi (total adjusted score: 44.8%).

#### **4.4 Discussion**

Six DESIs for amniocentesis were identified. Their quality was variable across IPDASi domains with lower scores on the Probabilities, Values, Development and Plain Language domains. The evaluations in randomised controlled trials or before and after studies had considerable scope for improvement. Only a small proportion of DESIs for amniocentesis were used and implemented in clinical settings. This reflects the emergent nature of the field



of shared decision making and DESI development, and highlights the need for rigorously developed interventions.

Existing DESIs for amniocentesis represent a small proportion of the total number of interventions developed in other healthcare contexts worldwide (over 500) (O'Connor et al. 2007a). There may be several reasons to account for the scarcity of DESIs for amniocentesis. First, the amniocentesis decision may not be perceived as generating as much anxiety and decisional conflict as screening or treatment decisions for directly life-threatening conditions. However, there is ample evidence that amniocentesis generates peak levels of anxiety at a time of heightened sensitivity (Glover et al. 2008; Sarkar et al. 2006; St-Jacques et al. 2008) and that maternal stress may be associated with poor outcomes for the mother and foetus (i.e., gestational complication, foetal growth retardation) (Nakamura et al. 2008; Reading 1983). Second, the difficulty to assess the decision making process surrounding amniocentesis testing may account for the small proportion of interventions available. Only 5 to 10% of women who undertake prenatal screening tests will be offered an amniocentesis (Benn et al. 2006; Gidiri et al. 2007). Given heightened levels of stress and anxiety, approaching women at the early stage of the decision making process to inform DESI development may be difficult.

Findings of this review revealed that existing DESIs for amniocentesis were barely used or implemented in routine clinical practice. Interventions that were primarily developed by researchers (i.e., lack of user involvement) might be unable to meet the practical requirements and decision support needs of patients and professionals who are expected to use those interventions. The lack of user involvement in DESI development is a plausible explanation to recurrent implementation difficulties encountered in this field (Holmes-Rovner

et al. 2000; Silvia et al. 2008). A systematic review of barriers and facilitators to DESI implementation suggested that the lack of applicability between shared decision making interventions, patients' characteristics and the clinical situation, was an important barrier to implementation (Gravel et al. 2006). Elwyn et al. used the normalisation process model (May et al. 2007) to illustrate the influence of principal stakeholders (patients, physicians, managers) and their respective knowledge, in using and implementing DESIs (Elwyn et al. 2008). These results point to the need to involve physicians and patients at all stages of development in order to produce interventions that have better goodness-of-fit with the clinical situations they intend to support.

The IPDASi evaluation emphasised the variable quality across interventions and domains. Most DESIs were effective and reliable information resources (see IPDASi scores on the information domain). They provided adequate information on the amniocentesis decision, on the features of a diagnostic test, guided expectant parents in making a decision and used scientific evidence. However, the communication of outcome probabilities, the expression and clarification of values, the development process, the evaluation and the use of plain language could be significantly improved. Lower scores on those domains may reflect the complexity and specialisation of the domains' requirements combined with the recent development, inexperience and implementation difficulties in the field of shared decision making (and DESI development). This analysis subsequently revealed that most interventions had a theoretical origin, a finding inconsistent with existing DESI reviews (Bekker et al. 1999; Bowen et al. 2006).

The DESIs' evaluations in trials of varying size and methods revealed poor quality evaluations and pointed to the difficulty to assess DESIs' effectiveness. The match between

their stated goals and the results of the evaluation was poor. Most DESIs were shown to facilitate information processing but failed to reduce emotional burdens associated with the amniocentesis decision (e.g., anxiety) and did not systematically increase knowledge. This may reflect a growing tendency to develop DESIs in short timeframes and promote their use on the internet without rigorously evaluating their impact on decision making outcomes. This raises concerns as to the use of poor quality interventions by expectant parents who are expected to make high stake decisions at a time of considerable emotional upheaval.

Systematic reviews conducted in other healthcare contexts corroborate our findings (Evans et al. 2005; Volk et al. 2007; Williams et al. 2008). Systematic reviews of prostate cancer screening revealed that DESIs increased knowledge but did not impact on other decision outcomes nor reduced emotional burdens (Evans et al. 2005; Volk et al. 2007). A systematic review of interactive decision aids for breast cancer genetic testing identified a small number of poor quality interventions which had rarely been evaluated and implemented in clinical settings (Williams et al. 2008). The IPDASi scores of DESIs for breast cancer genetic testing reached lowest scores on the same domains as DESIs for amniocentesis (i.e., communication of outcome probabilities, value expression and clarification, development process and evaluation).

## **4.5 Conclusion**

This chapter discussed the quality, effectiveness and implementation of existing DESIs for amniocentesis testing. Compared with other healthcare contexts (e.g., breast cancer, heart disease), little attention has been given to the decision to undertake amniocentesis testing. However, the complexity of the information provided, the high stakes of the decision, and associated emotional strain emphasise the need for high quality DESIs. The DESIs' assessment against the IPDASi domains suggested that the development process, presentation

of probabilistic information and elicitation of patients' values could be significantly improved. Only one DESI for amniocentesis had been developed in the UK and was not used or implemented in clinical settings. Further, the majority of DESIs included in this review were developed without patients' and professionals' involvement (e.g., needs assessment, field-testing). Interventions that are developed for a specific group of users (e.g., pregnant women considering amniocentesis testing) should be tailored to their information and decision support needs. There is therefore room for developing a DESI for amniocentesis tailored to the UK practice and policies, using a systematic development process and adapted theoretical framework.

# Chapter 5

## User Perspective

### 5.1 Introduction

Pregnant women facing a decision to undertake amniocentesis generally report peak levels of stress and anxiety (Sarkar et al., 2006). As noted previously, between 5 to 10% of pregnant women in the UK will face a decision to undertake amniocentesis. Assessing the needs of a small proportion of pregnant women at a time of acute stress and sensitivity may prove difficult. To date, research on how best to provide information on amniocentesis testing is limited and associated information and decisional needs have rarely been documented (Marteau 1995; St-Jacques et al. 2008; Hunt et al. 2005). Studies investigating information and decision needs associated with prenatal testing have not specifically addressed the decision to undertake amniocentesis testing (St-Jacques et al. 2008). The amniocentesis decision has generally been confounded with the decision to undertake prenatal screening tests. Although those decisions are closely related, amniocentesis is an invasive procedure that involves risks, far reaching consequences, complex information and uncertainty. The information and decision support needs associated with this procedure differ from those associated with prenatal screening tests. There is therefore scope for evaluating specific information and decision support needs associated with the amniocentesis decision.

The quality criteria produced by the IPDAS collaboration specify that the DESI development should include a needs assessment with individuals who are currently facing the decision. In the clinical context of amniocentesis testing, conducting users' need assessment, also known as user perspective, involves examining the specific decision needs of women who have been offered amniocentesis testing. Assessing user perspectives while they face the

decision is essential in determining the content of the intervention and ensuring that women's basic information and decision support needs are addressed. The aim of this chapter was to assess pregnant women's information and decision support needs associated with amniocentesis testing.

## **5.2 Methods**

### **5.2.1 Participants**

A qualitative study design was adopted using semi-structured interviews with pregnant women who had been offered amniocentesis testing. Pregnant women were identified and approached by midwives or screening midwives in two participating antenatal clinics (University Hospital Wales and Llandough Hospital, Cardiff). In addition, to ensure that the views of women who had experienced chromosomal abnormality (after having accepted or declined an amniocentesis) were represented, an advert was posted in the journal of the Down's Syndrome Association. The advert described the study and invited women who had been offered amniocentesis testing to take part in a phone interview, by contacting the researcher (author of this thesis) to ask any questions they had about the project and to agree a telephone interview date. In the antenatal clinics, women (any age) who had been offered an amniocentesis after screening tests for Down's syndrome, advanced maternal age or mid-pregnancy ultrasound scan, were informed of the study by midwives. Information leaflets describing the study were distributed by the midwives during the counselling session where women were offered an amniocentesis. Women interested in taking part gave verbal agreement for their contact details to be passed onto the research team. They were later contacted by a researcher to answer any questions they had about the project and to agree an interview date. Pregnant women were not invited into the study if they had been offered another diagnostic test, such as CVS, since this procedure involves different risks. In the journal of the Down's Syndrome Association, the research advert was directed to all women

who had been offered an amniocentesis (whether or not they accepted the test) and who received a diagnosis of chromosomal abnormality. The study protocol and materials were reviewed and approved by the research and development committees of the participating sites, by the School of Psychology Ethics Committee (Cardiff University) and by the National Research Ethics Service.

### **5.2.2 Data Collection**

Interviews with pregnant women were carried out in the antenatal clinics, at the participant's home or over the phone, from May 2007 to February 2008. The semi-structured interview schedule consisted of 13 open-ended questions exploring women's experience of the amniocentesis decision, their information and decision support needs and how information and risks should be framed to facilitate understanding. Their reasons for accepting or declining amniocentesis, and their attitudes (e.g., satisfaction, regret, misunderstanding etc) following the decision were examined. Special attention was paid to new topics emerging such as the difficulties women faced in making their decision, the influence of others (e.g., partners, family) or the satisfaction/dissatisfaction with the information and support provided (see Appendix 2 for interview schedules). All interviews were recorded digitally and transcribed by the same researcher (author of this thesis).

### **5.2.3 Data Analysis**

The transcribed interviews were coded using a two-step thematic content analysis derived from descriptive phenomenology (Denzin and Lincoln 2000; Holloway 2005; Pope et al. 2000), assisted by the computer software ATLAS-ti (ATLAS-ti 5.2). First, the transcripts were coded to identify information, decision support and emotional needs. In a second and more detailed analysis, the interview transcripts were coded according to all the themes discussed in the interviews, including spontaneously emerging themes. Similar codes were merged and subsequently grouped into families of codes and networks. Six interview

transcripts chosen for being representative of the overall sample, were coded by two independent raters (M-A D and MS) in order to ensure reliability of coding and to agree the themes and family of codes for all remaining interview transcripts. Discrepancies among raters were discussed until agreement was reached.

### **5.3 Results**

In the participating antenatal clinics, 18 women who had recently been offered an amniocentesis were recruited and 12 agreed to be interviewed. The reasons for declining the interview were the impossibility of making an appointment after the counselling session (n=4), the lack of time (n=1), and the stress and anxiety associated with this decision (n=1). Participants were interviewed in the antenatal clinics (75%) or at the participant's home (25%). Most women attended the interview alone (66.7%) and four women came with their partners (33.3%). Participants were interviewed within two weeks after they had been offered to undertake amniocentesis. Among women who were approached through the journal of the Down's Syndrome Association, seven women were recruited and five took part in a telephone interview. Two women had been offered CVS (first trimester diagnostic test) and were therefore excluded from the study. Women were interviewed between one and seven years after having been offered amniocentesis testing. Interviews lasted between 10 and 50 minutes (23 minutes on average).

In total, 17 pregnant women who had been offered amniocentesis took part in the study. Ten women decided to undergo amniocentesis and seven declined the test. Among women who undertook amniocentesis, seven women received a normal result and three women received a diagnosis of Down's syndrome. Among women who received a diagnosis of foetal chromosomal abnormality, one pregnant woman out of three decided to terminate the pregnancy. Five out of seven women who declined an amniocentesis had a healthy baby and



two women had a baby with Down's syndrome. The mean age of women in the sample was 36.4 years (range 28-47 years, standard deviation 6.6). Most women were British (n=15), one was Turkish and one was Indian. The demographic characteristics of the participants are summarised in Table 5.1.

### **5.3.1 Information Needs**

Twelve out of 17 participants described the amniocentesis experience as a stressful, complex and upsetting decision making process. For those who undertook prenatal screening, the high chance result and subsequent offer of an amniocentesis was a shock, generating intense strain and anxiety.

“They rang me at 8:30 at night and told me that the screening gave me a result of 1 in 10 which for somebody of my age should have been 1 in 600. I was completely shocked, obviously, I didn't expect anything like this and I didn't even know what Down's syndrome was.” (F, age 33, declined amniocentesis)

Five out of 17 participants were satisfied with the overall information and decision support provided. All remaining participants (n=12) expressed various unmet information and decision support needs (see Table 5.2).

Most pregnant women felt that more detailed information about the risks involved; the risk of miscarriage, the risk factors for miscarrying and other associated risks (e.g., infections, long term consequences) should be provided. They reported the need for increased consistency regarding the miscarriage rate as it was misleading to be given different percentages. The national miscarriage rate is one in a 100 procedure (1%) but most antenatal clinics will quote a local rate, generally lower than the national rate but based on limited scientific evidence (Gaudry et al. 2008; Tabor et al. 1986). Regarding the overall quantity of information provided, women's opinions diverged. Some women experienced information

Table 5.1 *Characteristics of Pregnant Women Interviewed (User Perspective)*

<b>Characteristics</b>	<b>Sample size</b>
Amniocentesis	
- Accepted	10
- Declined	7
Marital status	
- Married	15
- Cohabiting	2
Number of existing children	
- 0	6
- 1	8
- 2	3
Existing children with a chromosome disorder	2
Obstetric History	
- Previous miscarriage	2
- Previous amniocenteses	2
- In vitro fertilisation pregnancy	1
Outcome post-amniocentesis in n=10 women who had the test	
- Miscarriage	0
- Normal result	7
- Down's syndrome diagnosed	3
- Termination of pregnancy	1
Outcome when amniocentesis declined (n=7)	
- Miscarriage	0
- Healthy baby	5
- Baby with Down's syndrome	2

Table 5.2 *Themes Identified in Interviews With Pregnant Women (User Perspective)*

<b>Themes</b>	<b>Sub-themes</b>
Information needs	<ul style="list-style-type: none"> <li>- Information about the risks</li> <li>- Information about the procedure</li> <li>- Information about the screening tests</li> <li>- Personalised information</li> </ul>
Emotion and decision support	<ul style="list-style-type: none"> <li>- Heightened stress and anxiety</li> <li>- Addressing emotional difficulties</li> </ul>
Reason	<ul style="list-style-type: none"> <li>- Reasons for accepting an amniocentesis</li> <li>- Reasons for declining an amniocentesis</li> </ul>
Decision making process	<ul style="list-style-type: none"> <li>- Deciding with a partner</li> <li>- Outcomes (e.g., satisfaction, regret)</li> </ul>

overload while others lacked information and actively looked for further information elsewhere (e.g., internet, books).

“I found particularly that when I was going to the appointments you get so much information bombarded that you can’t get it all in.” (F, age 28, undertook amniocentesis)

“The information I had clearly wasn’t enough because I did search on internet. I didn’t have enough for me to make a definitive decision in my mind. I did have to go away and do a bit more research.” (F, age 36, declined amniocentesis)

Regarding the procedure, participants were generally satisfied with the quantity of information provided but would have liked more information about the results, consequences and implications of an amniocentesis (i.e., abnormalities detected, termination of pregnancy). Women reported difficulties understanding what the test may or may not detect and lacked information about the characteristics and timeframes of each test (i.e., PCR test and karyotype test).

“We were a bit confused about what the second test was for, because I thought originally that the first test, showed you pretty much whether it was ok or not, but actually it wasn’t, it was a definite no for Down’s syndrome but I couldn’t work out what the second test was for.” (F, age 32, undertook amniocentesis)

Prior to undertaking prenatal screening for Down’s syndrome, 8 out of 17 women would have liked more information about the screening tests available, their purpose, the uncertainty associated with the results, and the implications of a high chance result (i.e., amniocentesis testing offered). Three women who undertook prenatal screening and were subsequently offered amniocentesis regretted their screening decision and blamed it on a lack of information pre-screening test. They did not expect to receive an increased risk result.

“For me, that was a terrible rollercoaster, and I wish I’d never even had the blood test. So I do feel that before you even have the blood test, more information should be given. Don’t have the blood test if you don’t know the rest of the consequences.” (F, age 35, declined amniocentesis)

Most women interviewed wished to receive information tailored to their individual needs and presented in multiple ways to account for individual differences (e.g., educational levels, ethnic backgrounds, culture).

I personally found that just having the figures was enough, that was fine, you could work it out on the figures. The midwife did show us some graphs, you know with coloured dots and so forth which I'm sure would be useful for other people as well cause everybody visualises these things differently, don't they?" (F, age 36, declined amniocentesis)

Women interviewed felt that both verbal and written information should be provided as the stress and anxiety experienced during the counselling session significantly limited their capacity to assimilate and recall complex information. Most participants believed that probabilistic information would be better understood if framed in multiple ways, using diagrams, flow charts, percentages and frequencies with identical denominators to facilitate the comparison between the risk of chromosomal abnormality and the risk of miscarriage. Visual elements such as images, videos (e.g., video of the amniocentesis procedure) should be made available to women, provided viewing remains optional. Five participants expressed the need to know about other women/couples' experiences, to be informed about support groups or reliable internet forums.

"It might be an idea if perhaps; you could even see an amniocentesis procedure, so you know the whole stage of what is involved. And I think the kind of consequences of having it, because you are still going to be faced with a situation of: Right, ok now, am I going to do something about it or am I just going to live with the information till the baby is born? I think it would be good if you had people's experiences of the whole process and how they dealt with it." (F, age 35, declined amniocentesis)

While unmet information needs were identified, most women reported satisfaction with the counselling provided and interaction with their healthcare professionals.

### **5.3.2 Emotions and Decision Support**

Most women reported heightened stress and anxiety. The emotional stress and worry experienced between the offer of an amniocentesis and the results of the chromosome tests, (or until the birth, for women who declined an amniocentesis) were reported to fluctuate but never disappeared. Peak levels of anxiety were reported immediately after a high risk screening test result or offer of an amniocentesis, and when waiting for the chromosome tests results. Nine out of 17 women experienced great difficulties dealing with overwhelming stress, anxiety or regret while being pregnant, when they never anticipated facing such a

difficult decision. In addition, amniocentesis related anxiety seemed exacerbated by the increased sensitivity most women experienced at this stage of the pregnancy.

“It is a very stressful time, a very worrying time and I think that perhaps people who deal with it every day don’t realise what the average person is going through.” (F, age 28, undertook amniocentesis)

“I was really upset. I was told about the figure which was 1 in 220, which I understand is quite a low risk but sometimes I don’t think you see that anyway, you just think of Down’s syndrome, what would I do next basically, even though it was still a small risk, I was still really worried and it was a horrible horrible experience.” (F, age 34, undertook amniocentesis)

Most women felt that emotional difficulties should be addressed and more decision support made available through support groups or relevant charities. Some women felt that decision support could also be provided through discussion with their partner, family, friends, or healthcare providers. Three women reported that unbiased decision support should be provided whatever the decision may be. Two women felt forced into having an amniocentesis and received little support, if not disapproval, for declining the test or continuing the pregnancy. Three women felt that health professionals lacked neutrality regarding disability and pregnancy termination.

“It was all negative at the time, I have either got a Down’s syndrome baby that I may have to abort or I am going to have a miscarriage of a healthy baby. That was all I could see at that point.” (F, age 32, undertook amniocentesis)

“My experience has been on both occasions that you are expected to screen for abnormality and do something about it, which isn’t always how people think, not how I think. It would go against my belief really, to do that. In my experience, there are a lot of judgements made.” (F, age 47, declined amniocentesis)

### **5.3.3 Reasons for Accepting/Declining an Amniocentesis**

In total, 24 reasons to accept or decline amniocentesis were reported. Amongst 10 women who undertook amniocentesis, six participants chose to have the test to find out if the baby had a problem and avoid recurrent stress and anxiety for the rest of the pregnancy. Five women opted for an amniocentesis as they felt incapable or unwilling to look after a disabled child. Three out of 10 women undertook the test to have the option to terminate the

pregnancy if a problem was found. Three women had an amniocentesis to prepare for the birth of a baby with a chromosomal abnormality. Three out of 10 women decided to have the test as they already had children and were concerned about the impact of a disabled child on siblings. Women's reported reasons for or against an amniocentesis are presented in Table 5.3.

All women who declined an amniocentesis (n=7) based their decision on the risk of miscarriage and conviction that they would not terminate the pregnancy if a problem was found. Additional reasons for declining the test were medical complications such as bleeding during the pregnancy or twin pregnancy (n=2), previous obstetric history such as an in vitro fertilisation (IVF) pregnancy or difficulties getting pregnant (n=2) and the risk of miscarriage being higher than the risk of chromosomal abnormality (n=2).

#### **5.3.4 Making a Decision**

When deciding about amniocentesis, four out of 17 women disagreed with their partners.

"My husband was very keen for me to have the test. Over the time, I decided I didn't really want this test. So, it was very difficult, it did cause conflicts between us because obviously, at the end of the day, it is my body and I don't want to be in a position where I am feeling guilty, if anything happened. So that was a difficulty, it really was." (F, age 35, declined amniocentesis)

All women who decided to have an amniocentesis, including women who received a diagnosis of chromosomal abnormality, reported no regret. Despite weeks of constant worry, none of them regretted having had an amniocentesis.

"I am happy that we went ahead with it, the results were clear so we got reassurance from that. Because up until that point, I don't feel that we could have started planning adequately for the baby." (F, age 39, undertook amniocentesis)

Women whose amniocentesis results showed Down's syndrome (n=3) felt positive about their decision, as the test enabled them to prepare for the birth of a disabled child or to

Table 5.3 *Pregnant Women's Reasons Influencing Decision Making About Amniocentesis*

<b>Reasons reported by pregnant women</b>	
<i>Reasons for accepting amniocentesis</i>	<i>Reasons for declining amniocentesis</i>
- To find out if the baby has a problem (stress of not knowing)	- To avoid anything that may harm the baby (risk of miscarriage)
- Capacity/willingness to look after a disabled child	- View on termination (would not terminate the pregnancy if problem was found)
- To have the option to terminate pregnancy	- Capacity/willingness to look after a disabled child
- To prepare if a problem is found	- Medical complication/Obstetric history
- Existing children	- Previous miscarriage
- Family history of chromosome disorder	- Difficulty getting pregnant (IVF pregnancy)
- Risk of miscarriage compared to risk of a problem	- Risk of miscarriage compared to risk of a problem
- Knowledge and/or experience of children with Down's syndrome	- Knowledge and/or experience of children with Down's syndrome
- Partner's views	- Partner's view
- Age	- Age
- Obstetrician's expertise in conducting amniocenteses	- Religious beliefs
- Views of friends, family	- Views of friends, family
- Adjusted risk compared to risk in similar age group	- Adjusted risk compared to risk in similar age group
- Difference between the woman's screening result and screening cut-off limit	- Difference between the woman's screening result and screening cut-off limit
- Previous amniocentesis	- Previous amniocentesis
- Existing child with Down's syndrome	- Existing child with Down's syndrome
- Anomalies detected on the mid-pregnancy scan	- Practical reasons (husband away, unable to rest for a few days)

terminate the pregnancy. All women who decided not to have an amniocentesis were satisfied with their decision and did not experience regret. However, most women reported recurrent anxiety regarding the risk to give birth to a baby with a chromosomal abnormality.

"I feel that I've made the right decision. I suppose I am a little bit nervous. It is a worry I've got to be honest with you, it's gonna be 6 months of worry thinking what if." (F, age 39, declined amniocentesis)

### **5.3.5 Synthesis**

Most women reported significant emotional and cognitive difficulties making a decision about amniocentesis testing (i.e., stress, anxiety, difficulty assimilating probabilistic information and information overload). They felt that the provision of better services would reduce the emotional challenges of this period. They unanimously highlighted the need to address decision difficulties by providing personalised and interactive information, presented in multiple ways (e.g., numbers, diagrams, videos, women's experiences), in order to account for individual differences in processing complex information. Most women felt that emotional difficulties should be addressed and that decisional and emotional support should be made available through support groups, relevant charities, or other interventions. Women wished to receive comprehensive information about the risks involved, the results of the chromosome tests and potential consequences of an amniocentesis. The majority of women interviewed wished to be informed about the termination of pregnancy. Finally, they highlighted the need to provide detailed information about amniocentesis before the screening tests, as existing information and consent arrangements appeared insufficient.

## **5.4 Discussion**

The findings revealed pregnant women's unmet needs for information, decision and emotional support when deciding about amniocentesis testing. Significant decision making difficulties were reported (e.g., difficulty assimilating probabilistic information, information overload). Pregnant women also highlighted elevated stress and anxiety that was triggered by the decision and its aftermath, and expressed the need for reinforced emotional and decisional support.



While a minority of women were satisfied with the overall information and support provided, most participants expressed the need for personalised and interactive information (e.g., images, video clips or forums). Potter et al. (2008) described the importance of women's values and of different types of knowledge in deciding about prenatal testing. Pregnant women highlighted the need for testimonials from others facing a similar dilemma. Previous studies suggested that watching or hearing experiences of women who have made a decision about prenatal testing and have experienced different outcomes (e.g., healthy baby, miscarriage, detection of chromosomal abnormality) was considered beneficial in making a decision (Moyer et al. 1999; St-Jacques et al. 2008). In addition, women believed that information should be framed in multiple ways to address individual differences in processing complex information and to facilitate understanding. Difficulties understanding probabilistic information and making sense of the risks have been extensively documented (Kuppermann et al. 2006; Howe et al. 2000). Pilnick et al. (2004) pointed to the difficulties and stress experienced by women attempting to make sense of the nature and significance of screening test results. They concluded that difficulties understanding risks figures and probabilities may be eased by adequate non-directive counselling. Further research suggested that the way in which risk information was framed influenced decisions to accept or decline amniocentesis testing (Marteau 1989). Marteau established that pregnant women were more likely to have an amniocentesis if the risk of foetal chromosomal abnormality was framed negatively (i.e., 5% or 1 in 20 chance of having a baby with Down's syndrome) rather than positively (i.e., 95% chance that there is no abnormality).

Further, the majority of women interviewed wished to receive comprehensive information about the risks involved and implications of amniocentesis (i.e., termination of pregnancy and chromosome abnormalities potentially detected). Research showed that health

professionals counselling women about prenatal testing generally focussed on the prenatal testing process rather than on the condition being screened or tested for (Williams et al. 2002a). Observations of routine antenatal consultations revealed that information about the range of abnormalities tested for was generally not provided by obstetricians (Marteau et al. 1993). Further research showed that elective termination of pregnancy was not routinely mentioned or discussed with women who had been offered an amniocentesis (Bernhardt et al. 1998).

Pursuant to prenatal testing policies in the UK, accepting or declining amniocentesis should be the result of an informed choice. Nonetheless, several participants felt pressured into undertaking prenatal testing. They considered that health professionals' attitudes lacked neutrality and non-directiveness. This finding is consistent with previous study of prenatal testing decisions, where participants pointed to health professionals' pressure to undertake the tests (Potter et al. 2007). Further research highlighted health professionals' tendency to shape women's understanding and uptake of prenatal screening and diagnostic tests (Press and Browner 1997). A questionnaire and interview study of 211 women undergoing amniocentesis or CVS indicated that most participants found it difficult to decline prenatal diagnostic tests when offered (Sjogren and Uddenberg 1988). While women did not generally report external pressures to undertake prenatal diagnostic tests, they highlighted the difficulty to opt out, once amniocentesis or CVS had been offered. This may be related to social pressures to conform to normality and use technologies which are increasingly routinised and presented as non-controversial (Sjogren and Uddenberg 1987). Since the technology exists and is widely available, there is an implicit pressure to undertake those tests (Ettorre 2000). Pregnant women may also feel that undertaking prenatal testing is part of their parental duty and societal responsibility to engender non-diseased and genetically normal off springs.

The decision to undertake amniocentesis was associated with recurrent stress and anxiety. Heightened anxiety was reported after a higher chance screening test result and prior to receiving amniocentesis results, as has been documented previously (Beeson and Golbus 1979; Cederholm et al. 2001; Michelacci et al. 1984; Sun et al. 2008; Susanne et al. 2006). Women consistently reported the need for further emotional support. Emotional difficulties associated with prenatal testing have been extensively documented and are consistent with the present finding (St-Jacques 2008; Susanne et al. 2006). Emotional difficulties and ways of coping with extreme anxiety may be addressed in a DESI, by providing contact details of relevant support groups and charities or by enabling women to communicate with others in a similar situation (e.g., message board). In addition, several women reported difficulties agreeing with their partners, which in turn, triggered increased stress and anxiety. A study of women's attitudes towards prenatal diagnostic procedures showed that 38% of women interviewed reported divergent opinions between partners (Potter et al. 2008). Finally, several women reported regretting the decision to undertake screening tests for Down's syndrome. Press and Browner (1997) revealed that 85% of women considering prenatal diagnostic tests could not precisely articulate the reasons for undertaking prenatal screening tests for Down's syndrome. Further research suggested that pregnant women undertaking screening tests for Down's syndrome did not fully comprehend the potential consequences and implications of the test (Baillie et al. 2000). A lack of understanding of the consequences and reasons for undertaking prenatal screening may lead to increased emotional stress and regret in the case of an increased risk of foetal chromosomal abnormality and subsequent offer of invasive diagnostic tests.

The risk of miscarriage was the most often reported reason for declining an amniocentesis while finding out if the baby had a problem was the most common reason for

undertaking the test, as has been reported previously (Cederholm et al. 1999; Priest et al. 1998). In the literature, the most commonly reported reason for undertaking prenatal testing is maternal age (Cederholm et al. 1999; Kukulcu et al. 2006; Moyer et al. 1999). Given screening tests for Down's syndrome are routinely offered to pregnant women in the UK, factors such as the influence of existing children, the possibility of having options if a problem is found and pregnant women's views on termination or on disability, had a stronger impact on women's decisions than maternal age.

The strengths of this study were the heterogeneity of the sample and semi-structured format of the interviews. The interview sample included women who declined the test, women who received normal and abnormal amniocentesis results, women with experience of chromosomal abnormality or pregnancy termination. The structure of the interview gave women freedom to broaden the themes of the interview schedule while still focussing on the decision making process.

Limitations of the study were the differences between the sample of patients recruited in the antenatal clinics and recruited through the Journal of the Down's Syndrome Association. Women recruited through the Down's Syndrome Association were interviewed between one and seven years after having been offered an amniocentesis. The proportion of women receiving a diagnosis of chromosomal abnormality was therefore higher than in the general population. In addition, the passage of time, involving a possible change in clinical practice and provision of information, is likely to introduce biases. Finally, women recruited through the journal were self-selected from a specific population whereas women recruited consecutively in the clinic were systematically approached by screening midwives, and this may have introduced biases.

## **5.5 Conclusion**

This chapter presented a qualitative assessment of information and decision support needs of women facing amniocentesis testing, with the objective to develop a DESI for amniocentesis. The majority of women interviewed highlighted information and decision support needs which could be addressed in a web-based intervention: neutral, balanced and interactive information, testimonials from women who faced a similar dilemma, probabilistic information framed in multiple ways etc. Participants expressed the need for further information pre-screening as several women admitted regretting their screening decision. The offer of an amniocentesis and subsequent test results were often associated with heightened stress and profound worry. Women did not anticipate to be offered an amniocentesis and were generally unprepared to face a decision with far reaching consequences and uncertain outcomes. Greater attention should be diverted to unmet emotional and decision support needs as they appear from these interviews, to be as important as unmet information needs. There is scope for developing interventions that provide non-directive and evidence-based information but also address emotional and decisional difficulties, with the aim to enable pregnant women to make an informed choice; one that is consistent with their values and preferences.

# **Chapter 6**

## **Stakeholder Analysis**

### **6.1 Introduction**

Deciding whether or not to undergo amniocentesis is a complex and highly distressing decision, often undertaken with little systematic decision support. DESIs have been developed to support individuals when they face complex healthcare decisions such as amniocentesis testing. According to the IPDAS collaboration, the first step in developing DESIs is a needs assessment with health professionals from relevant disciplines (Elwyn et al. 2006). In the clinical context of amniocentesis testing, conducting a needs assessment, also known as stakeholder analysis, involves examining the professionals' evaluation of women's information and decision support needs associated with amniocentesis testing. Stakeholders include all health professionals who inform and counsel women about amniocentesis testing or have an in depth understanding of the prenatal testing process. While a stakeholder analysis does not replace the direct assessment of potential users' needs (i.e., needs assessment with women facing amniocentesis testing), it offers a general overview of commonly reported needs. It also involves assessing the counselling needs and difficulties professionals may experience when advising women about amniocentesis testing. Such analysis is essential in ensuring that the DESI's content is clinically accurate, consistent with professionals' daily practice, and therefore acceptable in conjunction with existing counselling.

