

**Being a Carrier and Living with the BRCA  
Gene Mutation: An Interpretive  
Phenomenological Study of the Experiences  
of Women Who Elect Risk-Reducing  
Bilateral Mastectomy, their Husbands and  
Family**

**Thesis submitted in partial fulfilment for the Doctor of Philosophy by  
Nicola West**

**School of Healthcare Sciences  
Cardiff University**

**2016**

DECLARATION

This work has not previously been accepted in substance for any degree and is not concurrently submitted in candidature for any degree.

Nwest 8.8.17

Signed ..... (candidate) Date

**STATEMENT 1**

This thesis is being submitted in partial fulfilment of the requirements for the degree of Ph.D.

Nwest 8.8.17

Signed ..... (candidate) Date

**STATEMENT 2**

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

Nwest 8.8.17

Signed ..... (candidate) Date

**STATEMENT 3**

I hereby give consent for my thesis, if accepted, to be available for photocopying and for inter-library loan, and for the title and summary to be made available to outside organisations.

Nwest 8.8.17

Signed ..... (candidate) Date

## **ABSTRACT**

**Background** - Women who inherit the BRCA1/2 gene have up to an 85% lifetime chance of developing breast cancer and a 63% chance of developing ovarian Cancer. Since the publication and update of the UK Nice National Guidelines (2015) for women with a strong family history and Angelina Jolie, the famous American actress declared her status and her surgery, many more women are seeking testing in order to undergo bilateral risk-reducing mastectomy and oophorectomy. Very little is known about the longer-term effect on the woman her partner and her relatives following diagnosis and bilateral mastectomy from a qualitative research perspective.

**Aim** - The aim of this study, therefore, was to explore, interpret and develop an understanding of the experiences of women and their relatives living with the BRCA gene who elect to have bilateral risk-reducing mastectomy in an attempt to improve clinical practice by providing a new horizon of understanding.

**Methods** - An interpretive hermeneutic phenomenological study was conducted guided by the philosophy of Gadamer (2004) with its emphasis on a fused horizon of understanding with eight BRCA positive women, five of their husbands and five of their relatives. This study is a prospective study that took place in a large teaching hospital in Wales.

**Findings** - This study provides a new fused horizon of understanding of what it means to be a BRCA Positive woman and a relative living with the BRCA gene

who elect to undergo bilateral mastectomy as a risk-reducing modality. A hermeneutic text of interpretation exposed three main horizons (Gadamer, 2004). 'The price of survival', which includes the journey of the overall desire to survive, not just prevent cancer. 'The altered child's trajectory' which involves the transference of the fear of cancer onto (for) children and future generations and 'disembodied, a separation from self' which includes the effects of surgery on sexuality, femininity and identity. These horizons unite the experiences of the women and her relatives resulting in a new fused horizon of understanding, 'being disembodied'. These findings add knowledge and understanding for clinicians, researchers and policy makers working in the field of breast care with many implications for practice.

## **DEDICATION**

Dedicated to my dear mother, Suzette.

1944 - 2001

## **ACKNOWLEDGEMENTS**

When I set out to do this study over five years ago, I knew it would not be an easy task working full time with a family that depends on me. There have been many moments of happiness but also many moments of sadness having lost a number of relatives on the way. I could not have completed this very important and relevant piece of work however without the following acknowledgements.

Firstly, I would like to thank all the women (probands), their husbands, and the family members who took part and made this study possible. Without their honesty, complete trust in me and their willingness to talk about their very personal experiences, the findings from this study would not have been possible. A huge thank you.

I would also like to thank Professor Helen Sweetland and Professor Robert Mansel for their ongoing support throughout my nursing career, their belief in me and for their ongoing enthusiasm to make clinical practice always better for our patients.

A huge thank you to my supervisors, Dr Katie Featherstone and Professor Jane Hopkinson and, for their patience, their support, their encouragement and their specialist experience.

I would also like to thank the Florence Nightingale Foundation, The Band Trust, The Garfield Foundation and The Stephanie Williams Trust for three consecutive research scholarships that enabled me to continue with the study.

To my darling husband Charles and my beautiful daughter Charlotte for always carrying on without me and never once making me feel guilty. Thank you, Charles, for always supporting every challenge I have set myself in our thirty years together and there have been many!

To Karl Rowe in the IT Department who, despite thousands of calls, always responded and taught me so much on the computer. He never once made me feel that I was incompetent or a complete novice with computers when embarking on such an enormous piece of work. Thanks also to Dr Dave Clark, especially to Rosemary Williams for her constant help to put the whole thesis together at a very stressful time and to Mari Ann Hilliar for her superb librarian skills.

Finally, a huge thank you to all my friends who have seen me disappear early from functions without questioning me and to Dr Amie Hodges, my fellow Ph.D. student, for aiding my vice and supplying those skinny café lattes in Costa Coffee.

## CONTENTS

ABSTRACT.....	iii
DEDICATION.....	v
ACKNOWLEDGEMENTS .....	vi
CONTENTS .....	viii
OVERVIEW OF THE THESIS.....	xviii
CHAPTER ONE – Breast Cancer, Genetic Risk and BRCA Mutations 1 and 2. 1	
1.1 Introduction.....	1
1.1.1 Implications of the BRCA Mutation for Individuals and Their Families .....	2
1.1.2 The Decision of Testing and Disclosure on the Family Dynamics .....	3
1.1.3 Implications of the Decision to Elect to have Risk-Reducing Breast Surgery for the Woman.....	6
1.2 Access to Testing Policy, Practice and the Media .....	9
1.2.1 Policy and Practice .....	9
1.2.2 The Influence of Media .....	10
1.3 Overview: Breast Cancer, Mastectomy and Reconstruction.....	12
1.3.1 Treatment for Breast Cancer .....	12
1.3.2 Psychological Effects of Breast Cancer .....	13
1.3.3 Breast Cancer and BRCA Mutations.....	14
1.3.4 Treatment Decisions of Women with Breast Cancer and the BRCA Mutation.....	14
1.3.5 Breast Cancer, Mastectomy and Reconstruction.....	16
1.3.6 Partners and Reconstruction .....	17



1.4 The Function of the BRCA Gene .....	18
1.4.1 The Discovery and History of the BRCA Gene Mutation.....	19
1.4.2 The Risk of Breast Cancer with a BRCA1 and 2 Mutation .....	20
1.5 Genetic Risk, Screening and Counselling .....	22
1.5.1 Genetic Risk .....	22
1.5.2 Family Risk and Responsibility .....	25
1.5.3 BRCA Screening.....	27
1.5.4 Genetic Counselling.....	28
1.5.5 The Role of the Breast Nurse Specialist .....	29
1.6 The Impact of Testing and Disclosure .....	31
1.6.1 The Impact on Women.....	31
1.6.2 The Impact on Men .....	34
1.6.3 Family Disclosure .....	36
1.6.4 Impact on Younger Women .....	38
1.6.4 Impact on Adolescence.....	40
1.6.5 Impact on Partners and the Family .....	41
1.7 Prevention and Risk Management Options in the UK .....	42
1.7.1 Surveillance and Screening .....	43
1.7.2 Chemoprevention .....	44
1.8 Surgery: Risk-Reducing Mastectomy .....	45
1.8.1 The Psychological Effects of Risk-Reducing Mastectomy .....	48
1.8.2 The Decision to Elect to have Breast Reconstruction .....	49
1.8.3 Surgery Versus Screening Programmes.....	51
1.9 Summary .....	52

CHAPTER TWO – The literature review (The Current Horizon of Understanding, Gadamer, 2004).....	54
2.1 Introduction.....	54
2.2 Systematic Search Strategy .....	55
2.2.1 Preliminary Search January 2011 until April 2011 .....	55
2.2.2 Inclusion Criteria .....	57
2.2.3 Exclusion Criteria.....	57
2.2.4 Expanded Search .....	58
2.2.5 Final Search.....	58
2.4 Risk and Embodiment .....	59
2.4.1 The Significance of the Breasts .....	65
2.4.2 A Symbol of Femininity - The Social Significance of the Breast.....	66
2.5 Pre-Surgery – Decision Making.....	70
2.5.1 The Timing of Surgery .....	73
2.5.2 The Decision to Elect Breast Reconstruction Following RRM.....	75
2.6 Post-surgical Impact.....	76
2.6.1 Regret Following Surgery .....	76
2.6.2 The Impact on Body Image.....	79
2.6.3 The Longer-Term Impact on Body Image .....	85
2.6.4 The Impact on Sexuality .....	90
2.6.5 The Experience of Partners Following Risk-Reducing Mastectomy.	96
2.6.6 Summary of Literature on Body Image and Sexuality.....	99
2.8 Long-Term Impacts of Risk-Reducing Mastectomy.....	100
2.8.1 Quality of Life.....	100

2.8.2 A Summary of the Review .....	104
2.8.3 Conclusion .....	107
CHAPTER THREE – Methodology and Methods.....	109
3.1 Introduction.....	109
3.2 My Starting Point - The Emergence of the Research Question from Practice .....	110
3.2.1 Situating the Researcher .....	112
3.2.2 Situating the Others .....	115
3.2.3 My Academic Role .....	116
3.2.4 The Research Question and Aim .....	118
3.3 Hermeneutic Philosophy.....	118
3.3.1 The Philosophical Beliefs Guiding the Research .....	121
3.3.2 Ontological Position .....	121
3.3.3 Epistemological Position .....	124
3.4 Methodology and Theoretical Approach.....	126
3.4.1 Phenomenology .....	126
3.4.2 Interpretive Phenomenology .....	129
3.4.3 The Work of Hans-George Gadamer (1900 - 2002) .....	132
3.4.4 Philosophical Hermeneutics.....	134
3.5 Methods.....	137
3.5.1 A Qualitative Approach .....	137
3.5.2 In-Depth Interviews .....	139
3.5.3 Design.....	140
3.5.4 The Interview Approach: Hermeneutic Interviews.....	141

3.5.5 Reflexivity and my Role in the Interview .....	144
3.5.6 The Relationship Between the Researcher, The Study and The Participants - My Clinical Role .....	145
3.5.7 Advantages of a Dual Role .....	146
3.5.8 Planning.....	148
3.5.9 The Potential Power Imbalance in the Interview .....	149
3.6 Setting, Sampling, Recruitment and Participants .....	150
3.6.1 The Setting .....	150
3.6.2 Sampling.....	150
3.6.3 Sampling Decisions .....	151
3.6.4 Recruitment .....	153
3.6.5 The Study Participants.....	154
3.6.6 Ethical Considerations and Approval .....	155
3.7 Analysis and Interpretation .....	157
3.7.1 The Act of Dialogue .....	159
3.7.2 The Hermeneutic Circle .....	159
3.7.3 The Fusion of Horizons.....	160
3.8 Phases of Analysis .....	162
3.8.1 Phase One.....	162
3.8.2 Phase Two.....	164
3.8.3 Phase Three .....	165
3.8.4 Phase Four .....	166
3.9 Rigour in the Study .....	166
3.9.1 Balanced Integration .....	169

3.9.2 Openness .....	170
3.9.3 Concreteness.....	171
3.9.4 Resonance.....	172
3.9.5 Actualisation .....	172
3.9.6 A Flowchart of the Summary of the Research Process.....	174
3.9.7 Chapter Summary.....	175
CHAPTER FOUR – The Findings.....	176
4.1 Introduction to the Findings Chapters.....	176
4.2 The Price of Survival - Pre-surgical Experiences .....	177
4.2.1 Getting to Grips with the Possibility of a BRCA Mutation .....	178
4.2.2 Conflict Within the Family About How to Respond.....	182
4.2.3 Responsibilities, Obligations and Needing to Know .....	183
4.2.4 The Price: Divisions Within the Family.....	185
4.2.5 The Price: The Unexpected Impact and Implications of the Result	189
4.2.6 The Price: Living with and Making Sense of Being BRCA Positive	190
4.2.7 The Price: Disposing of Dangerous Breasts .....	195
4.3 The Price - No Choice, Surgery is the Only Option .....	196
4.3.1 The Price: A Visibly Altered Body .....	198
4.3.2 The Price: No Longer My Whole Self.....	200
4.3.3 The Price: Being without Nipples .....	203
4.3.4 The Price: Loss of Intimacy.....	208
4.3.5 The Price: Living with the Consequences of their Decision .....	210
4.3.6 The Price: Marital Conflict.....	212
4.3.7 The Price: The Fight for Surgery.....	213

4.4 The Altered Child's Trajectory .....	215
4.4.1 The Kids Sake - Cancer Free Worry .....	216
4.4.2 Preserving the Childs Trajectory - Maintaining a Normal Childhood.....	219
4.4.3 Hope for Future Developments in Medicine .....	222
4.5 A Separation from Self - Post Surgical Experiences .....	223
4.5.1 Separation - The Fear Has Gone.....	224
4.5.2 Separation - No Return to Normality .....	225
4.5.3 Separation - Regret and Loss of a Former Self .....	228
4.5.4 Separation - No Longer a Feminine Body .....	229
4.5.5 Separation from Self - Disembodied .....	232
4.5.6 Separation from The Intimacy .....	234
4.5.7 Separation - Nipples to Preserve and Restore Femininity .....	238
4.5.8 Separation - Still Not Me! .....	240
4.5.9 Help to Restore Self .....	242
4.6 Conclusion of Probands Findings.....	247
CHAPTER FIVE – The Husbands' Experiences .....	248
5.1 Introduction.....	248
5.2 The Price of Survival - Pre-Surgical Experiences.....	249
5.2.1 The Price - Finding Out About a Gene in the Family .....	249
5.2.2 The Price – Having the Test and Understanding the Implications .	252
5.2.3 The Price - The Meaning of Being BRCA Positive .....	255
5.2.4 The Price - Strategies for Getting Through the Day .....	260
5.3 The Price - Losing the Breasts .....	260

5.3.1 The Price - Physical and Emotional Impacts of the Decision .....	262
5.3.2 The Price - Undergoing Breast Reconstruction.....	264
5.3.3 The Price - Uncertainties about Changes to the Body .....	266
5.3.4 The Price - Turning the Family Upside Down .....	268
5.4 A Separation from Themselves - Post-Surgical Experience .....	270
5.5.1 Separation - The Fear Has Gone.....	270
5.5.2 Separation - Not Who They Were? .....	271
5.5.3 Separation - No Return to Normal.....	274
5.5.4 Separated - Not in Unity.....	275
5.5.5 Separation from her Self .....	278
5.5.6 Sex Talk - A Need to Reunite .....	279
5.6 The Altered Child's Trajectory .....	280
5.6.1 Maintaining A Normal Child Trajectory.....	280
5.6.2 Introducing Fear into the Children's Lives? .....	281
5.7 Summary .....	284
CHAPTER SIX – The Family Members .....	286
6.1 Introduction.....	286
6.2 The Price of Survival .....	287
6.2.1 The Price - Being on The Alert.....	287
6.2.2 The Price - It's A Female Thing .....	289
6.2.3 The Price - Having a Test and Being BRCA Positive.....	293
6.2.3 The Price - Losing the Breast .....	294
6.3 The Price - A Separation from Themselves.....	295
6.3.1 The Price - Separation from The Marriage.....	298

6.3.2 The Price - Feeling the Guilt .....	299
6.4 The Altered Child’s Trajectory .....	301
6.4.1 Preserving the Normal Child Trajectory .....	304
6.5 Summary of Experiences .....	306
CHAPTER SEVEN – Discussion and the Expanded Horizon of Understanding .....	308
7.1 Research Question Revisited .....	309
7.2 The Price of Survival .....	311
7.3 The Altered Child’s Trajectory .....	320
7.4 A Separation from Self - ‘Disembodied’ .....	323
7.5 Strengths and limitations of this Study .....	334
7.6 Concluding Statement .....	336
7.7 Recommendations and Implications for Practice.....	337
REFERENCES .....	339
APPENDIX ONE - A Short Autobiography .....	384
APPENDIX TWO - NICE (2013) Guidelines .....	388
APPENDIX THREE - Literature Review Search Strategy .....	397
APPENDIX FOUR - Publication and Dissemination Plan .....	406
APPENDIX FIVE - National and International Presentations .....	409
APPENDIX SIX – Reflections and Diary .....	411
APPENDIX SEVEN - MINDMAPS .....	415
APPENDIX EIGHT – NHS R&D Approval.....	433
APPENDIX NINE – REC Approval.....	435
APPENDIX TEN – REC Provisional Approval .....	438



APPENDIX ELEVEN – Potential Interview Topics .....	442
APPENDIX TWELVE - Patient Consent Form Version 2 .....	444
APPENDIX THIRTEEN - Patient Information Sheet .....	445
APPENDIX FOURTEEN - Patient Information Sheet Final .....	449
APPENDIX FIVETEEN – Partner Consent Form Version 2 .....	453
APPENDIX SIXTEEN – FN Award Letter .....	454
APPENDIX SEVENTEEN – FN Award Letter 2013/14 .....	457
APPENDIX EIGHTEEN – FN Award Letter 2012/13.....	460

## **OVERVIEW OF THE THESIS**

This section provides a summary of the seven chapters within this thesis.

### **Chapter One – Setting the Scene and Laying the Foundations**

This chapter sets the context of the study and provides the background information in relation to the prevention of breast cancer and the decision to elect to have bilateral risk-reducing mastectomy. It includes information on the BRCA gene mutations 1 and 2, the risk, the family, health care providers, decision-making including breast reconstruction. In addition, a short autobiography of the researcher and the motivation for the study is offered in Appendix One. The terms BRCA mutation, gene mutation, mutation positive or mutation carriers are used interchangeably.

### **Chapter Two - The Current Horizon of Understanding (Gadamer, 2004)**

This chapter provides an overview of the current literature reviewed, what Gadamer (2004) calls, ‘the current horizon of understanding’<sup>1</sup>. Thus the pre-understandings and prejudices formed from that literature, are highlighted and my research questions emerged.

### **Chapter Three – Methodology and Methods**

This chapter presents my epistemological and ontological position, my methodology and methods of conducting the research including the decision and

---

<sup>1</sup> Knowledge attained from the literature review and before this study was conducted.

advantages of utilising a qualitative approach; this is reflected upon throughout the study. Examples of my reflections are provided in Appendix 6.

#### **Chapter Four - The Probands' Experiences**

This chapter presents the analysis and findings of the text generated from the eight probands<sup>2</sup> who were interviewed pre-surgery, at six and twelve months post-surgery and a subsample of four probands at eighteen months. It provides the three horizons<sup>3</sup> of understanding that developed from the data and which unite the experiences of the women, their husbands and their family.

#### **Chapter Five - The Husbands' Experiences**

This chapter presents the analysis and findings of the text generated from the five husbands who were interviewed pre-surgery and at twelve months post-surgery. Three horizons of understanding developed from the data and are exposed.

#### **Chapter Six - The Family Members**

This chapter presents the analysis and findings of the texts generated from the five family members (one mother, two fathers, one sister and one brother) which contributes to understanding the phenomena of living with the BRCA gene and the surgery from the family's perspective. Thus, three horizons of understanding

---

<sup>2</sup> Women tested and who have the BRCA mutation.

<sup>3</sup> A 'horizon' is the range of vision that includes everything that can be seen from a particular vantage point.

are exposed from interpreting the parts, in this study (husbands and family) and the whole (probands) and which unite the experiences.

### **Chapter Seven - The Expanded Horizon of Understanding**

This chapter revisits the pre-understandings and prejudices<sup>4</sup> highlighted at the start of the study and puts forward an expanded horizon of understanding as a result of engaging in the hermeneutic experience (gaining a deep understanding of what it means to be a woman with a BRCA mutation who elects to remove both of her breasts), by viewing the parts (the husbands' and the families' experiences) in terms of the whole (the probands' experiences) and engaging in the hermeneutic circle<sup>5</sup> of understanding. A discussion of the findings and conclusion are offered together with the implications for practice.

---

<sup>4</sup> Prejudice refers to the productive pre-understandings that enlighten future understanding of the phenomenon, gleaned from experience and the current literature.

<sup>5</sup> The hermeneutic circle is the back and forth movement between partial understanding and the more complete whole.

# **CHAPTER ONE – Breast Cancer, Genetic Risk and BRCA Mutations 1 and 2**

This chapter provides the background and context of the study: the BRCA mutation 1 and 2 and the decision to elect to have preventative breast surgery. It provides information about the genetics of breast cancer risk for individuals and the implications for their wider family. It presents an overview of the many factors involved in the prevention of breast cancer and the decision to elect risk-reducing mastectomy (RRM) including the role of health care professionals, laying down the foundations for the chapters that follow. To enable greater understanding of RRM as a preventative intervention and to put the study into context, breast cancer and its treatments are also considered.

## **1.1 Introduction**

The focus of this study is on understanding the experiences of women who have inherited a mutation in either of the two breast cancer susceptibility gene mutations (BRCA1 and BRCA2) and who have elected to have bilateral risk-reducing mastectomy (RRM), which involves the complete removal of both breasts, and the impact on their families. This is the first qualitative study to my knowledge that focusses specifically on BRCA positive women and the decision to have risk-reducing mastectomy that includes exploring the experiences of their partners and family members. The desire to conduct the study was born of nursing practice and a lifelong career caring for individuals

with breast cancer and their families, witnessing the devastation of diagnosis, treatment and death. It also resulted from seeing an increasing number of young women attending services and deciding to undertake bilateral risk-reducing mastectomy with no studies to date detailing the specific experiences and informational needs of younger women undergoing RRM (Glasse et al., 2016). Yet currently, breast care teams do not fully understand the effects of this surgery on these women or the wider impacts on their relationships with their family, especially their partners. The overall goal of this study is to inform a clinical service that meets the needs of BRCA positive women and their families by understanding their experience.

### **1.1.1 Implications of the BRCA Mutation for Individuals and Their Families**

Although women with the BRCA mutation do not have cancer, they have up to an 85% lifetime risk of developing breast cancer and up to a 60% lifetime risk of developing ovarian cancer (Barcenas et al., 2006; Chen and Parmigiani, 2007). The identification of a mutation means that they have to face many life changing, difficult decisions, including the decision to reduce this risk by removing their healthy breasts and ovaries, which carries benefits but also risks (Howard et al., 2009). Although accepting cultural differences in the perception of body image and decision making, many women in the western world are provided with the option to consider surgery to remove both of their breasts and believe it is the only option to prevent cancer and the debilitating

treatment that it involves (Metcalfe et al., 2005b; Brandberg et al., 2008; Lostumbo et al., 2010). In the systematic review by Howard et al., (2009), forty-three published articles were reviewed identifying three main factors that influence high risk women's decisions about surgery. These include medical factors, for example, being BRCA positive, psychological factors including the increased anxiety following the death of relatives to breast cancer and social factors which involve their perceived obligations to the family, all of which cannot be viewed as independent or in isolation. How these factors affect women and their family in their everyday lives and how they influence decisions about genetic testing and preventative treatment, however, is less understood.

### **1.1.2 The Decision of Testing and Disclosure on the Family Dynamics**

Understanding of the inheritance of a BRCA mutation and the implications of testing is highly complex and multifaceted for women and their families. Katapodi et al., (2013) found that both individual and family characteristics influence the decision to be tested and that testing was positively associated with 'family hardiness', that is, a family's cohesion and ability to overcome adverse events. However, many individuals still find it particularly difficult knowing when and how to disclose information about testing and risk-reducing surgery to the family, and children in particular, (Kenen et al., 2006; d'Agincourt-Canning, 2006a) due to the emotional impact for themselves and their children.

Rowland et al., (2016) in their qualitative study with eleven families that included thirteen children and young adults aged between ten and twenty-one, found that the majority of mothers disclosed their own risk but avoided disclosure of their children's risk, disclosing even less to their sons. Importantly, many of the women who had undergone surgery avoided disclosing any information about the negative psycho-social effects because they tried to protect their children from the impacts of surgery. However, this withholding and downplaying of information inadvertently made it more difficult for their children to have an awareness of the risks and the implications of having a BRCA mutation and surgery. As a consequence, many of these young adults had a blasé attitude towards the surgery with reconstruction and often failing to differentiate this from cosmetic augmentation, additionally preventing them from fully understanding the implications and the risks of major surgery (Rowland et al., 2016).

Thus, in addition to coming to terms with the inheritance of a gene mutation and the implications of risk-reducing surgery, women also have to consider the wider consequences, which involve and affect the whole family. A qualitative study examining the impact of BRCA testing on family dynamics and relationships (eleven women and one male from eight different families with the mutation), demonstrated emotional consequences for both the individual tested (the index case) and their relatives, impacting negatively on the family dynamics and their relationships. Discussions within the family also changed and some members of the family were found to become less connected to



other family members and reported that they felt burdened to disclose their results within their family (Douglas et al., 2009).

Crotser and Dickenson (2010) in their qualitative hermeneutic study of nineteen women from seventeen nuclear families, gained an understanding of the experiences of younger women with the BRCA mutation (n=12) and the communication within their families. Receiving and responding to the BRCA result was found to have more of an effect than expected, evoking a paradox of both reported fear and empowerment for these young women. This caused an additional ripple effect throughout their families. Relationships changed as a result of their limited communication about the BRCA result and a need to protect family members, resulting in a change to the sense of connectedness to family members, with some young women seeking support and help outside of the family. The reported emotions amongst family members who receive news of a mutation also included sadness, guilt, resentment, anger and blame (Bradbury et al., 2007).

How these factors affect women and their families in their everyday lives and how they influence decisions about genetic testing and preventative treatment, however, is less understood. Thus, it is important for health care professionals working with BRCA individuals and families who are seeking testing and surgery to be aware of the implications of this process for the wider family dynamics and the difficulties they may have already experienced in their pathway to the clinic and surgery. In order to fully understand and support

women in the context of their family, a deeper exploration of these experiences and impacts using qualitative research perspectives can aid understanding and benefit clinical practice.

### **1.1.3 Implications of the Decision to Elect to have Risk-Reducing Breast Surgery for the Woman**

Bilateral mastectomy as preventative risk-reducing surgery is not without consequences and has been associated with physical complications, psychological problems, decreased quality of life (Gahm et al., 2010; Hallowell et al., 2012; den Heijer et al., 2012; Gopie et al., 2013) and in some cases, regret (Borgen et al., 1998; Frost et al., 2000; Payne et al., 2000). Indeed, Bresser et al., (2006) refer to this type of surgery as very radical. Furthermore, decisions about surgery have often been found to reflect an emotional heuristic approach, that is, a reflection of a woman's own fear, worry and perceived control rather than a perceived rational weighing up of both risk and benefits (Kenen et al., 2003a; Beesley et al., 2013; Heiniger, et al., 2015).

A great deal of uncertainty and stress has been associated with the decision about risk-reducing surgery including what surgery to undergo and when. A qualitative study carrying out in-depth interviews with thirty-two women (of which eight were BRCA positive) to determine their experience of making such decisions, identified that these women initially turned to their doctors to address their uncertainties, reporting that some providers were too directive

whilst others too non-directive. Women were left feeling very disappointed and turned to family, friends and the wider breast cancer community for direction (Klitzman and Chung 2009). These decisions are often already made by women by the time they and their families attend the surgical appointment. However, an understanding of how women come to their decision is necessary in order to identify their expectations, their informational and support needs.

McQuirter et al., (2010) in their qualitative study of ten BRCA positive Canadian women found that the decision to proceed with surgery was an emotional and intuitive process that involved interpersonal factors, contextual factors and informed by relationships rather than a 'rational' weighing up of the pros and cons of surgery. A literature review by Glassey et al., (2016) also found that women are not well informed prior to making a decision about RRM identifying that younger women needed more information and support. Overall, fear and worry strongly influence decision making (Beesley et al., 2013; Heiniger et al., 2015).

However, health care professional's especially surgical teams, often underestimate the impact of the test result or how women and families came to the decision of surgery. They also underestimate the consequences and experience of such surgery for women and families with a BRCA mutation because they do not have cancer (Rolnick et al., 2007; DiMillo et al., 2013). Rolnick et al., (2007) surveyed nine hundred and sixty-seven women who had a prophylactic mastectomy and found that of the 80% who had bilateral

mastectomy, two thirds wanted more information and to be better prepared for the physical changes experienced post-surgery. Moreover, in a qualitative study by Patenaude et al., (2008), that explored the psychological consultation with women who were considering RRM, more than 50% of the one hundred and eight women felt that psychological consultation pre and post-surgery was required to aid decision making and prepare them for surgery. Despite this finding, formal clinical psychological services for women and their families with a BRCA mutation vary considerably in the UK and is not standard practice in many areas (Glasse et al., 2016). In Wales, for example, the cancer genetic clinic may be on a different hospital site to the surgical clinics and there is no formal clinical psychological consultation before surgery.

Furthermore, differences in service provision from breast care nurse specialists exist in the UK, for example, in many breast care services in the UK and in many parts of Wales, there is no routine pre and post-operative care from a specialist nurse dedicated to these patients or their families. This gap in service provision is because they do not have cancer, their risk has been removed by surgery and their physical care and psychological support needs have yet to be fully identified. Thus, the precise physical, psychological and informational needs from testing through to surgery and beyond from a clinical perspective for the woman and her family is less understood. Indeed, only one study, with seven participants, has included Welsh women with a BRCA gene, these were aged eighteen to thirty, and the study focussed on genetic testing and not the experiences of surgery (Brunstrom et al., 2016).

This first section introduced the study, the start of the journey from genetic testing to decisions about risk-reducing mastectomy, demonstrating that this process involves not only individuals but also their wider families. It introduces the justification for the need to conduct this study and the importance of clinical practice. The remainder of Chapter One provides background information that puts the study into context.

## **1.2 Access to Testing Policy, Practice and the Media**

### **1.2.1 Policy and Practice**

The updated family history guidelines by the National Institute for Health and Care Excellence (NICE, 2013) has widened the eligibility criteria for testing meaning that many more women have been able to request genetic testing for a BRCA mutation. These guidelines have lowered the threshold for testing from a greater than 20% risk to now include individuals assessed as having at least a 10% risk of carrying a mutation, making more women eligible to be tested. (NICE, 2013). The criteria for testing in the UK can be seen in Appendix Two.

These guidelines also require that discussions about risk-reducing surgery should be more implicit throughout the process of assessment and testing. Although surgery is not a recommendation, it is presented as an option for risk management for women with a BRCA mutation that should be discussed (See Appendix Two).

Importantly, this expanded eligibility criteria has prompted a dramatic increase in referrals for both genetic testing and risk-reducing surgery. In the UK for example, there has been a two-and-a-half-fold increase in referrals and the demand for testing has doubled (Skytte et al., 2010; Evans et al., 2015), Indeed in Wales, the number of tests being performed has quadrupled. Moreover, in 2009, there were one thousand one hundred and sixty three women having RRM in the UK (Neuberger et al., 2013) and numbers are now estimated to be much higher. Within the UK breast centre where this study took place, since 2013 there has been a threefold increase in the number of referrals or surgery and without any associated increase in resources for nursing care and psychological support for this group.

### **1.2.2 The Influence of Media**

In addition, wider knowledge of the BRCA genes and availability of testing via the media has also resulted in many more women with a family history of breast or ovarian cancer attending cancer genetic clinics seeking screening and preventative surgery (Parks, 2014; Sherkow and Scott; 2014; Evans et al., 2015). Media coverage of Angelina Jolie, the American actress, and her decision to disclose her BRCA mutation and surgery (New York Times, May 2013) is just one example. The role that the media play in both normalising and influencing risk-reducing mastectomy decisions should not be under-estimated. As with the Angelina Jolie story, Crabb and Le Couteur

(2006) utilise discursive analysis to demonstrate how a popular magazine account portrays one woman's story of the removal of her healthy breasts as morally acceptable. 'Fiona Farewells her breasts' situates Fiona as a responsible self-sacrificing mother who is removing her asymptomatic breasts in order to survive, intimating that if she does not do it, then she will die. She is presented as both a rational and reasonable individual.

These portrayals contrast to some of the earlier suggestions that women's decision to have risk-reducing surgery is influenced by an over-estimation of risk or intense anxiety and viewed by professionals and family, as extreme behaviour (Meiser et al., 2000). The fact that woman and their families are now able to accurately determine the probability that they will contract breast or ovarian cancer with the diagnosis of a mutation in either the BRCA1 or 2 genes also makes it a more acceptable decision and less controversial.

To put the study into context and prior to examining what is already known about the experience of a BRCA mutation and RRM, it is pertinent to give a brief overview of the wider literature examining the experiences of women with a diagnosis of breast cancer who have a mastectomy and reconstructive surgery to identify any similarities, but also to examine and understand why women seek preventative surgical interventions.

### **1.3 Overview: Breast Cancer, Mastectomy and Reconstruction**

In the United Kingdom (UK) there are approximately fifty-four thousand new cases of cancer a year and one hundred and fifty cases a day, of women diagnosed with breast cancer. It is the most common cancer occurring in females accounting for 31% of all new female cancers. One in eight women will be diagnosed with breast cancer during their lifetime and although rare in men, approximately four hundred men are diagnosed each year in the UK. Even though mortality rates have fallen by 40% since 1989, breast cancer remains the second leading cause of cancer deaths in women (Cancer Research UK (CRUK), 2013).

#### **1.3.1 Treatment for Breast Cancer**

The treatments for breast cancer can be lengthy and debilitating, consisting of surgery and adjuvant therapies such as chemotherapy, radiotherapy, hormone treatments or any combination that can take up to a year to be completed. Both surgery and adjuvant therapy can have immediate and long-term negative effects on an individual, for example depression, anxiety, body image problems, especially from breast and hair loss and weight gain, all associated with psychological problems (King et al., 2000; Avis et al., 2005; Mols et al., 2005; Kayl et al., 2006; Mehnert and Koch, 2008; Lemieux et al., 2008) and a reduced quality of life (Crompvoets, 2003; Ohsumi et al., 2007; McLachlan, 2009; Karaoz et al., 2010; Adams et al., 2011; Rosenberg et al., 2013).



### **1.3.2 Psychological Effects of Breast Cancer**

Cancer related distress has been identified as especially high following breast cancer diagnosis (Voorwinden and Jaspers, 2014) and associated with reports of anxiety, depression and adjustment (Walker et al., 2014). It has been estimated that up to 38% of these women suffer from anxiety and depression (Vahdaninia et al., 2010) with up to 50% experiencing sexual problems (Fobair et al., 2006). Breast cancer treatments can impose unwanted changes to the body following mastectomy. This can distance a woman from her body even with reconstruction, challenging femininity and identity (Rosenblatt, 2006; Everson, 2009; Piot-Ziegler et al., 2010). Furthermore, although breast conservation is an option for women diagnosed with breast cancer, it offers a modest benefit when compared to mastectomy, of experiencing cancer related fears and changes in body image. Body transformation from the scars of both surgeries and the effects of chemotherapy, constantly remind women of the impact of the treatment on their lives (Thomas-Maclean, 2005). Women with breast cancer can often access psychological counselling, either provided by cancer charities or from a referral to a trained counsellor in a General Practitioner (GP) practice. In contrast, women electing to have RRM, find it difficult to access such services because they do not have cancer.

It is widely acknowledged that mastectomy following a breast cancer diagnosis is associated with considerable psychological morbidity resulting specifically in dramatic alterations in body image (Petronis et al., 2003; van Oostrum et al., 2003; Marshall and Kiemle, 2005; Hawkins et al., 2009; Manganiello et al.,

2011; Rosenberg, et al., 2013). Studies have identified that women with a BRCA mutation are aware of this morbidity and feel a responsibility to act on their opportunity to prevent cancer from occurring (Howard et al., 2009; McQuirter et al., 2010).

### **1.3.3 Breast Cancer and BRCA Mutations**

Importantly, breast cancers that develop in women with the BRCA1 mutation can be more aggressive, develop at a younger age when compared to those with BRCA2 (Skytte et al., 2011), and can be triple negative cancers. Triple negative cancers do not have receptors for the hormones oestrogen or, progesterone or for the HER2 protein, which means that women have limited options of adjuvant treatment (hormonal therapy and Herceptin) post-surgery that will help to prevent cancer from recurring (CRUK, 2013). Between 10% and 30% of cancers in women with a BRCA1 mutation will be triple negative and increasingly, in clinical practice, more BRCA mutations are being identified in women who present with breast cancer and who, because of examining tumour pathology, are found to have a triple negative cancer (Vargas et al., 2010).

### **1.3.4 Treatment Decisions of Women with Breast Cancer and the BRCA Mutation**

Many of the women found to have triple negative breast cancers request genetic testing for a BRCA mutation and if a mutation is identified, choose to

have bilateral mastectomy (Jeffers et al., 2014). It is also known that younger women diagnosed with breast cancer choose the most aggressive treatment in order to reduce their risk of further disease (Finch et al., 2013). Women with breast cancer have different motivations and expectations to healthy at-risk women when it comes to testing and surgery. Affected women, for example, want to identify why they developed cancer thus trying to prevent family member's from developing it, whereas healthy at-risk women prioritise assessing and understanding their own risk (Iredale et al., 2003). In addition, those women with breast cancer and a BRCA mutation who choose RRM, do not report statistically significant reductions in their cancer worry following surgery compared to BRCA women unaffected by cancer (Brandberg et al., 2004; Borreani et al., 2014). Nevertheless, those diagnosed with breast cancer with an additional BRCA mutation are increasingly choosing to have bilateral risk-reducing mastectomy.

In a grounded theory study by Jeffers et al., (2014), women with breast cancer who also had a BRCA mutation, chose to remove their breasts and their ovaries in order to 'maximise survival'. The process of maximising survival was found to begin even earlier than the genetic test, during their cancer journey, with a focus on survival for the sake of themselves but importantly, for their family. These women's focus moved from living with a cancer diagnosis to living with an inherited disease which demonstrates the significant psychosocial impact of living with both genetic inherited disease and cancer (Jeffers et al., 2014). Kwong and Chu (2012) also found in their qualitative study of twelve Chinese

women with breast cancer (of which eleven were BRCA positive) and who chose to have the other breast removed in response to this genetic information, a quarter were found to experience regret and had psychological problems up to three years post-surgery, despite breast reconstruction.

### **1.3.5 Breast Cancer, Mastectomy and Reconstruction**

Breast reconstruction following surgery can have both positive and negative effects on quality of life and cosmetic satisfaction (Brandberg et al., 2000; Malata et al., 2000; Nissen et al., 2001; Harcourt et al., 2003; Elder et al., 2005; Bresser et al., 2006; Isern et al., 2008). Within some studies, significant dissatisfaction and regret have been identified (National Mastectomy and Reconstruction Audit, 2011, Harcourt and Rumsey, 2004; Sandham and Harcourt, 2007; Sheehan et al., 2007).

Reconstruction can never replace a woman's breast and women's expectations are often unclear, with decisions based on individual preferences (Denford et al., 2011), and influenced by a woman's concern for her husband. Piot-Ziegler et al., (2010) conducted a qualitative study using semi-structured interviews with nineteen women who had mastectomy following breast cancer and nine had been offered reconstruction. Many reported that they had reconstruction for their husband's sake. The authors found that despite reconstruction, mastectomy led to a body that was deconstructed, which challenged the women's gendered identity causing feelings of

embarrassment, loss, mutilation and dissonance from her former integral body.

It is not unsurprising, therefore, that most young women with breast cancer decide not to reconstruct their breast. However, evidence also suggests that those women with breast cancer who do not have reconstruction following mastectomy, report a negative impact on their relationships and on psychological wellbeing. Within one study over 30% of women feel that their partners perceive them as less attractive and over 80% covered up their bodies during intimacy (Andrzejczak et al., 2013). Importantly, it has been suggested that there may be an over reliance on using surgical reconstruction in an attempt to solve the bodily loss caused by this surgery (Holland et al., 2014). Indeed Fang et al., (2013) and Harcourt et al., (2003) advise that reconstruction should not be seen as a panacea for the distress caused by mastectomy.

### **1.3.6 Partners and Reconstruction**

The literature examining the experiences of partners or family members following mastectomy and reconstruction is particularly limited and has focused on the experiences of having a family member diagnosed with breast cancer, not those who are living with risk. However, these studies have demonstrated negative outcomes for partners especially with regard to their relationship, sexuality, and body image adjustment (Sandham and Harcourt,

2007; Rowland and Metcalfe, 2014). They report mourning the loss of their partners' breast with experiences of distress, distaste and avoidance of intimacy. The systematic review by Rowland and Metcalfe (2014), reported that men found it particularly challenging talking to their partner about alterations to body physique and image and requested more information and support from professionals to prepare them personally for the change but also to help them to support their partners. In response, the inclusion of partners in this study will explore some of these issues and examine the experience of breast loss for partners in the context of risk reduction.

#### **1.4 The Function of the BRCA Gene**

Cancers develop as the result of a disruption in the normal functioning of genes and in the majority of cases, they occur by chance, from exposure to environmental factors or problems in the replication of DeoxyriboNucleic Acid (DNA). DNA is a self-replicating material present in all living cells and a major component of chromosomes (National Institute for Health (NIH), 2016). The two main breast cancer genes BRCA1 and BRCA2 produce tumour suppressor proteins and these have been located on chromosomes 17 and 13, respectively. These proteins help repair damaged DNA thus ensuring that the cells genetic material is stable.

When either of the genes is mutated, or there is an alteration, it results in the protein being unable to function properly. DNA damage is not repaired

properly; as a result, cells develop additional genetic alterations that can lead to cancer (NCI, 2015). In this case, there is the uncontrolled growth of breast and ovarian tissue because the tissue does not respond to the body's normal signals to stop growing.

#### **1.4.1 The Discovery and History of the BRCA Gene Mutation**

BRCA1 and BRCA2 are the most common genes that put an individual at the greatest risk of breast and ovarian cancer (CRUK, 2013). The region of the genome that became known as BRCA1 was discovered, in 1990, on chromosome 17 in a laboratory in Berkeley, USA by Mary-Claire King, Professor of Genome Sciences and her colleagues, at the University of California. Four years later, the gene was cloned at the University of Utah and by Myriad Genetics. In 1995, the BRCA2 gene was also discovered and cloned by Myriad Genetics, a company in Salt Lake City who patented the genes in 1998 and was the only company to exclusively offer a genetic test from the late 1990's until 2013, when the 'US Supreme Court revoked' the patent (NCI, 2015).

The discovery of both genes has made genetic testing possible for women and their families. Genetic testing refers to the examination of an individual's DNA, focusing on one or more specific genes and testing for BRCA gene mutations became available in the UK in 1996 (Bancroft et al., 2010). Since the NICE (2013) guidelines were implemented in Wales in 2015, those with a personal

history of breast cancer (affected) and testing of unaffected individuals with a strong family history of a greater than 10% chance of having a mutation, has been possible. Women can also fund the test themselves if they do not meet the criteria for testing within the National Health Service (NHS). The costs vary considerably, but in Wales, private testing costs approximately five hundred pounds. In some parts of the UK, women and their families are self-funding private genetic testing, especially where their family history is not available for example in cases of adoption or if there are estranged relationships within families or where communication problems exist. Women diagnosed with breast cancer within a family with high risk can initiate a test if they are also assessed as having a 10% or greater risk of having a mutation; this is called a diagnostic test and takes forty days for the results (UK Genetic Testing Network (UKGTN), 2016). This, in turn, has increased the case load of multidisciplinary breast teams especially breast care specialist nurses. Cancer genetics centres in the UK offer predictive testing to those unaffected by cancer over the age of 18 if there has been an identified BRCA mutation in a family member. A blood test is performed and results are expected in twenty days (NICE, 2013)

#### **1.4.2 The Risk of Breast Cancer with a BRCA1 and 2 Mutation**

Hereditary breast cancer accounts for approximately 5-10% of all breast cancers and BRCA1 and BRCA2 mutations account for 20-30% of hereditary breast cancer in families (Ford, 1998; NCI, 2015). These cancers usually occur below the age of fifty (Rogozinska-Szczepka et al., 2004) and put the affected



person at risk of other cancers, particularly ovarian cancer, accounting for approximately 15% of ovarian cancers overall (NCI, 2015).

Although women with a family history of breast and ovarian cancer generally overestimate their risk (Meiser et al., 2000; Kenen et al., 2003; Braithwaite et al., 2005) with younger women found to have higher cancer related distress (Vodermaier et al., 2010; Patenaude 2013;), the risk of developing breast cancer is significantly higher for women with a BRCA mutation. Women who have inherited a BRCA1 or BRCA2 mutation have an approximate lifetime risk<sup>6</sup> of 60-85% of developing breast cancer and a 40-60% chance of developing ovarian cancer compared with a 12% risk of breast cancer in the normal population (Barcenas et al., 2006; Chen and Parmigiani, 2007).

These cancers develop pre-menopausally and women have up to a 64% chance of developing bilateral breast cancer (Evans, et al., 2009). In comparison, men with a BRCA mutation have up to a 16% risk of prostate cancer and approximately an 8% risk of breast cancer (Evans et al., 2009). Prostate cancers that develop in men with a BRCA mutation have poorer survival rates than those who test negative because the cancers are more aggressive and develop faster (Narod et al., 2008). This is important in the context of this study because there were three men in the sample who tested

---

<sup>6</sup> Risk calculated up to the age of eighty years.

positive for a BRCA mutation and thus had to consider their own risk as well as their family members.

The inheritance pattern of the BRCA mutation is autosomal dominant, which means that both females and males have an equal chance of inheriting the abnormal copy of the gene from their parents. Thus, a mutation in BRCA1 and BRCA2 can be inherited from either an individual's mother or father with each child having a 50% chance of inheritance (NCI, 2015). In families where there are many incidences of young female cancers and within certain ethnic groups such as the Ashkenazi Jewish population, there is much more likely to be an alteration in BRCA1 and BRCA2 (Bleiker et al., 2003; d'Agincourt-Canning, 2006b; Julian-Reynier et al., 2010; Gopie, 2013). In the Ashkenazi population, for example, there is a tenfold increase in the mutation compared to the general population. Clusters present in families within the Jewish populations and these are primarily located in Iceland, Sweden and Quebec (Frank et al., 2001; d'Agincourt-Canning, 2006b; John et al., 2007).

## **1.5 Genetic Risk, Screening and Counselling**

### **1.5.1 Genetic Risk**

Over the past twenty years, there has been a growing interest in the concept of risk amongst health care professionals. Risk information informs both public health policy and clinical practice with its focus on prevention of disease and management of risk. With increasing access to genetic risk information, it is

imperative that clinicians understand what it is like living with increased health risks and thus the information and support individuals require. One of the most significant findings from a qualitative study exploring the experiences of women living with an increased risk of breast and ovarian cancer was that they wanted more information from health care practitioner's (McEwan, 2011).

Many women will make decisions about risk-reducing surgery based on their perceived rather than their actual risk. Within health care, the term 'risk' refers to the probability that members of a certain category will develop or contract a disease and is a term interpreted by some as 'harm', relating to many uncertainties, including the future (Zinn, 2005). Indeed, Palmer and Sainfort (1993) conceptualise risk as *'a combination of probability and something adverse, unpleasant or dangerous'* (pg. 275). However, more recent research would argue that for some individuals, the concept of 'risk' can be a motivating force that positively affects future actions (Horlick-Jones and Prades 2009; McEwan, 2011). Within this study, 'risk' is that which concerns women and their wider family members with a BRCA1 or 2 mutation at risk of developing breast, ovarian and prostate cancer either now or in the future. Thus, for many of the participants in this study, 'risk' may represent both harm and a motivating force. Harm because they have a confirmed significant risk of getting cancer and motivation because they have the ability to reduce that risk and have elected to have risk-reducing breast surgery.

Increasing biomedical knowledge of the genetics of breast cancer alongside an increasing ability to access risk assessment technology such as genetic testing and risk-reducing surgical techniques has had profound implications for women with a family history of breast cancer and their wider family. Firstly, it exposes women to make choices about medical and surgical interventions, when there is no actual disease. Secondly, it has the potential to affect both men's and women's emotional wellbeing and sense of self. Thirdly, it can raise moral and ethical issues of disclosure and secrecy of risk information that can impact on the wider family network (Hopwood, 2005; Featherstone et al., 2006). Indeed, genetic risk and predisposition to a disease are a complex family matter with considerations that go beyond the family member who initiates testing. These include ethical and moral issues, confidentiality, who 'counts' as family and the possession of different levels of knowledge and understandings. Thus, it is never an individual matter (Featherstone et al., 2006). Furthermore, despite the ways 'risk' is communicated to individuals in terms of either percentage's or ratio's, other factors are also involved in how individuals make sense of that risk and they will have different interpretations.

Despite evidence that genetic counselling for cancer risk improves the accuracy of an individual's understanding of risk (Meiser and Halliday, 2002 Keller et al., 2008). Sivell et al., (2008) systematic review found that not only do individuals find it difficult to accurately quantify their risk, but the way they understand risk is strongly affected by their lived experience. Kenen et al., (2003b) in a qualitative UK study with twenty-one women at high risk of breast

and ovarian cancer found that most women perceived their risk as a 'chronic risk' which resulted in a change in their self-identity. Those participants whose mothers or sisters had died of breast cancer experienced the saliency of their risks much more acutely than those whose family members had recovered from their cancers. These are important factors that are likely to influence women requesting risk-reducing surgery.

### **1.5.2 Family Risk and Responsibility**

Genetic testing and the resulting information about the risk of developing a condition involves a consideration of an individual's relationships, both biological and social (Hallowell, 2009). Receiving genetic test results often bestows individuals with a responsibility to manage their risk and thus their bodies, in order to prevent any potential health issues (Hallowell, 1999; d'Agincourt-Canning, 2006a; Bury, 2008., McEwan, 2011). Therefore, a BRCA mutation will affect the way in which women and families think about their bodies. This, in turn, will affect their sense of self-identity and even before the risk is identified, some women may already have an uneasy relationship with their bodies. This uneasiness will affect how they perceive themselves and their body image leaving them feeling vulnerable, especially when faced with decisions regarding risk management and in particular, risk-reducing surgery. This, in turn, can have an impact on relationships, dating and marriage (Hamilton et al., 2009, Matloff et al., 2009; Hamilton and Hurley, 2010).

The responsibility of risk, testing and disclosure of genetic information is considered a matter for the individual and their family as opposed to the professionals, but the timing and the content of the disclosure, with children especially, varies within families. The difficulties and challenges of this have been explored in a qualitative study by Hallowell et al., (2005b), using semi-structured interviews with twenty-nine family members. Parents described how they felt it was their responsibility and obligation to undergo the test and to disclose the results to their children.

However, individuals perceive 'family' in many different ways. Family life involves a number of different relationships, which can lead to complexities and vulnerabilities (Hughes and Ferguson, 2004). Families who are assessed as at high risk of a disease are particularly vulnerable because the risk exposes them to difficult decisions. This can be challenging, particularly if there are changes in family relationships with the potential for secrecy and the associated difficulties in communicating genetic risk information (van Oostrum et al., 2007). This may even lead to a re-evaluation of the self and Novas and Rose, (2000) suggest that genetic testing creates 'new categories of persons' who have an obligation to manage themselves and others once provided with genetic risk information.

Communication and disclosure of genetic results within families, especially when male carriers are involved, is felt to be a family matter (Hallowell et al., 2005b). However, Featherstone et al., (2006) also remind us that it is not as

straight forward as we may think. Although practitioners may assume that the genetic risk information is disseminated unproblematically through families, there is no way of knowing who, what, or how family members are told or indeed, who counts as family in the context of genetic risk information. This is important for clinical teams because many of the individuals and families who seek surgery are related and although some attend surgical consultation together, others attend separately and do not have clear lines of communication.

### **1.5.3 BRCA Screening**

If an individual's family history of breast and ovarian cancer indicates that this may be associated with a mutation in a BRCA gene, the clinical team will construct a detailed family history or 'pedigree'. Screening for a mutation is based on family history and the Breast and Ovarian Analysis of Disease Incidence and Carrier Estimation Algorithm (BOADICEA), is one example of a computer programme used to calculate a person's risk, recommended by NICE (Lee et al., 2013, NICE, 2013). The National Institute for Health and Care Excellence (NICE) Guidelines (2013) on family history, recommend that in both primary and secondary health care, a history should be taken and if it meets the criteria, that a referral is made to a cancer genetic service.

#### **1.5.4 Genetic Counselling**

In the UK, in line with current guidelines (NICE, 2013), before performing a genetic test, an NHS clinic requires that at least two sessions of genetic counselling take place with the individual and their family seeking testing. This is the case in Wales where the study took place but often only one session is attended. The woman and her family will see a geneticist or a trained counsellor who discuss with them the implications of both a negative and a positive test result, to help them understand the available risk management options, and make the best possible adjustment to the risk presented to them. Genetic counsellors are specialists who assess risk and explain inheritance to those who seek information. They discuss choices with individuals and their work is directed at families (Featherstone et al., 2006).

Genetic counsellors are trained in counselling and communication and provide psychosocial assessment and support to those attending the genetic clinic (Gregory et al., 2010). In many units across the UK, but especially in Wales, following the genetic test result, if a woman requests to see the surgical team to discuss the surgical options regarding risk-reducing mastectomy, she is referred. However, in Wales, the surgical consultation may not occur for some months or years after the genetic test. Furthermore, cancer genetic services vary across the UK in terms of inter-professional working and therefore some services will be integrated and on one site whilst in some parts of Wales, the surgical consultation is on a different site and as already mentioned, can occur months or years following a test. An interdisciplinary approach with good



communication is therefore essential because there is wide variation in the amount of information that geneticists and genetic counsellors provide about the surgical techniques of breast surgery and reconstruction. There is also a wide variation for women to have access to a specialist breast care nurse.

Genetic counsellors, as well as the surgical team, need to be up to date with information in order to give women and their families informed choices. This is important because the quality of the information given to them and their families regarding their surgical options and the way in which the information is communicated will have a major influence on the subsequent decisions that are made, including a decision to elect to have RRM. The issue of reconstruction is complex, with many different techniques to discuss and consider including the woman's and family expectations and post-operative recovery. It is therefore essential that professionals working with this patient group are up to date with their knowledge and trained to provide appropriate information and support to women and their families.

### **1.5.5 The Role of the Breast Nurse Specialist**

Nurse specialists working in breast disease are highly experienced key workers in the Multi-Disciplinary Team (MDT) (NICE, 2009). They provide specialist support and information to women with breast cancer and their families from diagnosis to recovery, having undergone extensive training. Their goal is to prepare individuals and the family for treatment and set realistic expectations,

for example, by showing photographs of the different stages of surgery. More frequently, many nurse specialists have undergone extended training to provide psychosocial interventions, for example, Cognitive Behavioural Therapy (CBT).

Evidence suggests that women with breast cancer who are cared for by specialist nurses have reduced psychological morbidity and an improved quality of life (McArdle et al., 1996; Rustoen and Begnum, 2000; Eicher et al., 2006). NICE guidelines on acute and advanced breast cancer (NICE, 2009) recommend that all women diagnosed with breast cancer should have access to a key worker such as a clinical nurse specialist. However, women and their families with the BRCA mutation in many parts of the UK, but especially in most of Wales, do not routinely have any pre-surgical support from a specialist nurse and the only contact may be with a geneticist or a surgeon. Furthermore, a multidisciplinary approach for patients with a BRCA mutation undergoing breast surgery has been shown to improve both the experience and the outcome for such patients (Alamouti et al., 2015).

Evidence demonstrates that decisions to proceed with surgery made by surgeons as opposed to the woman herself, result in higher levels of dissatisfaction and regret post-surgery (Borgen et al., 1998; Frost et al., 2000; Payne et al., 2000).

## **1.6 The Impact of Testing and Disclosure**

### **1.6.1 The Impact on Women**

When a woman receives a positive mutation result, she is confronted with difficult life-changing decisions that carry many risks both physical and psychological (Howard et al., 2009). The fear of cancer, uncertainty about the future, risk to family members and the consequences of surgery are just some of their concerns (Lodder et al., 2002; Van Dijk et al., 2003; Watson et al., 2004; Antill et al., 2006; Graves et al., 2012). The decision to pursue testing is complex and difficult for many women and their families to comprehend (Hamilton and Hurley, 2010). Interestingly, women at risk of breast or ovarian cancer from a paternal family history are less likely than those who have a maternal family history, to initiate a test or visit a genetic clinic, believing it unlikely that they will have an increased risk (d'Agincourt-Canning, 2001; Forrest et al., 2003). Consequently, many women fear rejection after disclosing a positive gene mutation result (Hoskins et al., 2008; Klitzman and Sweeney, 2011) and greater distress levels have been found in BRCA positive women when compared to those who tested negative or received uninformed results, up to five years after testing (Graves et al., 2012). Reports of the need for psychological counselling has been consistently reported (Vos et al., 2013) with long term distress noted up to seven years post-test (Hughes et al 2011).

To assess the impact of testing on women and men with a mutation, Watson et al., (2004) conducted a multi-centre prospective study in the UK using

questionnaires (obtaining a 92% response rate). A total of two hundred and sixty-one participants took part. Cancer worry, intrusive thoughts and a negative impact on quality of life were apparent for the ninety-one mutation carriers compared to the one hundred and seventy non-carriers, and this impact continued up to a year post-test. More importantly, despite 28% of carriers electing to have RRM, this did not reduce their cancer worry or the intrusive thoughts at twelve months.

This is in contrast to other studies (Hatcher et al., 2001; Lodder et al., 2002; Brandberg et al., 2008; Patenaude et al., 2008) which identified a significant reduction in cancer fear. Moreover, two non-carriers elected to have RRM before genetic testing and twenty-nine participants sought professional help post-surgery. It is unclear, however, how and why the non-carriers came to their decision, what the professional help involved, why they required it or how many of those seeking help had undergone RRM. Compared with the women, the twenty-five BRCA positive men in the study did not report poorer mental health scores, but three also required professional help post-testing. Again, it is unclear what professional help consisted of or any explanation of why it was necessary. The strength of the study lies in the prospective design and the inclusion of nine centres across the UK.

The concept of self and self-identity is very subjective for both men and women. In a qualitative study by d'Agincourt-Canning (2006b) in depth interviews were conducted with thirty-nine participants, five of whom were men. The study

attempted to explore the impact of a positive BRCA mutation on self and self-identity. Themes that emerged linked their positive mutation results to becoming much more aware of three types of self. Their embodied self (physical), their familial (relational) self and their (social) self. For the majority of participants, knowing their genetic results enabled them to take control and plan measures such as surveillance or surgery.

However in contrast, for a minority of younger women, they reported being thrust into an uncertain state of neither ill nor completely well, what Dagan and Goldblatt (2009) refer to as the *'twilight zone'* (pg. 273). In their study with seventeen Israeli BRCA positive women, a state somewhere between health and sickness was experienced which led to a reconsideration of the women's social identity, with one participant reporting regret in having had the genetic test. In d'Agincourt-Canning's study, (2006b), the knowledge of their genetic risk following a positive test result had a profound and limiting effect on the younger women who were extremely vulnerable and frightened by the certainty (a mutation) and the uncertainty (when they would get cancer and how severe it would be) of the genetic information. This information gave them a confirmed risk that they did not have before and a future that they perceived as having no control over, their expectations therefore was that of a grim future.

The strength of d'Agincourt-Canning's study highlights the fact that individuals do not interpret their risk or the information imparted in a neutral manner but as part of their wider social experience. What the study does not highlight

however is why women, especially younger women, experience such disruption whilst others report they are able to take control. Some limitations of the study include the following, the sample were all highly motivated to take part, many had already had cancer and received treatment and it only captured experience at one moment in time. Responses to genetic information, for example, may shift over time (d'Agincourt-Canning, 2006b).

### **1.6.2 The Impact on Men**

Compared to women, the rates of men seeking and receiving testing are much lower (Julian-Reynier et al., 2000; Foster et al., 2002). Hallowell et al., (2005a, 2006), explored the influences on the decision to be tested and men's role in the transmission of the gene mutation within the family. Similarly, to women, men were found to portray themselves as morally responsible for their children and testing was seen as a family duty, findings supported by other studies (Liede et al., 2000; d'Agincourt-Canning, 2001,2006a; Lodder et al., 2001).

Males who are BRCA positive can experience increased anxiety and stigma because of their risk of cancer (Daly et al., 2003, Heshka et al., 2008, Stromsvik, 2010) but it is not always related to the implications for themselves or their options, because currently there is a lack of preventative or risk-reducing measures for this group (Lodder et al., 2001, Hallowell, 2005a). Shiloh et al., (2013) studied fifty-one men with a positive mutation and thirty who were negative in a cross-sectional study, to examine the cognitive,

emotional and behavioural impact four years after receiving their test result. Telephone interviews identified that those with a BRCA mutation had increased perceptions of their risk and their distress levels were significantly higher than they were before the test, with this impact continuing after the four years.

Men were much more aware of their physical self and relational self, especially their responsibility to others. More significantly, their distress concerned and focussed on their children, especially their daughters, findings that are supported by other studies (Mc Allister et al., 1998; Lodder et al., 2001). However, the limitations of this study include the fact that many of the mutation negative men did not complete the full survey, the majority were from the Ashkenazi Jewish population who hold particular cultural beliefs, and there were no pre-test measures of distress to compare with. Using self-report measures to identify health behaviours and the length of time following testing is also a limitation.

Overall, men express less negative feelings about testing positive than women (Hesse-Biber and An, 2016) and in constructing their own perspective on the role they play in the inheritance of the mutation, they drew upon discourses of guilt, blame or fate even if they test negative (Hallowell et al., 2006). Significantly, if men are included in family conversations about women in the family with a breast cancer diagnosis, very few become involved in the decision about RRM (Leide et al., 2004). Indeed, men have been found to deliberately avoid any conversation regarding surgery (Mc Allister et al., 1998). However,

despite it being a difficult task, men describe feeling empowered about informing their children of the result because they were giving them the opportunity to act upon it. However, this was always weighed against the risk of causing their children additional emotional harm (Hallowell et al, 2006).

### **1.6.3 Family Disclosure**

Disclosure of a mutation within the family depends on individual personality traits that is, whether family members are described as information gatherers, disseminators or blockers of information (Peters et al., 2011), as well as having the knowledge and support to communicate with the family which will depend on the pre-existing culture of discussion about cancer within the family (Lafreniere et al., 2013). Lafreniere et al., (2013), in a retrospective study, examined the process and factors involved in influencing communication of genetic test results to family members. Although the interviews were conducted four to nine years post-testing, the study identifies three phases to the process and offers insight into the complexities. There is a decision-making phase where individuals take the time to decide whether to share and what they share, a disclosure phase where certain family members are told and the reaction phase, which will influence any subsequent disclosure.

Family members have also been found to adopt different communication strategies. Hallowell et al., (2005b) in their study identified three strategies. Complete openness where information to children was transparent, limited



disclosure where some information was shared and total secrecy where testing and the implications of the test was kept a guarded secret. All parents focused on protecting children from anxiety, with limited disclosure being adopted in the majority of cases. Unlike previous research which has focused on disclosure patterns within the wider family (Forrest et al., 2003; Claes et al., 2005), Hallowell's study focuses on the immediate family and on male mutation carriers in particular. One explanation for the use of different disclosure strategies within families is the guilt, blame and resentment identified in some individuals following testing for the gene (d'Agincourt-Canning, 2006b; Stromsvik, 2010; Shiloh et al., 2013).

However, the initial experience of having a genetic test is not always a negative experience; it has been found to empower women to make decisions and to take responsibility for their past, present and future family on moral grounds, such as the responsibility of being a good mother and doing the right thing (Hallowell, 1999; 2003). However, by taking this morally responsible perspective, Hallowell argues that this approach means women actually remove their right not to know, putting their own needs aside. Furthermore, if the result is positive, women then feel a moral obligation to act upon the result thus subjecting themselves to risk management options such as surgery with uncertain consequences that potentially cause physical and psychological harm, in addition to unexpected cosmetic results (Hallowell 2000; Hallowell et al., 2012). Consequently, following a positive BRCA test there is an increase in distress levels leading to anxiety and depression, a reduction in the quality

of life and many problems associated with an altered self-image (van Oostrum et al., 2003; Watson et al., 2004; Foster et al., 2007; Kenen et al., 2007; van Dijk et al., 2008; Crotser and Boehmke, 2009; Esplen et al., 2009). Often these increased distress levels continue in the long term (Watson et al., 2004; Graves et al., 2012).

#### **1.6.4 Impact on Younger Women**

Younger women, in particular, are often unaware of the serious implications of having a test that could identify an altered BRCA gene. They have less experience to draw upon in decision-making (Hamilton et al., 2009) and therefore suffer from higher than normal cancer related distress. Over a third of patients within two studies experienced high cancer related distress associated with uncertainty about the available options, the inheritance of the gene and concerns about its impact on childbearing (Linden et al., 2012; Patenaude, 2013).

In the study by Patenaude, forty daughters of BRCA positive women, some of who were told before they were eighteen years old, participated in a qualitative telephone interview to identify the impact of their mothers' result and their cancer related distress. Findings demonstrate that their genetic knowledge was suboptimal, having many uncertainties and misconceptions. For example, many believed that a negative mutation could still lead to a child with a mutation. Thus, despite having normal levels of general distress, their cancer

related distress, using well-validated measures, was high. However, this was a retrospective study, three years post-test and not having any pre-test distress scores, demonstrates its limitations. The daughters were of different age groups and all the mothers in this study were still alive.

Daughters of deceased women with a BRCA mutation have been found to demonstrate even higher cancer related distress (Patenaude et al., 2013). Young women who are BRCA positive face many difficult decisions including the disclosure within relationships, the issue of being single and childless and the uncertainty about breast cancer (Hamilton and Hurley, 2010). Indeed, the impact of uncertainty regarding decisions and future health following a positive mutation result has been compared and equalled to that of a diagnosis of breast cancer. Dimillo et al., (2013), conducted a grounded theory study with six BRCA positive women under the age of forty-five who had decided not to have surgery. This group was found to be constantly fearful of a cancer diagnosis with many uncertainties, feelings of powerlessness and vulnerability.

The negative consequences of testing, therefore, have to be weighed against the benefits (DiProspero et al., 2001; Beran et al., 2008). Younger women have been found to be particularly vulnerable as they misinterpret their lifetime risk as their short-term risk, making them believe that cancer is imminent (Werner-Lin et al., 2012). This is important as many younger women are seeking bilateral mastectomy. Evidence suggests that compared to older

women with the mutation, younger women are more distressed following a BRCA mutation result (Lodder et al., 2002; Watson et al., 2004) and find it more difficult to adjust (Lodder et al., 2002; van Oostrum et al., 2003; Watson et al., 2004).

#### **1.6.4 Impact on Adolescence**

In early childhood when young girls are observing their mother's body in comparison to their own, sexual self-image and sexuality are beginning to develop. Breasts should represent sexual maturity, pleasure, motherhood and breast feeding (Matloff et al., 2009). However, in families where a BRCA mutation exists, breasts for the young female, in particular, can often become objects of fear, anxiety, scars, reconstruction and fear of death (Hamilton and Hurley, 2010; Werner Lin, 2001). Indeed, in families where women have lost their breasts, the young female may experience feelings of guilt about her young, healthy, intact body (Matloff et al., 2009). For adolescents in families where a BRCA mutation exists, excitement about sexual development is clouded by the risk and fear of cancer (Matloff et al., 2009). Moreover, experiences of cancer with a significant family member at the time of puberty can have a negative impact on sexual development and psychosocial functioning. For example, the breasts may signify danger and these feelings can continue into adult life, affecting both partnering and family planning (Werner-Lin, 2008).

### **1.6.5 Impact on Partners and the Family**

The process of genetic testing and the decisions that follow will always have implications that go beyond the individual who seeks testing (Featherstone et al., 2006., Hallowell, 2009). The literature examining the experience of partners is limited, however it has been identified that partners of women diagnosed as BRCA positive find the process of testing and the decision to elect RRM particularly stressful (Metcalf, et al., 2002; Mireskandari et al., 2006; Matloff et al., 2009; Sherman et al., 2010). Studies have examined the role of the spouse in the decision to be tested (Kessler, 1993; Williams et al., 2000; Bluman et al., 2003) and found that they wanted to be more involved in this process and included by professionals (Metcalf, et al., 2002; Mireskandari et al., 2006). Indeed, some partners feel alienated from the testing process altogether (Sherman et al., 2010).

Feelings of lack of support at all stages of the process for partners and women with a BRCA mutation is associated with higher levels of distress (Sherman et al., 2010). The need for more support and information from health professionals around the time of disclosure and a wish to meet other partners has therefore been identified (Metcalf et al., 2002; Mireskandari et al., 2006). Studies that have assessed knowledge levels with regard to the whole process of testing, identify that knowledge in partners and family members is suboptimal. This lack of knowledge adds to their overall distress (Lerman et al., 1996; Wonderlick and Fine, 1997; Bluman et al., 2003; Mireskandari et al., 2007; Patenaude et al., 2013). Studies have also highlighted that the increase

in distress immediately following genetic testing and a positive mutation, concerns fears of breast cancer and their children's risk (Lodder et al., 2001; Metcalfe, 2002; Kenen et al., 2006; Mireskandari et al., 2006; Mireskandari et al., 2007; Jeffers, 2014; Mauer et al., 2015).

Support from health professionals, in general, is limited for women with the BRCA mutation and their partners, despite evidence that demonstrates elevated levels of distress and the need for more support (Lloyd et al., 2000; Metcalfe et al., 2002; Mireskandari et al., 2007). Importantly, the overall adjustment of the partner to the whole journey is significant in providing support to the BRCA positive woman, as support received from spouses and social support networks have been identified by women to aid their coping and adaptation (Wylie et al., 2003; Metcalfe et al., 2004a). However, no studies to date have focused specifically on the support needs and the whole experience for the woman, her partner and family member following a positive BRCA result and the experience of RRM.