With a view to develop a DESI for amniocentesis testing, the aim of this chapter was to examine health professionals' evaluation of women's information and decision support

needs, and to determine how the provision of information and communication of risks can be improved and tailored to current practice.

## **6.2 Methods**

### **6.2.1 Participants**

A qualitative approach was adopted. Information and decision support needs were assessed using in-depth semi-structured interviews with health professionals who counsel women about amniocentesis testing. A convenience sample of health professionals was approached and recruited in two antenatal clinics in Wales (University Hospital of Wales and Llandough hospital). To ensure a breadth of responses, professionals from different specialities were recruited: obstetrics, midwifery, genetics and counselling. Professionals from the Policy and Public Health sector as well as professionals from relevant charities were recruited through networking and steering group meetings in England and Wales. The study protocol and materials were reviewed and approved by the research and development committees of the participating sites and by the National Research Ethics Service. In total, 20 professionals were invited to take part. This included consultants in obstetrics and gynaecology, midwives, screening midwives, geneticists, coordinators of the national antenatal screening programme, and directors from charities.

### **6.2.2 Data Collection**

Semi-structured interviews with stakeholders were carried out in antenatal clinics or over the phone from April to September 2007. The interview schedule was structured around 11 open-ended questions investigating the professionals' assessment of women's information and decision support needs, their difficulties in making a decision, the factors they took into account when deciding and their attitudes post-decision. Potential issues and difficulties arising when counselling women about amniocentesis testing were also investigated. Special

attention was paid to the professionals' opinions and preferences regarding the presentation of information, framing of outcome probabilities and portrayal of risks. Interviews were conducted until data saturation was reached. All interviews were recorded digitally and transcribed by the same researcher (author of this thesis) (see Appendix 3 for interview schedules).

### **6.2.3 Data Analysis**

The qualitative analysis was as described in Chapter 5 (p. 74). The list of codes and an example of coded interview transcript are enclosed in Appendix 3. To ensure reliability of coding, six interview transcripts were coded by two independent raters (see Chapter 5).

## **6.3 Results**

Twenty health professionals were recruited and 17 were interviewed. The sample consisted of six consultants in obstetrics and gynaecology, four midwives, one screening midwife, two geneticists, two coordinators of the national antenatal screening programme, the local director of the Down's Syndrome Association, and the director of a national charity supporting parents during the antenatal testing process. Interviews lasted between 11 and 52 minutes (27 minutes on average). Five themes were identified in the interviews: information needs, emotions and decision support, reasons for accepting/declining an amniocentesis, perceptions and counselling difficulties (see Table 6.1).

### **6.3.1 Information Needs**

Primarily, most professionals believed that pregnant women needed to understand the harms, benefits and implications of each option and be aware of the risk of miscarriage. Some health professionals insisted on the necessity to balance and make sense of the risk of miscarriage against the risk of chromosomal abnormality. Professionals were inconsistent on



Table 6.1 *Themes Identified in Interviews With Health Professionals (Stakeholder Analysis)*

Themes	Sub-themes
Perceived information needs	<ul style="list-style-type: none"> <li>- Detailed information about the procedure</li> <li>- Consequences/implications of an amniocentesis</li> <li>- Screening test purposes</li> <li>- Multiple framing of probabilistic information</li> <li>- Lack of consensus around the risks involved</li> </ul>
Perceived emotional and decision making difficulties	<ul style="list-style-type: none"> <li>- Peak anxiety levels</li> <li>- Ways of providing decision support</li> </ul>
Professionals' evaluation of pregnant women's reasons for accepting/declining an amniocentesis	<ul style="list-style-type: none"> <li>- Women/professionals consistency</li> </ul>
Other issues and perceptions	<ul style="list-style-type: none"> <li>- Issues encountered when deciding with a partner</li> <li>- Improved understanding</li> </ul>
Counselling difficulties	<ul style="list-style-type: none"> <li>- Time constraints</li> <li>- Specialised knowledge</li> </ul>

the risk of miscarriage to quote (i.e., local rate or national rate). Paradoxically, they insisted on the need to provide consistent information across professionals.

“We quote a 1% risk of miscarriage, a 1 in 100 and that is the Welsh national risk so that is the only figure that we can give them and again it is not discerning between foetuses that have chromosomal abnormalities and those that have not.” (F, midwife)

“Well, the national Welsh recommendation is 1 in 100 and our unit figure is 1 in 300. So I usually say to them, it's between half and 1 percent and that sort of covers everything. I think, within our unit, I don't see why we shouldn't be using our own figures. If that is what the risk is in our unit, that is what the risk is in our unit!” (F, consultant obstetrician & gynaecologist)

Health professionals felt that practical and detailed information about the amniocentesis procedure, its consequences and implications should be offered to women and their partners. Eight professionals reported that information about the results, the type of abnormality

detected by each specific test and timescales of the QF-PCR and karyotype test results should be provided systematically.

“They need to know that the PCR is available within I think 3 working days, isn’t it? and that it only gives a limited result and that other things may come back after that, that it’s only excluding three basic trisomies so we need to make them clear that a good result at the end of the three days is good but it’s not saying everything is fine.” (F, consultant obstetrician & gynaecologist)

Two professionals believed that women should be aware that amniocentesis testing does not only test for Down’s syndrome but may detect a wider range of chromosomal abnormalities. Four professionals highlighted the need to specify that a normal amniocentesis result cannot guarantee a healthy baby. While the chromosome tests conducted on the amniotic sample will detect the most common chromosomal abnormalities, changes in single genes or microdeletions will not be diagnosed.

“It’s getting them to understand that you can have all of the tests done and have a very disabled baby born. The tests answer the question that has been asked: Are chromosomes 18, 13 and 21 structurally normal? And the answer is yes. That doesn’t say the baby is normal. And I think that is the key sentence and that’s the thing they don’t like us for; understandable.” (F, geneticist)

Furthermore, two professionals highlighted the need to specify that the chromosome tests will not provide information about the severity of the abnormalities detected.

“No tests will tell you how affected a child is going to be by Down’s syndrome. What families actually want to know is: is my child with Down’s syndrome going to be a very able child? Will he go to mainstream education? (...) And of course, there is no way of knowing that and I think that’s the thing that lots of families find particularly difficult when they are making that decision.” (F, midwife)

There was no consistency among professionals on the amount of information needed about potential chromosomal abnormalities and whether or not to raise the issue of elective pregnancy termination. Given amniocentesis is performed between 15 to 18 weeks of pregnancy, elective pregnancy termination involves induction of miscarriage and labour. Eight professionals believed that informing women about the procedure of terminating a pregnancy was essential before women consent to amniocentesis testing. Four professionals

felt that information about elective termination of pregnancy was unnecessary at this early decision making stage and could be provided later, to avoid information overload.

“I think health professionals should anticipate that level of not understanding and be very explicit about what a termination of pregnancy is and how you do a termination, because women may not understand, and are consenting to things they don't understand.” (F, coordinator of the national antenatal screening programme)

“I think that's just one step too far of the process. If you start describing the whole process including the details of how you do a termination, it's too much to take in.” (M, consultant obstetrician & gynaecologist)

Most professionals underlined the necessity to understand the screening test purposes, results and implications (i.e., deciding about amniocentesis, possible diagnosis of abnormality, decision to continue/terminate the pregnancy) before embarking on prenatal screening for Down's syndrome. Six professionals reported that the false positive and false negative results associated with prenatal screening, especially relevant with maternal serum screening tests, should be communicated and understood.

“With the screening test, low risk is not no risk and this is what I always tell them. Similarly, some of the highest chance results are going to be wrong, it's not a definitive test. Initially, not everybody understands that, but I do, I make a big thing of it actually.” (F, midwife)

Regarding the presentation and framing of information, most professionals highlighted the need to present information in multiple ways and to use different formats. The majority of professionals felt that information should be tailored to women's individual needs and account for individual differences in processing information.

“Different women have different needs, some are very numeric and some are not. I think you just got to ask the women really, how do they normally make decisions? If they normally make decisions in their heads, sort of comparing, sort of red apples and green apples, then you can do it that way.” (F, coordinator antenatal screening programme)

“You can't generalise at all. You get the neuroscientist who wants to know everything, even about the technique of culturing the cells, and what would happen if they had a positive result, and exactly how the termination is done. And you get the other woman who comes into the room and says: I want an amniocentesis and don't tell me anything more, I just want it done.” (F, consultant obstetrician & gynaecologist)

Similarly, professionals highlighted the need to gradually provide different levels of information, from the basic essential information to the specialised, peripheral information that some women were asking for. Preferred methods for communicating risks varied from one professional to another: hard facts only, analogies, diagrams, flow charts etc. Some professionals used verbal analogies while others preferred visual aids such as diagrams or flow charts.

“It’s not easy doing this, it depends on the individual. Very often, when they are sitting there with their husband, I’ll talk about things like, horse racing, betting, because 100 to 1 in betting... the husband will go, oh yeah, no chance! But, they will understand the risk, they’ve seen that sort of risk, you know, in a betting shop.” (M, consultant obstetrician & gynaecologist)

“When you are talking about risks of amniocentesis, risks of Down’s, I think the visuals, I don’t know what they’re called, but the little cards that the midwives have, which show what a risk of 1 in 100 means in terms of little spots, one of the spots is red and the rest are black. I think that’s quite a good visual impact of how risky your procedure is.” (F, consultant obstetrician & gynaecologist)

“The dots, I think, are widely used by the midwives, the screening midwives. I use the analogy of days of the week, or days of the month, or days of the year. I think a lot of people find that a lot easier to follow.” (M, consultant obstetrician & gynaecologist)

Furthermore, most professionals highlighted the need to provide updated information, using precise but simple language. A minority of professionals suggested that women should be given enough time to decide and should be reminded of the possibility of changing their mind at any time. One professional believed that people would assimilate more information if they were given more time to decide.

“As with anybody who is getting bad news, the key thing they need is time. I think it’s no good just telling them what they need to hear. It’s giving them the time to understand what they need to hear because people view risk in different ways and, hear it in different ways.” (F, geneticist)

### **6.3.2 Emotions and Decision Support**

Most health professionals recognised that the amniocentesis decision was associated with heightened stress, anxiety and subsequent difficulty to assimilate information. They believed that women experienced highest anxiety levels at the time of the screening test results.

“It’s just shock, horror, because they were two fit, healthy people. Why was that happening to them? They want answers, they want to know why. They wouldn’t have had the test I don’t think if they thought it was going to come back as high risk.” (F, midwife)

Four professionals felt that prompting women to reflect on the reasons for having/not having an amniocentesis (e.g., for reassurance, to be able to terminate the pregnancy if a problem is found) would facilitate decision making:

“I say: what would you do? Would you terminate the pregnancy or is it just for your information? because that’s what they need to know, isn’t it? They need to use that as part of their decision and very often they don’t know.” (F, screening midwife).

Alternatively, some professionals felt that the impact of a disabled child on the expectant parents’ life and family should be discussed and explicitly addressed during counselling. Finally, helping pregnant women decide what the worst possible outcome would be: to give birth to a child with a chromosomal abnormality, to miscarry a healthy baby or to terminate a pregnancy following a diagnosis of chromosomal abnormality, was perceived to facilitate decision making. Four professionals reported the tendency for women to ask what the professionals would do.

“I often get asked: what do I think they should do? And I always decline to actually give any kind of weighed personal opinion on that. The truth is, I don’t actually know what I would do myself faced with that decision. And secondly, the birth of a child with Down’s syndrome is likely to mean something very different to different people, it wouldn’t be right for a health professional to try to tell somebody what’s right for them.” (M, consultant in obstetrics)

### **6.3.3 Reasons for Accepting/Declining an Amniocentesis**

Based on their experience with the amniocentesis decision, health professionals identified 25 reasons presumably affecting women’s decision about amniocentesis. They believed that reasons most commonly influencing the decision to undertake amniocentesis were: the risk of miscarriage, existing children, knowledge or experience of Down’s syndrome, and perceived capacity to look after a disabled child. Other frequently reported reasons are listed in Table 6.2. The consistency between the reasons reported by women (see

Table 6.2 *Professionals' Assessment of Women's Reasons for or Against Amniocentesis*

<b>Reasons reported by health professionals</b>	
<i>Reasons for accepting amniocentesis</i>	<i>Reasons for declining amniocentesis</i>
- To find out if the baby has a problem (stress of not knowing)*	- To avoid anything that may harm the baby (risk of miscarriage)*
- Capacity/willingness to look after a disabled child*	- Capacity/willingness to look after a disabled child*
- Knowledge and/or experience of children with Down's syndrome*	- Knowledge and/or experience of children with Down's syndrome*
- Existing children*	- View on termination*
- To prepare if a problem is found*	- Previous miscarriage*
- Partner's views*	- Partner's views*
- Risk of miscarriage compared to risk of a problem*	- Risk of miscarriage compared to risk of a problem*
- To have the option to terminate pregnancy*	- Medical complication/Obstetric history*
- Family history of chromosome disorder*	- Difficulty getting pregnant*
- Age*	- Age*
- Obstetrician's expertise in conducting amniocenteses*	- Religious beliefs*
- Views of friends, family*	- Views of friends, family*
- Concerns about people's reactions	- Concerns about people's reactions
- Cultural characteristics	- Cultural characteristics
- Knowledge about amniocentesis	- Knowledge about amniocentesis
- Couple's stability	- Couple's stability
- Previous amniocentesis*	- Previous amniocentesis*
- Professionals' influence	- Professionals' influence
- Existing child with Down's syndrome*	

\* Reasons reported by both pregnant women and health professionals

Chapter 5) and professionals was high. Twenty out of 24 reasons reported by pregnant women were consistent with health professionals' assessment. However, women identified several reasons or factors which were not recognised as influential reasons by health professionals. Specifically, pregnant women felt that their individual risk of foetal abnormality compared to the risk in a similar age group, the difference between their individual screening result and the screening cut-off limit, and the anomalies detected on the mid-pregnancy scan, may influence their decision about amniocentesis.

### **6.3.4 Professionals' Perceptions of the Amniocentesis Decision**

Five professionals noted significant decision making difficulties between spouses (e.g., conflicts, incapacity to decide together) and believed that health professionals should act as facilitators. Three professionals observed that men generally left the decision to women.

“The other problem sometimes is when there are slightly different thresholds between the couple and it's interesting. Most of the time, it tends to be the women who are more likely to go ahead with the testing.” (M, consultant in obstetrics)

As a consequence of improved information and decision support over the past five years, some professionals believed that women were better informed. Three professionals felt that women had a good understanding of the procedure and of Down's syndrome. However, two professionals noted significant difficulties dealing with statistics and understanding the limitations of amniocentesis testing. Three professionals felt that making a decision in a short time frame and balancing the risk of miscarriage against the risk of a problem could prove difficult. Two health professionals believed that women experienced information overload.

### **6.3.5 Counselling Difficulties**

Finally, health professionals reported communication and counselling difficulties. Three health professionals reported difficulties dealing with specialised genetic information and occasional lack of research evidence. Research evidence surrounding the cause of miscarriage following amniocentesis is poor and health professionals did not always have answers to women's concerns.

“It's quite specialist counselling. People that know little about something, quite often have quite a lot to say. So GPs, obstetricians, midwives may all have plenty to say about the little bit of knowledge they have, whereas the geneticists who really understand... hum, you know, for the woman, her view has already been prejudiced.” (F, consultant obstetrician & gynaecologist)

“The evidence based is not very good and again it's hard to tell women that you're doing a routine procedure and actually we can't tell them exactly when it's most likely to miscarry, what the symptoms would be, what sorts of women are most likely to miscarry, the data is not there or not that we found.” (F, coordinator antenatal screening programme)

Three professionals experienced difficulties providing specific information about amniocentesis testing (e.g., detailed information about Down's syndrome and other abnormalities) within the time constraints of the consultation.

"It's quite difficult in a short consultation to get over the range of abnormalities." (M, consultant obstetrician & gynaecologist)

### **6.3.6 Synthesis**

Health professionals highlighted the need to understand the risks associated with amniocentesis testing (e.g., risk of miscarriage, risk of infection), and the characteristics and limitations of chromosome test results. Most health professionals underlined women's cognitive and emotional difficulties deciding about amniocentesis testing (e.g., stress, anxiety, difficulty assimilating probabilistic information and information overload). They unanimously highlighted the need to address women's information needs by providing personalised and interactive information, presented in multiple ways (e.g., numbers, diagrams and videos of women's experiences). Professionals were inconsistent regarding the miscarriage rate to quote (i.e., local or national rate), and whether or not to describe the termination of pregnancy. They insisted on the necessity to provide detailed information about amniocentesis before the screening tests, as existing information and consent arrangements seemed insufficient.

## **6.4 Discussion**

The present findings indicated variations in the evaluation of women's information and decision support needs and a tendency to prioritise information provision over emotional support. The assessment of women's information needs considerably varied between healthcare professionals.



Healthcare providers expressed divergent opinions regarding the nature and quantity of information needed about the risk of miscarriage, chromosomal abnormalities tested for, termination of pregnancy and uncertainty associated with the tests. Previous work showed that health professionals from different specialities had differing approaches and attitudes to chromosomal abnormalities (Marteau et al. 1994). Divergent opinions with regards to the provision of information may be explained by limitations of current policies and guidelines (i.e., miscarriage rate) but may also be imputed to gaps in health professionals' general or specialised knowledge (i.e., genetics). Indeed, several health professionals reported difficulties dealing with specialised genetic information. Existing research on professionals' knowledge about prenatal genetic testing corroborates our findings by revealing inadequate knowledge related to the presentation and meaning of prenatal screening test results (Carroll et al. 1997; Sadler 1997; Wilkins-Haug et al. 1999). Marteau et al. (1993) established that obstetricians counselling women about amniocentesis testing occasionally provided women with incorrect information. Further research has shown that health professionals tended to overestimate their own levels of knowledge (Hunter et al. 1998; Tracey et al. 1997).

Health professionals held strong views and control over the nature of information communicated (or not) to women facing a decision to undergo amniocentesis testing. Several professionals strongly believed that the issue of elective termination of pregnancy should not be addressed or even mentioned to pregnant women considering an amniocentesis. Others felt that pregnant women should be fully informed about the termination procedure and made aware that it involved induced labour as this may affect their decision. Professionals' attitudes with regards to specific topics (termination of pregnancy, range of abnormalities tested for) may be described as paternalistic. Possible factors influencing health professionals' attitudes and control over specific information topics include societal pressures

to conform and minimise human differences and complex ethical dilemmas associated with amniocentesis testing and elective termination of pregnancy (Garel et al. 2002; Strauss 2002). It has also been argued that the prospect and range of foetal abnormalities may be too distressing or alarming for health professionals and pregnant women to discuss in detail (Marteau 1993). Finally, professionals reported counselling difficulties, including time constraints and occasional lack of specialist knowledge, as has been previously documented (Hunt et al. 2005; Marteau 1993; Williams et al. 2002b). Their counselling difficulties and lack of specialist knowledge may be addressed by DESIs.

As highlighted in Chapter 5, pregnant women reported important decision making difficulties (e.g., difficulty assimilating probabilistic information, information overload). Both women and health professionals emphasised a period of elevated stress and anxiety that was triggered by the decision and its aftermath. However, the need to reinforce and strengthen existing emotional and decision support highlighted by the majority of women interviewed, was only identified by a minority of professionals. Instead, professionals insisted on providing comprehensive information about the screening tests, the risks, the results and implications of amniocentesis testing. They identified areas where efficient information provision was essential but tended to underestimate existing emotional and decision support needs. This may reflect areas of expertise that health professionals feel most competent in carrying out and a genuine gap in understanding of patients' emotional needs. These results also indicated a gap in perception between users and providers on emotional and decision support needs of couples considering amniocentesis and are consistent with existing literature in this area (Hunt et al. 2005; St-Jacques et al. 2008).

## **6.5 Conclusion**

This chapter discussed the professionals' assessment of women's information and decision support needs associated with amniocentesis testing. There is scope for improving the framing of information and risk communication, for providing consistent information across professionals and tailoring information provision to individual needs and differences. Existing information and decision support for women considering amniocentesis testing could benefit from high quality DESIs offered as a supplement to routine information and counselling.

Furthermore, the data collected with health professionals highlighted concerns about the quality of information provided to women prior to prenatal screening tests and about the validity of consent obtained. Information about the characteristics, limitations and consequences (i.e., further invasive diagnostic tests offered) of prenatal screening tests should be provided to women and addressed in a DESI for amniocentesis testing. Finally, with the objective to answer patients' needs and to create DESIs which are accepted by patients and implemented by healthcare providers in clinical settings, DESIs should be developed in collaboration with both patients and professionals. Those interventions do not aim to replace but supplement face to face interactions with health-professionals.

# Chapter 7

## Prototype Development

### 7.1 Introduction

While DESIs propose to guide and facilitate decision making, they do not normally build on theoretical descriptions and explanations of how individuals make decisions. DESIs have been researched and developed for over a decade but their theoretical underpinnings have only been examined in recent years (Bekker et al. 1999; Bowen et al. 2006).

Decision making theories exist and have increased our understanding of how individuals make decisions or ought to make decisions that lead to optimal outcomes. Theories or models of decision making are divided into normative or prescriptive theories, that address how individuals should ideally make decisions, and descriptive theories, that describe how individuals achieve decision making in real-world situations (Baron 2000). Normative models or theories of decision making specify from a rational standpoint how a decision should be made to achieve the best possible goals or outcomes, under ideal conditions. Descriptive theories provide a factual and behavioural account of how people make decisions in normal settings. They often involve heuristics or rules of thumb. Although empirical evidence on the impact of theory-based interventions is missing, it is hypothesised that more extensive use of decision making theory would facilitate progress towards helping individuals make difficult healthcare decisions (Bekker et al. 1999). The aim of this chapter was to describe the theoretical foundations underlying the development of amnioDex, its overall prototype development and first prototype intervention. AmnioDex (amniocentesis decision explorer) is a web-based DESI developed to support and facilitate decision making of pregnant women facing a decision to undergo amniocentesis testing.

## **7.2 A Theory-Based Decision Support Intervention**

As highlighted in Chapter 2, the transfer of theoretical constructs into the design and prototype development of DESIs and their components (e.g., deliberation tools) remains rare and has posed obstacles. Few decision making theories offer an explicit architecture or method for transferring theoretical approaches into practical interventions. Expected utility theory provides an explicit and systematic method for achieving such translation: decision analysis and the decision tree. This may explain why expected utility theory has been widely applied to DESI development (Montgomery et al. 2003; Rothert et al. 1997). However, the validity and appropriateness of interventions based on expected utility theory (i.e., using decision analysis) are questioned (Elwyn et al. 2001). In healthcare decisions involving high stakes, short timeframes and emotional strains; asking patients to process numerical information and integrate probabilities with weighted utilities may be too difficult, if not impossible. Evidence suggests that the results of utility calculations are often inconsistent with the patient's choice (Pauker and Pauker 1979; Elwyn et al. 2001). Normative theories, such as expected utility theory are generally derived from mathematical models and do not normally account for the individual's emotional, cognitive, environmental and/or time constraints. By contrast, descriptive theories or models of decision making recognise that decision makers have limited reasoning and computational abilities and examine ways of overcoming these difficulties. Interventions designed to facilitate decision making processes (DESI and its components) may benefit from building on key concepts of how individuals actually make decisions under risk. In the clinical context of amniocentesis testing, where high emotional demands and complex information limit people's capacity to quantify utilities of options, the assumption was made that descriptive models of decision making would fit this specific context better than normative theories.

Given the translation difficulties highlighted in Chapter 4 and the lack of literature documenting the transfer of theory into practical interventions, a decision was made to use theoretical frameworks that had previously guided the development of DESIs or DESI's components. In 2001, Feldman-Stewart et al. developed a DESI and embedded value clarification exercise for men diagnosed with early stage prostate cancer, based on the differentiation and consolidation theory (DiffCon) by Svenson (Feldman-Stewart et al. 2001; Feldman-Stewart et al. 2006). The Ottawa Health Research Institute developed DESIs based on prospect theory, a theoretical foundation embedded in the Ottawa decision support framework (O'Connor et al. 1999c). Those two theoretical frameworks; DiffCon and prospect theory, were chosen to develop amnioDex.

### **7.2.1 Differentiation and Consolidation Theory**

Svenson postulates that pre- and post-decision making processes are intrinsically linked and therefore describes two stages of decision making: differentiation (pre-decision) and consolidation (post-decision). Differentiation is an active process of gradually differentiating competing options until one option is deemed superior to other alternatives to allow decision making (Svenson 1992). Differentiation involves a range of strategies and rules of reasoning including, for example, the conjunctive, disjunctive or lexicographic rules (Svenson 1979). Those rules are used to judge options and their attributes in terms of their attractiveness and importance. If none of the offered alternatives is judged superior, the status quo is maintained. The conjunctive rule is derived from Simon's satisficing principle (see Chapter 8) and requires the decision maker to specify a set of criteria on the attributes that are considered important in making the decision (Simon 1955). The option which meets all or most of the criteria will be selected. A disjunctive strategy involves choosing the option with one or two highly ranked attributes. The lexicographic rule involves choosing the option according to the most important attribute only. In case of attribute ties, the second most

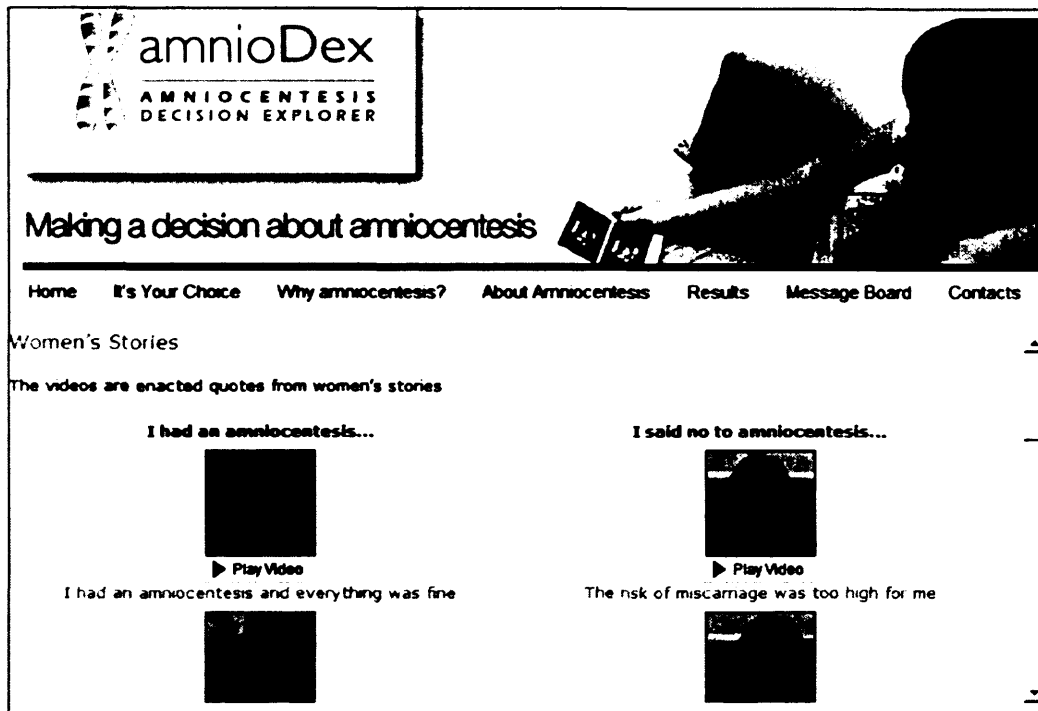
important attribute will discriminate between options. These strategies considerably overlap with Gigerenzer's "fast and frugal heuristics" (Gigerenzer and Todd 1999) (see Chapter 8). Finally, post-decision, the consolidation process occurs to protect the chosen alternative against future internal and external threats or decision doubts (Svenson 1992). Differentiation and consolidation theory predicts that sufficient restructuring (differentiation and consolidation processes) minimises or prevents the occurrence of regret and decisional conflict.

Following the prerequisites of differentiation and consolidation theory, information should be presented so the differentiation between options is facilitated. The comparison between alternatives should therefore be made easier. One method to achieve differentiation is the use of comparison tables that allow head to head comparison of attributes, either similar across options or unique (Feldman-Stewart et al. 2006). In light of this theoretical work, in amnioDex, two features were incorporated to facilitate differentiation. Firstly, a comparison table presented the harms and benefits of amniocentesis testing in equal detail. Secondly, balanced experiences of accepting and declining amniocentesis testing were provided. Video clips of enacted quotes from women's experiences illustrated all possible consequences of having or not having an amniocentesis. The videos were organised in two columns: "I had an amniocentesis" and "I said no to amniocentesis" (see Figure 7.1). The assumption was made that differentiation between options would be facilitated by providing balanced examples of all possible outcomes following the offer of amniocentesis testing (see Section 7.4).

### **7.2.2 Prospect Theory**

Kahneman & Tversky developed prospect theory to offer an alternative model to expected utility theory, as they believed that such normative model of rational choice failed

Figure 7.1 *Presentation of Video Clips Influenced by Differentiation and Consolidation Theory*



to describe human choices under risk (Kahneman and Tversky 1979). They noted that individuals systematically failed to match up to the rules of expected utility theory or other rational decision models and made use of heuristics (e.g., the influence of loss or gain frames and the impact of recently appraised information). Prospect theory postulates that decision making involves two phases: an early editing phase and a subsequent phase of evaluation of all available options until the option with the highest value is chosen (Tversky and Kahneman 1981; Tversky and Kahneman 1992). To simplify subsequent evaluation and decision making, the editing process consists of reformulating the cues available using several operations (e.g., preliminary analysis, framing and simpler presentation of the options by using heuristics). According to prospect theory, decision making is influenced by 1) the way in which information is presented and risks portrayed (i.e., framing of information), 2) a tendency to be risk averse when choosing between gains, and risk seeking when choosing between losses (i.e., certainty effect) and 3) the fact that losses generally loom larger than gains. Prospect theory posits that the way losses and gains are perceived depends on the



reference point. In the context of amniocentesis testing, being pregnant is the reference point; the loss therefore looms large (i.e., losing the baby).

The effects of information framing, initially characterised in prospect theory, have been documented in various contexts, including medical decision making (Kuhberger 1998; Marteau 1989; McNeil et al. 1982; Moxey et al. 2003). The framing effects in medical decision making imply that crucial treatment or screening decisions may be biased by the way in which harms and benefits are described. Framing effects may therefore induce biased or less informed decisions and lead to poor decision outcomes (i.e., decisional conflict or decision regret) (McNeil et al. 1982). With the aim to facilitate informed decision making and reduce decisional conflict, DESIs may include methods for reducing framing biases. It has been recognised that framing biases can be minimised by framing information in multiple ways: by language, by number (percentages and natural frequencies), by images and by diagrams (Edwards et al. 2001). Specifically, diagrams may be used to minimise framing effects by representing affected and non-affected icons in a visual diagram (O'Connor et al. 2005; Lipkus and Hollands 1999). To take account of framing effects, outcome probabilities in amnioDex were framed by language, number, and diagrammatic representations (see Figure 7.2). This decision was justified by the prerequisites of prospect theory but was also based on the results of the needs assessment conducted with pregnant women (Chapter 5), where women unanimously highlighted the need to address decision difficulties by presenting information in multiple ways. Further, during the needs assessment, pregnant women also reported difficulties understanding the screening test result. Therefore, to help women better understand their screening test result, and consistent with prospect theory, a functionality (“Understanding Your Result”) was added that framed women’s individual screening test result by language, number, diagrams

Figure 7.2 Example of Outcome Probabilities Framed by Language, Number and Diagram

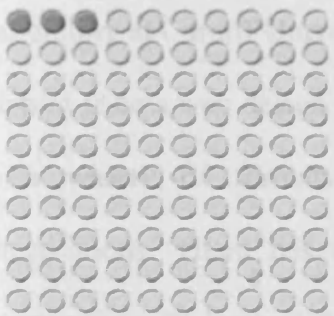
Why amniocentesis? About Amniocentesis Results Message Board Contacts

**Results**

The results of the chromosome tests (PCR and karyotype test) can detect Down's syndrome or other conditions.

It is estimated that 97% of women who undertake amniocentesis will have a healthy baby (ref 5).

A chromosome problem will be identified in 3 out of 100 amniocenteses performed



Your most important reason Talking to others Personal Stories





Figure 7.3 Screenshot of Understanding Your Result

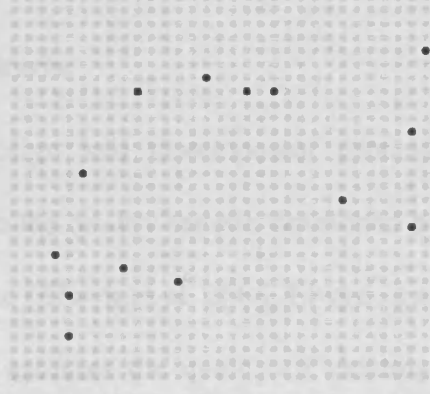
If you know the result, type your test result here: 1 in

For a 1 in 150 chance of having a baby with Down's syndrome, you have 149 out of 150 chances that the baby won't have Down's syndrome.

This also means that you have 7 out of 1000 chance of having a baby with Down's syndrome.



Imagine a jar with 149 coloured balls and 1 white ball. Imagine closing your eyes and picking up a ball from the jar. You picking the white ball corresponds to your chance of having a baby with Down's syndrome.



and images (see Figure 7.3). Women were asked to enter their individual screening test result. Their individual result was subsequently framed negatively and positively, converted into a diagram and illustrated using an image and analogy.

### **7.3 Developing AmnioDex**

AmnioDex (amniocentesis decision explorer) was developed by the Decision Laboratory at Cardiff University between September 2006 and July 2008, with technical support from a web design company. Over the past few decades, DESIs have been developed in a variety of formats: paper-based, audio, video, and more recently web-based interventions. There has recently been an increasing interest in developing web-based interventions (Schwitzer 2002). They offer the potential to individualise the provision of information and provide high adaptability and interactivity. In the case of amnioDex, a decision was made to develop a web-based intervention in order to include videos and other interactive elements, and because it offered great adaptability, reduced the cost of production (in the long term), and theoretically increased access and allowed wider dissemination. The amnioDex intervention is openly accessible online at [www.amniodex.com](http://www.amniodex.com).

#### **7.3.1 The IPDAS Criteria**

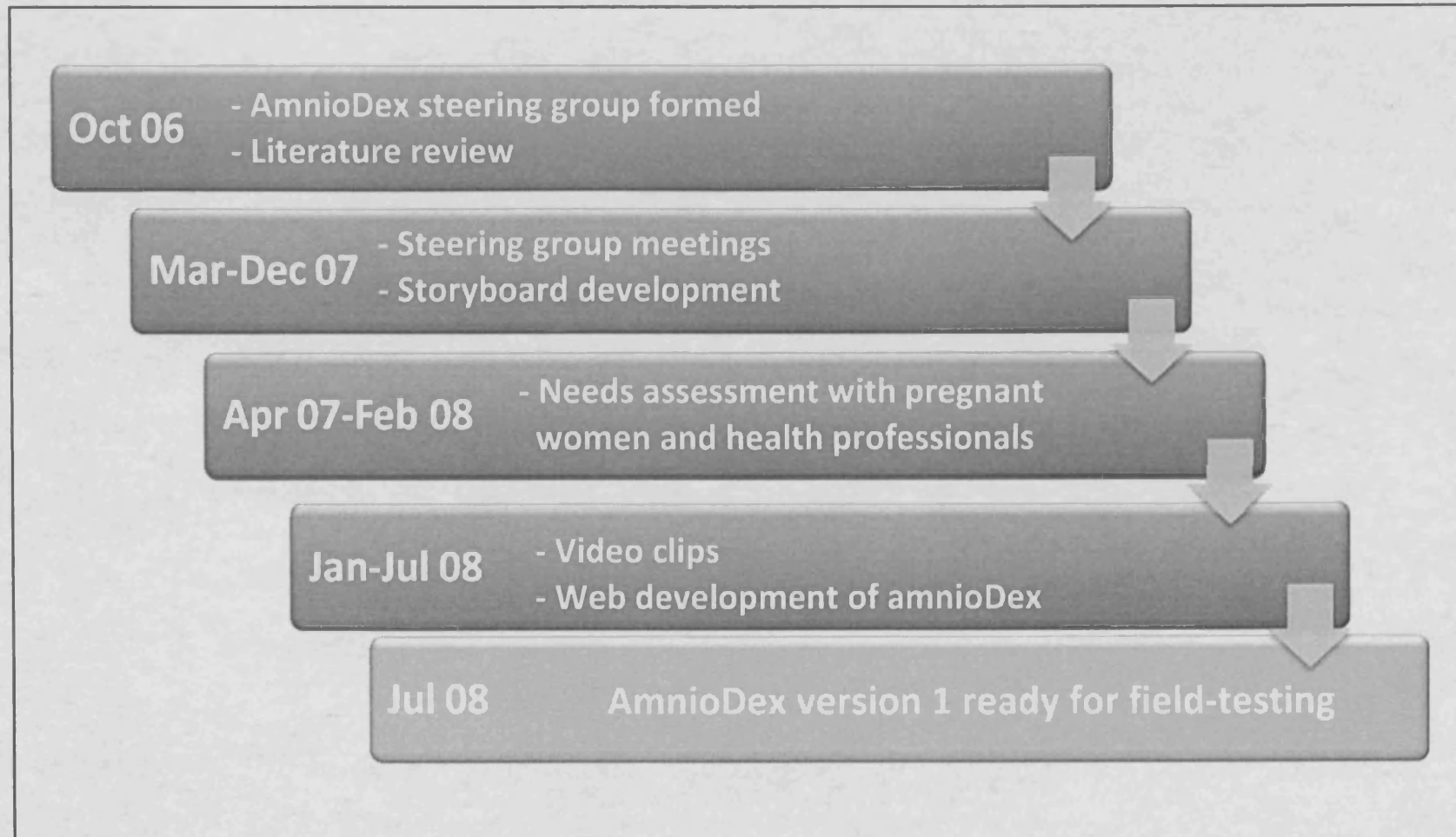
The general design and development process of the intervention was initially determined according to the quality criteria developed by the IPDAS collaboration and listed in the IPDAS instrument (Elwyn et al. 2006; Elwyn et al. 2009c). As described in Chapter 3, the IPDAS instrument assesses the quality of existing DESIs at 10 broad domain levels: Information, Test, Probabilities, Values, Guidance, Development, Evidence, Disclosure, Plain Language and Evaluation. While developing the intervention, an effort was made to fulfil as many domain requirements as possible. In compliance with the information domain, amnioDex provided comprehensive information about the decision at stake, the options

available, their harms, benefits, outcomes and potential consequences. The intervention also presented information about the specific features of the diagnostic test (amniocentesis) and implications of accepting or declining the test (Test domain). AmnioDex provided precise and balanced information about the outcome probabilities associated with each option (Probabilities domain). AmnioDex also included deliberation tools to facilitate the expression and clarification of values (Values domain) (see Chapter 8). In the section “Talking to Others”, amnioDex provided structured guidance towards making a decision by including a list of questions to discuss with their midwife or obstetrician (Guidance domain). As described in Chapters 5, 6, 8 and 9; amnioDex was developed following a systematic development process, involving pregnant women and health professionals at all stages of development (Development domain). The intervention was written using evidence-based information, synthesised from literature review and referenced throughout the website (Evidence domain). In section “More About AmnioDex”, the website provided information about developers’ credentials and sources of funding used to develop the intervention (Disclosure domain). Finally, the intervention was developed using plain language and will soon be reviewed by the Plain English Campaign (Plain Language domain).

### **7.3.2 The AmnioDex Steering Group**

A steering group of stakeholders and health professionals from disciplines relevant to the amniocentesis decision was created in October 2006 (see Figure 7.4). The amnioDex steering group was formed to ensure that the informational content was accurate and consistent with current clinical practice. Another objective was to make sure that it covered all issues women may have when deciding about amniocentesis while accounting for the difficulties health professionals may experience when counselling them. Comments and suggestions for improvements of steering group members were systematically examined by a stakeholder group of researchers, including the author of this thesis, before rejecting or

Figure 7.4 *Timeline Illustrating the Development of AmnioDex*



implementing suggested changes. The researchers who developed amnioDex retained editorial control of the intervention's content. The steering group consisted of 17 stakeholders: two consultants in obstetrics and gynaecology, nine midwives or screening midwives, one geneticist, three coordinators of the national antenatal screening programme (in Wales and England), one director of a national charity and one patient representative. Steering group members were initially involved in identifying essential themes and areas to be covered in the intervention. They were subsequently asked to review incremental prototypes of the intervention. Two steering group meetings were held in March and December 2007. Steering group members were frequently consulted via emails.

### **7.3.3 Textual Content**

The content of amnioDex was based on a literature review (see Chapters 2, 3 and 4), needs assessment conducted with pregnant women and health professionals (see Chapters 5 and 6) and steering group meetings, while complying with the IPDAS quality criteria. The decision to include or reject specific information, topics or sections was primarily based on available scientific evidence, data collected during the needs assessment, steering group meetings and theoretical underpinnings. However, the editorial control remained with the researchers who developed the amnioDex intervention (author of this thesis, GE and JB). In creative design involving multi-stakeholder consultation, such as amnioDex, editorial control appeared essential and inevitable.

In March 2007 (see Figure 7.4), a steering group meeting was held to discuss the content of the amnioDex intervention and identify critical information and essential topics to be covered. In September 2007, a storyboard presenting the informational content and graphic-based elements appearing on each page of amnioDex was created. The storyboard detailed the provisional layout, navigation structure, interactive elements and informational

content of the website. In parallel (April 2007-February 2008), a needs assessment was conducted with pregnant women and health professionals. The data was simultaneously analysed to guide the prototype development of the intervention. In December 2007, a second steering group meeting was held to review the storyboard, discuss essential informational content and identify areas and themes that needed particular attention. The steering group members commented on the wording and clinical accuracy of critical topics (e.g., section describing the risks, section about chromosomal abnormalities), on the presentation of information (e.g., use of pie charts, diagrams, images), framing of numerical data, general design and navigation of the website (as presented on the storyboard). They suggested the addition of a section displaying useful contacts and other information resources. Regular email exchanges were maintained with the steering group members to keep them updated of the changes made to the storyboard and verify the clinical accuracy of specific sections (e.g., section describing elective termination of pregnancy, section about the risks of miscarriage).

#### **7.3.4 Web Development**

In December 2007, a web design company was selected to develop the amnioDex intervention. Between January and March 2008, the video clips of women's stories and health professionals were filmed with actors and deliberation tools developed (see Chapter 8). The video clips of women's stories were developed using enacted quotes of women's experience about their decision to accept or decline amniocentesis testing. Ethical approval was granted by the National Research Ethics Service. Scripts were selected from 17 transcripts of interviews conducted with women who had been offered an amniocentesis and their partners (n=4). Ten actresses and one actor were selected to enact 14 video clips exclusively based on the interview transcripts. Two health professionals, a consultant in obstetrics and gynaecology (male) and a geneticist (female) were chosen to create 5 video clips addressing: the decision to undertake amniocentesis, the procedure, the risk of miscarriage, and

Figure 7.5 Screenshot of the AmnioDex Homepage



chromosomal abnormalities potentially detected. A video clip of a live amniocentesis procedure was filmed and added to the section “About Amniocentesis”. In parallel, the storyboard was finalised. The storyboard detailed all textual content appearing on the website and layout of all web pages including the message board, glossary and deliberation tools.

In March 2008, the latest version of the storyboard was circulated to all members of the steering group for comments. Steering group members were given three weeks to provide feedback and suggestions for improvement about the accuracy of the textual content, general layout of the website, quality and relevance of interactive elements, images and other graphic-based elements. Because of time constraints, the video clips were reviewed by 4 out of 17 members of the steering group: two coordinators of the national screening programme, one consultant in obstetrics and gynaecology and one geneticist. All video clips were



approved and uploaded onto the website. Between March and July 2008, the storyboard was converted into an interactive web-based intervention.

#### **7.4 The AmnioDex Intervention**

AmnioDex is an interactive web-based DESI intended for pregnant women (and partners) who have been offered amniocentesis testing, independently of their screening pathway (see Figure 7.5). AmnioDex may be used by pregnant women (and partners) who have been offered amniocentesis following a higher chance screening test result, an abnormality detected on the mid-pregnancy ultrasound scan, an inherited condition or a family history of a chromosome disorder.

AmnioDex ([www.amniodex.com](http://www.amniodex.com)) featured 63 pages of content divided into 10 sections. Four sections offered comprehensive information about the choice to be made (“It’s Your Choice”), the reasons for being offered an amniocentesis (“Why Amniocentesis?”), the amniocentesis procedure (About Amniocentesis), and the results of the chromosome tests (“Results”). Consistent with the results of the needs assessment (Chapter 5), where pregnant women reported the need for information about the elective termination of pregnancy, the section “If a Problem is Found” (in section About Amniocentesis) included information about continuing or ending the pregnancy. The section entitled It’s Your Choice, included six web pages featuring information about the harms and benefits of amniocentesis testing. Emotional and decisional support was provided in the following sections: “Making the Best Decision”, “Deciding With a Partner”, “Knowing Enough to Choose” and “Uncertainty and Anxiety”. The section entitled Why Amniocentesis? provided information about the reasons for being offered the test and included a tool to help users understand their screening test result (see Figure 7.1). Users were asked to enter their screening test result, and were subsequently shown different ways of framing the risk of chromosomal abnormality (i.e., positive versus

negative framing, diagram with dots and analogy). The section entitled about amniocentesis offered comprehensive information about the amniocentesis procedure (e.g., “What is it?”, “How is the Test Done?”, “After the Procedure”), the risks involved, the chromosome tests performed on the amniotic fluid and described all options available if an abnormality was detected (“Your Choices”, “Being Supported”, “Continuing the Pregnancy”, “Ending the Pregnancy”). The section entitled Results provided information about the results of the chromosome tests (“Getting the Results”, “Waiting for the Results”) about Down’s syndrome, other chromosomal abnormalities tested for and conditions that would not be detected following an amniocentesis. AmnioDex included a “Contacts” section with a list of relevant charities and alternative information resources available, a glossary and site map. The section entitled “More About AmnioDex” provided information about the development and authors of amnioDex, and a reference list.

Further, the intervention included several interactive elements and narratives. AmnioDex featured 14 video clips of enacted quotes of women’s stories, five videos of health professionals, and one video clip of a live amniocentesis procedure. The video clips of women’s stories were accessible throughout the website, in sections providing related information, or in the section “Personal Stories”, where all 14 video clips were presented according to four different themes: “I had an amniocentesis”, “I said no to amniocentesis”, “about the procedure” and “I miscarried after amniocentesis”. Five video clips of health professionals appeared in the sections: It’s Your Choice, About Amniocentesis and Results. To facilitate the expression and clarification of parents’ values with regards to the amniocentesis decision, amnioDex featured three deliberation tools: “weighing it up”, “your most important reason” and “talking to others”. Weighing it up and your most important reason were based on theory and required users to actively participate in the deliberation

process (see Chapter 8). Talking to others was developed without theoretical underpinnings but complied with the IPDAS recommendations by offering a list of questions and printable worksheet to discuss with their partners or health professionals. Finally, amnioDex included a message board divided into three web forums: “amniocentesis testing”, “making a decision”, “I’ve made a decision”.

## **7.5 The Virtual Reference Group**

Prior to evaluating amnioDex in a randomised controlled trial (grant outline submitted, see Appendix 5), a virtual reference group was convened to review the intervention after field test. In the context of amnioDex, the virtual reference group was a group of health professionals that provided advice and expertise by critically appraising the final content and presentation of the amnioDex intervention using a virtual review process (i.e., letters, email exchanges and telephone conversations). Members of the virtual reference group were identified through steering group meetings and contacts in England, Scotland and Wales, some of whom had already taken part in the stakeholder analysis (see Chapter 6). A letter was sent to 15 health professionals asking them to review the amnioDex website, and to comment on its content and format. Ten health professionals agreed to be part of the reference group: two consultants in obstetrics and gynaecology, a professor of obstetrics, a trustee from the Support Organisation For Trisomy 13/18 (SOFT), a midwife and lecturer in women’s health, a screening midwife, the operations director of the Down’s Syndrome Association, the director of Antenatal Results and Choices (ARC), a geneticist and the national programme director of the NHS Foetal Anomaly Programme.

# Chapter 8

## Development and Usability of Heuristic-Based Deliberation Tools

### 8.1 Introduction

How best to support people attempting to make difficult health decisions is an area of considerable research interest but debate about the methods for supporting and facilitating decision making remains. Numerous methods or strategies have been developed to assist patients in structuring their preferences to achieve a decision. Those methods propose to facilitate the expression and clarification of values so as to subsequently help patients select an option consistent with their stated values or preferences (Llewellyn-Thomas 2009). However, there is uncertainty and debate around the nature of cognitive processes that might help people express and clarify their values and attitudes in order to make informed preference-sensitive decisions (Dijksterhuis et al. 2006; O'Connor et al. 2007a; Wilson and Schooler 1991). Terms such as value, attribute, preference elicitation methods (Feldman-Stewart et al. 2006), value- or preference-clarification exercises (O'Connor et al. 1999b), probability-trade off techniques (Llewellyn-Thomas et al. 1996) and coaching or guidance methods can be found in the literature (Llewellyn-Thomas 2009). For the purpose of this and subsequent chapters, the generic term deliberation tools, was used to describe processes and methods that have been embedded into DESIs to facilitate deliberation and decision making. Over the past decades, deliberation tools have used a wide range of approaches to help patients express and clarify their values. Some have used decision analysis (based on expected utility theory) asking users to specify the numerical value of screening or treatment outcomes to identify the option with the highest expected utility based on their expressed

values and attitudes (Llewellyn-Thomas 1997). Other deliberation tools provide checklists, worksheets or balance scales for patients to complete, and indicate their values and preferences with regards to the decision (O'Connor et al. 1998b). Some feature video clips of patient experiences, illustrating the physical, social, and emotional effects of the harms, benefits and consequences of each option (Spunt et al. 1996). Some tools suggest a process of discussion with relatives or health professionals while others simply provide information about the harms and benefits of options, making the process of expressing and clarifying values implicit (Levine et al. 1992).

Most theory-based deliberation tools developed over the past decade either derived or made assumptions about rationality based on normative theories of decision making (Bekker et al. 2004; Holmes-Rovner et al. 1999; Llewellyn-Thomas 1997; Llewellyn-Thomas 2009). These theories or models of decision making assume unbounded rationality, that is, unlimited knowledge, time and computational capacities (Todd and Gigerenzer 2000). Normative theories such as these are derived from optimisation models that do not take account of emotional, cognitive, environmental and/or time constraints. People facing complex decisions such as those connected to health, often have a limited ability to process information and to integrate probabilities with weighted utilities. Bounded rationality models may be better suited to medical decisions under uncertainty. These models assume that humans have developed simpler adaptive strategies using limited information searching to arrive at effective decisions in time-efficient ways despite the aforementioned constraints (Simon 1956). Paradoxically, models of bounded rationality have rarely been used to develop decision support for patients and hence a research gap exists.

Gigerenzer et al. described a series of simple heuristics, which they called; fast and frugal heuristics, based on the theory of probabilistic mental models (Gigerenzer et al. 1991) and notions of bounded rationality (Gigerenzer and Goldstein 1996). Fast and frugal heuristics are non-compensatory; information search is limited and may be minimal, determined by simple stopping rules. The efficiency of heuristic-based methods has been evaluated in real world situations, including situations of medical decision making. Green and Mehr showed that simple heuristic-based methods outperformed complex rules of reasoning and improved physicians' decisions to admit patients to the coronary care unit (Green and Mehr 1997). There is growing empirical evidence that physicians may rely on fast and frugal heuristics without affecting the decision's quality. Fisher et al. showed that simple heuristic rules were successfully used to identify children at risk for pneumonia infections and to prescribe antibiotics (Fisher et al. 2002). Comparable results were found with decisions affecting prescription of antidepressants (Smith and Gilhooly 2006). The aim of the present chapter was to develop deliberation tools based on models of bounded rationality and to field-test these tools with researchers, healthcare professionals and pregnant women facing amniocentesis testing.

## **8.2 Methods**

The study was divided into four stages: (1) prototype development of two deliberation tools, (2) prototypes field-tested with researchers, (3) prototypes field-tested with health professionals, (4) prototypes field-tested with pregnant women facing a decision to undergo amniocentesis testing. The study protocol and materials were approved by the research and development committees of the participating sites (Cardiff and Vale NHS Trust, Velindre NHS trust) and by the National Research Ethics Service.

### 8.2.1 Prototype Development

Eight decision algorithms were considered as possible basis for the development of deliberation tools. These algorithms included four heuristic-based algorithms (Take the Best, Take the Last, Minimalist and Tallying), and four integration algorithms (Unit Weight Linear Model, Weighted tallying, Weighted Linear Model and Multiple Regression). In the literature, these decision algorithms (see Table 8.1) have been compared using simulations of performance on real-world questions under conditions of limited knowledge and time (Gigerenzer and Goldstein 1996). The criteria for selecting decision algorithms for translation into deliberation tools were the specified performance and predictive accuracy on simulated decision tasks (Gigerenzer and Goldstein 1996). Findings of that research and currently discussed research directions in this area (Wegwarth and Elwyn 2009) suggested that the heuristic-based algorithms: “Take the Best” and “Tallying” performed better than complex integration algorithms (Gigerenzer 2008). Both heuristic-based algorithms were therefore retained to guide the development of two deliberation tools embedded in amnioDex.

Key theoretical constructs of the Take the Best and Tallying heuristic-based algorithms guided the design of “your most important reason” and “weighing it up”. The deliberation tools were developed with technical support from a web-design company and collaboration with three researchers, over a period of 12 months. Several prototypes were developed for each deliberation tool. Each new prototype was discussed and adapted. Informal piloting occurred until the first usable prototype of the deliberation tools was ready for field-testing. The cognitive steps of each algorithm were isolated and translated into a graphic-based interactive deliberation tool. The first mental step of the Take the Best algorithm requires that one attribute is found that can discriminate between options (Gigerenzer and Goldstein 1996). In the second step, each attribute is reviewed, in order of

Table 8.1 Algorithms Considered for the Development of AmnioDex Deliberation Tools

Heuristic-based algorithms				Integration algorithms			
Take the Best	Take the Last	Minimalist	Tallying	Weighted Tallying	Unit Weight Linear Model	Weighted Linear Model	Multiple Regression
<ul style="list-style-type: none"> <li>▪ Cues<sup>3</sup> or attributes are subjectively ranked according to their validity/importance.</li> <li>▪ The highest ranking cue (best cue) is retrieved from memory.</li> <li>▪ If the cue discriminates between options, the search stops and an option is chosen.</li> <li>▪ If the cue does not discriminate between options, the process is repeated until a cue is found to discriminate.</li> </ul>	<ul style="list-style-type: none"> <li>▪ The cue that discriminated between options the last time a decision was made is chosen.</li> <li>▪ If this cue does not discriminate, the second most recently used cue is chosen.</li> <li>▪ The process is repeated until a cue is found to discriminate.</li> </ul>	<ul style="list-style-type: none"> <li>▪ The cues are selected in a random order until a cue is found to discriminate between options.</li> <li>▪ The minimalist heuristic requires even less knowledge than Take the Best and Take the Last.</li> </ul>	<ul style="list-style-type: none"> <li>▪ For each option, all positive cues are summed up, and the option with the largest number of positive cues is chosen.</li> <li>▪ All cues have the same value or importance with regards to the decision.</li> </ul>	<ul style="list-style-type: none"> <li>▪ Contrary to Tallying, where all cues weigh the same value, the Weighted Tallying algorithm involves weighing each cue according to its ecological validity. The ecological validity specifies the cue's predictive power, that is, the frequency with which the cue successfully predicts the choice. (Gigerenzer and Goldstein 1996)</li> </ul>	<ul style="list-style-type: none"> <li>▪ This algorithm is similar to Tallying, except that the assignment of option a and option b differs (see Gigerenzer and Goldstein 1996). This algorithm has been considered a good approximation of weighted linear models (Dawes 1979).</li> </ul>	<ul style="list-style-type: none"> <li>▪ This model is similar to the Unit Weight Linear Model and Tallying. However, the value of option a and b are multiplied by their ecological validity.</li> </ul>	<ul style="list-style-type: none"> <li>▪ Multiple regression accounts for the different validities of the cues.</li> <li>▪ Multiple regression assigns weights to each option corresponding to the covariance between the cues.</li> </ul>

<sup>3</sup> Cues refer to the attributes of each option.



importance, until one attribute is found that can discriminate between options. The Tallying algorithm requires that all determining attributes for each option are summed-up (Goldstein 1994). According to this algorithm, the option with the largest number of attributes will be chosen. All attributes have the same value or importance with regards to the decision.

### **8.2.2 Prototype Testing With Researchers**

The first working prototypes of the deliberation tools; weighing it up version 1 and your most important reason version 1, were piloted with a stakeholder group of researchers from multi-disciplinary backgrounds: medicine, psychology, health psychology, sociology and health informatics (n=15). The sample consisted of two researchers specialising in shared decision making, eight researchers specialising in health communication, two researchers specialising in health informatics, and three sociologists. A group interview was used to discuss each deliberation tool separately. The researchers were asked to comment on the design and usability of each tool. The data was recorded digitally and was analysed using thematic content analysis. Weighing it up and your most important reason versions 1 were amended following the researchers' comments to create the second working prototype of the deliberation tools (i.e., version 2).

### **8.2.3 Prototype Testing With Health Professionals**

The planned sample of health professionals (n=28) consisted of five consultants in obstetrics and gynaecology, a sonographer, a clinical nurse specialist, ten midwives, two geneticists, six coordinators of the national antenatal screening programme in Wales, England and Scotland, a patient representative and two professionals from national charities offering information and support during the diagnostic phase of pregnancy (Antenatal Results and Choices, Down's Syndrome Association). An email was sent to all 28 individuals, asking them to review the amnioDex website online, paying attention to the deliberation tools: weighing it up (version 2) and your most important reason (version 2). They were asked to

complete a short questionnaire. The 18-item questionnaire was divided into five sections: navigation, layout, video clips, deliberation tools and message board. Participants were asked to provide written feedbacks on each item. The data was analysed using thematic content analysis. For the purpose of this chapter, the analysis was focussed on the items addressing the deliberation tools only. The deliberation tools were amended according to the professionals' comments to develop weighing it up and your most important reason versions 3.

#### **8.2.4 Prototype Testing With Women Facing the Amniocentesis Decision**

Pregnant women who had been offered an amniocentesis were invited to use amnioDex and the deliberation tools (version 3). In one antenatal clinic (University Hospital of Wales, Cardiff), women (any age) who had been offered an amniocentesis after screening tests for Down's syndrome, advanced maternal age or mid-pregnancy ultrasound scan, were informed of the study by midwives. Pregnant women were excluded from the study if they could not read English. Women who indicated an interest in participating were consented and given an interview date. The interview was conducted in two phases. First, participants were asked to use the deliberation tools while verbalising their thoughts using the "think-aloud" method (Cotton and Gresty 2006; Davison et al. 1997). This method required participants to communicate their thoughts as they used the tools, indicating satisfaction, dissatisfaction, difficulties encountered and misunderstandings. The think-aloud technique provides insight into the usability of the products and impacts on cognitions and emotions of the steps required to navigate new technologies (Ericsson and Simon 1984; Fonteyn and Fisher 1995; Funkesson et al. 2007). Second, participants took part in a short semi-structured interview. The interview schedule consisted of eight open-ended questions focusing on women's reactions to the deliberation tools, navigation of the website, comprehension of content and suggestions for improvement. For the purpose of this chapter, the analysis was focussed on

their reactions to the deliberation tools. All interviews were recorded digitally and transcribed by the same researcher (author of this thesis). The interview data was qualitatively analysed using a two-step thematic content analysis (Denzin and Lincoln 2000; Holloway 2005; Pope et al. 2000), assisted by the computer software ATLAS-ti (ATLAS-ti 5.2). The deliberation tools were amended accordingly and weighing it up and your most important reason versions 4 were developed.

## **8.3 Results**

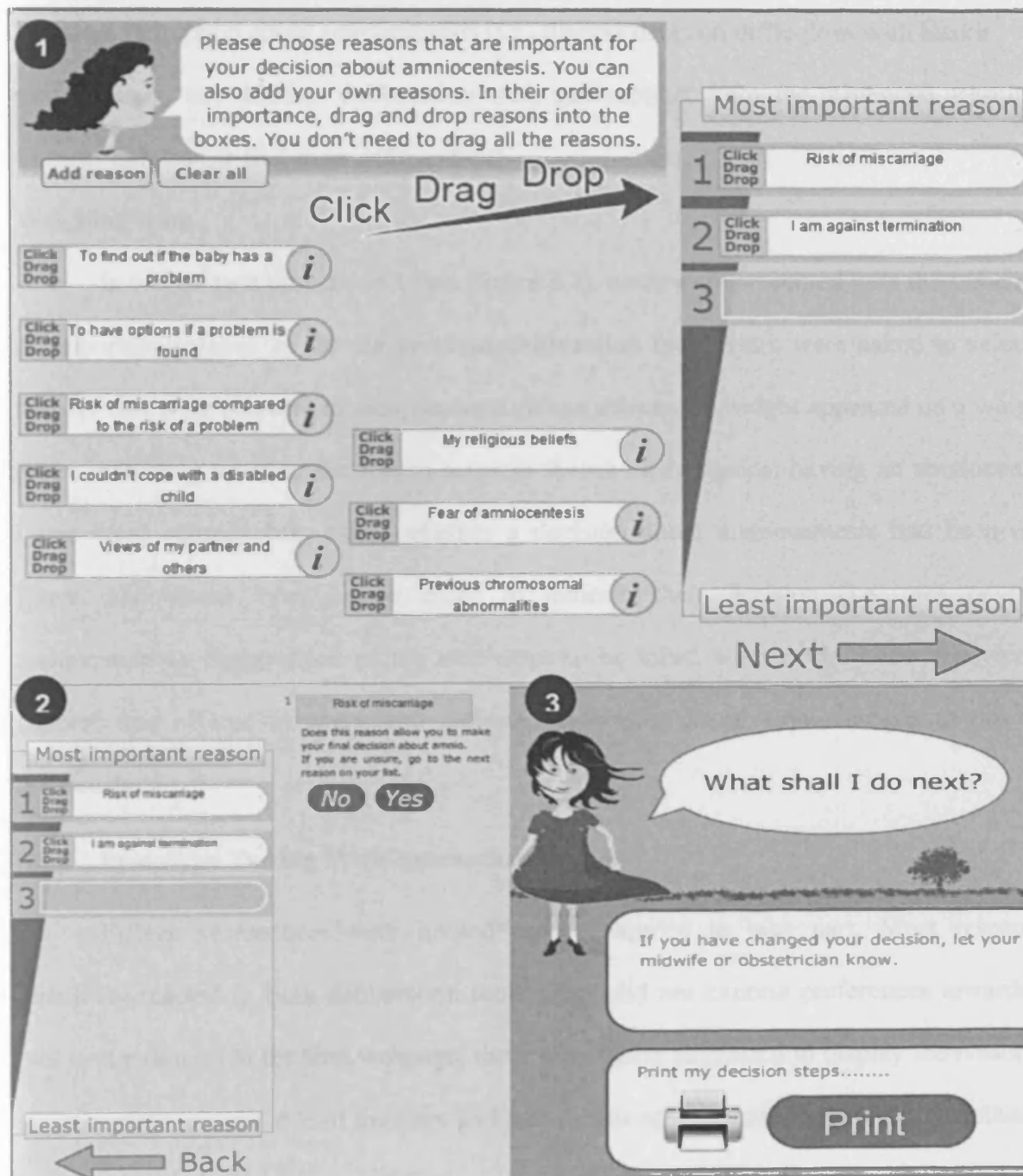
### **8.3.1 Prototype Development**

Key theoretical constructs of the Take the Best and Tallying algorithms respectively guided the design of your most important reason and weighing it up. To increase understanding and usability of the deliberation tools, the terminology was simplified. The term attributes, commonly used to describe the factors that could help form preferences, was replaced by the term reasons. The reasons displayed in both tools were selected from accounts provided in a detailed needs assessment reported in Chapter 5.