## **1.7 Prevention and Risk Management Options in the UK**

There are several available risk management options for women with a BRCA mutation, these include the decision of no intervention. Currently, women can choose to have screening in the form of mammography or magnetic resonance imaging (MRI), bilateral risk-reducing mastectomy (RRM) and/or bilateral salpingo-oophorectomy (RRSO) (Zakaria and Degnium, 2007). The updated

family history guidelines have also given more options for women and families in the form of chemoprevention, anti-oestrogen medication, Tamoxifen and Raloxifene (NICE, 2013). Risk management for men relies on breast awareness and screening for prostate cancer.

### **1.7.1 Surveillance and Screening**

Following a BRCA mutation result, women can choose to have surveillance in the form of mammography and Magnetic Resonance Imaging (MRI), which are images of the breast, performed yearly, commencing at the age of thirty. However, screening enables the early detection of cancer, but it is not risk-reducing. Consequently, for women who decide on screening, the early detection of cancer will still require the need for surgery, chemotherapy, radiotherapy and chemo-prevention medication. All of the treatment modalities that women with the mutation are trying to avoid (Hoskins and Greene, 2012). There is no evidence at this current time that such surveillance reduces mortality in high-risk women (Evans et al., 2009; De Brock et al., 2013). Indeed, a cumulative effect of yearly mammograms may compound the risk of breast cancer due to the radiation exposure. Furthermore, the fact that young women have dense breasts means that cancers can be missed on mammography. More significantly, the risk management options in the UK are limited for those women under the age of thirty who are BRCA positive. This is due to the fact that any form of available screening starts at the age of thirty as recommended by NICE (2013) and this leaves young women feeling

vulnerable, abandoned or stuck waiting for screening to begin (Brunstrom et al., 2016). It is proposed that this is one of the reasons why younger women are increasingly electing to have RRM.

For men with the mutation, risk management is very limited, consisting mainly of breast awareness and screening for prostate cancer. This latter option involves having a blood test at the age of forty for prostate specific antigen (PSA), a tumour marker, (Rebbeck et al., 2004; Madalinska et al., 2007; Nelson et al., 2009). PSA is a protein produced by normal cells in the prostate but also by prostate cancer cells and it is normal for men to have a small amount in their blood. A raised blood test result, therefore, may not necessarily indicate prostate cancer.

### **1.7.2 Chemoprevention**

Chemoprevention is the use of drugs, vitamins or agents in an attempt to reduce the risk of cancer or its recurrence. Women with the BRCA gene and those with a strong family history now have the additional option of taking Tamoxifen or Raloxifene, which are anti-oestrogen medications in the form of a tablet. They are known to reduce the risk of breast cancer occurring (Nelson et al., 2009) and have been recently approved by NICE (2013). However, the drugs have many side effects including weight gain, the risk of thrombosis (blood clots) and menopausal symptoms. Importantly, the drugs only offer a



40% reduction in the risk of developing breast cancer (De Leeuw et al., 2008; Beattie et al., 2009).

## **1.8 Surgery: Risk-Reducing Mastectomy**

Despite alternative options, a large proportion of women with a mutation, choose to have bilateral Risk-Reducing Mastectomy (RRM), which involves the complete removal of both breasts and/or Risk-Reducing Salpingo-Oophorectomy (RRSO), the removal of the ovaries and fallopian tubes. It is estimated that over 50% of women choose risk-reducing breast surgery but up to seventy-five percent choose ovarian surgery (Evans et al., 2009; Skytte et al., 2010).

RRSO is usually performed at a different time point to RRM, with some women choosing to have it before breast surgery, whilst others wait until after. The number of women seeking testing with the expectation of having risk-reducing surgery if they have the mutation has increased (Evans et al., 2009; Gopie et al., 2013). The fear of breast cancer and concern about caring for their children if a breast cancer occurred, has been found to be the biggest predictor for this decision (Lodder et al., 2002; Hatcher and Fallowfield, 2003; Antill et al., 2006; Howard et al., 2009; Haroun et al., 2011). Understanding what influences women and their families in their decisions is important for the breast and wider team who are in a position to provide decision making support because this surgery is irreversible and women are often very young when electing to have

it (Hamilton et al., 2009; Hamilton and Hurley, 2010; Hoskins and Greene, 2012; Glassey et al., 2016).

In the UK, new guidelines recommend that risk-reducing surgery should be discussed with all BRCA positive women and that a suitably trained professional should lead the discussion. The offer of immediate reconstruction should also be included (Evans et al., 2013; NICE, 2013). Women and their families are referred to a surgical team if they decide to proceed with surgery, but ultimately it is the surgeon's decision whether surgery goes ahead (Heemskerk-Gerritsen et al., 2007; Schwartz et al., 2012; Hampel et al., 2014).

Bilateral Risk-Reducing Mastectomy (RRM) leads to the greatest risk reduction of developing breast cancer in women with the BRCA mutation (Meijers-Heijboer et al., 2001; De Felice et al., 2015). Evidence has consistently demonstrated that RRM reduces risk by 90-95% (Evans et al., 2009; Rebbeck et al., 2009; De Felice et al., 2015). Risk-Reducing Salpingo-Oophorectomy (RRSO) also offers a significant decrease in the incidence of both ovarian and breast cancer, offering a 50% reduction in breast cancer and a 95% risk of ovarian cancer (Rebbeck et al., 2009; Evans et al., 2009). Furthermore, it appears to be a far more acceptable procedure than RRM because it leaves no visible outward bodily change, unlike breast surgery (Phillips et al., 2006; Crotser and Boehmke, 2009).

Recent evidence has shown that a combination of RRM and RRSO at the age of forty results in the best way of increasing survival rates for women with a BRCA mutation, a significant reduction in all cause breast cancer specific and ovarian cancer specific mortality (Domchek et al., 2010). This evidence also suggests that performing RRM any earlier has little impact on survival rates (Kurian et al., 2010). However, RRSO is not recommended until the age of forty because of the implications of early menopause, including the negative effects on bone health (Evans et al., 2009; Kurian et al., 2010). Increases in anxiety (van Oostrum et al, 2003), anger and distress (Dorval et al., 2000) as well as issues regarding quality of life and vulnerability have all been noted (Hallowell 2000; Hallowell and Lawton, 2002; Esplen et al., 2009).

By electing to have RRM, women have both breasts removed and also have to consider the removal of the nipple areolar complex (NAC). Currently, if all the breast tissue and the nipple is removed, there is up to a 95% chance that breast cancer will not occur. In some breast units in the UK, surgeons will preserve the NAC once a woman has been fully informed of the associated risks because if the nipple is preserved, the actual risk reduction is unknown (Reynolds et al., 2011; Yao et al., 2015). British, Dutch and American women are more in favour of RRM with over 50% choosing this option when compared with the rest of the western world (Evans et al., 2009; Gopie et al., 2013). Furthermore, international variation exists amongst physicians' attitudes towards RRM, in France and Germany for example only 30% and 27%

respectively, believe that it should be an option for BRCA women which may account for the difference in uptake (Gopie, et al., 2013).

### **1.8.1 The Psychological Effects of Risk-Reducing Mastectomy**

Studies have consistently reported a reduction in fear, reduced anxiety and reports of relief following risk-reducing mastectomy (McGaughey, 2006; Hallowell et al., 2012; Hagen et al., 2014; Glassey et al 2016). However, a literature review of thirteen studies examining the impact of RRM focussing specifically on body image identified that up to 50% of women suffer a negative effect on body image and experience adverse effects on sexual life (McGaughey, 2006). A reduction in quality of life has also been identified (Frost et al., 2000; Hopwood et al., 2000; Lloyd et al., 2000; Payne et al., 2000; Dowdy et al., 2004; Metcalfe et al., 2005; Geiger et al., 2007; Brandberg et al., 2008; Lostumbo et al., 2010; den Heijer et al., 2012; Gopie et al., 2013) with impacts on femininity and sexuality noted (McEwan, 2011). Many of these studies, however, fail to provide a deeper understanding of these impacts, especially in the context of the wider family. The family is an integral part of the decision and the experience (Sherman et al., 2010; Rowland and Metcalfe, 2014; Mauer et al., 2015). However, there are no qualitative studies to date that include the experiences of BRCA positive women specifically who have not had breast cancer and the experience of risk-reducing mastectomy, their partners and members of the family. Only three studies have been identified that include partners and RRM (Lloyd et al., 2000; Mireskandari et al., 2006.,

Mauer et al., 2015). It is argued that it is important for clinical practice to identify the informational and psychosocial needs of women including their family and to provide an appropriate care pathway that is in line with that experienced by women who have breast cancer.

### **1.8.2 The Decision to Elect to have Breast Reconstruction**

The majority of women in the UK elect to have breast reconstruction following RRM (Evans et al., 2005; Bresser et al., 2006). America, Australia and the Netherlands also have high rates of RRM with immediate breast reconstruction (Howard et al., 2009; Skytte et al., 2011; Nelson et al., 2012). The option and availability of breast reconstruction strongly influences a woman's decision to undergo RRM (Rolnick et al., 2007; McQuirter et al., 2010; Nelson et al., 2012). Nelson et al., (2012) conducted a survey of forty women who had elected RRM, utilising various reconstructive techniques.

Having the option of different reconstruction techniques strongly influenced 92% of the participants to have RRM. However, there were a few limitations of the study, its retrospective survey design and the inclusion of BRCA positive women who also had previous breast cancer and were removing their opposite breast (contra-lateral). The sample size was small and therefore the power of the study was low. The opportunity for further exploration of decision-making is also lost in the survey design.

Women have the option of either immediate or delayed reconstruction using various surgical techniques. This will depend upon their breast size, their general health, their preferred technique and the expertise of the surgeon. Breast implants are the simplest way to reconstruct the breast but advances in surgical techniques have given women the option of using their own tissue (autologous) alone or in combination with their own muscle (a pedicled flap) to reconstruct a breast. There is also the transfer of skin and fat (a free perforator flap) from either the abdomen, buttocks, back or the inner thigh and many women prefer this option as the final cosmetic result resembles a more natural looking breast (Majdak-Paredes and Fatah, 2009).

Counselling for such procedures takes time and women need adequate information and preparation in order for the final outcome to meet their expectations (Frost et al., 2000; Altschuler et al., 2008; Hallowell, 2012). Thus, although there have been reports of satisfaction following breast reconstruction, (Brandberg et al., 2000; Nissen et al., 2001; Harcourt et al., 2003; Elder et al., 2005; Hallowell et al., 2012) surgery has been shown to have negatively affected a woman's sense of self and identity, quality of life and psychological health (Hallowell, 1998; Patenaude, 2005; McCullum et al., 2007; Gahm et al., 2010; den Heijer et al., 2012; Gopie, 2013). However, a deeper exploration and understanding of these findings is required so that it can be acknowledged and addressed in clinical practice. Moreover, although RRM is now accepted as an option to offer women for risk management (NICE, 2013), some still consider it in parallel to mutilation (Press et al., 2005).

The more complex techniques require the input of a plastic surgeon and take up to ten hours of operation time (Isern et al., 2008) carrying the possibility of complications and several months of healing and recovery. It has been estimated, for example, that up to 64% of women have one or more complications (Barton et al., 2005), with many women requiring re-operation (Gahm et al., 2010; Arver et al., 2011). Two hundred and twenty-three women in a study by Arver et al., underwent RRM with two hundred and twenty electing reconstructions. 64% had unanticipated second operations as a result of infection, necrosis, capsular contraction of the implant and flap failure (Arver et al., 2011). These long-term problems and the experience for women and their family need further exploration.

### **1.8.3 Surgery Versus Screening Programmes**

Many studies have explored the emotional well-being of high-risk women attending screening programmes for breast cancer. These suggest that screening may actually increase anxiety (Lerman and Schwartz, 1993; Howard et al., 2009), while a range of studies suggest that women who elect RRM experience a decrease in cancer worries, anxiety and psychological distress (Stephanek et al., 1995; Josephson et al., 2000; van Geel, 2003; Brandberg et al., 2008; den-Heijer et al., 2012). However, Bresser et al., (2007b) found that women who experienced high levels of distress prior to RRM continued to experience high levels post-surgery and concluded that other factors such as personality and coping strategies may also be relevant, factors which are

important for psychosocial assessment. Furthermore, evidence suggests that many women electing to have RRM need more information and support than women having a risk-reducing contralateral mastectomy (the opposite breast to their breast cancer), pre-surgery, regarding all aspects of the process. This may also be one explanation for the high level of distress (Josephson et al., 2000; Bresser et al., 2006; Rolnick et al., 2007 Bonadies et al., 2011). In the UK, women with breast cancer who elect to have contralateral risk reduction will have also had support from their key worker which could explain their reduced need for information and support.

## **1.9 Summary**

This chapter has provided the background to the study and outlined the factors relevant to the study including the incidence and treatment of breast cancer, one of the main preventative aims of bilateral risk-reducing mastectomy. It has provided an overview of a BRCA mutation, the complexities, and impact of risk, genetic testing and decision making for the individual and the family. It has also provided an overview of the decision to elect to have risk-reducing mastectomy and what is known about some of the psychological impacts of this decision thus far. This chapter identified that the literature does not provide a deeper exploration of these impacts in the context of the wider experience for the woman, her partner and family. Thus, there is a need to add further insight and knowledge into the experience so that nursing care if necessary, can be tailored and developed to meet their needs. The next chapter will draw



upon more clinically focused literature to identify and establish what is known and what is yet to be explored.

## **CHAPTER TWO – The literature review (The Current Horizon of Understanding, Gadamer, 2004)**

### **2.1 Introduction**

This chapter focuses on the identification of the 'pre-understandings' (Gadamer, 2004) related to the phenomenon of living with a BRCA mutation and undergoing bilateral risk-reducing mastectomy from which my research question was developed. Examining the available literature (The Current Horizon of Understanding Gadamer, 2004), developed the foundations presented in Chapter One.

This literature review is not a systematic review, however, I used a systematic approach to locate relevant literature. In keeping with the phenomenological approach to my study, the focus of this literature review is understanding the phenomenon of living with the BRCA mutation and the experiences of RRM for the woman, her partner and the wider family. This approach was adopted to allow for more openness to the phenomenon studied by having fewer pre-conceptions, this added to the rigour of the study (Speziale and Carpenter, 2007).

## **2.2 Systematic Search Strategy**

### **2.2.1 Preliminary Search January 2011 until April 2011**

The literature search started in January 2011 before the commencement of the Ph.D. Teaching students inter-professionally and enabling them to shadow me in clinics, in addition to my personal interest, meant that I needed to stay up to date with the literature. I identified the literature for conferences and workshops that I was attending, observing that more women were being identified and referred for risk-reducing surgery. The team of surgeons with whom I worked were particularly concerned about performing the surgery without an identified BRCA mutation. These preliminary searches identified a lack of qualitative research on both young women with a BRCA women, who had not had cancer and were undergoing risk-reducing mastectomy and their partners and family. My research question and approach started to develop from this initial search.

The initial search focused primarily on women with a BRCA mutation undergoing bilateral risk-reducing mastectomy. This was too narrow and the focus of the review developed and broadened as the study progressed. Initial key words included 'BRCA1/2', 'proband', 'experience', 'risk-reducing mastectomy' and 'prophylactic mastectomy'. A broader search using the additional search terms, 'risk', 'partners', 'family', 'relatives', 'qualitative', 'interpretive phenomenology' and 'phenomenology' was adopted.

Much of the included literature was found in two databases Medline and Web of Science. Seventy-nine papers were found using the above search terms. An example of the Medline search can be seen in Appendix Three. As I developed my question, I expanded my search strategy to include other databases, PsycINFO, Cumulative Index to Nursing and Allied health Literature (CINAHL), Scopus and British Nursing Index (BNI). Initially, the search term 'proband' was used with the use of Boolean operators, no qualitative studies were located. By removing 'proband', 'qualitative' and 'relatives' and broadening the search to include 'risk', more literature became available

I accessed professional documents including professional nursing bodies Royal College of Nursing (RCN), Nursing and Midwifery Council (NMC) Department of Health databases (NICE), grey literature (Open Grey) and used back chaining.

The searches were repeated towards the end of the data collection in 2014 and again following analysis in 2015 to check for any new literature. I accessed specific qualitative journals, Psych-oncology and Qualitative Health Research and specific authors such as Nina Hallowell, Kelly Metcalfe and Regina Kenen who had done research in the field. There were no qualitative prospective studies found that included women, their husbands and family

members and specifically mastectomy. In particular, no studies were located that used Gadamerian or Interpretive Phenomenology.

### **2.2.2 Inclusion Criteria**

The search included literature in English but no limits on dates were set. Due to the limited number of qualitative studies specific to BRCA women who were undergoing risk-reducing mastectomy and who had not had breast cancer and the experience of family members, data was accessed which included women at high risk. This approach enabled research conducted prior to the introduction of clinical protocols and before genetic testing was more readily available, to be found. Articles were accessed that were primary research, where the experience of being BRCA positive and undergoing risk-reducing mastectomy for the woman and her family was the focus, peer reviewed where possible, including systematic reviews, meta-analysis and literature reviews. Below is a summary of the papers found in the various databases.

**Medline/Ovid-79, Psychinfo-8, Scopus-345, CINHAL-29, BNI-3, web of Science -161. Total - 622 papers.**

### **2.2.3 Exclusion Criteria**

The search excluded studies that were:

- Not written in English.

- Women only having bilateral risk-reducing oophorectomy.
- Women with breast cancer choosing risk-reducing, prophylactic or contralateral mastectomy.
- Satisfaction following breast reconstruction.

#### **2.2.4 Expanded Search**

The following additional databases were accessed, Google scholar, Orca, Cardiff University libraries and the Royal College of Nursing Libraries. One recently published literature review relevant to younger high-risk women electing to have risk-reducing mastectomy was found (Glasse et al., 2016). Two Ph.D. thesis, one from Australia, (Crabb, 2006) and one from New Zealand (McEwan, 2011) that focussed on living with the experience of increased breast and ovarian cancer risk and which included BRCA women who had undergone breast surgery, informed the study.

#### **2.2.5 Final Search**

In January 2017, I ran another search. An updated Medline search found 104 papers, 647 papers were assessed in total. After reading the abstracts and excluding many of the medically focussed or genetically focussed papers that were unrelated to the experience of BRCA positive women and their family undergoing mastectomy specifically, removing the duplicate studies and including studies found from back chaining all the papers, 21 papers were

included in the review and the table of included studies can be seen in Appendix Three.

This literature review explores what is known of the experiences of women identified as being BRCA positive and their families who elect to have bilateral mastectomy. However, an understanding of risk and embodiment is necessary in order to critically review the available literature on such experiences. The review has been divided into three sections: risk and embodiment, pre-surgery, and post-surgery experiences.

## **2.4 Risk and Embodiment**

This study involves the body, in particular, the bodies of women who carry a genetic mutation, which puts them at increased risk of cancer and specifically female cancers. For some women being at high risk of these cancers and of the decision to undergo surgery with reconstruction can be a stigmatising condition. Stigma refers to a negative attribute that sets an individual apart from what is considered 'normal' to other individuals in society. Kenen et al., (2007) over a four-year period reviewed the messages on the archived message board devoted to women at high risk for breast and ovarian cancer on the Facing Our Risk of Cancer Empowered (FORCE) website. Twenty-one women's posts were analysed using a qualitative thematic analysis and results found that women worried about whether or not to disclose any information

about their risk and surgery to friends and family because they feared negative and stigmatising comments especially in revealing their body and the loss of their natural breasts.

In trying to consider and understand risk, therefore, Kavanagh and Broom (1998) have identified three types. Environmental risks such as pollution and toxic chemicals, lifestyle risks such as cigarette smoking and alcohol and embodied (corporeal) risk such as the inheritance of a genetic mutation. They propose that environmental risks have very different social and personal consequences, for example, they are often viewed as out of the control of the individual. Lifestyle risks, on the other hand, are seen as occurring as a consequence of the person's behaviour. Importantly, the third type of risk, embodied or corporeal risks are distinguished from other types of risk by being located in the body of individuals, threatening them from within.

The inheritance of the BRCA gene mutation is one example of a corporeal risk which is perceived as being out of the control of individuals and less avoidable when compared to environmental or lifestyle risks. It poses a worry for the present and for the future, defining who a person is, for example 'being a carrier', rather than what they do. Furthermore, unlike environmental and lifestyle risk where both the body and the self are at risk together, with corporeal or embodied risk there may be a dissociation of the body from the self to counteract the threat of the particular body part (Kavanagh and Broom, 1998). In this sense, therefore, the decision to elect RRM could be initially



viewed as one way of managing the corporeal risk of a BRCA mutation and breast cancer.

However, the dissociation of body and self is more complex than that because the risk in some body parts may be easier to dissociate from the self than others. For example, having an abnormal pap test which indicates cervical cancer and removing the womb, as in the study by Kavanagh and Broom (1998), or having the ovaries removed to prevent ovarian cancer, may be seen as easier to dissociate from because these organs are not as visible (Hallowell and Lawton, 2002; Howard et al., 2011). The body in this sense can be thought of as an object, made safe after the removal of its threatening parts. All of the participants in Hallowell and Lawton's (2002) grounded theory study which included forty-nine high risk women of which twenty-three had undergone prophylactic ovarian surgery, felt that the decision to lose their ovaries was much easier because they were internal and invisible. They felt oophorectomy was not an explicit threat to body image and thus body integrity or self, compared to women having a mastectomy (Hallowell 2000).

Indeed, Howard et al., (2011) in their grounded theory study found that being at risk for breast and ovarian cancer and having to make decisions about risk-reducing surgery threatened the women in four aspects of self which were interconnected and which had different significance over time. The four dimensions of self were physical health, self-identity, relationships with others and the emotional self. Preserving these aspects of self-represented the most

salient themes and thus only three out of twenty-two women chose to elect RRM, compared to nineteen women who chose RRSO. This finding implying that the disconnection between the body (their breasts) and self was much more difficult and that preservation of self-identity for these three women was most important. The authors conclude that all aspects of self should be a consideration and seen as equally important. These dimensions of self are also important because they provide direction for appropriate targeted support from health professionals whose general focus is on physical health (Howard et al., 2011). This is significant because the overall adjustment to the loss of a breast is directly related to an individual's pre-existing construct of self, including feelings about one's femininity, womanliness and physical attractiveness (Lloyd et al., 2000; Piot-Zeigler, 2010).

It follows, therefore, that labelling previously healthy women into an 'at risk' category whether it be BRCA positive or those with an unknown gene status, leads to vulnerability and a tendency to think of the body as 'dangerous', thus exacerbating the Cartesian split between body and self. This disconnection between body and self may help to facilitate and justify the risk management decisions made by individuals as in the studies by Hallowell and Lawton (2002) and Kavanagh and Broom (1998) where it is argued that these women were able to implicitly dissociate from self because the ovaries and womb are considered as less obviously connected to feminine identity.

However, this splitting of self and body only appears to occur with particular types of risk such as 'cancer risk'. One explanation for this is proposed by Kavanagh and Broom (1998) who argue that within Western society and culture, cancer is commonly constructed as the 'other' and is frequently portrayed as an 'invader' of bodies. Therefore, these cultural constructions of cancer may be the reason for the Cartesian splitting observed in their study (Kavanagh and Broom, 1998). However, Hallowell (2000) points out that this is not observed in studies of women who have undergone mastectomy for breast cancer. The evidence suggests that although a disruption of self and body occurs during treatment, there is also a deep sense of loss for the breast, albeit, a cancerous breast (Hallowell, 2000; Rosenblatt, 2006; Sandham and Harcourt, 2007; Mehnert and Koch, 2008). As a result of this loss, a profound effect on the self is experienced which challenges a woman's identity leading to a need to reconstruct ones' self-identity (Langellier and Sullivan, 1998; Cromptvoets, 2003; Montazeri, 2008, Piot-Ziegler, 2010).

Moreover, women who experience a loss of their body integrity feel alienated and report less psychological and social well-being (Petronis et al., 2003). A few explanations for this loss are offered. The location of the risk within the body is significant in that some body parts may be easier to dissociate from than others, those body parts for example that are less visible to the social world (Hallowell, 2000; Howard et al., 2011). It is also argued that in the case of women at high risk or with a BRCA mutation who remove their breasts, it is more difficult than expected to split body and self because these women do

not have cancer. Finally, the cultural and symbolic significance of the body, particularly in regard to breasts and pressure to conform to the norms of female beauty, further prevents this splitting of body and self. As Douglas writes of the body:

*'The body itself is a highly restricted medium of expression. The forms it adopts in movement and repose express social pressures in manifold ways. The care that is given to it, in grooming, feeding and therapy, the theories about what it needs in the way of sleep and exercise, about the stages it should go through, the pains it can stand, its span of life, all the cultural categories in which it is perceived must correlate closely with the categories in which society is seen, in so far as these also draw upon the same culturally processed idea of the body' (Douglas, 1996; pg. 69).*

The importance of the body in respect of its physical, social and cultural significance has been highlighted by Douglas (1996). As the quote above illustrates, the body is a medium of expression in which appearance, function and compliance with cultural norms are judged. Disease and illness are not the only aspects of its focus and importance. Patient stories and personal lived experiences are now the focus of healthcare improvement.

The breasts, in particular, play a much larger role in the way women live their lives and the way they are perceived in society than is acknowledged. As a result of the richness of their associated meanings, for example, Douglas (1996) argues that the constituent parts of the body such as the breasts or genitals, are substrates for symbolism and play a part in justifying a position in society as part of a particular group, that of being a 'woman'. A social lived

body, therefore, must be acknowledged as having equal importance to the biological body as together these have consequences for an individual's overall experience of living.

#### **2.4.1 The Significance of the Breasts**

As Dworkin (1974) writes:

*'In our culture, not one part of a woman's body is left untouched, unaltered. From head to toe, every feature of a woman's face every section of her body is subject to modification'* (pg. 113-4).

Thus, breasts are the most visible important symbol of female sexuality and womanliness and a part of a female body subjected to much modification. Sexuality encompasses the whole self, including thoughts, desires, sensations, emotions and identities (Moloney and Kirkman, 2005). Jackson and Scott (2010) also remind us that sexuality is one of the ways in which we live our lives, is a part of everyday ordinary life, incorporating relationships and roles. Yet many health care professionals find it difficult to initiate conversations about sexuality and sexual functioning, believing that they are not trained to do so. This is especially so in the case of caring for cancer patients (Fobair et al., 2006; Hordern and Street, 2007).

Breasts not only provide women with a secure sense of identity and sexual pleasure, but they expose them to risk, uncertainty and fear, with the potential of threatening their existence. It follows therefore that being a real woman

means having two breasts and having a gender identity entails having a body which is not only seen to be feminine (the way it looks), but also dependent upon being a feminine body, for example experiencing a particular type of material embodiment (the way it feels) (Hallowell, 1998, 2000, 2012). Gender refers to the way an individual recognises themselves as either male or female, masculine or feminine (De Francisco and Palzowescki, 2007). Therefore, as the Dworkin quote suggests, women tirelessly work to ensure that they have a body which is feminine in both shape and appearance and which indicates that they are female even if it requires modification with surgery. Undergoing breast augmentation is just one example of the ways in which women seek to achieve this 'normal' looking feminine body. In this way, women are seen to be conforming to the cultural and social norms of being and looking like a woman (Bartky, 1998). Cultural expectations, for example, of what it means to be a woman, require women to make themselves sexually attractive to men, being constantly pressured to look and behave in certain ways and then judged to identify how well they fit with these norms (De Francisco and Palczewski, 2007).

#### **2.4.2 A Symbol of Femininity - The Social Significance of the Breast**

Breasts are also symbols of motherhood and of femininity, factors extremely important to women. Indeed, according to Pelters (2014), breasts are the '*iconic image of femininity*' (pg. 95). They represent the female potential for nurturing in addition to a sexual power and are the most common

representation of femininity. In McEwan's (2011) study, exploring the experiences of thirty-two New Zealand women at high risk of breast and ovarian cancer, the maternal representation of their breasts were privileged over the sexual aspects and the four women who chose risk-reducing mastectomy sacrificed these sexual aspects of femininity for the sake of their children. The symbolic significance and functionality of breasts therefore for individuals and families with a BRCA mutation, needs to be addressed.

Femininity is socially constructed (Wimberley et al., 2005; Everson, 2008) and is achieved from behaviours that make a body recognisable as feminine, such as moving or acting in a feminine way and by disciplining the body. Women constantly strive to achieve the optimum feminine body, shape, size and appearance (Bartky, 1998) and so losing such an organ can result in a crisis in a woman's gendered identity (Sabo et al., 1986; Piot-Ziegler, 2010). Femininity, however, can be experienced differently and queer this normative view. In a paper by Pelters (2014) on BRCA femininities, she presents some of her interview data where she examined the phenomenon of 'doing health' with BRCA positive German women, who construct an alternative and self-determined femininity. One woman for example who had already undergone a breast reduction years earlier to free herself from '*too much femininity*' (pg. 102), was left feeling liberated from the physical symptoms of back and neck problems and this helped to remove the sexually objectifying attention she was receiving from her very large breasts. Thus she was planning a RRM in the future and being '*breastless*' (pg. 102) was not viewed a problem because she

gained agency through her BRCA status and her self-regulating femininity. Her oophorectomy since a BRCA mutation result also freed her from '*way too many female hormones*' (pg. 102) and painful menstruation. For another participant in Pelter's study who chose screening instead of surgery, she also took up Karate in order to feel empowered. Karate represented toughness and a more masculine related femininity. This masculine performance was balanced by her ability to remain looking feminine but it also secured a heterosexual relation. This was done in order to be seen as equal in her relationship with men and not a sexually objectified stereotype that she had relied on in the past. Thus, she had expanded her range of gender expression (Pelters, 2014).

If one accepts that the body is socially constructed (Bartky, 1998; Fox, 2012), the location and the significance that different body parts have for an individual in terms of their identity, will greatly influence the effect on their self-identity. It may be easier for individuals to objectify those body parts such as the ovaries or uterus which are invisible and far less connected to feminine identity, without having an effect on self-identity, because they are perceived as separate, unlike the more visible breasts (Hallowell, 1998). Indeed, in Hallowell's (2000) study involving six case studies of women with a family history of breast and ovarian cancer, some of who pursued prophylactic mastectomy, the self was seen as intrinsically linked to the body and mastectomy was seen to be a major threat to self-identity and femininity. Although the women reduced their risk of the threat of cancer, mastectomy



with reconstruction caused a major change in the way they experienced their body and thus their self-identity, they described themselves as 'incomplete'. Moreover, femininity was seen to incorporate both how the breasts looked to be feminine as well as how breasts felt feminine (Hallowell, 2000). Young (1998) supports this and suggests that what matters the most to women is the feeling in the breast, not the look of them. Thus, even when reconstruction aims to hide the loss from a mastectomy, there is a loss of sensitivity in the breast and the loss of feeling like a woman feels in the world of other women with two breasts (Young, 1990). The consequences of mastectomy for identity and corporality has also been identified by Piot-Zieglar et al., (2010). Although this qualitative study involved women with breast cancer, the aim of this study was to understand the consequences of mastectomy and therefore it is argued that the findings are relevant to women undergoing a mastectomy to reduce their risk. Nineteen women aged between thirty-seven and sixty-two participated in semi-structured interviews in a prospective design. Despite ten women having a reconstruction, the findings demonstrated that mastectomy leads to a painful experience of 'body deconstruction'. Thirteen women described a mutilated body and mastectomy represented an 'amputation', leading to a deep identity crisis which threatened what is socially expected from a woman's appearance, that is, to look feminine. Therefore, mastectomy modified the relationship that women had with their bodies but also their relationships with other people. One of the reasons for this is offered by Weitz (2004), who concludes that:

*‘Those whose bodies differ in some critical way from the norm, must develop a self-concept in the context of a culture that interprets bodily differences as signs of moral as well as physical inferiority. The resulting stigma leads individuals to feel set apart from others’ (pg. 177).*

The significance of the breasts in society has also arisen from the many cultural meanings assigned to the breasts produced in the context of the wider social environment, including gender relationships (Robertson, 2000). It has been suggested that cultural and social factors are responsible for the international differences in the uptake of risk-reducing surgery (Howard et al., 2009). For example, the Ashkenazi Jewish population in Israel, where the incidence of the BRCA mutation is higher (King et al., 2003; Simchoni et al., 2006), view RRM as totally unacceptable (Dagan and Goldblatt, 2009). In the qualitative study by Dagan and Goldblatt (2009) seventeen Israeli BRCA positive women despite fearing breast cancer and ten of the seventeen women undergoing oophorectomy to reduce their risk of ovarian cancer, the decision of risk-reducing mastectomy was out of the question, choosing surveillance instead, fearing the consequences of such surgery on femininity.

## **2.5 Pre-Surgery – Decision Making**

The decision to elect to have RRM is a complex process with many factors that can influence a woman’s decision. These include her relational networks such as the family (especially the age at which family members died), health care professionals, friends and social systems (Donchin, 2000; Lloyd et al.,

2000 Hallowell et al., 2005b; d'Agincourt-Canning, 2006b). McQuirter et al., (2010) conducted a Canadian qualitative study with a convenience sample of ten BRCA women to explore their decision-making with regard to RRM. They concluded that these women did not carry out a rational weighing up of advantages versus disadvantages but rather a complex interplay of contextual factors, for example, personal prior cancer experiences or family members with cancer as well as interpersonal factors such as concerns about body image and support. Findings that support other studies (Beesley et al., 2013; Heiniger, 2015). These findings sit in contrast however to some of the earlier studies. Prevention of breast cancer and survival, for example, took priority over the cosmetic results of surgery in studies conducted by Hopwood (1998); Lodder (1998) and Julian-Reynier et al., (1996).

Decision-making is not separate from the complexities of a woman's life and there are pivotal points when the decision to elect RRM becomes clear. Testing positive for the BRCA gene and having a close family member diagnosed or die with cancer are two examples (McQuirter et al., 2010, McEwan, 2011). Findings that are consistent with other studies (Lodder et al., 2002; Metcalfe et al., 2008, Howard et al., 2009). The systematic review by Howard et al., (2009) reported that Australian and New Zealander women with the mutation were seven times more likely than women without it, to elect RRM and that previous cancer within the family including age at diagnosis strongly influenced decisions. Uyei et al., (2006) also found amongst American women

that having the mutation and the efficacy of the procedure, were the main reasons for electing RRM.

Health care providers may be unsure how to support women's risk-reducing mastectomy decisions and do not always have the time or in some cases, the training to respond to the needs of women electing RRM (McCullum et al., 2007). Indeed, women and their families find it difficult to translate the information they have been given by cancer genetic departments about risk-reducing surgery, when an altered mutation is found (Josephson et al., 2000; Smith et al., 2008; Hamilton et al., 2009). Yet women and families are expected to make life-changing decisions and as a result, many women report a lack of psychological support from different practitioners throughout the journey, especially with the decision to have surgery (Lloyd et al., 2000; Josephson et al., 2000; Payne et al., 2000; Hatcher et al., 2001; Rolnick et al., 2007).

Rolnick et al., in (2007) for example conducted a survey with nine hundred and sixty-seven women at high risk of breast cancer to identify what they wished they had known before their surgery. The findings demonstrated that many of their informational needs had not been addressed prior to making their decision. Most of what they wished they had been told revolved around the reconstruction of the breasts and the expectations of surgery including the look, shape and feel. Two thirds of these women felt unprepared for the changes to their body and wished that they had received more information and

support, especially women undergoing risk-reducing mastectomy, compared to women undergoing contra lateral mastectomy. Many women felt that the need for emotional counselling should not be optional. Although there are limitations to this study in that women completed the survey three to twenty-two years following their mastectomy, many of the women had also had breast cancer and none of the women had a proven BRCA mutation, the response rate was high with 71% responding and it included a substantial number of women from six healthcare systems. Many of the findings have relevance for clinical practice by highlighting the need for practitioners to aid decision-making, offering support before and after surgery. However, a deeper exploration and understanding of what type of support and emotional counselling needs further clarification.

### **2.5.1 The Timing of Surgery**

Studies have identified that women and families find the whole process of decision-making and surgery extremely difficult (Mireskandari et al., 2006; Rolnick 2007; Matloff et al., 2009; Sherman et al., 2010; Mauer et al., 2015). Many women have to construct a 'right time' to undertake the surgery. For example, some women can take as long as nine years following a mutation result before they proceed to surgery, prioritising their decisions in order to consider them one at a time. Howard et al., (2010) in a qualitative study utilising interviews with twenty two BRCA Positive women found that some women do not make immediate decision's about risk-reducing surgery

following genetic testing, taking from three months up to nine years before deciding. Their decisions will depend on many factors such as how much time they have had to think about the implications, where surgery fits in with their normal life, when and if all family conflicts have been dealt with and interestingly when the health service can accommodate them (Howard et al., 2010).

An additional influence on decision-making is the wider impact of a confirmed mutation on the whole family (Howard et al., 2010). Those with young children or whose relatives have developed or died of cancer will take far less time to decide on surgery because of the fear of leaving children behind (Lodder et al., 2002; Van Dijk et al., 2008; Haroun et al., 2011). They have very often decided on risk-reducing surgery even before the test or the result (Julian-Reynier, 2010).

Decisions about surgery may also shift over time and in response to receiving relevant and adequate information. McCullum et al., (2007) explored three women's decision-making experiences and outcomes using case studies, all of whom were BRCA positive and who made different decisions, with regard to RRM. They were selected in order to demonstrate the complexity of the process of decision making, showing how women deliberated and revisited their decision's over time. The study employed a mixed methods approach using standardised measurements in a questionnaire and open-ended interview questions. Participants received a booklet describing RRM that was

developed by the research team and which included benefits and risks to RRM. The women were followed up at three and six months after the booklet was given. Initially, all three women wanted to have RRM but after receiving the booklet, each woman reported a different decision. Case one decided against RRM, explaining that it was *'too radical'*, case two remained undecided and with time wanted more information to be able to make a final decision and case three had undergone a RRM by the time of her second follow up interview. Findings illustrate the complexities of decision-making, how they may shift over time and the need for sufficient information and support to assist in the process.

### **2.5.2 The Decision to Elect Breast Reconstruction Following RRM**

Compared to younger women with breast cancer who often choose not to reconstruct their breast (Holland et al., 2014), most women who have a BRCA mutation, take up this treatment option (Frost, 2000; Evans et al., 2005; Wasteson et al., 2011). Although there have been some reports of satisfaction with the way the breasts look following RRM and reconstruction, (McGaughey, 2006; Spear, 2008; Wasteson et al., 2011), there have been many reports of dissatisfaction, loss, (Altschuler et al., 2008; Gahm et al., 2010; Gopie et al., 2013) and some regret (Frost, 2000; Payne et al., 2000). Even though breast reconstruction has improved in both the technique and its availability, satisfaction with the aesthetic result amongst BRCA positive women is still varied and has been reported to range between just 40% and 60% satisfaction

(Metcalfe et al., 2004b; Bresser et al., 2006; Isern et al., 2008; Gahm et al., 2010; Wasteson, 2011). Moreover, in BRCA women, in particular, evidence suggests that the cosmetic expectations of surgery are very high and often not achieved. Post-surgery, much lower satisfaction rates are reported when compared to women who have had breast cancer (Gahm et al., 2010; Brandberg et al., 2012; Hallowell et al., 2012). The underlying factors for this lower satisfaction needs further exploration.

## **2.6 Post-surgical Impact**

The literature reviewed highlighted four overarching themes following risk-reducing mastectomy relevant to this study; regret following surgery, the impact of surgery on body image and sexuality, experiences of the partner and the long-term quality of life. Due to the importance of the themes identified for clinical practice, each will be examined in turn, with a number of these studies presented and critiqued in detail.

### **2.6.1 Regret Following Surgery**

Anxiety about cancer risk is not always reduced following RRM (Watson et al., 2004) and there has been some reports of regret (Borgen et al., 1998; Frost et al., 2000; Payne et al., 2000). A retrospective study by Payne et al., (2000) specifically explored regret following bilateral mastectomy with twenty-one women who had completed an initial questionnaire as part of enrolment into a national prophylactic mastectomy registry and who had reported significant



regret in having the surgery. Nineteen women agreed to have a telephone interview; an experienced psychologist and psychiatrist conducted the structured clinical interview, which focused on the decision of RRM. Fourteen women had undergone reconstruction with implants. The physical and psychological trauma, in addition to the lack of psychological support, were found to be significant reasons for regret although the exact nature and extent of the trauma is unclear. Regret also related to poor cosmetic results, complications of the surgery and the effects on body image and sexuality. The exact nature of these problems were not reported, however.

Although this study offers some insight into the experience of RRM, there are many limitations, none of the women were BRCA positive and all the women had a varied and elevated perception of risk. They reported a general overall fear of breast cancer based on what they had read, which did not reduce post-surgery. The sample were self-selected, resulting in a biased representation (Flick, 2014). Furthermore, the physician had initiated the discussion and decision of RRM rather than the women themselves which, evidence demonstrates, leads to higher levels of dissatisfaction and regret (Borgen et al., 1998; Frost et al., 2000). Using a clinical interview as opposed to a qualitative interview was also a limitation and as such, there are no quotations from the participants to aid trustworthiness or establish rigour. Importantly, had the women received more information or support about the consequences of surgery before making the decision, the majority reported that they would have reconsidered their decision.

These findings concur with the study by Borgen et al., 1998, who also found significant regret fifteen years following surgery, especially in women whose physician had initiated the surgery. However, this study is now over fifteen years old and since the introduction of protocols and shared decision-making, a different experience may be found. Furthermore, the sample relied on volunteers who answered an advertisement in a magazine and who completed a questionnaire. The selected sample may have included women with more positive or negative experiences, thus the actual degree of regret is difficult to determine. The sample did not include any BRCA positive women and many had undergone reconstruction, this results in many limitations of the study in its relevance to this study. Frost et al., (2000) also found that 18% of women in their long-term study were dissatisfied with the outcome and on reflection, would not choose to have the surgery again, implying a degree of regret.

A more recent retrospective Swedish study by Gahm et al., (2010) that utilised questionnaires to analyse the impact of RRM and reconstruction performed by a plastic surgeon, found that although the fifty-nine women did not explicitly report regret in having the surgery, ten women expressed that it was not the right decision and was not wise to have had the surgery, again implying a degree of regret. Acknowledging the limitations of this study in that it was a retrospective study using a quantitative approach two years post-surgery, the findings offer insights into the many complications and physical effects of RRM. Thus, the findings have implications for clinical practice that can inform

practitioners who care for these patients and women who are considering such surgery.

### **2.6.2 The Impact on Body Image**

There have been three prospective (Brandberg et al., 2008, 2012; Gopie et al., 2013) and two retrospective studies conducted (Hopwood et al., 2000; Metcalfe et al., 2004a) that provide insight into the experiences of women following RRM but they specifically focus on cosmetic result and the impact on body image. These studies demonstrate mixed findings, some report a negative impact on body image, whilst others report satisfaction. These studies utilised questionnaires that focused on pre-determined outcomes such as body image, cosmetic result or mental health and included BRCA positive and negative patients, which have limitations in its relevance to this study because the risks, circumstances and experience will be different for those who do not have a mutation.

The study by Brandberg et al., (2008) was prospective which is its strength, it assessed psychological reactions and body image following RRM and reconstruction. Well-validated questionnaires, with ninety women of whom fifty were BRCA positive, were completed and the response rate was 90%. However, attrition increased over time with only eight women completing the questionnaire at twelve months and the power to reach statistical significance was low and therefore a limitation. Women were seen by a psychologist

pre-operatively and overall the majority of women were satisfied with the cosmetic result, however, it is unclear what percentage of these satisfied women were BRCA positive. It is also unclear what type of reconstruction was performed, important factors that determine the overall cosmetic result.

Although anxiety levels decreased post-surgery, consistent with other studies (McGaughey, 2006; Hallowell et al., 2012; Gopie, 2013), there was no difference in depression scores. Importantly 50% of women reported problems one-year post surgery with 48% experiencing body image and sexual problems, sexual pleasure was rated lower and women felt less attractive and more self-conscious. Findings that concur with Lodder, et al., (2002). It is worth noting, however, that 25% of the women in Brandberg et al's (2008) study had undergone prophylactic oophorectomy, which could also have accounted for their experiences especially the effects on body image and sexuality. Although 61% reported a positive change in their life, 46% reported a negative impact on intimate situations and femininity.

One explanation for the contradictory findings of having a positive life change yet negative impacts on aspects of life is that the questionnaire was unspecific. One of the many limitations of this study is that cancer specific distress was not measured, this is important when studying women with the BRCA mutation because many are electing surgery in order to reduce their distress. Furthermore, many of the participants were not BRCA positive which affects the relevance of the findings to the current study, family members were not

included and pre-determined specific aspects of the experience were measured.

Cosmetic results and satisfaction with surgery were again the focus in Brandberg et al's (2012) study with the aim of assessing expectations before and after surgery. The study utilised questionnaires six months and one year following bilateral mastectomy and reconstruction. Forty-nine of the ninety-one participants were BRCA positive and measurements of cosmetic satisfaction, sexual activity and body image were conducted using instruments designed for cancer patients, a different group who would have very different experiences and needs. Although the majority of women reported being satisfied with the cosmetic result, a larger proportion of those with a BRCA mutation, reported unmet expectations with over 40% complaining about the feel and the look of their breasts. The prospective nature of the study is a strength but further exploration and a deeper understanding of what the expectations were and why expectations were not met in the BRCA positive women, in particular, would have increased knowledge, however, this exploration was limited by the use of questionnaires.

One would have expected that with the inclusion of a plastic surgeon, a nurse and a psychologist as part of a recommended multidisciplinary team approach, that both groups of women would have had similar expectations. Women had also undergone oophorectomy and the NAC had been preserved in the women who underwent reconstruction, which would have given a superior cosmetic

result, all factors which affect expectations. The inclusion of BRCA negative women would have affected the results because their experience and risk is different, resulting in some limitations of the study in terms of the relevance to the current study.

Similarly, Gopie (2013) specifically explored the effects on body image following RRM, using questionnaires with fifty women of which forty-four were BRCA positive and four women had a strong family history. Although fifty women initially consented to the study, two were later excluded as breast cancer was found in the surgical specimen, seven dropped out and nine did not respond. Having a heterogeneous group of women especially those with breast cancer, unspecified high risk and BRCA positive, one would view the results and therefore the relevance to this study, with some caution because of the difference in sample, circumstances and therefore experiences. The prospective design of this study is a strength with questionnaires completed pre-surgery, six months post-surgery and following completion of the reconstruction, with a median of twenty-one months. However, the techniques of reconstruction varied amongst the women, a factor known that affects different satisfaction and so the results were again, not comparable.

There was a high attrition rate, one of the limitations of surveys with only thirty-two completing the final questionnaire making the sample low and results less convincing. The study aimed to assess partner satisfaction with relationships but it appears that partners were not included and results relied

on the women to comment on this aspect, questioning the trustworthiness and rigour of the effects on partner relationship measurements. The women had undergone oophorectomy, which would also have affected their body image scores. Body image and sexual relationship satisfaction was found to be significantly decreased and remained unchanged for 30% of the participants, although no sexual dysfunction scale was utilised. Results demonstrated that 29% of the women were unhappy with their breast appearance and 21% felt embarrassed with their naked body. However, the lack of body image data in a comparable age matched healthy group makes it difficult to compare how these differ from normal scores. Moreover, the ability to gain a deeper exploration of their dissatisfaction and their embarrassment is one of the limitations of utilising a questionnaire.

Conversely, the retrospective Canadian study by Metcalfe et al., in (2004a), suggested that the majority of women were happy with their decision to undergo surgery although younger women less so. Their study utilised questionnaires with seventy-five women to determine the psycho-social functioning following prophylactic mastectomy. Of the sixty women who returned their questionnaire, only thirteen women had a BRCA mutation, making it difficult to draw comparisons with a heterogeneous group. A mean of five years had elapsed since surgery and thirty-eight of the women had undergone reconstruction with seven women having a subcutaneous mastectomy.

Most women without a mutation were not experiencing any abnormal levels of distress nor any major problems with body image or sexual activity. A third of the women with a BRCA mutation, however, were still experiencing high levels of both general and cancer related distress, but it is unclear why. The limitations of the study in terms of the relevance to this study, include its retrospective design as women were expected to remember how they felt five years previously, the inability to explore the reasons why cancer related distress was still evident especially in the BRCA positive women or why younger patients, in particular, were less satisfied. Moreover, the small sample size, the fact that women had different types of mastectomy including subcutaneous mastectomy (where the skin and the nipple are preserved), would also have affected satisfaction and are therefore limitations. There were no pre-operative measures of anxiety or quality of life to compare with post-surgery, which would have increased knowledge, had relevance for practice and contributed to the rigour of the study.

In support of this, Hopwood et al., in (2000) in Manchester UK, utilised questionnaires in a retrospective design with a 79% response, to assess the mental health and body image of seventy-six women, up to three years post RRM. Overall, the findings showed no evidence of significant mental health or body image problems but women felt less attractive, less feminine and there were negative effects on sexual relationships, the reasons for which were not reported. It is unclear from the findings what the 'serious psychological' or 'body image problems' were and how many women experienced them. The



women in this study were treated within the only known protocol at that time, for high-risk women in the UK and received two genetic counselling sessions, a psychological assessment, surgical consultation pre-operatively and an annual follow up. Interviews with the psychiatrist were conducted only when body image scores or general health scores were high at follow up.

The interview in this sense had a different function to that of a research interview, a therapeutic interview with the aim of treating psychological problems. There were only six BRCA positive women in the sample and three of these had breast cancer, the women underwent different types of reconstruction or no reconstruction, resulting in many limitations of the relevance to this study. The focus of their study was on pre-determined outcomes, rather than overall experience and the authors conclude themselves that it was not a systematic research evaluation and a baseline questionnaire was not routinely collected, with some reports of missing data.

### **2.6.3 The Longer-Term Impact on Body Image**

The following studies aimed to explore the psychosocial consequences of being BRCA positive and electing to have surgery in the longer term. They are both retrospective and prospective with changes in body image being the focus.

In contrast to the previous studies, van Oostrum et al., (2003) used a mixed methods approach that aimed to explore the long-term psycho-social consequences of being BRCA positive (twenty-three participants) and BRCA negative women (forty two participants) five years after the test in order to identify risk factors for long-term distress. Sixty-five women completed a questionnaire and fifty-one were interviewed (twenty positives and thirty-one negatives). More women with a mutation underwent RRM and reconstruction (twenty-one out of twenty-three) and had a less favourable body image with 70% reporting changes in sexual relationships. Of particular note is that 50% of the mutation positive women consulted a psychiatrist or psychologist for support during the five years following testing, potentially highlighting the difficulties experienced by those with a mutation. Long term distress was associated with higher risk perception pre-surgery, the loss of family members to cancer and less open communication within the family.

Well validated assessment tools were used to measure body image, anxiety and depression but questionnaires did not allow a further in-depth exploration of the experience. Women had also undergone bilateral oophorectomy, factors that could have affected the experience, and two of the women with a mutation were diagnosed with breast cancer during the study. Anxiety and depression scores between years one and five were increased for both mutation positive and negative women, suggesting that factors other than testing and RRM affect the long-term distress noted.

Limitations of this study include its retrospective design and the quantifiable measurement of outcomes as opposed to rich, qualitative data examining experiences, for example, there was no data from the interviews nor any quotations from participants to draw any conclusions about the experience. The inclusion of women with breast cancer whose issues will be different (Metcalfe et al., 2005) and women who had undergone oophorectomy in addition to a 24% dropout rate, were also limitations to increasing knowledge relevant to this study.

Similarly, the study by Altschuler et al., (2008) was retrospective, and examined the long term psychosocial experiences of bilateral mastectomy but also included women with breast cancer having a contra lateral (opposite breast to breast cancer) mastectomy. A total of nine hundred and sixty-seven women were sent a survey, three to twenty-two years following a mastectomy with a response rate of 71%. One hundred and ninety-five women had RRM and seven hundred and seventy-two had a contra lateral-mastectomy. The qualitative aspect of the survey used open-ended questions which were coded independently by three of the authors, this ensured trustworthiness and an audit trail. Compared to women having contra-lateral mastectomy there were over twice as many negative responses from the women who had undergone RRM. Reports of decreased sensation in the breast, body image with sexual problems and chronic pain, were all reported.

The limitations of this study include the long and varied time points since surgery in terms of expecting the women to recall their feelings or experience after twenty-two years, the majority of women had breast cancer and more importantly, it appears that none of the participants were BRCA positive, so it is difficult to make comparisons. Although direct quotes from the participants were taken from the open-ended responses, it is difficult to interpret the meaning or explore the responses. There were contradictions from the responses, for example, some women reported satisfaction in the closed ended section of the questionnaire, but in the open-ended section they reported negative psychosocial outcomes. It is also unclear if any of the women had reconstruction and if so, what type, as this would have affected the experiences.

Conversely and importantly, the majority of women in den Heijer et al's (2012) prospective long-term study were BRCA positive, offering results more relevant to this study. They explored the psychological distress following prophylactic mastectomy six to nine years post-surgery in thirty-six high-risk women who had undergone mastectomy with reconstruction. Questionnaires were completed shortly after consent for the study in (2003), pre-surgery, six months and finally at six to nine years post-surgery with all of the BRCA positive women completing the questionnaires at nine years. Well validated questionnaires were used to specifically identify anxiety and depression, body image scores and impact of events. Results showed a decrease in cancer specific distress and general distress long term. Findings consistent with other

studies (McGaughey, 2006; Hallowell et al., 2012; Hagen et al., 2014; Glassey et al., 2016).

Body image problems were still evident, however, at six months and in the long term. One explanation for the findings at six months is that reconstruction was incomplete as many women were still waiting for surgery. However, women were still dissatisfied and reported problems at nine years when surgery was completed. Women with body image problems pre-surgery were more vulnerable to having body image problems post-surgery, social support and having an active coping style was found to increase body satisfaction long term, important factors that have relevance for clinical practice.

Although this study is prospective and therefore, its strength, there is a long-time interval between the questionnaires given at six months and again at nine years. The expectation that women could accurately report how they felt in the previous nine years questions its trustworthiness. Furthermore, the outcome measures were pre-determined, specific for body image and psychological distress.

Although these measures are aspects of experience, the opportunity to explore the experience further is lost when a survey design is utilised. Within the sample, women had undergone both mastectomy and oophorectomy and this would be captured in the measurements. It also included women with breast and ovarian cancer, who would have different experiences and

concerns. Finally, the women had different reconstruction and not all the participants were BRCA positive, factors, which contribute further to some limitations of the findings.

#### **2.6.4 The Impact on Sexuality**

Sexuality has consistently been shown to be adversely affected following RRM, but in some studies without adequate explanation and with some contradictions. Importantly, both quantitative and qualitative studies have been conducted. Hatcher and Fallowfield, (2003) conducted a qualitative prospective study as part of a larger quantitative study by Hatcher et al., (2001) in the UK, where questionnaires with one hundred and fifty-four women were utilised in order to measure psychological and sexual morbidity following prophylactic mastectomy. Seventy-nine women chose surgery, sixty-four declined and eleven deferred making a decision. Findings demonstrated that overall, there were no significant changes in sexual pleasure post-surgery. However, these women were a heterogeneous group, not all women underwent reconstruction and those that did, had different techniques, important factors that could affect sexuality. Some of the women also had breast cancer, which would have had an influence on their satisfaction. For the qualitative aspect of this study, in depth interviews were conducted at home pre-surgery and again at six and eighteen months with sixty women who underwent surgery. Interviews were conducted pre-surgery and at eighteen months with the twenty women who declined surgery.

The aim was to identify the psychosocial implications of the surgery. Although anxiety was reduced post-surgery, the majority of women were not prepared for many of the effects of surgery. Findings consistent with Gopie et al., (2013) and den Heijer et al., (2012). Some women experienced pain, dissatisfaction with their sexual relationships and dissatisfaction with their reconstruction, with complications still being reported at eighteen months. Most of the women did not see any photographs of surgery, they had not received any information pre-surgery from a surgeon or any nursing staff, thus support and information were requested. No difference in sexual pleasure was noted which sits in contrast to other studies (Frost et al., 2000; Lodder et al., 2002; Bresser et al., 2006; Brandberg et al., 2008).

However, there were some contradictions in the findings. A third of women reported that their husbands did not want to touch their breasts and their sexual relationships were adversely affected. There were also limitations to this study, only eight of the women were BRCA positive and a proportion did not have surgery, making it difficult to compare the groups. Although it is a qualitative study and there is some explanation about the process of data analysis, it is unclear what the underpinning methodology was which questions the trustworthiness and rigour of the study. The study included BRCA positive and negative women in addition to patients with breast cancer, which is also a limitation because they are different groups of women with different circumstances and needs.

Bresser et al., in (2006) found adverse experiences in sexual relationships with altered feelings of femininity for over half of the participants in their study and they reported that were more likely not to choose the procedure again. It was a retrospective study using questionnaires three years following surgery with one hundred and fourteen women, sixty-three of whom were BRCA positive. The women underwent RRM or contralateral mastectomy with reconstruction and fifteen had previous breast cancer. Although there are limitations in this study in that it was retrospective and included a heterogenous group of women such as those with cancer, it does demonstrate the impact of RRM on femininity and thus sexual relationships. A deeper exploration into these findings with a qualitative design would add further knowledge.

Few pure qualitative studies have been conducted with BRCA positive women without cancer, specifically undergoing risk-reducing mastectomy but Nina Hallowell from the UK has studied high risk women extensively. In the most recent study, Hallowell et al., (2012) utilised a qualitative design, whose aim was to identify and understand the women's experiences with no fixed, forced questions. Semi-structured telephone interviews were conducted with forty Australian women three years after undergoing risk-reducing surgery. Twenty-five of the sample had a BRCA positive mutation and nineteen of the twenty-one women who had RRM mastectomy underwent reconstruction. Two themes of looking different and feeling different were developed from the data. Although the surgery was seen as positive in reducing risk and worry, as found in other studies (Frost et al., 2000; van Oostrum, 2003; Dowdy et al., 2004;



Brandberg et al., 2008; Miller et al., 2010), many negative consequences of surgery were reported. These effects had an impact on the women's sexuality and have important relevance for clinical practice, highlighting a need for those involved in their care to identify and address the issues, for example, the women reported being unprepared for many of the consequences of surgery including the altered sensations in the breast.

The majority of women were BRCA positive, this is a strength of the study, comparing a comparative group, but there were some limitations to consider. Women in Hallowell's study also underwent risk-reducing oophorectomy either alone or in combination with breast surgery and this would have affected their experience, especially their sexuality because of the implications of menopause on sexuality. The type of reconstruction that women underwent also differed which would have affected the satisfaction with their cosmetic result. Women who had implants, for example, described their breasts as feeling hard and different whereas women who had muscle flap surgery described how they were able to have a 'tummy tuck', which was seen as a bonus. The retrospective nature of the study is also a limitation because participants were trying to recall how they felt three years previously. The fact that they were all interviewed after the same three-year period, however, is a strength.

The qualitative study by McEwan, (2011) also has relevance to this study and for clinical practice. The experiences of thirty-two New Zealand women living

with an increased risk of breast and ovarian cancer were explored as part of a Ph.D., with the author working as both a practitioner and a researcher. The study investigated the impact on body image and sexuality following risk-reducing surgery by removing significant female organs associated with sexuality and femininity, the breasts and ovaries. Semi structured interviews were conducted and a narrative thematic analysis resulted in a core category 'getting on with it'. This was found to be a dominant theme in the way women approached their risk and their decision to have surgery. Nineteen women had undergone surgery and although the majority of these women had elected to undergo RRSO and therefore the experiences of an altered femininity and sexuality were mainly related to these individuals, only four women had elected RRM. 'Getting on with it' proved more difficult for some of the women in sacrificing the sexual aspects of femininity for the maternal representation of their breasts and the gendered representation of women as nurturers. Many of the women, for example, expressed a desire to 'be there for their children' as the main reason for their decision. Many women who had not had surgery, however, identified worries about RRM on body image and sexuality. For those who had undergone surgery, loss, changes to body image and sexual problems had been experienced mainly by women who had RRSO. Pressure from husbands to undergo some form of breast reconstruction to replace the loss was also reported in this study, demonstrating the importance of a woman's breasts for sexuality. The strengths of the study include that sixteen of the women were BRCA positive and the sample included two lesbian women who are not represented in the majority of studies on high-risk women

and whose breasts were equally as important to those in a heterosexual relationship. Five of the women in this study had breast cancer which would have affected their experience especially 'loss' and is one of its limitations. Further exploration of women's sexuality problems with regard to risk-reducing mastectomy in a prospective study with a larger sample size would add further knowledge.

Adding to current knowledge, similarly, Lloyd et al., (2000) conducted a grounded theory study three years post-surgery, the aim of which was to explore the personal experiences of surgery and psychological adjustment for the woman and her partner. Ten women and eight men were included, using semi structured interviews with a purposive sample. The time since surgery ranged from six weeks to three years and nine women had implant based reconstruction. Sexuality and feelings of womanliness had been adversely affected, although further exploration of the reasons why and how would have increased knowledge and informed practice.

The aim of grounded theory is to develop a theory that is grounded in the analysis (Flick, 2014) and thus a core category of 'suffering and countering multiple losses' was found to be central to their experiences, the loss of femininity and womanliness as well as the loss of close members of the family. However, the studies limitations include its retrospective design, only two women were BRCA positive and three of the ten women had also undergone oophorectomy which would have affected the results. Furthermore, all the

women in the sample were married; single women may have had a different experience. Participants waiting for surgery or in the decision-making process could have been asked to participate in this study and was also recognised as a limitation by the authors.

### **2.6.5 The Experience of Partners Following Risk-Reducing Mastectomy**

Currently, only three studies include the partners' experience following RRM in women known to be BRCA positive (Lloyd et al., 2000; Mireskandari et al., (2006); Mauer et al., 2015).

In the retrospective qualitative study by Lloyd et al., (2000), although sexuality was not explored specifically, the majority of the eight men felt surgery had no negative consequences on their sexual relationship. However, the men did report that their sexual relationships had deteriorated, resulting in some contradictions. There were only two quotes from the partners to validate any findings and aid trustworthiness and therefore, there were some limitations of the findings. Interviews were conducted between four months and three years' post-surgery and the aim was to determine the effect of surgery on their relationship and family life. Although partners did not feel that their relationship was negatively affected by surgery, the cosmetic result was described as '*shocking*' and they reported a need to '*adjust*' to it. It is unclear however what the shock entailed and in what way the sexual relationship had been affected or what they had to adjust to. Partners also described a strain on the relationship due to the extra work that had to be undertaken over the

post-operative period in trying to work yet simultaneously care for their children. These factors have importance for clinical practice in meeting the needs of the whole family and thus a further exploration of the deeper issues would add to the current knowledge.

Similarly, Mauer et al., (2015) identified problems with sexuality following RRM. An online retrospective survey was conducted with twenty-five male partners of women who were BRCA positive. Women were asked to forward the link to the study to their partner and the link was posted on a forum notice board. Eleven of the women had undergone mastectomy and ten had reconstruction, seven women had also undergone bilateral oophorectomy.

The focus of the study was on male partners' thoughts about changes in sexuality and thoughts about prophylactic surgery. Fourteen women had not undergone a mastectomy at the time of the study but when asked if they would want the women (partner) to have reconstruction after a mastectomy, the majority of the men said 'yes'. Although the results concluded that this sample of men was still attracted to their partner who had undergone mastectomy, there were problems and concerns with breast sensation loss, sexual problems and physical attractiveness.

The information was limited however and it is unclear precisely what the concerns were. It was not possible to gain a deeper understanding of the experience due to the limits of using a survey design. Women had also

undergone oophorectomy and many of the concerns regarding sexual problems may have been linked to this and menopausal symptoms. The sample size was small and it focused on men currently in relationships who were recruited via their partners. The results may have been very different if the men were single and had been contacted directly.

The study by Mireskandari et al., (2006) was a qualitative retrospective study, this aimed to explore the needs and concerns of partners of women at high risk of breast cancer. Fifteen male partners were interviewed by telephone, only seven had a partner who was BRCA positive and only one woman had undergone RRM compared to four women who had undergone bilateral oophorectomy. This latter surgery may have been responsible for the majority of relationship problems reported. Data analysis was conducted using methods described by Miles and Huberman and this is clearly documented, thereby aiding trustworthiness and an audit trail with a number of participant quotes. Relationship problems, greater distress and adjustment problems were reported in more of the men where there was a BRCA mutation. However, it is unclear what these relationship, distress or adjustment problems were.

Findings demonstrated that partners identified RRM as one of the most challenging decisions they had to make but there was no further exploration. They reported a need to meet other partners in the same situation as well as a need for information and support from health professionals, the reasons for

which were not explored. The time since testing for those who were positive ranged from one to sixty-six months and this was not accounted for in the analysis, which is another limitation. This variation in time would have affected the experience because the partners may have adjusted to the situation if interviewed at three years, as opposed to one year.

### **2.6.6 Summary of Literature on Body Image and Sexuality**

Although there have been some reports of satisfaction following RRM, overall the studies demonstrate a number of problems following RRM in terms of the negative effects on body image, identity and sexuality, for both the women and their partners. A literature review conducted by McGaughey (2006) summarises the research on body image specifically following RRM, concluding that 50% of women undergoing RRM suffer negative effects on body image, however, the exact reasons for which need further exploration and understanding. In some cases, there have been reports of regret, especially where there has been no support for women and the family or where the decision to elect RRM has been doctor led as opposed to being led by the women themselves. Following RRM, there is less anxiety and fear of developing breast cancer overall, but these studies demonstrate that there is still a surprisingly high level of anxiety and distress experienced by some women with a BRCA mutation and their partners post-surgery. This knowledge alongside the many negative impacts with identity and sexuality for BRCA women, in particular require further exploration.

## **2.8 Long-Term Impacts of Risk-Reducing Mastectomy**

The remaining literature considers the long-term effects and impact of risk-reducing mastectomy on the quality of life. Three retrospective studies utilised questionnaires and one qualitative study that also considered family members.

### **2.8.1 Quality of Life**

The assessment of Quality of life (QOL) has also been the focus of a few studies post RRM. Three such long-term retrospective studies utilised questionnaires. A descriptive study by Frost et al., in (2000) involved five hundred and seventy-two women who had undergone RRM but none of the women had a BRCA mutation. The main outcome measures were satisfaction with surgery and psychological quality of life although it is unclear which validated questionnaire was utilised. Time from surgery to the questionnaire was fourteen years and findings demonstrated that post-surgery, women had less concern about developing breast cancer, findings that are consistent with other studies (van Oostrum, 2003; Dowdy et al., 2004; Brandberg et al., 2008; Miller et al., 2010) and most women were satisfied with the procedure.

However, there were many negative consequences of surgery, problems with sexual relationships, feeling less feminine, lower self-esteem and 36% had a diminished overall satisfaction with their body, with 18% of women who would not choose to have the surgery if given the chance again. Women who reported that they had undergone surgery based on the advice of their



physician were the most dissatisfied, with adverse effects found in emotional stability and quality of life. Seven women developed breast cancer during the study and this is a limitation of the study because they were included in the analysis and their circumstances and experiences would be different. There was a fourteen-year time lapse since surgery and therefore a problem of recall bias is a consideration. Women had undergone different types of reconstruction or no reconstruction and this would have affected cosmetic satisfaction. Indeed, women who did not have reconstruction were the most satisfied. A deeper exploration of the reasons for dissatisfaction and reasons for low self-esteem and sexual problems would have added further knowledge.

Similarly, the study by Geiger et al., (2007) specifically looked at the quality of life after RRM for high risk women who had surgery between 1979 and 1999 but who did not have a BRCA mutation. This is important when considering the results because the needs of high-risk women will be different to those carrying a mutation. Three hundred and twelve women were emailed a questionnaire of which a hundred and seventeen were women with a high risk but who did not have surgery. These women had ovarian cancer and early changes within the breast thus not a comparative group. The response rate was poor with only a 58% response (one hundred and six who had surgery). Diminished quality of life was associated with dissatisfaction with sex life prior to the surgery and this was also evident in women who did not undergo surgery.

Although this is a long-term study, asking women to recall their feelings or experiences from twenty-three years prior is a major limitation and a consideration toward the trustworthiness of the findings. In addition, the accurate risk status of the women in the comparative group is unclear, an important factor that would affect QOL. Furthermore, without pre-mastectomy assessment of the quality of life or psychosocial factors, it is difficult to identify whether the two groups of women differed systematically before the procedures and if so, how much this would have affected the results. It is also unclear whether the comparison group were on any chemotherapy-prevention programmes or surveillance programmes due to their high risk, which again would have affected the results. The inclusion of women with ovarian cancer and early breast changes is also a limitation of the study because the difference in circumstances makes it difficult to compare to high risk women and BRCA positive women in particular.

Metcalfe et al., (2005) also report on the overall quality of life following RRM as part of a larger study assessing psycho-social functioning following RRM (Metcalfe et al., 2004a). This QOL study however specifically assessed the predictors of QOL with seventy five women age range from twenty to sixty-two of whom sixty returned the completed questionnaires. A mean of fifty-two months had elapsed since RRM and completion of the questionnaire, thirteen women were BRCA positive. The levels of QOL for the study participants were found to be slightly above the average levels for the normal population and higher than the mean levels reported for women with breast cancer.