#### **Your Most Important Reason**

In your most important reason version 1 (see Figure 8.1), users were presented with a series of important reasons that were considered influential in arriving at a decision to accept or decline amniocentesis testing. The reasons were displayed in boxes with clickable information buttons and more reasons could also be added. Users were asked to choose the reasons that were relevant to their decision making and to rank them in order of importance. The first important reason ranked was automatically selected and a short question generated asking: "Does this reason allow you to make your final decision about amniocentesis?" Users who chose "yes", were asked to indicate their decision (yes or no to amniocentesis). Suggestions of the next steps to be taken were made, such as informing their healthcare provider, reading more about amniocentesis, printing their deliberation pathway or watching

Figure 8.1 Screenshot of Your Most Important Reason Version 1 (Take the Best)



enacted quotes of women's stories. Further advice and support was offered to users who indicated indecision about amniocentesis (i.e., discuss decision difficulties with health professionals, use another deliberation tool, get support from the Antenatal Result and Choices helpline or find more information).

### **Weighing it up**

In weighing it up version 1 (see Figure 8.2), users were presented with the same series of important reasons as for the previous deliberation tool. Users were asked to select the reasons that were relevant to their decision. When selected, a weight appeared on a weighing scale, indicating whether the reason acted in favour of, or against having an amniocentesis. Users were subsequently asked whether a decision about amniocentesis had been made. Users who chose "yes", were asked to indicate their decision (i.e., yes or no to amniocentesis). Suggestions of the next steps to be taken were made. Further advice and support was offered to users who indicated indecision about amniocentesis as described previously.

### **8.3.2 Prototype Testing With Researchers**

Fifteen researchers were invited and 10 agreed to take part. Most researchers positively reacted to both deliberation tools. They did not express preferences towards one tool or the other. On the first webpage, three researchers suggested to display the reasons for amniocentesis on one side of the page and the reasons against amniocentesis on the other side because they believed this would facilitate the differentiation between options. All remaining researchers were satisfied with the way reasons were presented and felt that it was appropriate to display the reasons in a random order, without making a distinction between reasons for and against amniocentesis. To avoid increasing the number of reasons displayed or affecting the design and layout, the presentation of reasons was kept unchanged (i.e., reasons were presented randomly without distinguishing between reasons for and against

Figure 8.2 Screenshot of *Weighing it up Version 1 (Tallying)*

Please choose reasons that are important for your decision about amniocentesis. You can also add your own reasons. Drag and drop the reasons you think would be in favour of amniocentesis into the green bowl. Drag the reasons you think would be against amniocentesis to the red bowl. You don't need to drag all the reasons.

Click Drag Drop I couldn't cope with a disabled *i*

Click Drag Drop Previous chromosomal *i*

Click Drag Drop To find out if the baby has a

Click Drag Drop To have options if a problem is

Click Drag Drop Risk of miscarriage compared *i*

Click Drag Drop Views of my partner and *i*

Click Drag Drop Risk of miscarriage

Click Drag Drop I am against termination *i*

Click Drag Drop My religious beliefs *i*

Click Drag Drop Fear of amniocentesis *i*

Clear all    Add reason

**Click Drag Drop**

Choose the reasons that help you decide and put them in the weighing scales.

**Amniocentesis**

1

to amniocentesis

**No amniocentesis**

2

Next

What shall I do next?

If you have changed your decision, let your midwife or obstetrician know.

If you decide to have an amniocentesis, you might want to:

[Find out more about amniocentesis](#)

Print my decision steps.....

Print

[Women's stories...](#)

amniocentesis). Most researchers considered that instructions and textual content of weighing it up and your most important reason could be improved. The overall textual content and instructions appearing on the first page of each tool were amended to increase usability and meet the requirements of the Plain English Campaign (Cutts 1995). Furthermore, one researcher suggested the addition of a short demo for each deliberation tool, because it could not be guaranteed that users would systematically read the instructions before using the tools. A short demo was added, accessible after users clicked on the demo button appearing on each deliberation tool. All amendments described above were integrated into the deliberation tools to produce the second versions of weighing it up and your most important reason.

### **8.3.3 Prototype Testing With Health Professionals**

Twenty eight health professionals were invited and nine professionals agreed to review the website and embedded deliberation tools (version 2). The sample consisted of two midwives, a consultant in obstetrics and gynaecology, five professionals from the national screening programmes in England, Scotland and Wales, and the director of a national charity (Antenatal Results and Choices). Five health professionals expressed concerns regarding the clarity and usability of the tools. For both tools, the instructions were deemed unclear and confusing. Five out of nine professionals reported difficulties understanding how to use the deliberation tools. Two professionals even questioned the necessity to integrate such tools on the website, as they feared the tools would confuse rather than help pregnant women. One out of nine professionals reported preferring weighing it up to your most important reason. One out of nine professionals reported finding the tools very useful, after having read and understood the instructions. Four professionals considered that both deliberation tools would prove beneficial in clarifying women's thoughts and facilitating decision making. Two out of nine professionals insisted on the necessity to review the demo before using the tools and avoid the initial confusion they experienced. They suggested integrating a mandatory demo in

each deliberation tool. Following the professionals' comments, the textual content and instructions were amended to increase clarity and usability. The demo button was made more obvious to encourage users to watch the demo before using the tools. These changes were incorporated into the deliberation tools to produce the third versions.

### **8.3.4 Prototype Testing With Women Facing the Amniocentesis Decision**

In the participating antenatal clinic, 14 pregnant women who had recently been offered an amniocentesis were invited to take part and 10 women agreed to be interviewed. Pregnant women used the deliberation tools versions 3. Nine participants took part in a phone interview and one participant attended a face to face interview. Five women decided to undergo amniocentesis and five women declined the test. Interviews lasted between 17 and 75 minutes (29 minutes on average). The mean age of women in the sample was 36.7 years (range 34-41 years, standard deviation 2.4). Eight women were British, one woman was Filipino, and one woman was Algerian. The mean gestational age, at the time of interview, was 18 weeks (range 17-19 weeks, standard deviation 0.5). The demographic characteristics of the participants are summarised in Table 8.2. Five themes were identified: benefits of the deliberation tools, disadvantages of the deliberation tools, difficulties using the deliberation tools, preferences for one tool over the other, suggestions for improvement (see Table 8.3).

All participants used both deliberation tools in no particular order. Seven out of 10 pregnant women found the deliberation tools helpful: in weighing the pros and cons of options (n=4), in making a decision (n=2), confirming the decision made (n=2), providing a comprehensive list of reasons (n=2) and generally facilitating understanding (n=1).

"It was good to do that and see that, for me, everything went towards the no, not having it. I mean, it [weighing it up] just helped me make the decision basically. It's just nice to be able to make the decision by using different ways of doing it, just to understand it a little bit more." (F, age 37, declined amniocentesis)



Table 8.2 *Characteristics of Women Interviewed (Field Test of the Deliberation Tools)*

<b>Characteristics</b>	<b>Sample size</b>
Amniocentesis	
- Accepted	5
- Declined	5
Marital status	
- Married or engaged	5
- Living with partner	5
Nationality	
- British	8
- Other (Filipino, Algerian)	2
Number of children	
- 0	2
- 1	6
- 2	2
Existing children with a chromosome disorder	0
Obstetric History	
- Previous miscarriage	1
- Previous amniocentesis	0

Table 8.3 *Themes Identified in Interviews With Pregnant Women (Field Test of the Deliberation Tools)*

<b>Themes</b>	<b>Sub-themes</b>
Benefits of the deliberation tools	<ul style="list-style-type: none"> <li>- Decision making process facilitated</li> <li>- Decision outcome visualised</li> <li>- Increased clarity</li> <li>- List of reasons</li> </ul>
Disadvantages of the deliberation tools	<ul style="list-style-type: none"> <li>- Complexity</li> <li>- Incompatible with such emotional decision</li> <li>- Artificial process</li> </ul>
Difficulties using the deliberation tools	<ul style="list-style-type: none"> <li>- Usability</li> <li>- Understanding difficulties</li> <li>- Technical difficulties</li> </ul>
Preferences for one tool over the other	<ul style="list-style-type: none"> <li>- Advantages of weighing it up</li> </ul>

“I think it could be quite useful, because, I guess, to look at it nice and clear, it gives you a way of just putting it all in and putting your thoughts down, in front of you.” (F, age 37, declined amniocentesis)

Seven out of ten pregnant women felt the instructions were generally clear and the tools easy to use. The majority of users did not watch the demo but nine out of ten users thoroughly read the instructions.

“It was fine to use, dead simple!” (F, age 34, undertook amniocentesis)

Two users considered the list of reasons helpful in clarifying their thoughts about amniocentesis testing. While they already knew the main reasons/factors for accepting or declining an amniocentesis, visualising the list was deemed helpful in achieving decision making.

“The best bit which helped compared to the leaflets was those little cartoons down at the bottom [weighing it up and your most important reason]. It’s got all the reasons that you were thinking of, in your brain, that were all messed up, so it lists it, so you know what those reasons are, you just couldn’t think straight at the time.” (F, age 34, undertook amniocentesis)

Furthermore, seven out of ten users expressed preferences towards weighing it up over your most important reason.

“I like the weighing scales. I found that one a little better to use, just because it’s more visible as you do it rather than, the other one, you’ve got to wait till the end to know what the result is. But, yes, I like the weighing scales.” (F, age 34, declined amniocentesis).

They felt that weighing it up was more immediate, intuitive and helpful in visualising the decision. They perceived the movements of the weighing scales as facilitating the trade off between options. Pregnant women considered that weighing it up enabled them to visualise their decision making process (e.g., movements of the scales during deliberation) and the final outcome, as reflected by the arrow on the weighing scale: leaning towards amniocentesis or not.

“I think the first one [weighing it up] was more immediate in, kind of, putting it visually in front of you, in making a decision and putting down the pros and cons. I guess that just depends on how you... I don’t know. To me, the first one actually was better, the most useful, interesting.” (F, age 37, declined amniocentesis)

Three users indicated that your most important reason was more complex and instructions seemed less clear than weighing it up. They felt that your most important reason was less immediate and required more effort and focus than weighing it up. Two out of ten pregnant women reported difficulties ranking their reasons in order of importance.

Three pregnant women out of ten found the tools unhelpful in making a decision about amniocentesis. They felt the tools were overly complex and caused confusion instead of facilitating understanding. One woman considered the tools too clinical to use for such an important and emotional decision as amniocentesis.

“I think it’s such an emotive subject that, you know, in your head you go through it all and you work out what you want to do and how you want to deal with the problem if there is a problem. But would I like to actually physically weigh the pros and cons and things like that? No, because it feels too clinical and you know, I’m not deciding whether to keep a boyfriend or dump a boyfriend, I’m trying to decide what I’m going to do if my baby has got a problem.” (F, age 35, undertook amniocentesis)

Three out of ten women reported difficulties understanding how to use the tools. One woman out of three did not read the instructions before using the tools. They all considered that the layout and design of the tools were too complex and experienced significant difficulties navigating the tools. They also felt that the tools required a high level of concentration that was not necessarily possible at this stage of the pregnancy.

“Especially for people who are not working with computers, they’re going to find that hard. The thing is, sometimes when you’re pregnant, you are all over the place, do you see what I mean? My concentration is not as good.” (F, age 37, undertook amniocentesis)

Two out of ten women experienced difficulties dragging and dropping boxes in the column (i.e., your most important reason) or on the weighing scales (i.e., weighing it up).

“I am not sure about that [pointing to the click, drag and drop box] I find that quite complicated and I work on computers but I think until you’re familiar with it...I find that part quite difficult.” (F, age 37, undertook amniocentesis)

Finally, pregnant women experienced the most difficulty using your most important reason. Two out ten women reported difficulties selecting and ranking reasons in order of importance. They tended to select all reasons, including those that were not relevant to their decision. Both participants suggested to add a box labelled “does not apply” where all irrelevant reasons could be placed. The deliberation tools were modified according to women’s comments. The action to drag and drop boxes was replaced by a column where users ticked the reasons that applied to them. The overall design and layout of the tools was simplified to increase usability on the basis of these comments and version 4 of the deliberation tools (final version) was created (see Figures 8.3 and 8.4).

#### **8.4 Discussion**

The findings showed that heuristic-based algorithms could successfully guide the design of interactive deliberation tools but also indicated difficulties and challenges posed by the translation of theoretical constructs into usable interactive methods. The evaluation of the deliberation tools differed across the stakeholder groups. Most researchers and pregnant women positively evaluated the tools while the majority of health professionals expressed concerns about their clarity and usability. While your most important reason (Take the Best) was based on a simpler and presumably more intuitive decision algorithm, the majority of women explicitly preferred weighing it up to your most important reason.

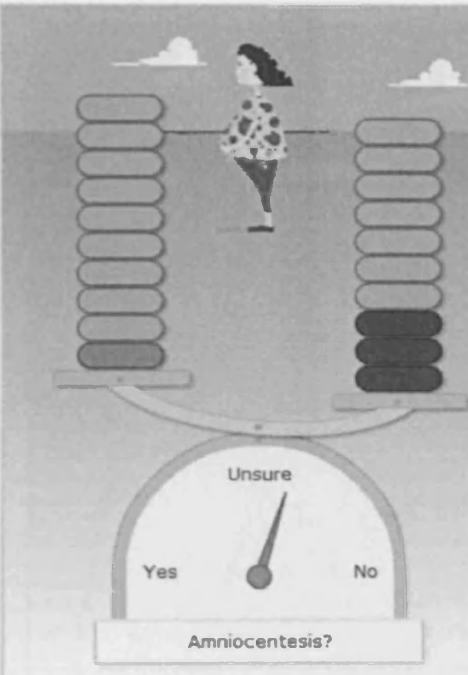
The research findings suggested that the success of this translation largely depended on effectively dealing with the challenges this process generated. Translating abstract mental steps into an acceptable interactive interface proved difficult for the web designers and researchers. Each mental step required extensive discussions and iterative modifications. To comply with the principles of bounded rationality, the tools had to remain simple and fast while mirroring each algorithm’s cognitive steps. However, creating a graphic

Figure 8.3 Screenshot of Weighing it up Version 4 (Tallying)

1
Click here for Demo

Reasons For and Against Amniocentesis	For	Against	Clear
To find out if the baby has a chromosome problem <span style="float: right; font-size: 18px;">?</span>	<input checked="" type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
To have options if a problem is found <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
I couldn't cope with a disabled child <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Previous chromosomal abnormality <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Views of my partner and loved ones <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Risk of miscarriage compared to risk of chromosomal abnormality <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Risk of miscarriage <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input checked="" type="checkbox"/>	<input type="checkbox"/>
I am against termination <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
My religious beliefs <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input checked="" type="checkbox"/>	<input type="checkbox"/>
Fear of amniocentesis <span style="float: right; font-size: 18px;">?</span>	<input type="checkbox"/>	<input checked="" type="checkbox"/>	<input type="checkbox"/>

Enter Other Reasons
▼
Next >

2


Among the reasons listed below, choose the ones that are important for you. Tick whether the reason is for or against amniocentesis. You don't have to tick every box.

The scales show you have more reasons in favour of not having an amniocentesis.

Have you made a decision about amniocentesis?

Yes
No

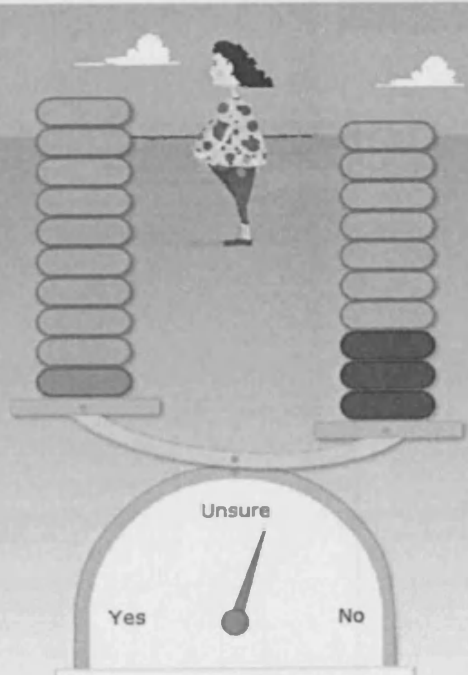


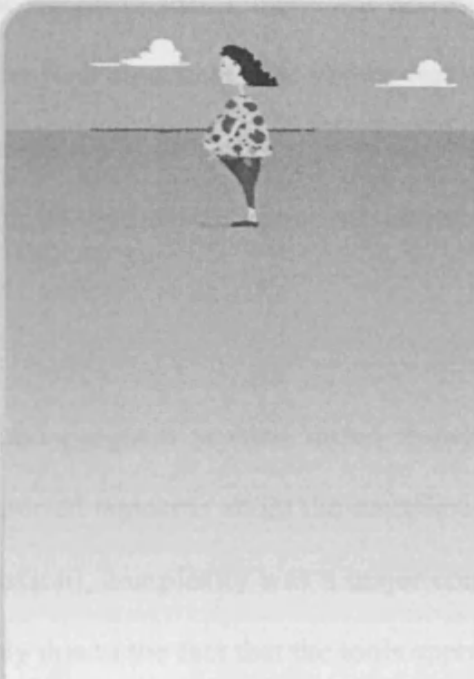
Figure 8.4 Screenshot of Your Most Important Reason Version 4 (Take the Best)

1
Click here for Demo

Please choose reasons that are important for your decision about amniocentesis. You can also add your own reasons. Select a reason for each box in their order of importance. You don't have to use all the boxes.

1	Please select ...	▼
2	Please select ...	▼
3	Please select ...	▼
4	Please select ...	▼
5	Please select ...	▼
6	Please select ...	▼
7	Please select ...	▼
8	Please select ...	▼
9	Please select ...	▼
10	Please select ...	▼

Reset
Next >



2
DEMO

	Your Reasons	Decision Reached?
1	To find out if the baby has a chromosome problem <span style="float: right;">?</span>	
2	I couldn't cope with a disabled child <span style="float: right;">?</span>	No
3	Previous chromosomal abnormality <span style="float: right;">?</span>	
4	Please select ...	
5	Please select ...	
6	Please select ...	
7	Please select ...	
8	Please select ...	
9	Please select ...	
10	Please select ...	

< Back

Does this reason allow you to make your final decision about amniocentesis?

Yes
No

representation of the second step of the Take the Best algorithm (where each attribute is reviewed in order of importance until an attribute is found to discriminate between options) was complex because it was abstract and ambiguous. While creating a graphic representation of the Tallying algorithm was straightforward (i.e., a weighing scale), there was no obvious and unequivocal graphic representation of the Take the Best algorithm. The choice was made to create a column where important reasons were dragged and dropped (version 1), without knowing whether the chosen representation would be optimal in terms of clarity and usability.

During the field test, health professionals and pregnant women raised translation issues. While only a minority of pregnant women reported concerns about the complexity of one of the deliberation tools (your most important reason), complexity was a major concern for health professionals. The difference here was likely due to the fact that the tools appraised by pregnant women (version 3) had been revised using health professionals' suggestions for improvement and consequently achieved higher usability. Divergent perceptions may also be attributed to the differing opinions, interests and information needs of pregnant women and health professionals. It was seen in Chapter 6 that health professionals had strong and often diverging opinions about the nature and quantity of information needed on the range of chromosomal abnormalities tested, elective termination of pregnancy and risk of miscarriage to quote. Given that the deliberation tools provided comprehensive information about potentially controversial topics (e.g., elective pregnancy termination) professionals may have feared that this resource would be inconsistent and interfere with information received during the medical consultation. This finding highlights, as documented in Chapter 6, the professionals' control over information and tendency to adopt paternalistic attitudes. Furthermore, processes or informational contents that are not specifically insightful or

relevant to health professionals may have been perceived as very relevant to decision making among pregnant women (e.g., clear list of reasons). The structure and guidance provided by the deliberation tools may have offered a form of decision support that health professionals did not necessarily identify or consider helpful (e.g., visualising the decision making process and outcome on the weighing scale), possibly because of their expertise and greater understanding of the consequences and implications. The gap between pregnant women's and health professionals' perceptions and interests has been documented in the literature and is consistent with the present finding (Hunt et al. 2005; St-Jacques et al. 2008).

While stakeholders and health professionals did not express clear preferences towards one tool or the other, most pregnant women reported preferences for weighing it up over your most important reason. The majority of pregnant women felt that weighing it up offered a more immediate and intuitive way of weighing the pros and cons of amniocentesis and visualising the decision. The movement of the weighing scales was deemed helpful in facilitating the trade off between options. In your most important reason, some women reported difficulties ranking the reasons in order of importance and comprehending the instructions. Your most important reason took longer to complete and required more concentration than weighing it up. According to the Take the Best algorithm and underlying principles of unbounded rationality, the task of ranking attributes (here called reasons) in order of importance and finding an attribute that discriminates between options, should be fast, simple and completed with limited cognitive effort. The translation of the Take the Best algorithm into your most important reason failed to comply with the above principles. Converting the Tallying algorithm into graphic based element resulted in a simple, intuitive and visually efficient object: the weighing scale. However, transferring the Take the Best heuristic into a simple, fast and intuitive tool proved more difficult. The present findings



point to a paradox. While the Take the Best heuristic is a simpler decision algorithm and one that requires less cognitive effort than Tallying, its translation into your most important reason was more complex and less intuitive. This highlights the difficulty of translating abstract theoretical constructs into usable tools and points to the necessity to field-test complex interventions before assuming that those interventions are appropriate and usable by patients/users. Although seven out of ten pregnant women expressed preferences towards weighing it up, not all comments concerning your most important reason were negative. Therefore, a decision was made to keep your most important reason for further empirical testing. Conceptually, and based on its specified performance and predictive accuracy on simulated decision tasks (Gigerenzer and Goldstein 1996), the take the best heuristic, which guided the development of your most important reason, works. The difficulty seems to lie in the translation and operationalisation of the heuristic into an interactive tool. Further work is needed to investigate the issues associated with the translation of abstract mental steps into usable tools.

In the literature, the development of heuristic-based deliberation tools has not been documented. However, one study compared the effectiveness of a heuristic-based DESI (Take the Best) with one based on the analytic hierarchy process (method derived from a normative theory of decision making) for a decision to undertake colorectal cancer screening (Galesic et al. 2008). The analytic hierarchy process DESI described options and attributes and consisted of pair-wise comparisons of all options and attributes (Dolan and Frisina 2002). The Take the Best version of the DESI described options and attributes and asked users to select the most important attribute and identify the option that best satisfied the chosen attribute. Participants were asked to read one of the three DESIs and to indicate their current screening decision. The results indicated that the Take the Best DESI predicted the final

decision better than the analytic hierarchy process. Since information about the transfer of the Take the Best heuristic into a practical intervention was missing, a comparison with your most important reason was not possible. However, the results suggested that heuristic-based approaches effectively predicted decision making in the context of health decisions.

The strengths of the study were the innovative approach used in developing deliberation tools and the diverse nature of the sample. As far as could be determined, heuristic-based algorithms have never been used to develop deliberation tools. The present study is pioneering as an attempt to transfer theoretical constructs into usable DESI's components. Furthermore, the deliberation tools were field-tested with three different groups of users: researchers, health professionals and potential users (women considering amniocentesis testing). One therefore expects that major dysfunctions and understanding difficulties would have been addressed from all relevant view points.

A limitation was the comparison difficulties generated by the iterative approach of the field test. The groups of users evaluated incremental versions of the deliberation tools, which subsequently compromised direct comparisons between the groups. Another possible limitation may be the multiple methods used to collect data. However, given stakeholders and health professionals' time constraints and overall recruitment difficulties, adopting methods that were convenient for each group seemed essential and non-negotiable.

## **8.5 Conclusion**

The translation of theoretical constructs into graphic-based deliberation tools was possible. However, field-testing revealed that the tool's usability highly depended on the accuracy and feasibility of the translation. The practical transfer of the Tallying algorithm into an interactive interface led to the development of weighing it up, which reportedly

facilitated decision making and complied with the principles of ecological validity. Weighing it up may be used in other healthcare contexts where a decision between two treatments or screening options has to be made. However, the translation of the Take the Best algorithm into your most important reason proved more problematic and pointed to the need to field-test complex interventions before evaluation and implementation in clinical settings. The translation difficulties inherent in this process may be the major obstacles in designing theory-based DESIs. If there is to be success in translating theory into practical interventions, there will need to be significant commitment among stakeholders and user-groups to collaboratively develop usable interventions. There is scope for examining the translation issues associated with theory-based interactive decision tools. At this stage, it is not possible to ascertain whether one approach (i.e., Take the Best or Tallying) is superior to the other. Further research is needed to compare how different interactive translations of heuristic-based algorithms may influence or even facilitate decision making.

# Chapter 9

## Field-Testing of AmnioDex

### 9.1 Introduction

Field-testing is increasingly recognised as a necessary assessment and validation of the quality and usability of DESIs. Field-testing is described as a “live” testing of a prototype DESI (Evans et al. 2007) which involves showing the newly developed intervention to potential users who comment on its content and usability in order to amend it accordingly. The necessity to field-test DESIs prior to evaluating and implementing them has been recognised by the IPDAS collaboration (Elwyn et al. 2006). Nevertheless, the value of field-testing is yet to be shown as only a minority of shared decision making interventions seem to undergo field-testing. As highlighted in Chapter 4, the majority of interventions included in the Cochrane systematic review of decision aids did not undergo field-testing prior to evaluation. Few studies to date have highlighted the necessity to field-test DESIs (Evans et al. 2007; Kim et al. 2005) and applied guidelines on how to field-test complex interactive interventions are not yet available.

The IPDAS collaboration suggests that DESIs should be field-tested with patients/users who are currently facing the decision addressed by the intervention and with health professionals from relevant disciplines. However, the IPDAS collaboration does not provide guidelines about methods for field-testing DESIs, about the number of users needed nor offers a clear definition of field-testing. Evans et al. developed a model of field-testing which distinguishes between (1) exploratory field-testing and (2) prototype field-testing (Evans et al. 2007). Exploratory field-testing consists of asking users to look at and comment on early prototypes of specific intervention’s components, before the first prototype DESI has

been developed. During prototype field-testing (stage 2), users are shown successive prototypes of the intervention. This model is the most detailed account of field-testing to date but does not provide clear guidance as to which methods to use to obtain optimal results.

In the broader context of human-computer interactions and interactive health technologies, usability tests are frequently conducted, using various methods and explicit guidelines (Gray and Salzman 1998). Both field tests and usability tests are intended to increase usability and acceptance of the newly developed intervention. However, field tests also examine the relevance and quality of the content and are not limited to web-based interventions. Contrary to field tests, usability tests involve specific and well defined expert-based or user-based methods. Heuristic evaluations and cognitive walk-through are the most widely adopted expert-based methods while the think-aloud technique is considered the most popular user-based method (Gray and Salzman 1998; Maguire 2001; Nielsen 1993). Heuristic evaluations consist of assessing an intervention twice against a list of usability principles or heuristics. In other words, experts or users are asked to assess and comment on the usability of the interface using simple rules or heuristics such as: (1) Does the interface/website use simple and natural dialogue? (2) Does it speak the user's language? (3) Is the language consistent throughout the interface? (Jaspers 2009). The cognitive walkthrough is an expert evaluation of the cognitive processes required by potential users to navigate the intervention. The expert is required to use the intervention without any guidance and behave as a novice user. Finally, the think-aloud method is a user-based method that requires users to navigate the intervention while systematically verbalising their thoughts (Cotton and Gresty 2006; Ericsson and Simon 1984). A study comparing the most common expert and user-based methods suggested that an expert-based heuristic evaluation was the simplest and most cost-

effective method and should be combined with a user-based think-aloud method for optimal usability testing (Jaspers 2009).

The objectives of this chapter were to: (1) field-test the entire amnioDex website with lay users, healthcare providers and women who had been offered an amniocentesis, (2) assess their reactions to the DESI's content, design and usability, and (3) make progress towards elaborating concrete guidelines for field-testing complex interventions.

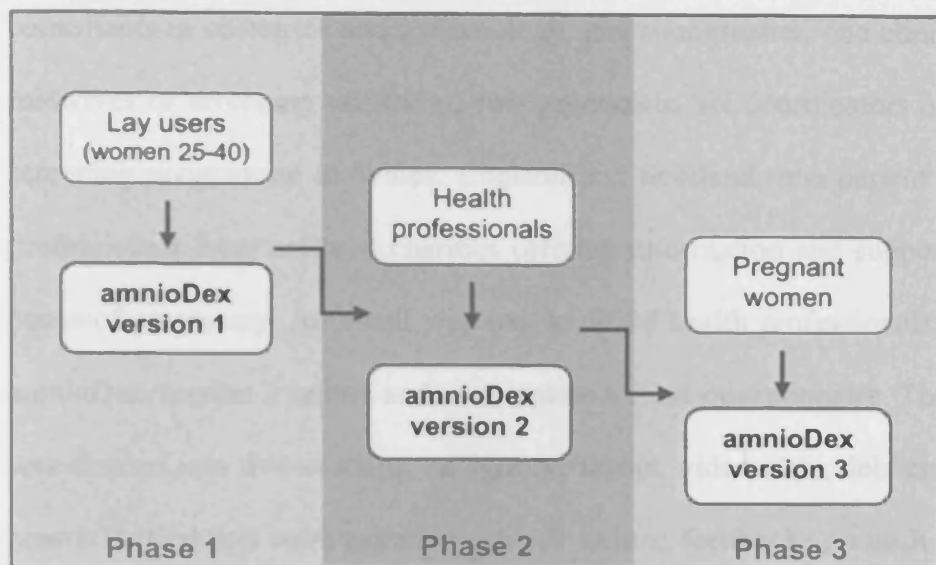
## **9.2 Methods**

The field test was divided into three independent phases to address issues from a technical, professional and user point of view: (1) field-testing with lay users, (2) field-testing with health professionals and (3) field-testing with pregnant women who had been offered amniocentesis testing. A qualitative approach was adopted. Participants were invited to use incremental prototypes of amnioDex and comment on the usability, textual content, potential dysfunctions and possible improvements. All findings related to the deliberation tools are reported in Chapter 8. An improved version of amnioDex was systematically developed before testing the intervention with the next group of users (see Figure 9.1). The study protocol and materials were reviewed and approved by the Research and Development committees of the participating sites (Cardiff and Vale NHS Trust, Velindre NHS trust) and by the National Research Ethics Service.

### **9.2.1 AmnioDex Version 1 Field-Tested With Lay Users**

In order to mirror the broad characteristics of women facing the amniocentesis decision, women aged 25 to 40 were recruited in the Department of Primary Care and Public Health (Cardiff University) on a voluntary basis. Lay users were invited to use amnioDex version 1 in the presence of a researcher (author of this thesis) and to verbalise their thoughts

Figure 9.1 Method Used to Field-Test AmnioDex



while navigating the website (think-aloud technique). The think-aloud technique required participants to communicate their thoughts while they used the intervention, indicating satisfaction, dissatisfaction, difficulties encountered and misunderstandings (Cotton and Gresty 2006; Davison et al. 1997). The think-aloud technique was selected for its widely recognised efficacy in usability tests of interactive health technologies (Jaspers 2009). Lay users were specifically requested to focus on the navigation, design of the website, potential dysfunctions, and ways of optimising usability. They were asked not to focus on the textual content given this was of low relevance to them. Field notes were taken while lay users navigated the website. Lay users were recruited until no new dysfunction was identified or new suggestion for improvement made. The data was qualitatively analysed using a thematic content analysis assisted by the computer software ATLAS-ti. The website was amended according to the lay users' comments to develop AmnioDex version 2.

### 9.2.2 AmnioDex Version 2 Field-Tested With Health Professionals

A sample of 28 healthcare providers and professionals from the Policy and Public Health sector identified through networking and steering group meetings in England and

Wales was invited to review the amnioDex website. The planned sample consisted of five consultants in obstetrics and gynaecology, one sonographer, one clinical nurse specialist, ten midwives or screening midwives, two geneticists, six coordinators of the national antenatal screening programme in Wales, England and Scotland, one patient representative and two professionals from national charities offering information and support during the diagnostic phase of pregnancy. An email was sent to all 28 health professionals, asking them to review amnioDex version 2 online and to complete a short questionnaire. The 18-item questionnaire was divided into five sections: navigation, layout, video clips, deliberation tools and message board. Participants were asked to provide written feedbacks on each item (see questionnaire in Appendix 4). The qualitative analysis was as described in section 9.2.1 (p. 149). AmnioDex version 2 was modified according to health professionals' comments and suggestions for improvement to develop amnioDex version 3.

### **9.2.3 AmnioDex Version 3 Field-Tested With Pregnant Women**

Pregnant women were identified and approached by midwives or screening midwives in one antenatal clinic (University Hospital Wales, Cardiff). All pregnant women (any age) who had been offered an amniocentesis after screening tests for Down's syndrome, advanced maternal age or mid-pregnancy ultrasound scan, were informed of the study by midwives. Pregnant women were excluded from the study if they could not read English. Women who indicated their interest to take part in the study were contacted by research staff to complete a consent process and were given an interview date. The interview was conducted in two phases. First, women were invited to use amnioDex version 3 while verbalising their thoughts using the think-aloud technique. Arrangements were made for them to view the website at home in the presence of a researcher, or during a telephone interview. After using amnioDex, participants took part in a semi-structured interview (face to face or telephone) investigating their reactions to the intervention (satisfaction/dissatisfaction), navigation or understanding



difficulties and suggestions for improvement. The interview schedule consisted of eight open-ended questions investigating women's reactions to the website, its navigation, design and usability. Special attention was given to women's understanding of information provided and suggestions for improving design, navigation and content. All interviews were conducted, recorded digitally and transcribed by the author of this thesis (see interview schedule in Appendix 4).

### **9.3 Results**

The presentation of results is organised in three sections: 1) amnioDex version 1 field-tested with lay users, 2) amnioDex version 2 field-tested with stakeholders and 3) amnioDex version 3 field-tested with pregnant women.

#### **9.3.1 AmnioDex Version 1 Field-Tested With Lay Users**

Eight women aged 25 to 40 (30.2 years old on average; range 25-38; standard deviation 5.1) agreed to navigate amnioDex version 1 using the think-aloud technique (see Figure 9.2). They spent between 10 and 45 minutes navigating amnioDex version 1 (28 minutes on average). Comments were categorised into four themes: design and layout, interactive graphic elements, presentation of information and technical difficulties (see Table 9.1). Six out of eight lay users identified major dysfunctions and suggested improvements. Two lay users considered that amnioDex version 1 was a good and user-friendly website and did not identify any dysfunctions or elements that needed improving.

Four out of eight lay users identified basic design and layout elements that needed improving: removing gaps and blank spaces (n=4), improving the navigation structure by making the bottom and top tabulations more salient (n=2), centring images and aligning the

Figure 9.2 Screenshot of the AmnioDex Homepage Version 1

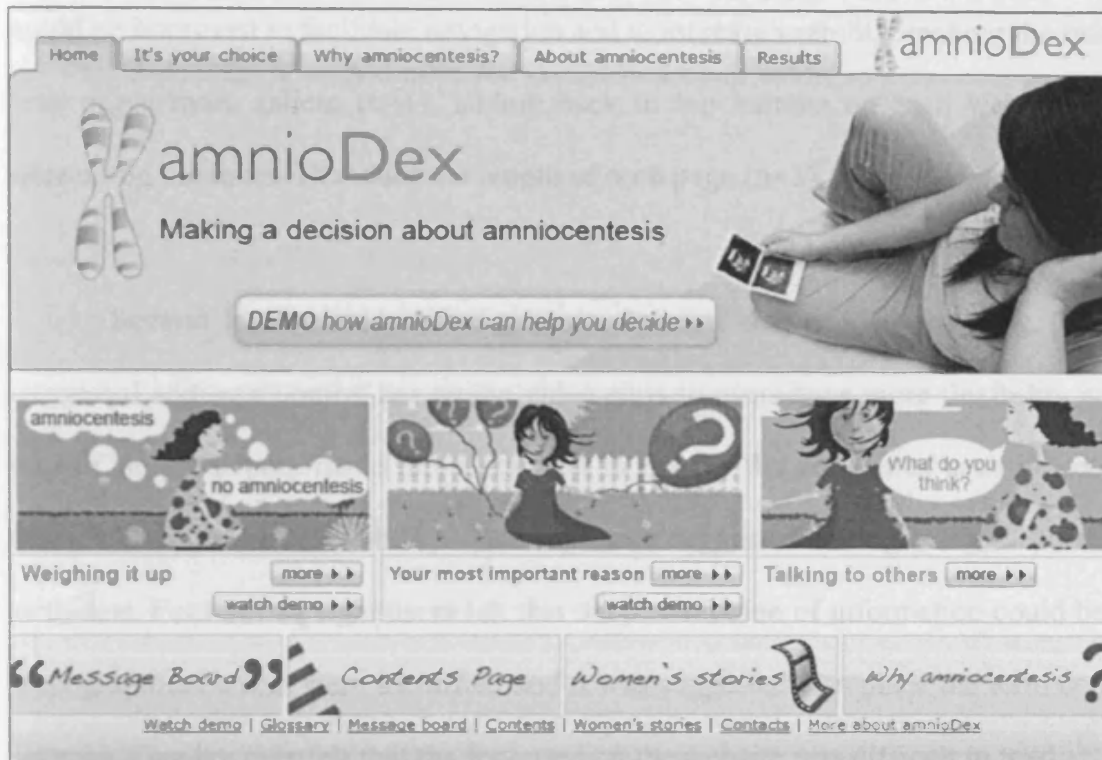


Table 9.1 Themes Identified in the Field-Testing With Lay Users

Themes	Sub-themes
Design and layout	<ul style="list-style-type: none"> <li>- Improving basic page lay out</li> <li>- Optimising the navigation</li> </ul>
Interactive graphic elements	<ul style="list-style-type: none"> <li>- Improving video clips</li> <li>- Improving diagrams</li> <li>- Images</li> </ul>
Presentation of information	<ul style="list-style-type: none"> <li>- Correcting typographical errors</li> </ul>
Technical difficulties	<ul style="list-style-type: none"> <li>- External/internal links</li> <li>- Interactive elements</li> </ul>

video clips with the textual content (n=2). Lay users identified elements of the design that could be improved to facilitate navigation and to increase usability: making the links to other web pages more salient (n=1), adding back to top buttons on each web page (n=1) and decreasing the indent to reduce the length of each page (n=1).

Several lay users identified graphic elements that needed changing. Two lay users suggested adding a control bar on the video clips so users have more flexibility watching the clips. One participant made the suggestion to display the content of each video clip in text version to increase accessibility. Two lay users suggested adding a legend to the diagrams with dots. Four out of eight users felt that the presentation of information could be improved. Typographical errors were identified and it was suggested to replace the term content list by sitemap. One lay user felt that the font used on the website was difficult to read.

Four out of eight lay users reported technical difficulties using the website and embedded interactive elements: inactive or inaccurate links, internal or external pages linked inconsistently across the site, elements partially or incorrectly displayed (e.g., diagram with dots, images) and difficulties registering on the message board. Suggestions were made to link internal and external pages in a consistent manner by systematically opening the new linked page in a separate window and to simplify the registration process by collecting minimum information only (i.e., username, email address and password).

### **9.3.2 Outline of Changes Made**

Drawing on lay users' comments, the design and layout of amnioDex was improved by removing all gaps and blank spaces, redesigning the top and bottom tabulations to increase salience (e.g., bolder colours, font size etc), aligning all photos with the textual content to reduce the length of the page and accentuating internal and external links. A control bar was

added on each video clip (e.g., demos, women's stories etc) and all internal and external pages (including video clips) were linked in a consistent manner to increase usability (i.e., page or video clip systematically opened in a new window). Legends were added to all diagrams and the term content list was replaced by site map. The presentation of information was improved by proofreading the website and page indents were reduced to decrease the size of the pages. Back to top buttons were added where necessary. All technical difficulties identified by lay users were addressed. Building on all amendments described above, amnioDex version 2 was developed.

### **9.3.3 AmnioDex Version 2 Field-Tested With Health Professionals**

Twenty eight health professionals were invited to take part and nine professionals agreed to review the website. The sample consisted of two midwives, a consultant in obstetrics and gynaecology, five professionals from the national screening programmes in England, Scotland and Wales, and the director of a national charity (Antenatal Results and Choices). Health professionals reviewed the website online and returned the 18-item questionnaire. Four themes were identified: design and layout, interactive elements, textual content, visual elements (see Table 9.2).

Most health professionals positively appraised the overall design and layout of the website while a minority identified elements that needed improving. Six out of nine health professionals felt the design of the web pages (including the homepage) was efficient and inviting. Six health professionals found the website easy to navigate and self-explanatory. They felt that the colour schemes were aesthetically pleasing and not offensive nor distracting.

“[The website is] colourful enough to be attractive but avoids being distracting.” (F, programme manager, NHS national services Scotland)

Table 9.2 *Themes Identified in the Field-Testing With Health Professionals*

<b>Themes</b>	<b>Sub-themes</b>
Design and layout	- Overall design and layout - Aesthetic - Navigation
Interactive elements	- Message board - Screening test result calculator
Textual content	- Typographical errors - Clarity
Visual elements	- Video clips - Technical difficulties - Images

Three out of nine health professionals considered that the top tabulation was not sufficiently salient, and should be made more prominent. One health professional reported difficulties finding a list of references and another felt that usability would be improved by adding a short description on the homepage of how to navigate the site (e.g., looking for information in the top tabulation, using the deliberation tools etc).

“I think it might be worth having some text on the first page explaining how you can use the resource. I think that as it stands, it’s great if you can hear the demonstration but you may not always be able to hear the sound.” (F, programme associate, Foetal Anomaly Screening Programme, England)

With regards to the interactive elements featured in amnioDex (see Chapter 8 for findings related to the deliberation tools), most health professionals felt that the message board embedded in the website was an excellent resource. They believed that the message board would be helpful to pregnant women facing a decision about amniocentesis as well as health professionals counselling them. Some professionals insisted on the necessity to moderate the message board and ensure that information posted on the forums was clinically correct and ethically/morally appropriate and reported concerns about the anonymity of users leaving messages on the forums.

“This is an excellent concept - is there any guarantee of anonymity in this section?” (F, regional coordinator, Antenatal Screening Wales)

Two out of nine health professionals felt that the functionality available in Understanding Your Result, where the screening result is framed in multiple ways, was extremely helpful and should be made more accessible to users.

Minor typographical errors were reported. One health professional felt that it was not immediately clear what an amniocentesis was and that textual content should be improved to increase clarity. One health professional felt that the website should provide some information about CVS (see Chapter 3). Two health professionals questioned the need for users to see the screening pathway (see Figure 9.3), as they felt this level of detail was not essential when making a decision about amniocentesis testing.

With regards to the visual elements featured on the website, most professionals believed that the video clips were an excellent resource, informative and realistic, with a wide range of experiences given (n=4).

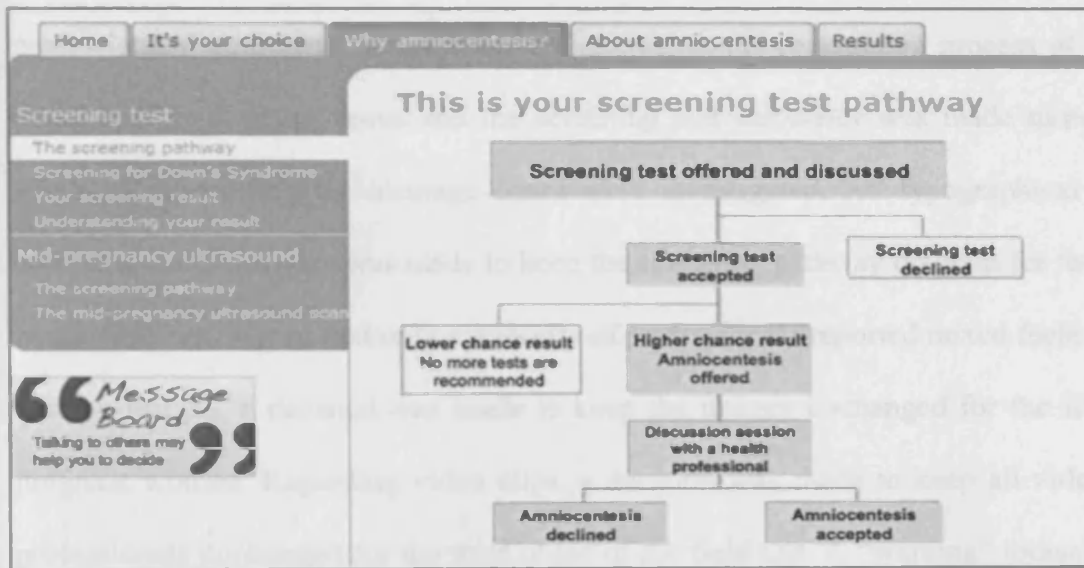
“I loved the videos; I thought they were very heartfelt. I think users would really identify with the video clips. They appear to have tried to give a range of views to give the tool balance.” (F, programme associate, Foetal Anomaly Screening Programme, England)

Several professionals felt that the videos of health professionals could be improved and believed that the genetics video was overly technical and complex.

“The only video that I feel is a bit too technical potentially is the one where the geneticist talks about microdeletions and how a test might not find everything that is wrong with a baby.” (F, programme associate, Foetal Anomaly Screening Programme, England)

One health professional felt that some video clips of health professionals were too subjective and should provide factual information about amniocentesis testing rather than the professional’s opinion.

Figure 9.3 Screenshot of Screening Pathway in AmnioDex Version 2



“These [video clips of health professionals] could be improved. For example the one with the geneticist needs to be providing the info the women required not just giving her opinion on what women should be told.” (F, programme manager, NHS national services Scotland)

Four professionals found the video clip of the amniocentesis procedure useful and informative although two of them expressed concerns about the methods used to perform the amniocentesis. One health professional felt that a live video of an amniocentesis procedure might be too frightening for pregnant women to watch. Several health professionals reported technical difficulties watching the video clips (e.g., poor sound quality, slow download) and identified video clips where the title needed changing. Finally, most health professionals positively reacted to the images displayed on the website although three health professionals reported mixed feelings about the use of cartoon images.

“Personally I found the image with the balloons looked like a child and not immediately obvious it was meant to be a pregnant woman.” (F, programme manager, NHS national services Scotland)

#### 9.3.4 Outline of Changes Made

The majority of health professionals were positive about the design and layout of AmnioDex version 2. Only minor amendments were made. The top tabulation was made more

salient and a more consistent font size was used throughout the website. Drawing on health professionals' comments about interactive elements, the registration process of the message board was made anonymous and the screening test calculator was made more accessible. Ways of moderating the message board were investigated. All typographical errors were corrected and a decision was made to keep the screening pathway diagram for the third phase of the field test. Given that only a minority of professionals reported mixed feelings about the cartoon images, a decision was made to keep the images unchanged for the field test with pregnant women. Regarding video clips, a decision was made to keep all videos of health professionals unchanged for the third phase of the field test. A "warning" message preceding the live amniocentesis video was added: "Warning, this video shows an amniocentesis taking place". All amendments were incorporated into the intervention to develop amnioDex version 3.

### **9.3.5 AmnioDex Version 3 Field-Tested With Pregnant Women**

In the participating antenatal clinics, 24 pregnant women who had recently been offered an amniocentesis were invited to take part and 15 women agreed to be interviewed. Fourteen participants took part in a phone interview and one participant attended a face to face interview. Pregnant women were interviewed between 4 and 20 days following the counselling session where amniocentesis testing was offered and discussed (10 days on average). The intention was to field-test the website with pregnant women who had not yet made a decision about amniocentesis testing. Although pregnant women were contacted on the actual day or the day following the higher chance screening test result (and subsequent offer of an amniocentesis), all pregnant women interviewed had already made a decision about amniocentesis testing at the time of the interview. It was not possible, for practical reasons and often because of heightened stress and anxiety, to schedule an interview date immediately after pregnant women had been offered an amniocentesis.



Ten women chose to undergo amniocentesis and five women declined the test. Interviews lasted between 17 and 78 minutes (32 minutes on average). Time spent navigating the website varied widely. Women spent between 5 and 60 minutes navigating amnioDex. The mean age of women in the sample was 34.9 years (range 27-41 years, standard deviation 3.7). Eleven women were British, one woman was Filipino, one woman was Chinese, one was Indian and one woman was Algerian. The mean gestational age at the time of interview was 17.3 weeks. The demographic characteristics of the participants are summarised in Table 9.3. Four themes were identified during the interview (think-aloud technique and semi-structured interview): positive features, negative features, benefits of amnioDex compared to routine counselling and suggestions for improvements (see Table 9.4).

#### **Positive Features of the AmnioDex Intervention**

Thirteen out of 15 pregnant women who used amnioDex perceived the website as a good and useful resource. Several women believed that amnioDex was an excellent tool, which they would recommend to women in a similar situation.

“I think it’s very good. I think it’s helpful. It’s quite, sort of, user friendly, and it seems quite straightforward and just puts things down fairly simply. Hum, it seems to, kind of address the key things you are worrying about the main things.” (F, age 37, declined amniocentesis)

“It’s good, it’s very good, and if I knew, anyone who was in my situation, I would tell them to go and have a look at it.” (F, age 35, undertook amniocentesis)

“It’s the most comprehensive website I’ve seen on amniocentesis, to be honest.” (F, age 35, undertook amniocentesis)

Women generally perceived the website as user-friendly. One participant valued the fact that amnioDex had been developed in the United Kingdom (as opposed to the United States) and was therefore better tailored to her needs.

“It’s nice, it’s inviting and it doesn’t look American, because when you look on the websites, you’re just trying to find something that relates to you, and what you’re going through, and you know, you’re happy to chat with people who are from abroad and all the rest of it, but actually, what you want to know is what are the things that happen in Wales and in England and what your choices are?” (F, age 35, undertook amniocentesis)

Table 9.3 *Characteristics of Women Interviewed (Field-Testing)*

<b>Characteristics</b>	<b>Sample size</b>
Amniocentesis	
- Accepted	10
- Declined	5
Marital status	
- Married or engaged	8
- Living with partner	6
- Single	1
Nationality	
- British	11
- Other (Filipino, Chinese, Indian Algerian)	4
Number of children	
- 0	4
- 1	9
- 2	2
Existing children with a chromosome disorder	0
Obstetric history	
- Previous miscarriage	1
- Previous amniocentesis	0

Table 9.4 *Themes Identified in the Field-Testing With Pregnant Women*

<b>Themes</b>	<b>Sub-themes</b>
Positive features	<ul style="list-style-type: none"> <li>- AmnioDex in general</li> <li>- Information</li> <li>- Layout and navigation</li> <li>- Graphic-based elements (images, videos, message board etc)</li> </ul>
Negative features	<ul style="list-style-type: none"> <li>- Information</li> <li>- Layout and navigation</li> <li>- Technical dysfunctions</li> </ul>
Suggestions for improvements	<ul style="list-style-type: none"> <li>- AmnioDex in general</li> <li>- Graphic-based elements</li> <li>- Implementation</li> </ul>
Benefits compared to routine counselling	<ul style="list-style-type: none"> <li>- AmnioDex in general</li> <li>- Information</li> <li>- Video clips</li> </ul>

With regards to the informational content, all participants found the information clear and easy to understand. Specifically, information was deemed simple, balanced and easily accessible. Most pregnant women found the information very comprehensive and detailed, and highlighted the quality and value of information addressing the risk of miscarriage and range of chromosomal abnormalities tested for.

“No, as I said, it seems to cover everything, all possible avenues on this. It’s simple to use as well, you don’t have to be a rocket scientist or anything to figure it all out, no I think it’s really good as it is.” (F, age 34, declined amniocentesis)

“I think it’s useful, yeah, no, it’s user friendly, it’s balanced, it’s not too clinical.” (F, age 34, undertook amniocentesis)

Eight out of 15 participants found the website aesthetically pleasing and attractive with a good layout and inviting colour schemes. Most participants (n=12/15) praised the navigation structure and organisation of information sections. They found the website easy to navigate and did not report difficulties finding specific information or simply browsing general information.

“I think that the website is very clean and very easy to navigate and you know, it’s a good resource really.” (F, age 35, undertook amniocentesis)

Concerning graphic-based elements, the majority of pregnant women positively reacted to video clips available on amnioDex, especially videos of women’s stories (n=12/15).

“I think maybe, by using this, especially by listening to women’s stories, you can see why people have it, why people don’t have it. I think it made me understand a little bit more.” (F, age 37, declined amniocentesis)

They felt that videos of women’s stories offered varied and balanced examples of women’s decisions with regards to amniocentesis testing. This feature was considered helpful in making a decision about amniocentesis.

“I watched them. It was good to see all the different kinds of opinions, views of why they wouldn’t do things and obviously some of the women had a Down’s syndrome baby and things like that, that was quite good. It did make you think.” (F, age 37, declined amniocentesis)

“I like the videos on there as well, people who have already had it or who haven’t and have decided against it. So it gives you an all round opinion.” (F, age 34, declined amniocentesis)

Six women considered that the functionality specifically developed to help them understand their screening test result was helpful.

“There’s a section where they give you, your sort of percentage, one in 170 and you can write it in. That was the best, that was the most useful. Understanding your results, that was it.” (F, age 37, declined amniocentesis)

A minority of women commented on the message board, live video of the amniocentesis procedure and diagrams. They perceived the live amniocentesis video as a good resource (n=3) and felt that the message board (n=4) was a good feature provided messages were posted on the forums. The multiple framing of outcome probabilities and diagrams with dots was considered helpful by several pregnant women.

### **Negative Features**

With regards to the informational content, one out of 15 participants considered that amnioDex did not provide comprehensive information.

“It does give you information, if you don’t know anything at all, it doesn’t give you comprehensive details, but then that’s not the aim is it? From there, you can go on to research more yourself. The questions [refers to talking to others] were unhelpful and annoying.” (F, age 34, undertook amniocentesis)

“[referring to the section about the karyotype test results] No it doesn’t really help, it doesn’t really say anything.” (F, age 34, undertook amniocentesis)

A minority of participants felt that the section describing the karyotype test results (n=2) could be clearer. One participant reported confusion regarding the different risks of miscarriage communicated in the antenatal clinic (i.e., national rate and rate of the hospital where the procedure is performed) and expected amnioDex to provide explicit information about which risk to trust. The need for further information about cystic fibrosis, about possible developments of non-invasive prenatal diagnostic tests and about the tests available

after declining an amniocentesis were reported by isolated participants. Finally, one participant reported the need for specific information about the difficulties encountered when deciding about amniocentesis (e.g., balancing the risk of miscarriage, against the risk of foetal abnormality, making a decision with uncertain outcomes).

“It should be specific. You should give some examples, hum, why people have difficulties making a decision, because they have to think, ok, you may lose the baby, you have to make a choice, like, if you have an abnormal baby, what’s your choice? My risk was 1 in 232. This is not high risk you know, because the normal cut off is 1 in 250, so my risk is not very high, and the national risk of miscarriage is 1%, so this is double, the risk is double, so I have to think, ok, should I take this risk, if I take this risk, losing my baby is a higher risk than my baby having down’s syndrome and I have to balance this.” (F, age 27, undertook amniocentesis)

A minority of participants reported technical difficulties using amnioDex and watching the video clips or other functionalities involving Flash Player. Three participants reported difficulties navigating the website (e.g., difficulty closing and opening video clip windows). Finally, a minority of participants experienced difficulties using the think-aloud technique while navigating the website and recurrently prompted the researcher for guidance of what to do next, for the researcher’s help or opinion.

“Hum, I’m finished with this now. So where shall I go?” (F, age 41, declined amniocentesis)

### **Suggestions for Improvement**

On the homepage, participants suggested to specify the meaning of the acronym amnioDex (amniocentesis decision explorer), to add relevant branding (i.e., logo of Cardiff University), and a short paragraph describing the decision at stake, the aims of the website and the principal functionalities.

“I mean, you wouldn’t have to put aims and objectives but to say, this website has been put together to try and help you make an informed decision, there’s no right or wrong answer but hopefully the information will help you map out your thoughts, whatever you want to phrase it, but it might be quite useful to sort of have that on the homepage.” (F, age 34, undertook amniocentesis)

A suggestion was made to add a search box and to provide further information about the developers of amnioDex (e.g., occupations, relevant expertise).

With regards to graphic-based elements, participants suggested to shorten the video of the amniocentesis procedure and to add an explanation indicating that all videos clips were quotes from women's stories enacted by actresses. A suggestion was made to modify the order of the video clips so all "negative outcomes" (i.e., detection of abnormality, miscarriage) were not presented consecutively, as some participants feared this might be daunting to watch.

Finally, participants suggested ways of facilitating access to and implementation of amnioDex. Several participants felt that the web address should be given prior to the counselling session.

"The point I think you need to have the information is perhaps, either when you are given the results, over the phone, or at the point of having the test initially actually, when having the blood test. Probably having the blood test is too soon, because obviously, at that point, hopefully you're not having a high risk result, so maybe at the time when you get that initial call, it's at that point I would be wanting this kind of website." (F, age 37, declined amniocentesis)

### **Benefits of AmnioDex Compared to Routine Counselling**

Compared to routine counselling, six out of fifteen participants felt that amnioDex facilitated decision making or reinforced their decision. Several participants considered that amnioDex offered more comprehensive information than usual practice.

"If you haven't made a decision, I'm sure you can go and spend absolutely ages looking though everything to give you the best possible help in your decision." (F, age 34, declined amniocentesis)

Several pregnant women praised the fact that amnioDex provided information that they could assimilate at their own pace and consult at different stages of the decision making process.

"Because when you're told about it, when you're told about the amniocentesis, then more than just having a leaflet that you can go through, you'd like to research it on the net, you'd like to see what happens to other women and their opinions, you'd like to get as much information as you possibly can." (F, age 35, undertook amniocentesis)

"It's easier to look at something rather than have a conversation with someone over the phone or even sit in a surgery because sometimes you just forget to ask things or you don't understand things properly, here you can go back and keep looking at it and it's quite simple to understand." (F, age 37, declined amniocentesis)

“Because you’re so stressed, not that much of it went in I don’t think. If I’d had this, it would probably have been more helpful.” (F, age 29, undertook amniocentesis)

Eight out of fifteen participants felt that amnioDex would be beneficial in addition to routine counselling, as it provided balanced examples of women’s experiences (i.e., video clips).

“You know, it’s being able to make an informed decision, having a resource that you can use, that shows you what’s gonna happen, when it’s gonna happen and what other people have been through.” (F, age 35, undertook amniocentesis)

The majority of pregnant women (n=14/15) would have used amnioDex if the resource had been available to them.

### **9.3.6 Outline of Changes Made**

AmnioDex version 3, field-tested with pregnant women, was amended to develop the final version of the intervention (see Figure 9.4). The top tabulation was redesigned and a dual navigation system was added to improve the navigation structure (i.e., sub-menus appeared on the left hand side of the page and on the top tabulation) (see Figure 9.5). A search box, relevant logos and the words; amniocentesis decision explorer, were added on the homepage. The video of the amniocentesis procedure was shortened. A disclaimer was added in the section Personal Stories: “The videos are enacted quotes from women’s stories”, and the order of the clips was modified following participants’ feedbacks. Technical difficulties associated with the video clips were addressed. Information contents perceived unclear or too brief were discussed with the steering group and some sections were amended.

## **9.4 Discussion**

While all stakeholder groups positively received amnioDex, substantial amendments were needed to increase usability and acceptability. Lay users made most criticisms. Suggestions for improvements and criticisms generally applied to the navigation structure and graphic-based elements. Health professionals and pregnant women positively reacted to

Figure 9.4 Screenshot of the AmnioDex Homepage Version 4



Figure 9.5 Screenshot of the AmnioDex Navigation Structure Version 4





the design, graphic-based elements (e.g., video clips, diagrams, images) and informational content (clarity and accuracy).

Both pregnant women and health professionals perceived amnioDex as a useful resource in adjunct to existing counselling. Video clips of women stories were considered particularly helpful and informative by professionals and pregnant women. Interactive elements such as the message board were positively reviewed by most stakeholder groups and deemed helpful in making a decision by pregnant women. There is documented evidence that speaking to others in a similar situation (e.g., via a message board) or hearing experiences of women facing similar dilemma can be comforting to women facing prenatal testing (Moyer et al. 1999; St-Jacques et al. 2008). Watching testimonials or reading messages of others in a similar situation may help pregnant women imagine the consequences and implications of all possible outcomes of amniocentesis testing. The tendency to think through all possible scenarios has been previously described as “imagining coping”, and may be facilitated by the presence of deliberation tools or balanced narratives (Potter et al. 2008).

A new navigation structure, search box, improved layout, and technical improvements were the main changes made to amnioDex. Most criticisms were made with regards to graphic-based elements and navigation structure, across all stakeholder groups. The latter finding is consistent with previous account of field-testing (Evans et al. 2007), where the navigation was considered most problematic. This highlights the need to thoroughly investigate usability issues associated with web-based interventions and to draw on existing methods used in usability studies such as the think-aloud technique (Dumas and Redish 1999). Field-testing web-based interventions with lay users prior to potential users (i.e., pregnant women and professionals) appeared essential in identifying basic technical and

usability issues. Pregnant women and health professionals were therefore able to focus on the informational content and interactive elements of the website without being distracted by basic usability issues (previously picked-up by lay users). Health professionals expressed divergent opinions regarding the video of the amniocentesis procedure, the videos of health professionals and raised concerns regarding the lack of control over the content of the message board. As highlighted in previous chapters, health professionals generally held strong opinions and fairly controlling or paternalistic attitudes about the nature and quantity of information needed prior to deciding about amniocentesis testing. A message board or video of the procedure may have threatened, to some extent, the physicians' sense of control over information. Health professionals' influence or authority surrounding prenatal testing has been extensively documented (Marteau 1993; Marteau et al. 1992). A study of women's reasons for undertaking prenatal testing showed that the decision to undertake maternal serum screening tests seemed imposed upon women and influenced by the physician's authority (Chiang et al. 2006). Further research revealed that healthcare providers tended to shape the meaning and purpose of prenatal testing (Press and Browner 1997). DESIs such as amnioDex may be effective in balancing the physicians' influence and authority.

Compared to routine counselling, amnioDex offered practical accounts of women facing a similar dilemma and comprehensive information about the risk of miscarriage and the chromosomal abnormalities tested for. Pregnant women believed that amnioDex was the most comprehensive web-based resource available on amniocentesis testing and a useful supplement to usual practice. Nevertheless, pregnant women raised issues associated with the implementation of amnioDex. Several women believed that amnioDex should be accessible at an early stage of the decision making process (i.e., when undergoing screening tests or receiving the results) to ensure that women can access the resource before making a decision.

Accessibility and lack of awareness of existing interventions have been described as significant barriers to the implementation of DESIs and recommendations have been made to increase accessibility (Graham et al. 2003; O'Donnell et al. 2006).

The present study offers some insight into practical ways of field-testing web-based DESIs. The think-aloud technique, widely used in cognitive psychology research and usability tests of computer-based interfaces, provided understanding into the navigation and understanding difficulties encountered while using amnioDex (Gray and Salzman 1998). This method highlighted usability issues which would not have been uncovered by observation or semi-structured interview alone. The majority of participants positively reacted to this approach. However, several pregnant women experienced difficulties continuously articulating their thoughts and prompted the researcher for guidance and instructions. The think-aloud technique has been described as highly adaptable (Davison et al. 1997). This method may therefore be adjusted to fit the requirements of DESI field-testing in order to overcome the methodological issues raised in the present study. Based on the amnioDex findings and previous research on the evaluation of e-learning, the level of guidance given to participants when using the think-aloud technique needs to be increased (Cotton and Gresty 2006). While Ericsson and Simon's methodology for the think-aloud approach provided minimum instructions, a more flexible use of instructions may be necessary to resolve methodological issues described above (Ericsson and Simon 1984). For instance, prompts may be used to collect specific data: "What do you think of this section?", "Why have you decided to go in this section?" Similar prompts were piloted by Cotton and Gresty (2006) and were considered useful in generating data. Finally, a short interview used in conjunction with the think-aloud technique may be essential in ensuring that all key issues are covered, as has

been documented previously (Cotton and Gresty 2006; Johnson et al. 2005; Peleg et al. 2009).