Importantly, two significant predictors of QOL were found, vulnerability and psychological distress. Those found to have higher levels of vulnerability were more likely to experience a lower level of QOL. Vulnerability included feelings of susceptibility to an invasion of the body by cancer and thus a loss of trust in the body to stay healthy. Psychological distress such as continuing to perceive high breast cancer risk predicted lower quality of life. These findings offer important insights for members of the multidisciplinary team who care for women who elect RRM by providing understandings of how the overall psychological health of women can be promoted. QOL was measured after a period of adjustment possibly accounting for the above average scores seen. Had QOL been measured immediately following RRM for example, transient effects may have been observed.

Wasteson et al., (2011) has carried out the only study examining long-term satisfaction following bilateral mastectomy that includes perspectives of the family. This was the long-term study building on an earlier study (Josephson et al., 2000). Semi-structured interviews were conducted in a qualitative design with thirteen women who also took part in the earlier study, only one of whom was BRCA positive. All participants had undergone reconstruction using implants and three main areas were explored, daily activities, risk perception and cosmetic results.

Although all women reported satisfaction with their decision to have surgery and there was a reduction in their risk, negative consequences of surgery were

reported mainly due to a change in the women's bodies which affected relationships. Family relationships had been adversely affected especially with regard to their spouse and for many, there were ongoing complications from surgery, which necessitated further surgery. There are a few limitations to this study that need consideration. The retrospective design would have affected the ability to recall experience ten years earlier as there may have been many other factors prior to the study that influenced their overall experience. Only one woman had a BRCA mutation, which may have affected the satisfaction rates as BRCA positive women appear to be less satisfied following RRM and have different concerns and experiences (van Oostrum et al., 2003; Gopie, et al., 2013; Heijer, et al., 2012). The underlying methodology in the study is unclear and there are no quotations from participants to support trustworthiness in the analysis, auditability or rigour. In addition, there were no assessments of psychological measurements using any well validated assessment tools either pre or post operatively.

### **2.8.2 A Summary of the Review**

The focus of the literature examining the experience of women with BRCA1 and 2 has primarily concentrated on using 'satisfaction' and 'body image' as key outcome measures following breast removal rather than overall experience. Satisfaction with surgery overall was found to be variable with many studies adopting quantitative methodologies. Some studies were predominantly conducted before any clinical protocols were established and

across a wide range in time points following the surgical intervention. Thus, asking women to recall how they felt many years after the experience, results in the problem of recall or response bias (Borgen et al., 1998; Frost et al., 2000; Hatcher et al., 2001; Hatcher and Fallofield, 2003; Bresser et al., 2006; McGaughey 2006; Brandberg et al., 2008; Gahm et al., 2010).

Only seven studies addressed some of the longer term effects and again, these primarily concentrated on using specific outcome measurements associated with the surgery, included women of different risk categories but did not include the experiences of their wider family (Frost, 2000; van Oostrum et al., 2003; Metcalfe et al., 2005, Geiger et al., 2007; Altschuler et al., 2008; Wasteson, 2011 and den Heijer et al., 2012). Only six studies have used qualitative methods to examine the experiences (Lloyd et al 2000; Hatcher and Fallowfield, 2003; Mireskandari et al., 2006; Howard et al., 2011; McEwan, 2011; Hallowell et al., 2012).

The majority of the studies were retrospective and included BRCA positive women and women with breast cancer, different categories of high risk or women who had undergone contra-lateral mastectomy as a risk-reducing procedure. This makes it difficult to identify the experiences of BRCA positive women alone. In addition, the studies included women who underwent different reconstruction techniques at different time points and women who did not have reconstruction. Thus, these studies focus on a very small part of the experiences of a heterogeneous group of women. This makes overall

comparison difficult. However, the review does offer valuable insight and knowledge in terms of identifying some of the impacts of RRM on body image, sexuality and quality of life for women with and without the BRCA mutation, leaving scope for further research with BRCA positive women specifically, including their family.

Many of the studies used structured questions, this can mean that the results reflect the researcher's focus, rather than the focus of understanding the experiences of participants. The use of assessment tools that have not been well validated in some of the studies also raises issues of credibility and validity. The sample size varies within studies and the methodology in some of the studies is unclear. Only three qualitative studies have included the partners' experiences, these were retrospective and the data is limited. Many of the studies, therefore, lack a deeper understanding of the experience of a BRCA positive woman, in particular, electing RRM in the context of her wider family. The family perspective, especially those of the partner, is important for clinical practice as the decision to elect RRM and to maintain a positive mental health post-surgery, is a family matter and research with this particular group is required to increase knowledge and understandings.

None of the studies identified in this or the previous chapter offered a deeper examination of the experiences of body image, sexuality and quality of life and thus how practitioners can support women and their families. Finally, there were no studies found that adopted a phenomenological approach, in

particular, an interpretive design utilising Gadamerian philosophy, that explores the phenomenon of living with the BRCA mutation and electing RRM.

### **2.8.3 Conclusion**

The current horizon of understanding, therefore, highlights many limitations, some contradictions and some gaps in the literature. A table of the included studies is summarised in Appendix Three. These and other factors led to the identification of the professional clinical need to conduct research that aims to provide a deeper exploration of the meaning of being a BRCA positive woman who elects RRM in the context of the wider family. The overall aim is to find a new horizon of understanding that will benefit patients in clinical practice. One consistent finding is the need for support and information from health care providers. However, the reasons for and detailed requirements underlying this support, still require exploration. An interpretive phenomenological approach I believed, would achieve this deeper understanding and the research questions therefore emerged.

In particular, what does it mean to be a BRCA mutation carrier and what is it like living with someone who is a carrier? How do women and their families make sense of the BRCA mutation, come to the decision of surgery and what are their experiences? What is the experience of undergoing RRM and why is body image, identity and sexuality so adversely affected? What are the

needs of the women and their families in terms of information, assessment and nursing care?

The next chapter introduces the research process and the methods used in the study.



## CHAPTER THREE – Methodology and Methods

### 3.1 Introduction

This study employs an interpretive hermeneutic phenomenological approach guided by the philosophy of Gadamer. It was conducted using in-depth, longitudinal qualitative interviews with eight BRCA mutation positive women (probands), following their results (pre-surgery) and at six and twelve months post-surgery. A final interview was conducted at eighteen months with a subset of four of the probands to capture an indication of the longer-term effects of RRM. To explore the wider impacts of their decision making and surgery, five of their husbands were interviewed pre-surgery and again at twelve months and five members of their family were interviewed pre-surgery. A total of forty-three interviews were conducted. Due to limitations of the requirements of the Ph.D., and the amount of data that would be generated, it was felt that interviews at two-time points with the partners and one with the relatives would add a contribution to knowledge. Analysis was conducted using the hermeneutic circle guided by the philosophy of Gadamer (2004).

To promote, '*credibility, depth and coherence*' as explicated by Finlay, (2006 pg. 17) and to put the study into context, the theory underpinning this study and its methodology needs to be discussed. This chapter will, therefore, include the starting point and decisions made by the researcher including the choices that were made throughout the study. Here, the researcher and participants are situated, the research question is defined and the methods

used are explicated. The ontological and epistemology standpoints are also included (Durant Law, 2005; Guba and Lincoln, 2005).

### **3.2 My Starting Point - The Emergence of the Research Question from Practice**

It is necessary, as the researcher, to situate myself within the study but also to provide some reflection on my starting point and what drove my desire to conduct the study. It also gives me an opportunity to give you, the reader, a sense of what it was like to undertake this study and the thesis generally. In Appendix One, I also offer a short autobiography of myself in order to reveal a little more about my background. Watts, (2009) contends that situating the researcher and providing an autobiography enables a better understanding and awareness of how one's own prior experience can influence the values, beliefs and decisions made along the journey.

Reflection is also an important aspect of any form of awareness and indeed in nursing practice generally. In trying to understand and interpret human experience about any phenomenon, a reflection on pre-understandings or what Gadamer calls '*prejudices*' is necessary. It is important and worth noting at this point that the term prejudice should not be viewed with any negativity as in the modern use of the word. In this context, it represents the productive pre-understandings that aid any future understanding (Fleming and Robb, 2003). Thus, not only does this reflection legitimise the pre-understandings, it

also helps to contain any influence that they may have on future understanding (McManus Holroyd, 2007).

Pre-understanding, therefore, derives from knowledge of the current literature (The Current Horizon of Understanding, Chapter Two) from my previous experience as a nurse and from my place as a woman in society. Furthermore, history and culture are very much a part of one's pre-understanding. According to Gadamerian philosophy, it becomes impossible to step outside of history, to look at the past. Indeed, a researcher's personal experience and pre-understanding should be viewed positively in studies underpinned by hermeneutics (which is the science of understanding human experience and meaning gained through interpretation) by providing some context to the study (Fleming et al., 2003). Furthermore, Durant-Law (2005) proposes that the inclusion of a researcher's experience enables them to situate 'self' within the study. It also makes visible the close relationship between the researched and the researcher.

The term hermeneutics is the philosophy of understanding gained through 'interpretation' and has been used in both ancient and modern times. (Dahlberg et al., 2008; Vandermause and Fleming, 2011). Philosophers such as Heidegger, and especially Gadamer, viewed the historicity of existence including background, pre-understanding and co-constitution as central to being able to understand 'being' in the world. Within the hermeneutic circle where Gadamer describes how understanding takes place by considering the

'parts' of the phenomenon with the meaning of the whole', the interpreter (researcher) brings their historicity into the interpretation. Furthermore, Gadamer also placed emphasis on culture and was interested in what culture brought to the situation. How it happened to be, for example. For all these reasons, therefore, it becomes important to situate the researcher (Gadamer, 2004).

### **3.2.1 Situating the Researcher**

I qualified as a Registered General Nurse in 1985 following training that involved learning whilst working in the ward environment. It was as a ward sister that my interest in breast care started when I observed that women would unsympathetically be told that they had breast cancer on a Friday afternoon in a dreary side ward, by a surgeon with limited communication skills and where maintaining confidentiality was a problem due to the unsuitable environment. The women were then admitted on the Monday for a mastectomy (complete breast removal) with no psychological care or general information pre-surgery.

At this time, the specialist nurse role in breast care was in its infancy and running a ward with sufficient nursing staff was a daily challenge. Furthermore, specialist nurses were seen as an expensive extra. However, as a ward sister I had more autonomy to influence service delivery and consequently, I made the first effort to set up some ward based psychological

care for the patients with breast disease. This involved listening to the women and supporting them and their family in a private room.

In 1991, I became the first specialist nurse in breast care in a large teaching hospital in Wales. It was from here that I had the autonomy, authority and opportunity to introduce a service for women and their families with breast disease with support from a surgeon who had excellent communication skills and valued the specialist nurse role. Much of the attention and care from a nursing point of view was directed at those with breast cancer both symptomatic<sup>7</sup> and screen detected<sup>8</sup> and little attention was given to those with a strong family history. This was mainly due to workload priorities and only one breast care nurse specialist in post. Furthermore, the Breast Cancer Susceptibility Gene (BRCA1/2) was not identified until 1994 and thus requests for risk-reducing surgery at that time was not possible and an uncommon practice.

In Wales, since 1996 when the test became available and since NICE introduced family history guidelines, women with a positive BRCA mutation who elect to have RRM have been referred to a surgical breast care unit. They are referred from a cancer genetic unit to discuss reconstruction options and undergo the surgery. The women are often positioned on a ward with women having surgery for breast cancer and although the women will have a

---

<sup>7</sup> Women presenting with breast cancer symptoms to a designated clinic.

<sup>8</sup> Women found to have breast cancer on routine mammography as part of the national screening programme.

consultation with a breast surgeon, they may not see a specialist nurse or have access to any pre-surgical support or information. Psychological care is limited for those women with breast cancer and in many areas of practice, non-existent for women with the BRCA mutation. At the time of commencement of this study, there was only one known unit in the UK offering psychological assessment prior to surgery.

From my experience as a Consultant Nurse working in breast care, I know that women elect to have their breasts removed following the detection of the BRCA gene mutation but I do not know what it is like to be given that result. Nor do I know how women make sense of it or how they are able to make the decisions that they do because of it. Furthermore, I do not know what it is like living long term with the knowledge of carrying such as mutation, the effect on the family or importantly, the long-term effects of the experience, both psychological or physical following bilateral RRM surgery. It was my pre-understanding for example that the fear of breast cancer may be reduced, but at what expense? However, neither I nor the team knew the answer to this and many more questions.

All resources are allocated to women with breast cancer and until recently, it was a general assumption in clinical practice in many different areas of the country, that women at high risk or carrying the gene were not a priority because they do not have cancer. In addition, in the past in Wales, many of these women have had to fight for an opportunity to see a surgeon because

they have not been regarded as a priority. This has now changed with the National Institute of Care and Clinical Excellence (NICE) guidelines and the increase in testing. The guidelines clearly state that for those women with the BRCA1/2 gene mutation, risk-reducing surgery with immediate breast reconstruction should be discussed (NICE, 2013). Therefore, the number of referrals nationally have trebled in the last few years. Colleagues welcomed the knowledge and understanding that would be gained from this study and the insight that would be generated in order to establish a service dedicated to BRCA positive women and their families.

### **3.2.2 Situating the Others**

Without the individuals that took part in this study, making a further contribution to gaining knowledge would not have been possible. To ensure anonymity, the women in this study are referred to as probands, the partners as husbands and the family members as relatives. I resist the title of patients despite the fact that I came to know them in a clinical capacity because not all the participants were patients at a given time and the probands were only in the role of being a patient for a very short period.

This study was therefore purposely conducted in order to explore the experiences of a different group of patients to those with breast cancer and those whom I felt were a forgotten, but important group. The inclusion of husbands and family members in addition to the women in the study is novel

and it was felt that to fully understand the experience for the woman with a BRCA1/2 mutation, it was necessary to position her in her social environment that engages with significant others. These others would inevitably be contributing to and affecting in some way the decisions made after the test and before subsequent surgery because I had witnessed this in clinical practice. They would also be living the experience and it was this experience that I wanted to capture, understand and learn from to meet their clinical needs. Many of the family members (including partners, parents, sisters and brothers) for example, also attended the clinic for the surgical consultation. Furthermore, little if anything is known about the effects of the whole process for the woman and her immediate family, including their past history, especially the consequences of RRM on the family. Thus, a gap in knowledge was identified. I imagined that the psychological effects were on a scale that matched the breast cancer patients, but I did not know if this was really the case. I felt privileged to be a caring, empathic nurse and to have access to their world and an opportunity to understand it.

### **3.2.3 My Academic Role**

My role as a lecturer in adult nursing complements the desire of lifelong learning by conducting this study as part of my Ph.D. The motivation to lead and improve clinical services for patients and to bridge the theory practice gap, I believed, would also benefit students. These include student nurses, medical students and inter-professional groups that I teach both pre and



post-registration. My interest in high risk women and those with the BRCA gene mutation has followed on from this lifelong learning and a desire to improve clinical services by attempting to understand real people who live in a world that has social, cultural and political connotations. It soon became apparent that as the main breast unit treating high risk and BRCA positive women and performing RRM much more frequently, that we knew very little about their experience.

An opportunity to research this as a Ph.D. student came to me as part of the necessary 'desirable' criteria for appointment as a Consultant Nurse and ultimately wanting a career as a clinical academic, I seized the opportunity. I was also fortunate to apply and gain a 'Florence Nightingale Research Scholarship' three years in a row in order to conduct this study.

As the researcher, therefore, I felt my knowledge, experience of the speciality, my involvement with the women and my counselling training, would add to rather than detract from the findings of the study. This belief was grounded in the fact that I, as a researcher, would be acknowledging throughout the study the factors that could influence the study and the constant reflection process would help to ensure the rigour of the study. Furthermore, reflexivity and the researcher's use of self is central to hermeneutic phenomenology and Gadamerian philosophy (Greatrex-White, 2008).

### **3.2.4 The Research Question and Aim**

My research questions developed from clinical practice and from identifying the gaps in knowledge from the literature. My overall aim, therefore, is to explore, interpret and develop an understanding of the experiences of women and their families who elect for bilateral RRM after inheriting a BRCA mutation. My overall research question is *'What are the experiences of living with the BRCA gene mutation and electing bilateral risk-reducing mastectomy for the women, partners and family?'*

I attempt to contribute to the body of knowledge and find a new fused horizon of understanding by exploring the whole experience from diagnosis to surgery and beyond as lived and experienced by the women primarily, and family members. I also endeavour to contribute to the introduction of a minimum standard of care for BRCA positive women and their families that reflect the needs identified in my study and develop a service that meets the needs of the participants and their families.

### **3.3 Hermeneutic Philosophy**

The philosophy of hermeneutics underpins interpretive phenomenological methodology which informs this study (Gadamer, 2004). This research is, therefore, an interpretive hermeneutic phenomenological study underpinned and guided by the philosophy of Gadamer, (2004). Interpretive phenomenology seeks to reveal and convey deep insight and understanding

of concealed meanings of everyday experiences (Annells, 1996; Vandermause and Fleming, 2011). Its goal is to increase understanding of the multiple interpretations of the meaning of human experience (Crist and Tanner 2003; van Manen., 2014) and can be used to challenge pre-understandings in order to understand experience differently. It is not used to prove or disprove the theory (Tapp, 2004; Cresswell, 2014).

Moreover, Gadamer posited that the interpretivist approach does not seek to establish a concrete view of a phenomenon but seeks to enable the researcher to understand the phenomena in itself. Thus, there is no such thing as absolute truth and no such method to human truths (van Manen, 2014). For Gadamer (2004) hermeneutic phenomenology is about how people understand the world they live in and he proposes a hermeneutic circle of understanding that considers the 'parts' of a phenomenon and the relationship to the 'whole' of the phenomenon.

The hermeneutic circle is the back and forth movement between partial understanding and the more complete whole. The 'parts' in this study, therefore, refer to my pre-understandings gleaned from experience and from the literature, the experiences of the women, the husbands and the family. The 'whole' refers to the understanding that emerged from 'being' a woman with the BRCA mutation who elects to have RRM in the context of her family and the interpretation of all the texts. The 'whole' in this sense is the united experience for the woman and her family living with the BRCA mutation,

electing to have RRM and the fusion of horizons that contribute to the body of knowledge.

Understanding is therefore at the heart of Gadamer's philosophy where understanding and interpretation are intrinsically linked and occur as a consequence of *'being in the world'* (Gadamer, 2004). Understanding is said to occur when the horizon of the researcher fuses with the horizon of the participants under study. A horizon is *'the range of vision that includes everything that can be seen from a particular vantage point'* (Gadamer, 2004, pg. 301). Furthermore, the researchers' preconceptions, biases and assumptions are clarified and they become an integral part of the study findings; (Whitehead, 2004; Koch, 2006). The findings of studies using an interpretive approach are consequently never neutral or value free and so this is made explicit right at the start.

This hermeneutic study, therefore, does not intend to establish a truth about the phenomenon or create a theory about it, but to challenge and question pre-understandings in order that new understandings and knowledge can emerge. In this way, practice and assumptions may be challenged to improve care for women and their families. Indeed, within Gadamer's philosophy and 'fusion of horizons', there is a strong movement away from traditional research and its focus on method and methodology. His focus was on trying to build a new bridge back to the humanist tradition and the notion of culture, which he believed constitutes knowledge. Gadamer (2004) believed that the

importance of culture was abandoned at the expense of the dominance of method (Grondin and Plant, 2014).

### **3.3.1 The Philosophical Beliefs Guiding the Research**

When conducting a study using a qualitative design, it is crucial to position the researcher in terms of their beliefs within the study (Cresswell, 2014) and there are three philosophical areas that need explication which both Crotty (1998) and Durant Law (2005) highlight. These include the ontological (what exists, how things really are?) and epistemological (what is known and what is the relationship between the knower and what can be known?) considerations, the theoretical perspective and the methodological considerations (how do we find out?).

### **3.3.2 Ontological Position**

When reflecting upon the influences on my Ph.D., I have been guided and influenced by philosophers such as Husserl, Heidegger, Merleau-Ponty but particularly by Gadamer (2004). Authors such as Guba and Lincoln (1998), Annells (1996), Crotty (1998), Ritchie et al., (2013) and David and Sutton (2011) also helped in establishing my positioning and beliefs which is that multiple realities exist but that experiences can be shared. There can be commonalities between individuals, only those who experience the BRCA mutation result and the surgery can really know the reality and these realities will depend upon their experience, their social and cultural background and

their beliefs. The ontological position, therefore, sitting in a relativist perspective because relativism proposes that knowing can only be achieved by being (Gadamer, 2004). Reassuringly, both Heidegger and Gadamer with their emphasis on hermeneutic philosophy took a relativist ontological approach by the explication of their notions of historicity and understanding (Annells, 1996). Guba and Lincoln's (1998) view of relativism also contends that realities exist in multiple constructions and these mental constructions are socially based and depend on the person who holds them.

Ontology is concerned with the nature of reality and knowing about the world in which we live (Ritchie et al., 2013). It asks what kind of being is the human being (Denzin and Lincoln, 2011). Some ontological questions inquire about how things really are? What is the real experience? What does it all mean? These questions were, therefore, a driving force in my research. A number of the studies examining women with the BRCA mutation were mainly driven by an objectivist, realist perspective (Hopwood et al., 2000; Frost, 2000; van Oostrum et al., 2003; Metcalfe et al., 2004a; Metcalfe et al., 2004b; Geiger et al., 2007; Wasteson, 2011) where there is a belief that an external reality exists and this is independent of our own beliefs or of our understanding (Ritchie et al., 2013).

This would assume that all women with the BRCA mutation have the same experience and that it could be objectively measured, I did not believe this to be the case. In addition, the research questions and aims in the

above-mentioned studies were very different to the aims and objective of this study. This may then account for the lack of true understanding about being BRCA positive and the experience of living with the mutation and electing RRM.

At the opposite end of realism is the belief that reality is mind dependent and only knowable through socially constructed meanings, known also as relativism (Ritchie et al., 2013). By keeping a reflective diary and following many discussions and much debate with my supervisors, I was able to identify and clarify my own ontological and epistemological standpoints. I, therefore, believe that this study is positioned in a relativist subjective ontology and that no external reality can exist outside of our beliefs or our understandings.

It is a reality that women have the BRCA mutation and elect surgery, but the construction of the meaning and the experience of this, I believe, is different for each person. However, I also propose that multiple realities exist, but there can be a shared understanding. Furthermore, these realities can change over time and can be influenced by the social, cultural and individual past, present and future experiences. Gadamerian philosophy is therefore primarily ontological and focuses on being, as opposed to knowing (Annells, 1996; Gadamer, 2004).

### **3.3.3 Epistemological Position**

There are two main philosophical theoretical frameworks that underlie research methodology, these can be broadly divided into positivist and interpretivist paradigms (David and Sutton, 2011; Silverman, 2011). Epistemology is concerned with ways of knowing about the world we live in and about what contributes to that knowledge and its sources. It also attempts to discover the relationship between the inquirer and the known (Denzin and Lincoln 2011; Ritchie et al., 2013). There was a need for me, the researcher, to understand the experiences of the participants and to learn from them and use this information to increase knowledge. There also existed a link between the subject and myself because of my role as a Consultant Nurse in breast disease, my clinical knowledge and my place as a woman in society. I knew what the literature had identified as to why women removed their breasts and the issues surrounding body image, but I did not actually know what it was like to have breasts removed or make that decision. Moreover, I did not know what body image and sexuality issues existed or why, nor did I know what it means to be a carrier, live with a carrier or what the whole experience entails.

This study fitted most closely with an interpretivist, constructivist epistemology, totally removed from the objectivist / positivist perspective advocated by Husserl whose focus was more epistemological than ontological (Annells, 1996; Vandermause and Fleming, 2011). The justification for a constructivist perspective developed for a number of reasons; there was a link between the researcher and the topic being researched, the belief that multiple realities



exist which are socially constructed and a belief that there could be a shared reality.

Constructivism also recognises the uniqueness of experience (Crotty, 1998; Guba and Lincoln, 2005; David and Sutton, 2011; Ritchie et al., 2013) incorporating a relativist ontology. Studies conducted using hermeneutical phenomenology fall ontologically, epistemologically and methodologically into the constructivist paradigm (Guba and Lincoln, 2005; Ritchie et al., 2013). Constructivism focuses on understanding experience from those that live it and that knowing the reality is dependent on those experiencing it. It proposes that knowledge is actively 'constructed' by humans rather than it being a passive role and constructivist researchers believe that in order to understand the experience, it must be interpreted (David and Sutton, 2011; Ritchie et al., 2013).

Both interpretivism and constructivism reject universal laws and value neutral observations and therefore sit at the opposite end of positivism (Guba and Lincoln, 1998; David and Sutton, 2011). The interpretivist view is grounded in philosophy and acknowledges the social world, which I believed, was crucial and pertinent in this study because of the social importance of breasts and the meaning of 'being a woman'. In considering the theoretical perspective of this study, I, therefore, believed an interpretivist approach would be more appropriate in achieving the aims of the study.

### **3.4 Methodology and Theoretical Approach**

The methodology of any research involves a discussion about the way in which the research data were gathered (research methods) and the underlying theoretical approach that influenced the research (David and Sutton, 2011). This study is adopting a phenomenological approach, in particular, an interpretive hermeneutic approach, initially guided by Heidegger (2010) but more importantly, influenced and motivated by Gadamer (2004). There is, however, a need for clarification of the adopted philosophical stance because, without this, there is an absence of methodological clarity (Lowe and Prowse, 2001).

#### **3.4.1 Phenomenology**

Phenomenology cannot be simplified to one research method (Crotty, 1996; Rafter and Robinson, 2003; van Manen, 2014), which is often the reason for many of its criticisms. Nurse researchers, for example, have been criticised for misinterpreting and presenting the methods of phenomenology without full understanding (McNamara, 2005; Thomas, 2005). They have been accused of having '*done a phenomenological study without knowing phenomenology*' (Porter, 1998 pg. 18). As a consequence, nurses have been advised and encouraged to explain the philosophical underpinnings of their research (Paley, 2005; Cooper and Endocott, 2007; Duncan et al., 2007). This lack of understanding may in part be due to the nature of phenomenology (Dowling, 2007) and to the difficulties of interpreting the original texts, written in German,

by some of the most famous founders of phenomenology. Writers such as Husserl, Heidegger and Gadamer.

Part of the confusion amongst researchers may also arise from the fact that phenomenology is seen both as a philosophy and as a methodology (Moran, 2002; David and Sutton, 2011; Creswell, 2014). It is an umbrella term applied to research by many researchers who have a range of values, beliefs and assumptions and in many ways, it is not a clear defined rule based method or one recognised in the usual scientific sense (Finlay, 2012; van Manen, 2014). One only has to look at the different interpretations by writers such as Ricoeur, 1976; Gadamer, 2004; Georgi, 2006; Heidegger, 2010; van Manen, 2014; to understand this confusion. There is a big difference, for example, between phenomenology and hermeneutics (van Manen, 2014) yet the terms are often used interchangeably within the literature, without adequate explanation.

Phenomenology is the science of phenomena and involves the human world, it can be described as a philosophical method for questioning and understanding rather than drawing concrete conclusions (van Manen, 2014). Finlay (2012) reminds us that the main concern for most phenomenological researchers is the embodied, experiential meanings and they generally agree that understanding the rich descriptions of a phenomena as lived by participants, is one of the main aims of phenomenological research. The debates around phenomenology, however, involve how best to conduct the

research because some approaches emphasise description, whereas others, emphasise interpretation (Finlay, 2012).

Hermeneutics is a branch of philosophy concerned with the study of the interpretation of human behaviour, the structures of society and how they function within society, including the exposure of hidden meanings (Byrne, 2001). Many nursing researchers have utilised hermeneutics to study phenomena (Byrne, 2001; Spence, 2001; Dowling, 2004; Spence, 2005; Dahlberg et al., 2008). Rather than attempting to generate new theory, hermeneutics strives to create new possibilities and to understand things differently by being open to the phenomenon, constantly questioning (Gadamer, 2004; Tapp, 2004).

Phenomenology has its roots in both psychology and philosophy and the key terms many researchers will be familiar with include essence, experience, understanding and meaning (Moran and Mooney, 2002; Speziale and Carpenter, 2007). Hermeneutic scholars are also interested in the lived experience but address the interpretation of 'meaning' and 'being' rather than focusing purely on descriptive experience. Furthermore, hermeneutic scholars reject Cartesian dualism of the split between subject/object or mind/body which is important in the context of this study. Finlay (2012) suggests that there are five iterative processes that are helpful in uniting the divergent methodologies and these are; embracing the phenomenological attitude, entering the life-world, dwelling with horizons of implicit meanings, explicating

the phenomenon holistically and dialectically and integrating frames of reference.

When first embarking on my research, I described it as a phenomenological study, naively not realising or understanding that there are as many phenomenological philosophies as there are phenomenologists (Caelli, 2000; Creswell, 2014). Indeed, eighteen different forms of phenomenology have been identified (Caelli, 2000). This is important because as mentioned earlier, what we understand to be reality (ontology) drives the epistemology (how we know what we know about the world) and the methodology (how we go about obtaining it). Furthermore, with the many perspectives offered, epistemologically, phenomenology can be located in the positivist (Husserl), post-positivist (Merleau-Ponty), interpretivist (Heidegger) and constructivist (Gadamer) paradigms (Racher and Robinson, 2003). Thus, there has also been a lack of explanation and therefore criticism, in many phenomenological studies as to where these assumptions fit in with the beliefs of the researcher (Paley, 2005; Thomas, 2005). The researcher has to account, during all stages of the research, for their underlying assumptions and ensure that their methods fit with these assumptions.

### **3.4.2 Interpretive Phenomenology**

Primarily this study set out to explore, but importantly understand, the phenomenon of living with the BRCA gene and the experience of electing RRM

for women and their family. For this reason, a phenomenological approach seemed the obvious choice. I had no theory to prove, no laws or scientific rules to adhere to and nothing to measure. As van Manen points out:

*'Phenomenology is primarily a philosophic method for questioning, not a method of drawing determinate conclusions. But in this questioning there exists the possibilities and potentialities for experiencing openings, understandings and insights giving us glances of the meaning of phenomena' (van Manen, 2014 pg. 29).*

I had to decide however whether the study would be descriptive or interpretive. The main aim of this study is to understand the experience and interpret meaning, so a purely descriptive approach I believed, would not completely help achieve my aims or do justice to the phenomenon. It would not provide a deeper understanding of the phenomenon nor would it identify the cultural and historical interpretations of the world. I felt an interpretive approach was more appropriate (Silverman, 2011; van Manen, 2014). Indeed, there is a debate about whether or not it is at all possible to describe a phenomenon, without interpretation (Pringle et al., 2011).

Heidegger, went one step further in his explication of phenomenology which demonstrated the emphasis on interpretation and the belief that it was more than 'to the things themselves'. Phenomenology is an inquiry that involves a dynamic play of showing and hiding. Thus, to Heidegger, phenomenology meant:

*'To let what shows itself be seen from itself, just as it shows itself from itself. That is the formal meaning of the type of research that calls itself 'Phenomonology'. But this expresses nothing other than the maxim formulated above. To the things themselves' (Heidegger, 2010 pg. 32).*

He proposed that the *'let show itself'* was something that was hidden and concealed but belonged to the lived experience and to the *'what shows itself'*. In other words, its true meaning and ground that needed to be uncovered (van Manen, 2014).

The aim of phenomenology, therefore, is to capture the richness of a phenomenon as it presents to the person who experiences it without any prior scientific hypothesis. It is a meaning-giving form of inquiry and a way of accessing the world as we experience it. It can be seen as a philosophical method for questioning in order to gain understanding (van Manen, 2014). However, Crotty (1998) emphasises that phenomenology 'seeing' is not easy. The researcher has to select purposively people who are equal and who are co-researchers, genuinely wanting to inquire into their own experience of the phenomenon in order to elucidate the essential elements (McNamara, 2005).

Again, I did not feel that this could be done without interpretation because to describe the experiences of living with the BRCA mutation would not be challenging the taken for granted assumptions or understandings of the participants. The main schools of phenomenology that have often influenced and been utilised by nurse researchers are Husserlian, Heideggerian and

Gadamerian phenomenology. All three schools of philosophy in some context influenced this study but Gadamer was most influential and will now be explicated.

### **3.4.3 The Work of Hans-George Gadamer (1900 - 2002)**

Although both Husserl and particularly Heidegger influenced my research approach, Gadamer (2004) and his work offered the missing piece and gave me the overall inspiration that provided the main lens to guide my research. It allowed me to both interpret and clarify the experiences. As Dahlberg explains:

*'Hermeneutics is the philosophy of understanding gained through interpretation to explain something and to clarify it'*  
(Dahlberg et al., 2008, pg. 66).

Through hermeneutics, beliefs, values and commitments become known. Understanding is achieved when the researchers' horizon fuses with the entity or person under study (Gadamer, 2004). Thus, in considering the women and the family in this study, in addition to Heidegger, Gadamer's (2004) philosophy fitted more closely with my aim and my personal philosophical beliefs. My aim attempted to gain a deeper understanding of how participants and their families made sense of a BRCA mutation. How for example did they come to the decision of surgery and electing to lose their breasts, what was it like living after the surgery in a world where meanings of risk, identity, femininity and embodiment are already laid down and experienced. This approach was adopted because an interpretive approach would look for *'culturally derived*



*and historically situated interpretations'* of the world (Crotty, 1998 pg. 67) in addition to how life has been socially constructed (Miles, et al., 2013) and these were deemed important in this study.

In addition, on reading further into Gadamerian philosophy, Caelli (2000) reveals that Heidegger was very critical of the role that culture and tradition has on understanding a phenomenon and did not initiate exploration of the cultural meanings for individuals. Heidegger seeks out the event that enables being, but Gadamer searches for the fusion of horizons between the past and the present. As my study involved culture and tradition, especially the culture and tradition of being a woman, I felt that Heidegger's beliefs did not completely satisfy me. I also believed that the participants' past experiences had influenced their present 'being' especially the decision to remove their breasts. Furthermore, as the women and family in my study are socially and historically conditioned, where breasts and illness have particular importance in a given culture, a hermeneutic study allowed me as a researcher to try and understand how the individuals interpret their world in a given context.

Thus, Gadamer's work inspired my thinking and bridged that gap. Moreover, hermeneutic understanding attempts to transport the horizon of the past to the horizon of the present (Crotty, 1998). For Gadamer, the horizon of the present cannot emerge without knowledge of the past. Alongside the researcher, Gadamerian hermeneutics consists of dialogue as opposed to individual phenomenology, where interpretation uncovers every activity including the

social, cultural and gender implications. I believed this to be very important in my study (Koch, 1999; Gadamer, 2004).

Gadamer and his work is now referred to as philosophical hermeneutics which expanded on the work done before him by Husserl and Heidegger and which was the focus of his magnum opus, *Truth and Method* (1960, 2004). Human beings according to Bryne (2001) experience the world through language and through language we gain both understanding and knowledge. These two assumptions form the basis of hermeneutics. Taking a step further, Gadamer regarded both conversation and the oral tradition as pre-suppositions for understanding written texts and believed that we are conversational beings where language is the reality (Gadamer, 2004). He believed that only through language and openness to the experiences of others, can understanding be achieved (Vandermause and Fleming, 2011).

#### **3.4.4 Philosophical Hermeneutics**

Philosophical hermeneutics considers how an individual, who is socially and historically conditioned, interprets their world in a given context and so I believed this fitted more accurately with the aim, philosophy and my participants. According to Gadamer, human beings are self-interpreting historical creatures who through tradition and historical life, find a means of understanding (David and Sutton, 2011).

Hermeneutics, advanced by Gadamer, involves two important elements, pre-judgement which is a person's pre-understandings or prejudices that make understanding possible and universality. According to Gadamer, it is an advantage not to be freed from prejudices or pre-understandings and he argued that the past, including all its traditions, must not be ignored because it is these prejudices, that makes understanding possible. Furthermore, if prejudices are not recognised, there is a possibility that one could fail to understand meaning (Fleming et al., 2003). As Gadamer explains:

*'A person who believes he is free of prejudices, relying on the objectivity of his procedures and denying that he is himself conditioned by historical circumstances, experiences the power of the prejudices that unconsciously dominate him' (Gadamer, 2004. pg. 354).*

Universality acknowledges the fact that the person who expresses and the person who listens is connected by a common consciousness, making understanding possible (Gadamer, 2004). Philosophical hermeneutics, therefore, made it possible to aid the process of understanding the experiences of the women, the husbands and the family because there was an existing link and bond between myself as a researcher and the phenomenon being interpreted. The bond as referred to by Gadamer (2004) is the relationship that must exist between the researcher who reads the text and the text itself, which details the phenomenon. The text must have relevance and meaning for the researcher, including the historicity of the research subjects.

I felt this historicity was very important, not least because of the women's past history of losing close family members to breast cancer. There existed a strong bond between myself and the participants. This involved my role as a nurse but also as a woman living in the same world. Through interpretation using Gadamerian hermeneutics, the researcher is able to consider the social, cultural and gender implications by permeating every activity (Crist and Tanner, 2003; Koch, 2006).

The hermeneutic circle of understanding is a circular movement of understanding and interpretation whereby knowledge is developed (Koch, 1999; Gadamer, 2004). In this way, researchers approach a topic with preconceptions, these are then re-examined in light of '*what the things themselves*' reveal to us (Gadamer, 2004, pg. 267). With this new understanding, we then return to a further exploration. The topic is understood by viewing the '*whole in terms of the detail and the detail in terms of the whole*' (Gadamer, 2004, pg. 291).

The hermeneutic circle was therefore utilised in this study to aid analysis and understanding. The hermeneutic circle is not to be reduced to the level of a vicious circle but viewed as a positive circle (Gadamer, 2004). The hermeneutic circle is the back and forth movement between partial understanding and the more complete whole. Gadamer's notion of the circle includes pre-understanding, historicity, the linguistics of understanding and the fusion of horizons (Annells, 1996; Gadamer, 2004).

In following Gadamerian philosophy, therefore, it is important for researchers to demonstrate a research process that reflects the underpinning philosophy despite there being no prescribed, fixed method or methodological structure (Gadamer, 2004). In fact, Gadamer pointed out that there are no methods to human truth and that as soon as we start to employ a method to experience, we turn it into an object where the truth of the lived experience will be difficult to reach (van Manen, 2014). However, to reach understanding, Gadamer (2004) did believe that there was a need for some direction from a systematic approach and so Fleming et al., (2003) offered this direction and many researchers have utilised Gadamerian philosophy and principles using different systematic approaches (Annells, 1996; Crist and Tanner,2003; Tapp, 2004; Howlin, 2008; Grassley and Nelms, 2008).

## **3.5 Methods**

### **3.5.1 A Qualitative Approach**

Depending on the question, David and Sutton (2011) and Silverman (2011) explains that any method chosen for a study should be the most appropriate for answering the question. Both quantitative and qualitative methods can, therefore, be viewed as merely tools that solve research problems. In current health care, there is now much more of an emphasis on acknowledging the experiences of patients and their families (Department of Health, 2010) lending itself more to a qualitative paradigm. Furthermore, because historically

much of the current literature on BRCA women, and indeed on healthcare research generally, has focused on quantitative designs and measurement, I wanted to capture the deeper meaning of experience.

It is also my belief that when studying experience, one cannot separate the multiple influences that have contributed to that reality, such as personal values, culture and social interaction. To answer the research question, therefore, I believed the most appropriate approach would be a qualitative one as I attempted to understand 'meanings' rather than make causal connections within the data, test hypothesis or generate figures (Ashworth, 2008; Silverman, 2011; Ritchie et al., 2014). Although accepting that both a qualitative and quantitative approach offer value in increasing knowledge, I did not and do not believe that to reduce the experiences of the women and families into a quantifiable analysis would be doing justice in providing a deep understanding of their experience. Knowledge in this sense can be derived through context specific understandings and therefore fitted well with the use of a qualitative method (Denzin, 2009; David and Sutton, 2011).

Polkinghorne (2006, pg. 73) offers a definition of qualitative research:

*'Qualitative study is an activity whose intended goal is the production and communication of an insightful and disclosing understanding of human phenomenon. Qualitative researchers engage in those activities that they believe will bring about the accomplishment of this goal as it relates to the particular phenomenon they are studying'.*

Furthermore, qualitative researchers stress the socially constructed nature of reality and the intimate relationship between the researcher and what is being studied (Denzin and Lincoln, 2011). As Mason (2002) stresses, the qualitative approach is suitable when the phenomenon being studied is a social process or has social meaning. In addition, the researcher uses themselves as the research instrument as a means of gaining a deeper understanding of the phenomenon (Fleming et al., 2003).

Taking a positivist view would assume that a reality about the experiences of the participants in my study already existed and could be generalised (Guba and Lincoln, 1998; Crotty, 1998; Denzin and Lincoln, 2011) and that value free research is possible (Ritchie et al., 2013). The researcher, in this case, would be seen as independent of the phenomenon of study and would have no influence over the research and the findings. This was impractical and did not fit with my philosophical beliefs. There was nothing to test, no laws that I needed to identify nor any known fixed reality about the experiences. Furthermore, a positivist approach would assume a theory of truth about the experiences that already existed.

### **3.5.2 In-Depth Interviews**

Interviews conducted within a qualitative paradigm aim to add to the body of knowledge from the meanings gained from participants (DiCicco-Bloom and Crabtree, 2006; David and Sutton, 2011) and in qualitative research, the

interview is the most frequent data collection approach utilised (Sandelowski, 2002; Nunkoosing, 2005; Silverman, 2011). For Gadamer (2004), some flexibility is required when applying methods that suit the phenomena under study and he favoured the spoken word in the interview as opposed to written texts thus allowing a two-way conversation and a deeper understanding.

The aim of this study set out to understand the whole experience for the BRCA woman and her family and to consider how this experience may change over time. Therefore, in justifying my decision-making across the research process and being strongly influenced by Gadamer (2004), I felt interviews would allow this conversation leading to the generation of written texts from transcribing the interview data. Compatible with hermeneutic philosophy, interviews can be unstructured or semi structured (Cohen et al., 2000).

### **3.5.3 Design**

In order to obtain full understanding of the particular historic situation in line with Gadamer (2004), speaking to participants on more than one occasion is recommended. Thus, the design used in-depth semi-structured, prospective interviews. There were at least three interviews with each proband as advocated by Seidman (1991) and they were conducted pre-surgery and again at six and twelve months following surgery. A final interview was conducted at eighteen months with four of the probands. The husbands were interviewed pre-surgery and again at twelve months and the relatives were



interviewed on one occasion pre-surgery due to time constraints. In total forty-three interviews were conducted and transcribed.

All interviews took place either in the clinical area in a quiet private room or in their own homes. Interviews lasted between forty-five and sixty minutes and a digital recorder was used. All interviews were transcribed and analysed. Initially, I transcribed the interviews to familiarise myself with the data and then because of my time constraints, used my funding to employ a senior member of the faculty to transcribe the remaining interviews.

#### **3.5.4 The Interview Approach: Hermeneutic Interviews**

Within hermeneutic interviews, the goal of the researcher is to co-create the findings with the participants through an engaged genuine conversation so that a fusion of ideas emerge (Binding and Tapp, 2008). In keeping with Gadamer's philosophy, a dialogic interaction takes place and a text develops. It is through this dialectic of questions and answers and the genuine open conversation which results, that Gadamer believed a topic could be more fully understood. Being present to and guided by the text and open to where the text takes you, is one of the goals in hermeneutic dialogue (Gadamer, 2004). The interview in this sense becomes reciprocal, dynamic and historical where a stance of openness to the meanings offered, is demonstrated (Crist and Tanner, 2003). Hermeneutic interviewing has been described as a unique, interactive, reflexive activity that is concerned with interpreting and describing

rich, ever day lived experience in order to achieve understanding (Walker, 2011).

The prospective nature of this study can therefore be seen as a strength. In a prospective study design, I could follow up on questions from previous interviews which are in keeping with Gadamer's assumption that understanding relies on the particular historic situation (Seidman 1991; Gadamer, 2004). My reading of some of the early sociological theories (Dingwall, 1997; Gubrium and Holstein, 1997) about how the interview can increase understanding of the social world through language and social interaction, also influenced my decision to interview. The beliefs of such writers reflect my own in the sense that the researcher in a qualitative interview is very much part of the research process, viewed as a co-participant. Data is co-created by the researcher and the participant because language construction is a two way process, especially when the researcher is knowledgeable of the phenomena under study.

Gadamer (2004) emphasised that to understand the person is to understand the subject matter, not to relive the experience. He reminds us that the purpose of the hermeneutic interview is not to replicate the experience but to create a new understanding of the phenomenon by both researcher and interviewee becoming immersed in the subject (Fleming et al., 2003). Streubert (2002) and Brinkmann and Kvale (2011) further purport that at an emotional and cognitive level, positive feelings of respect and curiosity in a reciprocal process between

interviewer and interviewee can lead to a deeper understanding of the topic, thereby generating rich data for analysis.

Interviewing, therefore, has been a well-known feature of phenomenological research and philosophical hermeneutic interviewing in qualitative research is now becoming extremely popular in health care (Baillie, 1996; Vandermause and Fleming, 2011; Walker 2011). The qualitative interview according to Ritchie and Lewis (2003) requires the interviewer to have a clear logical mind, an ability to listen, a good memory, curiosity and an ability to establish a good rapport and be able to demonstrate empathy. The same elements which are required in a competent counsellor. Empathy is particularly relevant because the interviewer has access to the world of the interviewee and has the ability to check or clarify understanding with the individual. Kvale (1996) also suggested that in order to show empathy, the interviewer makes use of the 'self' and becomes a research instrument.

Although there has to be boundaries, there has to be a transparency of 'self' in order to allow such engagement. In addition, the values, background, prior knowledge and pre-understandings of the topic area regarding the researcher are believed to be connected to the research by influencing the responses to the participants as well as the generation and analysis of the rich data (Ashworth, 2008; Creswell, 2014). The outcome of the research is therefore influenced by both participant and researcher. As Ashworth points out:

*'There is no escape at all from the fact that the research interaction is a genuine human encounter and that nothing can be done to stop the behaviour of the researcher being meaningfully communicated'. (Ashworth, 1987, pg. 18)*

### **3.5.5 Reflexivity and my Role in the Interview**

Researcher 'self-awareness' or reflexivity is crucial in qualitative research and an essential element of Gadamer's philosophy (Gadamer, 2004; Binding and Tapp, 2008; David and Sutton, 2011). It ensures rigour, credibility and trustworthiness (De Witt and Ploeg, 2006). This requires the constant reflection of beliefs, assumptions and prior knowledge and an insight into how these can affect the research (David and Sutton, 2011; Ritchie et al., 2013). Brinkmann and Kvale (2015) contend that acknowledging a subjective perspective can actually improve the findings by constructing multiple perspectives. Moreover, according to Byrne (2001), the experience of the researcher in hermeneutic studies should not be viewed negatively, it should be seen as a way of providing some context for the study. Ashworth (2008) further contends that the interviewer in qualitative research needs to be able to understand the talk and not be a passive recorder.

That said, it would be naive and unrealistic to imply that I could remain totally impartial from the participants and the phenomena that I was studying. An important element of any study is for the researcher to remain reflexive throughout the whole study and this I have endeavoured to do by keeping a

diary of my thoughts and actions, continually questioning my decisions and findings and discussing the issues at supervision.

### **3.5.6 The Relationship Between the Researcher, The Study and The Participants - My Clinical Role**

A key epistemological question in qualitative research concerns the relationship between the researcher and the researched and the influence that this could have on the values and the findings. On one side, there is an argument that the researcher can remain objective and value free. The other argument is that in the social world participants are not only affected by being studied but that the researcher cannot remain neutral and it is an interactive process. Value free objective research is seen as almost impossible (Ritchie et al., 2013).

The women in this study had already decided upon having breast surgery following their BRCA mutation result in the cancer genetics clinic. The purpose of the initial breast clinic appointment was to meet their surgeon and to discuss surgery. This included the opportunity to see photographs of reconstruction and discuss surgery with the consultant nurse (researcher). Family members also accompany the women to the clinic.

Following a referral from the cancer genetics service to the surgeon, as part of my role as a Consultant Nurse, I would have met the women in my study

approximately two to five months prior to the research interview. I therefore believed that a relationship had already been formed. My perception was that this would be an advantage. The advantages would be two-fold. For the participant, I believed that they would feel at ease talking to me because they had already met me and from a clinical perspective, I understood their BRCA result, having knowledge of the subject. They had volunteered to be part of my study, which indicated that they were interested in talking to me. Secondly, it would be an opportunity for the participants to contribute to and influence the care and service for future women and their families. For the researcher, the findings would be contributing to knowledge and lead to the possibility of a novel service. I, therefore, perceived that my relationship with them both as a nurse and a researcher would add value to the relationship.

### **3.5.7 Advantages of a Dual Role**

Leslie and McAllister (2002) and Fontana and Frey (2000) emphasise the benefits of dual professional roles. In this case, nurse and researcher. They state that nurses should reclaim their 'nursedness' and that the benefits of this would allow the participants to talk about social taboos because of the intimacy and immediacy developed within the relationship, thus allowing such disclosure but always being mindful of confidentiality and doing no harm (Nursing and Midwifery Council, 2009). Moreover, as a trained counsellor, in addition to my nurse training, I believed this was advantageous in being able to deal with any intimate conversations confidently and effectively.

Furthermore, Leslie and McAllister (2002) claim that to listen empathically, reflect, gently probe and successfully clarify (which are all components of a counselling interview) can enhance, rather than hinder the research interview. For me, as the researcher, I anticipated and hoped that the women and the men in my study would want to talk about the intimate aspects of losing the breasts and therefore I would be able to draw upon my 'nursedness' and my counselling skills if necessary. This would also be calling upon the social, cultural and historical pre-understandings of both the researcher and participants that Gadamer so often refers to, by opening up the conversation in order to fully understand the topic (Binding and Tapp, 2008). That said, I did not know what they would actually feel or disclose to me, but I felt prepared for any sensitive issues that may arise.

In line with Gadamer's philosophy (2004) I, therefore, remained reflexive throughout the study as I did not want the interviews to become a clinical interview or a counselling session and I was conscious of this before I started. I reflected on my own pre-understandings before the interviews began, in addition to the pre-understandings that were generated from the review of the literature. I was then able to be 'open to learn' from the participants in my study (Gadamer, 2004).

### 3.5.8 Planning

Before the interviews began, I had a few questions and a loose interview guide in order to meet the aims of my research. I did not want the interviews to follow a directive rigid format (Dahlberg et al., 2008) nor did I want to be influenced by what the literature had revealed as I felt this would bring assumptions into my approach and the interviews (Norlyk and Harder, 2010). To address this, as recommended by Fleming et al., (2003), I engaged with various colleagues at work in order to provoke my pre-understandings and my beliefs. I started the initial interviews with an open question. *'Tell me about your experience of the BRCA gene'*. Subsequent interviews involved open questions such as *'Tell me about your experience of having surgery'* and *'would you like to tell me about your experience so far'*. I was able to listen tentatively and follow up on points over the course of the interviews.

Through an engaged conversation, a text began to emerge and a fusion of ideas began to take place (Crist and Tanner, 2003). Although Brinkmann and Kvale (2015) argue that an interview will not slip into therapy because research interviewers have neither the time nor the training, I tend to disagree. Firstly, I do have the training and secondly, as a novice researcher, I could have allowed the interview to develop into a counselling session. However, conscious of this, I approached the interviews with the understanding that the aim of the research interview was very different to that of a therapeutic or clinical interview. This resulted from my ability to be reflexive throughout my decision-making and throughout this study.



I believe that the aim of a therapeutic interview is to bring about some form of 'healing' in a psychological way and that the person is being 'helped', with the helper being the interviewer. In a research interview, however, the roles are reversed. The participant is the helper with the potential to increase knowledge, not just for the interviewer or the interviewee but also for the academic and the professional clinical community (Bulpitt and Martin, 2010).

### **3.5.9 The Potential Power Imbalance in the Interview**

Due to the nature of the study, I anticipated that the participants may become upset talking to me about the sensitive subject but I was prepared for any emotion as this type of reaction is experienced in clinical practice on a regular basis. I was also mindful that there was further support (referral for counselling) that I could offer participants if necessary. More importantly, I was also aware that there could potentially be a power imbalance in the interview due to my role as a Consultant Nurse. In response, I began by explaining the difference in my role as a researcher and my role as a nurse and allowing the participants to dictate where the interviews took place and on which day and time; they were therefore in control. The fact that the participants had been approached by the research nurse and had voluntarily contacted her to agree to the study, further ensured that they remained in control and were not influenced to take part.

## **3.6 Setting, Sampling, Recruitment and Participants**

### **3.6.1 The Setting**

The study was based within a UK National Health Service (NHS) university trust in a purpose-built breast centre. Each year four thousand symptomatic patients are referred with breast problems. Approximately four hundred and fifty breast cancers are diagnosed a year. It is the main referral site for BRCA women and their family who elect to have surgery and there are now approximately one hundred patients referred per year with the BRCA mutation.

### **3.6.2 Sampling**

In phenomenological studies, in particular, Norlyk and Harder (2010) highlight the wide variation in how sampling is applied. It appears that little agreement exists in the literature on exactly what sampling should be (Curtis et al., 2000) and as a result, sampling in qualitative research has become a complex issue. There were a number of useful texts to draw upon when considering the sample in this study (Mason, 2002; Silverman, 2005, 2011; Holloway and Wheeler, 2010; Bryman, 2012).

The sample was purposive, as in many qualitative studies where the aim is criteria-based or to serve a purpose (Bryman, 2012). With this in mind, the sample were approached because they had particular characteristics, the BRCA mutation, which allows the deep exploration and understanding of a given subject (electing to have RRM). This will enable answers to be given to

the researcher's questions (*what is the experience of being and living with a mutation carrier and undergoing RRM*). Furthermore, as Dahlberg et al., (2008) point out, there has to be sufficient exposure to those experiencing a phenomenon in order to fully understand and thus husbands and family members were also included. The husbands and family members were included in addition to the women because the literature had identified a gap in knowledge of their experience especially their experiences of RRM. The knowledge gained of their experiences would help practitioner's both understand and address their clinical needs, thus also help the women.

### **3.6.3 Sampling Decisions**

As part of the decision-making process, case studies of the families was also a consideration. However, due to Wales being a small geographical area and with a prime obligation of maintaining confidentiality (thus preventing families with the BRCA mutation from being identified), I decided against case studies. In addition, I wanted to look at the experiences of the women primarily, in the context of their family and case studies would have focussed more on the family, taking the emphasis away from the experience of the women undergoing RRM. I decided to look at their experience as three separate groups.

Unlike quantitative research where sampling is based on having adequate representation and numbers in order to be able to generalise the findings,

reduce bias or produce statistically significant findings (Silverman, 2011), generalisability in qualitative research is not the objective. It is therefore important when assessing or judging a sample in qualitative research that the appropriate criteria are applied (Ritchie et al., 2014). If the purpose of a qualitative study is to explore an experience, Brinkmann and Kvale (2015) point out that just one participant is enough.

Prior to the study in 2011, approximately eight women with the BRCA gene mutation were referred yearly from the Cancer Genetics Department. Due to low available numbers referred each year, it was felt that a sample size of eight would be appropriate given that this number was the yearly local referral rate and that my approach was to gain in depth understandings of their experiences. Again, due to the limits of the Ph.D., the low numbers of potential families and following discussion with supervisors, five partners, and five relatives were deemed sufficient to get a glimpse of their experiences. The prospective nature of the study was felt to be a major strength of this approach, however, it was impossible due to constraints of the Ph.D., to interview all participants up to the eighteen-month time interval. It was also felt that this may be asking too much commitment from the family. It was therefore decided to interview the women at three-time points in order to gain an understanding of the long term effects of surgery, the partner's pre and post- surgery and the relatives just once. The limits of the Ph.D. and the amount of data that would be generated was a major factor in the decision process and one of the reasons why only a sub sample of four women (the first four who came

forward) were interviewed at eighteen months. The goal was to gain an in-depth understanding of the experiences with a further plan of post-doctoral work to develop the findings. The eligibility criteria outlined in Figure One were set for the study.

<b>Figure One: Inclusion Criteria</b>	<b>Probands</b>	<b>Family</b>
• Positive genetic test result for BRCA1/2.	X	
• Undergoing Bilateral Mastectomy.	X	
• Physically and mentally able to give informed consent.	X	X
• Eighteen years of age or over.	X	X
• Able to communicate.	X	X
• Willing to give informed consent.	X	X
• Able to communicate in English.	X	X
• Be a family member of the proband		X

### **3.6.4 Recruitment**

Following diagnosis of the BRCA mutation and after the appointment with the surgeon and consultant nurse, women were approached by the research nurse and provided with a study information sheet. After reading about the study, they could telephone the research nurse who would then inform the researcher of the participants. The participants were then contacted by the researcher to discuss the study further and to agree a time and place to conduct the interview. A consent form was signed once any questions about the study had been answered and the participants were satisfied. Permission was then sought from the women so that their husbands and family could be

approached to participate in the study. When family members contacted the research nurse to take part in the study, the researcher was contacted and the family were seen after agreeing on a suitable time and a convenient place to conduct the interview.

### 3.6.5 The Study Participants

The age of the probands in the sample ranged between the ages of twenty-three and fifty-eight years and the time since mutation results ranged from six to twenty-four months (see Table One). No restriction for inclusion was put on the elapsed time since initial results to ensure a deeper understanding and to include a difference in time range.

**Table One – Probands’ Information**

<b>Probands</b>	<b>BRCA Status</b>	<b>Time Since Mutation result</b>	<b>Age</b>	<b>Occupation</b>
<b>Proband 1</b>	BRCA2	2 Years	23 Years	Skilled Manual
<b>Proband 2</b>	BRCA1	2 Years	36 Years	Clerical
<b>Proband 3</b>	BRCA1	6 Months	34 Years	Skilled
<b>Proband 4</b>	BRCA2	1 Year	33 Years	Professional
<b>Proband 5</b>	BRCA2	18 Months	40 Years	Unskilled
<b>Proband 6</b>	BRCA2	6 Months	45 Years	Unskilled
<b>Proband 7</b>	BRCA2	18 Months	58 Years	Unemployed
<b>Proband 8</b>	BRCA1	6 Months	35 Years	Professional

Seven of the probands were undergoing immediate breast reconstruction and six of these had an implant-based reconstruction. One proband had an implant and strattice (pig’s skin) reconstruction and she also preserved her NAC. One

proband underwent a delayed implant reconstruction twelve months after her initial mastectomy. Four surgeons were involved in performing the surgery.

The husbands' ages ranged from between twenty-four years and forty-four years. The relatives' ages ranged from between thirty-eight years and sixty-five years and included one mother, two fathers, one sister and one brother. The details of the sample can be seen in Tables Two and Three.

**Table Two – Husbands' Information**

Husbands	Age	Occupation
Husband 1	24 Years	Skilled Manual
Husband 2	37 Years	Professional
Husband 3	36 Years	Professional
Husband 4	35 Years	Professional
Husband 5	44 Years	Skilled Manual

**Table Three – Relatives' Information**

Relatives	Relation to Proband	Age	Occupation
Relative 1	Father	60 Years	Skilled Manual
Relative 2	Mother	48 Years	Unskilled
Relative 3	Father	65 Years	Professional
Relative 4	Sister	59 Years	Skilled Manual
Relative 5	Brother	38 Years	Clerical

### **3.6.6 Ethical Considerations and Approval**

I attended a full Ethics Committee Meeting in December 2011 following submission of all the necessary documentation requirements online to The

Integrated Research Application System (IRAS) and the Trust Research and Development Department (R&D). IRAS supported the completion of multiple application forms for the Wales NHS Research and Ethics Committee (WREC), the sponsor and the Local Health Board (R&D). I attended the Research Committee meeting alone and it took one hour to discuss my research and satisfy the Committee. It was a very positive and exciting experience. All their questions were addressed and the committee had no ethical concerns. Minor amendments were made to the information sheets and full ethical approval was granted in January 2012 by The South East Wales Ethics Committee.

Annual reports have been submitted to both South East Wales Ethics Committee and the Trust Research and Development Department (R&D) since commencing the study. The Nursing and Midwifery Council (NMC, 2008) code of conduct for ensuring compliance with standards for conduct, performance and ethics were adhered to throughout this study, as was the Royal College of Nursing research ethics guidance for nurses (RCN, 2009).

Confidentiality, anonymity and participants' well-being were my main consideration throughout the study. Every effort was taken to protect the participants' identity by using letters instead of names, 'P' for Probands, 'H' for Husbands and 'R' for Relatives. Having password protected access to a computer and following the Nursing and Midwifery Council code of professional practice standards for conduct, performance and ethics (NMC,



2009) were also ensured. All records were kept confidentially and securely and recordings were erased from the digital recorder as soon as they were saved. Participants were treated with respect and dignity and the researcher was honest and open throughout the study thus upholding the principles of research governance and ethics. The study throughout followed the ethical principles laid down by the Welsh Assembly Government Research Governance Framework (2009), Cardiff University Research Governance for Health and Social care in Wales (2009) and Cardiff University Research Governance (2010). There was a constant awareness of the potential to cause psychological distress from engaging in the hermeneutic circle and the back and forth process of partial and complete understanding.

I was always mindful of the length of the interview and of the need to stop the interview if probands or relatives became distressed as a result of raising awareness. All study participants were in control and could stop the interview at any point. They were very keen however to continue with the interviews, even if they became upset. On reflection, participants were able to express their feelings openly in a safe and confidential environment.

### **3.7 Analysis and Interpretation**

A number of writers have suggested methods for undertaking hermeneutic studies and analysis including Colaizzis, 1978; Giorgi, 1989; van Manen, 1990 and Koch 1996, and all methods were reflected upon and considered during

this study. However, due to the diversity of their underlying principles, they were not entirely consistent with Gadamerian philosophy. Colaizzi's (1978) method did not view pre-understandings as essential to understanding the participants. He believed that the only benefit of this was in generating research questions. Equally, van Manen suggested that the knowledge gained through pre-understandings be turned against itself thus exposing its shallow character (Fleming et al., 2003).

All authors, however, indicate that when applying methods, the importance of analysis is to discover an approach that helps to guide an inquiry. As with most philosophers, therefore, Gadamer offers a guide to help researchers direct their research, rather than advocate a rigid procedure (Tapp, 2004). Thus for Gadamer, hermeneutic phenomenology had a tendency to be too dominated by a procedure and he advocated the use of an appropriate method rather than a single, right method where flexibility and modification are seen as acceptable (Gadamer, 2004). However, it is important for researchers to be explicit and transparent in the way the research is conducted. They need to explicate the philosophy underpinning the analysis and how analysis of the texts occurred, leading to the fusion of horizons. Thus, Gadamer's three metaphors of understanding, the act of dialogue, the hermeneutic circle and the fusion of horizons were utilised (Thompson, 1990; Gadamer, 2004).

### **3.7.1 The Act of Dialogue**

The act of dialogue is aimed at understanding that which the participants speak about, for example, being a BRCA mutation carrier, living with the mutation, the decision to elect surgery and life after the surgery. A fusion of horizons comes about through the act of dialogue (Gadamer, 2004).

### **3.7.2 The Hermeneutic Circle**

The hermeneutic circle considers the 'whole' in terms of the 'parts'. It is a continuous passing to and from parts of the text to make sense of the whole text and in doing so, the researcher interprets the whole text. The 'parts' therefore are many and include my pre-understandings, the literature including the current horizon of understanding, the individual participants' experiences expressed in the texts and the texts as a whole. They also include the social and cultural influences and past experiences' of both the researcher and the participants as well as the meanings, understandings and horizons that develop from the interpretation of the texts. The 'whole' in this sense therefore is the woman, her husband and the family living with the BRCA gene mutation and the experience of RRM. Simplistically, the researcher approaches a topic with preconceptions and prejudices. These are examined and revised in the light of what the things themselves tell us, (the participant accounts) and the current horizon of understanding (the literature). In light of this new understanding, further exploration is sought and the topic is then understood by *'viewing the parts and the whole in terms of the detail and the detail in terms*

*of the whole*' (Gadamer, 2004, pg. 291). In this way, it was possible to gain a totality of the meaning of the experience.

Thus the main aim of hermeneutic phenomenology is to reveal a *'totality of meaning in all its relations'* (Gadamer, 1997; pg. 471) by going *'beyond what is directly given'* (Spiegelberg, 1982; pg. 712). It seeks a deeper understanding of human experience by opening it up and rediscovering. Interpretive inquiry is therefore about being 'struck' by something and Gadamer (2004) believed that our past experiences structure how we deal with and interpret subsequent experiences.

Interpretation and understanding go hand in hand, it should not be viewed as a separate task. Interpretation begins in the interview and Gadamer (2004) proposes that there must be a harmony between the parts and the whole. In this case the participants in the study and the meaning of the whole experience. If harmony is not achieved, then understanding will not occur.

### **3.7.3 The Fusion of Horizons**

A fusion of horizons occurs as a result of the researcher constantly questioning the meaning of the text thus allowing the text to provide answers, leading to an interpretive dialogue between the two. In keeping with Gadamer's philosophy the art of hermeneutic understanding, including analysis, should not follow locked steps (Dahlberg et al., 2008) but should at least demonstrate

congruity between the framework used for analysis and the findings of the study. Furthermore, the interpreter in this context carries on the conversation with the voices beneath the text in order to identify that which is unspoken. The text is constantly questioned with the aim of lifting from it what the text is trying to say (Gadamer, 2004).

It is worth noting at this stage, that understanding others can never be totally achieved because understanding is constantly evolving. In addition, the whole analytic process using the hermeneutic circle can go on indefinitely, because understanding changes as time goes on. As Gadamer (2004) highlighted, however, to be able to understand, one has to be able to understand differently.

The fusion of horizons leads to a new horizon of understanding and this process of understanding results from the encounter between the researcher and the topic of inquiry. Interpretation and understanding cannot be separated and in this way, two standpoints come together. Therefore, in keeping with Gadamerian philosophy, we let the other (the participants in the study) speak to us and we are willing to be influenced by their perspective and we learn (Thompson, 1990; Gadamer, 2004). Hermeneutic phenomenology, however, asserts that all research is value laden and that the researcher brings bias, prejudices (pre-understandings) and assumptions to it (Lincoln and Guba, 1985; David and Sutton, 2011) but that these prejudices become a positive part of the encounter.

I, therefore, had to examine my own prejudices so that the text could assert its truth against my own pre-understandings (Gadamer, 2004). The genuine conversation that occurs in the interview brings the researcher closer to identifying their presuppositions and prejudices because of the potential to show up one's own weaknesses. This I documented in a reflective diary.

Pre-understandings or prejudices in this sense are not viewed negatively but as enlightening any new understanding (Binding and Tapp, 2008). Traditions and the past should not be seen as a negative but as an aid to overall understanding by shaping any interpretation (Gadamer, 2004). Furthermore, and more importantly, Gadamer believed that the historicity of an experience is lost if the aim is to objectify that experience by trying to repeat it or verify it.

### **3.8 Phases of Analysis**

The following phases demonstrate the approach to analysis, this was guided mainly by frameworks suggested by Crotty, 1998; Crist and Tanner, 2003; Fleming et al., 2003 and Lindwall and von Post, 2008.

#### **3.8.1 Phase One**

The individual texts from the interview transcripts of the probands, the husbands and the relatives were read and re-read with an open mind and there was a sense of the experience emerging (Lindwall and von Post, 2008).

This phase Crotty (1998) refers to as empathic reading. At the same time, the digital recordings were playing so that the two partners (researcher and participants) (recordings and transcripts) were working together to create a common understanding (Fleming et al., 2003). In this phase, there needs to be a continuation of openness from the researcher and an ability to see the participants' point of view and what the whole text is saying. Notes were taken as I progressed through the transcripts, prompting questions that I could follow up on with subsequent interviews. Individual texts (Probands n=8, Husbands n=5, Relatives n=5) were examined to form a whole text by moving from the parts to the whole and back again (the hermeneutic circle).

The first series of interviews with the probands were analysed before the second, third and fourth series and the cycle was repeated (Crist and Tanner, 2003). In this phase meanings emerged from the whole text, these became the focus of interpretation in order to form concepts that represented the meaning as a whole.

The interview transcripts were coded for themes by going through each transcript line by line. Initially, Nvivo8 Software was used to generate codes, but due to the enormous amount of data obtained, I found a much simpler way of categorising the text using Microsoft Word and a facility known as Spike. This method involved highlighting pieces of text that corresponded to a specific theme, moving it into a spike until the spike is emptied into a separate document. This left the original text intact allowing further categorisation,

whilst the selected sections of text were reflected upon by the researcher. Main categories then began to emerge from the themes. Mind maps of each participant's story and their experiences were developed and used in the analysis to reflect the final three horizons (see Appendix Seven).

### **3.8.2 Phase Two**

This phase involved interactive reading (Crotty, 1998) of the text where I questioned the text and the concepts that began to develop in relation to my pre-understandings, my research aims and the interpretation of the first interviews. This was done in an attempt to clarify meanings, challenge why I had come to that meaning and look for any inconsistencies from the previous interviews. Questions were asked, for example, 'what does this text say about real life and the phenomenon?' I continued to engage in the hermeneutic circle, moving from the parts of the text (all interview transcripts) to the whole as further questions transpired.