Strengths of this study were the methodological approach used to field-test amnioDex and heterogeneity of the sample. The think-aloud method seemed to provide more insight into the intervention's usability than classic observational techniques. In spite of minor methodological limitations, this method appeared versatile enough to be adapted to DESI field-testing.

This study may be criticised for the limited number of pregnant women recruited to field-test the intervention. Considerable recruitment difficulties were encountered throughout the field test. Many women approached by midwives in the antenatal clinic declined to take part in the study. As highlighted in Chapter 8, a further limitation may be the iterative approach of the field test. The stakeholder groups evaluated incremental versions of the deliberation tools, which subsequently compromised direct comparisons between the groups.

## **9.5 Conclusion**

Pregnant women and health professionals positively appraised the amnioDex intervention. The website was deemed useful, clear and comprehensive. Usability was considerably increased by criticisms and suggestions made by lay users. Field-testing is a fundamental assessment of the DESI's acceptability and usability prior to evaluation and dissemination, and becomes especially relevant when developing web-based interventions. Finally, this study provided some insight into possible methods for field-testing web-based interventions. Field-testing with lay users, professionals as well as potential users may be required as each stakeholder group identified different dysfunctions or potential improvements. Previous research into usability testing of interactive healthcare interfaces

suggests that the think-aloud method may be adapted and combined with qualitative methods to yield optimum results.

# **Chapter 10**

## **General Discussion**

The studies described in this thesis aimed to assess information and decision support needs of women facing amniocentesis testing in order to develop and field-test, in collaboration with them and their healthcare professionals, an interactive theory-based DESI. This chapter presents an overview of the principal findings, discusses strengths and weaknesses of the methodological approach, and considers clinical implications and future research.

### **10.1 Translating Theory Into Practical Interventions**

There is an apparent lack of conceptual and theoretical underpinnings to the development of DESIs. The majority of DESIs included for review did not build on models or theories of decision making (Chapter 2). The findings highlighted the difficulty to transfer abstract theoretical constructs into the practical development and evaluation of DESIs. There may be several interpretations to the lack of theoretical underpinnings underlying DESI development; difficulty translating abstract theoretical constructs into practical interventions, lack of empirical evidence of the efficiency of theory-based interventions or time constraints and cost-effectiveness imperatives. It is worth noting that the IPDAS instrument does not assess whether the DESI development was informed by theory (Elwyn et al. 2009c). The theoretical and research work presented in this thesis advances the field by describing a developmental pathway that can be followed to design a DESI that is acceptable to patients and health care professionals.

Chapter 7 and 8 described the transfer of descriptive decision making theories and heuristic-based algorithms into the design of amnioDex and interactive deliberation tools. The findings indicated that carefully selected theoretical frameworks can successfully guide the development of interactive interventions but also underlined conceptual and technical challenges generated by this innovative and abstract translation. Prospect theory influenced the framing of information (e.g., risk of miscarriage, chance of chromosomal abnormality) and motivated the development of the screening test result calculator. To comply with the principles of differentiation and consolidation theory (DiffCon theory), information was presented in a way that facilitated differentiation (i.e., head to head comparison, balanced examples of women's stories presented in adjacent columns). The development of those theory-based elements was guided by existing attempts at integrating Prospect and DiffCon theories into decision aid design (Feldman-Stewart et al. 2001; O'Connor et al. 1999c). The transfer of theory into practice was also facilitated by the fact that only selected elements of the intervention were guided by the aforementioned theories. All elements developed according to prospect and DiffCon theories were positively reviewed by the majority of users involved in the field test (Chapter 9). However, the subjective and equivocal nature of the transfer of theory into DESI development means the value of translational work needs to be monitored. First, there is a need for in-depth evaluation of the effect of theory-based components on selected decision outcomes. Second, the impact of theory-based components or interventions on decision outcomes needs to be evaluated against atheoretical interventions or routine care as empirical evidence in this area is currently missing. Third, there is scope for developing theory-derived outcome measurements to examine whether theory-based components achieve their stated aims (Elwyn et al. 2009d). The lack of well validated and widely accepted measures of DESI effectiveness is a considerable obstacle to DESI evaluation and implementation. Although multiple measures of DESI effectiveness exist

(Simon et al. 2007), there is no consensus on the most relevant constructs or criteria on which their effectiveness should be assessed (Kennedy 2003; Ratliff et al. 1999; Dy 2007). Theories or models of decision making could provide useful indications of relevant constructs to measure when assessing the quality of decisions and DESIs' effectiveness.

While the transfer of prospect and DiffCon theories into DESI elements was straightforward and fairly systematic, translation difficulties arose when attempting to develop heuristic-based deliberation tools. Fast and frugal heuristics are simple rules of reasoning that have demonstrated the same or superior predictive accuracy compared to complex mathematical models in similar decision tasks (Green and Mehr 1997; Todd and Gigerenzer 2000). Heuristic-based algorithms are fast and non-compensatory rules of reasoning broken down into simple cognitive steps. One would therefore expect the transfer of heuristics into deliberation components to be intuitive and unequivocal. This research showed that transferring heuristics into graphic-based elements was a difficult and iterative process where the usability of deliberation components was dependent on the accuracy of the translation (Chapter 8). Although heuristic cognitive steps provided some degree of guidance for designing the deliberation tools, there was ambiguity regarding how best to graphically represent each step. There is evidence that the Take the Best algorithm, when tested on real-world questions under conditions of limited knowledge and time, performs as well as the Tallying algorithm (Gigerenzer and Goldstein 1996). One would therefore expect the deliberation tools derived from the Take the Best and Tallying heuristic-based algorithms to be equally usable and efficient. Paradoxically, the simplest algorithm (i.e., Take the Best), when transferred into a graphic interface, was least usable. The difficulty therefore seemed to arise from the translation and operationalisation of these algorithms, and of the Take the Best algorithm in particular. There is scope for investigating alternative ways to operationalise the



Take the Best heuristic-based algorithm, and create an interface which is more intuitive and usable. One way of addressing this issue would be to use brainstorming techniques with web design experts as well as lay people (possibly pregnant women considering amniocentesis testing) to envisage novel ways of translating the algorithm's abstract mental steps into graphic-based elements.

In spite of translation difficulties, field-testing with pregnant women facing amniocentesis showed that when translation was accurate (e.g., weighing it up), theory-driven deliberation components were favourably appraised by stakeholders (Chapter 8). The structure provided by the tools (e.g., a clear list of reasons, an efficient visual analogy; the weighing scale) was deemed helpful in achieving a decision. Although deliberation components may be helpful in eliciting values and making a decision, the deliberative processes of decision making remain poorly understood. Further research is needed to clarify deliberation processes and identify ways of facilitating it.

Finally, the findings highlighted a significant theory-practice gap. Most theories or models of decision making have focused on explaining or describing how humans cognitively approach and achieve a decision rather than on how tools could be designed to help them make decisions. Crossing the bridge from decision theories to the design of usable and effective interventions presents important translation difficulties and challenges that have not yet been resolved. Based on the experience of developing amnioDex, it appears that no single theory or model alone can guide the design of all DESI components (i.e., information component, deliberation component and outcome measurements). Some theories provide a relevant framework for designing information components (e.g., prospect theory) while others can be used to inform the development of deliberation tools (e.g., heuristic-based

algorithms) or outcome measurements. If the translation of theory into practice is to become widespread and systematic, there is a need for developing a taxonomy of theoretical constructs transferable into practice and elaborating possible methods for achieving this ambitious translation (Elwyn et al. 2009d). Such taxonomy exists for behavioural interventions (Abraham and Michie 2008). However, there is currently no taxonomy of decision making theories used in DESI development and the translation of decision making theories or models into usable interventions has rarely been documented (Feldman-Stewart et al. 2001). Finally, usability issues highlighted in the field test of heuristic-based deliberation components entail sustained collaboration between patients and principal stakeholders to develop interventions that are acceptable and usable while providing structured guidance to clarify values and achieve decision making.

## **10.2 Information and Decision Support Needs of Women Facing Amniocentesis**

It is widely recognised that deciding whether or not to undergo amniocentesis should be the result of an informed choice (General Medical Council 1999; Marteau 1995), namely, “one that is based on relevant knowledge, consistent with the decision maker’s values and behaviourally implemented” (Dormandy et al. 2002, p.1). Achieving informed choice promotes patients’ autonomy and patient centred care, and reduces medico legal costs. It has therefore become a cornerstone of the British National Health System. There are documented concerns that existing information and decision support available in the UK does not enable pregnant women to make informed decisions about amniocentesis testing (Chapter 3), and that the information provided does not address their own interests (Hunt et al. 2005; Rostant et al. 2003; Cederholm et al. 1999; Jaques et al. 2004; Marteau 1995). In situations of equipoise such as amniocentesis testing, DESIs have been shown to increase knowledge, improve the perception of risks and involvement in decision making (O’Connor et al. 2006). Six DESIs for amniocentesis of variable quality and limited effectiveness were identified

(Chapter 4), none of which are currently available in the UK. Most interventions were developed without systematic input from principal stakeholders (pregnant women and health professionals) and rarely field-tested or piloted with pregnant women and health professionals. The apparent tendency to empirically develop interventions without a systematic development process casts doubt on the clinical accuracy and reliability of those interventions. Those findings emphasised the need to involve patients and health professionals at all stages of the development process. The necessity to involve relevant stakeholders prior to developing (needs assessment) and evaluating interventions (field-testing) has been recognised by the IPDAS collaboration (Elwyn et al. 2006).

The needs assessment conducted with pregnant women and health professionals (Chapters 5 and 6) exposed unmet needs for information, emotion and decision support and highlighted the gap between the professionals' assessment and women's reported needs. Women repeatedly expressed the need for information about the range of abnormalities tested and potential consequences of amniocentesis testing (e.g., elective termination of pregnancy), while professionals focussed on describing the amniocentesis procedure, and explaining the risks of foetal abnormality and pregnancy loss. Healthcare providers expressed divergent opinions regarding the nature and quantity of information needed about chromosomal abnormalities potentially detected and elective termination of pregnancy. Several professionals believed that the sensitive issue of pregnancy termination and detailed information about abnormalities should only be raised and discussed when a problem was found, post-amniocentesis. Recorded observations of 25 routine antenatal consultations for amniocentesis testing suggested that obstetricians focussed on describing the risks of abnormality and miscarriage and provided little or no information about the conditions tested for and about the possibility of terminating the pregnancy (Marteau et al. 1993). Down's

syndrome was mentioned in most consultations but was never described. Other conditions were mentioned in only a third of the consultations. It has been argued that providing sufficient information about abnormalities potentially detected and implications of an amniocentesis could reduce confusion and distress experienced in the diagnostic phase of pregnancy (Marteau et al. 1993). The lack of time and lack of specialised knowledge reported by health professionals most probably influenced the presentation of information and tendency not to discuss those sensitive topics. Health professionals' influence on the presentation of information can be balanced by a DESI for amniocentesis, such as amnioDex, which presents comprehensive information in a neutral manner, and provides balanced examples of women's decisions about amniocentesis.

Further, health professionals tended to focus expertise and energy on the provision of information, at the expense of emotional and decisional support. By contrast, pregnant women reported high emotional strain and anxiety and emphasised the need for reinforced emotional support. Until relatively recently, emotion has been viewed as an impediment to effective decision making, at best a distraction and at worst a source of bias. This view is being challenged (Damasio 1994; Evans 2002; Marnett 2004). The role of emotions in decision making is increasingly advocated (Anderson 2003; Wilson and Gilbert 2005). The Attend, React, Explain, Adapt (AREA) model of affective forecasting developed by Wilson et al. assumes that decision making is strongly influenced by how individuals anticipate their emotional reactions to future events, namely affective forecasts. Research indicates that affective forecasts are generally flawed, and that individuals often overestimate the impact of negative events and the intensity of related emotional reactions (Gilbert et al. 2000; Gilbert et al. 2004; Wilson et al. 2005). Further research is needed to examine the extent to which affective forecast errors can be addressed in DESIs.

The present study suggested that healthcare providers exerted an explicit and almost paternalistic control over specific information topics. Information related to the range of chromosomal abnormalities detected, the elective termination of pregnancy and the miscarriage rate appeared controversial and not systematically addressed by all professionals. The present findings are consistent with those of Bernhardt et al. (1998) and Marteau et al. (1993). It is worth noting that the professionals' control over information was also reflected in the steering group meetings held with health professionals prior to developing amnioDex (Chapter 7). Professionals involved in the steering group and stakeholder analysis (Chapter 6) expressed views which frequently differed with women's reported information and decision support needs. The question arose as to which view should be integrated into the intervention. Although editorial control remained with the researchers who developed amnioDex, deciding whether to present information requested by pregnant women (e.g., information about the elective termination of pregnancy, local miscarriage rate) or to follow health professionals' opinions was difficult. On the basis that amnioDex was specifically designed for pregnant women deciding about amniocentesis, a decision was made to prioritise women's information needs over the professionals' views. However, prioritising women's views or legitimate information at odds with local practice may have significant repercussions on the implementation and dissemination of amnioDex in the NHS (see section 10.5, p. 183 for further discussion).

Finally, several pregnant women reported feeling pressured into undertaking prenatal testing (i.e., screening tests and amniocentesis), some of whom experienced regret about their screening decision. Pressures to undertake prenatal testing are inconsistent with current policies and standards advocating informed choice prior to undertaking prenatal screening or diagnostic tests (General Medical Council 1999). Health professionals' directiveness with

regards to prenatal testing has been extensively documented (Al-Jader et al. 2000; Press and Browner 1997; Santalahti et al. 1998; Marteau et al. 1993; Sjogren and Uddenberg 1987). Clarke (1997) argued that despite significant ethical and societal objections, prenatal genetic screening tests have become routine and are often undertaken by pregnant women who do not realise the objectives and implications of these tests (Williams 1995; Press and Browner 1997). It has been assumed that offering routine prenatal screening to a low risk population would be helpful to prospective parents. Routine prenatal screening may be in line with societal norms and ideals but is not necessarily beneficial at the individual level. It has also been argued that offering routine prenatal genetic screening may have reinforced negative attitudes towards disability in the general population (Shakespeare 1998). Most pregnant women undertaking screening tests for Down's syndrome are not aware of the objectives of the screening programme (including cost effectiveness objectives) and do not normally reflect on the overall usefulness of the test. Almost half of pregnant women interviewed during the needs assessment of amnioDex would have liked more information about the screening tests available prior to amniocentesis testing (Chapter 5). Reflecting on the objectives of genetic screening and considering its usefulness at the individual level may limit regret when receiving a higher chance result and promote informed choice. AmnioDex was specifically developed for women who had been offered amniocentesis testing, and although screening tests are mentioned and described in the section Why Amniocentesis? it does not specifically address the decision to undertake prenatal genetic screening. Although screening and testing for Down's syndrome are related issues, deciding whether to undertake screening tests for Down's syndrome or whether to undertake amniocentesis are two separate decisions which cannot be addressed in the same intervention. Addressing the decision to undertake screening tests in amnioDex did not seem appropriate or achievable and was beyond the scope of this thesis. There is therefore room for developing a DESI for prenatal

screening that describes the objectives and implications of screening and helps women consider the relevance of the test with regards to their values, attitudes and preferences. Increasing knowledge about prenatal screening is not sufficient; prospective parents should be aware of the objectives, limitations and underlying motives for offering prenatal genetic screening to pregnant women who have a low risk of foetal chromosomal abnormality (Clarke 1997).

### **10.3 Field-Testing Complex Interventions Prior to Evaluation**

Findings of this research suggested (Chapter 2 and 4) that only a minority of existing DESIs underwent field-testing or piloting prior to evaluation and dissemination (Drake et al. 1999; Evans et al. 2007; Nagle et al. 2008). There is a tendency to develop DESIs without ensuring that those interventions are usable and acceptable to users. The qualitative field-testing of amnioDex revealed that the intervention was deemed acceptable by principal stakeholders (i.e., pregnant women considering amniocentesis and health professionals), although the navigation, layout and graphic-based elements needed to be modified to address their comments (Chapter 9). This study is the first to carry out field-testing of a web-based DESI with three relevant user groups: lay users, health professionals and pregnant women facing a decision to undergo amniocentesis. Field-testing the intervention with lay users proved beneficial in identifying essential usability issues prior to field-testing amnioDex with principal stakeholder groups. Pregnant women and health professionals specifically praised the video clips of women's stories offering balanced examples of women's (and their partners') decisions about amniocentesis testing. All interactive elements embedded in the website, such as the message board, the screening test calculator or the deliberation tools were deemed helpful by pregnant women. Schwitzer et al. conducted a review of health decision support tools (Schwitzer 2002) and identified four key functionalities distinguishing web-based DESIs from other media: videos of patient stories, online community network

(e.g., message board), user-specific outcomes data (e.g., screening test result calculator) and public access. It is worth noting that amnioDex has all these key functionalities (Chapter 7). By contrast, Schwitzer established that none of the web-based interventions included for review delivered all four key functionalities and suggested that patients would benefit from more comprehensive web-based interventions.

Little is known about specific methods for field-testing DESIs. This research provided some insight into practical guidelines for field-testing web-based DESIs (Chapter 9). The findings suggested that web-based interventions should be tested with principal stakeholders (i.e., professionals and patients) as well as lay users as they critically appraised the website from three different angles: usability, clinical relevance and users' acceptability. The think-aloud technique may be used to investigate the process of cognition and emotions while users navigate the intervention. The field test of amnioDex and previous accounts of usability tests (Cotton and Gresty 2006) indicated that this method should be adjusted to better fit the requirements of DESI field-testing. More guidance should be given to participants prior to using the intervention and clear prompts may be used to collect specific data or attract users' attention to specific sections of the website. Further, evidence suggests that combining qualitative methods with the think-aloud technique would yield optimal results (Cotton and Gresty 2006; Peleg et al. 2009). For instance, a semi-structured interview addressing general usability and acceptability issues may be used after participants navigate the website using the think-aloud technique. Finally, the necessity to field-test complex interventions has been recognised by the IPDAS collaboration, but is also consistent with the Medical Research Council (MRC) framework for the design and evaluation of complex interventions to improve health (Campbell et al. 2000). The new MRC guidance recommends greater attention to the development process and piloting phase (i.e., field-testing) (Craig et al. 2008).



## 10.4 Strengths and Weaknesses

The research methods were specifically selected to examine and address information, emotions and decision support needs associated with amniocentesis testing, following the MRC complex intervention framework and IPDAS quality criteria. Two literature reviews were conducted: a theoretical review and systematic literature review. The theoretical review examined the conceptual and theoretical frameworks underlying the conception, prototype development and evaluation of DESIs included in a Cochrane systematic review. The second literature review investigated the principal characteristics, quality, effectiveness and implementation of existing DESIs for amniocentesis testing. Interventions included for review were rated against the 10 domains of the IPDAS instrument. Qualitative research methods were chosen to capture women's experiences of deciding about amniocentesis and professionals' assessment of pregnant women's needs. Semi-structured interviews were conducted with pregnant women facing a decision to undertake amniocentesis and health professionals from relevant disciplines. The amnioDex intervention was subsequently developed using an iterative approach with multi-disciplinary input. Building on existing methods of usability testing of interactive healthcare interfaces, amnioDex and embedded deliberation components were field-tested using the think-aloud technique and semi-structured interviews. This study was the first to translate heuristic-based algorithms (Take the Best and Tallying) into interactive deliberation tools. Finally, strengths of this study were the novel approach adopted to integrate theory into the practical development of an interactive intervention and embedded deliberation components.

This research may be criticised for the low number of pregnant women recruited in the needs assessment and field-testing of amnioDex. Significant recruitment difficulties were encountered throughout the study. Women facing amniocentesis testing are a very small

proportion of pregnant women in the UK (5 to 10%) and generally experience extreme stress and anxiety when deciding about amniocentesis and waiting for the results (Susanne et al. 2006; Tercyak et al. 2001). Pregnant women approached in the participating antenatal clinics frequently declined to take part or withdrew from the study pre-interview due to stress and anxiety triggered by the amniocentesis decision, a finding which further strengthens the need to develop DESIs. Another limitation may be the low participation of male partners in the needs assessment and field-testing of amnioDex. Although this finding is consistent with male participation rates in other reproductive health contexts (Bunting and Boivin 2007), it nevertheless points to a need for more effort to be directed at recruiting men in future research of this type. The decision to undertake amniocentesis is shared and determined by the values and preferences of the pregnant woman as well as those of her partner, and the partner's influence in deciding about prenatal testing has been previously documented (Carroll et al. 2000; Jaques et al. 2004). A systematic review of decisional needs related to prenatal testing (St-Jacques et al. 2008) revealed that partners' decisional needs have rarely been assessed. Although only four partners participated in the needs assessment, their perceptions and preferences about amniocentesis testing were indirectly investigated by asking (in the partner's absence) pregnant women about their partner's views. A final limitation is that it would have been desirable to conduct a large scale online evaluation of the amnioDex intervention but this was beyond the scope of this project. Given considerable recruitment difficulties encountered during the field-testing of amnioDex (data collected until May 2009), it was not practically possible to set up an online trial. However, a proposal to evaluate amnioDex in a randomised controlled trial was developed. A grant funding application was submitted to HTA Clinical Evaluation and Trials (see Appendix 5 for outline protocol).

## **10.5 Clinical Implications and Future Research Directions**

The findings of this thesis indicated that a collaborative approach between researchers, potential users and health professionals can successfully lead to an intervention that is both usable and acceptable. If complex interventions such as amnioDex are to be accepted by patients and health professionals, the needs and concerns of those involved as well as practical considerations of the medical setting have to be taken into account. A systematic development process involving regular input from stakeholders and systematic piloting may facilitate usability and acceptability.

It transpires from the literature and experience of developing amnioDex, that the greatest challenge of all is to move from successful DESI development to implementing those interventions in clinical settings. Difficulties disseminating and implementing innovative healthcare interventions, of which DESIs are part, have been widely documented (Greenhalgh et al. 2004; Holmes-Rovner et al. 2000; Silvia et al. 2008). The most commonly reported barriers to implementation are the lack of awareness and support from health professionals and difficulties organising the DESI distribution in primary or secondary care settings (e.g., time constraints, lack of resources) (Silvia et al. 2008). Based on the experience of developing amnioDex, the disagreement between pregnant women (or patients in general) and health professionals as to what information should be provided in the DESI may have repercussions on whether and how a DESI is implemented and whether it actually meets the needs of those using it. For example, if the intervention provides information that health professionals judge unnecessary, disturbing or confusing, they may be less willing to implement the DESI, even if women want this information. A collaborative approach that reinforces the fit between professionals and patients by equal representation in the steering group, or awareness raising activities between health professionals and patients, seem

promising ways of ensuring that a DESI is acceptable to all. Second, as noted previously (Silvia et al. 2008), implementation may be limited by the lack of structure and resources available to identify potential users and distribute DESIs when patients need it most. Indeed, pregnant women indicated that the timing of amnioDex delivery was crucial. They believed that the intervention should be available early on, before or immediately after receiving the screening tests results, or else they would not use it.

Further, a study reporting the assessment of an educational intervention for prenatal screening indicated that implementing computer-based interventions in clinical settings potentially disrupted the flow of patients (Griffith et al. 2005). There may be less disruptive ways to implement DESIs, such as allowing out-of-office online access to the intervention, which would not require resources or time commitment from health professionals. However, this would have drawbacks in terms of access for those without computers. Increasing awareness about DESIs could be achieved by advertising the intervention in secondary care settings (e.g., cards advertising amnioDex distributed in antenatal clinics) and through existing online platforms (e.g., NHS Choices, pregnancy websites). Clearly preliminary discussions about DESIs need to take into account such practical considerations if they are to have any realistic chance of implementation.

The development and implementation of DESIs is in line with current trends in the NHS to promote patient autonomy, informed choice and access to services (Department of Health 2000). NHS Choices is a prime example of the NHS willingness to develop patient autonomy and involvement in healthcare. The NHS Choices website provides information about over 750 conditions and treatments, current health related topics and services available, with the aim of increasing patients, as well as the general population's, control of their

healthcare (NHS Choices 2009). Further, the NHS white paper *our health, our care, our say: a new direction for community services* emphasises the patients' rights to be informed and involved in their health related decisions: "You will be in charge of your own health. You will get better information so you can make choices about staying healthy and well" (Department of Health 2006, p. 7). However, those tools primarily provide information but do not currently offer the much needed decisional or emotional support. There is therefore an opportunity to reinforce NHS aims if DESIs can become routinely available to those who wish to be actively involved in their care. The difficulty here is to negotiate access to the NHS, which can best be obtained by collaboration with health professionals and NHS stakeholders at the early stage of the DESI design and development.

## **10.6 Conclusions**

The research presented in this thesis provided a developmental pathway for the design of a DESI for amniocentesis testing, demonstrating that it was possible to develop a theory-driven intervention that was ultimately acceptable to pregnant women and professionals alike. Given implementation and dissemination difficulties inherent to this field, it is imperative to involve health professionals and potential users in an attempt to maximise the clinical accuracy, relevance, usability and acceptability of those interventions to patients. While stakeholder involvement and iterative testing are no guarantee of successful DESI implementation, it will facilitate the dissemination process by accounting for practical requirements and specific needs of users and professionals while increasing professionals' awareness.

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## **Appendix 1**

**International Patient Decision Aids Standards Instrument.....Page 211**

## International Patient Decision Aids Standards Instrument

Domain Information	Strongly agree 4	Agree 3	Disagree 2	Strongly disagree 1	Score	Comments
1. The decision aid describes the health condition or problem (intervention, procedure or investigation) for which the index decision is required.	<i>There is a detailed description of the health condition or problem</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no description of the health condition or problem at all</i>		
2. The decision aid describes the decision that needs to be considered (the index decision).	<i>The decision, that the decision aid addresses, is specifically stated</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no description of the index decision</i>		
3. The decision aid describes the options available for the index decision.	<i>A comprehensive list of options related to the decision is provided</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no description of the available options</i>		
4. The decision aid describes the natural course of the health condition or problem, if no action is taken.	<i>There is a description of how the untreated condition is expected to develop if no action is taken</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no description of the natural course of the health condition or problem</i>		
5. The decision aid describes the positive features (benefits or advantages) of each option.	<i>A comprehensive list of benefits and/or advantages of each option is provided</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no description the potential benefits or advantages of the options</i>		
6. The decision aid describes negative features (harms, side effects or disadvantages) of each option.	<i>A comprehensive list of harms and/or side effects and/or disadvantages of each option is provided</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no description the potential harms or side effects or disadvantages of the options</i>		
7. The decision aid makes it possible to compare the positive and negative features of the available options.	<i>Potential harms and potential benefits are presented in a head-to-head comparison</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The presentation of potential harms and potential benefits does not allow for a head-to-head comparison</i>		
8. The decision aid shows the negative and positive features of options with equal detail (for example using similar fonts, order, and display of statistical information).	<i>The negative and positive features are presented with equal detail by using the same font, order, and display of statistical information</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The negative and positive features of the options are not shown with equal detail – leading to a perceived favouring or disfavouring of a specific option</i>		

Domain Test	Strongly agree 4	Agree 3	Disagree 2	Strongly disagree 1	Score	Comments
1. The decision aid describes what the test is designed to measure.	<i>The test is described in detail to provide the user with a complete picture of what it is designed to measure</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The test is only included as a label</i>		
2. The decision aid includes information about the chances of having a true positive test result.	<i>The natural frequency (event rate) of having a true positive test result is included</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no information about the probability of having a true positive test result</i>		
3. The decision aid includes information about the chances of having a true negative test result.	<i>The natural frequency (event rate) of having a true negative test result is included</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no information about the probability of having a true negative test result</i>		
4. The decision aid includes information about the chances of having a false positive test result.	<i>The natural frequency (event rate) of having a false positive test result is included</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no information about the probability of having a false positive test result</i>		
5. The decision aid includes information about the chances of having a false negative test result.	<i>The natural frequency (event rate) of having a false negative test result is included</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no information about the probability of having a false negative test result</i>		
6. If the test detects the condition or problem, the decision aid describes the next steps typically taken.	<i>There is information about possible follow-up actions when the condition or problem is detected</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no information about possible next steps when the condition or problem is detected</i>		
7. The decision aid describes the next steps if the condition or problem is not detected.	<i>There is information about possible follow-up actions when the condition or problem is not detected</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no information about possible next steps when the condition or problem is not detected</i>		
8. The decision aid describes the chances that the disease is detected with and without the use of the test.	<i>The probability of detecting the target condition both with and without screening is presented</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no information about the probability of detecting the target condition</i>		
9. The decision aid has information about the consequences of detecting the condition or disease that would never have caused problems if screening had not been done (lead time bias).	<i>There is explicit information about the possibility of screening leading to the detection and treatment of the condition or disease that might never have caused symptoms had it not been for the screening</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no information about the possibility screening leading to the detection of a condition or disease that may never have become symptomatic</i>		



<b>Domain Probabilities</b>	<b>Strongly agree 4</b>	<b>Agree 3</b>	<b>Disagree 2</b>	<b>Strongly disagree 1</b>	<b>Score</b>	<b>Comments</b>
<b>1.</b> The decision aid provides information about outcome probabilities associated with the options (i.e. the likely consequences of decisions).	<i>The decision aid clearly presents probabilities for all relevant outcomes of the options</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no reference to the magnitude (absolute or relative) of the likelihood of positive or negative outcomes</i>		
<b>2.</b> The decision aid specifies the defined group (reference class) of patients for which the outcome probabilities apply.	<i>The decision aid provides a clear definition of the population for which the outcome probabilities apply</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no definition at all of the population for which the outcome probabilities apply</i>		
<b>3.</b> The decision aid specifies the event rates for the outcome probabilities (in natural frequencies).	<i>Event rates for each of the positive and negative outcomes are presented</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no reference to event rates for positive or negative outcomes</i>		
<b>4.</b> The decision aid specifies the time period over which the outcome probabilities apply.	<i>The decision aid provides a clear definition of the time period for the given event rates</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no description of the time period for the given event rates</i>		
<b>5.</b> The decision aid allows the user to compare outcome probabilities across options using the same denominator and time period.	<i>The decision aid uses constant denominators and the same time frame for the outcome probabilities of the options</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The denominators for the risk events or the time frames for the outcomes vary across the options</i>		
<b>6.</b> The decision aid provides information about the levels of uncertainty around event or outcome probabilities (e.g. by giving a range or by using phrases such as 'our best estimate is').	<i>The uncertainty around the probability estimates is conveyed through ranges, 95% confidence intervals, or phrasing such as "our best estimate is..."</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>There is no acknowledgement of the uncertainty in the probability estimates or there is a failure to round off numbers – giving the false illusion of precision</i>		
<b>7.</b> The decision aid provides more than one way of viewing the probabilities (e.g. words, numbers, and diagrams).	<i>More than one method is used to present all of the outcome probabilities</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>Multiple methods to view the probabilities are not included in any instance</i>		
<b>8.</b> The decision aid provides information about event or outcome probabilities by using more than one framing method (e.g. positive or negative frames, loss or gain frames).	<i>Outcome probabilities are presented with more than one framing method</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>More than one framing method to view the probabilities is not included in any instance</i>		