This engagement with the hermeneutical circle is essential, because only when the whole of the text is understood can the meaning of the parts widen (Fleming et al., 2003). Answers to my questions began to emerge and an understanding of the participant's experience began to develop. The reality of the text became a part of me as the reader and new meaning developed (Lindwall and von Post, 2008). An example of my questioning related to why women chose to remove their breasts rather than choosing screening. Initially,



the answer to that question from my pre-understandings and first stage analysis was that they did so to prevent breast cancer developing.

On further interpretation, however, it was much deeper than that. They had come to that decision because of the desire to survive and only surgery could save them, they had a fear of leaving their children behind and they felt a deep sense of moral obligation whether this was as a mother or a father. Thus, there was a fusion of horizons between the researcher and the participants.

### **3.8.3 Phase Three**

A number of themes and categories emerged resulting in many new initial horizons, this was a consequence of gaining an understanding of the phenomenon from all three groups. This was achieved by being open to what the texts were telling me through more interactive reading and looking for the hidden meanings or for what had not been said (Gadamer, 2004). Concepts then began to emerge that represented the deeper understanding and meaning.

Quotes from the participants were identified in order to demonstrate the meaning and provide resonance with one's own experience. Quotes also enable readers to judge the trustworthiness of the interpretations. The quotes, therefore, demonstrated an understanding of the interpretation and not merely a description of the experience (Norlyk and Harder, 2010). It was at this stage

that it became necessary to identify the horizons that connected and united the probands and the family, this was achieved by comparing the parts to the whole. Therefore, the final three horizons captured the analysis of all three groups of participants.

#### **3.8.4 Phase Four**

In this phase the new fused horizons were synthesised with the pre-understandings, prejudices from the literature and my own experience and a new horizon of understanding developed. Norlyk and Harder (2010) suggest that researchers should connect the different viewpoints from the analysis when forming an interpretation. This was achieved by viewing the new horizons alongside the pre-understandings and the literature. In this way, the pre-understandings were either confirmed, refuted or a different perspective suggested. Conclusions were then made about the experiences and implications for practice highlighted.

### **3.9 Rigour in the Study**

All research demands a demonstration of trustworthiness and rigour (Koch 2006). In qualitative research, in particular, the research process and truthfulness of the analytical process needs to be transparent because it affects the legitimacy of nursing science (de Witt and Ploeg, 2006; Porter, 2007). Nursing research has attracted much concern and debate for many years (Crotty, 1996; Paley, 1996; Annells, 1999; Turner, 2003; Whitehead,

2004; de Witt and Ploeg, 2006; Koch, 2006; Porter, 2007) not least because of its definition and misinterpretation of what constitutes rigour.

Rigour is defined as the '*Goodness*' of qualitative research (Emden and Sandelowski, 1999, pg. 2) and '*trustworthiness*' is recommended in trying to establish this '*goodness or quality criteria*' especially when taking a *constructivist approach*' (Guba and Lincoln 1998, pg. 210). Koch (2006) argues that trustworthiness is established if the reader can audit the events, actions and influences of the researcher. Some form of criteria of rigour is therefore essential to ensure high quality research methodology (Morse et al., 2002). However, some of the previous concern centres on the fact that applying a generic set of criteria to establish rigour in qualitative research does not sufficiently take into consideration the different philosophies and methodologies and the inconsistencies in articulating these philosophies. Sandelowski and Barroso (2002) argue against using 'epistemic criteria' for exactly that reason, preferring instead to use wise judgement and keen insight in recognising a trustworthy piece of work. This may then account for some of the criticism generated (De Witt and Ploeg, 2006).

Researchers utilising Gadamerian philosophy, therefore, have to establish the trustworthiness of their research and the truthfulness of their analysis especially so because it has not been traditionally utilised in nursing research. However, when referring to rigour, Gadamer (2004) proposes that

hermeneutic experience has a rigour of its own, that of, '*uninterrupted listening*' (Gadamer 2004, pg. 461).

In keeping with Gadamerian philosophy, this study will demonstrate trustworthiness and goodness in the process of '*uninterrupted listening*' by presenting the participants' stories as true and as faithful as possible by accounting for every decision made throughout the research process. The broad set of traditional criteria for rigour used by many nurse researchers includes trustworthiness, auditability, credibility, confirmability and transferability (Fleming et al., 2003, Whitehead, 2004; Koch, 2006).

However, a review of the literature shows that using generic qualitative research criteria of rigour for interpretive phenomenology can cause obstacles to the full expressions of rigour because of the philosophical inconsistencies mentioned earlier (de Witt and Ploeg 2006). The notion of multiple truths in interpretive phenomenology, for example, challenges the reader, making it difficult to decide which study is more rigorous than the others. There is no doubt, therefore, that a systematic process of accounting for the decisions made throughout a study underpinned by interpretive phenomenology is necessary and this has been a theme in the literature for many years (Crist and Tanner, 2003; Fleming et al., 2003; Turner, 2003).

As an alternative, van Manen (1997) and Madison (1998) propose the use of the term expressions of rigour, rather than strict criteria of rigour, because of

the unique features of interpretive phenomenology. Therefore, to demonstrate trustworthiness in this study and to preserve the integrity and legitimacy of interpretive phenomenology, a proposed framework by de Witt and Ploeg (2006) was utilised.

These expressions of rigour are balanced integration, openness, concreteness, resonance and actualisation (de Witt and Ploeg, 2006). The framework was developed and based on the work of Madison (1988) and reflects phenomenological schools of thought informed by Gadamerian philosophy. Madison (1988) argues that hermeneutic phenomenology is systematic and rigorous but not scientific and he justifies the bases of his set of criteria on what he calls persuasive (practical) reasoning. This comes in the form of reasoned argumentation rather than on demonstrative (scientific) reasoning. This is useful for nurse researchers because it facilitates application to the many philosophers and philosophies with its logic of argumentation rather than any specific philosophical tenet (de Witt and Ploeg, 2006). What follows, therefore, is an explanation of how the expressions of rigour were applied in this study and how trustworthiness was established.

### **3.9.1 Balanced Integration**

Balanced integration in this study involved articulating the philosophical approach, demonstrating how it fits with the researcher and the topic. My epistemological and ontological standing made explicit, including the

underpinning philosophy of Gadamer applied throughout. (Gadamer, 2004) (Chapters One and Three). A brief autobiography is also offered in Appendix One. Gadamer's philosophical concepts of pre-understandings, (prejudices) the hermeneutic circle, and the fusion of horizons are entwined within this study's methods and findings and there is a balance between the participants' voices (Chapters Four, Five and Six) and the philosophical explanations. A range of experiences (husbands and relatives) are identified and represented in order to understand the whole experience (Chapters Five and Six). The research process is therefore reflected upon throughout my study giving the reader an overall understanding of the decisions made, thus demonstrating congruency with the philosophical tenet and promoting trustworthiness.

### **3.9.2 Openness**

Openness is demonstrated by the overt explanation and reflection of the research process and the decisions made, including the rationale for my decisions and evidence of maintaining all ethical principles. It is recognisable through the systematic account of the multiple decisions made, thus providing an audit trail. Situating myself in the study and an introduction to the study (Chapter One) and extracts from my diary reflections are offered in Appendix Six as well as a short autobiography (Appendix One). Chapter Three includes a detailed explanation of the research process. Pre-understandings are reflected upon throughout the study (Chapters Two and Seven) and frequent honest open discussions took place with supervisors throughout the study.

An example of this openness was having a series of long conversations with my supervisor regarding the importance of including partners and relatives in the study which I consistently defended and which I believed were an important part of understanding the overall experience. My justification to include them was based on a gap in clinical practice and on the identification of my pre-understandings from the literature.

### **3.9.3 Concreteness**

Concreteness addresses the research outcome and is recognisable when the study's findings are written in such a way as to situate the reader in the context of the phenomenon, it gives the reader a real life feel of the experience by connection. In this study, participant voices and experiences in the form of text are presented in the findings chapters under the heading of Horizons starting with the probands, the husbands and finally the relatives. The process of interpreting the texts is provided (Chapters Three, Four, Five, Six and Seven). Frequent discussions of the findings with supervisors and presentations at meetings allow the reader to relate to the experiences and stimulate the audience, thus allowing them to judge whether the interpretations are congruent with the findings. An example of this was giving a presentation at a National breast cancer conference and having a 'complete audience' of both nurses and clinicians, with extremely positive comments because of the implications of the findings for clinical practice.

### **3.9.4 Resonance**

Resonance addresses the research outcome and is the experiential or felt effect of the findings upon the reader. Van Manen (2014) calls this the '*epiphany*' or the '*sudden perception and intuitive grasp of the life meaning of something*' (pg. 364) is where self-understanding takes place. This is recognisable when the findings are presented at conferences or meetings and resonate with the audience. It is also evident when supervisors or BRCA patients themselves read the findings. It was agreed with the participants in this study that on completion of the final study, a summary of the work would be given back to them to read. Bradbury-Jones et al., (2010) support this method of participant feedback. Presentations at conferences and meetings within the university have on numerous occasions, and especially as the research progressed, stimulated much discussion and genuine interest with extremely positive feedback given. The results not only resonate with those in the speciality but in health care generally.

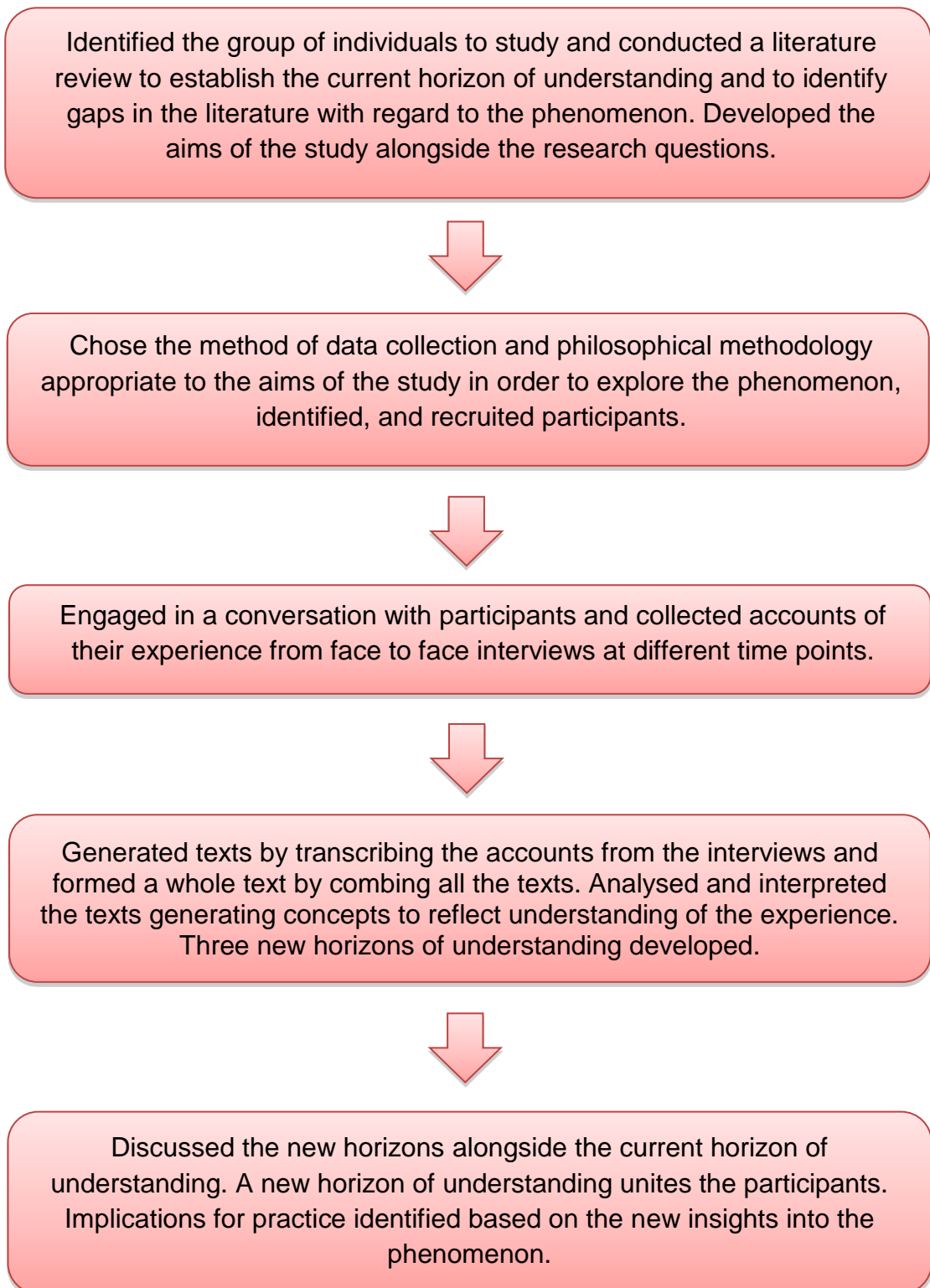
### **3.9.5 Actualisation**

Actualisation addresses the research outcome and is where the study's findings do not end when the study ends. It addresses the future realisation of the resonance of such findings and is recognisable when recommendations for practice are made and changes in practice occur. In Chapter Seven, the new horizon of understanding is presented and recommendations for practice



are made. A new way of working has been introduced in clinical practice and a multidisciplinary joint clinic has been established.

### 3.9.6 A Flowchart of the Summary of the Research Process



### **3.9.7 Chapter Summary**

This chapter located the researcher in the study and demonstrated the rationale for the methodological decisions made, including the ontological and epistemological standing. The philosophical underpinnings of the study were described and the study design, participant characteristics, recruitment and analysis were discussed. Rigour and how trustworthiness was promoted within the study were given and there was a summary of the process of interpretation and understanding. Four phases of analysis were used to guide the discussion of analysis and the fusion of Horizons. The following chapters present the findings of the study starting with the probands, husbands and finally the family.

## **CHAPTER FOUR – The Findings**

### **4.1 Introduction to the Findings Chapters**

This study sets out to understand what it is like to be a BRCA mutation carrier, live with the mutation and elect to have bilateral RRM for the woman in the context of her family. The following three chapters detail the findings from the interpretation of the experiences of the women (probands), the husbands and family members.

Staying true to my methodology, findings are presented as ‘horizons’ (Gadamer, 2004), with three main horizons of understanding identified through interpretation that capture the stories and experiences of all three groups. These horizons are, the price of survival, the altered child trajectory and a separation from self. In all three chapters, each horizon of understanding is presented along with the sub themes that led to that horizon of understanding.

However, I am aware that there were a number of ways in which I could examine and present their experiences. For example, an alternative way of presenting these findings would be by giving the different time points primacy, with chapters focussed on the period’s pre-surgery, six, twelve and eighteen months or by presenting all the findings under the three main horizons. I decided against this because of the limits of the Ph.D. and instead decided to capture and summarise those time points in pre-surgery and post-surgery experiences as not all the participants were interviewed at the different time

points. The focus of my study is primarily on the woman's experience in the context of her family and I felt the women's experiences if presented under each horizon would have been diluted. I, therefore, chose to study the experiences and present them as three separate groups.

This first chapter examines the experiences of the eight probands and are interpreted through these horizons. The probands ages ranged twenty-three and fifty-eight years and two were sisters. They all had children, girls and boys except for one proband who did not yet have any children. Seven were married and one was divorced. All except one proband was employed. The first horizon of understanding 'the price of survival' is the longest horizon because it interprets the complex experiences and journey from first learning of a mutation, leading up to the surgery. This horizon will now be presented, followed by the 'altered child trajectory' and 'a separation from self'.

#### **4.2 The Price of Survival - Pre-surgical Experiences**

'Surviving' was the main priority for all participants in this study but there was a price to this survival. The literature review, especially the work of Lodder et al., 2002; Van Dijk et al., 2008; Howard et al., 2010; Haroun et al., 2011, identified that women want to prevent breast cancer and survive but it did not offer an understanding of the price women and their families pay for this survival throughout the whole journey. This study, therefore, bridges that gap in knowledge and includes the experiences of a long journey of acceptance that a gene existed within the family, the difficulties in understanding the

implications of a mutation including the difficulties that participants encountered as a result of having inherited it. The need to be tested, the complexities of family communication and dynamics, the shock of the results and the implications of the decision to elect to have RRM, for example, caused major changes to the participant's lives, which they were not prepared for. The following sections provide the reader with that journey. The experiences have been divided into pre and post-surgery.

#### **4.2.1 Getting to Grips with the Possibility of a BRCA Mutation**

The awareness of a specific breast cancer gene within families became evident in different ways for the probands in this study. All the probands were aware of a family history of breast cancer, however, none had initially heard about this being associated with a specific BRCA mutation and in some cases, individuals had no idea that family members were going for a test or indeed that there was a test. It all began as a general awareness when someone in the family became concerned about the risk and sought risk assessment and a test for themselves. An identified family mutation therefore came as a major shock especially if there had been a lack of involvement from the start, making it more difficult for probands to understand all the implications. Proband two, for example who was most shocked by the news of a mutation in the family, explained how her sister had not told anybody in the family what she was doing. She only came to hear about a gene because of her sister's actions:

*'Finding out about the BRCA gene, I sort of stumbled across it, I sort of stumbled across it because my sister was err anxious about my nan having cancer' (P2).*

Most of the probands had not initiated the process of risk assessment or testing themselves but had become involved because of others. As Proband three pointed out *'she (cousin) started talking about this BRCA gene mutation and a test when my aunt died'* (P3). Another reported how as a result of her sister going for the test and then persuading her, she went to the clinic. *'If it wasn't for my sister I wouldn't have gone down there'* (P5). The diagnosis of cancer in young female close relatives, however, had made probands much more conscious of their own potential risk of cancer and of their own survival. Proband four, for example, explained how the loss of her aunt and knowledge of her young cousin's terminal diagnosis of breast cancer with an additional BRCA mutation, expedited and heightened her awareness:

*'But it was only when obviously x passed away I think it was three years ago now this month, her daughter x who was thirty-two last year, her diagnosis came along and that for me brought it forward by a decade. I knew that breast cancer would be something that I would have to consider and be aware of' (P4).*

Indeed, for many of the probands, there was an expectation that they would follow the family pattern in developing either breast or ovarian cancer. This depended upon what particular cancer the family member had died of. One proband for example, because of the family pattern of breast cancer explained how *'she knew breast cancer would be coming her way'* (P4). Another believed

it inevitable that she would get it because it had come down through the female generations. *'I was bound to be in line'* (P8).

The focus of concern, however, was the type of cancer which they knew was in the family and initially, they did not link this to the wider risk of a BRCA mutation with both breast and ovarian cancer so this posed an additional shock with more difficult decisions to make. Despite having genetic counselling about risk, it was surprising to see that for many of the probands, the shock about a BRCA gene was related to the sudden connection being made between the mutation and the risk to the breasts. Thus ovarian, rather than breast cancer, was the main fear if family members had died of ovarian cancer and three of the probands had lost relatives to ovarian cancer. Indeed, five of the eight probands had already had their ovaries removed following their BRCA result and before breast surgery, reducing their risk of ovarian cancer but many probands had not connected a BRCA mutation to the risk of breast and ovarian cancer proband two, for example, had never worried about her breasts:

*'I don't think I ever really connected breast cancer and ovarian cancer as being together so I'd never, ever had an issue with thinking about my breasts and would never have even crossed my mind that that needed to be looked at'* (P2).

Other probands soon realised that the risk of breast cancer was higher than for ovarian cancer which they were unprepared for and which caused additional fear and shock because they now had more to worry about and the decisions to be made, were much more difficult. Two probands explain:



*'And I felt fine about it because I just thought well umm I am going to have a hysterectomy anyway, and although it sounds strange to say, I wasn't as worried about breast cancer at that point because my cousin was very much worried about breast cancer because of her mum and I was much worried about ovarian because of my mum. But you know, as I have sort of found out information about the breast cancer, it become apparent that actually, that is, you know, something that is more likely to happen than the ovarian' (P3).*

*'So the biggest, I think the biggest shock for us really is the percentage of getting it (breast cancer) was quite a lot higher than I'd thought, so that was quite a shock, you know it's not like you're saying oh you've only got a 15-20% more, your risk is so much higher that that was quite a shock and although there was a thought around, I don't think I ever really connected breast cancer and ovarian cancer' (P7).*

Even in families where there was more awareness of a possible genetic link and reports of more open communication, there were still difficulties experienced concerning risk and the breasts in particular. Proband three, for example, had some awareness that there may be a genetic link and promised her mother that she would have a hysterectomy after her family was completed. Her mother died of ovarian cancer and had the foresight to provide a sample of her blood when she was alive in case a test ever became available. However, despite this communication and planning with her mother, proband three like many others, explained how it was all *'a bit of a taboo'* and therefore hard to discuss with anybody else in the wider family. Why was this so difficult to discuss? It was difficult because it concerned the breasts, which were seen as taboo and which had wider implications and consequences for the whole family. Proband three explained how difficult it was:

*'And whilst she was alive she um sent some of her DNA umm to be tested and to set me up with genetic counselling umm and so after she died, I saw the genetic counsellor and they went through umm our family history and umm said that they would test my DNA against my mum's and do a test basically. And then with the breasts it different really because it affects so many more people you sort of feel and it's hard for my family. I talked to my cousin about it a lot but not many other people sort of ask me about it sort of family wise because I think they all see it as a bit of a taboo' (P3).*

#### **4.2.2 Conflict Within the Family About How to Respond**

Many probands highlighted family communication difficulties with knowledge of a mutation, for example, not all family members, supported the probands' decisions to seek genetic testing. Despite close relatives having had breast cancer themselves, some probands were discouraged from starting the process which would lead to a test or surgery, for example, their family viewed it as *'meddling'* or *'potching with yourself'* (P5). Family members could not understand why some probands would want to start a process that could lead to the loss of their breasts when they did not have cancer. Family members who had to deal with their own cancer diagnosis and surgery were identified as being particularly reticent. As a result, initially, there was an unwillingness by some family members to give their blood in order for a mutation to be identified. This caused problems and tension within family relationships with some probands having to fight for and justify their decision. For example, it took some time before the mother of two of the probands agreed to give a sample of her blood so that they could have the test. Proband six used her need to survive to convince her mother:

*'No, my mother's answer was stop potching with yourself when there's nothing wrong with you, stop meddling. I'm like well because I value my family and I've got a new grandson and I want to be here for them. I said I don't want to die young if I can prevent it.' Yes and they (Mother and sister) wouldn't give it (blood) in the beginning, I'd forgotten about that, neither of them would give it and I said well still, I'm still going to go down for the counselling and I was going there for a long time before . For some unknown reason, my mother decided to give it and she said I will give it if you really want me to' (P6).*

Why would parents not want to help prevent their children from getting cancer?

It was too painful for some parents to acknowledge that they had passed on a mutation. In justifying their mother's initial reaction to testing, one proband believed that her mother felt guilty and responsible thus wanting to ignore it.

Proband five explained:

*'I dunno, my mother blamed herself type of thing, obviously which you do, don't you, because she passed it out through the genes and um she sort of wasn't happy in the beginning for us to go for Genetic testing. She said like things like that should be left alone' (P5).*

#### **4.2.3 Responsibilities, Obligations and Needing to Know**

Once it was evident, however, that a BRCA mutation existed within their family, probands felt an obligation and a responsibility to act upon it and various events contributed to the difficult decision to be tested. The death of a relative from a younger generation (P3, P4, P5 and P6), a memory of a close relative suffering at the end of their life (P2, P8), the recognition of the sheer number of women diagnosed in the family (P1, P7) or a need to know for their children's

sake in order to be able to take care of them (all), prompted probands to seek testing. A close family diagnosis increased the need to know for two probands whose mother and sister were diagnosed within weeks of each other. Their fear increased and the diagnosis had a catastrophic effect on the whole family thus initiating the test for the two probands. The fear of breast cancer was very vivid for one proband who believed she was next:

*'mum had her results during July of 09 and was told she had breast cancer which was like, devastated the whole family because up until then my mother had never had any previous history of illness and we were all fit and healthy. That hit us for six for a start, two weeks later my sister was diagnosed with breast cancer so not only one but two within weeks, we were all just, didn't know what to do with ourselves and then all of sudden you're lying in bed and I couldn't sleep thinking of them, thinking oh my god if it's mother, my sister, what about me, what about me' (P6).*

There were a range of rationalisations therefore for the decision to have the test and the subsequent decision to have RRM. Most probands had a constant fear of cancer but it was more than avoiding cancer, they wanted to survive and they believed they had an opportunity and a responsibility to do so. As proband two explained *'Someone and something has given me this, so why would I ignore it' (pg, 2)*. In addition, during data collection in May 2013, Angelina Jolie disclosed that she was BRCA positive. The probands were able to relate the meaning of being BRCA positive to her, making it less of a stigma. Some felt she had also sent out a message to the world that it was the right thing to do as a responsible mother and that even for famous people, there was an obligation and a price to survival, having the test and removing healthy

breasts. This famous disclosure had a range of impacts on the probands. Most probands thought she was brave in having told the world about her gene status and subsequent RRM. This had made it easier for probands to talk to people and explain their own story by normalising it. Proband eight explained:

*'Well it made it easier to talk to people about it, you know, you say well you know I've got what Angelina Jolie's got so I think it was very brave of her. Well, there's somebody who's famous, it can happen to anyone, yeah, precisely' (P8).*

However, some probands were angry about her disclosure because they felt she was giving false hope to women who would have expected to look like her. They also felt that she was being put on a pedestal for what she was doing. Proband five was particularly angry:

*'Oh I just thought it was a bit, it didn't affect me at all, I just thought it was sad that they put it all over the press, you know she was a martyr, she's this she's that, I mean she's not the only woman in the world who's had it done is she, you know. I was a bit angry really I think because they were saying like she was a martyr because she'd had it done you know' (P5, 6 months).*

#### **4.2.4 The Price: Divisions Within the Family**

The gene testing and the disclosure of results caused many rifts within families because of collusion, silence and secrecy. The majority of probands explained how communication about a gene was complex within the family. This was because of the secrecy of the topic and beliefs and rules within the family regarding who is permitted to know and who must be protected from this knowledge. Proband seven, for example, explained how a gene had never

been discussed properly in the family because she had not revealed that she had gone for a test. Her grandmother had not been tested but it was obvious that her grandmother had passed it down the generations to her mother, her aunt and then onto her. The secrecy and collusion was because nobody wanted to upset her grandmother or make her feel guilty. Proband seven explained the complex situation:

*'I'm not supposed to know that my gran, I'm not supposed to know that my gran has the gene, she's never formally told me that and she's never told that to my aunt, Yeah and so she told, it's kind of all a bit hush hush, we don't discuss it with Gran although I think now it's fairly obvious that gran has got it but we don't actually say that because I think she was upset about the concept that she might have passed it down to my mum and also to me, which is ultimately what has happened isn't it. But there's nothing, as you say there's nothing you can do about it' (P7).*

Many other probands explained the complexities within the family. They gave accounts of individuals having a test and the wider impact this had within the family. The issue of testing for one proband, for example, resulted from a forced disclosure from her sister. Her sister had secretly gone for the test and subsequently revealed that she did not have the mutation. As a consequence of her sister's actions, proband two and her brother felt they had almost been forced to consider the test themselves without any discussion, forewarning or individual control. The resulting impact of that one test had devastated the whole family because of the wider impact the results had on them all. The

decision to seek testing was seen as a family decision and not an individual one to be taken. Proband two justified her anger at having no control:

*'And this is the thing, we feel, well me and my brother discussed this and we said it is very canny that x has pushed for this and went into this with her eyes open and requested it through her GP and everything, and we have been sort of, it's been forced upon us, we have, in a way'. This was a family decision to proceed with this test. My sister should have never embarked upon it on her own and in isolation. She should have discussed this with us all because it has impacted on all of us' (P2).*

Proband two continued to explain the family difficulties about the issues of who could be told and who had control over this information. Similar to other probands, it was expected by some individuals in the family that a gene result would be kept secret from other family members but this did not feel comfortable or right for some of the probands, causing much distress. Proband two for example despite resistance, morally, could not keep it from her father:

*'But I felt disloyal because I was holding something about my dad, and him and his family because my sister didn't want him to know. He has a right to know. And I found that difficult' (P2).*

Thus, she explained the 'mess' and how unpleasant it had all become and how the family had been divided. It, therefore, took a long time after the positive test results to pluck up the courage to go and tell her father that both she and her brother were positive for a BRCA mutation. Like many of the probands, she did not want family members to feel guilty about passing on a mutation.

Furthermore, her father would not have known that he had passed the mutation on because he had not been for the test himself. It was also a 'mess' because she now had to think about acting upon her own result which brought many difficulties in itself. She explains:

*'So I think around the end of February I took the courage to go and tell my parents because it meant that I had to start making some decisions about myself which was awful, it was a dreadful mess, only because I am more protective of how they are going to feel because it is always about well it's my fault I gave it to you, but my answer back to my dad was you didn't ask for it either so why are you blaming yourself for' (P2).*

Why was there so much secrecy within the family? Issues of secrecy and who to tell were reported by all probands and a range of reasons were given to justify it. For example, probands wanted to protect parents or siblings from worrying about them. Indeed, in some cases going for a test and the decisions they had made were kept a secret right up until the time of surgery. Probands also needed to be sure of their decisions. Proband one, in particular, was scared to tell her family, especially her siblings, because it would highlight that they too may have inherited the mutation and they did not want to know. Proband one was, therefore, trying to avoid a division in the family:

*'But um I thought it would be better that way because save them like obviously save them worrying all year bout my decisions. I wanted to make sure I knew what I was doing'. I didn't actually tell my family, I was scared to tell my family, so I actually left it the whole year and I literally told them about 3 weeks ago (laughs) um cause I was scared, I was like, okay in myself and my husband knew ,and like two friends but I was*



*just afraid to tell my family because I didn't want to upset my brother and sister because obviously, it would come to them that they might have it and they would have to go for the test' (P1).*

#### **4.2.5 The Price: The Unexpected Impact and Implications of the Result**

Although most of the probands expected or prepared themselves for a positive mutation result, some had not. Three probands, two of whom had inherited a mutation from their father, were shocked, scared and devastated on hearing their result. Why were some probands more shocked, scared or devastated about the results than others? It was because they had not been involved from the start, had not realised the implications of the test or understood the significance of a mutation. Proband four, for example, expressed the fear that she may already have breast cancer *'I'm panicking in case it's already there now'* (P4). Proband five she did not expect her mutation result, believing it was down to luck. *'I'm always the one who is okay'* (P5) and proband two who did not understand that her father could pass it on, believed *'It's just one of those freak of nature things that has happened'* (P2).

Indeed, proband two had given little attention to the prospect that she may have a mutation. Like many of the probands who had not realised the full implications, she reflected on her naive decision to be tested:

*'I don't think I realised what I was getting myself into at all, I just thought oh well if x is doing it I will need to do it, thinking it will never happen to me' (P2).*

Following her result, like many of the participants, proband two was shocked and traumatised, questioning why she had the gene and initially feeling responsible for having inherited it. Coupled with the shock and the unforeseen effects of the result, in an attempt to make sense of it all, she subsequently felt a sense of failure in preventing it from occurring:

*'And I think maybe in a way I saw it as failure. Failing, that I've actually got this thing, this BRCA thing, you think it will never happen to you, why did it happen to me and also that I just felt so unwell you know, and I normally bounce back after things but this did hit me for six in terms of feeling rotten' (P2).*

#### **4.2.6 The Price: Living with and Making Sense of Being BRCA Positive**

Having embarked upon a journey therefore that led to a test and a positive mutation, probands now had to make sense of it all, deal with the result and its implications and live with the fear, which was life changing for them. For some, cancer was already an inevitability especially in families that had experienced the death of a close family member and so having a specific mutation, did not have the same impact as for those who had not witnessed death. Some probands, for example, were expecting to develop breast cancer without an identified gene. Proband eight explained why:

*'Yeah, before we even knew about the BRCA gene I always thought I'd be the one being the eldest, somehow, that I would get it but of course, my little sister got it instead. But when you've lived with cancer and it's been in the family for years*

*and years, it's not as, it's a horrible way to die but it's not as frightening as somebody that's had nothing to do with it and you mention the word cancer and they like nearly drop you know. Sounds funny but a lot of my family have died of cancer, a lot of them and...'* (P8).

However, for the majority, a BRCA mutation was more significant confirming the certainty of their bodies being invaded by cancer, which increased their fear. Indeed, some probands considered that they may already have cancer. As proband four explained *'breast cancer felt like it was imminent like it was around the corner if not there already, but it just felt inevitable, and with X getting it...'* (P4). For many probands, despite knowing that the risk of breast cancer with a mutation was up to an 85% lifetime risk for example and despite reassurance from professionals that it may never occur, all probands feared that it was now a confirmed definite. Two probands explain:

*'Yeah, I know that's like not a fact, but to me, that's how I felt, so as soon as I found out I just said look, I know now that I'm going to have cancer one day so but like, but doctors would say or you know there's nothing that to say your gonna have it, you might have it when your 70, you might never have it'* (P1).

*'I think my family history had always pointed in that direction but getting the news that I had the gene just confirmed that and cemented my opinion that I was going to get it (breast cancer) one day. When you look at the science and the genetic risk then yeah, I couldn't be definite about that at all and I know that, but it's just that kind of acceptance in my head'* (P4).

Despite the belief and inevitability of getting breast cancer and their individual understanding of having a mutation, probands also gave their rationalisations for expecting to be BRCA positive. The majority of probands for example who had expected to have a mutation believed it was because they resembled their mother and because of the number of women with cancer in the family. Thus, as well as inheriting other traits, they would also inherit a BRCA gene. They connected the same physical and personality traits of family members with the likelihood of testing positive. Two probands were convinced they had it and explained why:

*'but I was fairly certain in my mind that I had it because I'm so similar to my mother that, I know it doesn't necessarily you know, you're 50% your mother, 50% your father or whatever but the reality is I look the spit of my mother, I am. Mannerisms, everything is my mother so I was convinced that I had the gene' (P7).*

*'I don't know what it was, I think I knew I was going to have it because what I had seen with my mother as well, and me and my mother, because we are so close and alike I just thought if my mother has it, then I got to have it. 'I think it was something I always, cause my mother and her mother, I think it was 4 or 5 other women in the family had had it (breast cancer) and think it was something I always thought I knew' (P1).*

These rationalisations of risk also extended to their children and future generations. Proband seven, for example, was able to identify which of her children she believed she needed to worry about based on both the physical and personality traits she associated with her mother and the genetic pattern of inheritance within the family:

*'I mean I look at the kids and I think x is so much like my husband, it doesn't really matter, you know, but x is the spitting image of my mother, absolutely spitting image, personality and everything and I think oh I hope he doesn't have it. Yes, in my head that's how I think that's the worst thing is that the kids could have it' (P7).*

However, not all the probands could rationalise the positive mutation by linking it with their family resemblance or family history. Proband six unlike her sister who believed she was always lucky, believed she was always unlucky. She thought this view would help prepare her, but the result had much more of a negative effect than she anticipated:

*'Yeah, I prepared myself in as much as if anything ever goes wrong in this life, in this family, it's normally me. Like hysterectomies, three of us had it done within weeks, whose went wrong, mine, things like that and I'd sort of prepared myself to think well it is me and I will be okay about it, but when you hear it, it's different' (P6).*

Most probands were empowered and forewarned with the knowledge of a BRCA mutation, enabling them to be pro-active and take control, even if the price to pay was the removal of their healthy breasts. Proband four explained the benefits of knowing:

*'I felt as though I'd, I guess because if you know you've got the gene you know what your risks are and so, you've got a relatively good idea and I'm not somebody really who sits there panicking over, oh my god what if this happens or what if that happens, I kind of feel that you just deal with the problems that arise when they arise and there's nothing else you can do about it otherwise. So, I felt as though well at least I've been forewarned which is more of an empowering position to be in' (P4).*

Interestingly, unlike the other probands, one proband felt fortunate to have a mutation because she believed she would be safer, thus enabling her to receive superior care compared to those who had chosen other risk management strategies:

*'I said that I wasn't actually convinced that if I didn't have the gene whether that was a positive outcome for me because I very much saw it that if I had the gene something could be done about it, I would then come under the care of people who would look at it, would look after me and it would be something that I would actually get preferential treatment over somebody else who might just be on screening' (P4).*

This ability to be proactive was echoed by many other probands, who unlike some family members were lucky to have a choice in doing something about their risk. This had not been possible for some family members because they had already died and it was at the expense of these family members, that it was possible to prevent the same from happening to themselves. Proband eight explained:

*'It sounds strange, but it was bad that I have got the gene but good that I had the opportunity to do something about it. I feel lucky that they've found, but at my sister's expense, that they found out that we've got the BRCA gene to give me the choice' (P8).*

#### 4.2.7 The Price: Disposing of Dangerous Breasts

In acknowledging some of the benefits of being BRCA positive, all probands now had a different view of their bodies, in particular, their breasts and what they represented. Whereas most liked their breasts before the BRCA result, they used different metaphors to explain how they felt after the result. They reported a need to dispose of their breasts because they were dangerous, Two probands explain:

*'Because like I don't know, it's just like monsters and aliens inside you, just wanna like get rid, don't you' (P5).*

*'Mmm, they are my time bombs. They used to be my big voluptuous chest, they're now my time bombs, I want them gone' (P6).*

However, despite the need to dispose of their breasts, the majority of the probands were concerned about what people would think about their body looking so different. Many people, for example, had already started to judge them about their decision, especially if they were very young. It was important to probands for people to understand the preventative nature of their surgery and as such, they felt a need to justify their decision. The majority of probands, for example, did not want people to think that they were having surgery and reconstruction for breast enhancement because that would be for a different reason. Many were frightened that they would be judged and felt embarrassed. Two probands explain:

*'Yeah, and do you know what I'm more frightened of as well, that somebody thinks I've had a boob job for vanity purposes' (P2).*

*'I don't want people sat there thinking has she had a boob job' (P4).*

### **4.3 The Price - No Choice, Surgery is the Only Option**

The fear of cancer and not surviving dominated the experiences of all the participants but the price to pay for their survival was the need to elect to have RRM. Despite the understanding that the risk was not 100% reduced and the availability of alternative options such as surveillance programmes, RRM, although a difficult decision, was seen as the best and only option for preventing breast cancer and thus death for all of the probands. All of the probands felt that they had no choice because they wanted to survive. Two probands explain why:

*'The knowing that I'm not going to die of breast cancer or the risk is minimalized to such a low % by having surgery...' (P6, pre-surgery).*

*'Getting cancer? I have always, since my mother I was only young like very young but I've always worried so when I found out about the gene it was just like I'm definitely going to have it. Don't you know something, I felt like I knew that I was going to have cancer and that's why I feel like I have to do what I'm doing tomorrow (having surgery) because deep down inside I feel like she was like only 31, her mother was 34 and I feel like obviously, I'm going to be young having it, so that's why I want to do this now' (P1, pre-surgery).*



Most probands were, therefore, determined to proceed with surgery despite friends, family or professionals questioning their decision, trying to persuade them against it or suggesting that they wait until they were older. Proband one was determined to go ahead with surgery despite her young age and the resistance:

*'But yeah, my age, people like the genetic counsellor 'do you want to wait until you're older'? And then down here, you might not have cancer till you're into your 70's and I was like but I'm still going to have it' (P1, surgery).*

When questioned on why and how probands came to the decision of surgery, many reasons were given for choosing surgery over other options and most provided detailed accounts of losing close young family members to cancer. Proband eight reflected on the terrible distress of watching her sister die of breast cancer. Thus, like many of the probands, although she did not want to have her breasts removed, she felt she had no choice and this experience had convinced her to elect to have surgery:

*'What made up my mind that perhaps might not have made me so definite is I was watching my sister dying of it and the state that she was in. Quite honestly it was terrible' (P8).*

Most probands, therefore, believed that they had been given a great opportunity to ensure survival, which must be taken. Surgery would enable them to carry on living their normal lives, albeit at the price of losing their breasts, which initially was a secondary consideration. Indeed, all probands

believed that the decision and the opportunity to elect to have RRM was going to save them and they were grateful to have that option. RRM would also prevent them from living with a constant fear of breast cancer occurring. Two probands explain why it had to be surgery:

*'So, my way of thinking is life's for living, you've been given a second chance in a way, why would you want to waste it' (P2).*

*'I'm glad that we have found out now and that there is the surgery about, you know, to fix it. I do see it as lifesaving, yeah. I mean can you imagine if they had this years ago and they say oh yes you've got the BRCA2 gene and you'd have to live with the thought of that every day, constantly on your mind, so yeah it's a relief' (P5).*

However, having these opportunities to be proactive and secure their own survival at the expense of family members who had not survived, caused many probands to feel sadness and guilt:

*'I felt terrible guilt that what she had been through had meant that I was able to dodge it and it was unfair that it was something that she couldn't dodge' (P4).*

#### **4.3.1 The Price: A Visibly Altered Body**

As time elapsed and the reality of surgery approached, despite the perceived survival benefit, the decision of RRM was far more difficult for probands compared to removing their ovaries. If probands had completed their family,

removing the risk of ovarian cancer was a much easier decision. As proband two explained:

*'I didn't want more kids that's one thing, so that was a no brainer. And this is why the bottom half was much easier to make a decision on. But that was easier because I thought right I don't want any more children' (P2).*

Why was this an easier option especially in view of the known menopausal symptoms experienced following oophorectomy? Even though many probands experienced menopausal symptoms following RRSO, there was no visible evidence to the body of having had the ovaries removed compared with breast surgery, making it easier for the probands to accept and live with. Proband eight explained why:

*'Yeah, or I didn't mind having a hysterectomy because you can't see it so that didn't bother me' (P8).*

Breast surgery, on the other hand, was much more visible, perceived as a bigger operation psychologically and would result in a changed visible body. More anxiety was experienced by all probands about the expected outward appearance because people would be able to see what had been done to their body. Indeed, it was this fact that had been a dilemma, making it more difficult for some probands to decide to proceed with surgery despite believing it was the right thing to do. It was also more difficult to accept if breast cancer risk

had not been their main fear. Proband three was particularly worried about living with the changes to her body:

*'It was you know a no brainer situation, you know. I had to do it (have ovarian surgery) and that's all there is to it. It's harder to make the decision about the breast cancer because my mum didn't have breast cancer and because it's an outward thing. And time to recover and you know all of those sorts of things but I don't want to choose to have this operation and then feel sad that I don't, I don't feel like I can you know, sort of be pleased with my body' (P3).*

#### **4.3.2 The Price: No Longer My Whole Self**

Probands wanted to survive, but they also wanted to remain looking like a woman. For the majority of probands, their breasts were very important and defined who they were, representing being 'whole' women. All probands wanted breast reconstruction in order to remain feminine, whole and womanly and it was an expectation that breast reconstruction would enable them to do so, looking like a woman should. Pre-surgery all probands were given an opportunity to see realistic photographs of the surgical outcome and discuss the operation. All but one proband was either shocked, horrified or devastated at these images of the cosmetic result of breast loss, even with reconstruction because it was not what was expected. The images looked so different to normal breasts and did not resemble 'themselves'. Two probands share their expectations and devastation after seeing the photographs:

*'What! I really, this is what I expected, scar underneath, pop it all out, pop something in and do it up. But we thought cosmetic*

*and preventative was one and the same thing but they're not. O they were shocking, they were shocking' (P6).*

*'In the beginning, before I seen the photos I thought oh yeah you know, you'll have a nice lovely looking pair but I don't know I just haven't got any expectations now. Ooh, I don't like the look of them, I thought, Oh fuck, look at that, really when I saw the photos. 'In the beginning, before I seen the photos I thought oh yeah you know, you'll have a nice lovely looking pair but I don't know I just haven't got any expectations now. Ooh, I don't like the look of them, I thought, oh fuck, look at that, really when I saw the photos. I was gutted. I thought, Yeah, scoop it out or something you know and I don't know, and just put a bit back in. Where I got that from I don't know, but that was I was thinking' (P5).*

Most of the probands initially thought that after reconstruction, the breasts would look normal, possibly like women who have plastic surgery and they expected minimal evidence of surgery. The majority of probands were shocked to see the extent of the scars and the final shape of the breasts. All probands wanted an acceptable cosmetic result in order to be able to live with themselves. Thus, although they were very shocked, they were grateful to see the photographs to help prepare them for the cosmetic result. Two probands explain why:

*'I didn't realise it was so ugly. I thought that they could rebuild them with all the cosmetic, you know with all the plastic surgery around I thought that I could have them rebuilt back to normal, except for perhaps little scars' (P8).*

*'Yeah, so I think it's really good because I think you know, if you didn't show those pictures you would be like oh shit, what the hell is that' (P2).*

Looking normal was therefore very important to all of the probands but many were distressed and viewed the surgical results as mutilating because there were no nipples on the women in the photographs. This was one probands reaction on first seeing the photographs without nipples:

*'One thing that shocked, I think probably increased my shock and upset was I think the surgeon's results are very extreme surgery, isn't it, or in my mind, it is because of losing your nipples' (P7).*

These reactions led me to the question of why they were so shocked and what was it that they were expecting? The pictures of reconstruction on the internet were very different because the women in these images had kept their nipples. The perception of plastic surgery had also strongly influenced their initial expectations. For example, initially, some of the probands thought that the operation was a quick, relatively small procedure that gave an opportunity to improve their original breast shape, being more of a cosmetic procedure. This is precisely what proband seven was expecting:

*'I'm trying to view it, the breast side of things, as a really positive thing and that hopefully I get a fantastic pair of boobs and I come from having always had big boobs, of having to have enormous bras with ridiculously wide straps on my back and never wearing a backless top to now I'll be able to wear backless tops because I'll have a pair of, okay they'll be false, but you can't see that in a top and actually it's not a bad thing, they don't flop all over the place when you take your bra off' (P7).*

However, although acknowledging the need to see the photographs, after seeing them, the reality of the surgery had an impact that was unforeseen with many now reporting much anxiety about what they were going to look like. Why was this so important to them when surviving was the main reason for surgery? The potential impact on their femininity and identity was just as important as the need to survive, and this suddenly became real for them. Two probands explained why:

*'We need to see pictures so that we are going to be sort of happy to live with the cosmetic result. Even though you know it is only vanity but it's your femininity as well isn't it you know. And things like that do bother me and ah you want to, still want to feel like you can be feminine Umm I think umm the only thing I would say was that when I saw the pictures Umm I did find it quite a shock and umm (thinks) even though I do recognise that I shouldn't have done, because I knew what it was going to look like, it wasn't that, it's just that it was the reality hitting home' (P3).*

*'So I don't know, I just always think what are they are gonna look like after, and there's not gonna be any feeling in them, and to touch them and think oh I'm not going to have that. The scarring and I know I'm not going to be left with any nipple and things like I worry about all of that' (P5).*

#### **4.3.3 The Price: Being without Nipples**

The issue of losing the nipples, therefore, became a major dilemma for the probands. They all believed that they had no choice but to lose their nipples in order to maximise the reduction in risk, but removing the nipples was going to be a huge loss for them. The majority of probands did not want to lose their

nipples but equally, they did not want to worry about breast cancer developing later. One proband expressed the dilemma:

*'I wouldn't get rid of them (nipples) if I had a choice, but I haven't got the choice so' (P8).*

Why were the nipples so important? Having nipples was seen as 'normal' for women and represented their 'breasts'. They represented the look of femininity and womanliness. Despite believing that it was the right thing to do, the thought of waking up after the operation without nipples frightened many probands because they would not look like 'normal women'. The nipples were seen as the normal representation of a breast. Two Probands explain their concerns:

*'I am going to be scared not having the nipples which has scared me the most, um but like I say, I know I am doing the right thing even though it's going to be hard, I know I have done it for the right reasons Just the thought of like, like I have never liked my boobs, they not very attractive I've never liked my own boobs but um just the nipples is what like you look normal in clothes but when you take your bra off not having any nipples it will feel, it won't look right ,it will just be like the scars but I obviously know like that I can have reconstructive nipples like afterwards' Yeah, like what I was scared of was waking up after the operation and just not seeing any, I don't know why, but it was just the nipples which was' (P1).*

*'My biggest thing is how I am going to look. You know if I look at it clinically, will I feel like a woman not having any ovaries, fallopian tubes or real breasts' (P2).*



Indeed, one proband felt so concerned about life without her nipples, that despite knowing the risks, she chose to preserve them. She was not able to imagine living without her nipples especially after seeing a patient volunteer who had her nipples removed. Consequently, she was willing to live with the additional risk of getting breast cancer in order to maintain her femininity, identity and ultimately her happiness:

*'The scars didn't look as bad as I thought but I did think it looked odd that she didn't have nipples and that's been my main concern because I can cope with the plastic appearance of implants but I think not having a nipple would, is the thing I can't cope with. 'I've made the decision to keep my nipples because I kind of decided that you have to have an element of risk in life, I think we've talked about this before didn't we, you have to have an element of risk and I think that's, that additional risk would mean I would be happier with myself and I have to still live with myself going forward and have to look at myself in the mirror and I think that, at this stage would be too much of a shock for my system, not to say maybe in five years' time when nipple reconstruction hasn't improved again I might make the decision that actually I'll take them off and I'll have prosthetics or I'll have it reconstructed, I don't even know if that's an option. But at this stage, that's too much for my brain to deal with' (P7).*

The expected impact of the loss of the nipples and the breasts, however, very much depended on the significance of the breasts and the meaning attached to them. If for example, breasts did not define who the proband was, it was perceived to be less frightening. Only one proband explained how her breasts did not define her or her body image, her partner was very supportive which she believed made it less likely that she would be negatively affected:

*'So, in which case it wasn't scary because I kind of thought, and ultimately perhaps mixed in with my own feelings on my body image, that it's just flesh, it's something that I knew wasn't going to, or I hope still isn't going to massively affect me because I don't hold great store in these in defining me, I'm not somebody who gets my cleavage out or wears low tops so I don't know how much things are going to change for me in that sense. It's only recently I've thought about it as well in terms of I'm in a strong relationship where x has had his health issues and I know that it's only made us stronger and so I'm, you know it's now that tables are turning slightly. I don't know whether my decision would have been different and I would have felt differently if I was single, it's only recently I've thought about it in that way. I don't know whether I'd have had different body image issues if I didn't know that there's somebody at home who is going to be supportive' (P4).*

Was this the same for women who were single? One proband who was single had to wait for a delayed breast reconstruction because of other medical problems but she needed her breasts in order to feel feminine and to secure a relationship. She talked about how a *'flat chest'* is not regarded as feminine and was concerned about being *'dead flat'*, as this would restrict the clothes she could wear that made her feel more feminine. She, therefore, anticipated that the wait for reconstruction would be very difficult for her:

*'Well, they're (my breasts) quite important because I'm still single. I think the most stressful bit will be waiting for the reconstruction. It's just not feminine is it.? No, I wouldn't want dead flat because I know the problems you get with that and you're restricted by what you can wear, you can't wear a bikini and stuff like that, which I still do' (P8).*

Like the majority of probands, her breasts did define who she was and the anticipated loss of her breasts and her nipple, in particular, was also a major

concern. She was anxious about the effect that all this could have on a potential partner because she feared it would scare him away:

*'I think they'd (men) run a mile if I met somebody because they look so horrible. You never know, even at my age, that's the main thing really is that I'm still single and if I did get a new partner what would they you know, think? And when would you tell them and you know, it's that more than anything, it was the length of scars and it, and also what to me is important is that they disregard your nipple, they can't sew it back on' (P8).*

Despite probands acknowledging that the purpose of RRM was to survive, the cosmetic result was far more important than they initially disclosed. Some probands, for example, had tried to convince themselves that the surgery was not being done for cosmetic benefits but they still hoped that the result would be better than the photographs had portrayed. Proband five explained this dual importance:

*'Even though the surgery is to reduce the risk of breast cancer, the cosmetic result is still very important. When people say it doesn't matter what they look like, it does. You think to yourself I don't care, I'm having it done because of the risk-reducing bit and whatever but it does, it does matter. I think you look at those pictures right but you think no, mine will probably look better than that, I don't know, you look at those pictures and you think no and you still go ahead with it, don't you. It wasn't for cosmetic reasons anyway so, but still, once you have it done, you know it's still a bit of a shocker then when you see the result' (P5).*

#### 4.3.4 The Price: Loss of Intimacy

Although the majority of their husbands were supportive of their decision to remove both breasts, most probands did not know what their husbands thought about the actual loss of breasts and the effect it could have on their relationship. As one proband claimed *'he just doesn't want to talk about it'* (P7). The effect on intimacy especially caused concern because the majority did not discuss it as a couple and they had to guess what their husbands would think. Proband five, for example, was curious to know what her husband thought:

*'I don't know because he's not very open with me, so he wouldn't so much tell me but probably you know, if you've got on the phone to him he might say it to you, but not to me because like men are really deep in there, you know, so'* (P5).

Therefore, in addition to their own concern about how they would look, all the probands worried pre-surgery about how their husbands would feel about the change in their physical appearance, if they would be less attracted sexually to them and if they would be compared with other women. Proband three explained:

*'It's more about him as well as me you know and when he looks at other women, will he think of gosh you know, I haven't got such a feminine partner any more, you know all those things...'* (P3).

Probands talked openly and honestly about their anxieties of how their husbands would cope without their breasts because they were very much part of their sex life and it was difficult to *'read'* their husbands. For example, one proband described her husband as *'deep'* (P5). Others justified their lack of discussion as *'he didn't want to upset me'* (P4). Proband two was frightened that her husband would not be able to accept the breast loss:

*'Yeah, he (husband) has really been my biggest issue, he has been my biggest fear in all of this because I am constantly thinking what is he thinking, what is he feeling. How is he going to cope' (P2).*

Why was he her biggest fear? One of her main concerns was that her husband was only able to accept the RRM if he thought of it as a breast enhancement and this attitude concerned her because he was avoiding the real issues of her losing her own breasts, suggesting that he was not coping

*'He said so let's think of it this way, you're going in for a boob job' (P2).*

All probands and their parents were concerned about the effect of RRM on intimacy after surgery and were concerned that it may lead to sexual problems in the marriage and have the potential to end the marriage. Thus, in order for one proband to prepare herself for the changes which she perceived to be inevitable, pre-surgery she initially stopped including her breasts intimately

and then went on to stop having sex completely. The difficulties experienced were explained by two probands who were preparing themselves for the loss:

*'Right, my sex life has gone out of the window. I can't explain it, it's, I think, we're all human and I think well they're going to be gone soon so if I don't involve them in a sex life now I'm not going to miss them Right, so it's like I put them totally out of bounds and fair play to my husband he was like right okay, because let's be fair they're part of your sex pattern. I totally cut them out, I was like don't touch them, don't bother with them, I even stopped having sex, right, and it affected my sex life totally' (P6).*

*'And like my parents were worried about my husband's reaction like, um how will he um cope after it cause she said people break up and it's an intense thing and she said life isn't the same after it she said you will never look at yourself the same and he may not look at you the same' (P1).*

#### **4.3.5 The Price: Living with the Consequences of their Decision**

The decision to remove healthy breasts had therefore prompted different reactions from people, many negative. Most of the probands were questioned about their decision because of how they would look, even by those who had a previous breast cancer. This surprised many of the probands because they thought these individuals would understand the reason for their decision. Proband four shared a story about a difficult conversation with a work colleague who could not understand her decision:

*'one girl in particular who was a couple of years older than me but she'd had breast cancer a couple of years ago, she turned to me and said why are you doing that, you know you haven't got cancer why are you doing this to yourself' (P4).*

The majority of probands were under the age of forty and were being additionally judged because of their age. This was a consistent finding in their stories and experiences. Proband one like many, found it extremely stressful trying to convince people that it was the right decision for her. She constantly had to justify why her young age did not matter:

*'Then I got, felt like I was getting judged because of my age but still to me if I carry the gene, age doesn't matter to me because if you got it, it could happen at any time can't it'? Well, it's just the age people would say you don't need to do anything rash because obviously, I was, how old was. I was 23, I was 22 at the time um, I'm 23 now 24 in a couple of months. I was worried about their reaction because it was something they were like or don't do that yet you know you're too young but inside I knew I had to do it' (P1).*

Why was there so much resistance to their decisions from colleagues, family, and friends? Many people disagreed with the probands' decision to remove their healthy breasts because of the unnecessary effects on the body, comparing it to the removal of other body parts. Many people could not understand their decision because there was nothing wrong with them and they did not have cancer. Proband three explained why she had to do it but also the resistance she experienced:

*'Before I had the operation people did say to me well you can't go around chopping parts of your body off and things like that. But I was just so clear that it was the right thing to do for me so I kind of thought well it doesn't really matter if that's what other people think, I've just got to do this for myself. I think that is definitely a consideration that you think people might think you are being drastic...' (P3).*

Why did RRM attract so much resistance from family members? In particular, fathers worried about electing to have RRM so young and the effect on their daughter's body and relationships. Parents believed the loss would be significant. Proband three relayed her father's reaction to her decision:

*'And he (father) thinks, I think umm, you know I am still a young person, you know, he doesn't want it, it's all for me, it's not because he's being horrible or anything (laughs) I think, you know, to begin with, I think he was just like well, you know this is very drastic. He could understand the hysterectomy but this is very drastic, you know' (P3).*

#### **4.3.6 The Price: Marital Conflict**

As a consequence of the resistance from family, probands talked about the length of time it took to decide on breast surgery and how the decision was made more difficult when there was a lack of support or communication from their husbands and the wider family. The majority of probands had full support from their husbands, who were able to contribute to their decision. Some probands however experienced conflict. One proband experienced many problems in her marriage as a result of the decision to elect to have RRM. Her husband did not want her to have the surgery and thus could not contribute to the decision. She shared his response for example to their initial discussion on RRM, *'I think you should stay on screening'* (P2). Proband two explained the marital conflict:



*'My husband has been my biggest stumbling block since I have had this Ummm and I think he thinks that there is nothing wrong at the moment so why do anything about it. You know if it's clear now, why you are doing that. Umm, I think he is in denial, he finds it extremely difficult to communicate and we have had, recently we have had some, very, not nice conversations about it, because I am saying you can't go through life and not have any feeling or an opinion. Your opinion can't be the statistics are this and therefore you do this. There's got to be some emotion and some feeling and you have to talk about it, otherwise, it won't go away' (P2).*

As a consequence of this conflict and difference in views about RRM, there were tensions and problems in the marriage which persisted and which made her whole experience more distressing. She shared her sadness:

*'It's very difficult. You feel like lonely and isolated in your own life and marriage and home. There were times when I really didn't want to go home; I used to sit in here until 8-9 o'clock. You just think I just don't want to go home; I can't talk to him so what's the point' (P2).*

#### **4.3.7 The Price: The Fight for Surgery**

Despite the difficult decision to elect to have RRM and the journey leading up to surgery, it was difficult for some probands to have access to it. For example, two probands had to move hospital and change consultant in order to have the surgery. The first surgeon seen was not keen to perform surgery and tried to persuade them to have screening. They had to fight for surgery and were made to feel guilty because they did not have cancer. Proband five explained:

*'It's like you feel guilty. I dunno, for going there because there isn't anything wrong with you and you, they (the first surgeon) make you feel like if there's nothing wrong with you, why do you want to do it type of thing. Yeah, it's like she (first surgeon) was going why don't you just have the mammograms and whatever you know, it's not, they like sort of tell you, obviously it's not going to be like a boob job type of thing, and you know, I knew that before I went down there. I knew it was reconstruction. I think they are testing you. I think they think you know are you going to have these, perhaps you think you are going to have these lovely boobs when this finishes and it aint like that' (P5).*

Why did women have to fight for surgery when they were proven to have a mutation and guidelines had suggested that it should be offered? The reason for this reluctance was the psychological effects that breast loss could potentially have on the women, with little understanding of its long-term consequences. These probands had, therefore, waited a long time for the surgery because they had to move hospital and find a surgeon who was willing to perform the surgery. This experience had exacerbated their anxiety and they were angry at the unacceptable waiting time for surgery. The long waiting time was echoed by many of the probands and there was a general feeling that women electing to have RRM were not a priority. Proband six was very angry:

*'Treated differently, you feel like umm, I totally understand that people with breast cancer have got to be seen before me but when you're in the position you also feel, excuse me, I'm still here and I'm still waiting and there's always going to be people diagnosed daily with breast cancer so when do I fit in' (P6).*

#### **4.4 The Altered Child's Trajectory**

The literature review identified the moral obligations of parents to children once a mutation is identified within the family. Work by authors such as Liede et al., 2000; Lodder et al., 2001, Hallowell et al., 2005, d'Agincourt-Canning, 2006a, and McEwan., 2011, remind us of such obligation to be tested and complexities when considering children. This thesis, however, offers an understanding of how parents viewed the child's trajectory as altered as a result of disclosure, of living with risk and living with a mother who has elected to have risk-reducing surgery. In this study, probands, husbands, and relatives in addition to feeling a moral obligation to their children to do the right thing, were extremely concerned about their children's future. Parents feeling the most responsible for the inheritance of a mutation, believing that people would now treat and view their children differently. As a consequence, they experienced guilt and fear and this thesis provides an explanation for that guilt and fear. The horizon of the altered child's trajectory, therefore, resulted from an interpretation of the fears and concerns that the participants now believed were facing their children and future generations by introducing risk into their lives, including having surgery. It also resulted from the potential of getting cancer and the possibility of their children being without a mother, thus how children's lives would be altered. This next section will focus on the probands' experiences of concern for their children and how the interpretation evolved leading to an 'altered child's trajectory'. It will demonstrate the attempt made by the probands to preserve the normal child trajectory.

#### 4.4.1 The Kids Sake - Cancer Free Worry

Before embarking on a mutation test, surprisingly, probands reported that the initial focus and the importance of their need to know was for their children's sake, rather than for themselves. They talked of a moral obligation as a mother to be tested and to be pro-active, for their children's sake. This concern for their children was echoed by the majority of probands who were afraid of not surviving or being able to care for their children as this would alter their child's life trajectory. Two probands explain:

*'Um, and I was quite paranoid, I kept thinking any symptom, this could be cancer because I knew I could have been carrying the gene, any little thing was like setting my mind off thinking or it's something you know, is seriously wrong with me. I was back and forth to the doctors just quite paranoid really thinking or this could be cancer so I needed to know if I carried it or not and I wanted to be able to then do something about it cause I got two kids as well and I wanted to be able to say to them if anything does happen, mammy done everything she could to try and stop it so' (P1).*

*'you know, and feeling that for our children and x knows that I've always like thought that I'll never see, even when we were younger I used to say well I'll be the first to die, you know. I just always used to think I'll never see them get married, you know which is awful isn't it' (P3).*

The genetic test and subsequent surgery was therefore seen by the majority of probands as a means of preventing their death for their children's sake. It was also seen as the right way to behave and the right thing to do as a result of being armed with the knowledge. Proband six strongly believed that having

the test and surgery would save her life and the lives of others and she urged other women with children to do the same:

*'I would say to people that have cancer in their family, is for immediate family to go down the same route, to go to the genetics, get themselves tested, have this (surgery) as an option because there is help out there, don't hide in the corner and wait to die, because that's what you could be doing if you don't go. So in my experience, it's better to get yourself looked at' (P6, 12 months).*

As with the decision to be tested, probands believed they also had a moral obligation to make the decision to elect surgery for their children's sake. Throughout their stories, probands gave various accounts of their decision to have RRM and their moral obligation to protect their children as a mother. Proband one, for example, reflected on the day she received the test result and thought about RRM, she thought *'I've got to do it for my kids'* (P1). Why did probands believe that they had to do it for their children and not themselves and only RRM would allow them to be there for their children?

Probands believed that surgery would prevent breast cancer from occurring rather than waiting for cancer to develop which they all believed was inevitable. They feared cancer could be missed on screening mammography. Two probands had experience of cancer being missed on both their mothers' and their sisters' mammograms. As for all probands, they wanted to avoid breast cancer and treatment at all costs, to protect their children. Proband six explained why it had to be surgery:

*'So, I thought no I can have a mammogram and still end up with breast cancer and chemotherapy like my sister, so no, let's do the preventative like. I said well they've offered us preventative surgery or regular mammograms but because my mother and sister had misdiagnoses, in the beginning, the mammograms missed the cancers. I don't trust mammograms' (P6).*

They were electing RRM for their children's sake because it was the means of preventing any chemotherapy or distressing treatment that the children would have to witness, in addition to the possibility that the probands could die. For example, RRM they believed, would prevent their children from having any psychological suffering, allowing them to have a cancer worry free, childhood. They did not want to burden them with this worry. What suffering were they trying to prevent? If breast cancer did occur, their children would have to witness the distressing effects of the treatment such as vomiting from having chemotherapy and the stigma associated with hair loss, thus a disruption to normal family life. They wanted to prevent that from happening because of the perceived long lasting psychological effects. Proband two reflected on the effects of cancer treatment on her son at the age of five when her mother-in-law had breast cancer and the need to protect him:

*'Umm, I don't want them to see me go through lots of surgery with chemotherapy and losing my hair. My son was five when my mother in law had breast cancer and lost her hair and he talks about it now so it is obviously buried in his head. I don't want to put that risk in their lives; I don't want them to go through it' (P2).*

#### 4.4.2 Preserving the Child's Trajectory - Maintaining a Normal Childhood

Maintaining a normal childhood for their children by excluding any risk or stigma attached to having cancer and undergoing treatment was poignant for many of the probands but particularly proband one, who explained what it was like for her as a child whose mother was undergoing treatment. She had vivid memories of how disruptive this was on family life. She wanted to prevent that happening to her children at all costs:

*'I said to my mother if you could go back in time and like, if my father would have had the choice, would he have wanted you to prevent this so obviously he wouldn't have had to go through and my mother wouldn't have to and us kids cause it was hard and I just can remember being a kid and I don't wanna do that, I would rather them see me like this for the next couple of months, getting over it than going through the chemo and we were going to like people's houses and we weren't seeing, you know, when she was going for chemo and everything, we were out of the house a lot and I would rather do this now' (P1).*

Probands were also concerned about their children's risk of cancer. Why were so worried about their children's own risk of cancer especially being so young or yet to be born? Probands soon realised that their children, both girls and boys had the same 50% chance of inheriting the gene which they perceived as extremely high. This meant that in addition to worries about possible cancer treatments if they themselves were to get breast cancer, they were now faced with fears about their children having a positive mutation and getting cancer. This fear had now changed their child's life trajectory. For proband two this was her main concern:

*'I think like I say, you don't think it's you, so my first reaction when I was told was what about my children because this 50% thing obviously because it's your genes, was more of an issue at the time for me' (P2).*

Thus, all probands were extremely anxious and frightened about passing a gene to their children and to future generations. They were particularly scared that once the children knew, they would feel differently about themselves, be seen as different by others and be burdened with the need to make major decisions about having a test and having children of their own. Proband six explained the extra worries she now had:

*'The worry, it's not just about myself, I've got a daughter and a grandson and a son who wants children, it's about them as well' (P6).*

However, most of these concerns and fears were gender specific, with a focus on girls and their future risk and decision-making. Their fear concerned their daughters, specifically the decision of electing to have RRM which consumed the majority of probands. Probands experienced fear and sadness when thinking about their daughters because of the implications for their breasts. This meant that they could not relax and enjoy their children growing up. Two probands share their fears:

*'And then you've got to think about your own kids, that's the biggest thing for me and I think oh my god I'd hate her to go through all this so what I'm really, really frightened of is*



*watching my daughter even contemplate going through it. I look at her and I just want to cry because she's got to make these decisions one day and I find it so hard' (P2).*

*'Yeah, just the way, if I'm in the bath and she walks in trying to cover myself, trying to dress, trying to make these stupid decisions which you know and I think oh my god, I'd hate her to go through all this' (P5, eighteen months).*

The process of testing, disclosing a mutation in the family, decision-making, and fears about RRM for all probands had now shifted to concern for their children. All these factors had changed their children's life trajectory. Their anxieties related to the timing of telling their children, but mainly to the decision of electing to have breast surgery, including the effect it could have on the way they and others would perceive them. Proband four who did not yet have children was already worrying for example about dealing with a daughter who asks for advice, concerned about the amount of pressure that her daughter would be under and the effect it could have on her daughter's future life and relationships:

*'Yeah, I've got a risk in life, for life, it's easier for me at the moment as well because I don't have children, when children come along that, and that's made me worry as well about the relationship I'm in because I'm very comfortable and confident but if a twenty year old daughter of mine, you know at what point do you tell them, at what point do they decide I can't do this yet mum, you know no boy will look at me and at what age, it puts a pressure on them and you think oh we didn't get together until I was thirty but it could be that, you know at thirty the risks are still high and if they're still single as I was at thirty*

*and career wise as well, I don't know it will affect their career so there are all those kind of things to worry about' (P4).*

#### **4.4.3 Hope for Future Developments in Medicine**

In order to manage the worry and fear which persisted up to the eighteen-month interviews, about their children's altered life trajectory, many probands wanted to believe that their children had inherited their husband's genes or hoped for alternative future treatments that either replaced the need for surgery or could stop the gene from being passed on. Proband two could only deal with it all by thinking this way:

*'And then you've got to think about your own kids, that's the biggest thing for me. I've said at the moment we cannot control it and in my head I'm saying she hasn't got my genes, she's got his and that's what makes me get through it but now and again something will click and I'll think oh my god and I'm looking at her thinking. You know there might be a pill to fix the faulty gene' (P2, eighteen months).*

These concerns for their daughters were echoed by all of the probands. Why was this the case when their daughters may make different choices or may not have a mutation? It was because of the devastating effects of surgery on the body and potential relationships. Many hoped that there would be an alternative to surgery for their daughters and future generations because of their own experience and effects to the body. One proband relayed a difficult conversation that she had when her daughter had already started to worry about her own risk instead of enjoying her youth:

*'Yes because she (daughter) said to me today, what could I do and I said perhaps like things will be different and it'll just be like a little tablet she can take when it comes to, but she said what would you do and I said I don't know, I don't think I would have done it(had surgery) if I had the choice again. Had I known now, I would have felt like this, it's just physically, I am absolutely drained' (P5, 6 months).*

#### **4.5 A Separation from Self - Post Surgical Experiences**

The final horizon of understanding, 'a separation of self' resulted from the interpretation of the experiences following surgery including a separation from the fear of breast cancer. The literature review had identified that body image, sexuality including femininity and identity are impacted as a result of RRM (Hatcher and Fallowfield, 2003, van Oostrum et al., 2003, Brandberg et al., 2012; Gopie et al., 2013). In particular, the latest studies by McEwan, 2011 and Hallowell et al., 2012 identify the consequences of such surgery and offer useful insight into the experiences. However, the latest studies also focus on the experience of undergoing RRSO and include women with breast cancer, whereas the focus of this study is BRCA positive women and RRM. Thus, the interpretation of the experiences of the eight probands in this study, offer insight and a contribution to existing knowledge as to the reasons why women experience such impact, which led to the expanded horizon of understanding. The next section, therefore, presents both the positive and negative effects of electing RRM through the interpretation of the probands experience leading to the third horizon of understanding 'a separation of self' and the expanded horizon of understanding that of 'disembodiment'.