<b>Domain Values</b>	<b>Strongly agree 4</b>	<b>Agree 3</b>	<b>Disagree 2</b>	<b>Strongly disagree 1</b>	<b>Score</b>	<b>Comments</b>
<b>1.</b> The decision aid describes the features of options to help patients imagine what it is like to experience the physical effects.	<i>The decision aid provides clear detail (through personal stories or in the main narrative of the decision aid) about the possible impact of harms and benefits on someone's physical life</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>Harms and benefits are brief, factual descriptions or labels only</i>		
<b>2.</b> The decision aid describes the features of options to help patients imagine what it is like to experience the psychological effects.	<i>The decision aid provides clear detail (through personal stories or in the main narrative of the decision aid) about the possible impact of harms and benefits on someone's psychological life</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>Harms and benefits are brief, factual descriptions or labels only</i>		
<b>3.</b> The decision aid describes the features of options to help patients imagine what it is like to experience the social effects.	<i>The decision aid provides clear detail (through personal stories or in the main narrative of the decision aid) about the possible impact of harms and benefits on someone's social life</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>Harms and benefits are brief, factual descriptions or labels only</i>		
<b>4.</b> The patient decision aid asks patients to think about which positive and negative features of the options matter most to them.	<i>The decision aid provides clear direction to consider personal preferences in making the decision. (This may be achieved through balanced examples of how others value the features of each option in order to illustrate how different values may lead to different choices or through explicitly measured values guiding patients to rate or trade-off different features of options)</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The discussion of harms and benefits does not include reference to the personal importance of harms and benefits (only providing the chances of the outcomes happening)</i>		
<b>5.</b> The decision aid suggests ways for patients communicate what matters most to them to others involved in the decision (e.g. health professionals, family members).	<i>The decision aid provides clear strategies to facilitate communication of personal values</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provided direction or suggestions of how to share personal values with others involved in the decision</i>		

Domain Guidance	Strongly agree 4	Agree 3	Disagree 2	Strongly disagree 1	Score	Comments
<p>1. The patient decision aid provides a step-by-step way to make a decision.</p>	<p><i>The decision aid provides clear guidance to the decision-making process through one of the following methods: (a) the structure of the decision aid is stepwise (implicit guidance); or, (b) there is explicit guidance in the form of a worksheet or specified steps; or, (c) there is a thought experiment e.g. "Imagine that you have chosen option A, write down your expectations of the outcomes and how you would feel about them and share this with your practitioner"; or, (d) strategies are included for making /progressing with the decision e.g. an action plan for progressing with the decision</i></p>	<p><i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i></p>	<p><i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i></p>	<p><i>The decision aid does not provided any features of structured guidance</i></p>		
<p>2. The patient decision aid includes tools like worksheets or lists of questions to use when discussing options with a practitioner.</p>	<p><i>The decision aid provides a worksheet or list of questions that is clearly intended to be shared with others involved in the decision</i></p>	<p><i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i></p>	<p><i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i></p>	<p><i>The decision aid does not provided any means to facilitate communication of views/situation to others</i></p>		

<b>Domain Development</b>	<b>Strongly agree 4</b>	<b>Agree 3</b>	<b>Disagree 2</b>	<b>Strongly disagree 1</b>	<b>Score</b>	<b>Comments</b>
<b>1.</b> The development process included finding out what clients' / patients' need to discuss options or decide on courses of action.	<i>The decision aid provides or supporting documentation provides clear evidence of a clients' / patients' needs assessment or involvement in topic selection</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid or supporting documentation does not provided any information about clients' / patients' involvement in the development process</i>		
<b>2.</b> The development process included finding out what health professionals' need to discuss options or decide on courses of action.	<i>The decision aid provides or supporting documentation provides clear evidence of a health professionals' needs assessment or involvement in topic selection</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid or supporting documentation does not provided any information about health professionals' involvement in the development process</i>		
<b>3.</b> The development process included expert review by clients' / patients' not involved in producing the decision aid.	<i>The decision aid provides clear information about review of the decision aid by clients' / patients' not involved in the development process</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid was not reviewed by clients' / patients' or does not provide any information about review</i>		
<b>4.</b> The development process included expert review by health professionals not involved in producing the decision aid.	<i>The decision aid provides clear information about review of the decision aid by health professionals' not involved in the development process</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid was not reviewed by health professionals' or does not provide any information about review</i>		
<b>5.</b> The decision aid was field-tested with patients who were facing the decision.	<i>The decision aid provides clear information about field-testing the decision aid among patients who were facing the decision</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid was not field-tested or does not provide any information on field-testing</i>		
<b>6.</b> The decision aid was field-tested with practitioners who counsel patients who face the decision.	<i>The decision aid provides clear information about field-testing the decision aid among practitioners who counsel patients facing the decision</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid was not field-tested or does not provide any information on field-testing</i>		

<b>Domain Evidence</b>	<b>Strongly agree 4</b>	<b>Agree 3</b>	<b>Disagree 2</b>	<b>Strongly disagree 1</b>	<b>Score</b>	<b>Comments</b>
<b>1.</b> The decision aid (or available technical documentation) provides citations to the studies selected.	<i>The decision aid or supporting documentation provides citations to the scientific evidence used</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provide any information on the scientific references</i>		
<b>2.</b> The decision aid (or available technical documentation) describes how research evidence was selected or synthesised.	<i>The decision aid or supporting documentation explicitly describes the methods for identifying and appraising the evidence</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provide any description of the methodology for collection and appraisal of evidence</i>		
<b>3.</b> The decision aid (or available technical documentation) provides a production or publication date.	<i>The decision aid or supporting documentation clearly states the date of last update</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provide a date of last update</i>		
<b>4.</b> The decision aid (or available technical documentation) provides information about the proposed update policy.	<i>The decision aid or supporting documentation provides clear information on the procedure for updating the evidence</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provide any information on the update policy</i>		
<b>5.</b> The decision aid (or available technical documentation) describes the quality of the research evidence used.	<i>The decision aid or supporting documentation provides an explicit rating of the quality of the scientific evidence used to describe the benefits and risks</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provide any description of the quality of the scientific evidence</i>		

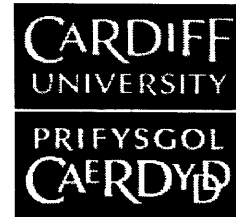
<b>Disclosure and transparency (Disclosure)</b>	<b>Strongly agree 4</b>	<b>Agree 3</b>	<b>Disagree 2</b>	<b>Strongly disagree 1</b>	<b>Score</b>	<b>Comments</b>
1. The decision aid (or openly available technical documentation) provides information about the funding used for development.	<i>The decision aid or supporting documentation explicitly states sources of funding for development of the decision aid</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provide any information about sources of funding</i>		
2. The decision aid includes authors' / developers' credentials or qualifications.	<i>The credentials of individual developers are given clearly in the decision aid itself OR if the credentials of those individuals directly responsible for the development and content of the decision aid are given clearly in supporting materials</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provide any information about credentials of the authors / developers</i>		
<b>Using plain language</b>	<b>Strongly agree 4</b>	<b>Agree 3</b>	<b>Disagree 2</b>	<b>Strongly disagree 1</b>	<b>Score</b>	<b>Comments</b>
1. The decision aid (or available technical document) reports readability levels (using one or more of the available scales).	<i>The decision aid or supporting documentation clearly reports its readability level</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid does not provide any information about readability level</i>		
<b>Evaluation (Evaluation)</b>	<b>Strongly agree 4</b>	<b>Agree 3</b>	<b>Disagree 2</b>	<b>Strongly disagree 1</b>	<b>Score</b>	<b>Comments</b>
1. There is evidence that the decision aid improves the match between the features that matter most to the informed patient and the option that is chosen	<i>The decision aid or supporting documentation clearly reports that evaluation study results demonstrate an improved match between values and choice</i>	<i>Use this rating if you think the patient decision aid fulfils the criterion but there is room for improvement</i>	<i>Use this rating if you do not think that the patient decision aid fulfils this criterion or if unclear</i>	<i>The decision aid or supporting documentation does not provide evidence of evaluation or fails to demonstrate evidence of improved match between values and choice</i>		

## **Appendix 2**

**Interview Schedule for User Perspective.....Page 220**

**Example of Coded Interview Transcript.....Page 223**

**List of Codes (User Perspectives).....Page 224**



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Participant code: \_\_\_\_\_

Occupation: \_\_\_\_\_

Date: \_\_\_\_\_

Marital status: \_\_\_\_\_

Date of birth: \_\_\_\_\_

**Obstetric history:**

Previous pregnancies: \_\_\_\_\_

Number of children: \_\_\_\_\_ Children with a chromosome disorder: \_\_\_\_\_

Previous experience with amniocentesis: \_\_\_\_\_

**Patient Interview Schedule (User Perspective)**

- Introduction to the research topic and aims

*"Thank you very much for agreeing to participate in the research. By participating, you are helping us design a tool that will help pregnant women who are faced with a decision to undergo amniocentesis. By creating a decision tool, we aim to give them accurate information about the options they are offered and see how they react to the support provided".*

- Anonymity and informed consent

*"The interview will last between 30 and 60 minutes. I am recording it in order to analyse it afterwards. Your names will remain anonymous and all the information collected during the interview will be kept strictly confidential."*

*I am working within the rules and regulations that Cardiff University has set."*



➤ Before we start, do you have any questions about this interview and its aim, or something you would like to have more explanation about?

1. Could you now tell me about your experience of having the blood tests for Down's syndrome?

(Probes: Did you understand the process? Do you know why this test is done?)

2. Could you tell me what you were told about the results of the blood tests for Down's syndrome?

(Probes: What did you understand about those results? How did you interpret the information about the risk?)

3. What sort of information do you think women need about the blood test?

4. How do you think this information should be given?

(Probes: Using diagrams or graphics which illustrate the risk?)

5. How were you introduced to the offer of an amniocentesis?

(Probes: Did you understand why you were offered amniocentesis?)

6. What did you already know about the amniocentesis procedure?

7. What are the issues, preferences, or factors that you took into account when you had to make a decision regarding amniocentesis?

(Probes: How did you come to this decision? What helped you make this decision?)

8. We are also interested in your partner's views about this decision. What were the issues, preferences, or factors which he brought up when you discussed amniocentesis testing?

9. How do you feel about your decision regarding amniocentesis?

10. What sort of information do you think women need about amniocentesis?

11. Amniocentesis carries a risk of miscarriage, what information do you think women need regarding this risk?

12. How do you think this information should be given?

(Probes: Using diagrams, graphics or decision trees which illustrate this risk?)

13. Do you have any further questions you would like answered or comments you would like to make regarding this study?

*"Thank you very much for your time and participation".*

# Example of Coded Interview Transcript Using ATLAS.ti (User Perspective)

File Edit Documents Quotations Codes Memos Networks Views Tools Extras Windows Help

P-Docs P-7: Interview Quotes 1:1 Yes, yeah Codes 'put my mind a Memos

01 Interview patient 8

02

03

04 R: So, could you tell me about your experience of having the blood test for Down syndrome?

05 I: Right, hum, we had the, I had the blood test at 16 weeks, hum, basically, it's just a normal blood test, went for it, the results came back, I had a telephone call and then, the lady just explained everything, like my results and everything to come in and see her and that was the initial blood test.

06 R: Did you understand why that test was done for?

07 I: I did understand, but this being my first child, I don't think I probably understood the consequences, hum, and, the worry that ... basically

08 R: You had enough information.

09 I: yes, information was fine.

10 R: What about the results, what were you told?

11 I: Hum, when they phoned me up and gave me the message I thought tat was quite distressing really, the lady on the other end of the phone said to me that the result had come back high and that I needed to come in and see her straight away, hum, which obv I was worried about. I was quite devastated about it to be honest and then when I got to the hospital to speak to the specialist, it was a totally different ballgame, there was no need for me to worry that much when she went into all details, hum, so I wasn't very happy at first with the initial phone call.

12 R: How did you interpret this information, what did you think, what were your thoughts when she told you you're at risk?

13 I: My initial thoughts were straight away. Right, where do I stand? Am I gonna be having a DS children, not maybe I am having one, I thought straight away, yes I am gonna be having one. Hum, I didn't really understand it all to be honest with you. Looking back now, I sometimes regret having this screening test, hum, because obv of the worry that I've got now.

14 R: What sort of information do you think women need about this blood test?

15 I: The paper work was fine, I had the opportunities to ask questions as well. I just don't feel that I did. Hum, I don't feel that I asked, (...), but being young, being like my first child, it's all exciting, I mean, it's the last thing that you want to think about really. But, I mean I should have asked a few more questions.

16 R: How do you think this information should be given to women?

17 I: I don't think the results should be given over the phone. I don't think that should be done. But, there again, I don't think it should be given by a letter either cause I don't think that would be very nice. Hum, it's a noone situation really about the results. As for information, I don't know really. I mean I had a one meeting with a midwife before I had the blood test and as I said, I should have just asked more questions really, or with knowing that it was my first child, maybe she could went in with a little bit more detail for us. Cause she didn't ask if it was my first child. She obviously knew it by my notes and everything, but if she had have asked I suppose she might have knew that I was a bit vulnerable really.

18 R: What about the numerical data, the risk in itself, do you think it would help if it was presented using, pictures or?

19 Hum, she did, she had a little graph, 250 spots on and in the middle was one red spot. Now, as soon as I've seen that my mind was totally, not totally at ease, but was eased a lot more than what it was when she said, look this is your result over the phone, my result was 206 so when she showed me these dots and everything I did feel a lot better. But, when she said over the phone, see I didn't know if it was 1 in 206, I didn't know how the ratio worked, you see, straight away, initially, so yeah, I mean, I was pretty worried at first. But, Hum, I like the dots idea, that was good. I liked that.

- difficulty: did not consider implications of screenin
- satisfaction: information provided
- emotions: panic/stress/upset after "high chance"
- emotions: awful experience, highly distressing
- information needs: immediate information when
- perception: High chance result = baby has DS
- perception: parents seem to consider the wor
- decision: recurrent anxiety after having decid
- emotion: regret re screening test
- difficulty: did not consider implications of screenin
- information needs: screening test purposes and c
- framing of information: visuals needed
- framing of information: visuals needed emo

## List of Codes (User Perspective)

Amniocentesis results: anxiety diminished after 1st result  
Amniocentesis: uncomfortable but not painful  
Attitude: did not expect to get a high chance result  
Attitude: laid back about screening test because 3rd pregnancy  
Creating sense about why screening test came back positive  
Decision support: husband supported her decision  
Decision support: more counselling available if a problem is found  
Decision support: need for more decision support  
Decision support: professionals with unbiased/neutral opinions  
Decision support: support from relevant associations  
Decision support: taking enough time to make a decision  
Decision support: talking to others (family, professionals)  
Decision: agreement between partners  
Decision: differing opinions within the couple (conflicts)  
Decision: immediate decision was to have amniocentesis/ then changed her mind  
Decision: recurrent anxiety after having declined amniocentesis  
Decision: satisfied declined amniocentesis (no regret) even after child born with Down's syndrome  
Decision: satisfied had amniocentesis (even after diagnosis of Down's syndrome)  
Decision: satisfied not to have had amniocentesis although moments of anxiety  
Decision: satisfied with decision to have had amniocentesis  
Decision: shared with husband  
Difficulty: balancing the risk of miscarriage against the risk of Down's syndrome  
Difficulty: conflicts between local/national miscarriage rate  
Difficulty: felt forced into having an amniocentesis  
Difficulty: making a decision in a short time window  
Difficulty: interpreting/understanding the numerical value  
Difficulty: to make a decision based on probabilities  
Difficulty: did not anticipate/prepare for a high chance result  
Difficulty: did not consider implications of screening test  
Difficulty: did not get enough information on screening tests  
Difficulty: did not realise that the screening test result was not certain.  
Difficulty: did not what she would do with the results if problem was found  
Difficulty: high chance result is misleading  
Difficulty: hospital unable to provide updated leaflets about Down's syndrome  
Difficulty: information overload  
Difficulty: lack of information  
Difficulty: lack of information and support about Down's syndrome  
Difficulty: lack of neutral, unbiased information from medical staff  
Difficulty: lack of understanding around risk of miscarriage (foreign patient)  
Difficulty: lack of understanding of what the results (PCR + karyotype) mean  
Difficulty: lack of understanding screening test result  
Difficulty: poor knowledge about amniocentesis before screening  
Difficulty: rushed into making a decision  
Difficulty: strong stereotypes around children with down's syndrome  
Difficulty: total lack of support re continuing pregnancy  
Difficulty: total lack of tact from medical staff  
Difficulty: very biased information, pro-termination  
Difficulty: very difficult decision  
Difficulty: waiting for results  
Difficulty: not informed about possibility of nuchal translucency scan

Dissatisfaction: medical team judgemental, insulting about child with chromosome Disorder  
 Dissatisfaction: amniocentesis results given over the phone  
 Dissatisfaction: delay re-amniocentesis results  
 Dissatisfaction: no real choice as to continue/terminate pregnancy  
 Dissatisfaction: offered amniocentesis over the phone  
 Dissatisfaction: pressure to have a termination  
 Dissatisfaction: pressure to opt for screening test + amniocentesis  
 Dissatisfaction: professional's attitude, choice was not offered, professional's views imposed on her  
 Dissatisfaction: standard of care received in hospital  
 Dissatisfaction: total lack of professionalism from healthcare professionals  
 Emotion: regret re screening test  
 Emotions: affective forecasting  
 Emotions: anxiety decreased after PCR  
 Emotions: awful experience, highly distressing  
 Emotions: felt guilty for not having screening test  
 Emotions: no regret re amniocentesis  
 Emotions: no regret re not having amniocentesis  
 Emotions: panic, shock after diagnostic of Down's syndrome  
 Emotions: panic/stress/upset after "high chance" result  
 Emotions: stress, emotions, vulnerability in deciding  
 Emotions: too emotional to take information in  
 Emotions: very emotional decision  
 Emotions: very upsetting time  
 Framing of information: convert risk of Down's syndrome into a number out of 100  
 Framing of information: multiple ways of presenting the data (to account for individual differences)  
 Framing of information: verbal information needed  
 Framing of information: visuals needed  
 Framing of information: written information needed  
 High chance result: did not feel directly concerned  
 High chance result: panic, stress  
 High chance result: sense of urgency=stress  
 High chance result: too little immediate information  
 Information given: was given local miscarriage rate as well as national miscarriage rate  
 Information needs: impact of IVF on screening test results  
 Information needs: address emotional aspects, difficulty in deciding  
 Information needs: adjust/personalise information to individual differences and background  
 Information needs: amniocentesis does not guarantee a healthy baby  
 Information needs: average Down's syndrome risk in similar age group  
 Information needs: balanced examples of other people's experiences  
 Information needs: be given a choice  
 Information needs: being called back to clinic after high chance result is a routine procedure  
 Information needs: benefits, joys of having a child with a chromosome problem  
 Information needs: consequences of amniocentesis  
 Information needs: detailed information about risk of miscarriage  
 Information needs: finding a balance in the quantity of information given  
 Information needs: for analogies (lottery) (men especially)  
 Information needs: forum, local support group, message board  
 Information needs: hard facts  
 Information needs: how the amniocentesis procedure is done  
 Information needs: immediate information when given high chance result  
 Information needs: infections do not always result in miscarriage (antibiotics etc)

Information needs: information about pain during procedure

Information needs: information about range of problems detected

Information needs: information about what amniocentesis does not detect

Information needs: information around results, implications etc

Information needs: information given before screening test

Information needs: information should be optional (e.g., video of procedure)

Information needs: information to take home

Information needs: karyotype test result and problems detected

Information needs: local miscarriage rate

Information needs: more information about what screening test is looking at

Information needs: multiple ways of framing information

Information needs: neutral, balanced information

Information needs: No detailed information about screening test process (chemicals in the blood etc)

Information needs: No need for overload of information about consequences of amniocentesis, termination etc.

Information needs: number of women who have child with Down's syndrome after high chance result

Information needs: out of all women who are given high chance result, how many undertake amniocentesis

Information needs: practical aspects, arrangements around amniocentesis

Information needs: professionals' opinions

Information needs: range of screening tests available

Information needs: reasons why they are having screening test/amniocentesis

Information needs: reliable information

Information needs: risk factors for miscarriage

Information needs: risks involved

Information needs: screening test purposes and consequences

Information needs: screening test results

Information needs: screening test results are not definite

Information needs: characteristics of IVF pregnancies in relation to amniocentesis

Information needs: understand the screening test results

Information needs: videos (procedure), visuals

Information: read about amniocentesis

Information: search on internet

Information: understood the information

Knowledge: knew a little about amniocentesis before

Perception: amniocentesis is uncomfortable but not painful

Perception: does not know why had screening test

Perception: enough information given before screening test

Perception: experienced pains after the procedure

Perception: extremely reassured by local rate

Perception: High chance result = baby has Down's syndrome

Perception: leaflets were useful but did not help her make a decision

Perception: national average misleading

Perception: negative stereotypes, images on children with Down's syndrome

Perception: never think that screening test result will come back high chance

Perception: painful procedure

Perception: parents seem to consider the worst case scenario: Down's syndrome baby or miscarriage of healthy baby

Perception: risk of miscarriage minimised

Perception: very quick decision

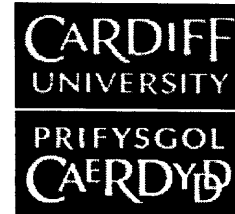
Perceptions: amniocentesis as a reassurance for the parents not the baby

Professional's tendency: systematic provisional amniocentesis booking  
 Reason: avoid anything that may harm baby  
 Reason: being able to prepare if problem is found  
 Reason: age  
 Reason: anomalies detected on the mid-pregnancy scan  
 Reason: compare adjusted risk with cut-off limit  
 Reason: difficulty getting pregnant  
 Reason: does not want to be confronted with other difficult decisions (pregnancy termination)  
 Reason: existing child  
 Reason: existing child with Down's syndrome (no to amniocentesis)  
 Reason: family history of chromosome disorders  
 Reason: have the option to terminate pregnancy  
 Reason: husband away, not here to support her  
 Reason: knowledge, understanding, experience of children with Down's syndrome  
 Reason: medical complication (no amniocentesis)  
 Reason: no termination if a problem was found  
 Reason: not wanting to look after a disabled child  
 Reason: obstetric history  
 Reason: partner's views  
 Reason: practical reasons, being able (or not) to rest for two days  
 Reason: previous experience of amniocentesis  
 Reason: previous miscarriage  
 Reason: reassured by obstetrician expertise  
 Reason: religious beliefs, faith  
 Reason: risk for her age compared to risk in similar age group  
 Reason: risk of infection  
 Reason: risk of miscarriage  
 Reason: risk of miscarriage compared to risk of a problem  
 Reason: stress of not knowing  
 Reason: the numerical value, the risk of a problem  
 Reason: to find out, definite answer  
 Reason: views of friends, family etc  
 Satisfaction: visuals (diagram with dots)  
 Satisfaction: information around amniocentesis procedure  
 Satisfaction: overall information and support  
 Satisfaction: amniocentesis was a positive experience: extra scan...  
 Satisfaction: being able to decide straight away  
 Satisfaction: good information about Down's syndrome  
 Satisfaction: good risk communication  
 Satisfaction: information around screening test  
 Satisfaction: information leaflets  
 Satisfaction: information provided  
 Satisfaction: the diagram with dots  
 Satisfaction: timeline for results  
 Satisfaction: understanding of screening test purposes  
 Screening test: understood why was offered the test  
 Support: friends

## **Appendix 3**

<b>Interview Schedule for Stakeholder Analysis.....</b>	<b>Page 229</b>
<b>Example of Coded Interview Transcript.....</b>	<b>Page 231</b>
<b>List of Codes (Stakeholder Analysis).....</b>	<b>Page 232</b>





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Participant code: \_\_\_\_\_

Position occupied: \_\_\_\_\_

Date: \_\_\_\_\_

## Interview Schedule (Stakeholder Analysis)

- Introduction to the research topic and aims

*“Thank you very much for agreeing to participate in the research. By participating, you are helping us design a tool that will help pregnant women who are faced with a decision to undergo amniocentesis. By creating a decision tool, we aim to give them accurate information about the options they are offered and see how they react to the support provided”.*

- Anonymity and informed consent

*“The interview will last between 30 and 60 minutes. I am recording it in order to analyse it afterwards. Your names will remain anonymous and all the information collected during the interview will be kept strictly confidential.  
I am working within the rules and regulations that Cardiff University has set.”*

☞ Before we start, do you have any questions about this interview and its aim, or something you would like to have more explanation about?

1. What sort of information do you think women need regarding the result of the screening test for Down's syndrome?

2. In what way do you think this information is best presented?

3. What important issues, preferences or factors influence women when they have to make a decision regarding amniocentesis?
4. What difficulties do women come to you with?
5. If there is one thing that women ask you, what is-it?
6. What is the best way to describe and explain the risk of having a baby with Down's syndrome and the false positive and false negative results associated?
7. How do you think this information should be given?  
(Probes: Using charts, diagrams, decision trees? Using number or words?)
8. What sort of information do you think women need regarding the amniocentesis procedure?
6. What do they need to know regarding the PCR test and the karyotyping procedure?
7. What sort of information do you think women need about the risk of miscarriage?
8. How do you think this information should be given?  
(Probes: using diagrams or charts which illustrate this risk? Using number or words?)
9. What do they need to know regarding the results of the amniocentesis and its implications?
10. How do you think this information should be given?  
(Probes: using decision trees, or charts?)
11. Do you have any further questions, or comments you would like to make regarding women's information needs when they are offered amniocentesis?

*"Thank you very much for your time and participation"*

# Example of Coded Interview Transcript Using ATLAS.ti (Stakeholder Analysis)

health professionals interviews april 08 - ATLAS.ti

File Edit Documents Quotations Codes Memos Networks Views Tools Extras Windows Help

P-Docs [P-2: Interview B Beattie] Quotes [2:1 The triple t] Codes [couple's decision] Memos [ ]

01 Interview transcript B Beattie

02

03 R: What sort of info do you think women about the results of the blood test for DS?

04 I: The triple test, I think they need to know what their age related risk of having a DS baby would be and then what the adjusted risk is. I think one of the problem is to try to put that into some sort of prospective and it is useful to use analogies like for a risk of 1 in 360 you could say something like, If i pick one day in the next year and you can guess the same date it gives them a better idea of what the concept of 1 in 360 is. The other thing is that sometimes we use cards with dots on them to try and give an idea of what the risk would be. Hum, percentages are not particularly helpful bc the risk seen of 1 in 200, 1 in 300, people find I think difficult to understand the concept of half a percent, a third of a percent, so it is probably easier to use overall numbers. The problem that they usually have is trying to make a decision about whether to go ahead with the amniocentesis so I think an equally important part of that is understanding the concept of the risk of miscarriage from the amnio and I would often draw them a set of skills and show that on the one hand, you are balancing the risk of miscarriage if they have the amnio, and the other hand, the risk of missing a baby with DS if you don't, and tell them to try and make that judgement

05 R: ok , You mentioned the percentages and the dots, how do you think this info is best presented

06 I: I think depends on the patients. The dots I think are widely used by the midwives, the screening midwives, I tend to think of it, I use the analogy of days of the week, or days of the month, or days of the year. I think a lot of people find that a lot easier to follow.

07 R: ok,

08 I: Depends on the patients. If somebody is a solicitor, a lawyer, high intelligent, they may be quite happy just with row figures.

09 R: What important issues or factors or preferences do you think women take into account when they have to make this decision about going for amniocentesis?

10 I: One of the big problems is that a lot don't understand fully what DS is, so a simple thing is to review with them what their understanding of what the condition is, hum, bc if they consider that DS is something which is very serious then they are more likely to have an amniocentesis. If on the other hand they think that it's something with a very variable presentation and they may have even have experience of another child who has DS in the family or as a neighbour and they may have different views. I think it is important that they understand what DS is and that even if we diagnose it on an amniocentesis, we can't predict the severity of the outcome.

- information needs: to give adjusted numerical risk of
- information needs: use of analogies to communicate
- perceptions: overall numbers easier than percentage
- information needs: understanding the risk of miscarriage
- difficulty: balancing the risk of miscarriage against the
- framing of information: analogies are better than flo
- perception: "clever individuals understand row figure
- information needs: to explain and understand DS
- difficulty: limitations of amniocentesis testing (no pre

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## List of Codes (Stakeholder Analysis)

Couple's decision: couple come together  
Couple's decision: disagreement within couple  
Couple's decision: men tend to follow/support their partner's decision  
Decision support: consider the impact of child with Down's syndrome for the long term  
Decision support: anticipate what will do if problem is found (termination of pregnancy)  
Decision support: consider impact of disabled child on family  
Decision support: couple need to decide why they are having amniocentesis (reassurance, to be able to terminate...)  
Decision support: leave them time to decide  
Decision support: parents need to be reassured about amniocentesis  
Decision support: parents need to know what the worst outcome is (having an affected child/losing a healthy baby)  
Decision support: tendency to seek professional's advice  
Decision support: to facilitate discussion within couple  
Decision support: discuss condition with specialist  
Difficulty: understanding numerical value because no fixed denominator  
Difficulty: lack of understanding of what amniocentesis is testing for  
Difficulty: understanding false positive/false negative results  
Difficulty: "putting the pregnancy at risk"  
Difficulty: accessing the information anytime  
Difficulty: balancing the risk of miscarriage against the risk of a problem  
Difficulty: being rushed into making a decision  
Difficulty: couples/women have to make a difficult decision (responsibility in deciding)  
Difficulty: dealing with statistics  
Difficulty: emotional arousal, understanding gaps  
Difficulty: emotional difficulty in deciding  
Difficulty: information overload  
Difficulty: limitations of amniocentesis testing (no prediction of Down's syndrome severity)  
Difficulty: making a decision without knowing the outcome  
Difficulty: risk of Down's syndrome is equivalent to risk of miscarriage  
Difficulty: understanding the full implications of amniocentesis  
Emotions: very emotional and difficult decision  
Emotions: emotional upheaval (hormonal changes etc)  
Emotions: high chance screening test very emotional "panic, horror"  
Emotions: high stress and anxiety levels  
Emotions: very emotional when called in for counselling session, difficulty assimilating information  
Framing of information: analogies are better than flow charts  
Framing of information: analogies facilitate understanding  
Framing of information: diagram and flow charts are not appropriate  
Framing of information: different sources of information and formats needed  
Framing of information: face to face discussion  
Framing of information: interactive information (internet)  
Framing of information: multiple ways/formats to present information  
Framing of information: need for illustrations  
Framing of information: numerical data framed in different ways  
Framing of information: visuals, pictorials (i.e., diagrams, flowchart) needed  
Framing of information: written information to take away  
Information needs: practical and detailed information around procedure  
Information needs: to describe Down's syndrome condition  
Information needs: understanding potential implications of screening/amniocentesis

Information needs: accounting for the individual variability, personalised information

Information needs: accounting for individual variability in interpreting numerical value

Information needs: addressing screening issues in amnioDex

Information needs: amniocentesis cannot guarantee healthy baby, does not detect all problems

Information needs: amniocentesis does not only test for Down's syndrome

Information needs: anticipate consequences of each option

Information needs: benefits of having amniocentesis

Information needs: both men and women information needs should be addressed

Information needs: chromosome test can reveal gender of the baby

Information needs: consistent information across professionals

Information needs: describe (brief overview) range of abnormalities that may be detected

Information needs: describe the different degrees of impairment of people with Down's syndrome

Information needs: detailed information about risk of miscarriage (who miscarries, why?)