#### 4.5.1 Separation - The Fear Has Gone

In support of many of the studies identified in the literature (McGaughey, 2006; Hallowell et al., 2012; Hagen et al., 2014; Glassey et al 2016), all probands felt a great sense of relief post-surgery because the risk and fear of breast cancer had gone. This was an immediate effect especially if no cancer had been found, they, therefore, experienced a separation from the fear. One proband, for example, explained how she now *'never gives it a second thought'* (P4). Another commented on how the *'risk has gone and so I never think about it'* (P2) and a third made the point *'Great, I have got nothing to worry about now'* (P8). Proband three explained the overwhelming resolution of fear when she woke up from surgery and the enormous amount of relief knowing that no cancer had been found:

*'Fear has totally gone, in like a massive way, in a way that, because I remember friends saying to me oh you'll have such relief when you've done it and I must admit at the time I thought I'm sure I will feel relief but I think I will feel scared about what's happened to my body. But actually, when I woke up I just felt such relief and no fear about what had happened to my body, it was quite sort of empowering really I just thought oh you know, no cancer now' (P3 6 months).*

*'And felt a huge sense of relief afterwards, huge. And to know that it had all come back clear, and x had her mastectomy and reconstruction in early November/late October but it didn't come back clear. Umm so it came back, first of all, they said that they that there was pre-cancerous cells, and then they actually called her in to say they had found a tumour in the nipple duct' (P3, twelve months).*

This resolution of fear continued for all probands and was evident at eighteen months post-surgery. It was a huge and difficult decision to elect to have RRM but many probands had feared cancer for such a long time and had now been separated from the fear. They had elected such surgery in order to be freed from this fear of cancer. Proband five explained:

*'I don't even think about the fear any more. I think of all that I've done and to me, that has eradicated that because I haven't gone through all that to have fear so that's gone. Oh god, yes for me it has, yeah, I don't even think about that' (P5, 18 months).*

#### **4.5.2 Separation - No Return to Normality**

Although the fear of cancer had gone, the majority of probands expected to recover from surgery more quickly than they did and return to normal life. They underestimated the pain and the length of time necessary for recovery. This delay in recovery occurred for a number of reasons. If there were complications with infection (P2, P4, P5, P6, P7, P8) or probands were dissatisfied with the cosmetic result (P2, P4, P5, P6, P8), or needed further surgery, this delayed a return to normal life. One proband, for example, lost both of her implants due to infection and this further delayed recovery. For all probands, it was important to get back to work and to normality but many were experiencing and dealing with complications over a year post-surgery and waiting for more surgery. Proband four explained her problems:

*'The infection I had, actually that put me back in hospital for 10 days and that was more painful than any surgery I have had. Well I guess over the last twelve months it's hard to ignore the fact that the implants had to come out, so it's a year on, I hoped at the time when I was making the decision originally that it would be one operation all done and dusted, twelve months on I would be you know, kind of well and truly over it and moving on whereas at the moment now I'm still waiting for my final operation. So, the cycle isn't complete yet, so the downsides have been that yes it hasn't quite been the smooth process that I'd planned. and the other side of the coin in that period after the implants had to be removed I had something like three or four months with nothing at all, with completely flat, no implants in and I found that quite liberating and its only kind of just served to remind me that I'm quite comfortable in my skin, it was definitely the right decision for me' (P4, twelve months).*

The majority of probands also experienced the surgery itself as much more painful than expected and there were a number of unexpected physical effects even after six months, which prevented them from returning to normal. Many experienced a lack of strength and mobility in their arms for example and also expected the recovery to be as quick as the ovarian surgery. It was therefore frustrating and a big shock when they were not as active or could not do their usual activities. Two probands explain:

*'I can't put any weight on my arms; I couldn't get up from that position if I tried. I noticed through physical things when I try to do something the lack of strength or the pull or the aching because of, because I use my arms a lot but I don't sit and think look at the state of me or nothing like that' (P6, six months).*

*'I probably under estimated the amount of pain and the mobility if that makes any sense, because from the surgery I had before(hysterectomy) within two weeks I felt fine, I was*

*up and about driving and things but this time after five weeks I still wasn't driving, which for me is a massive shock because I'm so active but in my mind and physically I knew I couldn't do it because it was too much pain and I'm not sure, because I've researched it as well post op, whether the pain I was getting was due to the muscles because they were tight or whether it was the surgery and probably a bit of both. It hurts all the time and it's the weirdest feeling because it feels heavy; it feels really, really heavy' (P2, six months).*

Indeed, the pain, the fatigue and the effect on their lives continued for most probands beyond twelve months and this emphasised what an ordeal it had been for them. It also emphasised that they had not realised the enormity of the surgery or had not prepared themselves for it. Furthermore, despite all the physical and emotional effects experienced following surgery, many of the probands soon realised that they had returned to work too soon. On reflection, they had not given themselves enough time to come to terms with what had happened. Thus, for many probands, this long process was continuing as there were still outstanding operations to complete their reconstruction which some family, friends and work colleagues did not appreciate. Two probands explain their experiences and the unexpected consequences of surgery:

*'I think I under estimated it, yeah, I think I did and also because it's been longer than I thought it would be. I think in hindsight it was a stupid thing to do because I didn't give myself the opportunity or the time to come to terms with what went on. Because as soon as my butt was sitting on that chair, because I was back in work and I feel that, not that I want anyone to feel sorry for me because I'm not like that or I don't want people saying are you okay, because I'm not like that either but I feel as if, not just work but my family, have certainly oh well she's back to herself, where behind closed*

*doors, they don't appreciate what I've still got to go through' (P2, six months).*

*'I think I was completely and utterly naïve in terms of the pain and I am not a very patient person and so it's about a year of your life really and I don't think I really appreciated the level of pain, how rubbish I feel at points, not all the time but how all-encompassing it was going to be' (P7, twelve months).*

#### **4.5.3 Separation - Regret and Loss of a Former Self**

Despite the long recovery and emotional effects, seven of the eight probands had no regrets about having elected RRM and believed it was the right decision. Proband one was particularly sure:

*'I am so glad I done it; it was definitely the right decision (laughs). Like my mother was saying, everyone was saying, any regrets? I said no I will do it all again tomorrow if I had to, for me it was like 150% the right thing to do' (P1).*

For one proband, however, there was a deep sense of regret in electing RRM. The drastic changes to her body and the way she now looked, made her question if it was worth doing. It had affected her intimate relationship with her husband and she wanted to hide away. Proband five believed that screening would have been the better choice because she now found her body repulsive:

*'I'd say about two or three months ago I was sorry I had it done, I kept looking at them and thinking oh my god they're gross and whatever. He can't touch. No, no. He wouldn't want to anyway, the way they look. He doesn't say anything*



*but. Yeah, I wouldn't let him touch them. I don't like him looking at them, no they are disgusting. Yes disgusting, they are, honestly, they are. I wouldn't have had it done if I had known I would feel like this, would put up with the risk and just be tested regularly. Yes I wish I'd done that to be honest with you (chosen screening) rather than go through with all this because life is so short and you don't realise how short life is and then you think you're putting yourself through all this and I don't know, is it worth it' (P5, twelve months).*

Why did she find her body so disgusting? Despite seeing the photographs, like many probands, she was expecting 'proper' breasts. Instead, she felt she 'looked like a man', masculine, not feminine and this was not regarded as normal. To be a woman was to have breasts that both looked and felt like breasts. These expectations were echoed by many probands. Proband two for example, similarly believed she was 'looking like a boy' because she had no nipples and her breasts were flatter. Proband five explained why she thought she looked like a man and what she was expecting:

*'I didn't think it met my expectations after I had it done because you know what it's like, I think you have this picture in your mind, you're going to have proper boobs and it's not, it's basically not like that is it. Because there was no nipples and I thought it looked a bit like, like a man' (P5).*

#### **4.5.4 Separation - No Longer a Feminine Body**

Looking feminine and like a woman should look was important for all the probands who were still relying on reconstruction to replace their breasts and make them feel feminine. However, this was not the case and many probands

hated the way they looked. As a consequence, the majority now avoided looking at themselves and did not allow their husbands to see them undressed. Thus, many probands tried to disguise their bodies by wearing clothes that were different to what they were used to. Proband five particularly hated her body:

*'I just don't look at them; I try not to look at them because I think if I looked at them too much that would bother me. I'd probably cry because of the way they look. Yeah, obviously miss the way they look because they obviously looked better before didn't they. Oh my god, I hate my body. I don't know, I didn't mind. I suppose you dress differently. I think I hide under baggy clothes and stuff now. Oh my god, I hate the way they look. It's just their totally shapeless and there's like, I mean I've got that much hanging skin underneath so there's more hanging skin than there is boob, too be honest' (P5, twelve months).*

The efforts to hide their body and maintain different disguises was exhausting for many probands especially if they had not told their children. One proband, for example, whose husband had insisted on not telling their children about the surgery, was constantly under pressure to hide away because of the shame she felt and she did not want to upset her children. Proband two explained why it was so difficult for her:

*'So, I, as much as I disagree, it was easier to agree (not to tell the children). The frustrating thing about it is for me with it, is getting changed and bathing because my son walks in and out and all sorts right and last night he unlocked the door and came in the bathroom and in the mornings, we've always done it, the kids get up in the morning and come into our*

*bedroom, in our bed and watch TV. I find it difficult now because all my clothes, his are in a separate room, his clothes, he sleeps in our room, all my clothes, everything are in my bedroom and I'm trying to get out of the bath, across the landing and I'm like this with the towel and I'm finding that that pissed me off this morning really bad because the kids were in bed, in our bed, I get out the bath and I've got to adjust a towel around me and I'm trying to hide' (P2, six months).*

Although seven of the probands had no regrets about RRM, many of these probands were also 'unhappy' with their body despite reconstruction. One proband who was advised to undergo a delayed reconstruction equally found her mastectomy scars 'horrible' (P8). Why did she think they were they so horrible? The scars were uneven and looked 'unfeminine' because they were 'flat'. This had adversely affected her confidence resulting in a total life change in which she became totally housebound:

*'After I had the mastectomy, it made a bigger difference than I thought and I didn't go out, I haven't been out because I don't have any confidence. I haven't gone out and I used to go out quite often with my sister, which I am going to do again once things are finished. Even sunbathing out the back, because of the lumps, it is just horrible. It's just not feminine is it' (P8, six months).*

Why did the mastectomy scars affect proband eight so much and result in her being housebound? Whilst waiting for her reconstruction, a flat uneven chest made her look like neither man nor woman. This was unattractive, making her too conscious to go out, scared of what people would think. She felt and looked different, she explains:

*'It's ugly. You look like a man, don't you? I don't feel female any more if that makes sense? I was very conscious of it, that I looked like a transvestite or something' (P8, twelve months).*

#### **4.5.5 Separation from Self - Disembodied**

Looking 'normal' and feeling normal like a 'real woman' should was crucial for all the probands in terms of 'being women' and maintaining a sense of self. However, the majority were devastated about the way they looked because they did not feel themselves. This was because their bodies had drastically changed, they had many scars, they had no feeling in the reconstructed breasts and their breasts looked unreal. It had therefore been a major life-changing event with many experiencing that the loss of their natural breasts had split them in two, separated them from their whole self and their husbands, as though they had been disembodied. Proband two explained:

*'But I don't like what I see, yeah just looking and thinking what can I wear to hide this today. I just see scars and 'broken', I don't know, especially because I've got so much surgery I just look, like dot to dot and I just feel as if, I don't know. He always says to me it doesn't matter, you're still you, he's said that from day one in fairness to him and he doesn't matter you've haven't got them, it's fine. But I think deep down it does, with him and I think that makes me think differently about myself. Yeah, I think it just looks a bit, I don't know what the word is, 'broken' I guess. Whereas before you would have had feeling in your chest but I've got no feeling in my chest, I've got nothing there. Not real, it doesn't feel real' (P2, twelve months).*

The majority of probands therefore looked and felt very different about themselves and their bodies, which did not significantly improve over time.

Many continued to avoid looking at themselves in mirrors or avoided taking off their clothes because it was too traumatic, reminding them of the intact body they once had. It also reiterated the drastic life changing effects on their body and the loss of who they once were. Proband six explained:

*'You look at yourself differently. I often stand there, you know through the stages and look in the mirror to have a look, and they're obviously not as pretty as a normal breast would be out of clothes, I look at myself and think oh, don't look in the mirror. I tend to look and then look away, but not just because I don't know, it makes you look at yourself differently. You sometimes feel the same and yet you look different and I think people need to be prepared for that. It's not a pretty picture then, it isn't' (P6, six months).*

Consequently, as a result of this drastic change to the body and the way probands now felt about themselves, there had been major life changing consequences, especially for relationships. Proband six explained the effects on her relationship:

*'Because everything is different. My feelings are different, my body is different and everything is different for the two of us' (P6, twelve months).*

Why did probands feel so different about themselves and why had relationships changed? It was difficult for most of the probands to accept the reconstructed breasts as 'their breasts' or refer to them as 'my breasts' because the breasts did not move like their real breasts, look like real breasts or behave like real breasts for example even with time, the sensation was still altered which affected their relationships. The majority felt disconnected from

their breasts, disembodied, as they tried to accept a new self. Proband four explained how she viewed herself:

*'It's weird because when I talk about them its 'these don't move', but I haven't thought about it in the sense of these are mine, it is just this is me now. More sensation has come back and so that makes it easier. They still feel numb in certain places. I have knocked into things and not realised which is fine because it doesn't hurt but I realise when I am about to knock a chair over or something. I couldn't feel them. They don't fully feel like me' (P4, eighteen months).*

The probands did not feel 'themselves' anymore because the drastic change in shape, size and sensation of their breasts especially when they were undressed, was not who they once were and reminded them of what they had lost. Proband six explained why:

*'It doesn't feel like me anymore because I was voluptuous, I was a big girl and now I haven't got any and I feel a little bit that I'm unfeminine because of my shape. Not so much in clothes, in clothes, I can handle it but out of clothes because they're behind that pectoral muscle, if you bend or make a movement they distort so badly that that's when I don't like looking at them' (P6, twelve months).*

#### **4.5.6 Separation from The Intimacy**

The most drastic changes in relationships involved the lack of intimacy and probands were willing and needed to discuss how their sex life had been negatively affected. The majority of probands complained that there was no feeling for them in the breast thus could not get aroused or feel any pleasure.

Their breasts did not feel natural to those who touched them, making probands self-conscious and embarrassed. The implants also felt firm which meant that their husbands found it difficult to touch them, often avoiding their breasts. Many probands, therefore, continued to cover themselves up or avoid intimacy. Proband two explained:

*'And if you touch it it's really, really strange because there's no feeling there. I think he struggles that I avoid, if we are intimate I've always got a vest on, he finds that really hard. He doesn't touch at all. He doesn't even touch me. Because he's never tried, so I don't think he can. I think he thinks it's horrible but he won't tell me that. He won't say that to me, but I know that' (P2, twelve months).*

Wanting to look and feel womanly, feminine and attractive especially during intimacy was essential for all the probands but many probands avoided sex because they did not feel attractive and were ashamed. Even for the minority of probands who did not have any complications with the surgery, who were pleased with the outward appearance of the breasts and who did have sex post-surgery, it was still very difficult for them to feel feminine and attractive. Why was this the case? There appeared to be a mismatch between what they saw in terms of an acceptable breast shape and what they felt. It is proposed that these women, like those who avoided sex felt this way about themselves due to feeling 'disembodied'. Proband three for example, despite being pleased cosmetically, still did not feel like a woman:

*'As time's gone on we've found, you know I've been totally happy with the reconstruction, really happy with the scarring*

*and everything else but I have found it difficult from a womanly perspective and that is something that I think, you know initially so relieved to get over the operation, so pleased about the cosmetic result and things but actually you know as time goes on and your mind settles I do find it hard to feel womanly which is odd because from the outside nobody can tell anything different but I think just psychologically I do find that quite hard. Yes I think it's just feeling like a woman, it's hard you know, sexual relationships is hard and because there's not much feeling, you know at the end of the day, it's hard. I don't really feel attractive you know, even though I've got a much better cleavage than I had before' (P3, eighteen months).*

Indeed, as a result of many probands feeling this way, some reported taking anti-depressants in order to cope with the effect that RRM and reconstruction had on their sex life. The anxieties that probands had pre-surgery about their husbands finding them less feminine and less attractive were still vivid. Many probands believed that the lack of sex in their lives was because their husbands were struggling to adjust to their bodies. For one proband this had a major psychological effect which continued long term. She explains:

*'I look for reassurance, I just look to see if that's what does put my husband off (sex) or not and he says I love you anyway, but I look for reassurance. I tend to cover up a lot more, clothing wise. I have, I had to go on Citalopram. I'm still on citalopram but I think without that I would be low, I do need a lift still. It has taken the stuffing out of me basically' (P6, six months).*

*'Well I didn't really know how I would feel, I was hoping I would feel better for it which I said I do because we eradicated the fact that I shouldn't have cancer now. I didn't know how I was going to feel, I suppose okay, the only thing I would say is affected in my life is my sex life as I told you before' (P6, twelve months).*



Wanting to discuss their sex life and warning women about the long-term effect on the sexual relationship was something that many of the probands felt strongly about and requested that it be an integral part of the pre-surgical information. Proband five explained the significant effect the surgery had on her sex life even after eighteen months and why women should be forewarned:

*'I think perhaps you should bring into your talk beforehand the fact that it does affect your sex life because obviously it's a sexual organ and it'll be gone, never to be replaced, it's not just gone for a day, it's gone for life and it's a big effect on your life, perhaps you should mention that to people' (P5, eighteen months).*

Many of the probands, therefore, felt different, very unattractive and unfeminine. In order to try and accommodate these drastic changes to their body and to appear more sexual to their husbands, some probands adopted various strategies to try and look more feminine. Proband four, for example, wore pretty underwear to disguise the results of the reconstructed breasts:

*'Just keeping a little bit of the ugly away. Although I don't think of it as ugly but they are not attractive. It's not to say the ones I had before were that brilliant either but just keeping away the scars and just wearing a nice pretty little cami top or something like that it actually helps to bring back a lot more of the feminine, sexy back into the relationship' (P4, eighteen months).*

#### 4.5.7 Separation - Nipples to Preserve and Restore Femininity

To improve the way they looked and to feel more feminine, probands chose to preserve the nipple (P7) or to have a nipple reconstruction with the areolar complex tattooed (P1, P3, P6). Why did the nipple make so much difference? Nipples helped to make the breasts look more real, like they should look. They also differentiated probands from looking male and masculine, a factor which was extremely important to them. Nipples also helped take the attention away from the scarring and the fact they looked unnatural. Two probands explain:

*'I'm thrilled to bits, I'm so happy with it as well. I didn't know whether I felt like I needed them done, but I'm glad now., I've always been happy from day one so it's just like the finishing touch I just can't believe how real they now look' (P1, six months).*

*'Because I've had the nipples tattooed on haven't I, so that made me feel a bit better immediately because you had a focus, you looked before a big bump, now you look and there's a centre of attention. I think it makes them more realistic' (P6, twelve months).*

Indeed, some probands who were frightened about losing their nipples pre-surgery were able to get on with their lives, feeling more *'normal'* after having the nipples reconstructed and tattooed. The husbands of these probands were also very pleased with the nipple surgery and tattooing which was important for the probands, who very much considered what their husbands thought of their bodies. Proband one explained:

*'Marvellous, it's like everything was normal like, even from, he was brilliant, he always has been but as soon as I was back to normal in myself, everything else went back to normal. Yeah, completely part of me, I think he feels the same, well I don't, I think he's really happy and you know when I had the nipples he was thrilled, they were amazing, you know they couldn't have done a better job if they tried' (P1, eighteen months).*

However, having just a nipple reconstruction without the tattooing did not have the same effect because this still made the probands still feel masculine. The tattooing of the areolar therefore completed the process and made the breast look more female and thus the probands feel more feminine. Proband three explained the difference an areolar made to her femininity:

*'This is good and it has now all come together. It just shows how important the tattooing is, just a nipple on its own made me feel in some way masculine because men have pecs, the nipple maybe you don't see, it's the areola. I don't really know why psychologically I felt it because obviously, I had something, I had a nipple whereas previously I hadn't had a nipple but I just felt a bit manly, I couldn't really put my finger on why and I thought that's really strange because I was so excited about having nipples and it wasn't like I was unhappy with anything because the result was great but I just didn't feel very womanly for some reason' (P3, eighteen months).*

The effect of seeing nipples made a huge difference to the way probands were able to live their lives. As predicted pre-surgery, many probands confirmed the positive difference having nipples would make. Proband seven, who was the only proband to preserve her nipples, reflected on her decision and the result. It had helped her to cope by disguising the scarring from the outset. She believed that her breasts looked more normal with her own nipples:

*'Yes, I'm really happy about it. I think it was the right decision for me at the time and actually I, yes, I think so and I think it's helped disguise the scars as well because they're obviously a bit darker the scar just doesn't look red and you can't see the scar at all. So, I think visually it's helped and I think it's helped me cope with it all. They look to me like normal breasts' (P7, twelve months).*

However, there was a difference in seeing a nipple and feeling sensations in the nipple, which remained a challenge. Because there was now no sensation even in the normal nipple, coupled with the reconstruction, proband eight like many other probands also felt disconnected from her breasts:

*'The nipple area I can't feel anything, it's literally just the sensation of the bag pushing on the inside so that's strange and I suppose I'm never going to, that's not ever going to come back, is it. There's nothing there. It is a bit weird that you can't feel anything and that's a bit odd really' (P8, twelve months).*

#### **4.5.8 Separation - Still Not Me!**

Despite the efforts that probands made to look and feel more feminine, many were still experiencing problems with the way they looked and felt about themselves and their bodies at eighteen months. Some probands who had completed their surgery and thus were living with the final cosmetic result were now very different people. Proband two, for example, who was extremely self-conscious believed the reconstructed breasts were not part of who she once was and she shared her experience of how much she missed her own breasts:

*'I don't know, I think they're just part of you (your own breasts) and you don't realise until they've gone, I didn't realise and I don't think I'll ever be the same, I don't think I was very open anyway if that makes sense, I wasn't one to walk around my house or with my friends and just strip off, I was never like that. This has just pushed me completely over the other side you know. I feel extremely conscious because I don't like the look of them' (P2, eighteen months).*

Indeed, those probands still waiting for further surgery could not move on with their lives until it had been completed because they hoped it would make them feel more like themselves. As a consequence, probands continued to be very self-conscious about their bodies. Proband seven, for example, who was obsessed about her appearance needed to have her reconstruction finished in order to feel and be herself:

*'Well I'm so fanatical about my appearance, not that you'd know it looking at me at the moment but I'm very self-conscious about my weight, always have been, very self-conscious about my stomach even down to like my appendicitis scar which hasn't healed particularly well, I've got a lot of loose skin on my tummy and to not have any breasts and I've always had very large breasts for my size so I've normally been, I've always been about a double D or an E, possibly going up to an F at some points. So, at the moment, I've got a very small chest for me at the moment so I can't wait for them to be finished' (P7, twelve months).*

The difference in the way probands perceived their bodies was not only related to the physical aspects of the surgery, but to the whole process they had endured. This included the inheritance of the gene, the constant fear of cancer, electing to have the surgery, the worry about children and the process of coming to terms with everything. Most probands felt that they had now

changed as a person. They had become far more concerned and anxious about their family and the whole journey had been traumatic and all consuming. Proband three reflected on the journey:

*'I still think that when you are in a big worry sort of mode and this has been going on for years because of Mum and x and then all the decisions around this, I think it changes you to a point. It makes you more serious and worry about the people that you love, you just want to protect everybody and I think that now I have done everything, I would like to get back to a point where I am, sometimes I think I just feel a bit low but not because I am depressed I think it is just the amount that has happened. That has been over a period of 13 years and it hasn't all been hard but there has been a lot of difficulties and I would like to feel a bit more lifted and I think that will come now that everything is moving on' (P3, eighteen months).*

#### **4.5.9 Help to Restore Self**

As a result of all these changes to their bodies, all probands expressed a need for support, information and psychological support from the professionals both before and following surgery. Some probands did receive support from their family and friends with physical activities such as shopping and house work, but in other families where there was tension about their decision to proceed with surgery, little physical or psychological support was received. Proband six explained:

*'They (family) don't ask really, no. Even though they've been through it, like I say we've always been there for them, there's us we've had cancer, there's you, you haven't and you've done this of your own accord, so they don't really ask us how we are' (P6, twelve months).*

What Psychological support was needed by probands and why did they feel this was necessary? Access to counselling and information was deemed important especially post-surgery, including access for their husbands who many probands felt were not coping. Many of the probands recognised that their husbands did not have anybody to discuss the whole experience with, especially any intimate problems, especially if their husbands had been sworn to secrecy. In particular, probands worried about how their husbands were coping with the loss of their breasts, especially when parents had also shown concern that the relationship may not survive. One proband explains:

*'with him, I do worry about him because he does keep himself to himself and I do worry he's not telling me everything he's feeling, cause my family were worried that he may not feel the same cause my mother has seen herself being down in that in that, men don't stay with their wives after stuff like that. But I worry that he hasn't had enough people to talk to or get support from throughout the journey of it all' (P1, twelve months).*

Furthermore, because many probands had experienced a separation from self thus feeling less feminine and disembodied, the need for counselling and support for themselves was also highlighted. Why did they feel counselling was necessary? All probands needed to be prepared by professionals for the effects of RRM on their sexual relationship in particular and be able to discuss their concerns pre-surgery. The sexual aspect of their lives pre and post-surgery was not discussed and more preparation about the potential sexual difficulties with the ongoing support post-surgery to deal with the

relationship issues was necessary. Proband six who needed that support particularly made the point:

*'Nobody tells you that bit, nobody tells you what's going to happen after. there's fantastic support all the way up, yeah, and you get told what's going to happen, when you're going to do it but as soon as you have the operation in terms of how you're going to feel sexually or physically, who tells you that bit' (P6, twelve months).*

As many of the probands were shocked and devastated with the cosmetic results, on reflection, they believed that an opportunity to see a patient volunteer who had undergone the surgery would have helped them psychologically by setting realistic expectations and easing the blow. They particularly wanted to see the final cosmetic result of a range of real people who had been through the same experience, but who had different surgical outcomes, both good and bad. Others also believed that having this opportunity may even challenge decisions, questioning whether the technique is the right one, or indeed whether surgery is the right decision. Some probands, for example, may not have gone ahead with surgery and may have taken a different path had they seen the results on a real person. Two probands explain:

*'I think having more access to people who've had it done and seeing a range of what it looks like would help because it helps your understandings and shapes your expectations for the op' (P7, six months).*



*'You know you should ask people like me and x to show the results, instead of photos, ask us to show the results, real people. Because I don't know, perhaps like me, they might think do I really want to put myself through that. You don't know, do you? I think perhaps if I'd seen the real thing I would not have gone through with it' (P5, twelve months).*

Probands again reflected and acknowledged the importance of having the opportunity to see the photographs pre-surgery and the chance to discuss their options. For all probands, they had benefitted because this had given them some idea what would be happening to their body and gave them an opportunity to change their minds. Proband three, for example, after seeing the photographs and having the opportunity to go over the implications of reconstruction, soon realised that she was not ready for the surgery:

*'I think us talking it through, has made my mind set slightly different as well and just sort of thinking you know because obviously when I last saw you I was thinking yes lets go for the summer and then after seeing the photos and talking to my cousin, and thinking, and talking to my husband obviously, I just thought right actually you need more time, if time is on our side, but just when, so that our minds are ready for it and also what type of operation' (P3).*

In contrast, the disadvantages of not seeing any photographs were also highlighted. One proband for example, who was fortunate to elect to have a new procedure, with an implant based reconstruction and pigs skin (strattice), did not see any photographs because of its novel technique. Although she

had a plastic surgeon involved in her surgery, she had little understanding of what to expect and little idea of how she was likely to look and feel:

*'I don't think I really saw any photographs in x, he drew pictures so I could understand it but I didn't see any photographs other than I was being reassured I would have that natural droop, that was kind of all the reassurance that I had, that was it. What I'm still, what I struggle with sometimes is if I'm getting a pain or a feeling or a sensation and I think oh gosh what's going on now, here we go, I think for me I would have liked an even more detailed understanding of what they did, to understand, for quite a while it was pulling in some places and I was feeling that really quite, actually physically unbearable, I didn't like the sensation it was really horrible' (P7, six months).*

However, seeing the photographs was only part preparation for the extreme changes to their bodies. All probands, therefore, talked about the need for ongoing support, more consultations especially if there were complications with the surgery or a need for more surgery and counselling to adjust to their bodies. Ongoing support from women who had personal experience of such surgery or from support groups was also identified as being beneficial to restoring self. Two probands suggest what could help them and others:

*'Yes, but you need more consultations and not be too far apart because what I'm finding is the consultations are so far apart you forget what you've asked, you forget the information, you kind of want them a lot closer. So, you want a couple of weeks to mull something over and then go back, not go back six to nine months after, there's too long in between' (P7, twelve months).*

*'I don't know, perhaps a little bit of counselling after maybe, you know. Sort of like, like some ladies who've had it done meet up and then discuss. Yeah, like a support group or*

*something like that. It would be nice for people to know what they've been through' (P5, twelve months).*

#### **4.6 Conclusion of Probands Findings**

The probands' stories were powerful accounts of the impact of the inherited BRCA mutation and RRM. Although their fear of breast cancer had resolved, the fear was now for their children, who they saw as having an altered child trajectory. As a result of RRM, despite reconstruction of the breast and nipple, all probands experienced an alteration in themselves as a person and a drastic change in their bodies as women with one proband experiencing deep regret in having elected to have RRM. Thus, by electing to have RRM and losing their breasts, probands experienced a 'separation from self', their whole self, as a result of 'disembodiment' which did not improve significantly over time and for which they required support and counselling long term. This new fused horizon of understanding, that of 'disembodiment' offers a new contribution to existing knowledge by offering an explanation as to why women experience such negative effects on identity, femininity and sexuality and feel separated from their whole selves. These findings have implications for practice and the wider breast care community and thus, identifying the symbolic significance and functionality of the breasts for women with a BRCA mutation who elect to have RRM, is essential. The next chapter presents the experiences of the husbands.

## **CHAPTER FIVE – The Husbands’ Experiences**

### **5.1 Introduction**

This chapter explores and interprets the experiences of the five husbands in this study. The men were aged between twenty-four and forty-six years and they were all in full time employment except for one who had left the Army. Two were professionals, two were manual workers and one was ex-military. All but one of the men had children. The men were interviewed either in their own homes or in the breast centre and they were interviewed just once before and once post-surgery due to time constraints. It was felt that at twelve months, they would have had enough exposure to the lived experience that would enable them to tell their stories and increase knowledge.

The probands who took part in this study had asked their husbands to consider the study and contact the research nurse if they wanted to participate. Five husbands came forward. This chapter interprets the experience of living with a BRCA mutation and surgery from the husbands’ perspectives. Three horizons of understanding found in the experiences of the probands unite the experiences of the husbands and the family members, the price of survival, the altered child's trajectory and a separation from themselves. Each horizon will be presented alongside the interpretation of their stories which led to that horizon of understanding. The price of survival will be presented first, followed by a separation from themselves and the altered child's trajectory. The pre-surgical experience will be followed by the post-experience.

## **5.2 The Price of Survival - Pre-Surgical Experiences**

The Survival of their wives and their children was the main focus of the experiences of the husbands in this study. However, there was a price to this survival which included the long journey of discovering and making sense of a mutation in the family, the difficult decision of testing and the decision to elect to have risk-reducing mastectomy, concern about the effects of RRM on the relationship and living with ongoing concerns about their children. The next sections provide the reader with that journey.

### **5.2.1 The Price - Finding Out About a Gene in the Family**

The husbands in this study only became aware of a BRCA gene mutation once the probands had attended the clinic to discuss their risk, this was the result of other family members initiating a test. Although family history was a term they were familiar with, the link with an actual gene that caused breast and ovarian cancer was all new information that shocked them and took some time to comprehend. They were aware of young family members dying of breast or ovarian disease and that cancer was in the family but many husbands had not attended the genetic clinic appointments with their wives thus the full implications of a BRCA gene, for example, the cancer risk and the surgical options, was difficult to understand even after time had elapsed. As husband five explained:

*'It is in the family; a lot of her family have passed away very young through cancer you know'. Well I heard of breast cancer but I never heard of like the BRCA gene sort of thing*

*before, I couldn't understand a lot of that; I still don't understand a lot of it now' (H5).*

Why was this so difficult to understand? Although some of the family had been seen by a genetic counsellor and verbal information had been given, the majority of husbands were not aware of the availability of a particular test for a BRCA gene and it had all happened very quickly for some of them. Many were gathering information from their wives or were directed by family events, for example, it was only after the death of his wife's young cousin that a test for a BRCA mutation became apparent to husband four:

*'I've known about, my nan died of breast cancer, I know, I've heard of, I didn't know about the BRCA gene or what it was called but I've heard of families where the risk of breast cancer is really high, in media and stuff but I didn't know that there was, you know the gene testing going on and things like that until her cousin' (H4).*

For others, if a close family relative had died young of breast or ovarian cancer, there was some thought given to the possibility of a link to this and the next generation. Husband three had some suspicion of a family link because his mother in law had ovarian cancer and died:

*'Unfortunately, her mum died of ovarian cancer when we'd been together for three or four years and you know without knowing the science behind it that did; it did seem quite obvious to us that there would seem to be some kind of link even though we couldn't really prove it' (H3).*

As the husbands gathered and received more information from their wives, they reflected on the family history and became much more aware of the increased chances of getting breast and ovarian cancer with a positive BRCA test result. They then became extremely worried about their wives' survival because of this high risk and the link with cancer. There was a price to pay however for acting upon the genetic information and ensuring survival. By embarking on the journey to find out if a mutation existed in the family, this led to many anxieties with difficult decisions to make. These anxieties included the BRCA gene itself, a decision to be tested, a potential positive result, the possibility of surgery with loss of the breasts and the risks for their children. For all husbands, this information was overwhelming and the majority needed more than a verbal explanation. One husband was so shocked, he did not understand the full implications, *'what it all means'* and could not take everything in when he first learnt of a mutation in the family:

*'So, I suppose you know going back to the day when we first got the news, umm, there was that initial ok I understand what you have just told me but don't understand what it means so in a way, where's the leaflet' (H2).*

Husbands needed more information and time to assimilate the information because the majority wanted to be able to support their wives. What did they need to know? They needed to understand for themselves the implications of the test and the result. They needed to discuss and understand the actual risks, they needed to discuss and assess the urgency of the test or indeed, decide if their wives should have the test. However, this was difficult for the

husbands if their wives did not want to disclose their risk information more widely or if they did not have the necessary information. This secrecy caused many problems because there was nobody for them to talk to or find answers to their questions. One husband was really struggling to help his wife:

*'It has been hard like but you have nobody to talk to them and explain/ask questions and different things so it just been kept close between us like so it been a bit hard at times and obviously she been stressed about it and what do I do and all that, but I can only give so many answers like and that's, it been awkward at times like, and that' (H1).*

### **5.2.2 The Price – Having the Test and Understanding the Implications**

Once the husbands gained more information, with time, the risk of cancer and some of the implications were beginning to be realised but the test became a real concern because they feared it may be positive. However, all of the husbands concluded that their wives should have the test. They gave various reasons for their decision, for example, if a young family member had died of cancer or if the probands themselves were pushing to have the test, the husbands were strongly influenced. For one husband whose wife's cousin had recently elected to have RRM and was found to have the early stages of breast cancer, this also reinforced his decision making him more anxious that his wife would get cancer. He wanted the test to be done immediately:

*'But I think what really kind of brought it into really sharp contrast was her cousin when they did the double mastectomy they found pre-cancerous cells in her milk ducts and her cousin is quite a bit younger than her as well, she's only twenty-nine so I think that really, so I think that was really the thing that completely swung it you know, I think we went from*



*an initial reaction of you know, breast cancer is quite treatable, it's quite curable, it's not guaranteed that you'd get it, if you do you can deal with it, to lets have the test now' (H3).*

Although there was a desire for their wives to have the test and know the results, some acknowledged that the implications of having the test had still not been fully considered or understood, for various reasons. One husband, for example, needed to know the test result before he could think about the next step. He encouraged his wife to have the test despite his wife not being sure about it. In hindsight, he reflected that this may not have been the best approach because they now had to consider the bigger picture of the surgery.

Husband two explained:

*'Umm, and she wasn't sure whether she should have the test because she wasn't sure what she would do with the result if she got it. Yeah I'm a, I'm quite a practical person by nature, so my stance on that sort of thing and I think I said this to her at the time, why don't you just have the tests and then figure out what you do with the result once you got it, there is no point trying to figure out what you would do with the result if you don't know what the result, In hindsight maybe that wasn't helpful, I don't know, umm but from a practical sense, that to me made sense' (H2).*

Other husbands did not have time to think about the implications because the decision to be tested, the timing of the test and the urgency of having it done depended on how anxious their wives were and what was happening in the family. For some husbands, therefore, the gene test came about too quickly because their wives had initiated it and there had been little time to think about

it. Husband four who was distracted with his own health problems, was suddenly faced with the whole process of testing before he had time to think:

*'I suppose it all came about a bit quick 'I wasn't really thinking of the gene testing and family history and it seemed to, because there was so many things going on with my own health at the time I wasn't paying so much attention to it. X got to be tested first because that's where it's coming from sort of thing, so it was put off for a couple of months but still on a back burner so to speak and then it came.' But yes, with the age of X getting it so young and then the gene testing, that came about pretty quick from when she was diagnosed and then getting tested and then her sister getting tested, then her dad getting tested and then you know all the way down to X' (H4).*

One way of helping the husbands understand some of the implications of the BRCA gene was Angelina Jolie's disclosure that she was BRCA positive and that she had elected to have RRM. Her disclosure had helped the majority of husbands because it had raised awareness, they could now relate to it and they could talk about it. Husband five who had never witnessed cancer in his own family was comforted by the Angelina Jolie story. Her story had raised his awareness:

*'Well it's definitely opened my eyes up to it because like I'd never really known anyone who's had breast cancer or anything you know, so I've never had to witness anything and then with Angelina Jolie in the news, it really helped...' (H5, six months).*

Why had this helped them? Most husbands had difficulty in telling people about the gene and the surgery, not only because it was a private matter and they had been sworn to secrecy but because they had to understand it first for

themselves. Having celebrities and public figures like Angelina Jolie and Sharon Osbourne as examples of people having the same gene and surgery made it easier to explain to people and normalise it, especially when they were nervous about telling people. These husbands also saw Angelina Jolie as a role model in having a BRCA mutation, thus it was more acceptable and reassuring for their wives to have the gene and the surgery knowing that celebrities also had it. Two husbands share their experiences of how the celebrities' disclosure had helped them:

*'I think in terms of being apprehensive about telling people, I think that crossed my mind, yes definitely but I think if you can explain it and having the Angelina Jolie story is something to help you explain, I think is great, to be honest' (H3).*

*'whereas Angelina Jolie is a global superstar and doesn't need the publicity so I think in that respect it's been very, very good and very useful and she's obviously such a glamorous sort of Yeah, yeah, married to one of the heart throbs of Hollywood so you know it's, almost I suppose to some extent a perfect role model in terms of other people sort of seeing what can be done and that somebody and that it can affect somebody like that and then I think there have been a couple of others, I think Sharon Osborne has had the surgery as well' (H4).*

### **5.2.3 The Price - The Meaning of Being BRCA Positive**

The majority of husbands had considered the strong probability that their wives would be carrying a BRCA mutation once tested. This was influenced by how much their wives had convinced them that they would carry it and by reflecting upon the number of female relatives already diagnosed with breast cancer.

For example, if there had been many female deaths within the family, there was an expectation that their wives would have the mutation. Two husbands explain how they came to that conclusion:

*'We, therefore, decided that we would find out whether she had the BRCA gene and again even though it seemed, you know, like a 50/50 chance, I think we both kind of thought that she would have it because it did seem to pass down the female side of her family and sure enough that was how it panned out' (H3).*

*'I think in the back of her mind she always knew, because her mother's mother I think it was, died of cancer, obviously the gran, the mother's sister's daughter died two or three years ago, from cancer, at a young age forty-two, forty-four. X father is a twin, his twin brother died of cancer only six, seven years ago, you know, so it is. It is in the family, it's really scary' (H5).*

In addition, following the test results, although the risk and the likelihood of getting breast cancer with a mutation was explained to the husbands in percentages and in terms of a lifetime risk, the majority believed that cancer was inevitable and again, this perceived risk was associated with the number of relatives who had already died. Husband three gave his reasoning:

*'65% to 85% but we felt it would be 85% and part of the reason I think we felt that was because x auntie had died of breast cancer as well, which is her mum's sister and at the same time as finding out that x had the BRCA gene, she was very close to her cousin, her auntie's daughter and they had both lost their mums, and were very close' (H3).*

All these husbands were not just fearful of the risk of getting breast cancer but were constantly anxious about their wives' dying, perpetuated by their wives'

anxiety and strong beliefs about getting cancer and dying. In fact, the fear of cancer, in general, was very vivid in all the husbands' experiences. Two share their fears of losing their wives:

*'Really high and I'm still worried that she still might get something, because, although you're saying it's like the breast cancer and the ovaries or whatever, is it if you remove them' things which will lower it, is there still a chance she's going to get it anywhere else, I don't even know them questions you know' (H5).*

*'It was more like, with her it was more of a worry of when am I going to have it like. With her and her side of her family, it was all coming down in generations and coz her sister obviously didn't want to be tested, it was like, is it going to miss her like and get me and I was like so you really think it's going to happen, like you know so I was like, to me at first, I thought she was thinking oh she's going to have it in the next couple of years like which was a bit like what am I going to do if I lose her?' (H1).*

Thus, for the majority of husbands, a mutation was more frightening than having a strong family history because it meant it was more serious, the risk of getting cancer was almost inevitable and importantly, they were likely to lose their wives. Husband five was very frightened:

*'Well, it was quite frightening to be honest with you because just my, lucky enough we've never had it in my family at all so I don't really understand much about cancer apart from what you hear on the news and the telly and everywhere else. But the minute you find that something like that (BRCA gene) is involved in your family, then it's quite frightening because the last thing you want to do is lose your wife' (H5).*

Indeed, the BRCA mutation result devastated some of the husbands despite being aware of the strong family history and the probabilities of testing positive. One husband for example, although he had encouraged his wife to have the test, like many of the husbands, he had not thought about the implications of a positive result. Thus, it was too painful for him to believe the result hoping that a mistake had been made when his wife was given the result. His disbelief was evident:

*'And sort of, you know, are you sure you know. Could this have been misdiagnosed you know, again with the greatest respect to your colleagues, you hear these odd stories about people who go in to have the right kidney taken out and the left comes out instead and you think well could there have been a mistake' (H2).*

Thus, although having the test made sense to the husbands, the result of a positive mutation had a significant impact on the husbands for a number of reasons. It meant having to make decisions about surgery, a need to consider future generations and a constant worry about their own children's risk. Their experiences involved a constant fear of the probands' not surviving. Therefore, even though many had expected the result of the test, they experienced considerable negative impact and devastation. For example, one experienced *'a feeling of isolation'* (H2), another was constantly feeling *'quite upset'*, whilst others were pre-occupied with thoughts about the test result *'five days a week'* (H3), *'it was on my mind, playing all the time'* (H1).

These negative experiences mainly related to the significant risk of both breast and ovarian cancer, which threatened their future and importantly their wives' survival. For example, the impact of knowing his wife had the BRCA mutation and was at risk of two potentially life-threatening cancers frightened husband three who shared what it meant to him to have a long life together:

*'In terms of how it impacts on me, obviously being supportive of her and wanting for us to be married for a very long time is the key driver behind everything really and you know, I think it's very difficult because part of you sort of thinks, I think particularly when you first hear about the BRCA1 gene and you hear about your kind of the percentage probabilities of different types of cancer, ovarian and breast, you realise that you know you're not being dealt a particularly great hand' (H3).*

The pre-occupation with the results and the negative effects on their day to day life was like having 'the sword of Damocles hanging over them' both in terms of the fear of getting breast cancer and the long wait for surgery. Most husbands were consumed by the result which affected everything they did. Husband two explains:

*'You know there is not a day that goes by and probably not an hour that goes by without this being there, you know this is the sword of Damocles that is there from the moment we awake to the moment we go to sleep' (H2).*

#### **5.2.4 The Price - Strategies for Getting Through the Day**

To deal with the gene result, all of the husbands were trying strategies to keep themselves healthy and well. These behaviours included blocking negative thoughts out by distracting themselves or some focussed on their own health and getting fit. The test result had made some much more aware of the need to stay fit and healthy for their wives' sake. For others, it was too painful to think about any of the implications and so they tried to ignore it. Two husbands adopt different strategies to deal with the situation:

*'Okay, so first it's been about 2 years since the diagnosis was first derived, it's been a strange experience. I suppose umm lots of conflicting emotions umm at one end of the scale, we effectively have been able to block it out, probably incorrectly but nevertheless, we are busy people day to day there are so many things to do, it's quite easy to just fill your day with stuff that blocks this problem out' (H2).*

*'Well I suppose it depends on the circumstance you know, I think what it has done is given me quite a clear insight that in my own personal sort of health is you know, really important and that there's no point her going through all of this if I go and keel over with a heart attack so I think you know, I've been really a bit more involved with the gym and football and stuff so I think that's a good release for me that I think' (H3).*

#### **5.3 The Price - Losing the Breasts**

In order for their wives to survive, the husbands believed that screening with mammography could detect cancer, but not prevent it or prevent their wives from dying. Thus, the majority believed that although the loss of the breasts



with all the implications, was a price to pay, RRM was the only option in maximising their survival. Primarily it was their wives who had made the decision about surgery, but some husbands had strongly influenced or encouraged their wives because they were so frightened of them dying and because they would not have forgiven themselves if cancer did occur later in life when they had this opportunity to prevent it. Two husbands explain why it had to be surgery:

*'That's exactly the right decision (surgery) and I think you know, I also think that if we were to not have the operation and to find ourselves five or ten years down the line and her getting it I think we'd never forgive ourselves because you know we need to be there for each other, for our children and you know it's, it could be that almost, potentially it could be almost the best case scenario that you'd end up having this operation reactively rather than proactively. Anyway, so that is, you know, when you kind of weigh it all up then it just, you know it didn't seem as though there was an alternative really to us' (H3).*

*'Oh yes, well to be honest I virtually pushed X all the way you know, I think I most probably made 90% of the decisions for her because you know I don't want to lose her, her mother decided that she was going to keep hers you know, so she said she's coming up to 70 years of age now so it didn't make a difference whether she passed away or not. It's quite a brave thing to say to be honest with you but I just want to hold on to my wife a bit longer' (H5).*

The majority of husbands, therefore, believed that surgery was a chance to be proactive rather than reactive. They believed that they had been given a better

chance of surviving than other family members and should take that opportunity. Husband three shared the long-term benefits of this decision:

*'I'm trying to very much see it is as a positive because it gives us an opportunity that perhaps people have not had in the past, you know, people have obviously developed cancer of different forms and not had the opportunity to have proactive surgery that you know, so, it doesn't always feel like it but I think we've tried to be really glass half full about it and so it's been something that we're really trying to embrace as being an opportunity and although it means obviously a difficult next few weeks, hopefully, it will make for a much more beneficial time in years to come' (H3).*

### **5.3 1 The Price - Physical and Emotional Impacts of the Decision**

Although surgery was the right decision, it was still a very difficult decision and their experiences leading up to the decision of electing surgery was very intense for all of the husbands. However, the majority were able to contribute to their wives' decision-making even if they questioned the timing of it. For example, some husbands wanted their wives to go ahead with surgery immediately but others wanted their wives to wait until they were older. Why did age matter to them? They were concerned that losing the breasts so young would have greater consequences on their sex life and their relationship. One husband explains:

*'She had to do something about it, and I was like give it a couple of years but she was adamant that sooner is better kind of like. At first, I was a bit like you know, do you think it's a bit soon and you're so young? Do we have them off and all that like, because she is so young and I am like. I was like what is it going to be like without them and all that. It was just, is it going to be the right option, is it going to be the sensible thing to do or is it going to push us apart' (H1).*

Indeed, for this reason, the decision about RRM was far more difficult than the decision to have the ovaries removed. For example, one husband who agreed to his wife's ovarian surgery, was not able to contribute at all to his wife's decision about RRM, why was this the case? He did not think it was the right decision for them and he explains why:

*'I said to her, I am not going to advise you because I can't, I am not the right person to, so you will at some point come to me for advice and I'm afraid I won't be able to give it. I will give support but not advice and I'm not sure we are ready for this' (H2).*

Why was the removal of the ovaries an easier decision and more acceptable? The breast surgery was far more difficult than the decision to elect risk-reducing ovarian surgery because of the visible effect of breast loss on appearance. In fact, all of the husbands were concerned about the effect that the breast loss would have on their wives' psychological state especially if their wives were very conscious of their body appearance. Two husbands explain their concerns:

*'obviously, we had a decision to make about whether we sort of followed that through with a mastectomy and that I think was a harder decision because the hysterectomy is kind of, whilst obviously it's a serious operation, there's no kind of outward appearance' (H3).*

*'I think the one with the breasts is more, not so much a worry for me, but for x, because women like their figures and she does cry a lot and then saying' what am I going to look like and this, that and the other' (H5).*

Thus, although RRM was believed to be maximising their wives' survival, some husbands felt that RRM was mutilating and were initially reluctant about it because of the drastic effects and deformity to the body. Why was the surgery viewed as producing a deformity? The husbands acknowledged the importance of the breasts for a woman's femininity, sexuality, and identity, acknowledging that the decision was difficult for them and their wives because their wives would look different to other women. Two husbands explain why they were concerned:

*'Why would you do that to yourself basically, why would you kind of I suppose, kind of deform your body that was the initial reaction' (H4).*

*'Because I think a woman's breasts are a very visible part of their persona, their femininity, I think you know they are just a very overt physical sign of somebody's I suppose their being, their sexuality, their femininity, everything you know and I think that you know, I daresay in a jumper or in a dress or something like that then you know, it may not be particularly evident to the untrained eye I'm sure' (H3, twelve months).*

### **5.3.2 The Price - Undergoing Breast Reconstruction**

The husbands understood the need to have a reconstruction to maintain their womanhood. They expected and relied upon reconstruction to maintain this femininity and restore who their wives were before the surgery:

*'I just feel desperately sorry for her. So, yes, it's a dreadful situation for her, for all of us, there is a route out of it which is*

*the, I think it's the reconstruction. It's difficult for a man but for a woman to have your breasts removed, I just can't imagine anything worse because it's kind of who you are' (H2).*

Why were the husbands so reliant on breast reconstruction to fix everything? The cosmetic result of the surgery was also very important to all the husbands and they were very honest about their concerns. They were expecting breasts that resembled their wives own breasts. However, the photographic images they had seen of the proposed surgery and reconstruction were very different to their expectations, especially seeing breasts without nipples. Although acknowledging that they needed to see photographs in order to prepare them for the devastating outcomes, the images were shocking (H2, H3, H4) and graphic (H4, H1). Husband four was overwhelmed and shocked when he saw the results:

*'The one thing that, I didn't know what they would like afterwards and so we saw photos beforehand, Yeah and it's a big operation and it's not what I thought, It was a bit like woah, but it didn't last long. Just taken aback, I can imagine some blokes being, oh fuck sort of thing, massively so I think when a male thinks yeah implants, he's not thinking what the actual thing is, I didn't realise the nipple is gone. Yeah, I suppose so, and you know the scarring is big and some of it looked quite graphic, I can see some, for me, it's not really a problem I honestly don't think but I can imagine some men might have a real sort of problem with it because it is fairly big. Yes, I think that's really important, to get the photos in your head straightway, really straightaway so that you can prep yourself' (H4).*

Why were the photographs so shocking and devastating? The majority of husbands felt that the images did not resemble normal breasts and they could

understand why women or their husbands would choose not to have the surgery. Husband four explains why the images would affect most men:

*'I just think, you know we saw some photos of what it looks like afterwards and it just, they're just lumps of flesh, and we were both of the same sort of attitude. I can understand women who don't want to have it done, I really can, especially if they're more concerned about their looks and I don't mean that in a vanity sort of way and especially if they're, I can imagine it would be difficult if they were single, I mean if you're not with a partner, your life partner so to speak, you'd be thinking that's going to put men off and all the rest of it and if you're strong enough to think well it'll be alright, I'll just see if I get cancer, I'll deal with it then, if you can live like that and just have the tests every six months to a year, then all power to you but I know I couldn't live like that and I don't think she could either, everyone's different' (H4).*

### **5.3.3 The Price - Uncertainties about Changes to the Body**

After seeing the photographs, therefore, husbands were very concerned about how their wives' bodies would change and how they would react, so much so that one husband had asked himself *'am I going to accept it?'* (H1). Others tried to imagine how the new breasts would actually look compared to the images in the photographs. They were not convinced that the photographs were a true representation of the final result, hoping that they would be better. Husband three tried to imagine how different his wife would look:

*'She will look different, it's hard to imagine, she said they are going to be firmer and higher but you just see pictures and see people and it's what will it really look like, I don't know' (H3).*

Thus pre-surgery, all the husbands were concerned about the effect that RRM would have on their relationship, especially their sex life. They were concerned that the changes to the body would change their wives and their normal life, making their wives very self-conscious and less likely to want any intimate contact:

*'With sex I, well its, is it going to be the same obviously, like before everything was fine. That it's going to be ok? is it going to be the same or is it going to be more, I want to keep my top on or I feel uncomfortable. Is that going to push me away or will things be fine, you know' (H1).*

All husbands therefore expressed concerns about the long-term impact of RRM on them and their wives. What long term effects were they worried about? They all recognised for example that breasts are an integral part of a woman's body and the photographs of reconstruction did not resemble 'normal breasts'. Husband three was concerned therefore that if his wife disliked her body, this would affect her mental state long term:

*'Well I know that a wife who is healthy in body you know and you know fundamentally it is something we'll deal with because in years to come we'll look back on this as just another thing that we've gone through I'm sure of that but I think my concern is obviously just, yeah, for the operation itself obviously for her physical wellbeing but then in the future for her mental wellbeing' (H3).*

Like many of the husbands, he wanted to maintain his wife's confidence because that was part of who she was and in many ways, he was envious of and applauded the gift she had. He was afraid of the surgery changing her as

a person. Other husbands were also concerned about how their wives would feel about and view their body, but the majority believed that if the cosmetic result was acceptable and their wives were pleased with their body, then everything else would take care of itself. Two husbands explain:

*'The cosmetic result, I think if she's happy with that it'll be all over, you know, I think it'll be all over. She won't think anything of it again you know. I think she'll be fine, to be honest with you like, as long as she's happy with the outcome after you know, I think she'll be over the moon to be honest with you because it's something that she's always wanted, you know. Because she's never liked her breasts anyway, you know, so she, I wouldn't care if she didn't have anything at all, to be honest with you, it would never bother me at all. Yes, as long as she's happy with her appearance that's all I'm bothered about, you know' (H5).*

*'I just think that you know, confidence is huge because if you don't have a lot of confidence then you sort of become withdrawn and potentially go into your shell, don't you? she's a very bubbly person, she's got lots of friends, she mixes well with people, you know you put her in a room with people that she doesn't know and she'll hit it off with people, it's a gift, to be honest that I don't have and I would hate to see her lose that because I think that's part of who she is' (H3).*

#### **5.3.4 The Price - Turning the Family Upside Down**

In addition to the concern about physical appearance and psychological well being, there were also practical considerations and uncertainty about the recovery and how much disruption would be caused to family life. For example, there were uncertainties about how much help would be required and how long recovery would take. Husband one was particularly anxious because of his small children:



*'We have got two kids as well? Obviously how long is she going to be off for and not be able to do things and stuff like that' (H1).*

However, once a decision had been made to elect for RRM, husbands wanted to get on with it and with their lives. The wait for surgery varied, but it was at least five to six months after their initial clinic consultation and some operation dates had been cancelled. The wait was extremely traumatic and disruptive to family life because they had planned everything around the date. Husbands were also scared that their wives would develop cancer during that time. For others who did not yet have a date, life was on hold. They found it difficult to see a future until the surgery had been done. Two husbands explain the effects of having to wait:

*'Awful, I think the waiting is the worse, The waiting is definitely stressful, I think it's a, I can't think of a better word than stressful but it's kind of, in some ways because it's always looming, it can almost become unbearable and you just something that's kind of been the hardest bit has been the waiting and that's what I think the tunnel vision approach has kind of been the right thing for us and therefore that we just can't wait to be out the other side and I think then you kind of cope with it in your own way from there. Whereas I think for me I need to see past the operation to the future' (H3).*

*'Because that's another thing, sort of date wise because I said the whole year is on hold at the moment because we like to go on holidays every year and at the moment we haven't booked anything at all because they're saying it could be middle to the end of summer, you know what I mean so' (H5).*

## **5.4 A Separation from Themselves - Post-Surgical Experience**

As with the probands experiences, the initial focus of the husbands' experiences post-surgery was the reduced fear of cancer, but their main focus was the effects of RRM on their wives' bodies and their relationships and the ongoing concern for their children, who they now believed had an altered child trajectory. The focus on their wives bodies included the effects of RRM on their sex life, on how their wives now perceived themselves and how they believed that their wives had changed. These experiences dominated the post-surgery journey. The following section takes the reader through that journey.

### **5.5.1 Separation - The Fear Has Gone**

Post-surgery, the majority of husbands (H1, H3, H4, H5) were very relieved at not having to worry about the fear of cancer. Indeed, the fear of their wives dying from cancer had resolved for all the husbands. Two husbands share their experiences of the relief:

*'Which as I say everyone is completely different and entitled to their own view, but I think that being out the other side now, it might have been a hard couple of years but it doesn't half feel good to think that's not a worry for us' (H3, twelve months).*

*'It's been something that we've put behind us and put some real fears to bed as well' (H4, twelve months).*

Despite some of the uncertainties before surgery and any reluctance about the decision, all the husbands believed that for the benefits of the resolution of fear

alone, it was the right decision to elect for RRM and reconstruction. Without hesitation, one husband explains:

*'It's definitely, 110%, it was the right decision to... Oh yeah, yeah, without a doubt, especially with regards to what's happened to her cousin at the moment, you know: So yeah, definitely it was the right decision, there's no hesitation about that' (H4, twelve months).*

All the husbands, however, recognised that it was still a difficult decision to make, having the utmost admiration for their wives. The majority, for example, commended their wives for being *'very brave'*. Why did they think it was brave? Most believed that their wives had sacrificed their femininity and identity for the sake of their family and it acknowledged that it was not an easy operation to undergo. One husband expresses his gratitude:

*'I think she's every bit as beautiful now as she was when she had the operation so, in fact in many ways more so because I actually, I think what she's done for our family is so brave really ...' (H3, twelve months).*

### **5.5.2 Separation - Not Who They Were?**

However, despite being prepared for the cosmetic appearance, the majority of husbands were still very shocked at first seeing the changes to their wives bodies and surprised about the effect it had on them. Why were they so shocked? It made them realise the extent of the surgery and that their wives did not look like themselves. Most of the husbands took time adjusting to the new look of the breasts and their wives bodies because of the severe bruising,

the stitches and they looked so different. Two husbands share why they were so shocked at what they saw:

*'Probably a bit of the shock, I didn't really realise how taken aback I'd be by it when you see them' (H4, twelve months).*

*One of the things that I think was difficult was seeing how she looked immediately after the operation, because that was you know, I guess like with any operation there were stitches and bruising and you know that looked really, really painful and really I suppose shocking in a way' (H3, twelve months).*

As time elapsed, the look of their wives' reconstructed breasts became more acceptable to them. If nipple reconstruction and nipple tattooing had been completed, for example, some husbands were even more pleased because the breasts started to resemble normal breasts. However, this depended on how the appearance was viewed by their wives and the stage of the surgery. At twelve months, for example, the scars had settled, some of their wives had nipple reconstruction and the cosmetic result was better than some were expecting. Two husbands were pleasantly surprised:

*'Didn't know what to expect, I thought it was going to be scars and looking a bit awful but when I looked at them I was, that's pretty good that is, quite impressed' (H1, twelve months).*

*'I think the result as it stands today is fantastic, you know I genuinely think that x could be in a changing room and somebody could catch sight of her and not realise that anything has happened operation wise. So that's an incredible testament to the process I suppose and the surgery and the aftercare, I think having the nipples tattooed in the end has actually made an incredible difference as well*

*because although previously you know, the shape was good but there were scars, actually the tattoos have covered up most of the scars as well so I think you know, the physical appearance is incredible' (H3, twelve months).*

However, some husbands were struggling to come to terms with the 'new breasts' at twelve months because they were still so shocking and looked so different to normal breasts. They were finding it difficult to accept the new breasts and they had to adjust slowly because it was such a drastic change to the body and to how their wives once looked. In trying to find strategies that enabled them to cope with the drastic changes, they had to keep reminding themselves of why the operation was being done, to prevent cancer and maximise survival. For those husbands who could think in this way, it was far easier to accept but those who could not rationalise it in this way found it more difficult to accept. Husband four explains why he was struggling to get used to the way his wife now looked:

*'I knew what they were going to look like from pictures so, you know wasn't expecting breasts again, you know, get that idea out of your head straightaway. It was quite, it's still, and you know it does knock you back a bit when you see them but it's just a case of getting used to seeing them. I don't know, they still, yeah, they still kind of surprise me a bit. I mean with time I'm going to get used to them and I'm still all for her having them done: It's not something that's that easy to get used to because it's something so physical, the only way that I could sort of compare it to is if you lost a limb and do you know if you look at someone with like a false limb and then all of a sudden you can find yourself sort of staring just because you're not used to it and you're taken aback by it. So there is that element of it still' (H4, twelve months).*

### 5.5.3 Separation - No Return to Normal

In addition to affecting the final appearance of the breasts, if the wives had complications post-surgery, this made the experience far more difficult for the husbands. Many of their wives, for example, were experiencing pain or infection with six of the eight experiencing complications. This meant that there was a long delay in recovery and they could not move on with their lives.

Husband four was just one of many who was frustrated and annoyed:

*'Shit happens, I mean it was really annoying and really hard to see her in quite a lot of pain and with my own experiences of pain on a day to day basis with breathing exercises, she couldn't do that because she couldn't breathe properly so it was quite hard to try and help her with it. But what can you, you know it's not really preventable, it's just something that happens with surgery. You know it's really delayed us with stuff and life but there's nothing that can be done' (H4, twelve months).*

Why were they so annoyed? They had to wait and be patient and it was out of their control. This delay also meant that life was on hold because there were more operations to follow and the journey was still not over. For all the husbands, the whole process had taken far longer than expected and had not gone to their plan. Many did not expect the number of complications and life had been turned upside down. Two husbands explain:

*'Well yeah, no not really, not until she's had her final surgery and then we can try and get into a groove of what our life is going to be like. It's been so up and down, what with myself and with her, yeah it's a bit annoying in a way, like I say it's annoying in a way that it hasn't gone, it's been so up and down with her and we can't move on and we've got operations hanging over our head and so I mean once this last one is out*

*of the way and hopefully she heals up fine and we can move on' (H4, twelve months).*

*'I really wasn't expecting the surgery to have the complications that we've had. I don't know why I think you just sort of think yeah everything will be fine then, it will be done and then bobs your uncle it's all sorted in a month or so, so I wasn't really expecting all the problems' (H5, twelve months).*

#### **5.5.4 Separated - Not in Unity**

The surgery, complications and delayed recovery had taken its toll on many of the husbands but the majority were in unity and able to work together. This long process, however, was more difficult if there were problems in the marriage before the surgery, or as a result of the surgery. One husband, for example, who was not ready for his wife to have the surgery and who had problems in his marriage before the surgery, explained how the experience of surgery was the last straw, causing more divisions in their relationship:

*'So, I think, we were close to the edge anyway, the surgery has taken us over the edge so you can't just blame the surgery, it's everything that takes you up to that cliff edge rather than the thing that takes you over it. So it's just the combination of all of that' (H2, twelve months).*

What had taken them over the edge and why were they so close to the edge?

The journey of discovering a BRCA mutation and surgery for husband two had been extremely difficult, exacerbated by insensitivities of other family members who were invading their lives by expecting support for their own ongoing problems. He felt this intrusion on top of the surgery, had put him

and his wife under extra pressure and had put a strain on their marriage. He shared his frustration:

*'There is too much invasion, that perhaps too strong a word, intrusion, there's too much intrusion from other people needing stuff. People come and take whatever they need and we get what's left and it's not enough' (H2, twelve months).*

He also expressed the personal price of the experience, the experience of much sadness and his struggle to live a normal life with his wife. Even after twelve months, he was just existing, rather than living a happy life:

*'For me it's, I suppose it's, I suppose I'm just trying to find a way of maintaining sanity amongst the status quo, so it's not a great existence but you just deal with it and you carry on. It's a struggle I think is the short answer, it hasn't got easier over the last twelve months, there are still the same basket of challenges that there was twelve months ago and none of them have really moved forward I don't think so, we spend most of our time doing what we need to do on a day to day basis without really resolving any of the more challenging issues. They just tick along in the background' (H2, twelve months).*

Furthermore, he felt strongly that input from the professionals was essential in order to prepare and guide families in dealing with such a difficult journey with potential family conflict. Whereas the surgery should have been the end of the long difficult journey for husbands and wives and an opportunity to move on, husband two believed it was the beginning of the process:

*'It's, you know it's not an easy journey to go through, I think if there's a learning from all of this it's preparing the family as well as the patient so more guidance from professional people to the parents to say you've got to give her space here,*



*whatever other issues you might have take them somewhere else because your daughter is no longer the person who can do this for you. She now needs to focus on her. If she'd had something more sinister, so if she'd had a heart attack or something or if she'd actually had cancer then maybe it would have been different. But there is this impression that oh well she's had the surgery therefore it's done and it's not done, the surgery is the start not the end of this process But if there's something that the medical profession could do at the outset which helps prepare the wider family for all of this' (H2, twelve months).*

The wider family conflicts on top of what they were trying to deal with had not been helpful for them as a couple or his wife personally. He was concerned about everything she had to cope with and how devastating it had been, she had drastically changed:

*'you know she was not in a great place before all this happened and you know, her going through what she's gone through would be challenging on its own, when you overlay all the other stuff it's, I don't want to sound like I'm over exaggerating here but I think it's basically destroyed her as a person' (H2, twelve months).*

Husband two, therefore, felt strongly that continued support after the surgery was necessary because of the trauma of the whole journey. One suggestion was that the professionals need to put a care pathway in place for men and families in order to help the marriage survive:

*'Yes, it's been hell, there's no doubt about that, I think one of the points that you made right at the start is there is no, there is no patient pathway or patient journey for the man in this, other than the steps that you personally have taken because you've seen a gap in the NHS provision. So that doesn't exist and I think there are knock on effects because that doesn't*

*exist, there will be marriages that fail because of this' (H2, twelve months).*

### **5.5.5 Separation from her Self**

For the majority of husbands, 'getting back to normal' was judged in terms of the intimate side of their relationship. Despite the wives concerns pre-surgery that their husbands would not find them attractive or desirable, husbands still felt attracted to their wives sexually and there was no change in the way they felt about their wives. Husband three explained:

*'No, no, it's changed nothing, if anything I suppose it's made me feel more protective of her I suppose which, yeah it certainly hasn't had any negative ramifications in terms of how I feel about her sexually' (H3, twelve months).*

One husband even acknowledged that his wife had made efforts to be intimate because she understood that he had sexual needs. Despite it being difficult the first time they had sex, he explained how his sexual life was getting back to normal:

*'I don't think it's changed it to be honest with you, at first just obviously adapting to it but then everything is so back to normal it's like, you wouldn't think she'd had anything done like. Everything has just carried on as normal. It was a little difficult at first but was easily overcome. No, at first it was a big thing like, you know when it came to sex and things but then once you get over, then everything was fine. But she was kind of accepting too, understanding then that I've got kind of needs' (H1)*

For the majority of husbands, however, sexual intercourse occurred far less often and for some, was non-existent, especially if their wives were unhappy

with their bodies. The husbands believed the reason for this lack of intimacy was because their wives were 'self-conscious' about the way they looked, still covering themselves up and hiding away with embarrassment, even after a year had passed:

*'I don't see them that often, she keeps them covered up, although she is getting better, she does it on purpose I know, they're like rocks, I mean, but I know with time she'll get more comfortable' (H4, twelve months).*

Others believed that the problems with intimacy and having sex was a result of their wives feeling different and not themselves, emotionally and physically disconnected from their body. Husband two explained:

*'Yeah, you know we've had sex a couple of times since the operation but there isn't that emotional connection. I think it's her body, you know she doesn't like the way she looks, I think the operation has killed off any sort of sexual appetite perhaps, she has become more withdrawn in herself, so she is less, if willing is the right word, she seems to not want an intimate relationship any more' (H2, twelve months).*

### **5.5.6 Sex Talk - A Need to Reunite**

Having support before and after surgery was welcomed by most of the husbands and seen as extremely important. They wanted the opportunity to talk about their feelings, especially the sexual issues, with people who have been in similar situations. Various other suggestions for the support they required were also given for example, in addition to access to the specialists within the team who could understand what they were going through, many of

the husbands wanted access to a support network. Two husbands give their views:

*'You know, someone to talk to about it, someone who gives you a bit of understanding and someone else, you know, to talk to mainly about it, like I have really felt like talking to people like, and your feelings of like you know ,the sexual issues and that and different stuff like that and obviously you have times when you are down and think about it and you have nobody to talk to like and it's like we talk to each other but you can only talk so much about it like to each other. A male preferably because it is easier to talk male to male' (H1, six, twelve months).*

*'But umm, if there was some kind of support network, it wouldn't appeal to everyone but for me, it would have helped' (H2, twelve months).*

## **5.6 The Altered Child's Trajectory**

### **5.6.1 Maintaining A Normal Child Trajectory**

Concern about children and future generations was a focus for the husbands as well as their wives. Although the fear had resolved regarding their wives, it was now for their children. The heritability of the gene was seen as an ongoing problem for many of the husbands because of the risk for their children and a there was a desire to maintain a normal childhood. All the husbands for example now feared for their children, in particular, they had anxiety about their daughters' risk because of the need to have the breasts removed. Many were totally pre-occupied with and extremely concerned about their daughter's risk and believed they now had a different life trajectory, a life of uncertainty,

worry and difficult decisions. For example, despite the fact that she was only eight years old, one husband believed the risk and potential surgery would ruin his daughter's life. This constant worry invaded his whole existence:

*'It even invades the dreams, it's there all the time, and it's not just with x..., it's obviously with my daughter now and her risk'. 'Umm, you know my daughter is eight soon, umm, at what age do I destroy her life by telling her that she might have this problem. I have no idea how to answer that question' (H2).*

### **5.6.2 Introducing Fear into the Children's Lives?**

Many of the husbands were constantly pre-occupied and afraid of telling their children. They did not want to alarm them, yet at the same time, they believed they had a responsibility. They felt responsibility as a parent to tell their children about the BRCA mutation, especially their daughters, but they felt it would change their life trajectory, their care free attitude and their potential relationships. It would also change the way people viewed them. Not only did they struggle with identifying the best time in the life trajectory to tell their children, but how to tell them. They were also fearful that they would get cancer. One husband was so afraid of his daughter finding a breast lump before they had actually told her. He shared all his fears:

*'and we still have my daughter and we need to find a way of telling her about this when she's old enough to understand, in a way which is not going to terrify her and I don't have the answer to that at the moment but that's at the back of my mind all day every day' (H2).*

*'Do we wait until she is married then her husband might say well you knew about this before we got married, why didn't*

*you tell me. You know, are we going to wake up one day and she finds a lump at the age of 10?' (H2).*

This fear caused many problems in the marriage because he felt so upset and responsible about the risks to his children and the issues that the mutation now brought. He started to question their decision to have had children and he explained why:

*'I think if we had known about this gene before we had children then we might not have had children, simply because you don't want to pass on the gene' (H2).*

Major concerns were echoed by many other husbands, the thought of their children taking a test, having a mutation, their daughters electing to have surgery or importantly their children dying as a result of getting cancer, filled them with fear. Husband five explained:

*'My daughter now, well she will have to be tested and my son as well so, it's quite scary, to be honest. That's my biggest concern, if anything god forbid ever happens to her you know, that I lose my daughter through breast cancer Yeah because we've seen it in the family on x side now and x cousin, years ago, she died of it, forty-two, you know' (H5).*

However, the fear for their sons did not appear to be as strong. Although all their children had an equal chance of inheriting a BRCA mutation, husbands focused on their daughters because they believed it was less likely that their sons would have it. Why was this the case? They were less fearful about the consequences for their sons because they did not have the double risk of

ovarian and breast cancer and they would not need such radical surgery as breast removal. Husband three explained why he had far less concern about his boys:

*'also, although I know that the BRCA gene can be passed down to the boys I suppose to some extent, firstly there's no guarantee that it will have been and secondly, as boys, they're prone to obviously different things – they don't have ovaries. I suppose that is something that maybe at some stage we'll need to consider whether we go down similar routes with them in terms of genetic testing and seeing whether we need to kind of have them screened for maybe, I'm not sure' (H3).*

Some of the older children had been given limited information about the mutation and the surgery but other children had not been told. Despite his wife not agreeing with him, one husband did not want to tell his children, causing extra strains in his relationship. He rationalised his decision because he believed this could mean they would get bullied. However, his main reason was that it would ruin their lives by changing the way they would see themselves and the way other people would see them. He had personal experience to base his decision upon. He shared his own story:

*'We have made a decision that we are not going to tell the children what surgery she is having. She will obviously tell them she is going into hospital umm I don't think the kids need to know so we have decided we are not going to tell them necessarily. Umm, I think back to when I was about thirteen or fourteen, umm children could be very cruel' (H2).*

Indeed, for some of the husbands, the concerns and fears for their children were still evident at twelve months and revolved around their risk of getting cancer and not surviving. Husband five feared losing his whole family:

*'That's my biggest worry out of the whole lot since she has gone through it, I know obviously she's a lot older than my daughter obviously and I already understand a lot more and sort of she's done it, by the time well, we haven't done our time exactly but you know it's not nice sort of getting prepared for my daughter to see what the test will be. Well that's was my biggest fear before I pass away, do I end up losing my wife, my daughter, my son before me, and you know' (H5, twelve months).*

Thus for all the husbands, their initial fear of losing their wives had resolved, but for many, the fear had now transferred to their children, making life uncertain. Husband two summed it up:

*'Yeah, has life got better no, has the fear gone, it's moved, so the fear is now for the children rather than for her' (H2, twelve months).*

## **5.7 Summary**

Finding a BRCA mutation in the family with all its implications had been a major life event for all of the husbands. Following news of their wives' positive result, there was a preoccupation and a negative impact for the majority of husbands even if they anticipated the result, especially in families where there were complex relationships. There was an overriding fear of losing their wives and their children to cancer. Although it was a very difficult decision, the majority of husbands believed that RRM was the only option to maximise survival but



they all acknowledged concerns about the effects of surgery on their wives and potentially, their children's psychological health including their own relationships and their future children's relationships.

The majority of husbands took time to adjust to their wives new reconstructed breasts because their wives looked so different and felt so different. Even with time, the appearance of the breasts for many of the wives did not improve and thus husbands were also living with the consequences of their wives 'separation from themselves' and feeling 'disembodied'. As a result, some husbands reported negative changes to their relationship and intimacy long-term for which they required help and support.

Post-surgery the fear of cancer they experienced for their wives had resolved but had transferred to their children. This fear, primarily for their daughters concerned the implication of breast loss but also included the fear of losing their children with cancer and the fear of how their life trajectory had changed. These findings have important implications for clinical practice and will be discussed in the last chapter.

## **CHAPTER SIX – The Family Members**

### **6.1 Introduction**

This chapter provides the analysis and interpretation of the five family members who took part in the study. Due to time constraints and the overall aim of the study to understand the probands' experience in the context of their family, they were interviewed just once. The interviews took place in their own homes either before or following the probands surgery and after their own genetic test. Of the family members interviewed, two were fathers of the probands who also had a BRCA mutation; one brother carried the mutation and one sister did not. One mother had breast cancer at the age of thirty-one a mutation was later identified.

The family members had agreed to be interviewed once they had been approached by the probands and this particular sample came forward offering a wide range of experiences. The age of the relatives ranged from thirty-six to sixty-five years. The two fathers had retired; the mother was in full time employment as was the brother and the sister. One father of the probands and her brother were part of this study. All the family members were therefore in some way, living with a BRCA mutation. In considering the 'parts in terms of the whole', three horizons of understanding united their experiences and those of the probands and the husbands. These are the price of survival, a separation of themselves, and the altered child's trajectory. Each of the horizons will be presented alongside the themes that led to those horizons.

The price of survival will be followed by a separation from themselves and finally, the altered child trajectory.

## 6.2 The Price of Survival

### 6.2.1 The Price - Being on The Alert

Preventing or surviving cancer was the main concern for the relatives as it was for the probands and the husbands in this study. All the relatives had been aware of living with a family history of breast cancer, indeed one relative had been treated for it, but none had ever heard of a specific gene. For the female members, there had been a constant message to the girls in the family *'to be careful'* and *'examine themselves regularly'* (R4). Relative four, for example, had always been alerted of the family history by her mother who believed she and her sisters had to be on *'their guard'*:

*'She would say to us (mum) you have to watch yourself, girls, you have to be careful there's something in our family. You know mum never knew anything about genetics or whatever she just yes, she just said there's something in the family because there'd been a history so I guess that's where it started' (R4).*

On their guard for what? There was a general feeling that a link existed between the number of cases of cancer in the family and the number who had died, but nobody knew what the link was. This coupled with a few cancer scares themselves or a personal diagnosis of cancer made the female

relatives much more aware of their risk. One relative who had a cancer scare remembers it vividly:

*'And I mean I've always been very aware, when I was thirty, thirty I think maybe thirty-one I had a lump in the breast so they checked it out and they said it doesn't look malignant but I had a, I had my second baby and I had to stop breast feeding her for them to remove the lump because they weren't sure. Again I don't know if that was something to do with the history that they were extra careful, but it was nothing, but I mean the awareness has always been there' (R4).*

Although relatives could see a pattern emerging within the family by the number of cases of breast cancer amongst the females, the implications of this with an additional BRCA mutation had only become apparent when other members of the family had been tested or had breast cancer. Male relatives like all of the participants initially had never heard of a BRCA mutation. There was much secrecy about cancer which was only ever 'whispered' about in the family. Two relatives share how it all unfolded and they came to know:

*'Nothing, nothing before, I think x and the other cousin, her other cousin x, were both very interested in this following x experience of breast cancer and it was through them that we came to hear about it. Probably my father's sister, his oldest sister x, born 1920, she had breast cancer in her early thirties which was 1950 something when I was born. So this was something that was always whispered about in the family, I wasn't party to it but it came to the front then when my sister had breast cancer, the first time, she was probably in her early forties. It was only then when the younger of my niece's, x, found a lump a few years ago that the genetics people took an interest in the case and it's only since then that we've come to know about the BRCA gene' (R3).*

*'I thought oh gosh this is a big thing, you know and out of the blue then they rang me and said oh you know can we have a meeting she said, we've got some news for you. So right okay then, so she came along to the house, she done another blood test and she said look she said, we've had this new testing, it's literally just in from America and she said we do this gene testing now and it's more or less instant she said, we'll have a result for you now within a few weeks. So we done that, a couple of weeks later and this, BRCA2 gene was mentioned' (R2).*

Thus, the full details of inheritance and the implications of a specific mutation were not fully understood by any of the relatives, for example, the knowledge that each person had a one in two chances of inheriting it. One relative had her own understanding of the inheritance chances:

*'Well I wasn't aware of the 50/50 so it was kind of x was positive, x was positive and I think in the back of my mind it was somebody's got to be negative, you know and I wasn't quite aware that it was absolutely straight 50/50 you know, everybody had exactly the same chance of having the gene or not' (R4).*

### **6.2.2 The Price - It's A Female Thing**

All relatives had limited understanding of the implications of being BRCA positive. The male relatives, in particular, were generally less well informed for example, they thought it was only females that were at risk or should be concerned about inheriting a BRCA mutation because of the risk to the female organs. As a consequence, they paid little attention to their own risk or potential consequences of having inherited it. Relative five, like others, believed that being healthy put him at less risk:

*'I just thought well it's a female thing, you know touch wood I've always been healthy, I'll be fine, you know, I thought I'd be negative.*

*I haven't really looked, I think when I went for the test I had a quick google, that was it so I didn't look at anything about it and' (R5).*

Initially, therefore the men in this study did not think that they would need to be tested and just ignored what was going on in the wider family because it had no personal consequences for them and they had no idea of the full implications. Two relatives explain:

*'No, if I was honest I probably just put it to the back of my mind and thought well personally I'm not going to get tested' (R5).*

*'I didn't realise what it was, the implications of it so I didn't really know much about it at all' (R1).*

The secrecy within the family also made it more difficult for them to gather information for example, for one relative neither he nor his wife were aware that one of his daughters had already been for the gene test and tested negative and another daughter was waiting on results. It was only when one of his daughters came to tell him her result, did he realise what was happening. He explained how he came to know:

*'I obviously know my mother's family history and she had cancer and her sisters and her mother but we didn't, we weren't even aware that there was a BRCA Gene. So, x came in and told us she had news, umm and then consequently we found out that our eldest daughter went for tests' (R1).*

In terms of their own mutation test and result, some relatives were shocked and yet numb when they were first told because they had expected to test

negative. They did not feel sadness or fear about their own positive results, the men rationalised that nothing had changed for them because nothing could be done. They took the view that there was no screening, no surgery and cancer was common. Relative threes' concern was always for his daughter:

*'So the actual day, going in for the result I was surprised at my reaction, I thought that I was a little bit stronger but no, I felt myself quite wobbly going in, certainly a bit shocked coming out. A few days of feeling uncomfortable but then the realisation well nothing has changed, this doesn't make any difference to me but it's X now she's got that 50/50 chance' (R3).*

Why were the men so concerned about their daughters rather than for themselves? The men took the view that their odds of surviving had not changed by having a mutation. All the men had, therefore, accepted their mutation result, taking a fatalistic view and rationalised that they had no control over it or its implications. In addition, they believed that a mutation did not have the same consequences or gender implications, whereas it did for the females, breast loss with all its implications. This was the main concern for all of the relatives. Two relatives were very blasé about their own risk:

*'There's more, you know we're all going to die of something, like 2.7 people are affected by cancer, we look at the rates because we do it for illness and income tax and stuff so we look and we have a lot of the stuff coming out from all sorts of different people so I thought well you know, my odds haven't got any worse but all I thought is mainly my daughter because I know it's all to do with your breasts and whatever' (R5).*

*'I have sort of, x (wife) doesn't like hearing it, but I have come to the conclusion that if it's going to happen it's going to happen and there is not a lot I can do about it'. You know in*

*20 years' time there might be something around or they might be able to eradicate it so there is a bit of a positive there' (R1).*

Unlike the probands, they were in no hurry to be proactive about a positive mutation, regardless of age. They took the view that there was treatment available if cancer did ever occur. Their focus being on prostate cancer, but not cancer of the breast. Unlike the women who focused on prevention and urgency, the men took the view that if cancer occurred, it would be slower growing and responsive to treatment. Relative three explained why he had accepted it and took a more passive approach:

*'There's not a great deal that I would be able to do, (risk of prostate cancer) I would go with the same process that an old school friend has recently finished a course of chemotherapy, a cousin on my mother's side, similar age to me, yeah I've just got to accept that blokes of my age have these issues and why shouldn't I be one of them. Yeah, my perception of cancers is that the older you are the slower the cancer takes' (R3).*

As such, in terms of their own risk, the men carried on with their normal life which for some, was far more complicated. One relative, for example, was going through a divorce and this took priority for him:

*'Since I had the result and I had the letter, I don't know where the letter is, it's filed away and I've just got on with other things that have been probably been taking a precedence because it's been a tough couple of years and it's just, I suppose I haven't got time to think about it really' (R5).*



### 6.2.3 The Price - Having a Test and Being BRCA Positive

As more information was obtained and understood, the majority of relatives recalled how fearful they were of the probands having the test, the timing of the test, having the mutation and the decision to elect RRM. Why was this the case? After all, this would be ensuring their survival? There was a price to this survival, however, making a difficult decision to be tested that had major implications. Relatives were concerned about what they were taking on at such a young age. One relative was devastated for example when her daughter wanted to be tested because she knew the implications and the decisions she would ultimately make, lose her breasts:

*'And then she came to me and she said I want to be tested and once she said it, it hit me like a brick. No that's something I don't want to know now, although I've done all the preparation for it, for when they were older, when she comes to me, it's not that I didn't want her to, it's I didn't want her to so soon. Young girl, that frightened me big time and I know she's headstrong as well in a totally opposite way to my other daughter' (R2).*

Despite having always encouraged her girls to be on the alert for signs of cancer because she had been diagnosed with breast cancer and deep down believing they would be carrying a gene; the reality of being told was still very frightening, because she knew what the whole process would involve and that her daughter would begin the process that would lead to RRM:

*'I think that's why I was so frightened, I didn't want to hear the fact that she would be carrying the gene but I guessed she would, do you know what I mean, hearing it and thinking about it, and you know, knowing the facts and not are two different*

*things aren't they in one respect. But yes, I suppose I was just frightened for the whole situation really' (R2).*

### **6.2.3 The Price - Losing the Breast**

All relatives, like probands and husbands, were frightened of cancer and of not surviving. They believed that probands did not have any choice other than to remove their breasts because it would lead to the best survival outcome and was, therefore, the right decision that probands were making. However, their initial thought of losing the breasts was one of 'horror'. Why did losing the breasts fill them with horror? They all thought RRM was a huge decision for any woman to have to make, with serious consequences to their body and thus their life. Relative three explained:

*'Well finding out that she has inherited it, that's, yeah and subsequently then her decision to have the bilateral mastectomy, Yes, yeah this is your little girl and a pretty serious operation. Initially, the thought of it was one of horror, but, when you begin to look at the probabilities, the survival rate, the probability, as my sister had it three times, the recurrence and then eventually you begin to realise that this is the only decision you can make. So from that point of view, we've come to accept it, it's, we would recommend it' (R3).*

As with the probands and the husbands, all relatives were reliant upon surgery to not only prevent cancer but prevent death and guarantee survival. They saw RRM as lifesaving. The price to pay for this survival, however, was risk-reducing mastectomy, but this was easier to accept than losing the

probands. Relative one shared his despair at the thought of losing his daughter, explaining why it had to be surgery:

*'If anything did happen to her or she did end up having cancer before all this, it would kill me. If it prolongs her life, if it stops the chance of her getting breast cancer, she's got to go for it' (R1).*

Although the thought of surgery frightened the relatives, having to cope with a diagnosis of breast cancer and treatment such as chemotherapy, equally frightened them. Therefore, all relatives believed that unlike family members who had died or who were undergoing treatment, probands had an opportunity to be proactive and prevent cancer and that they should take the opportunity. Relative three explained why surgery was the right decision for his daughter:

*'Yes, absolutely, I would not want to see her go through that (breast cancer treatment). She had the opportunity that my niece did not, because she had to have the mastectomy at a time when she was also having chemotherapy, so my daughter is in a much better position to recover, to bounce back' (R3).*

### **6.3 The Price - A Separation from Themselves**

Why was the whole process and surgery so frightening for relatives, even those who had been treated for cancer or done so much preparation for cancer occurring one day? They were frightened about the consequences of breast loss for the probands, leading to a different body and a different person. One relative who had personal experiences of the consequences of breast loss and the whole experience based her fear upon her own difficult experience. She

was deeply traumatised for example when she had her breasts removed, she found it very difficult to accept the way her body looked and who she had become, especially sexually. She would hide away her body from her husband. She had the same concerns for her daughter:

*'I wouldn't show my body to my husband or anything, it just broke my heart really, just seeing myself you know, coming out of the shower looking like that and I just, being so young, you know, I felt young, I didn't feel, I don't know, you're not old at thirty-one, thirty-two, are you? And I thought I'm still a young woman and I, I'm still sexually active and you know, and I just couldn't, I was just hiding away and I just' (R2).*

Indeed, the timing of the decision to have RRM was also a concern for the relatives. They worried about the loss of the breasts and the effect it could have on their marriage. Many relatives wanted the probands to wait until they were older, believing they would be more able to deal with it. Relative two wanted her daughter to take it in stages and not rush into it so that she was more mature to deal with it:

*'So, I said look, as long as you are only thinking of having the gene, the testing done and you're not going to take it any further now at your age. I said wait at least until you're late twenties, I said you're young, newly married, you know, x is so young. I mean my husband was older, he was thirty-five my husband so he was older and more mature to handle the situation' (R2).*

To deal with what? Like many of the relatives, she wanted her daughter to wait until she was older and based on the family pattern of cancer, felt time was on her side. This wait, she believed would enable her and her husband to

deal with the implications of breast loss, such as a radically altered body and a change in the way she would feel about herself. Relatives were filled with sadness as well as fear at the thought of the probands having to lose their breasts so young. They were concerned about how the drastic body changes would affect the probands' husbands or potential partner. Why were they so concerned about the husbands? Many relatives had also seen the photographs of the surgery because they had attended clinic with the probands. Thus, after attending clinic with their daughters and seeing the photographs, they were so shocked at the images of RRM, that they were immediately concerned about their son in laws. They were fearful that they would also be very shocked and avoid physical contact. Two relatives share their fears:

*'I am conscious of my scar so, with (pauses) so from I think the photographs, if she has a swimsuit on or a bra on you are not going to see any difference so you know, but I think she and I am worried about what x is going to be like with her. I think (pauses) I think he will sort of umm, I hope I'm wrong, but I think he will stand back a bit at first and she might feel it then. And I think you know, I don't mean in a sexual way, but you know like just a cuddle or something like that, you know and its, I think that's when she is going to feel it the most' (R1).*

*'Scared the life out of me, broke my heart, I really didn't want her to go ahead with it there and then. I really wanted her to wait until she was at least about twenty-eight. I say twenty-eight, not thirty-one because the generations have got younger. My mother was thirty-six, I was thirty-one, so I knew sort of in my mind that she had to do it a little bit sooner but I just wanted her to sort of more or less get her twenties out of the way you know' (R2).*

### 6.3.1 The Price - Separation from The Marriage

The concern about the impact on the husbands' was a constant focus throughout the relative's stories. Indeed, the majority of relatives expressed concern that relationships could end because the husbands may not be able to deal with it. Relative two was concerned about her young son in law staying in the marriage:

*'And her relationship and everything, young children, I didn't want, I mean x is as good as gold and I didn't doubt him supporting her in any way but there's always that percentage that it could have gone wrong, it could have broken up, because a lot of men can't handle it' (R2).*

Why did she feel he may not handle it, handle what? She was concerned that he was too young and immature to handle the changes to the body and the sexual difficulties that they would encounter. She relayed a conversation that she had with her daughter when she first learned of the surgery:

*'I said you're young, newly married, you know, x is so young. I mean my husband was older he was thirty-five my husband so he was older and more mature to handle the situation' (R2).*

The degree of concern that relatives had about the marriage surviving also depended upon how the probands would feel about their altered bodies and the changes to self. Relatives worried if they would be able to cope with the changes. As with the experiences of the husbands, all relatives believed that image was important to all of the probands and knowing what the results would look like, they were concerned about the psychological affect it would have on

probands and ultimately their marriages. The long-term psychological effect of RRM for the probands and their relationships was, therefore, a major concern for all relatives. Two relatives share the reasons why they were concerned:

*'Yeah because I know it's alright for me to turn round and say well this is why she should be ok you know she hasn't changed really, it's still her inside, but she is so obsessed with her flat stomach and how she looks. That's what's worrying I think, it will be one of the major worries as well, and how will she cope afterwards' (R1).*

*'she says she will be alright but, she seems to be ok handling it, but I think she is a bit you know, reading between the lines and I think she is a bit concerned and I think psychologically, I don't know how she's going to cope with it later if I'm honest' (R3).*

### **6.3.2 The Price - Feeling the Guilt**

In addition to sadness and fear, all relatives experienced guilt, not just in relation to passing on the mutation but because of the need to elect to have RRM and the implications on the probands bodies and relationships. The degree of this guilt was demonstrated by relative one who remembers the day he was told the result by his daughter. He could not apologise to her enough:

*'So, when we first got told, I was on the list for a transplant, I didn't connect anything to do with me and it wasn't until you know a couple of minutes after she told us, because obviously, we got very upset straight away, that I realised that I must have passed the gene onto her and the first emotion I had was guilt' (R1).*

*'The initial feeling was guilt, guilt and sorry for her, sadness for x. It was a mixture of, as I said earlier, guilt, sort of feeling*

*sorry for her and apologetic to her. And I've spoken to her and said oh I'm so sorry for passing it on and she said well it's not your fault' (R1).*

This guilt was even more salient if probands were not dealing with their experience of RRM and had changed as a person. Relative five for example who also had a mutation and who was very close to his sister, felt very guilty about what his sister was going through especially because she was not the one who had started the whole process:

*'You think well I'd rather be worse off than her, you know, that's what you think, you think, there's a bit of guilt there as well and I'm not sure about my other sister because she was the one who started the whole ball rolling and she was negative and doesn't seem to get involved that much' (R5).*

Why did he feel guilty about her? He missed his relationship with his sister which had changed drastically because she had changed as a person following the RRM. Everything was the same for him despite having a mutation but the result of RRM on his sister's body had been devastating for her and it was difficult to watch the psychological effect it was having on her:

*'I don't think it's a physical thing, all the stuff she's had done or maybe the future risks, it's probably what she's going through more than anything else after the surgery, Yeah and psychologically as well, you know it is affecting her, you know she's conscious of her clothes, it's not nice to see, she's quite a bubbly confident person and that's taken it off her, she doesn't like admitting it, she doesn't like talking and I think she buries herself in her work a bit, to get over that fact. She's so different now' (R5).*



Other relatives also felt guilty because the probands had to deal with the whole process from testing through to surgery and some were concerned that they would not be able to do it. Relative four, for example, who tested negative and who also felt guilty for not having the mutation, was concerned about her sister who had a history of depression. Although relieved to be able to keep her own breasts as these defined who she was, she felt very guilty because her sister was also very conscious of her image and liked her breasts. Relative four explained why she was so worried about her sister coping with everything and why she felt so guilty:

*'I'm worried about her dealing with it because it's her, it's, so my first thought was she's never going to deal with it. I kind of kept saying to her don't be surprised if they don't test you because of your mental state, you know her kind of very fragile state at that time. Basically, how the hell is she going to cope with it, but I mean obviously she got tested in the end, I was negative and she was positive' (R4).*

*'whilst I'm negative I do have that guilty feeling because it's oh god, I also believe I have nice breasts and I rarely if ever wear a bra, I sometimes have to wear one for work, other than that I don't wear one and you know then seeing her, I feel guilty because I'm going in there full of health' (R4).*

## **6.4 The Altered Child's Trajectory**

As with the probands and the husbands, relatives were focussed on the risk and implications for children in the family. Guilt, fear, sadness and shock were feelings that many of the relatives experienced after realising the inheritance pattern because of the wider implications for their own children and future

generations. For one relative, the devastation and guilt of passing the gene to his children and potential grandchildren was vivid:

*'Umm we stood in the kitchen and x said oh I've had a test and I have BRCA1 gene, so I said, what! She said well x is clear, I got it. So I said what you mean BRCA1 gene. She said it's a gene that causes breast cancers and ovarian cancers. I said what, how you got that. The first thing I said was how you got that, being a bit naïve'. 'But it was (thinks) how can I put it, it was when she came across and told us she had the gene and then told us the implications, its it is quite a kick in the stomach, it's very you know, it's the feeling of guilt and you know' (R1).*

Why did they feel such guilt and responsibility? They felt responsible as parents for passing the gene on especially if there was more than one child that could be affected and because grandchildren were involved. They felt guilty because of the decisions and the altered life trajectory that their children and future generations were now faced with. Relative one had two out of three children who were positive which increased his guilt because there were, even more, children to consider:

*'He came back as positive and I thought oh no, not another one you know but as you say, by then I had sort of, obviously, as I said earlier when she told us, there was the feeling of guilt that I passed it on to her and then more guilt' (R1).*

Passing on a mutation to future generations meant that the consequences of this had to be considered by the probands. Relatives felt they had burdened their children with extra worries and dilemmas about their own life trajectory and their potential offspring. Relative three, for example, believed he had introduced extra problems for his daughter and future family:

*'She's now got that 50/50 chance and so as hers came closer, again a repeat of those guilty feelings and it sort of, everything goes a bit quiet when she gets the result that she's got it too because you know then, that she's got all the decisions to be made. My daughter is in a different position in that if she should choose to have children, she actually knows. My father and I and how many generations we didn't know, had no inkling, so it's uncomfortable, it does put quite a burden on her and x that we didn't have' (R3).*

Although the relatives realised that they had no control over whether or not their children (including the probands) had a mutation, their main concerns were for their children and future generations. They did not want the normal childhood trajectory to be affected by introducing constant fear in their lives nor their future grandchildren getting cancer. However, the experience of a mutation in the family and the subsequent risk for children had in many ways altered their life trajectory. For example, despite still being very young or even babies, they were already anxious about them having a mutation. Two relatives share their anxieties:

*'I've got my son and I don't, he doesn't want to know, which it's no, I'm a boy, I said but you know it can affect you, you can carry the gene and you have got a little girl. He's like mum, maybe in the future, I do not want to think of, at the time she was only like a baby, well she's only three now. I don't want to think for the next twenty odd years that my daughter, I could have passed a gene onto my daughter that could give her breast cancer. I can't worry about that for the next twenty odd years, well when it comes that she is older...'* (R2).

*'I didn't have any thoughts about it apart from oh no, the kids have got a 50/50 chance and that was about it. I didn't think anything personally about myself or the implications, all it was, well I hope the kids haven't in years to come type of thing'* (R5).

#### 6.4.1 Preserving the Normal Child Trajectory

All the relatives were, therefore, trying to protect their children and grandchildren by ensuring life was as near to normal as possible. Like all the family, relatives did not want to burden the children or spoil their childhood with the worry. Relative two, for example, explained how although she had prepared herself and her daughters that breast cancer would occur one day, she had particularly tried to protect her children from having an altered childhood, interrupted by constant worry or disruption. This was achieved by not telling them the whole story of her breast cancer when they were younger:

*'But I've prepared myself my whole life for it and I think the girls have as well, you see, because they've all, I've never rammed it down their throats and when they were children always protected them, never give them a full awareness of the whole situation, you know' (R2).*

Indeed, it was precisely for this reason that the wife of one of the relatives did not want him to be tested because it would introduce risk and fear into their children's lives, including awareness of their own risk. He explains:

*'my wife didn't want me to have it done because of the kids and that's about as much as I knew, to be honest until and then x said she was going, and x was negative and x was positive' (R5).*

Relatives also had a fear that their children and grandchildren would get cancer because of a mutation and that they may lose them. This would also mean an altered life trajectory for their grandchildren, witnessing the

devastation of cancer treatment and death. Relative one tried to imagine what would happen if that were the case:

*'god what's going to happen with the kids, will she see them, you know, will she be there to see the kids and see them married and will she become a grandmother and obviously thinking about that type of thing umm' (R1).*

Passing the gene mutation onto future generations was, therefore, a concern for all relatives, especially for the females who would have the difficult decision of losing their breasts. This concern from the relatives was influenced by having witnessed the experiences of the probands. Relative five, for example, was concerned for his own young daughter and electing RRM based on seeing the impact on his sister:

*'So I wasn't so much concerned about x which is weird because they're both my children, it was more x because she was a female and that's probably because of the impact on my sister. That's probably why subconsciously I'm thinking well me and x are just going to live through it but you know, prostate or whatever, but the thing is it's more impacting on a woman isn't it' (R5).*

The relief of not having a positive gene and not having to worry about passing it on to future generations was also evident. Relative four, for example, who tested negative, was so delighted her daughter's lives would not be turned upside down:

*'I mean I was obviously extremely happy about it, simply because of my daughters, I'm not giving something horrible or there's not a chance that I'm giving this something horrible to*

*my daughters. They've just got other things that they have to deal with sort of thing' (R4).*

## **6.5 Summary of Experiences**

For the majority of relatives, a positive BRCA mutation in the family was devastating and they were mainly concerned with their children's (including probands) and grandchildren's risk and survival. Importantly, they felt responsible and guilty especially the parents in the sample, for the need of their daughters and future grandchildren to elect to have RRM in order to survive, which was a result of them passing on a mutation. They were especially concerned about the real and potential problems caused from a drastic change to the body and relationships as a result of RRM. This fact being the main reason for their continued guilt and sadness. Literature had identified that family members feel guilty in passing on a gene mutation (d'Agincourt-Canning, 2006b; Hallowell et al., 2006; Stromsvik, 2010., Shiloh et al., 2013) but the above reasons for this guilt, offers a new contribution to knowledge.

Relatives feared for current and future generations in terms of the timing of the test, the test result and the effect on their normal childhood trajectory, including their potential decision to elect RRM, believing that all these factors had now altered their life trajectory. They all feared for these children's relationships and the effects of breast loss on their body, their confidence, their marriage and their self-identity. At the same time, they also feared a diagnosis of breast

cancer for any family member and the effect this would have on the children's normal life trajectory.

Despite testing positive for the mutation, themselves and being at risk of both breast and prostate cancer, the concern for their own risk was secondary to their children. They believed that some form of cancer was inevitable and that the older it was contracted, the better the outcome.

## **CHAPTER SEVEN – Discussion and the Expanded Horizon of Understanding**

The three horizons of understanding that unite the probands, husbands and the relatives resulted from the interpretation of the most salient aspects of the experiences. Each horizon developed from the data by viewing the parts in terms of the whole (Gadamer, 2004). The new fused expanded horizon of understanding, 'disembodiment', the experience of living with a different body, disconnected physically, emotionally and socially from the original body, offers an explanation of why women undergoing RRM experience such negative impact on femininity, sexuality and identity. This new contribution to knowledge resulted from the interpretation of the experience of RRM for the probands primarily, but also draws on the husbands and relatives experiences of the consequences of RRM for the probands and their relationships; - a separation of themselves, leading to 'disembodiment'.

This is the first longitudinal qualitative study, to my knowledge, that explores the experiences of BRCA positive women, electing to have RRM, their husbands and their relatives. Utilising the hermeneutic circle of understanding, forty-three interviews were interpreted, they revealed a text that connected the experiences of the women, their husbands and relatives, providing a new fused horizon of understanding of their experience. In this discussion, the united interpretations and the three horizons of understanding including the



expanded horizon, 'that of disembodiment', will be explored by revisiting the research questions.

## 7.1 Research Question Revisited

My overall question was, *'What are the experiences of living with the BRCA gene mutation and electing bilateral risk-reducing mastectomy for the woman, her partner and her family'*. In particular, *'what does it mean to be a BRCA mutation carrier and what is it like living with someone who is a carrier?'* *'How do women and their families make sense of the BRCA mutation, come to the decision of surgery and what are their experiences?'* *'What is the experience of undergoing surgery and living beyond surgery?'* *'Why is sexuality, femininity and identity so adversely affected?'* Finally, *'what are the needs of both the women and their families in terms of information, assessment and nursing care?'*

It would be impossible within the remit of this Ph.D. to discuss all the horizons found within the data but as Gadamer advises, in order to understand experience, it is necessary *'to seek out those elements that we have found in our analysis of experience that are general'* (Gadamer, 2004; pg. 352). Three main horizons, therefore, united the experiences of the probands, husbands and family and underpinned the new fused horizon. The price of survival, the altered child's trajectory and a separation from self-leading to the new expanded fused horizon of understanding 'disembodiment'.

### **New Fused Horizon of Understanding**

Being a woman or a family member living with the BRCA gene can mean living a life of constant worry and fear of not surviving and this is not just individual, but is a complex family affair. Taking all available steps to survive fulfils a moral obligation as a wife and mother to act responsibly to this risk information and is not just for the sake of self, but is also part of a wider responsibility to their family. Doing so however, means making the difficult decision to elect to have bilateral risk-reducing mastectomy. The results of this surgery for probands in this study was both socially and culturally undesirable and had far-reaching consequences for the whole self because of the importance of the breast for identity, sexuality and gender. Thus, proceeding with bilateral mastectomy, despite reconstruction was associated with a separation of the whole self and disembodiment, an experience of living with a different body, separated from who one was and who one is perceived by others. This new state of being affects the woman and her wider family. There is a need for preparation, assessment, information and importantly, ongoing support.

The three horizons will now be discussed alongside the research questions.

## 7.2 The Price of Survival

- *What does it mean to be a BRCA mutation carrier and what is it like living with someone who is a carrier?*

Preventing cancer when given a diagnosis of a BRCA mutation has already been identified as a priority in the literature (Howard et al., 2009; Lostumbo et al., 2010; Brandberg, et al., 2012; Jeffers et al., 2014). Fear of cancer and an uncertain future drives the decision to seek testing and elect RRM (Lodder et al., 2002; Van Dijk et al., 2003; Watson et al., 2004; Antill et al., 2006; Graves et al., 2012). This was also found in the current study but extends the existing knowledge by highlighting that it is not just a matter of prevention, but one of survival. All participants in this study believed that being BRCA positive had more serious consequences and therefore without surgery, they would not survive. Having a BRCA mutation meant 'being BRCA'. This meant having both the current risk and the future risk, of which they perceived breast cancer and death as inevitable if they did not act upon the information. Being BRCA positive without surgery signified cancer and death. There was never a question of if participants got cancer, for them, it was a matter of when.

Probands, husbands and relatives believed that RRM in particular was life-saving and that they had no other choice. This finding challenged some of the existing pre-understandings, the focus being on preventing cancer, preventing the need to undergo surgery and chemotherapy in the future (Hoskins and Greene, 2012) or where other options such as surveillance are taken (Howard

et al., 2011). In this current study, there was a sense of not trusting their bodies to stay healthy or fight cancer should it occur and surveillance was therefore out of the question.

Importantly, this current study demonstrates that there is a 'price to this survival' for probands and their family. By seeking a genetic test, they experience an unexpected long difficult journey that leads to the identification of a positive mutation and the complexities of the experience unfold as the journey continues. Conflict within the family, the unexpected impact of results, difficult life changing decisions which affect probands, husbands, children and the wider family, electing to have RRM and living with the consequences of surgery, are just part of this price.

In this study, more than half of the probands had witnessed the death of their mother, therefore, the fear of cancer and of not surviving was even more salient. Findings that support other studies which show that a perceived high risk influences uptake of both testing and surgery (Hallowell et al., 2001; Antill et al., 2006; van Dijk et al., 2008) and that the death of a mother, in particular, has greater significance, leading to definitive risk-reducing action (Hamilton et al., 2009; Werner-Lin, 2012). For the family members in this study, there was also a constant fear of the probands and future generations dying of cancer. The majority of the husbands and relatives were totally preoccupied with the fear of a BRCA mutation, but interestingly, the male relatives, who also carried a BRCA mutation, were not concerned about their own cancer risk or survival.

This is in contrast to the findings of Shiloh et al., (2013) who found that 74% of participants in their study increased their surveillance for cancer and were still significantly distressed four years after testing, compared to BRCA negative men.

Probands with (7) and without (1) children were heavily influenced to have the test and elect surgery for their children's sake, a concern that persisted because they described their responsibility for their children's lives. Findings consistent with other studies (d'Agincourt-Canning et al., 2001, 2006a; Hallowell, 2003; McEwan, 2011). Probands also felt a moral obligation to their own and any future children in maintaining the ability to care for them by doing the 'right thing' as responsible parents, findings supported by Crabb and Le Couteur's, (2006) discursive analysis of how one woman's decision to elect RRM and remove healthy breasts was portrayed as morally responsible and self-sacrificing. These findings are also consistent with other studies where women privileged motherhood and the maternal representation of their breasts (Hallowell, 1999, Bury, 2008, McEwan, 2011). In these studies, women described a strong moral obligation to their kin to do the 'right thing', in doing so, however, they relinquished their right not to know, forcing them to act upon the information. Due to the complex family dynamics and initial secrecy concerning who should and should not know and when they should know, in this current study, some probands were also forced to act upon the genetic test results of others which meant they were presented by other family

members with a moral obligation to seek testing for their children's sake, and this caused divisions within the family.

In other cases within this study, where probands wanted to pursue testing and elect to have surgery, surprisingly, some family members could be obstructive, unwilling to provide a sample of blood for testing and relationships were complex. This raises many ethical considerations as identified by Dupras and Ravitsky (2013). On the one hand there is the right of the patient to privacy and confidentiality but on the other hand, there is the right of family members to receive clinical information if it affects and involves them personally. Furthermore, Hallowell makes the point that having a genetic mutation is not about individuals, it involves biological relationships and it is a family affair with far reaching ethical and moral implications such as who to tell, how and when (Hallowell, 2006). Moreover, participants in Hallowell's (1999) study with women at 'high risk' of breast and ovarian cancer but who were not BRCA positive, demonstrated that in doing the right thing, women felt an obligation to put the needs of others before their own.

This was even more salient for the women within this study who were not just categorised as 'high risk' but had a BRCA mutation, which meant that the probands and family once given a mutation result, felt they had done everything they could for their kin by electing to have RRM. Probands strongly considered and acted upon their obligation to their children, their husbands and their family members, putting their own needs second. This supports but

also extends knowledge specifically for BRCA women, because it also demonstrates how family responsibility, including gender responsibilities such as motherhood and social obligations, strongly influence decisions around testing, preventing cancer and importantly electing to have RRM.

At the time of disclosure of the mutation result, surviving cancer and the moral obligation to children was therefore at the forefront of all decisions. The effects of RRM on the body was not as significant. One explanation for this is that the breasts were initially seen as 'dangerous objects' that needed to be removed, findings consistent with Hallowell's (2000) study. Probands and family members in this study unlike many other women with a BRCA mutation who choose surveillance, did not trust that mammograms would detect cancer. This extends knowledge and was the experience for five of the probands.

The majority of participants in this study reported the need for more information and support. Although Some of the previous literature had suggested that more information surrounding RRM and the psychological effects on wellbeing was required, research limitations had resulted in conflicting conclusions, making it difficult for women to be well informed (Lloyd et al., 2000; Payne et al., 2000; Josephson et al., 2000; Hatcher et al., 2001; Bresser et al., 2006; Rolnick et al., 2007; Isern et al., 2008; Spear et al., 2008; Gahm et al., 2010; Wasteson et al., 2011, McEwan, 2011). Glassey et al (2016), identified that for younger women, in particular, electing to have RRM has unique information needs, yet there has been little focus on their experiences. This current study,

therefore, adds a new contribution to knowledge by identifying when and why younger women, in particular, need more information and support. They needed information and support specifically before testing to understand the implications of a positive test result, whilst trying to accept their result, whilst making difficult decisions about surgery and whilst dealing with concerns about their children. Post –surgery counselling was especially required when they were dealing with sexual and sensitive issues as a result of the drastic changes to their bodies. There was also a desire to meet other women with a BRCA mutation to see real life images of the expected surgical outcome and to discuss personal feelings. Both findings offering a new contribution to knowledge with important implications for practice. Future Counselling was specifically recommended by participants to prepare other BRCA women and their husbands for the drastic changes to the body and support to deal with the negative changes to sexual relationships.

- *How do women and their families make sense of the BRCA mutation, come to the decision of surgery and what are the experiences of risk-reducing mastectomy?*

Despite genetic counselling, making sense of the BRCA mutation varied amongst the participants especially the younger probands and family members because of their limited knowledge of inheritance, genetics and because a specific BRCA gene mutation was unknown to them. The link between breast and ovarian cancer risk was a shock for many of the



participants as was the link between risk-reducing breast and ovarian surgery, they were not aware that this latter procedure had also decreased their risk of breast cancer by 50%. For all participants, having a BRCA mutation was a life changing experience. They had breasts and ovaries to worry about but the decision to elect to have RRM was by far the most difficult. Losing their ovaries did not have the same impact and many of the women following their results decided to have their ovaries removed first because they perceived this as the easier option, less visible and less threatening to body integrity. Findings supported in the study by Hallowell and Lawton (2002) and Howard et al., (2011). Why was losing the ovaries an easier decision?

In addition to being aware of the lifestyle and environmental risks for breast cancer, the probands, experienced a corporeal risk as described by Kavanagh and Broom (1998) and Hallowell (2000). In Kavanagh and Broom's study of abnormal pap smears, they identified a Cartesian splitting of body and self in their participant's, where the body was objectified and the self was not seen of as 'at risk'. This enabled the objectifying of their 'risky' body parts (cervix) and thus their disposal (removal of the cervix), as not having any implications for 'self'. It is proposed that the probands in this study were also able to objectify their ovaries. However, with regards to their breasts, this was far more difficult. The probands in this study viewed their breasts not just as 'risky', but as dangerous and life threatening and in order to make and justify their decision to undergo RRM, and control their 'dangerous' body parts (their breasts), it is proposed that initially, they also experienced a Cartesian splitting of body and

self, where the risk was seen of as residing in the 'body' but not as of the 'self'. Many of the women viewed their breasts as '*ticking time bombs*' that they '*wanted gone*' and thought it would be easy to dissociate from because of the physical risk. However, it was not as simple as this and after seeing images of surgery, it soon became evident that RRM and the loss of the nipple especially, as potentially threatening other aspects of self, their self-identity as a woman and their emotional self.

On one level all participants felt a great need to preserve their physical health and stave off the risk of death because they perceived they would not survive and they had to act upon the information they had been given. They believed that electing to have RRM was the only way of managing and preserving their physical health. This reflects a finding in the study by Howard et al., (2011), who reminds us of the four dimensions of self; physical health, self-identity, relationships with others and the emotional self. Preserving self, could be understood in various ways in relation to the four aspects identified by Howard. Women elect to have RRM in order to preserve the physical health aspect of self as opposed to those women who choose other risk management strategies such as surveillance, these women are attempting to preserve their self-identity and also their emotional self (Howard et al., 2011). Instead of choosing surveillance which the majority of women did in the study by Howard et al., (2011), the women and families in this study, with their focus on survival, privileged their physical health, by electing to have surgery.

This choice was made, however, without realising the full extent of the effect that RRM would have on their other aspects of self, namely self-identity, relationship with others and their emotional self. It is argued that the reason for this was that they anticipated breast reconstruction would restore the aspect of self-identity and preserve their emotional self and the relationship with others.

Without realising it, decision-making, therefore, went beyond preserving their physical health because of the implication of the other three elements of self as identified by Howard et al., (2011), that of self-identity as a woman, relationships with others and the emotional self. The probands and family, for example, showed concern about aspects of the decision to elect surgery, how family and friends would perceive them without breasts and how they would appear physically, emotionally and socially. Probands, relatives and husbands in addition to wanting to preserve the physical health (their survival), soon became concerned about the emotional self and the effects of surgery on their self-identity. Thus, although they did not see any alternative to surgery in maximising survival, pre-surgery, they had reservations about physical appearance post-surgery and how femininity and sexuality would be affected.

The importance of identity and how the body is experienced socially, has also be highlighted by Fox (2012) and many other scholars with an interest in embodiment. Bartky (1990), Merleau-Ponty (2002), Butler (2004) and van Manen (2014) who remind us that gender plays a critical role in the way that

embodiment is experienced. More importantly, how society and culture construct gendered bodies, thus having breasts means 'being' a woman. There was a huge price to their survival therefore, for the women in this study—losing the breasts with all the social and cultural meanings attached to the breast, such as womanhood, femininity, identity and sexuality. Fox (2012) argues that we need to understand human bodies and 'selves', not so much as entities in their own right, but as connected to and always in relationship with both their physical and social environment.

Thus, for women with a BRCA mutation seeking RRM, it is the type of risk, the perceived danger, the location of the breasts and the social and cultural meaning of breasts, which present the fear and dilemma for women and their families and which is the price they pay for survival.

### **7.3 The Altered Child's Trajectory**

The family risk and fears continued onto the next generation and consequently, there was ongoing concern about all the children within the family, even though many were still very young. This was consistent amongst all participants and supports some of the pre-understandings (Lodder et al., 2001; Metcalfe et al., 2004a; Kenen et al., 2006; Mireskandari et al., 2007). However, this study extends knowledge by highlighting the focus of these concerns and fears. Fathers, in particular, were totally preoccupied by their children's risk of inheriting a BRCA mutation, especially their daughters and

were extremely anxious about how and when to tell them. Moreover, they were already beginning to worry about the next generation and their granddaughters.

My continual questioning of the text led to me ask why they were worried when their children were so young. Why were they already assuming that future generations would test positive when there was a 50% chance that they would not have the mutation? Instead of being able to enjoy their children and grandchildren, the present was clouded by what they perceived as 'problems that lay ahead of them'. Their biggest fears were that their children would not survive cancer but also, they feared their children having to elect to have RRM with all its implications for their future relationships. Thus, they viewed their children and future children's life trajectory in a different light, believing that others would also view them differently if they had a BRCA mutation. They believed that the children now been given a burden to live with and extra issues to consider on top of the challenges of puberty, adolescence and adulthood.

Some of the men in this study, therefore, found it difficult to share the results with their children, findings consistent with other studies (Daley et al., 2003; Hallowell et al., 2005) but increases knowledge by highlighting the reasons why. Parents, in particular, assumed that their daughters would have a mutation and were mindful of not disrupting the key transition periods in their lives (Hamilton et al., 2009). They particularly feared the thought of their

daughters losing their breast and the effect this would have on their marriage, based upon the experience of the probands. For this reason, some of the children had not been informed and the men in the family took on the responsibility for the decision of whether or not to tell and when to tell.

This is in contrast to other studies where women have taken on the main role of disclosure (d'Agincourt-Canning 2001; Forrest et al., 2003). The probands and family were less concerned however about their sons because they did not have to lose their breasts. For their daughters, they feared for their future husbands, marriage and their self-image. This led all participants to feel a huge amount of guilt and responsibility which was evident in all of the experiences.

Most of the relatives in this study, as well as feeling an obligation to be tested, felt a huge amount of guilt in having passed on or the potential to pass on, a mutation to their daughters (three of the probands), findings consistent with other studies (Liede et al., 2000; Lodder et al., 2001; Stromsvik et al., 2009). However, this study reveals that the guilt was because their daughters and granddaughters would have to lose their breasts, the implications for their body, their future, their relationships and opportunities for marriage. Furthermore, unlike previous studies (Mc Allister et al., 1998; Liede et al., 2000) where men deliberately avoided talking about RRM with the family, when asked the question '*tell me about the BRCA gene*', loss of the breasts, was the key focus and concern for all the men in this study. Siblings in this

study also experienced guilt, whether they did (1) or did not (1) have the mutation. This guilt was again because of the implications of breast loss for the women. Unlike findings in other studies (Leide 2000; Daley et al., 2003) where constant measures for surveillance were sought, relatives rationalised that they could do nothing about their own risk of cancer and appeared to have taken a fatalistic approach, accepting the fact. In this study, all their concerns focused on the probands and their future kin.

#### **7.4 A Separation from Self - 'Disembodied'**

- *What is the experience of undergoing surgery and living beyond surgery?*
- *'Why is sexuality, femininity and identity so adversely affected?'*
- *What are the needs of both the women and their families in terms of information and care delivery?*

Although RRM was a priority for the majority of participants, it was still an experience that caused great anxiety both pre and post-operatively. Pre-surgery the fear of breast cancer, but importantly, the fear of not surviving without electing RRM, dominated their experience, findings that add new knowledge. Post-surgery, participants experienced a separation from the fear, the fear of cancer had resolved for the majority, findings supported by other studies (Frost et al., 2000; van Oostrum, 2003; Dowdy et al., 2004; Brandberg et al., 2008; Hamilton et al., 2009; Miller et al., 2010; Hallowell, 2012), only to be replaced by pre-occupation with the body and their children. These findings

offer a new contribution to knowledge. Indeed, for some, there was complete resolution of fear regarding the probands' risk which continued long-term. However, the fear had shifted to the fear for their children, mainly daughters and concerned the issue of breast loss.

Post-surgery, the effect that RRM had on their bodies became the main focus and preoccupation for all participants because their expectations of the results of surgery were very different to the actual results. For the majority, they were expecting more of a cosmetically acceptable procedure despite seeing images of the surgery. All probands and their husbands hoped that their result would be better than the images had portrayed. The reality, however, was that for the majority, they were not. The breast without a nipple made a significant difference to the way probands and husbands perceived their post-surgical body. It signified a difference in looking normal and the meaning of being feminine and of being a woman. Despite believing it was the safest decision, the loss of the nipple had a major unexpected impact on how probands felt about their bodies. A nipple reconstruction later in their journey made some difference to increasing their confidence.

The anxiety about the effects of surgery on self-image, femininity and sexuality as found in many of the pre-understandings (Hopwood et al., 2000; Payne et al., 2000; Hatcher et al., 2001; Bresser et al., 2006) was very evident in this study especially from the experiences of the husbands and the family. Although electing to have RRM is regarded as a way of preventing cancer and



avoiding the devastating effects of treatments like chemotherapy, especially in younger BRCA positive patients (Hoskins and Greene, 2012), it also has its own devastating consequences on the body. For this reason, initially, all of the participants believed that breast reconstruction would be the answer and would prevent the emotional impact of losing the natural breast. In reality however and after seeing the likely cosmetic result without nipples, this was challenged. In support of Harcourt et al's, (2003) findings, post-surgery, the over reliance on breast reconstruction to restore the effects of the loss of femininity and self-image was evident and the majority of probands were dissatisfied. Furthermore, as found in some studies, although the majority of probands did not regret having the surgery (Metcalfe et al., 2004a; Brandberg et al., 2012; Gopie et al., 2013) and felt that they had done everything possible to survive, they all experienced a loss of their natural breasts (Crompvoets, 2003; Hamilton et al., 2009), especially nipple loss. Husbands and relatives were distressed and concerned about the effects that RRM had physically and emotionally on the probands, believing that they now looked very different, had changed in personality and for one husband '*destroyed his wife*'. Similar to the women in Piot-Zeiglers et al's (2010) study where mastectomy with reconstruction, provoked a painful experience of body deconstruction, the probands in this study endured a similar experience but one of 'disembodiment'. Like the participants in Piot-Zeigler's study whose aim was to examine the consequences of mastectomy on identity, probands experienced dissonance when comparing their former intact body with their now challenged, compromised body. Although women in Piot-Zeigler's study

had breast cancer, it is argued that the experience of feeling mutilated following mastectomy, was similarly experienced in this study. Furthermore, because probands in this study did not have cancer and were separated from their healthy breasts as opposed to diseased breasts, it is again argued that it was even more difficult to accept and integrate their new corporality.

Unlike the ovaries or cervix which are invisible and easier to dissociate from (Hallowell and Lawton 2003; Howards et al., 2011), the dissociation from the potentially cancerous breasts was far more difficult because of their overt visible presence, the social, cultural meaning attached to them and their connection to femininity and identity, and as such, probands and husbands grieved the loss. Relatives also highlighted the negative effects on the probands and how probands were suffering from the loss. This loss was a result of their breasts feeling different, they looked different and consequently, they felt differently about their bodies, especially sexually. Findings consistent with Hallowell et al's (2012) study. Most probands and some husbands talked of the negative effect on their sexual relationship and how the emotional connection was absent. It is argued therefore that the drastic changes to the body and the effect on femininity, identity and sexuality caused by electing RRM, resulted from women feeling that they had been unexpectedly, and permanently separated from their bodies. This separation leading to 'disembodiment'. It offers a new contribution to knowledge by demonstrating why women have such altered body image and report problems with femininity, their sexual relationships and identity, as highlighted in previous

studies (Hatcher and Fallowfield, 2003; Altschuler et al., 2008; Brandberg et al., 2008; McEwan, 2011). Their body image, sexuality and identity had been negatively altered because their breasts did not feel a part of who they now were, separated from their body and their 'selves' as women.

Many of the findings in this study also resonate with some of the pre-understandings and in particular, the findings of Hallowell et al., (2012) where women were found to experience a lack of sensation in the reconstructed breasts, yet feelings of hardness in the implant reconstruction. Many negative changes in their bodies were experienced which probands and husbands felt unprepared for. The reconstructed breasts even if they were 'looking like breasts' did not feel like breasts and were not seen as part of their body. Therefore, the majority of probands were inhibited sexually because of the lack of sensation, affecting their relationships. Maloney and Kirkman (2005) remind us that sexuality encompasses the whole self, including thoughts, desires, sensations, emotions and identities. In the study by Werner-Lin, (2008) similar findings were reported where women with a BRCA mutation were found to be sexually inhibited because the reconstructed breasts became triggers for the distress that they had encountered and the sacrifices they had to make to accommodate the risk. Moreover, the breasts were regarded as fake and the single women described themselves as damaged goods and imperfect. The probands in this study, despite reconstruction, reported feeling less feminine, '*masculine*', even '*boy like*'. They hid away and covered their bodies up feeling intimidated in the presence

of other women. I would argue, therefore, that many of the probands in this study also felt imperfect and that their breasts were experienced as fake. They also experienced a different gender, not recognising themselves as female or feminine, which did not fit it with what is culturally acceptable or what it means to be a woman as suggested by De Francisco and Palzowescki, (2007).

One explanation for these reactions is offered by Butler (2004). In order to produce a gendered subjectivity, Butler contends that femininity and masculinity are bodily styles that we incorporate and are ones in which we act out, not just ourselves but in relation to others and which others act out in relation to us. Pelters (2014) supports this view in her study where women were found to have both a normative view of femininity in line with gender functionality and an alternative self-determined femininity. Bartky (1990) also reminds us of the ideal and properly feminine body, a body with two breasts and how this is socially constructed. She argues that the properly feminine body is maintained and produced as a result of disciplinary practices such as dieting, exercise, makeup and plastic surgery. This is evidenced by how many women in society constantly pursue the ideal body image, seeking approval through behaviours such as breast enhancement or cosmetic surgery. Many of the probands in this study believed their surgery would be more cosmetically acceptable despite seeing photographs, this adds a contribution to knowledge and has important implications for practice in preparing women and setting realistic expectations.

As a result of this discipline described by Bartky (1990), the feminine body looks a particular shape and moves in a certain way providing individuals with a sense of identity, that of being and looking '*natural*' like a woman should. Bodies in this way are disciplined to represent and comply with a social ideal and women discipline their bodies to derive pleasure and avoid social punishment (Bartky,1990). Thus, a body that is undisciplined and appears a different shape (as the women in this study describe) is seen as deficient, non-feminine and at odds with social normalisation. Furthermore, it is proposed that this management and maintenance of the feminine body also entails managing the whole self, which is an integral part of the body.

It may be therefore, that when making the decision to remove their natural looking breasts, women in this study were temporarily able to objectify their breasts and experienced an initial intentional Cartesian splitting of body and self. They did this in order to preserve their physical self from their dangerous (potentially cancerous) breasts believing that it would have little or no effect on the other aspects of self as identified by (Howard et al., 2011) self-identity, relationships with others and the emotional self. The reality, however, is that it did have a significant impact on all aspects of the self because probands could not easily part with their breasts because they are integral to the whole self. As a result of their loss therefore, probands felt 'disembodied,' with family members also witnessing this substantial negative change in the way probands felt about themselves and lived their lives post -surgery.

Thus unlike the ovaries, it makes it impossible to objectify the breasts or dispose of them without major implications for all aspects of the whole self; body and self are intrinsically one. Although it is possible that the women in this study who underwent oophorectomy may have felt unfeminine and masculine because of the effects of their oophorectomy, they did not report this as so. On the contrary, it is argued, they were more able to objectify their internal (dangerous parts) less visible body parts as described by Kavanagh and Broom, successfully separating the ovaries from self, especially for probands who had completed their family.

In this study, a feminine looking body, as well as a feminine feeling body (which normal women possess), was the initial expectation following reconstruction, by all participants, in order to balance the dilemma of disposing of their *'ticking time bombs'* and maintaining a feminine body, which they perceived as the ideal. Like the women in Hallowell's, (2000) and (2012) studies, women in this study experienced a lack of breast sensitivity, a lack of movement and a change in texture of the reconstructed breasts as compromising their femininity and as having profound consequences on their sex life, even if their appearance looked acceptable. For although women in this current study may have had breasts which looked normal to the gaze of the outside world, they did not feel or experience their body as feminine or natural.

Furthermore, like the women in Hallowell's studies and in congruence with Bartkys' analysis, pre-surgery the probands were more concerned initially about survival and less concerned with the materiality of the body, how their breasts would feel and function. They were more concerned with having a body that looked or appeared feminine. However post-surgery, probands and their family were concerned with both the materiality of their breasts as well as how they looked. This sits in contrast to some of the pre-understandings that focus on the importance of looking feminine (McGaughey, 2006; Spear et al., 2008).

Moreover, in the current study, the probands' reconstructed breasts may or may not have looked like breasts, but they did not perceive them as real breasts, even for the proband who preserved her nipples. In support of Hallowell's (2000) study and the participants in McEwan's, (2011) and Pelter's (2014) studies, it follows that femininity is not only dependent on how a body complies with the normalised looking feminine body, but involves a particular type of material embodiment (how feminine the body feels). If this material body is in some way changed or reconfigured, as in the case of breast removal and reconstruction for the eight women in this study, then it is reasonable to suggest that it had major repercussions for gender identity and was therefore responsible for the disembodiment that they experienced.

Furthermore, when individuals experience a disturbance between their body and their world, as is the case with all the participants in this study, their whole

existence is profoundly shaken (Merleau-Ponty, 2002). The probands, husbands and relatives, initially hoped that breast reconstruction would enable the probands to look both natural and feel natural, to themselves and to others. They hoped this would restore their self -identity as women.

However, many probands experienced problems with healing and had multiple operations which affected the way the reconstruction eventually looked and felt, having considerable consequences on identity and sexual relationships. Indeed, some probands felt unprepared for these difficulties and wanted to be able to discuss their sexual difficulties. As a consequence, all the participants requested support and help in this regard. These findings support Hallowell's (2012) latest study in which few of the women felt entirely prepared for the reality of the procedure. In adding to the body of knowledge, women in this study reported that despite seeing photographs of surgery, it did not completely prepare them for how the new breasts would feel and more importantly how they would experience the disruption to self. In agreeing with Hallowell's (2012) study and others (den Heijer, et al., 2012, Gopie, et al., 2013) it is recommended that women and their family with a BRCA gene mutation should not only be provided with information gleaned from this and other studies that enable them to decide on the best options, but that they should have access to psychosocial care both pre and post-surgery if required.

As time has elapsed and with more women seeking risk-reducing surgery nationally, the inclusion of a clinical psychologist in the multidisciplinary team is now becoming routine practice, highlighting the problems experienced.



Furthermore, women with a BRCA gene mutation need to be assessed and cared for in the context of the wider family who also need support because these decisions are a family affair. Breast care nurse specialists are ideally placed to provide care that includes support from genetic counselling through decision making, surgery and beyond and many are also trained in psychological therapies. However, in a time of austerity, there has to be strong evidence that this is necessary and it is proposed that the fused expanded horizon of understanding 'disembodiment' found in this study adds new knowledge, contributing to that evidence. Further research in this area is also recommended as it is acknowledged that this study involved a small sample and may not be typical of all BRCA women electing to have RRM, resulting in some further gaps in knowledge and limitations to this discussion.

It may also be that with the newer techniques of reconstruction, more women preserving the NAC and increased access to plastic surgeons in a combined multidisciplinary team, that women may report more favourable experiences. However, it is also proposed that in concurring with Harcourt and colleagues (2003), there may be an over reliance on the role that breast reconstruction offers in the restoration of a woman's femininity and sense of self especially following RRM which women and families need to be prepared for. Finally, with more units across the country providing assessment of and intervention for potential psycho-social problems pre and post risk-reducing surgery, more women and their families will be spared some of the negative, life changing consequences of electing to have risk-reducing mastectomy.

## **7.5 Strengths and limitations of this Study**

The strength of this study lies in its prospective qualitative design and the focus on understanding the experiences of eight women who all have a BRCA mutation and who elected to have a risk-reducing mastectomy with implant reconstruction. In contrast, the majority of literature (pre-understandings) has focused on pre-determined outcome measurements using quantitative designs that have included women with different categories of high risk, including those with breast and ovarian cancer, in addition to BRCA positive women.

Its other major strength is that it offers a new contribution to the body of knowledge at a significant time when breast clinics, Nationally and Internationally, are experiencing a threefold increase in referrals for testing and those electing to have RRM, both with the BRCA gene alone and women with an additional diagnosis of breast cancer. These findings have important implications for clinical practice and the way in which women and their families are cared for.

The study's final major strength is that it includes the experiences of the family corpus, that of the husbands and a sample of different family relatives including mothers, fathers and siblings who are an integral part of the experience. There are no other qualitative studies, to my knowledge, that have included all three groups of the same family, in the same study.

As a result of this study, the new fused expanded horizon of understanding can be shared with others and has already had a major impact on service delivery within the author's clinical setting. For example, women and their family members are now offered at least three meetings with the surgeon, the consultant nurse and a plastic surgeon prior to surgery. There is now a dedicated joint clinic, which runs each month, that incorporates the geneticist, the surgeons (including a plastic surgeon), the consultant nurse and new aspiring specialist nurses.

Women now have the option of all the surgical reconstruction techniques, for example, implant techniques using both internal or external skin to support the implant (dermal sling and strattice), muscle flaps including using the abdomen, buttock and inner thigh fat and preservation of the nipple areolar complex (NAC). More importantly, and in line with the recommendations from this study, the potential for body image, gender identity, and sexual issues are at the core of the discussions and assessment that take place (Bonadies et al., 2011).

In reflecting on the limitations of the study, this was a small group of women, in one area of Wales, who offered to take part in the study. All the probands in this study had implant only surgery. Different reconstructive techniques may have found different experiences and thus different horizons.

Furthermore, not all of the husbands of the eight probands took part. It could also be argued that these women and their families are not representative of the whole of the BRCA population due to geographical differences. A different group of BRCA women and their family at a different time may yield different experiences. Furthermore, it is accepted that a different researcher may find different horizons that connect and unite the women and their family. However, it was not the intention of this study or to qualitative inquiry, to generalise these findings to all women and their family with the BRCA mutation, in all areas. This study does, however, offer a new horizon of understanding that contributes to the body of existing knowledge, helping members of the multidisciplinary team to learn from those who experience living with a BRCA mutation and who elect to have risk-reducing mastectomy and reconstruction.

## **7.6 Concluding Statement**

Staying true to Gadamerian philosophy (Gadamer, 2004) in offering an 'understanding' of 'being' a BRCA women who elects to have RRM in the context of her family, this thesis expounds the experience of living with the consequences of bilateral risk-reducing mastectomy and the effects on gender identity, body image and sexuality for the woman and her family. In having a moral obligation to survive for the sake of self and others, women make a difficult, brave and selfless decision to elect to have risk-reducing mastectomy, but in doing so, this leads to 'disembodiment', having consequences for the whole family.

## **7.7 Recommendations and Implications for Practice**

Future research conducted in this area is proposed with women and their families who are BRCA positive but who decide against surgery and choose surveillance. Future studies could also focus on the children of those who elect to have RRM. A longitudinal study with husbands and female partners of those women with a BRCA mutation would also add a valuable contribution to knowledge in the future. With more patients attending cancer genetic clinics in the future, there may also be a need to capture the experiences of a cohort of men with a BRCA mutation.

1. It is recommended that women with the BRCA mutation and their families electing to have risk-reducing surgery have appropriate information and support both verbal and written pre and post-surgery, from a suitably trained team of clinicians and nurses as part of a multidisciplinary team.
2. Patients are given time and access to the multidisciplinary team in order to make appropriate informed decisions about risk-reducing mastectomy and reconstruction with an allocated key worker.
3. Body image, gender identity and sexuality should be discussed openly pre and post-surgery including the assessment or appropriate referral for psychosocial intervention if necessary.

4. Women with the BRCA mutation should have access to onco-plastic reconstruction and be given the opportunity to meet patient volunteers to discuss and observe the cosmetic surgical outcome. Preserving the nipple areolar should be at the forefront of the discussion.
5. Patients and their families should be signposted to support groups if required and their information needs should be individually assessed and met.
6. Future research is required exploring the experiences of women and their families who elect to have RRM using different types of flap reconstructions.