Information needs: give balanced, neutral information

Information needs: give national miscarriage rate only

Information needs: having enough time to decide

Information needs: how the screening result is calculated

Information needs: inform women about limitations of local rate (scientific validity, little data etc)

Information needs: information given before screening test

Information needs: information needs to be optional/gradual (different levels of information offered)

Information needs: know all options available (possibility of changing their minds)

Information needs: make sure crucial information is understood

Information needs: making sense of the risk/of the statistical chance

Information needs: mention and explain termination

Information needs: mention the range of abnormalities

Information needs: need to consider worst case scenario

Information needs: need to explain complete package of screening

Information needs: no detailed description about chromosome problems potentially detected

Information needs: no detailed explanation about termination

Information needs: no mention of termination

Information needs: provide numerical value (screening test)

Information needs: risk to lose a healthy baby

Information needs: screening test does not give a definite answer

Information needs: screening test is not 100% accurate

Information needs: show the pathway post-screening test

Information needs: timescale of amniocentesis, results etc

Information needs: to communicate false negative and false positive results

Information needs: to distinguish screening and diagnostic tests

Information needs: to explain and understand Down's syndrome

Information needs: to give adjusted numerical risk of Down's syndrome + age related

Information needs: to give the local AND national miscarriage rate

Information needs: to know what is detected by the karyotype test

Information needs: to know what the PCR test detects (+ timescale)

Information needs: to understand risks involved and where difficulties may arise

Information needs: to understand the limitations of screening

Information needs: understand implications of a positive amniocentesis result

Information needs: understand implications of high chance result

Information needs: understand purposes of screening test

Information needs: understand screening test results

Information needs: understanding the risk of miscarriage

Information needs: updated information

Information needs: use of analogies to communicate numerical risk

Information needs: use very precise and appropriate language

Information needs: verbal information backed up with written material

Information: internet search

Information: simple written English (plain language)

Information: tailor the information to man/women's needs

Information: mention termination but do not give details

Miscarriage rate: tendency to rely more on local rate than national rate

Perception: some women want more information

Perception: people assimilate more if not rushed in making a decision

Perception/miscarriage rate: no value to the Wales national figure

Perception/miscarriage rate: operator's rate more relevant than Welsh national figure

Perception/professionals' difficulties: practical demands determine quantity of information given

Perception: "clever individuals understand row figures"

Perception: amniocentesis is a straightforward procedure

Perception: amniocentesis is the woman's decision

Perception: amniocentesis is useful in order to prepare (when termination is not an option)

Perception: better to give information verbally

Perception: conflict between husband and wife

Perception: decision rushed in the past (bad practice)

Perception: describing what the test picks up is too much information

Perception: false positive and negatives are not explained to patients

Perception: good understanding of Down's syndrome

Perception: good understanding of amniocentesis procedure

Perception: in general, understanding of screening test result

Perception: increase in uptake of screening test (lack of information pre-screening)

Perception: information acquired and understood progressively

Perception: karyotype + PCR detect everything (100%)

Perception: the majority of women are very upset, cry

Perception: male partner needs to be involved in decision

Perception: men tend to leave the responsibility to decide to the women

Perception: miscarriage is less traumatic than having a baby with Down's syndrome

Perception: no need for a lot of verbal info about amniocentesis

Perception: no need to describe termination

Perception: no need to discuss the condition with specialist if decision to terminate pregnancy is made

Perception: no value in communication of false positive and false negative estimates

Perception: not essential to describe Edwards' and Patau's syndromes

Perception: numerical data (risk) influences decision

Perception: poor understanding/knowledge of what Down's syndrome is

Perception: pregnancy termination is not the only reason for amniocentesis

Perception: risk of miscarriage depends on operator

Perception: some people feel insulted by analogies

Perception: the decision should be shared within the couple

Perception: the karyotype test result is misleading.

Perception: video is not necessarily the right way to convey information

Perception: women are aware of what Down's syndrome is

Perception: women are given too much information

Perception: women are satisfied with quantity of information

Perception: women better informed

Perception: women carry the main burden of care  
 Perception: women have amniocentesis for reassurance  
 Perception: women mainly rely on the PCR test results  
 Perception: women more inclined to undertake amniocentesis  
 Perception: women need to be reassured by the consultant's expertise  
 Perception: women never anticipate getting a high chance result "it can't be me"  
 Perception: women undertake screening tests without thinking about implications  
 Perception: detailed information about Down's syndrome should be given after problem found only  
 Perception: information and decision support hugely improved in the past five years  
 Perception: local rate should be used very carefully (little research data available)  
 Perception: local rates should not be used  
 Perception: men and women have different information needs (i.e., different ways of interpreting numerical value)  
 Perception: overall numbers easier than percentages  
 Perception: the majority undertake amniocentesis for reassurance  
 Perception: couples not worried about procedure but about consequences (i.e., miscarriage)  
 Perception: most people have amniocentesis to be able to terminate if problem  
 Professionals' difficulties: confusion regarding definition of false positive and negative results  
 Professionals' difficulties: specific and complex information provided by professionals who lack the expertise (ex: limited expertise in genetics)  
 Professionals' difficulties: conflicting screening procedures (NHS/private sector)  
 Professionals' difficulties: describing Down's syndrome in enough details in a consultation  
 Professionals' difficulties: lack of information about factors causing miscarriage or consequences of amniocentesis  
 Professionals' difficulties: lack of neutral judgement  
 Professionals' difficulties: more knowledge about Down's syndrome than other conditions  
 Professionals' difficulties: time constraints  
 Professionals' difficulties: which screening test to recommend  
 Professionals' difficulties: lack of expertise around genetic problems (especially midwives)  
 Professionals' difficulties: too much information is perceived as paternalistic  
 Professionals' role: to make sure that people understand  
 Reason: impact of a disabled baby on life, family etc  
 Reason: existing children  
 Reason: against pregnancy termination  
 Reason: age  
 Reason: concerned about people's reactions to their decision  
 Reason: contact with, understanding of Down's syndrome  
 Reason: couple's stability  
 Reason: cultural characteristics  
 Reason: desire to be prepared  
 Reason: difficulty getting pregnant  
 Reason: existing child with disability  
 Reason: find out if there is a problem  
 Reason: gut feeling, personal values, experience  
 Reason: how to cope with a disabled child  
 Reason: knowledge about amniocentesis  
 Reason: obstetrician's experience  
 Reason: partner's views  
 Reason: previous experience of amniocentesis  
 Reason: previous history of genetic abnormality  
 Reason: previous miscarriage, abortion  
 Reason: previous obstetric history

Reason: professionals' influence  
Reason: religious beliefs  
Reason: risk of miscarriage  
Reason: risk of miscarriage compared to risk of a problem  
Reason: to be given options if problem found (i.e., termination)  
Reason: to prepare if baby has problem  
Reason: view on termination  
Reason: opinions of family and friends  
Risk of miscarriage: realistic evaluation about local miscarriage rate  
Risk of miscarriage: relevance of operator miscarriage rate  
Screening test/ emotions: regret  
Screening test/difficulty: lack of understanding of screening test purposes  
Screening test: acceptance without understanding it  
Should people be offered alternatives to NHS screening procedure?  
Women's concern: operator's expertise  
Women's concern: pain during procedure  
Women's concern: risk of miscarriage  
Women's concern: why people miscarry (predisposing factors?)  
Women's/ couples' attitudes: seek assurance that making the right decision

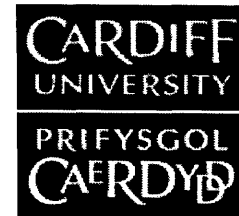


## **Appendix 4**

<b>Questionnaire for Field-Testing With Health Professionals.....</b>	<b>Page 238</b>
<b>Interview Schedule for Field-Testing With Pregnant Women.....</b>	<b>Page 239</b>
<b>Example of Coded Interview Transcript (Field-Testing).....</b>	<b>Page 241</b>
<b>List of Codes (Field-Testing With Pregnant Women).....</b>	<b>Page 242</b>

## Questionnaire (Field-Testing With Health Professionals)

<b>amnioDex version 1</b> <u><a href="http://www.amniodex.com">www.amniodex.com</a></u> We are looking at the:	<b>Comments</b> (+ suggestions for improvements)
<b>1. Navigation</b> - Is the website easy to navigate? - Is the structure of navigation self-explanatory? - Any comments on the demo or contents list? - Other comments or suggestions...	
<b>2. Design/Layout</b> - What do you think of the homepage? - What do you think of the colours used? - What do you think of the images? - What do you think of the general design of the website? - Other comments or suggestions...	
<b>3. Video clips</b> - What do you think of the video clips of women's stories? - What do you think of the videos of health professionals? - What do you think of the video of the amniocentesis procedure? - Other comments or suggestions...	
<b>4. Deliberation tools</b> - Weighing it up; - Your most important reason; - Talking to others. - Other comments or suggestions...	
<b>5. Message board</b> - Comments or suggestions...	



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## Interview Schedule (Field-Testing With Pregnant Women)

Participant code: \_\_\_\_\_

Occupation: \_\_\_\_\_

Date: \_\_\_\_\_

Marital status: \_\_\_\_\_

Amniocentesis: \_\_\_\_\_

Date of birth: \_\_\_\_\_

### Obstetric history:

Previous pregnancies: \_\_\_\_\_

Number of children: \_\_\_\_\_ Children with a chromosome disorder: \_\_\_\_\_

Previous experience with amniocentesis: \_\_\_\_\_

Number of weeks pregnant: \_\_\_\_\_

- Introduction to the research topic and aims

*"Thank you very much for agreeing to participate in the research. By participating, you are helping us pilot a tool that will help pregnant women who are faced with a decision to undergo amniocentesis. By creating a decision tool, we aim to give women accurate information about the options they are offered and see how they react to the support provided".*

- Anonymity and informed consent

*"The interview will last between 30 and 60 minutes. I am recording it in order to analyse it afterwards. Your name will remain anonymous and all the information collected during the interview will be kept strictly confidential.*

*I am working within the rules and regulations that Cardiff University has set."*

☞ Before we start, do you have any questions about this interview and its aim, or something you would like to have more explanation about?

1. I am going to show you the decision tool for amniocentesis, amnioDex. Would you please have a look at it?

☞ *The researcher will observe how amnioDex is used. The "think-aloud" technique will be used as the user navigates through the website. Users will be asked to describe their thoughts as they use the intervention.*

2. What do you think about this decision tool?

3. Do you find it easy to use and understand?

(Probes: is the content clear and easy to follow? Is the information regarding risks comprehensible and well presented?)

4. In what way do you think it can help women who are offered amniocentesis?

(Probes: What are the advantages of this decision tool?)

5. Are there areas in this decision tool that need changing?

(Probes: language, graphic elements, information clarity, risk information, navigation?)

6. Do you think that the information presented in this decision tool is easily understandable for all women who are offered amniocentesis testing?

7. Do you have any suggestions about ways of improving this decision tool?

8. Do you have any further questions you would like answered or comments you would like to make?

*"Thank you very much for your time and participation".*

# Example of Coded Interview Transcript (Field-Testing With Pregnant Women)

field testing phase 2+ - ATLAS.ti

File Edit Documents Quotations Codes Memos Networks Views Tools Extras Windows Help

P-Docs P14: P Quotes 15:3 It's fine, it gives you enough.. (45:45) Codes Benefi Memos

009 I: yeah, I've got it.

010

011 R: what I'd like you to do, if that's ok with you, is to take a look at the website, as if you hasn't made a decision yet and look at the sections that are of interest to you, and tell what you're doing and what you think when you're doing it. It's called the think aloud technique, so, I'm just asking you to think aloud, whenever you're doing something on the website. Does it make sense?

012 I: yeah

013 R: you've made a decision about amniocentesis already haven't you?

014 I: yeah. You should have let me know the website three days before. It may have helped me make a decision.

015 R: did you have the amniocentesis?

016 I: yes I have

017 R: so just tell me what you're doing when you're doing it and then, when you've seen enough, I'll ask you a few questions. So, where are you now?

018 I: I am in the first section, it's your choice. Harms and benefits.

019 R: ok, so if you tell me what you think about the information, or whether anything could be improved, that would be great.

020 I: Hum, I think it's quite straightforward, easy to read, information is simple and clear. Hum, not like I mean, not like most research, I mean like, what's that called, professional knowledge that people may not understand fully because it's just simply, it's clear what amniocentesis is. (long silence)

Instructions: think aloud technique

Using amnioDex: yes n=9 (2,3,4,5,6,9,11,12)

Instructions: think aloud technique

satisfaction: easy to understand, dear info

## List of Codes (Field-Testing With Pregnant Women)

Benefits of amnioDex: deliberation tools  
Benefits of amnioDex: facilitate/increase understanding  
Benefits of amnioDex: information tailored to UK population  
Benefits of amnioDex: informed decision + reinforces decision  
Benefits of amnioDex: more information than a leaflet (e.g., other conditions detected)  
Benefits of amnioDex: possibility of looking for information at your own pace  
Benefits of amnioDex: simple information, easy to understand  
Benefits of amnioDex: testimonials from people in similar situation  
Benefits of amnioDex: balanced information  
Benefits of amnioDex: the message board  
Benefits of amnioDex: very comprehensive information  
Deliberation tools: too clinical  
Deliberation tools: weighing it up helps visualising the decision  
Deliberation tools: decision is too important to use tools  
Deliberation tools: difficulty dragging, dropping  
Deliberation tools: beneficial to have both tools  
Deliberation tools: clear instructions, easy to use  
Deliberation tools: cover all important reasons  
Deliberation tools: difficulty using your most important reason  
Deliberation tools: difficulty using weighing it up  
Deliberation tools: emotional subject, not inclined to weight pros and cons  
Deliberation tools: facilitate understanding  
Deliberation tools: getting as much information as possible is crucial  
Deliberation tools: help making a decision  
Deliberation tools: helpful to have reasons listed  
Deliberation tools: helpful to weight up pros and cons  
Deliberation tools: helpful/confirmed decision  
Deliberation tools: it takes longer to work out your most important reason  
Deliberation tools: your most important reason is confusing  
Deliberation tools: your most important reason: good concept  
Deliberation tools: your most important reason: too complex/understanding difficulties  
Deliberation tools: need to have a demo built in  
Deliberation tools: preferred weighing it up  
Deliberation tools: quite useful  
Deliberation tools: unable to use the tool independently  
Deliberation tools: weighing it up is more immediate  
Deliberation tools: weighing it up is more intuitive/more useful  
Deliberation tools: weighing it up = useful tool  
Deliberation tools: weighing it up increases clarity  
Difficulty: think-aloud technique  
Difficulty: general navigation (e.g., closing new windows)  
Difficulty: technical difficulty (e.g., flash was not installed on user's computer)  
Difficulty: technical difficulty, (e.g., too slow, difficulty loading video clips)  
Difficulty: top tabulation not salient enough  
Difficulty: two different miscarriage rates  
Difficulty: seeing scroll bar in personal stories  
Dissatisfaction: deliberation tools  
Dissatisfaction: list of reasons is not comprehensive  
Dissatisfaction: information about karyotype test is not sufficient  
Dissatisfaction: It's your choice too general  
Dissatisfaction: no "doesn't apply" box

Dissatisfaction: not enough information

Dissatisfaction: some sections were too brief

Improvement: add information about developers' credentials/occupations

Improvement: add information about non-invasive diagnostic procedures

Improvement: add information about residual risk of miscarriage

Improvement: blurb on homepage with aims of website

Improvement: mention cultural differences

Improvement: more information about cystic fibrosis

Improvement: more information about risk of miscarriage

Improvement: more specific information

Improvement: not clear what talking to others is for?

Improvement: professional on message board

Improvement: specific information, examples of difficulties encountered

Improvement: video of woman who declined amniocentesis, positive outcome

Improvement: add a pop-up demo

Improvement: add branding to the website so users can trust this resource

Improvement: advertise website during Down's screening or after high chance

Improvement: amniocentesis procedure clip shorter

Improvement: change order of videos

Improvement: disclaim about video clips

Improvement: email talking to others

Improvement: info about blood sample potentially taken if contamination of amniotic fluid

Improvement: information about chromosomes test could be clearer

Improvement: more generic information about amniocentesis on homepage

Improvement: need to specify meaning of amnioDex

Improvement: instructions of weighing it up

Improvement: search box

Improvement: there need to be messages on the message board

Improvement: titles in top tabulation could be improved

Improvement: what's next after said no to amniocentesis

Limitations of think-aloud technique

Needs: information about what specifically happens in the UK

Needs: make an informed decision

Needs: more information (internet search etc)

Opinion: no new information on amnioDex compared to leaflets

Opinion: the leaflet covers a lot

Opinion: website does not replace face to face interaction

Opinion: website would have been useful if no leaflets given

Opinion: videos are interesting but not necessarily helpful

Satisfaction: section about conditions that will not be detected

Satisfaction realistic pictures of people

Satisfaction: addresses the most important issues

Satisfaction: aesthetically pleasing

Satisfaction: comprehensive info about chromosome problems

Satisfaction: comprehensive information about risk miscarriage

Satisfaction: contacts (ARC, Down's Syndrome Association)

Satisfaction: double navigation structure (top/side)

Satisfaction: easy to navigate

Satisfaction: easy to understand/clear information

Satisfaction: good information

Satisfaction: good layout

Satisfaction: good/useful resource

Satisfaction: inviting website

Satisfaction: section talking to others

Satisfaction: message board

Satisfaction: multiple ways of framing information (e.g., diagram with dots)  
Satisfaction: nice colours  
Satisfaction: right amount of information  
Satisfaction: screening test result calculation (very helpful)  
Satisfaction: section "frequently asked questions" very good  
Satisfaction: the website is not US based  
Satisfaction: user friendly, not too medical or clinical  
Satisfaction: very comprehensive information  
Satisfaction: very good resource  
Satisfaction: very good, would recommend to women in similar situation  
Satisfaction: video amniocentesis procedure  
Satisfaction: videos clips  
Satisfaction: videos offer varied examples, balanced opinions  
Satisfaction: written plain English  
Satisfaction: you can relate to the website/identify with women  
Using amnioDex: had partner using it as well  
Using amnioDex: no  
Using amnioDex: yes  
Using amnioDex: yes (provided the tool is available at the right time)



**Appendix 5**

**Outline Protocol of the AmnioDex Randomised Controlled Trial.....Page 246**

# Outline Protocol of the AmnioDex Randomised Controlled Trial

## Principal Investigator: Glyn Elwyn

### 1. Specification of research question:

Please state in one sentence the research question to be addressed.

Does a web-based decision support intervention increase informed choice for women offered amniocentesis compared to usual practice?

### 2. Importance of the health problem to the NHS:

Please describe the frequency of the health problem in the population and its impact on patients and the NHS.

#### A COMMON INVASIVE PROCEDURE WITH RISK OF MISCARRIAGE.

Amniocentesis is an invasive procedure that is offered to approximately 60,000 individuals / year in the UK and there is evidence that women are not sufficiently informed or supported prior to making a decision. Amniocentesis involves collecting amniotic fluid in order to obtain foetal cells so that DNA analysis and karyotyping can occur. The procedure is offered to between 5 - 10% of women who are calculated to have a 1 in 250 risk of foetal anomaly from ante natal screening tests (blood test), or because of advanced maternal age, history of chromosomal abnormality or anomaly detected on ultrasound. Screening tests for Down's syndrome are offered to all pregnant women in the United Kingdom. Undergoing amniocentesis is an emotionally charged decision, at a time of increased vulnerability, time pressure and with variable, often unsatisfactory support. The procedure leads to high levels of anxiety and stress because it is associated with an estimated 1% loss of normal pregnancies (post-procedure abortion) (Gaudry et al. 2008) and, in approximately 4% of women undergoing amniocentesis, a positive amniocentesis result (anomaly detection) will lead to a decision of whether or not to continue the pregnancy. However, the trade-off between the 1% miscarriage risk and the gain in information for 4% of women is not available in the current information to patients and there has been a general concern that more support is required to help women and their partners arrive at informed decisions.

#### LACK OF INFORMED CHOICE.

Achieving informed choice is a key principle in the NHS: it respects individual autonomy, demonstrates patient-centred care, reduces medico legal costs and potentially avoids unnecessary resource use where patients perceive inappropriate care. There are documented concerns that offers of amniocentesis either leave patients feeling unsupported in attempting to make a decision or that the procedure is regarded as routine, i.e. not requiring deliberation (Van Den Berg et al. 2006). In addition, there is evidence of health inequality: low health literacy leading to less opportunity to engage with information. The proposed intervention aims to address these problems.

### 3. A description of the technology and its possible effect on health status:

You should also discuss current and projected use in the NHS, with approximate costs.

**DESCRIPTION:** AmnioDex is a web-based decision support intervention designed to support women / partners to arrive at informed decisions about amniocentesis. AmnioDex has been developed and field-tested using multidisciplinary inputs over the last three years, using the MRC complex intervention framework, including theory, modeling, stakeholder needs assessment and evaluation (see URL [www.amniodex.com](http://www.amniodex.com)). AmnioDex contains information about the procedure, the risks, the potential results and the implications. It contains 14 patient videos and interactive elements to help patients consider harms and benefits. The intervention fulfils the International Patient Decision Aids Standards (IPDAS) (Elwyn et al. 2006).

**PROJECTED USE:** by women after antenatal staff referral. Costs are minimal, limited to update and dissemination activities (e.g. web links). Systematic reviews of decision support indicates that text / video versions of these interventions, when used alongside routine clinical support, increase knowledge, decision quality and increase the accuracy of risk perception (O'Connor et al. 2007)

### 4. Summary of the current evidence:

Please describe the current knowledge and outline other research taking place in this area. You should discuss how the proposed research will add to the existing evidence base. You must also consider any relevant published or ongoing HTA programme projects.

The Cochrane Review of patient decision aids contains the results from 55 randomised trials (O'Connor et al. 2007). These trials have been conducted over the last decade or so and the interventions have been either information booklets or videotapes (recently DVDs). These interventions are normally used as adjuncts to professional care. The results have been consistent: patients in the active arms achieve greater knowledge, greater accuracy of risk perceptions, greater involvement in decision making and improved decision quality. However, there is an increasing trend for these interventions to become web-based, thus reducing the cost of production (in the long term), and theoretically increasing access and allowing wider dissemination. A review of existing interventions for amniocentesis (Durand et al. 2008) identified six decision support interventions for amniocentesis worldwide, of variable quality and limited effectiveness, none of which are actively implemented in clinical settings. Two, US-based tools, are web-based. None are in current use in the UK. It is not known whether the beneficial effects demonstrated in the trials (across clinical domains) of decision support leaflets / linear videos can be replicated when these interventions move to the web, given potential barriers such as differential access and a variation in interest in using web-based materials and the lack of concurrent professional support and advocacy. Nevertheless, we know that there are significant concerns about individuals' ability to achieve informed choice and a recognised need to improve the availability of decision support. There is insufficient access under current arrangements to high quality evidence-based information about the decision to have or not have amniocentesis. This is complicated by the fact that the choice should ideally be made by the parents –to-be and not assumed to be a choice that a woman makes alone. Web-based resources allow much greater opportunity for partners and family members to review the information and arrive at an agreed decision, rather than a choice that a woman has to face alone in clinic as often occurs. We recognise though that in some situations this will require helping people gain web access via community based locations. This trial would aim to produce evidence that well-designed web-based decision support interventions are able to achieve: 1) decisions about undergoing amniocentesis that are deliberated, well-informed and better aligned with user preferences (informed choice); 2) that it is feasible to deliver information about a difficult choice by using an online medium, and; 3) that it is possible, by designing tools that are accessible and interactive, to reduce inequality attributable to low levels of health literacy. In addition, 4) the trial would provide data about the costs of the intervention and the gain in informed choice compared to current practice.

#### **5. What outcomes will be measured?**

1. The primary outcome is the construct of informed choice (Marteau et al. 2001). This measure postulates that informed choice is defined by a consistent and congruent attitude to the uptake (intention / behaviour) of the procedure, provided the individual has high knowledge; positive attitudes to the test and taking the test, or vice versa. Informed Choice will therefore be assessed using an adapted version of the Multidimensional Measure of Informed Choice that includes Attitudes and Knowledge subscales adapted from Goel (Goel et al. 2001).
2. Anxiety: Anxiety will be measured using the validated six-item short form of the Spielberger (State) Anxiety Inventory.
3. Amniocentesis uptake and reported service use.
4. Decision Regret (Brehaut et al. 2003). This is an important measure of decision outcome, which we will repeat at follow up.
5. A process measure (delibeRATE), a scale developed by the research team to estimate the level of deliberation achieved.

#### **6. Summary for the Non-Expert**

Please provide a summary of sections 1 to 5. This summary should enable the non-expert reviewer to understand how the proposal addresses a question important to the NHS, how and where the research will be carried out, what outcomes will be used to assess the success of the research, what if any, are the ethical issues involved in this study and arrangements for handling these, why this team is well placed to carry out the research and provide justification for the costs requested (including any NHS costs).

**PROBLEM:** Every year, 60,000 pregnant women in the UK are offered amniocentesis, where amniotic fluid from the uterus is examined for foetal abnormalities, specifically Down's, Patau's or Edwards' syndromes. If an affected baby is identified, women are given a choice to continue or terminate the pregnancy. However, amniocentesis carries a 1% risk of a miscarriage. The decisions – to have an amniocentesis with the possible decision about continuing the pregnancy – are both difficult and stressful. Many women report a lack of information and support at this time (Van Den Berg et al. 2006). **INTERVENTION:** Decision aids for patients have been developed to help individuals face difficult decisions. These interventions, when used alongside clinical services, increase knowledge, improve the accuracy of risk perception and increase informed choice. AmnioDex is designed to support women and their partners to decide whether or not to have amniocentesis – see [www.amniodex.com](http://www.amniodex.com). The tool contains information about the procedure, the risks, the expected test results, the abnormalities detected and the potential implications. **HOW & WHERE:** Women offered amniocentesis will be invited, consented, and randomised to usual practice or offered access to AmnioDex, at home, at the antenatal clinic or at community locations. At the point where they decide about the procedure, we will measure to what extent their choice is informed and aligned with their preferences. One month after the decision to have amniocentesis or not we again assess regret, anxiety and well-being. **ETHICAL ISSUES:** Patients who view amnioDex may become more aware of the dilemma and be more anxious. However, the NHS and the General Medical Council stresses the importance of informing patients of procedural harms as well as benefits. The patients will continue to have access to the counselling provided by the antenatal services. **TEAM:** The team has an international reputation for the design and evaluation of decision support interventions and is composed of obstetricians, midwives and psychologists who are experts in the field. Collaborators include UK-level policy advice (Foetal Anomaly Screening Programme), Antenatal Results and Choices (ARC) and Down's Association. **COST:** The intervention has already been designed and field-tested but requires evaluation in the NHS context. Costs are for a multi-site randomised trial in six antenatal services and the task of recruiting women.

**Objectives Please note: The following sections (D onwards) of the form are used (along with those earlier) in the second stage of the assessment process where the study design and scientific merit are also scrutinised. You should provide a clear explanation of your intended study.**

**Provide a clear summary of your research objectives.**

The objectives are:

- 1) To assess whether providing access to amnioDex, a web-based decision support intervention, leads to increased levels of informed choice (primary outcome) for people offered an amniocentesis. Informed choice will be based on the conceptualisation that when knowledge is high and where attitudes and intentions (or behaviours) are aligned with each other, then informed choice can be assumed, and will be assessed using the Multidimensional Measure of Informed Choice (Marteau et al. 2001).
- 2) To assess to what degree people who are offered the amnioDex resource, access it, use it and gain benefit (measured by web-log analysis, informed choice and secondary outcomes such as reduced decisional regret and the degree of deliberation achieved).
- 3) To gain information about the costs of developing, maintaining and updating a web-based decision support and whether the levels of access and the effects obtained are achievable and cost-effective.

### **Summary of Project**

**Please provide a summary of your proposed research using the headings listed in the Guidance Notes.**

**DESIGN:** A multi-centre randomised controlled trial with randomisation of women to intervention (access to amnioDex) versus usual practice trial arms (see flow chart).

**SETTING:** Hospital outpatients, specifically antenatal clinics, with access to intervention at home, antenatal clinic or by arrangements in other community locations.

**EVIDENCE REVIEW:** The Cochrane systematic review of decision aids has summarised the effects of 55 randomised controlled trials to date (O'Connor et al. 2007), although only three are of web-based tools. We have conducted a systematic review of decision support for amniocentesis, which shows a clear need for the development of better decision support that can be accessed with ease (Durand et al. 2008). A clear research gap exists.

**TARGET POPULATION:** Women offered amniocentesis are the target population. This target group is c. 6% of all women who undertake antenatal screening tests. Women are offered amniocentesis when antenatal serum screen identifies a women at 'high risk' (typically a 1 in 250 risk) of having a baby with a major chromosomal abnormality. Women offered amniocentesis after ultrasound scans (between 14-24 weeks) or because of advanced maternal age are also eligible for inclusion in the trial.

**HEALTH TECHNOLOGY:** The technology in this proposal is web-based decision support and has been developed after formal needs assessment (Durand et al. 2009) and extensive field-testing (see [www.amniodex.com](http://www.amniodex.com)). It contains 54 pages of information, 14 short videos giving patient experiences, two deliberation tools to support preference clarification and a users web-forum. We aim to compare the impact of this intervention against usual practice, i.e. the existing information and support. Decision support interventions are defined as interventions that describe and justify the conditions where clinical equipoise exist, they provide information about options and about the short, intermediate and long-term outcomes which have relevant and important consequences for decision-makers (Elwyn et al. 2006).

**MEASUREMENT OF COSTS & OUTCOMES:** Although the costs of developing the web-based intervention have already been incurred, these would be included in a cost-model, including the projected costs of decision support dissemination, of updating, maintaining and hosting the website over time. The primary outcome of the work will be the assessment of informed choice: Multidimensional Measure of Informed Choice. This measure is based on Marteau's conceptualisation that when knowledge is high (Marteau et al. 2001), and where attitudes and intentions (or behaviours) are aligned with each other, then informed choice can be assumed. Clearly, it is not our objective to reduce or increase amniocentesis rates, merely to ensure informed uptake. We would however hypothesise that, secondary outcomes, such as decision regret will be lowered. Data collection by call centre applied questionnaires (existing infrastructure at Cardiff University).

**SAMPLE SIZE:** In order to detect an improvement in informed choice from 50% to 65% with 90% power at a 5% significance level, a total of 460 women are required. In order to allow for loss to follow-up of 20% a total number of 575 women offered amniocentesis will be recruited. There is little evidence on the current level of informed choice in this group using this measure. Therefore a conservative value of 50% has been used.

**PROJECT TIMETABLES:** Total study duration 33 months. Months (M) 0-6: trial set up, recruitment of project staff and obtaining the involvement of 6 x 0.5 FTE NHS Trust research midwives to recruit patients, ethical and research and governance approval in the 6 NHS antenatal clinics required. In addition, the identification of local community locations for allowing women who have limited or no internet access alternative access points for AmnioDex. M 7-21: recruitment of women offered amniocentesis. Follow up: M 7-23. Data clean: M 7-25. Statistical analysis: M 25-30. Report writing: M 27-33.

**RECRUITMENT RATE:** We calculate the need to be able to include antenatal units that, over the recruitment period of 15 months, would care for 30,000 pregnant women, giving us an estimated 1,800 who would be offered amniocentesis. We have the support of six antenatal units (in the South West, Cardiff, Newport and Bristol, and around the West Midlands (South), including University Hospital of Coventry and Warwickshire, where co-applicants (Profs Fiander and Thornton) have existing collaborations and research infrastructure, comprising experienced research midwives. We therefore estimate 30-40% recruitment rate (600) and so are we confident of achieving the target sample of 575 women during the 15 month recruitment window.

**Please provide details about any related (planned or active) grants held by any member of your research team in this or similar research areas. You should include a clear explanation of how the research being proposed in this application will fit.**

This proposed work builds on two three year grants from CR UK to build decision support interventions for men considering PSA (see [www.prosdex.com](http://www.prosdex.com)), now in web-based clinical trial and for women facing breast surgery choices (see [www.bresdex.com](http://www.bresdex.com)), under development and evaluation. Glyn Elwyn co-leads the International Patient Decision Aids Standards Collaboration (IPDAS) and the decision laboratory at Cardiff University [www.decisionlaboratory.com](http://www.decisionlaboratory.com). AmnioDex represents a more sophisticated yet simple to use intervention and we wish to investigate whether this potentially very low cost method of dissemination can yield informed choice for women at a stressful time in their lives.

SEWTU is currently coordinating the Building Blocks trial, which is recruiting 2400 teenage mothers from maternity services across England to evaluate the Family Nurse Partnership. It also has a full submission with the NPRI for a trial of a lifestyle intervention for pregnant women who are obese. Neither of these studies competes with this trial, but this trial will build on the established working relationships of the unit with maternity and midwifery services across England and Wales.

**Please say with which of the UK Clinical Research Networks (<http://www.ukcrn.org.uk>) you intend to link for this research**

The proposal will be supported by CLRN in England & CRC Cymru. Prof Thornton, Clinical Director, West Midland CLRN, will mobilise a management and midwifery team to aid set-up and trial recruitment.

**Please list any benefits you may have identified from working with the network(s).**

The CLRN and CRC Cymru will provide help and guidance for study set-up and recruitment. The outline will be considered by the Specialty Group for Reproductive Health and will enrol additional sites and access CLRN support if these are required outside Wales and West Midlands (South). The benefits have been our ability to identify and obtain the co-operation of two clusters of ante-natal centres, and the advantage of being able to access the infrastructure support of research midwives.