## REFERENCES

- Adams, E. et al. 2011. The experiences, needs and concerns of younger women with breast cancer: a meta-ethnography. *Psycho-Oncology*, 20 (8) 851-861.
- Adern-Jones, A. et al. 2005. Too much, too soon? Patients and health professionals' views concerning the impact of genetic testing at the time of breast cancer diagnosis in women under the age of 40. *European Journal of Cancer Care*, 14 (3) 272-281.
- Altschuler, A. et al. 2008. Positive, Negative, and Disparate—Women's Differing Long-Term Psychosocial Experiences of Bilateral or Contralateral Prophylactic Mastectomy. *The Breast Journal*, 14 (1) 25-32.
- Anderson, B.O. 2001. Prophylactic surgery to reduce breast cancer risk: a brief literature review. *The Breast Journal*, 7 (5), 321-330.
- Andrzejczak, E. et al. 2013. Partner relationships after mastectomy in women not offered breast reconstruction. *Psycho-Oncology*, 22 (7) 1653-1657.
- Annells, M. 1996. Hermeneutic phenomenology: philosophical perspectives and current use in nursing research. *Journal of Advanced Nursing*, 23 (4) 705-713.
- Annells, M. 1999. Evaluating phenomenology: Usefulness, quality and philosophical foundations. *Nurse Researcher*, 6 (3) 5-19.
- Ansquer, Y. et al. 1998. Survival in early-onset BRCA1 breast-cancer patients. *The Lancet*, 352 (9127) 541.
- Antill, Y. et al. 2006. Risk-reducing surgery in women with familial susceptibility for breast and/or ovarian cancer. *European Journal of Cancer*, 42 (5) 621-628.
- Antoniou, A. et al. 2008. The BOADICEA model of genetic susceptibility to breast and ovarian cancers: updates and extensions. *British Journal of Cancer*, 98 (8) 1457-1466.
- Antoniou, A. et al. 2003. Average risks of breast and ovarian cancer associated with BRCA1 or BRCA2 mutations detected in case series unselected for family history: a combined analysis of 22 studies. *The American Journal of Human Genetics*, 72 (5) 1117-1130.
- Arver, B. et al. 2011 Bilateral prophylactic mastectomy in Swedish women at high risk of breast cancer: a national survey. *Annals of Surgery*, 253 (6) 1147-1154.

- Ashworth, P. D. 1987. The descriptive adequacy of qualitative findings. *The Humanistic Psychologist*, 15 (1) 38.
- Ashworth, P. 2008. *Conceptual foundations of qualitative psychology*. Qualitative Psychology: A practical guide to research methods 2, pp. 4-25.
- Avis, N.E. et al. 2004. Psychosocial problems among younger women with breast cancer. *Psycho-Oncology*, 13 (5) 295-308.
- Avis, N.E. et al. 2005. Quality of life among younger women with breast cancer. *Journal of Clinical Oncology*, 23 (15) 3322-3330.
- Baillie, L. 1996. A phenomenological study of the nature of empathy. *Journal of Advanced Nursing*, 24 (6) 1300-1308.
- Bainbridge, J. 2007. A new direction for breast cancer research. *British Journal of Midwifery*, 15 (7) 437-437.
- Balmer, C.E. 2002. BRCA1 and BRCA2 genetic testing for ovarian cancer: is it all good news? *European Journal of Oncology Nursing*, 6 (3) 162-168.
- Bancroft, E.K. 2010. Genetic testing for cancer predisposition and implications for nursing practice: narrative review. *Journal of Advanced Nursing* 66 (4) 710-737.
- Barcenas, C.H. et al. 2006. Assessing BRCA carrier probabilities in extended families. *Journal of Clinical Oncology*, 24 (3) 354-360.
- Bartky, S.L. 1990. *Femininity and domination: Studies in the phenomenology of oppression*. London: Psychology Press.
- Barton, M.B. et al. 2005. Complications following bilateral prophylactic mastectomy. *Monographs National Cancer Institute*, 35, p. 61.
- Beck, C. T. 1994. Phenomenology: its use in nursing research. *International Journal of Nursing Studies*, 31 (6) 499-510.
- Beattie, M.S. et al. 2009. Uptake, time course, and predictors of risk-reducing surgeries in BRCA carriers. *Genetic Testing and Molecular Biomarkers*, 13 (1) 51-56.
- Beesley, H., Holcombe, C., Brown, S.L. and Salmon, P. 2013. Risk, worry and cosmesis in decision-making for contralateral risk-reducing mastectomy: Analysis of 60 consecutive cases in a specialist breast unit. *The Breast*, 22:179-184.



Beesley, V.L. et al. 2013. Changes in supportive care needs after first-line treatment for ovarian cancer: identifying care priorities and risk factors for future unmet needs. *Psycho-Oncology* 22 (7) 1565-1571.

Beran, T.M. et al. 2008. The trajectory of psychological impact in BRCA1/2 genetic testing: does time heal? *Annals of Behavioural Medicine*, 36 (2) 107-116.

Bermejo-Pérez, M.J. et al. 2007. Effectiveness of preventive interventions in BRCA1/2 gene mutation carriers: A systematic review. *International Journal of Cancer*, 121 (2) 225-231.

Bielanski, T.E. 2001. Does prophylactic mastectomy in women at high risk for breast cancer provide a psychological benefit? *Journal of Family Practice*, 50 (4) 366-366.

Binding, L.L. and Tapp, D.M. 2008. Human understanding in dialogue: Gadamer's recovery of the genuine. *Nursing Philosophy*, 9 (2) 121-130.

Bleiker, E.M.A. et al. 2003. Psychosocial issues in cancer genetics. *Acta Oncologica*, 42 (4) 276-286.

Bleret, V. et al. 2011. Bilateral prophylactic mastectomy for women at very high risk for breast cancer: Beyond the technique. *Mastectomie bilatérale prophylactique chez les patientes à très haut risque de cancer mammaire: Au-delà de la technique* 66 (5-6) 250-253.

Bluman, L.G. et al. 2003. Attitudes, knowledge, risk perceptions and decision-making among women with breast and/or ovarian cancer considering testing for BRCA1 and BRCA2 and their spouses. *Psycho-Oncology*, 12 (5) 410-427.

Boenink, M. 2011. Unambiguous test results or individual independence? The role of clients and families in predictive BRCA-testing in the Netherlands compared to the USA. *Social Science and Medicine*, 72 (11) 1793-1801.

Bonadies, D.C. et al. 2011. What I wish I'd known before surgery: BRCA carriers' perspectives after bilateral salpingo-oophorectomy. *Familial cancer*, 10 (1) 79-85.

Borreani, C. et al. 2014. The psychological impact of breast and ovarian cancer preventive options in BRCA1 and BRCA2 mutation carriers. *Clinical Genetics* 85 7-15

Borgen, P. I. et al. 1998. Patient regrets after bilateral prophylactic mastectomy. *Annals of Surgical Oncology*, 5 (7) 603-606.

Bradbury,A.R. et al. 2007 How often do BRCA mutation carriers tell their young children of the family's risk for cancer? A study of parental disclosure of BRCA mutations to minors and young adults. *Journal of Clinical Oncology* 25 (24), 3705-3711

Bradbury-Jones, C. et al. 2010. Phenomenology and participant feedback: convention or contention? *Nurse Researcher*, 17 (2) 25-33.

Braithwaite, et al. 2005. Development of a risk assessment tool for women with a family history of breast cancer. *Cancer Detection and prevention*29(5) 433-439

Brandberg, Y. et al. 2004. Preoperative psychological reactions and quality of life among women with an increased risk of breast cancer who are considering a prophylactic mastectomy. *European Journal of Cancer*, 40 (3) 365-374.

Brandberg, Y. et al. 2008. Psychological reactions, quality of life, and body image after bilateral prophylactic mastectomy in women at high risk for breast cancer: a prospective 1-year follow-up study. *Journal of Clinical Oncology*, 26 (24) 3943.

Brandberg, Y. et al. 2012. Less correspondence between expectations before and cosmetic results after risk-reducing mastectomy in women who are mutation carriers: A prospective study. *European Journal of Surgical Oncology*, 38 (1) 38-43.

Brawarsky, P. et al. 2012. Use of Annual Mammography Among Older Women with Ductal Carcinoma In Situ. *Journal of General Internal Medicine*, 27 (5) 500-505.

Breast Cancer Care 2016 - <https://www.breastcancercare.org.uk/information-support/facing-breast-cancer/diagnosed-breast-cancer/primary-breast-cancer/triple-negative-breast-cancer> [Accessed September 2016]

Bresser, P.J.C. et al. 2006. Satisfaction with prophylactic mastectomy and breast reconstruction in genetically predisposed women. *Plastic and Reconstructive Surgery*, 117 (6) 1675.

Bresser, P.J.C. et al. 2007a. The course of distress in women at increased risk of breast and ovarian cancer due to an (identified) genetic susceptibility who opt for prophylactic mastectomy and/or salpingo-oophorectomy. *European Journal of Cancer*, 43 (1) 95-103.

Bresser, P.J.C. et al. 2007b. Who is prone to high levels of distress after prophylactic mastectomy and/or salpingo-ovariectomy? *Annals of oncology*, 18 (10) 1641.

Brinkmann, S. and Kvale, S. 2015. *Interviews: learning the craft of qualitative research interviewing*. Third edition. London: Sage Publications.

Brown, C. and Seddon, J. 1996. The social body and the biomechanical body: can they coexist in nurse education? *Journal of Advanced Nursing*, 23 (4) 651-656.

Brunstrom, K. et al. 2016. Experiences of women who underwent predictive BRCA1/2 mutation testing before the age of 30. *Journal of Genetic*, 25, 90-100.

Bryman, A. 2012. *Social Research Methods*. Oxford: Oxford University Press.

Bulpitt, H. and Martin, P.J. 2010. Who am I and what am I doing? Becoming a qualitative research interviewer: Helen Bulpitt and Peter J Martin discuss using reflexion to make research processes in studies transparent. *Nurse Researcher* 17 (3) 7-16.

Burke, E.E. et al. 2015. Prophylactic mastectomy: who needs it, when and why. *Journal of Surgical Oncology*, 111 (1) 91-95.

Burke, W. et al. 1997. Recommendations for follow-up care of individuals with an inherited predisposition to cancer: II. BRCA1 and BRCA2. *JAMA*, 277 (12) 997-1003.

Bury, M.R.1986. Social constructionism and the development of medical Sociology *Sociology of Health and Illness*, 8:137-169.

Bury, M.R.2008. Chronic illness as disruption. *Sociology of health and illness* June 28<sup>th</sup> <http://dx.doi.org/10.1111/1467-9566.ep11339939>. [Accessed September 2016]

Butler, J. 2004. *Undoing gender*. Psychology Press.

Butow, P. et al. 2005. Psychological Morbidity in Women at Increased Risk of Developing Breast Cancer: A Controlled Study. *Psycho-Oncology* ,14 (3) 196-203.

Butow, P.N. and Lobb, E.A. 2004. Analyzing the Process and Content of Genetic Counselling in Familial Breast Cancer Consultations. *Journal of Genetic Counseling*, 13 (5) 403-424.

Byrne, M. 2001. Hermeneutics as a method for textual analysis AORN journal 73 (5) 968-970.

Caelli, K. 2000. The changing face of phenomenological research: Traditional and American phenomenology in nursing. *Qualitative Health Research*, 10 (3) 366-377.

Calderon-Margalit, R. and Paltiel, O. 2004. Prevention of breast cancer in women who carry BRCA1 or BRCA2 mutations: A critical review of the literature. *International Journal of Cancer*, 112 (3) 357-364.

Cancer Research UK 2013. Let's Beat cancer Sooner [Online] <http://www.cancerresearchuk.org/health-professional/cancer-statistics/statistics-by-cancer-type/breast-cancer> [Accessed 8.9.2016]

Chapman, E. 2002. Perceptions of the body and genetic risk. In: Bainham, A. ed. *Body Lore and Laws: Essays on Law and the Human Body*. Oxford: Hart Publishing, p. 309.

Chapman, J.S. et al. 2015. Managing symptoms and maximizing quality of life after preventive interventions for cancer risk reduction. *Current Opinion in Obstetrics and Gynaecology*, 27 (1) 40-44.

Charalambous, A. and Beadsmoore, A. 2008. Ricoeur's hermeneutic phenomenology: an implication for nursing research. *Scandinavian Journal of Caring Sciences* 22 (4) 637-642.

Charmaz, K. 1995. The body, identity, and self. *The Sociological Quarterly* 36 (4) 657-680.

Chen, S. and Parmigiani, G. 2007. Meta-analysis of BRCA1 and BRCA2 penetrance. *Journal of Clinical Oncology*, 25 (11)1329-1333.

Cherry, M.C. et al. 2013. Understanding the Needs of Women Considering Risk-Reducing Salpingo-Oophorectomy. *Cancer Nursing*, 36 (3) E33.

Christopoulou, A. and Spiliotis, J. 2006. The role of BRCA1 and BRCA2 in hereditary breast cancer. *Gene Therapy and Molecular Biology*, 10 (1) 95-99.

Claes, E. et al. 2003. Communication with close and distant relatives in the context of genetic testing for hereditary breast and ovarian cancer in cancer patients. *American Journal of Medical Genetics, Part A* 116 (1) 11-19.

Claes, E. et al. 2005. Surveillance behaviour and prophylactic surgery after predictive testing for hereditary breast/ovarian cancer. *Behavioural Medicine*, 31 (3) 93-105.

Cohen, M. et al. 2000 *Hermeneutic phenomenological research: A practical guide for nurse researchers* London: Sage publication.

Cohen, M.Z. and Omery, A. 1994. Schools of phenomenology. Implications for research IN *Critical issue in qualitative research methods*. J. M. Morse. London: Sage.

Cohen, M. et al. 2000. Hermeneutic Phenomenological Research: *A practical guide for Nurse Researchers*. London: Sage publications.

Colaizzi, P. 1978 Psychological research as the phenomenologist views it. IN Valle and King (eds) *Existential-Phenomenological alternatives for psychology* pp48-71. New York: Oxford University Press

Cooper, S. Endacott, R. 2007 Generic qualitative research; a design for qualitative research in emergency care? *Emergency Medical Journal*, 24, 817-819.

Crabb, S and LeCouteur, A. 2006. 'Fiona Farewells Her Breasts' A popular magazine account of breast cancer prevention *Critical Public Health* 16 (1) 5-18

Creswell, J.W. 2014. *Research Design: Qualitative, Quantitative and Mixed Methods Approaches, 4<sup>th</sup> edition*. London: Sage.

Crist, J.D. and Tanner, C.A. 2003. Interpretation/analysis methods in hermeneutic interpretive phenomenology. *Nursing Research*, 52 (3) 202-205.

Cromptoets, S. 2003. Reconstructing the self: Breast cancer and the post-surgical body. *Health Sociology Review*, 12 (2) 137-145.

Crotser, C.B. and Boehmke, M. 2009. Survivorship considerations in adults with hereditary breast and ovarian cancer syndrome: state of the science. *Journal of Cancer Survivorship*, 3 (1) 21-42.

Crotser, C.B. and Dickenson, S, S. 2010 Women Receiving News of a Family BRCA1/2 Mutation: Messages of fear and empowerment. *Journal of nursing scholarship* 42 (4) 367-378

Crotty, M. 1996. *Phenomenology and nursing research*. South Melbourne, Vic.: South Melbourne, Vic. : Churchill Livingstone.

Crotty, M. 1998 *The foundations of social research. Meaning and perspective in the research process*. London: Sage.

Croyle, R.T. et al. 1997. Psychological responses to BRCA1 mutation testing: preliminary findings. *Health Psychology*, 16 (1) 63.

Culver, J.O. et al. 2011. Development and evaluation of a decision Aid for BRCA carriers with breast cancer. *Journal of Genetic Counseling*, 20 (3) 294-307.

Curtis, S. et al. 2000. Approaches to sampling and case selection in qualitative research: examples in the geography of health. *Social Science and Medicine* 50 (7) 1001-1014.

Cypowyj, C. et al. 2009. Subjective interpretation of inconclusive BRCA1/2 cancer genetic test results and transmission of information to the relatives. *Psycho-Oncology*, 18 (2) 209-215.

d'Agincourt-Canning, L. 2001. Experiences of genetic risk: disclosure and the gendering of responsibility. *Bioethics*, 15 (3) 231-247.

d'Agincourt-Canning, L. 2006a. Genetic testing for hereditary breast and ovarian cancer: Responsibility and choice. *Qualitative Health Research*, 16 (1) 97-118.

d'Agincourt-Canning, L. 2006b. A gift or a yoke? Women's and Men's responses to genetic information from BRCA1 and BRCA2 testing. *Clinical Genetics*, 70, 462-472.

Dagan, E. and Goldblatt, H. 2009. The twilight zone between health and sickness: A qualitative exploration with asymptomatic BRCA1 and 2 mutation carriers. *Women and Health*, 49 (4) 263-279.

Dahlberg, K. et al. 2008. Reflective lifeworld research (2<sup>nd</sup> edition) Hungary: Studentlitteratur.

Daly, P.A. et al. 2003. Predictive testing for BRCA1 and 2 mutations: a male contribution. *Annals of Oncology*, 14, 549-553.

Daly, M.B. 2009. The impact of social roles on the experience of men in BRCA1/2 families: Implications for counselling. *Journal of Genetic Counseling*, 18, 42-48.

David, M. and Sutton, C.D. 2011. *Social research: An introduction*. London: Sage.

De Bock, G. et al. 2013. Which screening strategy should be offered to women with BRCA1 or BRCA2 mutations? A simulation of comparative cost-effectiveness. *British Journal of Cancer*, 108 (8)1579-1586.

de Felice, F. et al. 2015. Bilateral Risk-Reduction Mastectomy in BRCA1 and BRCA2 Mutation Carriers: A Meta-analysis. *Annals of Surgical Oncology*, 22 (9) 2807-2809.

de la Peña-Salcedo, J.A. et al. 2011. Prophylactic Mastectomy: Is It Worth It? *Aesthetic Plastic Surgery*, 1-9.

de Leeuw, J. et al. 2008. Predictors of choosing life-long screening or prophylactic surgery in women at high and moderate risk for breast and ovarian cancer. *Familial cancer*, 7 (4) 347-359.

Denford, S. et al. 2011. Understanding normality: a qualitative analysis of breast cancer patients' concepts of normality after mastectomy and reconstructive surgery. *Psycho-Oncology* 20 (5) 553-558.

den Heijer, M. et al. 2012. Body image and psychological distress after prophylactic mastectomy and breast reconstruction in genetically predisposed women: a prospective long-term follow-up study. *European Journal of Cancer*, 48 (9) 1263-1268.

den Heijer, M. et al. 2013 International Variation in Physicians' attitudes towards prophylactic mastectomy-comparison between France, Germany, the Netherlands and the United Kingdom. *European Journal of Cancer* 49 (13) 2798-2805.

Denzin, N.K. 2009. *Qualitative inquiry under fire: toward a new paradigm dialogue*. Walnut Creek, California: Left Coast Press.

Denzin, N.K. and Lincoln, Y.S. 2000 Introduction: The discipline and practice of qualitative research in Denzin, N.K. and Lincoln, Y.S. (Eds), *Handbook of Qualitative Research* (2<sup>nd</sup> edition) Thousand Oaks, CA: Sage.

Denzin, N.K. and Lincoln, Y.S. 2011. *The SAGE handbook of qualitative research*. London: Sage.

Department of Health 2015. Policy paper. Government Policy; cancer research and treatment. <https://www.gov.uk/government/publications/2010-to-2015-government-policy-cancer-research-and-treatment/2010-to-2015-government-policy-cancer-research-and-treatment>. [Accessed September 2016]

de Souza, A.N. et al. 2014. Cultural aspects of healthy BRCA carriers from two ethnocultural groups. *Qualitative Health Research*, 1-17.

de Witt, L. and Ploeg, J. 2006. Critical appraisal of rigour in interpretive phenomenological nursing research. *Journal of Advanced Nursing*, 55 (2) 215-229.

DiCicco-Bloom, B. and Crabtree, B.F. (2006) Making Sense of Qualitative Research. *Medical Education*, 40, 314-321.

DiMillo, J. et al. 2013. Living with the BRCA genetic mutation: An uncertain conclusion to an unending process. *Psychology, Health and Medicine*, 18 (2)125-134.

Dingwall, R.1997. Accounts, interviews and observations IN *Context and Method in Qualitative Research* (Miller and Dingwall eds) 51-65. London: Sage publications.

DiProspero, L. et al. 2001. Psychosocial issues following a positive result of genetic testing for BRCA1 and BRCA2 mutations: findings from a focus group and a needs assessment survey. *CMAJ*, 164 (7) 1005-1009.

Domchek, S.M. et al. 2010. Association of risk-reducing surgery in BRCA1 or BRCA2 mutation Carriers with Cancer Risk and Mortality. *JAMA* 304 (9) 967-975.

Donchin, A. 2000. Autonomy and interdependence: quandaries in genetic decision-making.

Dorval, M. et al. 2000. Anticipated versus actual emotional reactions to disclosure of results of genetic tests for cancer susceptibility: findings from p53 and BRCA1 testing programs. *Journal of Clinical Oncology* 18(10), pp. 2135-2142.

Douglas, M. 1996. *Natural Symbols*, London: Routledge.

Douglas, H, A. et al. 2009 The effect of BRCA Gene; Testing on Family Relationships: A Thematic analysis of Qualitative Interviews *Journal of Genetic Counseling* 18 (5) 418-435

Dowdy, S.C. et al. 2004. Surgical risk reduction: prophylactic salpingo-oophorectomy and prophylactic mastectomy. *American Journal of Obstetrics and Gynaecology*, 191 (4) 1113-1123.

Dowling, M. 2004. Hermeneutics: an exploration *Nurse Researcher* 11 (4) 30-41.

Dowling, M. 2007. From Husserl to van Manen. A review of different phenomenological approaches. *International Journal of Nursing Studies*, 44 (1) 131-142.

Dreyfus, H.L. 1996. The current relevance of Merleau-Ponty's phenomenology of embodiment. *The Electronic Journal of Analytic Philosophy*, 4, 1-16.

Dubriwny, T.N. 2012. *The vulnerable empowered woman: Feminism, post feminism, and women's health*. Rutgers University Press.



- Dudok de Wit, A. et al. 1997. BRCA1 in the family: a case description of the psychological implications. *American Journal of Medical Genetics*, 71 (1) 63-71.
- Duncan, C. et al. 2007. Concept analysis: the importance of differentiating the oncological focus. *Journal of Advanced Nursing*, 58 (3) 293-300.
- Dupras, C. and Ravitsky, V. 2013. Disclosing Genetic Information to Family Members: The Role of Empirical Ethics. *eLS*, pp. 1-6.
- Durant-Law, G. 2005. Soft systems methodology and grounded theory combined: a knowledge management research approach. *Online Journal of Knowledge Management*, 2 (1) 13-23.
- Dworkin, A. 1974. *Woman Hating*. New York: In E.P. Dutton. 1979. *Pornography: Men Possessing Women*. New York: Plume.
- Easton, D.F. et al. 1995. Breast and ovarian cancer incidence in BRCA1 - mutation carriers. Breast Cancer Linkage Consortium. *American journal of human genetics*, 56 (1) 265.
- Eeles, R. et al. 1996. Prophylactic mastectomy for genetic predisposition to breast cancer: the proband's story. *Clinical Oncology*, 8 (4) 222-225.
- Eicher, et al. 2006. A nurse is a nurse? A systematic review of the effectiveness of specialist nursing in breast cancer. *European Journal of Cancer*, 42, 3117-3126.
- Eisen, A. and Weber, B.L. 1999. Prophylactic mastectomy—the price of fear. *New England Journal of Medicine*, 340 (2) 137-138.
- Eisen, A. and Weber, B.L. 2001. Prophylactic mastectomy for women with BRCA1 and BRCA2 mutations—facts and controversy. *New England Journal of Medicine*, 345 (3) 207-208.
- Elder, E.E. et al. 2005. Quality of life and patient satisfaction in breast cancer patients after immediate breast reconstruction: a prospective study. *The Breast*, 14 (3) 201-208.
- Emden, C. and Sandelowski, M 1999. The good, the bad and the relative, part two; goodness and the criterion problem in qualitative research. *International Journal of Nursing Practice*, (5) 2-7.
- Espenschied, C.R. et al. 2012. Closing the loop: action research in a multimodal hereditary cancer patient conference is an effective tool to assess and address patient needs. *Journal of Cancer Education*, 27 (3) 467-477.

Esplen, M. 2002. Pilot experience in women concerned by genetic screening of breast and ovarian cancer (BRCA1 et BRCA2 ) with supportive-expressive group therapy. *Revue Francophone de Psycho-Oncologie* 1 (3-4), 54-60.

Esplen, M. J. et al. 2009. The BRCA Self-Concept Scale: a new instrument to measure self-concept in BRCA1/2 mutation carriers. *Psycho-Oncology* 18 (11) 1216-1229.

Etchegary, H. et al. 2009. Decision-making about inherited cancer risk: exploring dimensions of genetic responsibility. *Journal of Genetic Counseling* 18 (3) 252-264.

Euhus, D.M. 2015. Risk-Reducing Mastectomy for BRCA Gene Mutation Carriers. *Annals of Surgical Oncology* 22 (9) 2807-2809.

Evans, D. et al. 1993. Perception of risk in women with a family history of breast cancer. *British Journal of Cancer*, 67 (3) 612.

Evans, D. et al. 1994. The impact of genetic counselling on risk perception in women with a family history of breast cancer. *British Journal of Cancer* 70 (5) 934.

Evans, D. et al. 2014. The Angelina Jolie effect: how high celebrity profile can have a major impact on provision of cancer related services. *Breast Cancer Research*, 16 (5) 442.

Evans, D.G. et al. 2013. Familial breast cancer: summary of updated NICE guidance. *British Medical Journal*, 346, p.3829.

Evans, D.G.R. et al. 1999. Utilisation of prophylactic mastectomy in 10 European Centres. *Disease Markers*, 15 (1-3), 148-151.

Evans, D.G.R. et al. 2005. Surgical decisions made by 158 women with hereditary breast cancer aged < 50 years. *European Journal of Surgical Oncology* 31(10) 1112-1118.

Evans, D.G.R. et al. 2009a. Risk-reducing mastectomy: Outcomes in 10 European centres. *Journal of Medical Genetics* 46 (4) 254-258.

Evans, D.G.R. et al. 2009b. Uptake of risk-reducing surgery in unaffected women at high risk of breast and ovarian cancer is risk, age, and time dependent. *Cancer Epidemiology Biomarkers and Prevention*, 18 (8) 2318-2324.

Evans, G., Wisley, J., Clancy, T. 2015. Longer term effects of the Angelina Jolie effect: Increased risk-reducing mastectomy rates in BRCA carriers and other high-risk women. *Breast Cancer Research*, 17:143.

Everson, C.L. 2009. *Reproductive decision-making in young women breast cancer survivors*. M.A. Thesis; Oregon State University.

Fallowfield, L. and Jenkins, V. 2004. Communicating sad, bad, and difficult news in medicine. *The Lancet*, 363 (9405) 312-319.

Fang, S.Y. et al. 2013. The effect of breast reconstruction surgery on body image among women after mastectomy: a meta-analysis. *Breast Cancer Research and Treatment*, 137 (1) 13-21.

Fanos, J.H. and Johnson, J.P. 1995. Barriers to carrier testing for adult cystic fibrosis sibs: The importance of not knowing. *American Journal of Medical Genetics*, 59 (1) 85-91.

Featherstone K, et al. 2006. *Risky Relations, Family Kinship and the Newer Genetics*. New York: Oxford International Publishers.

Fehniger, J. et al. 2013. Family communication of BRCA1/2 results and family uptake of BRCA1/2 testing in a diverse population of BRCA1/2 carriers. *Journal of Genetic Counseling*, 22 (5) 603-612.

Finch, A. et al. 2013 The impact of prophylactic Salpingo-oophorectomy on quality of life and psychological distress in women with a BRCA mutation. *Psych-Oncology* 22,212-219

Finlay, L. 2006. IN: Finlay, L. and Ballinger, C. 2006 (Eds) *Qualitative Research for Allied Health professionals*. Chichester: John Wiley and Sons Ltd.

Finlay, L. 2006. The body's disclosure in phenomenological research. *Qualitative Research in Psychology*, 3 (1):19-30.

Finlay, L. 2012. Unfolding the phenomenological Research process: Iterative Stages of 'Seeing Afresh' *Journal of Humanistic Psychology*, 53 (2) 172-201.

Firth, C. et al. 2011. Novel one-stop multidisciplinary follow-up clinic for BRCA1/2 carriers: patient satisfaction and decision making. *Psycho-Oncology* 20 (12) 1301-1308.

Fitzpatrick, M. 2000. *The tyranny of health: doctors and the regulation of lifestyle*. London: Routledge.

Fleming, V. et al. 2003. Hermeneutic research in nursing: developing a Gadamerian-based research method. *Nursing Inquiry*, 10 (2) 113-120.

Flick, U. ed. 2014. *The SAGE handbook of qualitative data analysis*. London: SAGE.

- Fobair, P. et al. 2006. Body image and sexual problems in young women with breast cancer. *Psycho-Oncology*, 15, 579-594.
- Folli, S. et al. 2012. Improved sub-areolar breast tissue removal in nipple-sparing mastectomy using hydro dissection. *The Breast*, 21 (2) 190-193.
- Fontana, A. and Frey, J.H. 2000. The interview: From structured questions to negotiated text. *Handbook of Qualitative Research*, 2 (6) 645-672.
- Ford, D. et al. 1998. Genetic heterogeneity and penetrance analysis of the BRCA1 and BRCA2 genes in breast cancer families. *The American Journal of Human Genetics*, 62 (3) 676-689.
- Forrest, K. et al. 2003. To tell or not to tell: barriers and facilitators in family communication about genetic risk. *Clinical Genetics*, 64 (4) 317-326.
- Foster, C. et al. 2002. Genetic testing for breast and ovarian cancer predisposition: cancer burden and responsibility. *Journal of Health Psychology*, 7 (4) 469-484.
- Foster, C. et al. 2004. Juggling roles and expectations: dilemmas faced by women talking to relatives about cancer and genetic testing. *Psychology and Health* 19 (4) 439-455.
- Foster, C. et al. 2007. Predictive genetic testing for BRCA1/2 in a UK clinical cohort: three-year follow-up. *British Journal of Cancer*, 96 (5) 718-724.
- Foucault, M. 1979. *Discipline and punishment*. Harmondsworth: Peregrine.
- Foulkes, W. et al. 2000. Primary node negative breast cancer in BRCA1 mutation carriers has a poor outcome. *Annals of Oncology*, 11 (3) 307-313.
- Fox, N.J. 2002. Refracting 'health': Deleuze, Guattari and body-self. *Health: Interdisciplinary Journal for the Social Study of Health Illness and Medicine*, 6 (3) 347-363.
- Fox, N.J. 2012. *The Body: Key Themes in Health and Social Care*. USA: Polity.
- Frank, T.S. and Critchfield, G.C. 2001. Identifying and managing hereditary risk of breast and ovarian cancer. *Clinics in Perinatology* 28 (2) 395-406.
- Friebel, T.M. et al. 2007. Bilateral prophylactic oophorectomy and bilateral prophylactic mastectomy in a prospective cohort of unaffected BRCA1 and BRCA2 mutation carriers. *Clinical Breast Cancer*, 7 (11) 875-882.

- Frost, C.J. et al. 2004. Decision Making with Uncertain Information: Learning From Women in a High Risk Breast Cancer Clinic. *Journal of Genetic Counseling*, 13 (3) 221-236.
- Frost, M.H. et al. 2000. Long-term satisfaction and psychological and social function following bilateral prophylactic mastectomy. *JAMA*, 284 (3) 319-324.
- Frost, M.H. et al. 2005. Satisfaction after contralateral prophylactic mastectomy: the significance of mastectomy type, reconstructive complications, and body appearance. *Journal of Clinical Oncology*, 23 (31) 7849.
- Fuller, S. and Anderson, R.C. 2006. Adjustment issues related to bilateral prophylactic mastectomy in women at elevated risk of developing breast cancer. *Plastic Surgical Nursing*, 26 (2) 60-65.
- Gadamer, H.G. 1997. *Truth and Method*, (2<sup>nd</sup> Revised edition) (J. Weinsheimer and D.G. Marshall, Trans. Rev.) New York: Continuum Publishing Group.
- Gadamer, H.G. 2004. *Truth and Method*, (2<sup>nd</sup> Revised edition, *translation revised by Joel Weinsheimer and Donald G. Marshall*) London, New York: Continuum Publishing Group.
- Gahm, J. et al. 2010. Bilateral prophylactic mastectomy in women with inherited risk of breast cancer—prevalence of pain and discomfort, impact on sexuality, quality of life and feelings of regret two years after surgery. *The Breast*, 19 (6) 462-469.
- Geiger, A.M. et al. 2007. Quality of life after bilateral prophylactic mastectomy. *Annals of Surgical Oncology*, 14 (2) 686-694.
- Geirdal, A.Ø. and Dahl, A.A. 2008. The relationship between coping strategies and anxiety in women from families with familial breast–ovarian cancer in the absence of demonstrated mutations. *Psycho-Oncology*, 17 (1) 49-57.
- Gifford, S.M. 1986. The meaning of lumps: a case study of the ambiguities of risk. *Anthropology and epidemiology*. Springer, 213-246.
- Giorgi, A. 1989. One type of analysis of descriptive data: procedures involved in following a phenomenological method. *Methods*, 1, 39-61.
- Giorgi, A. 2006. Concerning variations in the application of the phenomenological method. *The Humanistic Psychologist*, 34, 305-309.

Glasspool, D.W. et al. 2010. Interactive decision support for risk management: a qualitative evaluation in cancer genetic counselling sessions. *Journal of Cancer Education*, 25 (3) 312-316.

Glasse, R. et al, 2016 Decision making, psychological wellbeing and psychological outcomes for high risk women who choose to undergo bilateral prophylactic mastectomy-A review of the literature. *The Breast* 28 130-135

Goffman, E. 1990. *The presentation of self in everyday life*. London: Penguin.

Gold, R. et al. 2010 Myriad Genetics; In the eye of the policy storm. *Genetics in Medicine*, 12 (4).

Gooding, H.C. et al. 2006. Genetic susceptibility testing from a stress and coping perspective. *Social Science and Medicine*, 62 (8)1880-1890.

Gopie, J.P. et al. 2013. Body image issues after bilateral prophylactic mastectomy with breast reconstruction in healthy women at risk for hereditary breast cancer. *Familial Cancer*, 12 (3) 479-487.

Grann, V.R. et al. 1998. Decision analysis of prophylactic mastectomy and oophorectomy in BRCA1-positive or BRCA2-positive patients. *Journal of Clinical Oncology* 16 (3) 979-985.

Grann, V.R. et al. 1999. The quality of life associated with prophylactic treatments for women with BRCA1/2 mutations. *Cancer Journal from Scientific American*, 5 (5) 283-292.

Grassley, J.S. and Nelms, T.P. 2008. Understanding maternal breastfeeding confidence: A Gadamerian hermeneutic analysis of women's stories. *Healthcare for Women International*, 29 (8-9) 841-862.

Graves, K.D. et al. 2012. Long-Term Psychosocial Outcomes of BRCA1/BRCA2 Testing: Differences across Affected Status and Risk-Reducing Surgery Choice. *Cancer Epidemiology Biomarkers and Prevention*, 21 (3).

Greatrex-White, S. 2008. Thinking about the nature of research findings: A hermeneutic phenomenological perspective. *International Journal of Nursing Studies*, 45 (12) 1842-1849.

Green, J. and Thorogood, N. 2013. *Qualitative Methods for Health Research*. 3rd edition. London: Sage.

Gregory, M. et al. 2010. *Timescapes*. IN Family communication about genetics: Theory and Practice. Gaff, C (eds), Oxford University Press.

- Grondin, J. and Plant, K. 2014. *The philosophy of Gadamer*. Routledge.
- Gronwald, J. et al. 2007. A survey of preventive measures among BRCA1 mutation carriers from Poland. *Clinical Genetics*, 71 (2) 153-157.
- Guba, E.G. and Lincoln, Y.S. 1998. Competing paradigms in qualitative research IN Denzin, N.K. and Lincoln, Y.S. (Eds), *The landscape of qualitative research; Theories and Issues*. Thousand Oaks: CA: Sage.
- Guba, E.G. and Lincoln, Y.S. 2005. Paradigmatic controversies, contradictions, and emerging confluences. In: Denzin, N. *The SAGE Handbook of Qualitative Research*. 3<sup>rd</sup> Edition. California. Sage.
- Gubrium, J.F. and Holstein, J.A. 1997. *The new language of qualitative method*. Oxford University Press on Demand.
- Hagen, A.I. et al. 2014. Risk-reducing mastectomy, breast reconstruction and patient satisfaction in Norwegian BRCA1/2 mutation carriers. *The Breast*, 23 (1) 38-43.
- Hall, S.E. and Holman, C.D.J. 2003. Inequalities in breast cancer reconstructive surgery according to social and locational status in Western Australia. *European Journal of Surgical Oncology, (EJSO)* 29 (6) 519-525.
- Hallowell, N. 1998. 'You don't want to lose your ovaries because you think 'I might become a man'. Women's perceptions of prophylactic surgery as a cancer risk management option. *Psycho-Oncology* 7 (3) 263-275.
- Hallowell, N. 1999. Doing the right thing: genetic risk and responsibility. *Sociology of Health and Illness*, 21 (5) 597-621.
- Hallowell, N. 2000a. A qualitative study of the information needs of high-risk women undergoing prophylactic oophorectomy. *Psycho-Oncology*, 9 (6) 486-495.
- Hallowell, N. 2000b. 'Reconstructing the body or reconstructing the woman? Problems of prophylactic mastectomy for hereditary breast cancer risk' in L. K. Potts (ed) *Ideologies of breast cancer: feminist perspectives* London: Macmillan Press: 153–180.
- Hallowell, N. 2001. Surveillance or surgery? A description of the factors that influence high risk premenopausal women's decisions about prophylactic oophorectomy. *Journal of Medical Genetics*, 38, 683-691.
- Hallowell, N. 2003. Ethics and evidence eLS. [Online]. Available at:<http://onlinelibrary.wiley.com/doi/10.1002/9780470015902.a0005959/full>

Hallowell, N. 2009. Consent to genetic testing: a family affair? IN Corrigan, O (eds) *The Limits of Consent: A Socio-ethical Approach to Human Subject Research in Medicine*, p. 185-197.

Hallowell, N. et al. 2003. Balancing autonomy and responsibility: the ethics of generating and disclosing genetic information. *Journal of Medical Ethics* 29 (2) 74-79.

Hallowell, N. et al. 2004. Accommodating risk: responses to BRCA1/2 genetic testing of women who have had cancer. *Social Science and Medicine*, 59 (3) 553-565.

Hallowell, N. et al. 2005a. Men's decision-making about predictive BRCA1/2 testing: the role of family. *Journal of Genetic Counseling*, 14 (3) 207-217.

Hallowell, N. et al. 2005b. Communication about genetic testing in families of male BRCA1/2 carriers and non-carriers: Patterns, priorities and problems. *Clinical genetics*, 67 (6) 492-502.

Hallowell, N. et al. 2006. Guilt, blame and responsibility: men's understanding of their role in the transmission of BRCA1/2 mutations within their family. *Sociology of Health and Illness*, 28 (7) 969-988.

Hallowell, N. et al. 2012. Looking different, feeling different: women's reactions to risk-reducing breast and ovarian surgery. *Familial Cancer*, 11 (2).

Hallowell, N, et al. 2013. The responses of research participants and their next of kin to receiving feedback of genetic test results following participation in the Australian Ovarian Cancer Study. *Genetics in Medicine*, 15 (6) 458-465.

Hallowell, N. and Lawton, J. 2002. Negotiating present and future selves: managing the risk of hereditary ovarian cancer by prophylactic surgery. *Health*, 6 (4) 423-443.

Hamilton, J G, et al. 2009 Emotional distress following genetic testing for breast and ovarian Cancer: A meta –analytic review. *Health Psychology* 28 510-518

Hamilton, R. 2012. Original Research: Being Young, Female, and BRCA Positive. *The American Journal of Nursing*, 112 (10) 26-31.

Hamilton, R. et al. 2009. Life trajectories, genetic testing, and risk reduction decisions in 18–39 year old women at risk for hereditary breast and ovarian cancer. *Journal of Genetic Counseling*, 18 (2) 147-159.

Hamilton, R. and Hurley, K.E. 2010. Conditions and consequences of a BRCA mutation in young, single women of childbearing age. *Oncology Nursing Forum*, 37 (5) 627-634.



Hampel, H. et al. 2014. A practice guideline from the American College of Medical Genetics and Genomics and the National Society of Genetic Counsellors: referral indications for cancer predisposition assessment. *Genetics in Medicine*, 17 (1) 70-87.

Harcourt, D.M. 2004. Mastectomy patients' decision making for or against immediate breast reconstruction. *Psycho-Oncology*. 13 (20) 106-115.

Harcourt, D.M. et al. 2003. The psychological effect of mastectomy with or without breast reconstruction: a prospective, multicenter study. *Plastic and Reconstructive Surgery*, 111 (3) 1060-1068.

Harcourt, D.M. and Rumsey, N. 2001. Psychological aspects of breast reconstruction: a review of the literature. *Journal of Advanced Nursing*, 35 (4) 477-487.

Harcourt, D.M. and Rumsey, N. 2004. Mastectomy patients' decision-making for or against immediate breast reconstruction. *Psycho-Oncology*, 13 (2)106-115.

Haroun, I. et al. 2011. Reasons for risk-reducing mastectomy versus MRI-screening in a cohort of women at high hereditary risk of breast cancer. *The Breast*, 20 (3) 254-258.

Hatcher, B and Fallowfield, L. 2003. A qualitative study looking at the psychosocial implications of bilateral prophylactic mastectomy. *The Breast*, 12 (1) 1-9.

Hartman, A.R. 2005. The problems with risk selection; Scientific and psychosocial aspects. In: Senn, H.J. and Morant, R. eds. *Tumor Prevention and Genetics Iii*. Vol. 166,125-144.

Hartmann, L.C. et al. 2004. Prophylactic mastectomy for BRCA1/2 carriers: Progress and more questions. *Journal of Clinical Oncology*, 22 (6) 981-983.

Hartmann, L.C. et al. 1999. Efficacy of bilateral prophylactic mastectomy in women with a family history of breast cancer. *New England Journal of Medicine*, 340 (2) 77-84.

Hatcher, M.B. et al., 2001. The psychosocial impact of bilateral prophylactic mastectomy: prospective study using questionnaires and semi-structured interviews. *British Medical Journal*, 322 (7278)76.

Hatcher, M.B. and Fallowfield, L. 2003. A qualitative study looking at the psychosocial implications of bilateral prophylactic mastectomy\* 1. *The Breast*, 12 (1) 1-9.

Hawkins, Y. et al. 2009. Changes in sexuality and intimacy after the diagnosis and treatment of cancer: the experience of partners in a sexual relationship with a person with cancer. *Cancer Nursing*, 32 (4) 271-280.

Heemskerk-Gerritsen, B.A. et al. 2007. Prophylactic mastectomy in BRCA1/2 mutation carriers and women at risk of hereditary breast cancer: long-term experiences at the Rotterdam Family Cancer Clinic. *Annals of Surgical Oncology*, 14 (12) 3335-3344.

Heemskerk-Gerritsen, A. et al. 2012. Efficacy of Bilateral Risk-reducing Mastectomy on Primary Breast Cancer Risk in Healthy BRCA1 and BRCA2 Mutation Carriers. *European Journal of Cancer*, 48, p.s75.

Heemskerk-Gerritsen, B. et al. 2013. Substantial breast cancer risk reduction and potential survival benefit after bilateral mastectomy when compared with surveillance in healthy BRCA1 and BRCA2 mutation carriers: a prospective analysis. *Annals of Oncology*, p. mdt134.

Heidegger, M. et al. 2010. *Being and time*. New York: SUNY Press.

Heiniger, L. et al. 2015. Intuition versus cognition: a qualitative exploration of how women understand and manage their increased breast cancer risk. *Journal of Behavioural Medicine*, 1-13.

Heshka, J.T. et al. 2008 A systematic review of perceived risks, psychological and behavioural impacts of genetic testing. *Genetics in Medicine*, 10,19-32.

Hesser-Biber, S., An, C. 2016. Genetic Testing and Post-Testing Decision Making among BRCA-Positive Mutation Women: A Psychosocial Approach *Journal of Genetic Counseling*, 25 (5) 978-992.

Holland, F. et al. 2014. Younger women's experiences of deciding against delayed breast reconstruction post-mastectomy following breast cancer: An interpretative phenomenological analysis. *Journal of Health Psychology*, 1-12.

Holloway, I. and Wheeler S. 2010. *Qualitative research in Nursing and Healthcare*. 3<sup>rd</sup> edition. Chichester: Blackwell.

Hooker, G.W. et al. 2011. Longitudinal changes in patient distress following interactive decision aid use among BRCA1/2 carriers: A randomized trial. *Medical Decision Making*, 31 (3) 412-421.

Hopwood, P. et al. 1998. Psychological support needs for women at high genetic risk of breast cancer: some preliminary indicators. *Psycho-Oncology*, 7 (5) 402-412.

Hopwood, P. et al. 2000. Clinical follow-up after bilateral risk-reducing ('prophylactic') mastectomy: mental health and body image outcomes. *Psycho-Oncology* 9 (6) 462-472.

Hopwood, P. 2005. Psychosocial aspects of risk communication and mutation testing in familial breast-ovarian cancer. *Current opinion in oncology*, 17 (4) 340.

Horlick-Jones, T and Prades, A. 2009. On interpretative risk perception research: some reflections on its origins; its nature; and its possible applications in risk communication practice. *Health, Risk and Society* 11 (5) 409-430

Hoskins, L.M. et al. 2008. Disclosure of positive BRCA1/2-mutation status in young couples: The journey from uncertainty to bonding through partner support. *Families, Systems and Health*, 26 (3) 296.

Hoskins, L.M. and Greene, M.H. 2012. Anticipatory loss and early mastectomy for young female BRCA1/2 mutation carriers. *Qualitative Health Research*, 22 (12)1633-1646.

Howard, A.F. et al. 2009. Women's decision making about risk-reducing strategies in the context of hereditary breast and ovarian cancer: A systematic review. *Journal of Genetic Counseling*, 18 (6) 578-597.

Howard, A.F. et al. 2010. Women's constructions of the 'right time' to consider decisions about risk-reducing mastectomy and risk-reducing oophorectomy. *BMC Women's Health*, 10 (1) 24.

Howard, A.F. et al. 2011. Preserving the self: The process of decision making about hereditary breast cancer and ovarian cancer risk reduction. *Qualitative Health Research*, 21 (4) 502-519.

Howlin, F. 2008. Understanding advocacy in children's nursing: a Hermeneutic Study. *Journal of Children's and Young People's Nursing*, 2 (3). 115-125

Hughes, H. et al. 2011 Long term reactions to genetic testing for BRCA1 and BRCA2 mutations: does time heal women's concerns? *Journal of Clinical Oncology*. 29 4302-6

Iredale, R. et al. 2003. The information and support needs of women at high risk of familial breast and ovarian cancer; how can cancer genetic services give patients what they want? *Familial Cancer* 2 119-121

Isern, A.E. et al. 2008. Aesthetic outcome, patient satisfaction, and health-related quality of life in women at high risk undergoing prophylactic

mastectomy and immediate breast reconstruction. *Journal of Plastic, Reconstructive and Aesthetic Surgery* 61 (10) 1177-1187.

Iwakuma, M. 2002. The body as embodiment: An investigation of the body by Merleau-Ponty. *Disability, Postmodernity, Embodying Disability Theory*, pp. 76-87.

Jackson, S and Scott, S, 2010 *Theorising Sexuality*: Open University Press

Jacobson, N. 1998. The socially constructed breast: breast implants and the medical construction of need. *American Journal of Public Health*, 88 (8)1254-1261.

Jeffers, L. et al. 2014. Maximising survival: The main concern of women with hereditary breast and ovarian cancer who undergo genetic testing for BRCA1/2. *European Journal of Oncology Nursing*, 18 (4) 411-418.

Jemal, A. et al. 2011. Global cancer statistics. *CA: A Cancer Journal for Clinicians*, 61 (2) 69-90.

Jemal, A. et al. 2008. Cancer statistics, 2008. *CA: A Cancer Journal for Clinicians*, 58 (2) 71-96.

Jenkins, N. et al. 2013. Inter-embodiment and the experience of genetic testing for familial hypercholesterolaemia. *Sociology of Health and Illness*, 35 (4) 529-543.

Jolie, A. 2013. My medical choice. *New York Times* [Accessed: September 2016] <http://www.nytimes.com/2013/05/14/opinion/my-medical-choice.html>.

Josephson, U. et al. 2000. Initial experiences of women from hereditary breast cancer families after bilateral prophylactic mastectomy: a retrospective study. *European Journal of Surgical Oncology, (EJSO)* 26 (4) 351-356.

Julian-Reynier, C. et al. 1996. Attitudes towards cancer predictive testing and transmission of information to the family. *Journal of Medical Genetics*, 33, 731-736.

Julian-Reynier, C. et al. 2000. Uptake of hereditary breast/ovarian cancer genetic testing in a French national sample of BRCA1 families. *Psycho-Oncology*, 9 (6), 504-510.

Julian-Reynier, C. et al. 2010. Time to prophylactic surgery in BRCA1/2 carriers depends on psychological and other characteristics. *Genetics in Medicine*, 12 (12).

Kaas, R. et al. 2010. Prophylactic mastectomy in BRCA1 and BRCA2 mutation carriers: Very low risk for subsequent breast cancer. *Annals of Surgery*, 251 (3) 488-492.

Kakkori, L. 2010. Hermeneutics and phenomenology problems when applying hermeneutic phenomenological method in educational qualitative research. *Paideusis*, 18 (2) 19-27.

Karaoz, B et al. 2010. A qualitative study of the information needs of premenopausal women with breast cancer in terms of contraception, sexuality, early menopause, and fertility. *International Journal of Gynaecology and Obstetrics*, 109, 118-120.

Katapodi, M, C. 2013 Individual and family characteristics associated with BRCA1/2 genetic testing in high risk families *Psycho-Oncology*, 22 1336-1343

Kavanagh, A.M. and Broom, D.H. 1998. Embodied risk: my body, myself? *Social Science and Medicine*, 46 (3) 437-444.

Kayl, A. and Mayers, C. 2006. Side effects of chemotherapy and quality of life in ovarian and breast cancer patients, *Current Opinion in Obstetrics and Gynaecology* 18: 24–28.

Keller, M. et al. 2008. Psychosocial outcome following genetic risk counselling for familial colorectal cancer. A comparison of affected patients and family members *Clinical Genetics*. 74(5) 414-424

Kelly, K. et al. 2005. Using the common sense model to understand perceived cancer risk in individuals testing for BRCA1/2 mutations. *Psycho-Oncology*, 14 (1) 34-48.

Kenen, R, et al. 2003a. Family stories and the use of heuristics: women from suspected hereditary breast and ovarian cancer (HBOC) families. *Sociology of Health and Illness*, 25 (7) 838-865.

Kenen, R. et al. 2003b. Living with chronic risk: healthy women with a family history of breast/ovarian cancer. *Health, Risk and Society*, 5 (3) 315-331.

Kenen, R et al. 2004 Healthy women from suspected hereditary breast and ovarian cancer families: the significant others in their lives *European Journal of cancer care* 13 169-179

Kenen, R. et al. 2006. "Social separation" among women under 40 years of age diagnosed with breast cancer and carrying a BRCA1 or BRCA2 mutation. *Journal of Genetic Counseling*, 15 (3) 149-162.

Kenen, R.H. et al. 2007. Women with BRCA1 or BRCA2 mutations renegotiating a post-prophylactic mastectomy identity: self-image and self-disclosure. *Journal of Genetic Counseling*, 16 (6) 789-798.

Kessenich, C.R. et al. 2014. Genetic testing for BRCA1 and BRCA2 genes. *The Nurse Practitioner*, 39 (6), 8-10.

Kessler, S. 1993. Forgotten person in the Huntington disease family. *American Journal of Medical Genetics*. 48 (30) 145-150.

King, H.M. 2009. *Risk reduction decision making in women with BRCA1/2 gene mutations*. Dissertation Abstracts International: Section B: The Sciences and Engineering.

King, M. et al. 2000. Quality of life three months and one year after first treatment for early stage breast cancer: influence of treatment and patient characteristics. *Quality of Life Research*, 9 (7) 789-800.

King, M.C. et al. 2003. Breast and ovarian cancer risks due to inherited mutations in BRCA1 and BRCA2. *Science*, 302 (5645) 643-646.

Kinney, A.Y. et al. 2001. Knowledge, attitudes, and interest in breast-ovarian cancer gene testing: a survey of a large African-American kindred with a BRCA1 mutation. *Preventive Medicine*, 33 (6) 543-551.

Klitzman, R. and Chung, W. 2010. The process of deciding about prophylactic surgery for breast and ovarian cancer: Patient questions, uncertainties, and communication. *American Journal of Medical Genetics, Part A*, 152 (1) 52-66.

Klitzman, R.L. and Sweeney, M.M. 2011. "In sickness and in health"? Disclosures of genetic risks in dating. *Journal of Genetic Counseling*, 20 (1) 98-112.

Koch, T. 1995. Interpretive approaches in nursing research: The influence of Husserl and Heidegger. *Journal of Advanced Nursing* 21 (5) 827-836.

Koch, T. 1996. Implementation of a hermeneutic inquiry in nursing: philosophy, rigour and representation. *Journal of Advanced Nursing* 24 (1) 174-184.

Koch, T. 1999. An interpretive research process: Revisiting phenomenological and hermeneutical approaches. *Nurse Researcher* 6 (3) 20.

Koch, T. 2006 Establishing rigour in qualitative research: the decision trail *Journal of Advanced Nursing*, 53 (1) 91-100.

Koch, T. and Harrington, A. 1998. Reconceptualizing rigour: the case for reflexivity. *Journal of advanced nursing* 28(4), pp. 882-890.

Koehly, L.M. et al. 2008. Sisters in hereditary breast and ovarian cancer families: communal coping, social integration, and psychological well-being. *Psycho-Oncology*, 17 (8) 812-821.

Kriege, M. et al. 2004. Efficacy of MRI and mammography for breast-cancer screening in women with a familial or genetic predisposition. *New England Journal of Medicine*, 351 (5) 427-437.

Kurian, A.W. et al. 2005. Opinions of women with high inherited breast cancer risk about prophylactic mastectomy: An initial evaluation from a screening trial including magnetic resonance imaging and ductal lavage. *Health Expectations: An International Journal of Public Participation in Health Care and Health Policy*, 8 (3) 221-233.

Kurian, A.W. et al. 2010. Survival analysis of cancer risk reduction strategies for BRCA1/2 mutation carriers. *Journal of Clinical Oncology*, 28 (2) 222-231.

Kvale, S. 1996. *Interviews: An introduction to qualitative research*. Thousand Oaks, CA: Sage.

Kwong, A and Chu A. 2012 What Made Her Give Up Her Breasts: a Qualitative study on Decisional Consideration's for Contra lateral Prophylactic Mastectomy amongst Breast Cancer Survivors Undergoing BRCA1/2 Genetic Testing *Asian Pacific Journal of Cancer Prevention* 13 (5) 2241-7

Lafrenière, D. et al. 2013. Family communication following BRCA1/2 genetic testing: a close look at the process. *Journal of Genetic Counseling*, 22 (3) 323-335.

Landis, S.H. et al. 1999. Cancer statistics, 1999. *CA: A Cancer Journal for Clinicians*, 49 (1) 8-31.

Landsbergen, K.M. et al. 2010. Female BRCA mutation carriers with a preference for prophylactic mastectomy are more likely to participate an educational-support group and to proceed with the preferred intervention within 2 years. *Familial Cancer*, 9 (2) 213-220.

Langellier, K.M. and Sullivan, C.F. 1998. Breast talk in breast cancer narratives. *Qualitative Health Research*, 8 (1) 76-94.

Lapointe, J. et al. 2012. Influence of the family cluster effect on psychosocial variables in families undergoing BRCA1/2 genetic testing for cancer susceptibility. *Psycho-Oncology* 21 (5) 515-523.

Laverty, S. M. 2003. Hermeneutic phenomenology and phenomenology: A comparison of historical and methodological considerations. *International Journal of Qualitative Methods*, 2 (3) 21-35.

Lee, A. et al. 2014. BOADICEA breast cancer risk prediction model: updates to cancer incidences, tumour pathology and web interface. *British Journal of Cancer* 110 (2) 535-545.

Lee, E.H. et al. 2010. Effect of BRCA1/2 mutation on short-term and long-term breast cancer survival: a systematic review and meta-analysis. *Breast Cancer Research and Treatment*, 122 (1) 11-25.

Leide, A., et al. 2004. Cancer risks for male carriers of germline mutations in BRCA1 or BRCA2 : a review of the literature. *Journal of Clinical Oncology* 22 (40) 735-742.

Lemieux, J. et al. 2008. Chemotherapy-induced alopecia and effects on quality of life among women with breast cancer: a literature review. *Psycho-oncology*, 17 (4) 317.

Lerman, C. et al. 1996. BRCA1 testing in families with hereditary breast-ovarian cancer: a prospective study of patient decision making and outcomes. *JAMA*, 275 (24) 1885-1892.

Lerman, C. and Schwartz, M. 1993. Adherence and psychological adjustment among women at high risk for breast cancer. *Breast Cancer Research and Treatment*, 28 (2) 145-155.

Leslie, H. and McAllister, M. 2002. The benefits of being a nurse in critical social research practice. *Qualitative Health Research*, 12 (5) 700-712.

Lewis, Z.K. et al. 2009. Pancreatic cancer surveillance among high-risk populations: Knowledge and intent. *Journal of Genetic Counseling*, 18 (3) 229-238.

Liebens, F.P. et al. 2007. Management of BRCA1/2 associated breast cancer: A systematic qualitative review of the state of knowledge in 2006. *European Journal of Cancer*, 43 (2) 238-257.

Liede, A. et al. 2000. Evaluation of the needs of male carriers of mutations in BRCA1 or BRCA2 who have undergone genetic counseling. *The American Journal of Human Genetics*, 67 (6) 1494-1504.

Lim, J. et al. 2004. Short and Long-Term Impact of Receiving Genetic Mutation Results in Women at Increased Risk for Hereditary Breast Cancer. *Journal of Genetic Counseling*, 13 (2) 115-133.

Lincoln, Y.S. and Guba, G.G.1985. *Naturalistic Inquiry*. Thousand Oaks, CA: Sage Publications.



Linda Wasserman, R. 2013. BRCA Genetic Testing: An RN's Personal Story. *Clinical Journal of Oncology Nursing*, 17 (4) 449.

Linden, W. et al. 2012. Anxiety and depression after cancer diagnosis: Prevalence rates by cancer type, gender, and age. *Journal of Affective Disorders*, 141 (2) 343-351.

Lindenmeyer, A. et al. 2010. 'The family is part of the treatment really': A qualitative exploration of collective health narratives in families. *Health: An Interdisciplinary Journal for the Social Study of Health, Illness and Medicine*, pp. 1-15.

Lindwall, L. and Von Post, I. 2008. Habits in perioperative nursing culture. *Nursing Ethics*, 15 (5) 670-681.

Lloyd, S. et al. 2000. Understanding the experience of prophylactic bilateral mastectomy: a qualitative study of ten women. *Psycho-Oncology*, 9 (6) 473-485.

Lobb, E. and Meiser, B. 2004. Genetic counselling and prophylactic surgery in women from families with hereditary breast or ovarian cancer. *The Lancet*, 363 (9424) 1-4.

Lobb, E.A. et al. 2005. Differences in Individual Approaches: Communication in the Familial Breast Cancer Consultation and the Effect on Patient Outcomes. *Journal of Genetic Counseling*, 14 (1) 43-53.

Lobb, E.A. et al. 2009. Attendance of men at the familial cancer clinic: what they value from the consultation. *Genetics in Medicine*, 11 (6) 434-440.

Lodder, L.N. et al. 2001. Psychological impact of receiving a BRCA1/BRCA2 test result. *American Journal of Medical Genetics*, 98 (1) 15-24.

Lodder, L.N. et al. 2002. One year follow-up of women opting for pre-symptomatic testing for BRCA1 and BRCA2: emotional impact of the test outcome and decisions on risk management (surveillance or prophylactic surgery). *Breast Cancer Research and Treatment*, 73 (2) 97-112.

Lostumbo, L. et al. 2010. Prophylactic mastectomy for the prevention of breast cancer. *Cochrane database of systematic reviews*.  
<http://onlinelibrary.wiley.com/doi/10.1002/14651858.CD002748.pub3/full>  
DOI: 10.1002/14651858.CD002748.pub3

Lowes, L. and Prowse, M.A. 2001. Standing outside the interview process? The illusion of objectivity in phenomenological data generation. *International Journal of Nursing Studies*, 38 (4) 471-480.

Lynch, H.T. et al. 2006. Patient responses to the disclosure of BRCA mutation tests in hereditary breast-ovarian cancer families. *Cancer genetics and Cytogenetics*, 165 (2) 91-97.

Lynch, J.A. et al. eds. 2015. *Genetic tests to identify risk for breast cancer*. Seminars in Oncology Nursing. Elsevier.

Mackey, S. 2005. Phenomenological nursing research: methodological insights derived from Heidegger's interpretive phenomenology. *International Journal of Nursing Studies*, 42 (2) 179-186.

Madalinska, J.B. et al. 2007. Predictors of prophylactic bilateral salpingo-oophorectomy compared with gynaecologic screening use in BRCA1/2 mutation carriers. *Journal of Clinical Oncology*, 25 (3) 301-307.

Madison, G 1988 IN; de Witt, L. and Ploeg, J. 2006. Critical Appraisal of rigour in interpretive phenomenology nursing research. *Journal of Advanced Nursing*, 55 (2) 215-229.

Maggs-Rapport, F. 2001 'Best Research Practice'; in pursuit of methodological rigour. *Journal of Advanced Practice*, 35 (3) 373-383.

Maheu, C. et al. 2014. Which factors predict proposal and uptake of psychological counselling after BRCA1/2 test result disclosure? *Psycho-Oncology*, 23 (4) 420-427.

Mahon, S. 2000. Factors affecting genetic testing and decisions about prophylactic surgery. *Clinical Journal of Oncology Nursing*, 5 (3) 117-120.

Mahon, S.M. 2013. Large genomic rearrangements in BRCA1 and BRCA2: implications for patient care. *Oncology Nursing Forum*, 40 (3) 220-222.

Majdak-Paredes, E.J. and Fatah, F. 2009. Hereditary breast cancer syndromes and clinical implications. *Journal of Plastic, Reconstructive & Aesthetic Surgery*, 62 (2) 181-189.

Malata, C. et al. 2000. Immediate breast reconstruction after mastectomy for cancer. *British Journal of Surgery*, 87 (11) 1455-1472.

Manganiello, A. et al. 2011. Sexuality and quality of life of breast cancer patients post mastectomy. *European Journal of Oncology Nursing*, 15 (2) 167-172.

Marshall, C. and Kiemle, G. 2005. Breast reconstruction following cancer: Its impact on patients' and partners' sexual functioning. *Sexual and Relationship Therapy*. 20 (2) 155-179.

Martin, E. 1989a. The cultural construction of gendered bodies: Biology and metaphors of production and destruction\*. *Ethnos*, 54 (3-4) 143-160.

Martin, E. 1989b. *The Woman in the Body*. Buckingham: Open University Press.

Mason, J. 2002. *Qualitative Researching* (2nd edn). London, Thousand Oaks, New Delhi: Sage Publications.

Matloff, E.T. et al. 2000. What would you do? Specialists' perspectives on cancer genetic testing, prophylactic surgery, and insurance discrimination. *Journal of Clinical Oncology*, 18 (12) 2484-2492.

Matloff, E.T. et al. 2009. Unravelling the next chapter: sexual development, body image, and sexual functioning in female BRCA carriers. *The Cancer Journal*, 15 (1) 15-18.

Mauer, C. et al. 2015. Exploration of Male Attitudes on Partnerships and Sexuality with Female BRCA1/2 Mutation Carriers. *Journal of Genetic Counseling*, pp. 1-8.

McAllister, M.F. et al. 1998. Men in breast cancer families; a preliminary qualitative study of awareness and experience. *Journal of Medical Genetics*, 35, 739-744.

McAllister, M.F. 2001. Grounded theory in genetic counselling research. *Journal of Genetic Counseling*, 10 (3) 233-250.

McArdle, J.M. et al. 1996. Psychological support for patients undergoing breast cancer surgery: a randomised study. *British Medical Journal*, 312 (7034) 813-816.

McBride, K.A. et al. 2015. Timing and context: important considerations in the return of genetic results to research participants. *Journal of Community Genetics*, p.1-10.

McCann, G.A. and Eisenhauer, E.L. 2015. Hereditary cancer syndromes with high risk of endometrial and ovarian cancer: Surgical options for personalized care. *Journal of Surgical Oncology*, 111 (1) 118-124.

McConnell, T. et al. 2009. Husserl and Heidegger: Exploring the disparity *International Journal of Nursing Practice* 15 (1) 7-15

McCullum, M. et al. 2007. Time to decide about risk-reducing mastectomy: A case series of BRCA1/2 gene mutation carriers. *BMC women's health* [Online] 7(3). Available at: <http://www.scopus.com/inward/record.url?eid=2-s2.0->

58749102454&partnerID=40&md5=66c24af6384877500b9fd10e25112fe9  
[Accessed: 14/05/2016].

McEwan, A. 2011 'Getting on with it' New Zealand Women's Experiences of Living with an increased Risk of Breast and Ovarian Cancer. PhD Thesis. University of Otago. Available at:  
<https://ourarchive.otago.ac.nz/bitstream/handle/10523/1908/mcewenalison2011phd.pdf?sequence=3>. [Accessed 29/1/17].

McGaughey, A. 2006. Body image after bilateral prophylactic mastectomy: an integrative literature review. *Journal of Midwifery and Women's Health* 51 (6) e45-e49.

McLachlan, K. 2009. Information and support needs of young women with breast cancer. *Nursing Practice*, 8 (8) 21-24.

McManus Holroyd, A.E. 2007. Interpretive hermeneutic phenomenology: Clarifying understanding. *Indo-Pacific Journal of Phenomenology* 7 (2) 1-12.

McNamara, M.S. 2005. Knowing and doing phenomenology: The implications of the critique of 'nursing phenomenology' for a phenomenological inquiry: A discussion paper. *International Journal of Nursing Studies* 42 (6) 695-704.

McQuirter, M. et al. 2010. Decision-making process of women carrying a BRCA1 or BRCA2 mutation who have chosen prophylactic mastectomy. *Oncology Nursing Forum*, 37 (3) 313-320.

McTiernan, A. et al. 2008. Breast cancer prevention in countries with diverse resources. *Cancer*, 113 (8, Suppl) 2325-2330.

Mehnert, A. and Koch, U. 2007. Prevalence of acute and post-traumatic stress disorder and co-morbid mental health disorders in breast cancer patients during primary care: a prospective study. *Psych-oncology* 16(3)181-188.

Mehnert, A. and Koch, U. 2008. Psychological comorbidity and health-related quality of life and its association with awareness, utilization, and need for psychosocial support in a cancer register-based sample of long-term breast cancer survivors. *Journal of Psychosomatic Research*, 64 (4) 383-391.

Mehnert, A. et al. 2007. The association between neuropsychological impairment, self-perceived cognitive deficits, fatigue and health related quality of life in breast cancer survivors following standard adjuvant versus high-dose chemotherapy. *Patient Education and Counselling*, 66 (1) 108-118.

Meijers-Heijboer, H. et al. 2001. Breast cancer after prophylactic bilateral mastectomy in women with a BRCA1 or BRCA2 mutation. *New England Journal of Medicine*, 345 (3)159-164.

Meiser, B. et al. 2003. Attitudes to Prophylactic Surgery and Chemoprevention in Australian Women at Increased Risk for Breast Cancer. *Journal of Women's Health*, 12 (8) 769-778.

Meiser, B. 2005. Psychological impact of genetic testing for cancer susceptibility: an update of the literature. *Psycho-Oncology*, 14 (12) 1060-1074.

Meiser, B and Halliday, J. 2002. What is the impact of genetic counselling in women at increased risk of developing hereditary breast cancer. A meta-analytic review. *Social Science and Medicine* 54 1463-1470

Mellon, S. et al. 2009. Predictors of decision making in families at risk for inherited breast/ovarian cancer. *Health Psychology*, 28 (1) 38.

Merleau-Ponty, M.1962. *Phenomenology of Perception* (C Smith translated) London: Routledge and Kegan Paul.

Merleau-Ponty, M, 2002. *Phenomenology of Perception*. London: Routledge.

Metcalfe, K.A. 2004. Prophylactic bilateral mastectomy for breast cancer prevention. *Journal of Women's Health*, 13 (7) 822-829.

Metcalfe, K.A. et al. 2002. Evaluation of the needs of spouses of female carriers of mutations in BRCA1 and BRCA2. *Journal of Clinical Genetics*, 62 464-469.

Metcalfe, K.A. et al. 2004a. Psychosocial functioning in women who have undergone bilateral prophylactic mastectomy. *Psycho-Oncology*, 13 (1) 14-25.

Metcalfe, K.A. et al. 2004b. Satisfaction with breast reconstruction in women with bilateral prophylactic mastectomy: a descriptive study. *Plastic and Reconstructive Surgery*, 114 (2) 360-366.

Metcalfe, K.A. et al. 2005. Predictors of quality of life in women with a bilateral prophylactic mastectomy. *The Breast Journal*, 11 (1) 65-69.

Metcalfe, K.A. et al. 2008. Predictors of contralateral prophylactic mastectomy in women with a BRCA1 or BRCA2 mutation: The hereditary breast cancer clinical study group. *Journal of Clinical Oncology*. 26 (7) 1093-1097.

Metcalfe, K.A. and Narod, S.A. 2002. Breast cancer risk perception among women who have undergone prophylactic bilateral mastectomy. *Journal of the National Cancer Institute*, 94 (20) 1564-1569.

Miki, Y. et al. 1994. A strong candidate for the breast and ovarian cancer susceptibility gene. *Science*, 266, 66-71.

Miles, M. 2013. Exploring Heideggerian hermeneutic phenomenology: A perfect fit for midwifery research. *Woman and Birth*, 26 (4) 273-276.

Miles, M and Huberman, A. 1994. *Qualitative Data Analysis*. London: Sage.

Miller, S. et al. 2010. New strategies in ovarian cancer: uptake and experience of women at high risk of ovarian cancer who are considering risk-reducing salpingo-oophorectomy. *Clinical Cancer Research*, p. clincanres. 2953.2010.

Mireskandari, S. et al. 2006. Evaluation of the needs and concerns of partners of women at high risk of developing breast/ovarian cancer. *Psycho-Oncology*, 15 (2) 96-108.

Mireskandari, S. et al. 2007. Psychological adjustment among partners of women at high risk of developing breast/ovarian cancer. *Genetics in Medicine* 9 (5) 311-320.

Mokbel, K. 2003. Risk-reducing strategies for breast cancer: A review of recent literature. *International Journal of Fertility and Women's Medicine*, 48 (6) 274-277.

Maloney, P. and Kirkman, A. 2005. Introduction: Mapping the sexual landscape. In A. Kirkman and P. Maloney (Eds) *Sexuality Down Under: Social and Historical Perspectives*. University of Otago Press.

Mols, F. et al. 2005. Quality of life among long-term breast cancer survivors: a systematic review. *European Journal of Cancer*, 41 (17) 2613-2619.

Montazeri, A. 2008. Health-related quality of life in breast cancer patients: a bibliographic review of the literature from 1974 to 2007. *Journal of Experimental and Clinical Cancer Research* 27 (1) 1.

Moran, D. 2000. *Introduction to phenomenology*. New York: Routledge.

Moran, D. and Mooney, T. 2002. (Eds) *The phenomenology Reader*. New York: Routledge.

Morse, J.M. 2002. Verification Strategies for establishing reliability and validity in qualitative research. *International Journal of qualitative methods*, 1 (2) 13.

Nagar, H. et al. 2011. Local-regional recurrence with and without radiation therapy after neoadjuvant chemotherapy and mastectomy for clinically staged T3N0 breast cancer. *International Journal of Radiation Oncology, Biology, Physics*, 81 (3) 782-787.

Narod, S. et al. 2001. Prophylactic mastectomy in carriers of BRCA mutations. *New England Journal of Medicine*, 345 (20) 1498-1500.

Narod, S.A. and Offit, K. 2005. Prevention and management of hereditary breast cancer. *Journal of Clinical Oncology*, 23 (8) 1656.

Nathanson, K.L. and Domchek, S.M. 2011. Therapeutic approaches for women predisposed to breast cancer. *Annual Review of Medicine*, 62, 295-306.

National Mastectomy and Breast Reconstruction Audit (2010). Third Report, Leeds, NHS Information Centre.

National Cancer Institute 2015

<https://www.cancer.gov/about-cancer/causes-prevention/genetics/brca-fact-sheet> [Accessed September 2016]

National Institute for Health and Care Excellence 2009. <https://www.nice.org.uk/guidance/qs12/documents/briefing-paper> [Accessed October 2016]

National Institute for Health and Care Excellence. 2013 (CG164). *Familial Breast Cancer. Classification, care and managing breast cancer and related risks in people with a family history of breast cancer. Updated 2017* <https://nice.org.uk/guidance/cg164> [Accessed 10.9.2016]

Nelson, H.D. et al. 2009. Systematic review comparative effectiveness of medications to reduce risk for primary breast cancer. *Annals of Internal Medicine*, 151 (10) 703-715.

Nelson, J.A. et al. 2012. Breast reconstruction in bilateral prophylactic mastectomy patients: Factors that influence decision making. *Journal of Plastic, Reconstructive and Aesthetic Surgery* 65 (11) 1481-1489.

Nettleton, S. and Watson, J. 1998. *The body in everyday life*. London: Routledge.

Neuberger J, MacNeil, F. Jeevan, R. et al. Trends in the use of bilateral mastectomy in England from 2002 to 2011: Retrospective analysis of hospital episode statistics. *British Medical Journal Open* 2013; **3**:e003179.

Nissen, M.J. et al. 2001. Quality of life after breast carcinoma surgery. *Cancer*, 91 (7) 1238-1246.

Norlyk, A. and Harder, I. 2010. What makes a phenomenological study phenomenological? An analysis of peer-reviewed empirical nursing studies *Qualitative Health Research*, 20 (3) 420-431.

Novas, C. and Rose, N. 2000 Genetic Risk and the birth of the somatic individual. *Economy and Society*, 29 (4) 485-513.

Nunokoosing, K. 2005. The problem with interviews. *Qualitative Health Research* 15 (5) 698-706.

Nursing and Midwifery Council (2008). The code: Standards of conduct, performance and ethics for nurses and midwives London: NMC.

Nursing and Midwifery Council (2015). The code: Standards of practice and behaviour for nurses and midwives. London: NMC.

Ohsumi, S. et al. 2007. Quality of life of breast cancer patients and types of surgery for breast cancer-Current status and unresolved issues. *Breast cancer*, 14 (1) 66-73.

O'Leary, Z. 2004. *Essential guide to doing research*. London: Sage.

Ozakinci, G. 2005. *Psychological and behavioural outcomes of genetic testing for BRCA1/2 mutations among Ashkenazi Jewish Women*. Dissertation Abstracts International: Section B: The Sciences and Engineering.

Paley, J. 1997. Husserl, phenomenology and nursing. *Journal of Advanced Nursing*, 26 (1) 187-193.

Paley, J. 1998. Misinterpretive phenomenology: Heidegger, ontology and nursing research. *Journal of Advanced Nursing*, 27 (4) 817-824.

Paley, J. 2005. Phenomenology as rhetoric. *Nursing Inquiry*, 12 (2) 106-116.

Palmer, C.G.S, and Sainfort, F. 1993. Toward a new conceptualisation and operationalisation of risk perception within the genetic counselling domain. *Journal of Genetic Counseling*, 2 (4) 275-294

Park, S.S. 2013. Gene patents and the public interest: litigating association for molecular pathology v. Myriad Genetics and lessons moving forward. *NCJL and Tech*. 15, 519.

Patenaude, A.F. et al. 2006. Sharing BRCA1/2 test results with first degree relatives: Factors predicting who women tell. *Journal of Clinical Oncology*, 24 (4) 700-706.

Patenaude, A.F. et al. 2008. Support needs and acceptability of psychological and peer consultation: attitudes of 108 women who had undergone or were considering prophylactic mastectomy. *Psycho-Oncology*, 17 (8) 831-843.

Patenaude, A.F. et al. 2013. Young adult daughters of BRCA1/2 positive mothers: What do they know about hereditary cancer and how much do they worry? *Psycho-Oncology*, 22 (9) 2024-2031.



Paterson, D.L.N. 2010. *Family, loss, and place: everyday experiences of hereditary cancer in a First Nation community*. University of British Columbia Vancouver.

Payne, D.K. et al. 2000. Women's regrets after bilateral prophylactic mastectomy. *Annals of Surgical Oncology*, 7 (2) 150-154.

Pelters, B 2014. Gender Trouble? Queering the medical normativity of BRCA femininities. In Gibbons, S et al. (eds). *Breast Cancer Gene Research and Medical practices Transnational Perspectives in the time of BRCA*. New York: Routledge

Peters,,J et al. 2011 Unpacking the blockers: Understanding Perceptions and Social Constraints of Health Communication in Hereditary Breast Ovarian Cancer ( HBOC) Susceptibility Families *Journal of Genetic Counseling* 20(5) 450-464

Petronis, V.M. et al. 2003. Investment in body image and psychosocial well-being among women treated for early stage breast cancer: partial replication and extension. *Psychology and Health*,18 (1) 1-13.

Phillips, M. and Pugh, D.S. 2000. *How to get a PhD: A handbook for students and their supervisors*. 3<sup>rd</sup> edition. Maidenhead: Open University Press.

Phillips-Pula, L. et al. 2011. Understanding phenomenological approaches to data analysis. *Journal of Paediatric Health Care*, 25 (1) 67-71.

Piot-Ziegler, C. et al. 2010. Mastectomy, body deconstruction, and impact on identity: a qualitative study. *British Journal of Health Psychology*, 15 (3) 479-510.

Polkinghorne ,D.E. 2006. An agenda for the second generation of qualitative studies. *International Journal of Qualitative Studies on Health and Well being*.1, 68-77.

Porter, E.J. 1998. On 'being inspired' by Husserl's phenomenology; reflection on Omery's exposition of phenomenology as a method of nursing research. *Advances in Nursing Science*, 21 (1) 16-28.

Potter, S. et al. 2013. Exploring inequalities in access to care and the provision of choice to women seeking breast reconstruction surgery: a qualitative study. *British Journal of Cancer*, 109 (5) 1181-1191.

Press, N. et al. 2005. That's like chopping off a finger because you're afraid it might get broken': Disease and illness in women's views of prophylactic mastectomy. *Social Science and Medicine*, 61 (5) 1106-1117.

Price, M.A. et al. 2007. Predictors of cancer worry in unaffected women from high risk breast cancer families: Risk perception is not the primary issue. *Journal of Genetic Counseling*, 16 (5) 635-644.

Pringle, J. et al. 2011. Phenomenological approaches: challenges and choices. *Nurse Researcher*, 18 (2) 7-18.

Puntoni, S. et al. 2011. Gender identity salience and perceived vulnerability to breast cancer. *Journal of Marketing Research*, 48 (3) 413-424.

Racher, F. and Robinson, S. 2003 Are phenomenology and post-positivism strange bedfellows? *Western Journal of Nursing Research*, 25 (5) 464-481.

Rebeck, T.R. et al. 2004. Bilateral prophylactic mastectomy reduces breast cancer risk in BRCA1 and BRCA2 mutation carriers: the PROSE Study Group. *Journal of Clinical Oncology*, 22 (6) 1055-1062.

Rebeck, T.R. et al. 2009. Meta-analysis of risk-reducing estimates associated with risk-reducing salpingo-oophorectomy in BRCA1 or BRCA2 mutation carriers. *Journal of the National Cancer Institute* 101 (2) 80-87.

Recio-Saucedo, A. et al. 2016. Information requirements of young women with breast cancer treated with mastectomy or breast conserving surgery: A systematic review. *The Breast*, 25, 1-13.

Reynolds, C. et al. 2011. Prophylactic and therapeutic mastectomy in BRCA mutation carriers; can the nipple be spared? *Annals of Surgical Oncology* 18 (3) 102-3109.

Richards, M.P. 1999. Genetic counselling for those with a family history of breast or ovarian cancer: Current practice and ethical issues. *Acta Oncologica* 38(5), pp. 559-565.

Ricoeur, P. 1976 *Interpretation theory: Discourse and the surplus of meaning*. Fort Worth: Texas Christian University Press.

Rini, C. et al. 2009. Cognitive and emotional factors predicting decisional conflict among high-risk breast cancer survivors who receive uninformative BRCA1/2 results. *Health Psychology* 28 (5) 569-578.

Ritchie, J. et al. 2014. *Qualitative research practice: A guide for social science students and researchers*. London: Sage.

Robertson, A. 2000. Embodying risk, embodying political rationality: women's accounts of risks for breast cancer. *Health, Risk and Society*, 2 (2) 219-235.

Rogozinska-Szczepka, J et al. 2004. BRCA1 and BRCA2 mutations as prognostic factors in bilateral breast cancer patients *Annals of Oncology*, 15 (9) 1373-1376.

Rolf, G. 2006. Validity, trustworthiness and rigour; quality and the idea of qualitative research *Journal of Advanced Nursing*, 53 (3) 304-310.

Rolland, J. 2006. Living with anticipatory loss in the new era of genetics. *Individuals, families, and the new era of genetics*. Norton, New York, pp. 139-172.

Rolnick, S. J. et al. 2007. What women wish they knew before prophylactic mastectomy? *Cancer Nursing*, 30 (4) 285-293.

Rosenberg, S.M. et al 2013. Body image in recently diagnosed young women with early breast cancer. *Psycho-Oncology* 22 (8) 1849-1955.

Rosenblatt, L. 2006. Being the monster: women's narrative of body and self after treatment for breast cancer. *Medical Humanities*, 32, 53-56.

Rowland, E. and Metcalfe, A. 2014. A systematic review of men's experiences of their partner's mastectomy: coping with altered bodies. *Psycho-Oncology*, 23 (9) 963-974.

Rowland, E et al 2016. Preparing young people for future decision-making about cancer risk in families affected or at risk from hereditary breast cancer. A qualitative interview study. *European Journal of Oncology Nursing* 25 9-15

Royal College of Nursing (2007) Research Ethics. Research Guidance for Nurses. London: RCN.

Rueth, N.M. et al. 2011. Preoperative Risk Assessment Among Women Undergoing Bilateral Prophylactic Mastectomy for Cancer Risk Reduction. *Annals of Surgical Oncology*, 18 (9).

Rumsey, N. and Harcourt, D. 2004. Body image and disfigurement: issues and interventions. *Body Image*, 1 (1) 83-97.

Rustoen, T. and Begnum, S. 2000. Quality of life in women with breast cancer: A review of the literature and implications for practice. *Cancer Nursing*, 23 (6) 416-421.

Sabo, D. et al. 1986. The male role and mastectomy: Support groups and men's adjustment. *Journal of Psychosocial Oncology*, 4 (1-2), 19-31.

Salant, T. et al. 2006. "Why Take It If You don't have Anything?" Breast Cancer Risk Perceptions and Prevention Choices at a Public Hospital. *Journal of General Internal Medicine*, 21 (7) 779-785.

Salhab, M. et al. 2010. Risk-reducing strategies for women carrying brca1/2 mutations with a focus on prophylactic surgery. *BMC Women's Health* 10 (1) 28.

Sandelowski, M. 2002. Finding the Findings in Qualitative Studies. *Journal of Nursing Scholarship*, 34 (3) 213-219.

Sandham, C. and Harcourt, D. 2007. Partner experiences of breast reconstruction post mastectomy. *European Journal of Oncology Nursing*, 11 (1) 66-73.

Sauven, P. 2004. Guidelines for the management of women at increased familial risk of breast cancer. *European Journal of Cancer*, 40 (5) 653-665.

Schlich-Bakker, K.J. et al. 2009. Distress in couples approached for genetic counseling and BRCA1/2 testing during adjuvant radiotherapy. *Psycho-Oncology*, 18 (9) 965-973.

Schover, L.R. 2008. A lesser evil: Prophylactic mastectomy for women at high risk for breast cancer. *Journal of Clinical Oncology*, 26 (24) 3918.

Schrag, D. et al. 1997. Decision analysis-effects of prophylactic mastectomy and oophorectomy on life expectancy among women with BRCA1 or BRCA2 mutations. *New England Journal of Medicine*, 336 (20) 1465-1471.

Schwartz, M.D. 2005. Contralateral prophylactic mastectomy: efficacy, satisfaction, and regret. *Journal of Clinical Oncology*, 23 (31) 7777.

Schwartz, M.D. et al. 2009. Randomized Trial of a Decision Aid for BRCA1/BRCA2 Mutation Carriers: Impact on Measures of Decision Making and Satisfaction. *Health Psychology*, 28 (1) 11-19.

Schwartz, M.D. et al. 2012. Long-term outcomes of BRCA1/BRCA2 testing: Risk reduction and surveillance. *Cancer* 118 (2) 510-517.

Sheehan, J., Sherman, K.A., Lam, T. and Boyages, J. 2007. Association of information satisfaction, psychological distress & monitoring coping style with post-decision regret following breast reconstruction, *Psycho-oncology*, 16 (4) 342-51.

Sheehan, J., Sherman, K.A., Lam, T. and Boyages, J. 2008. Regret associated with the decision for breast reconstruction: The association of negative body

image, distress and surgery characteristics with decision regret. *Psychology and Health*, 23 (2) 207-19.

Sherkow, J.S. and Scott, C.T. 2014. Myriad stands alone. *Nature biotechnology* 32, 620.

Sherman, K.A. et al. 2010. Psychological adjustment among male partners in response to women's breast/ovarian cancer risk: a theoretical review of the literature. *Psycho-Oncology*, 19 (1) 1-11.

Shilling, C. 2012. *The Body and Social Theory*. London: Sage.

Shiloh, S. et al. 2013. A follow-up study on men tested for BRCA1/BRCA2 mutations: impacts and coping processes. *Psycho-Oncology*, 22 (2) 417-425.

Siedman, I.E. 1991. *Interviews as qualitative research*. New York: Teaching college Press.

Silverman, D. 2005. *Doing qualitative research* (2<sup>nd</sup> edition) London: Thousand Oaks: Sage.

Silverman, D. 2011. *Interpreting qualitative data: A guide to the principles of qualitative research*. 4th edition. London: Sage.

Simchoni, S. 2006. Familial clustering of site-specific cancer risks associated with BRCA1 and BRCA2 in the Ashkenazi Jewish Population. *Proceedings of the National Academy of Sciences of the United States of America* 13 (10) 3770-3774.

Sivell, S. et al 2008. How risk is perceived, constructed and interpreted by clients in clinical genetics, and the effects on decision making: Systematic Review. *Journal of Genetic Counseling*, 17 (1) 30-63

Skytte, A.B. et al. 2010. Risk-reducing mastectomy and salpingo-oophorectomy in unaffected BRCA mutation carriers: uptake and timing\*. *Clinical Genetics*, 77 (4) 342-349.

Skytte, A.B. et al. 2011. Breast cancer after bilateral risk-reducing mastectomy. *Clinical Genetics*, 79 (5) 431-437.

Smith, A.W. et al. 2008. Psychological distress and quality of life associated with genetic testing for breast cancer risk. *Psycho-Oncology* 17 (8) 767-773.

Smith, K.L. and Isaacs, C. 2007. Management of women at increased risk for hereditary breast cancer. *Breast Disease*, 27 (1) 51-67.

Smith, N. 2011. My breasts will kill me. *Psycho-Oncology*, 6 (3) 90-106.

Solvi, A.S. et al. 2010. Motivational factors and psychological processes in cosmetic breast augmentation surgery. *Journal of Plastic, Reconstructive and Aesthetic Surgery*, 63 (4) 673-680.

Spear, S.L. et al. 2008. Prophylactic mastectomy and reconstruction: clinical outcomes and patient satisfaction. *Plastic and Reconstructive Surgery* 122 (1) 1-9.

Spence, D.G. 2001. Hermeneutic notions illuminate cross-cultural nursing experiences. *Journal of Advanced Nursing*. 35 (4) 624-630.

Spence, D.G. 2005 Hermeneutic notions augment cultural safety education. *Journal of Nursing Education*, 44 (9) 409-414.

Speziale, H.S. and Carpenter, D.R. 2007. *Qualitative Research In Nursing: Advancing the Humanistic Imperative*. (4<sup>th</sup> edition) Philadelphia: Lippincott Williams and Wilkins.

Spiegelberg, H. 1982. *The Phenomenological Movement*. Boston: Martinus Nijhoff.

Stefanek, M.E. et al. 1995. Predictors of and satisfaction with bilateral prophylactic mastectomy. *Preventive medicine*, 24 (4) 412-419.

Stefanek, M. et al. 2001. Risk-reduction mastectomy: clinical issues and research needs. *Journal of the National Cancer Institute*, 93 (17) 1297.

Stoppa-Lyonnet, D. et al. 2000. Familial invasive breast cancers: worse outcome related to BRCA1 mutations. *Journal of Clinical Oncology* 18 (24) 4053-4059.

Streubert, H.J. et al. 2002. Phenomenology as method IN *Qualitative Research in Nursing: Advancing the humanistic imperative* (3<sup>rd</sup> edn) Philadelphia: Lippincott

Streubert, H.J. and Carpenter, D.R. 2011. *Qualitative research in nursing: Advancing the humanistic imperative*. London: Lippincott Williams and Wilkins.

Strømsvik, N. et al. 2009. Men in the women's world of hereditary breast and ovarian cancer—a systematic review. *Familial cancer*, 8 (3) 221-229.

Strømsvik, N. et al. 2010. Stigmatization and male identity: Norwegian males' experience after identification as BRCA1/2 mutation carriers. *Journal of Genetic Counseling*, 19 (4) 360-370.

Sussner, K.M. et al. 2013. Barriers and facilitators to BRCA genetic counseling among at-risk Latinas in New York City. *Psycho-Oncology*, 22 (7) 1594-1604.

Tan, M.B.M. et al. 2009. Standard psychological consultations and follow up for women at increased risk of hereditary breast cancer considering prophylactic mastectomy. *Hereditary Cancer in Clinical Practice*, 7 (6) 1-7.

Tapp,D. 2004. Dilemmas of family support during cardiac recovery. Nagging as a gesture of support. *Western Journal of Nursing Research*, 26 (5) 561-580.

Taucher, S. et al. 2003. Preventive mastectomy in patients at breast cancer risk due to genetic alterations in the BRCA1 and BRCA2 gene. *Langenbecks Archives of Surgery*, 388 (1) 3-8.

Tercyak, K.P. et al. 2007. Quality of life after contralateral prophylactic mastectomy in newly diagnosed high-risk breast cancer patients who underwent BRCA1/2 gene testing. *Journal of Clinical Oncology*, 25 (3) 285.

Thomas, S. Through the lens of Merleau-Ponty: advancing the phenomenological approach to nursing research. *Nursing Philosophy* 6, 63-76.

Thomas–Maclean, R 2005. Beyond dichotomies of health and illness. Life after breast cancer. *Nursing Inquiry* 1 (3) 200-209

Thompson, J. 1990. Hermeneutic Inquiry. IN *Advancing nursing science through Research*.2. California: Sage.

Tong, A. Kelly, S. et al. 2015. Intentions for risk-reducing amongst high risk women. *Psycho-Oncology*, 24 (1) 33-39.

Turner,D.S. 2003. Horizons revealed: from methodology to method. *International journal of qualitative of qualitative methods*.2 (1) 1-17.

United Kingdom Genetic Testing Network (2017)  
<https://ukgtn.nhs.uk/find-a-test/search-by-disorder-gene/test-service/familial-breast-ovarian-cancer-62/>  
Accessed September 2016

Uyei, S. et al. 2006. Association between clinical characteristic's and risk-reduction interventions in women who underwent BRCA1 and BRCA2 testing. *Cancer* 107 (12) 2745-2751.

Vahdaninia, M. et al. 2010. What do predict anxiety and depression in breast cancer patients? A follow -up study. *Social Psychiatry and Psychiatric Epidemiology*, 45 (3) 355-361.

Vandermause, R.K. and Fleming, S.E. 2011. Philosophical hermeneutic interviewing. *International Journal of Qualitative Methods*, 10 (4) 367-377.

- van de Water, W. et al. 2013. Elderly postmenopausal patients with breast cancer are at increased risk for distant recurrence: a tamoxifen exemestane adjuvant multinational study analysis. *The Oncologist*, 18 (1) 8-13.
- van Dijk, S. et al. 2003. Genetic counselling and the intention to undergo risk-reducing mastectomy: Effects of a breast cancer risk assessment. *British Journal of Cancer*, 88 (11) 1675–1681.
- van Dijk, S. et al. 2005. What's the message? Interpretation of an uninformative BRCA1/2 test result for women at risk of familial breast cancer. *Genetics in Medicine*, 7 (4) 239.
- van Dijk, S. et al. 2008. Decision making regarding prophylactic mastectomy: Stability of preferences and the impact of anticipated feelings of regret. *Journal of Clinical Oncology*, 26 (14) 2358-2363.
- van Geel, A. 2003. Prophylactic mastectomy: the Rotterdam experience. *The Breast*, 12 (6) 357-361.
- van Manen, M. 1990. *Researching lived experience: Human science for an action sensitive pedagogy*. New York: State University of New York press
- van Manen, M. 1997. From meaning to method. *Qualitative Health Research* 7, 345-369.
- van Manen, M. 2014. *Phenomenology of practice: Meaning-giving methods in phenomenological research and writing*. California: Left Coast Press.
- van Oostrom, I. et al. 2003. Long-term psychological impact of carrying a BRCA1/2 mutation and prophylactic surgery: a 5-year follow-up study. *Journal of Clinical Oncology*, 21 (20) 3867.
- van Oostrom, I. et al. 2007. A prospective study of the impact of genetic susceptibility testing for BRCA1/2 or HNPCC on family relationships. *Psycho-oncology* 14 (4) 320-328.
- Vargas, A. et al. 2010. The contribution of breast cancer pathology to statistical models to predict mutation risk in BRCA carriers. *Familial Cancer*, 9 (4) 545-553.
- Vodermaier, A. et al. 2006. Prophylactic mastectomy in women at high risk for breast and ovarian cancer: Qualitative analysis of the decision-making process and long-term satisfaction of two women carrying a BRCA1-mutation. *Psychotherapie Psychosomatik Medizinische Psychologie* 56 (9-10) 351-361.



Vodermaier, A. et al. 2010. Can self-esteem, mastery and perceived stigma predict long-term adjustment in women carrying a BRCA1/2-mutation? Evidence from a multi-center study. *Familial cancer*, 9 (3) 305-311.

Voorwinden, J.S. and Jaspers, J.P. 2014. Prognostic factors for distress after genetic testing for hereditary cancer. *Journal of Genetic Counseling*, 1-9.

Vos, J. et al. 2013. The counselees' self-reported request for psychological help in genetic counselling for hereditary breast/ovarian cancer: not only psychopathology matters. *Psycho-Oncology*, 22 (4) 902-910.

Walker, W. (2011) Hermeneutic Inquiry: Insights into the process of interviewing. *Nurse Researcher*, 18 (2) 19-27.

Walker, J. et al. 2014. Prevalence, associations, and adequacy of treatment of major depression in patients with cancer: a cross-sectional analysis of routinely collected clinical data. *The Lancet Psychiatry*, 1 (5) 343-350.

Wasteson, E. et al. 2011. High satisfaction rate ten years after bilateral prophylactic mastectomy—a longitudinal study. *European Journal of Cancer Care*, 20 (4) 508-513.

Watson, M. et al. 1999. The impact of genetic counselling on risk perception and mental health in women with a family history of breast cancer. *British Journal of Cancer*, 79, 868-874.

Watson, M. et al. 2004. Psychosocial impact of breast/ovarian (BRCA1 /2) cancer-predictive genetic testing in a UK multi-centre clinical cohort. *British Journal of Cancer*, 91 (10) 1787-1794.

Watts, L. et al. 2009. Managing self in role: Using multiple methodologies to explore self-construction and self-governance. *Researching beneath the surface. Psycho-social research methods in practice*, pp. 215-239.

Weinsheimer, J. 1985. *Gadamer's Hermeneutics A Reading of Truth and Method*. New Haven: Yale University Press.

Weitz, R. 2004 *The Sociology of health , illness and health care .A critical approach*(3<sup>rd</sup> edn) Belmont:Wadsworth

Werner-Lin, A. 2008. Beating the biological clock: The compressed family life cycle of young women with BRCA gene alterations. *Journal of Social Work in Healthcare* 47 (4) 416-437.

- Werner-Lin, A. et al. 2012. 'Cancer doesn't have an age': Genetic testing and cancer risk management in BRCA1/2 mutation-positive women aged 18–24. *Health (London)*, 16 (6) 636-654.
- Wilcke, M. M. 2002. Hermeneutic phenomenology as a research method in social work. *Currents: New Scholarship in the Human Services*, 1 (1) 1-10.
- Wilkinson, I. 2006. Health, risk and 'social suffering'. *Health, Risk and Society*, (1) 1-8.
- Wimberly, S.R. et al. 2005. Perceived partner reactions to diagnosis and treatment of breast cancer: impact on psychosocial and psychosexual adjustment. *Journal of consulting and clinical psychology*, 73 (2) 300.
- Whitehead, I. 2004. Enhancing the quality of hermeneutic research: decision trail. *Journal of Advanced Nursing*, 45, 512-518.
- Williams, J. et al 2000. Psychosocial impact of predictive testing on Huntington disease on support persons. *American Journal of medical Genetics* 96 (3) 353-259.
- Wonderlick, A.L. and Fine, B.A. 1997. Knowledge of breast cancer patients amongst breast cancer patients and first-degree relatives of affected individuals. *Journal of Genetic Counseling*. 6 (2) 111-130.
- Wooster, R. et al. 1995. Identification of the breast susceptibility gene BRCA2. *Nature*, 378, 798-791.
- Wylie, J. E. et al. 2003. Eds. *Effects of spouses on distress experienced by BRCA1 mutation carriers over time*. American Journal of Medical Genetics Part C: Seminars in Medical Genetics. Wiley Online Library.
- Yao, K. et al. 2015. Nipple-sparing mastectomy in BRCA1/2 mutation carriers: an interim analysis and review of the literature. *Annals of Surgical Oncology* 22 (2) 370-376.
- Young, I. 1990 *Throwing like a girl and other essays in feminist philosophy and Social Theory*. Indiana University press
- Young, I. 1998 *Breasted Experience: the look and the feeling*. In R Weitz (ed), *The Politics of Women's Bodies*. Oxford University Press
- Zakaria, S. and Degnium, A. 2007. Prophylactic Mastectomy. *Surgical Clinics of North America*, 87 (2) 317-331.

Ziegler, L.D. and Kroll, S.S. 1991. Primary breast cancer after prophylactic mastectomy. *American Journal of Clinical Oncology* 14 (5) 451.

Zion, S.M. et al. 2003. Reoperations after prophylactic mastectomy with or without implant reconstruction. *Cancer*, 98 (10) 2152-2160.

Zinn, J. 2005 .The biographical approach. A better way to understand behaviour in health and illness. *Health Risk and Society* 7 (1) 1-9

## **APPENDIX ONE - A Short Autobiography**

This study is about women and their family with the Breast Cancer Susceptibility gene (BRCA1/2). They do not yet have cancer but are at a very high risk. My personal experience of risk and family life as well as my professional life as a nurse, have therefore in some part contributed to the motivation to conduct and continue with this study. As you begin to read this short autobiography therefore, my hope is that you get a feeling of the many factors that influenced my decision to conduct the study and why it was this particular group of participants.

Even as a small child I knew, I wanted to be a nurse. I am the eldest of five children and always took charge of them when growing up. My real father disappeared soon after I was born and I had the luxury of having my mother all to myself until the age of two. In the 1960's it was frowned upon to have a child whilst not being married and so my mother met my new father when I was two. After they married, they went on to have four more children so I have one brother and three sisters. I have always regarded them and referred to them as my siblings, not half-sister or brother. The importance of family life and normality therefore have always been a high priority in my life in maintaining a strong family unit. Even now, my family turn to me for advice and support because we lost our mother at the age of fifty-three and my father was an alcoholic, times were very tough. I cared for and nurtured my siblings alongside my mother until I started my nurse training in 1982.

Nursing is therefore a career I have embraced, adored and have never regretted. I have worked my way up the career ladder from staff nurse to ward sister, clinical nurse specialist and lecturer and I am very proud and privileged to be the only Consultant Nurse in Breast Care in Wales. Until October 2014 and during the time of my study, I was proud to claim an exemplary sick record with no sick time at all! However, during this study, I had major abdominal surgery where I underwent total abdominal hysterectomy, bilateral oophorectomy with removal of my omentum. I had to take three months off, and it was at this time that the PhD went on hold because the future was uncertain. The surgery was necessary as I had a large tumour in my womb which was indeterminate and with a strong family history of cancer, prevention was better than cure. At the same time as my surgery, I was told that I had possible liver secondaries. This was a terrible time in my life and my only concern was for my ten-year-old daughter. Sadly, there was no information or psychological care offered to my immediate family or myself because the surgery was seen as preventative.

Thankfully and eventually, after many weeks of deliberation and extreme anxiety, the histology and the results confirmed that it was a large benign tumour and that the liver lesions must be 'something else'. They apologised. However, I now have yearly MRI scans to monitor the lesions. The uncertainty and the constant worry, I have learnt to manage. The liver is rather a large organ to remove and so this was never a viable option for me! My counselling

background has no doubt helped me accept and deal with the ongoing uncertainty.

Along this journey of conducting my study, I have lost a few relatives and many young friends with a diagnosis of cancer. In my role as Consultant Nurse and conducting my own nurse led clinics, I am constantly breaking bad news to individuals whether it be early breast cancer or metastatic (non-curable) disease. An opportunity to study a different group of individuals was both refreshing and exciting because there existed a group of patients who I felt were marginalised and as they did not have cancer and there was no nursing care pathway. Important factors that motivated me to conduct this study.

The biggest loss in my life however has been my dear mum who was a fit, energetic, motivated and extremely resilient woman who sadly in 2000, developed both ovarian cancer and a separate bowel cancer and died at the age of 56. Alongside my siblings, I nursed her until her death eighteen months after her diagnosis.

Thus, many other motivations led to my decision to conduct this study. My experience as a specialist nurse in breast disease, my own personal experience of surgery, losing my mum at such a young age to cancer and constantly breaking bad news to individuals with cancer were just a few. The importance and complexities of family life and valuing a stable unit, in addition to wanting to set up a service for BRCA positive women and their family, were

also vital factors that contributed to my desire to conduct this study. I witnessed for myself the deep loss of a family member to cancer and the experience of undergoing major surgery to prevent getting cancer and dying of the disease. The importance of a solid family network and meeting the needs of the whole family was ever apparent and sadly lacking. In addition, having the bilateral salpingo-oophorectomy has been no easy journey especially whilst trying to write a PhD thesis and thus many of the findings in my study, I can personally relate to. Writing this short autobiography also fulfils aspects of the 'expressions of rigour' as identified by de Witt and Ploeg (2006), openness, concreteness and resonance.

## APPENDIX TWO - NICE (2013) Guidelines

### NICE (2013) Guidelines for referral to cancer genetics and secondary care

Referral from primary care

1.3.3 People without a personal history of breast cancer who meet the following criteria should be offered referral to secondary care:

- one first-degree female relative diagnosed with breast cancer at younger than age 40 years **or**
- one first-degree male relative diagnosed with breast cancer at any age **or**
- one first-degree relative with bilateral breast cancer where the first primary was diagnosed at younger than age 50 years **or**
- two first-degree relatives, or one first-degree **and** one second-degree relative, diagnosed with breast cancer at any age **or**
- one first-degree or second-degree relative diagnosed with breast cancer at any age **and** one first-degree or second-degree relative diagnosed with ovarian cancer at any age (one of these should be a first-degree relative) **or**
- three first-degree or second-degree relatives diagnosed with breast cancer at any age. **[2004]**

1.3.4 Advice should be sought from the designated secondary care contact if any of the following are present in the family history in addition to breast cancers in relatives not fulfilling the above criteria:

- bilateral breast cancer
- male breast cancer
- ovarian cancer
- Jewish ancestry
- sarcoma in a relative younger than age 45 years
- glioma or childhood adrenal cortical carcinomas
- complicated patterns of multiple cancers at a young age
- paternal history of breast cancer (two or more relatives on the father's side of the family). **[2004]**

1.3.5 Discussion with the designated secondary care contact should take place if the primary care health professional is uncertain about the appropriateness of referral because the family history presented is unusual or difficult to make clear decisions about, or where the person is not sufficiently reassured by the standard information provided. **[2004]**

1.3.6 Direct referral to a specialist genetics service should take place where a high-risk predisposing gene mutation has been identified (for example, *BRCA1*, *BRCA2* or *TP53*). **[2004]**



## Patient education and information

### Referral to a specialist genetic clinic

1.4.4 People who meet the following referral criteria should be offered a referral to a specialist genetic clinic.

- At least the following female breast cancers only in the family:
  - two first-degree or second-degree relatives diagnosed with breast cancer at younger than an average age of 50 years (at least one must be a first-degree relative) **[2004] or**
  - three first-degree or second-degree relatives diagnosed with breast cancer at younger than an average age of 60 years (at least one must be a first-degree relative) **[2004] or**
  - four relatives diagnosed with breast cancer at any age (at least one must be a first-degree relative). **[2004] or**
- Families containing one relative with ovarian cancer at any age **and**, on the same side of the family:
  - one first-degree relative (including the relative with ovarian cancer) or second-degree relative diagnosed with breast cancer at younger than age 50 years **[2004] or**
  - two first-degree or second-degree relatives diagnosed with breast cancer at younger than an average age of 60 years **[2004] or**
  - another ovarian cancer at any age. **[2004] or**
- Families affected by bilateral cancer (each breast cancer has the same count value as one relative):
  - one first-degree relative with cancer diagnosed in both breasts at younger than an average age 50 years **[2004] or**
  - one first-degree or second-degree relative diagnosed with bilateral cancer and one first or second degree relative diagnosed with breast cancer at younger than an average age of 60 years. **[2004] or**
- Families containing male breast cancer at any age **and**, on the same side of the family, at least:
  - one first-degree or second-degree relative diagnosed with breast cancer at younger than age 50 years **[2004] or**
  - two first-degree or second-degree relatives diagnosed with breast cancer at younger than an average age of 60 years. **[2004] or**
- A formal risk assessment has given risk estimates of:
  - a 10% or greater chance of a gene mutation being harboured in the family (see [recommendations 1.5.8–1.5.13](#)) **[new 2013] or**
  - a greater than 8% risk of developing breast cancer in the next 10 years **[2004] or**
  - a 30% or greater lifetime risk of developing breast cancer. **[2004]**

1.4.5 Clinicians should seek further advice from a specialist genetics service for families containing any of the following, in addition to breast cancers:

- [triple negative breast cancer](#) under the age of 40 years **[new 2013]**
- Jewish ancestry **[2004]**
- sarcoma in a relative younger than age 45 years **[2004]**

- glioma or childhood adrenal cortical carcinomas [2004]
- complicated patterns of multiple cancers at a young age [2004]
- very strong paternal history (four relatives diagnosed at younger than 60 years of age on the father's side of the family). [2004]

1.4.6 The management of high-risk people may take place in secondary care if they do not want genetic testing or risk-reducing surgery and do not wish to be referred to a specialist genetics service. [2004]

1.4.7 Following initial consultation in secondary care, [written information](#) should be provided to reflect the outcomes of the consultation. [2004]

### **Genetic counselling for people with no personal history of breast cancer**

1.4.9 Women with no personal history of breast cancer meeting criteria for referral to a specialist genetic clinic should be offered a referral for genetic counselling regarding their risks and options. [2004]

1.4.10 Women attending genetic counselling should receive standardised information beforehand describing the process of genetic counselling, information to obtain prior to the counselling session, the range of topics to be covered and brief educational material about hereditary breast cancer and genetic testing. [2004]

1.4.11 Predictive genetic testing should not be offered without adequate genetic counselling. [2004]

### **1.5 Genetic testing**

1.5.1 All eligible people should have access to information on genetic tests aimed at mutation finding. [2004]

1.5.2 Pre-test counselling (preferably two sessions) should be undertaken. [2004]

1.5.3 Discussion of genetic testing (predictive and mutation finding) should be undertaken by a healthcare professional with appropriate training. [2004]

1.5.4 Eligible people and their affected relatives should be informed about the likely informativeness of the test (the meaning of a positive and a negative test) and the likely timescale of being given the results. [2004]

### **Mutation tests**

1.5.5 Tests aimed at mutation finding should first be carried out on an affected family member where possible. [2004]

1.5.6 If possible, the development of a genetic test for a family should usually start with the testing of an affected individual (mutation searching/screening) to try to

identify a mutation in the appropriate gene (such as *BRCA1*, *BRCA2* or *TP53*) (see [recommendations 1.5.8–1.5.13](#)). [2004]

1.5.7 A search/screen for a mutation in a gene (such as *BRCA1*, *BRCA2* or *TP53*) should aim for as close to 100% sensitivity as possible for detecting coding alterations and the whole gene(s) should be searched. [2004]

#### **Carrier probability at which genetic testing should be offered**

1.5.8 Discuss the potential risk and benefits of genetic testing. Include in the discussion the probability of finding a mutation, the implications for the individual and the family, and the implications of either a variant of uncertain significance or a null result (no mutation found). [new 2013]

1.5.9 Inform families with no clear genetic diagnosis that they can request review in the specialist genetic clinic at a future date. [new 2013]

1.5.10 Clinical genetics laboratories should record gene variants of uncertain significance and known pathogenic mutations in a searchable electronic database. [new 2013]

#### **Genetic testing for a person with no personal history of breast cancer but with an available affected relative**

1.5.11 Offer genetic testing in specialist genetic clinics to a relative with a personal history of breast and/or ovarian cancer if that relative has a combined *BRCA1* and *BRCA2* mutation carrier probability of 10% or more. [new 2013]

#### **Genetic testing for a person with no personal history of breast cancer and no available affected relative to test**

1.5.12 Offer genetic testing in specialist genetic clinics to a person with no personal history of breast or ovarian cancer if their combined *BRCA1* and *BRCA2* mutation carrier probability is 10% or more and an affected relative is unavailable for testing. [new 2013]

#### **Genetic testing for a person with breast or ovarian cancer**

1.5.13 Offer genetic testing in specialist genetic clinics to a person with breast or ovarian cancer if their combined *BRCA1* and *BRCA2* mutation carrier probability is 10% or more. [new 2013]

#### **Genetic testing for *BRCA1*, *BRCA2* and *TP53* mutations within 4 weeks of diagnosis of breast cancer**

1.5.14 Offer people eligible for referral to a specialist genetic clinic a choice of accessing genetic testing during initial management or at any time thereafter. [new 2013]

1.5.15 Offer fast-track genetic testing (within 4 weeks of a diagnosis of breast cancer) only as part of a clinical trial. **[new 2013]**

1.5.16 Discuss the individual needs of the person with the specialist genetics team as part of the multidisciplinary approach to care. **[new 2013]**

1.5.17 Offer detailed consultation with a clinical geneticist or genetics counsellor to all those with breast cancer who are offered genetic testing, regardless of the timeframe for testing. **[new 2013]**

### **Surveillance for women with no personal history of breast cancer but with a high risk or BRCA gene**

#### **Mammographic surveillance**

1.6.3 Offer annual mammographic surveillance to women:

- aged 40–49 years at [moderate risk](#) of breast cancer
- aged 40–59 years at [high risk](#) of breast cancer but with a 30% or lower probability of being a *BRCA* or *TP53* carrier
- aged 40–59 years who have not had genetic testing but have a greater than 30% probability of being a *BRCA* carrier
- aged 40–69 years with a known *BRCA1* or *BRCA2* mutation. **[new 2013]**

1.6.4 Offer mammographic surveillance as part of the population screening programme to women:

- aged 50 years and over who have not had genetic testing but have a greater than 30% probability of being a *TP53* carrier
- aged 60 years and over at [high risk](#) of breast cancer but with a 30% or lower probability of being a *BRCA* or *TP53* carrier
- aged 60 years and over at [moderate risk](#) of breast cancer
- aged 60 years and over who have not had genetic testing but have a greater than 30% probability of being a *BRCA* carrier
- aged 70 years and over with a known *BRCA1* or *BRCA2* mutation. **[new 2013]**

1.6.5 Consider annual mammographic surveillance for women:

- aged 30–39 years at [high risk](#) of breast cancer but with a 30% or lower probability of being a *BRCA* or *TP53* carrier
- aged 30–39 years who have not had genetic testing but have a greater than 30% probability of being a *BRCA* carrier
- aged 30–39 years with a known *BRCA1* or *BRCA2* mutation
- aged 50–59 years at [moderate risk](#) of breast cancer. **[new 2013]**

1.6.6 Do not offer mammographic surveillance to women:

- aged 29 years and under
- aged 30–39 years at [moderate risk](#) of breast cancer

- aged 30–49 years who have not had genetic testing but have a greater than 30% probability of being a *TP53* carrier
- of any age with a known *TP53* mutation. **[new 2013]**

### **MRI surveillance**

1.6.7 Offer annual MRI surveillance to women:

- aged 30–49 years who have not had genetic testing but have a greater than 30% probability of being a *BRCA* carrier
- aged 30–49 years with a known *BRCA1* or *BRCA2* mutation
- aged 20–49 years who have not had genetic testing but have a greater than 30% probability of being a *TP53* carrier
- aged 20–49 years with a known *TP53* mutation. **[new 2013]**

1.6.8 Consider annual MRI surveillance for women aged 50–69 years with a known *TP53* mutation. **[new 2013]**

1.6.9 Do not offer MRI to women:

- of any age at [moderate risk](#) of breast cancer
- of any age at [high risk](#) of breast cancer but with a 30% or lower probability of being a *BRCA* or *TP53* carrier
- aged 20–29 years who have not had genetic testing but have a greater than 30% probability of being a *BRCA* carrier
- aged 20–29 years with a known *BRCA1* or *BRCA2* mutation
- aged 50–69 years who have not had genetic testing but have a greater than 30% probability of being a *BRCA* or a *TP53* carrier, unless mammography has shown a dense breast pattern
- aged 50–69 years with a known *BRCA1* or *BRCA2* mutation, unless mammography has shown a dense breast pattern. **[new 2013]**

### **MRI surveillance**

1.6.13 Offer annual MRI surveillance to all women aged 30–49 years with a personal history of breast cancer who remain at [high risk](#) of breast cancer, including those who have a *BRCA1* or *BRCA2* mutation. **[new 2013]**

1.6.14 Do not offer MRI surveillance to any women aged 50 years and over without a *TP53* mutation unless mammography has shown a dense breast pattern. **[new 2013]**

1.6.15 Consider annual MRI surveillance for women aged 20–69 years with a known *TP53* mutation or who have not had a genetic test but have a greater than 30% probability of being a *TP53* carrier. **[new 2013]**

### **Surveillance for women who remain at moderate risk of breast cancer**

1.6.16 Ensure that surveillance for people with a personal history of breast cancer who remain at [moderate risk](#) of breast cancer is in line with [Early and locally advanced breast cancer](#) (NICE clinical guideline 80). **[new 2013]**

### **Recommendations for all women having surveillance**

1.6.17 Offer support (for example, risk counselling, psychological counselling and risk management advice) to women who have ongoing concerns but are not eligible for surveillance additional to that offered by the national breast screening programmes<sup>[5]</sup>. **[2004, amended 2013]**

1.6.18 Before decisions on surveillance are made, discuss and give written information on the benefits and risks of surveillance, including:

- the possibility that mammography might miss a cancer in women with dense breasts and the increased likelihood of further investigations **[new 2013]**
- possible over diagnosis
- the risk associated with exposure to radiation
- the possible psychological impact of a recall visit. **[2004, amended 2013]**

### **Risk-reducing mastectomy**

1.7.56 Discuss the risks and benefits of risk-reducing mastectomy with women with a known or suspected *BRCA1*, *BRCA2* or *TP53* mutation. **[new 2013]**

1.7.57 For a woman considering risk-reducing mastectomy, include in the discussion of risks and benefits:

- the likely prognosis of their breast cancer, including their risk of developing a distal recurrence of their previous breast cancer
- a clear quantification of the risk of developing breast cancer in the other breast
- the potential negative impact of mastectomy on body image and sexuality
- the very different appearance and feel of the breasts after reconstructive surgery
- the potential benefits of reducing the risk in the other breast and relieving the anxiety about developing breast cancer. **[new 2013]**

1.7.58 Give all women considering a risk-reducing mastectomy the opportunity to discuss their options for breast reconstruction (immediate and delayed) with a member of a surgical team with specialist skills in oncoplastic surgery or breast reconstruction. **[new 2013]**

1.7.59 Ensure that risk-reducing mastectomy and breast reconstruction are carried out by a surgical team with specialist skills in oncoplastic surgery and breast reconstruction. **[new 2013]**

1.7.60 Offer women who have *BRCA1*, *BRCA2* or *TP53* mutations but who decide against risk-reducing mastectomy, surveillance according to their level of risk. **[new 2013]**

## **Risk-reducing bilateral salpingo-oophorectomy**

1.7.61 Discuss the risks and benefits of risk-reducing bilateral salpingo-oophorectomy with women with a known or suspected *BRCA1*, *BRCA2* or *TP53* mutation. Include in the discussion the positive effects of reducing the risk of breast and ovarian cancer and the negative effects of a surgically induced menopause. **[new 2013]**

1.7.62 Defer risk-reducing bilateral salpingo-oophorectomy until women have completed their family. **[new 2013]**

## **Risk-reducing mastectomy for women with no personal history of breast cancer**

1.7.30 Bilateral risk-reducing mastectomy is appropriate only for a small proportion of women who are from high-risk families and should be managed by a multidisciplinary team. **[2004]**

1.7.31 Bilateral mastectomy should be raised as a risk-reducing strategy option with all women at high risk. **[2004]**

1.7.32 Women considering bilateral risk-reducing mastectomy should have genetic counselling in a specialist cancer genetic clinic before a decision is made. **[2004]**

1.7.33 Discussion of individual breast cancer risk and its potential reduction by surgery should take place and take into account individual risk factors, including the woman's current age (especially at extremes of age ranges). **[2004]**

1.7.34 Family history should be verified where no mutation has been identified before bilateral risk-reducing mastectomy. **[2004]**

1.7.35 Where no family history verification is possible, agreement by a multidisciplinary team should be sought before proceeding with bilateral risk-reducing mastectomy. **[2004]**

1.7.36 Pre-operative counselling about psychosocial and sexual consequences of bilateral risk-reducing mastectomy should be undertaken. **[2004]**

1.7.37 The possibility of breast cancer being diagnosed histologically following a risk-reducing mastectomy should be discussed pre-operatively. **[2004]**

1.7.38 All women considering bilateral risk-reducing mastectomy should be able to discuss their breast reconstruction options (immediate and delayed) with a member of a surgical team with specialist oncoplastic or breast reconstructive skills. **[2004]**

1.7.39A surgical team with specialist oncoplastic/breast reconstructive skills should carry out risk-reducing mastectomy and/or reconstruction. **[2004]**

1.7.40 Women considering bilateral risk-reducing mastectomy should be offered access to support groups and/or women who have undergone the procedure. **[2004]**



## APPENDIX THREE - Literature Review Search Strategy

Terms Used for Search Strategy
--------------------------------

BRCA1 and 2	Mastectomy	Risk Reduction	Relatives	Experience
		Risk adj3 reduct*	Spouses	Phenomonol*
	Prophylactic mastectomy	Risk Reduction Behaviour	Family relations	Qualitative Research
			Husband	
			Partner	
			Family	
			Proband	

# ▲	Searches	Results
1	*Mastectomy/	7947
2	Mastect*.ti,ab.	16193
3	prophylactic mastectomy.ti,ab.	701
4	exp Genes, BRCA1/ or exp Genes, BRCA2/	5815
5	exp Risk Reduction Behavior/	9873
6	(risk adj3 reduct).ti,ab.	2
7	exp Spouses/	8213
8	family relations/	9417
9	husband.ti,ab.	4714
10	exp Family/	271020
11	relative.ti,ab.	606276
12	partner.ti,ab.	50009
13	proband.ti,ab.	8149
14	exp Qualitative Research/	29920
15	phenomen.ti,ab.	125
16	experience.ti,ab.	466380
17	or/1-3	19650
18	4 and 17	349
19	5 or 6	9875
20	18 and 19	30
21	or/7-13	919961
22	or/14-16	489674
23	20 and 21 and 22	0
24	or/1-4	25116
25	19 and 24	98
26	21 and 24	1240
27	22 and 26	79

## TABLE OF INCLUDED STUDIES

Author	Topic	Method / Sample	Main Findings
Altschuler et al (2008) American Study	Long term Psychosocial outcomes of bilateral or contralateral prophylactic mastectomy	Retrospective Qualitative open ended Mailed Survey 3-22 years post-surgery 967 Sample (195 RRM) (772 contra-lateral mx) No BRCA positive women.	Negative impact on body image, sexuality and dissatisfaction with surgery in women having RRM.
Brandberg et al (2008) Swedish study	Evaluate Body image, sexuality and emotional reactions after RRM	Quantitative Prospective Questionnaire Sample 90 women	Negative impact on body image and sexuality for BRCA positive women(50)
Brandberg et al (2012) Swedish Study	Cosmetic results and expectations post RRM and reconstruction in BRCA positive women	Quantitative retrospective questionnaire Sample 91 (49 BRCA Positive)	Higher expectations in BRCA women not met post-surgery. Satisfaction with overall cosmetic result varied

Bresser et al (2006) The Netherlands	Satisfaction with Prophylactic mastectomy and reconstruction in genetically predisposed women	Retrospective questionnaire design with 114 women 3 years post surgery.63 BRCA positive.	Adverse effects in sexual relationships and altered feelings of femininity found Included women with breast cancer electing prophylactic mastectomy
Den Heijer et al (2012) The Netherlands	Body image and psychological distress after prophylactic Mastectomy and reconstruction in genetically pre-disposed women	Prospective quantitative long term study using Questionnaires Sample 36 women (27 BRCA positive)	Psychological distress about cancer reduced. Body image problems evident long term( 9 years)
Frost et al (2000) American Study	Long term quality of life and satisfaction following bilateral prophylactic mastectomy	Quantitative Retrospective study 14 years post-surgery Sample 572 women (None BRCA)	Reduced concern about cancer. Most women were satisfied with surgery. However, 18% would not choose surgery again. Problems found with sexuality, body image and self-esteem.

Geiger et al (2007) American Study	Long-term Quality of life following prophylactic mastectomy	Retrospective Quantitative Questionnaire with a sample of 312 Women. 195 had procedure, 117 did not (none BRCA) 23 years post-surgery.	Bilateral prophylactic mastectomy neither positively nor negatively impacts long-term psychosocial outcomes. Dissatisfaction with quality of life was associated with pre- surgery sex life.
Gopie et al (2013) The Netherlands	Body image issues following prophylactic mastectomy and reconstruction	Prospective Quantitative study with questionnaires Sample size 48 (44 BRCA)	Cancer related distress lower. Body image scores decreased with continuing problems.
Hatcher and Fallowfield (2003) United Kingdom	Psychosocial implications of bilateral prophylactic mastectomy	Qualitative prospective study with in depth interviews. Sample 80 Women (60 surgery) 20 (declined) (8 BRCA positive)	Pain, anxiety and mixed satisfaction with cosmetic results and sexuality post-surgery.
Hatcher et al (2001) United Kingdom	Psychological and sexually morbidity	Prospective Quantitative study using questionnaires.	High levels of anxiety persisted in non - surgical group.

		Sample 143 women(79 surgery) (64 declined)	No significant change in sexual pleasure post-surgery
Hallowell et al (2012) Australia	Women's reactions to risk-reducing and ovarian surgery	Qualitative retrospective study with a sample of 40 women.	Risk and relief of cancer reduced. Positive and negative changes to body. More preparation and psychosocial input required
Hopwood et al (2000) United Kingdom	Body image and Clinical follow up after prophylactic mastectomy	Retrospective Quantitative study using questionnaires. Sample 76 women (6 BRCA)	Contradictory findings. No significant body image problems. Reduced feelings of sexual attractiveness, problems with self-consciousness and feeling less feminine. Minority had serious body image and psychological problems
Lloyd et al (2000) United Kingdom	Understanding the experience of Prophylactic bilateral mastectomy	Qualitative Grounded Theory retrospective study. Sample 10 women(2 BRCA	Findings demonstrated a core category of 'Suffering and countering multiple loss'.Including family

		positive) and 8 men.	member losses and loss of 'womanliness.' Sexual and image problems.
McEwan (2011)	Experiences of New Zealand Women living with increased risk of breast and Ovarian cancer	Qualitative study interviews with 32 women, 6 had breast cancer. 14 BRCA-4 RRM.	Women at increased risk are prepared to undergo risk-reducing surgery to 'get on with it' for the sake of children. RRM affected femininity and loss experienced. Adverse reactions from friends re RRM
Mauer et al (2015) American Study	Exploration of male attitudes of partnerships and sexuality with BRCA Women	Retrospective online survey. Sample 25 men	Intimacy and communication problems after disclosure and surgery
Mireskandari et al (2006)	Evaluation of needs of partners of high risk women	Qualitative retrospective study. Sample 15 men telephone interviews (7 of their partners BRCA positive) 1 had RRM	Main findings, partners of BRCA women more distressed. Concerns about the decision to have RRM and children were most challenging.

Metcalfe et al (2005) Canada	Predictors of quality of life in women following bilateral prophylactic mastectomy	Retrospective Quantitative questionnaire. Sample 60 women (13 BRCA)	Predictors of QOL related to psychological distress and vulnerability for body image problems
Metcalfe et al (2004a) Canada	Psychosocial functioning in women following prophylactic mastectomy	Retrospective Quantitative Questionnaire. Sample 75 women (60 returned (13 BRCA)	Most women were happy with their decision of surgery (with reconstruction) although younger women less so. No significant body image or sexual problems. Cancer related distress still high post-surgery for BRCA women.
Payne et al (2000) American Study	Regret following bilateral prophylactic mastectomy	Retrospective Quantitative questionnaire with 21 women (none BRCA Positive)	Physical and Psychological trauma of surgery and lack of support reasons for regret. Surgery initiated by physician and not the women.
van Oostrum et al (2003) The Netherlands	Long term psychological impact of carrying the mutation and	Mixed method using questionnaires and interviews. Sample 65	Reduced fear in getting cancer. BRCA Women had less favourable body image and sexuality



	having surgery.	women.23 BRCA positive and 21 had surgery.	scores. 70% reported sexual difficulties
Wasteson et al (2011) Swedish Study	Long term satisfaction following bilateral prophylactic mastectomy	Retrospective Qualitative study using interviews. Sample 13 women (1 BRCA positive)	High Satisfaction with decision to have surgery and overall satisfied with cosmetic result. Some family problems identified.

## **APPENDIX FOUR - Publication and Dissemination Plan**

When undertaking a PhD, it is imperative that the knowledge gained from the findings are disseminated and accessible for professionals both academic and clinical. I have been involved in strategic level work and service development for over twenty years and I have made close links with colleagues at a national and international level. I am also a nurse advisor for The National Institute of Health and Care Excellence (NICE). I have consistently been invited to speak at conferences nationally and internationally, meeting inspirational nurses from all over the world. One example of my collaborative work is that I am Principal Investigator on a multicentre national study involving women newly diagnosed with breast cancer seeking breast reconstruction and a scientific committee member for the European Breast Cancer Conference in Barcelona.

It is my intention to publish my research in relevant, targeted journals that will reach clinicians and members of the multidisciplinary team nationally and internationally, an example of one of my publications

West, N. (2014) New approaches to managing risk of familial breast cancer, *Primary Care Women's Health Journal*. May 2014.

### **Suggested Articles and Target Journals**

- **Article 1** - The experiences of BRCA women electing to have risk-reducing mastectomy. An Interpretive Phenomenological study.

- **Target Journal** - The Breast Journal/Journal of Advanced Nursing/Psych-oncology/ Journal of surgical Oncology.
- **Article 2** - The experiences of partners whose wives elect to have risk-reducing mastectomy due to the BRCA gene.
- **Target Journal** - International Journal of Social Research/Journal of Advanced Nursing/The Breast/Journal of Surgical Oncology.
- **Article 3** - The experiences of relatives living with the BRCA Gene mutation.
- **Target Journal** - International Journal of Social Research/ Journal of Advanced Nursing
- **Article 4** - The trials and tribulations of doing a PhD part time.
- **Target Journal** - Journal of Advanced Nursing
- **Article 5** - Gadamerian Philosophy and its contribution to qualitative nursing research
- **Target Journal** - Qualitative research/International Journal of Social Research

### **Further Dissemination**

All the participants in the study will be given an overview of the findings personally. Information will be disseminated at the following local meetings:

1. The All Wales Breast Cancer Conference and Network.
2. The Florence Nightingale Foundation.
3. The Cardiff and Vale University Health Board Research Conference.

4. Cardiff University.
5. Breast Cancer Care Network.
6. **Internationally** - 11<sup>th</sup> International Breast Cancer Conference, Barcelona (2018).

## **APPENDIX FIVE - National and International Presentations**

Paris, 2013	4 <sup>TH</sup> International Breast Cancer Conference
Antwerp, 2014	5 <sup>th</sup> International Breast Cancer Conference
London, 2014	Florence Nightingale Foundation Conference Poster Presentation
Lisbon, 2014	Advanced Breast Cancer meeting
Istanbul, 2014	Turkey Breast Cancer Conference
Helsinki, 2014	International Council of Nursing Conference
Bournemouth, 2015	British Association of Surgical Oncologists (BASO) Meeting
Cardiff, 2015	Cardiff and Vale University Health Board Research Conference
Amsterdam, 2015	10 <sup>th</sup> European Breast Cancer Conference
Manchester, 2016	British Association of Surgical Oncologists (BASO) Meeting
Barcelona, 2018	11 <sup>th</sup> European Breast Cancer conference

### **Awards Received**

- Leadership Award at the Welsh Chief Nursing officer Conference, 2012
- Royal College of Nursing Welsh Specialist Nurse of the Year, 2012
- Runner up for Oncology Nurse of the Year, 2013. British Journal of Nursing Award.
- Florence Nightingale Research Scholarship, 2013 (The Band Trust) £1995
- Florence Nightingale Research Scholarship, 2014 (The Garfield Weston Foundation) £1914

- Florence Nightingale Research Scholarship, 2015 (The Stephanie Thompson Memorial Trust) £1500

## **Research 2016**

**PI (PEGASUS STUDY) Patients' Expectations & Goals: Assisting Shared Understanding of Surgery**

## **APPENDIX SIX – Reflections and Diary**

### **Reflection One – April, 2014**

There were a few challenges reflected upon during this study. Being conscious of some of the pre-understandings and being involved with the whole family, I was mindful not to lead the participants during the interviews or pay particular attention to certain issues for example, body image. As a novice interviewer, it was very tempting to lapse into role of counsellor or consultant nurse and try to fix everything. As the interviews continued, I became more confident and said very little, my listening skills intensified allowing all participants to have their 'voice'. I was genuinely interested and open to whatever they wanted to tell me.

My reflective journal was therefore an aid demonstrating how my behaviour had changed over time. The pre-understandings became less conscious and less important as understanding developed. The biggest challenge was the analysis and seemed to take me a lifetime. The challenge has been 'letting go' of the data. I was so immersed in the data and inspired by what they were telling me, despite so many years in practice, that it was extremely difficult to prioritise what was important. To me, it was all very important. However, staying true to Gadamerian philosophy, engaging in the hermeneutic circle and considering the parts that made up the whole, gave me direction to identify the experience that united the three groups and which generated new knowledge. Using the mind maps to organise my data was invaluable

The participants in this study all described how they valued the chance to help me understand their experience, not just for the benefit of themselves but for the benefit of others after them. They were particularly vocal and helpful about how professionals could help in the transition of their altered self by receiving more preparation and ongoing support. There were many points in the interviews when the participants were upset and particularly with the family, I had to stop the recorder and after a short break, recommence the interview, at their request. I felt very privileged that they wanted to talk to me and very grateful. I also noted the trust that they had put in me and the ease at which they discussed very intimate sensitive information.

#### **Extract from Diary July, 2011**

Completing forms, protocol, ethical approval. Really slow on the computer. Need to attend as many study days as possible to catch up with other PhD students. Attending qualitative MSc module social sciences. Amanda Coffey and Paul Atkinson excellent speakers. Met with Amanda Coffey to discuss narrative analysis and methodology as a result of one of the inspiring lectures. Endnote training day 11.11.11. Want to include the husband's and family in the study.

#### **Extract from Diary January, 2012**

Ethical approval granted quickly after a panel meeting in which I was present. Quite daunting before the meeting but very excited to know that I can start the study. GP on the panel concerned about how I will recruit partners and what I would do if they separated from the women. Feel proud to have answered all questions. Coffee with Aimee at Costa to celebrate. Hope there will be enough BRCA women referred. Hope partners will want to



take part. Really keen to know about their experiences. Panel really pleased that partner's and relative's experiences included.

### **Extract from diary March, 2012**

Started interviews. Proband's so honest and forthcoming. Many women wanting to take part. Partners and relatives wanting to take part, so really excited. Emotional interviews. Started to type up interview transcripts, looking at the transcripts and beginning to look for what the experience is all about. So difficult to transcribe. Booked in for numerous study days including working with long documents, rapid reading, NVIVO training. Feel like the oldest student in the class. More interviews booked.

### **Extract from Diary August, 2013**

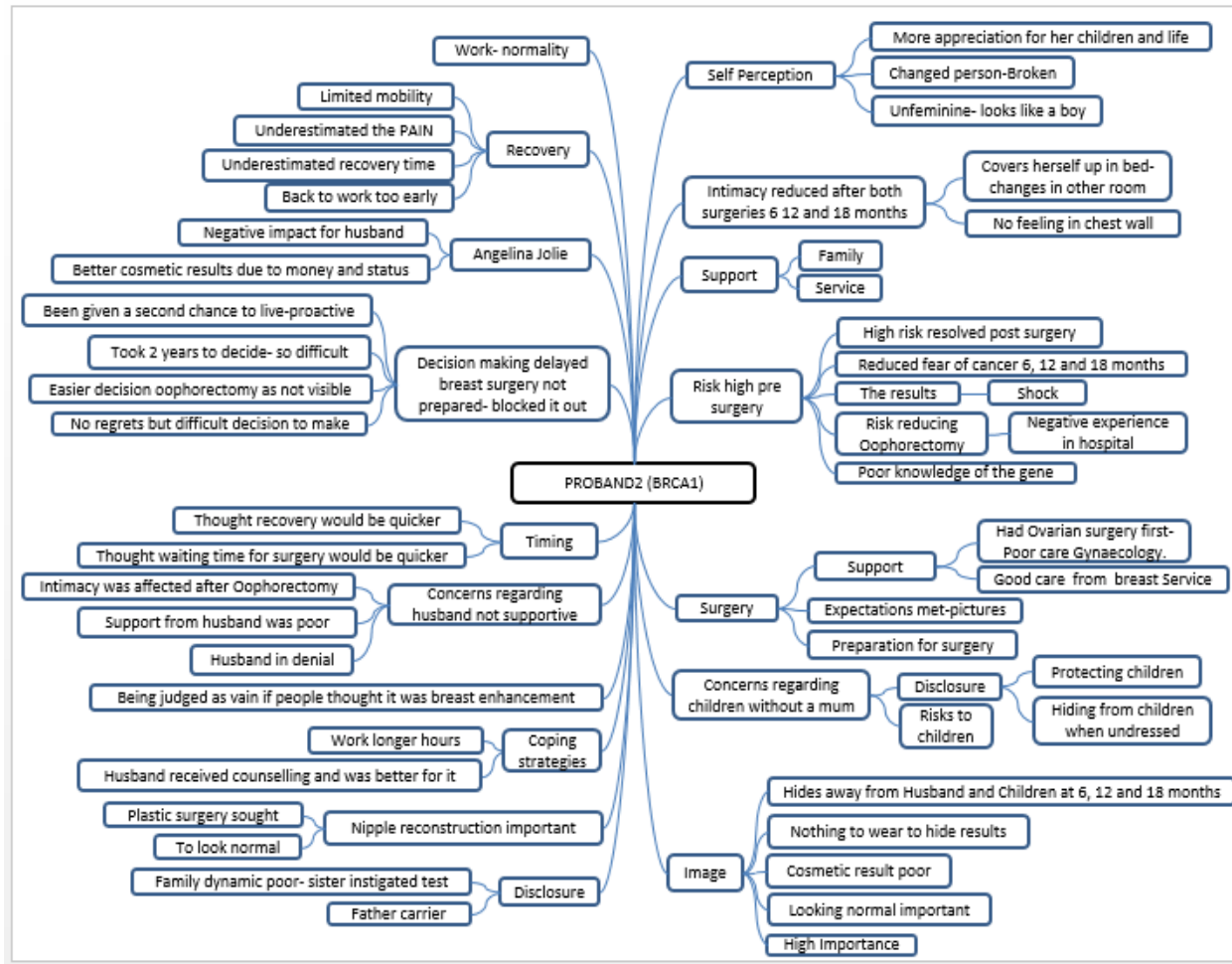
Really confusing trying to understand the difference between Ontology and epistemology. So many articles to read and difference of opinion. Reading too much, have to stop reading and get writing. Supervision session booked to clarify. Had a long conversation and debate with PhD colleague and it's like the blind leading the blind. Useful articles from Jane Harden that really helped to clarify the confusion. So much to learn and read. Ontology is the study of being. I strongly believe that there is not one reality and that reality is subjective but experiences can be shared. So excited to be doing a qualitative study. Gadamer's philosophy and his book 'Truth and method' fits more closely with my methodology and fits with a constructivist approach, although I recognise that it has not been utilised as much in nursing as Husserl's phenomenology. I have come to the conclusion that epistemology is the study of knowledge and asks the question, what is the nature of the relationship between the knower and what can be known. Is knowledge objective and measurable or is it subjective and interpretive. A bond or link exists between the participant's and myself as a nurse because of my experience in breast care and my place as a woman in society.

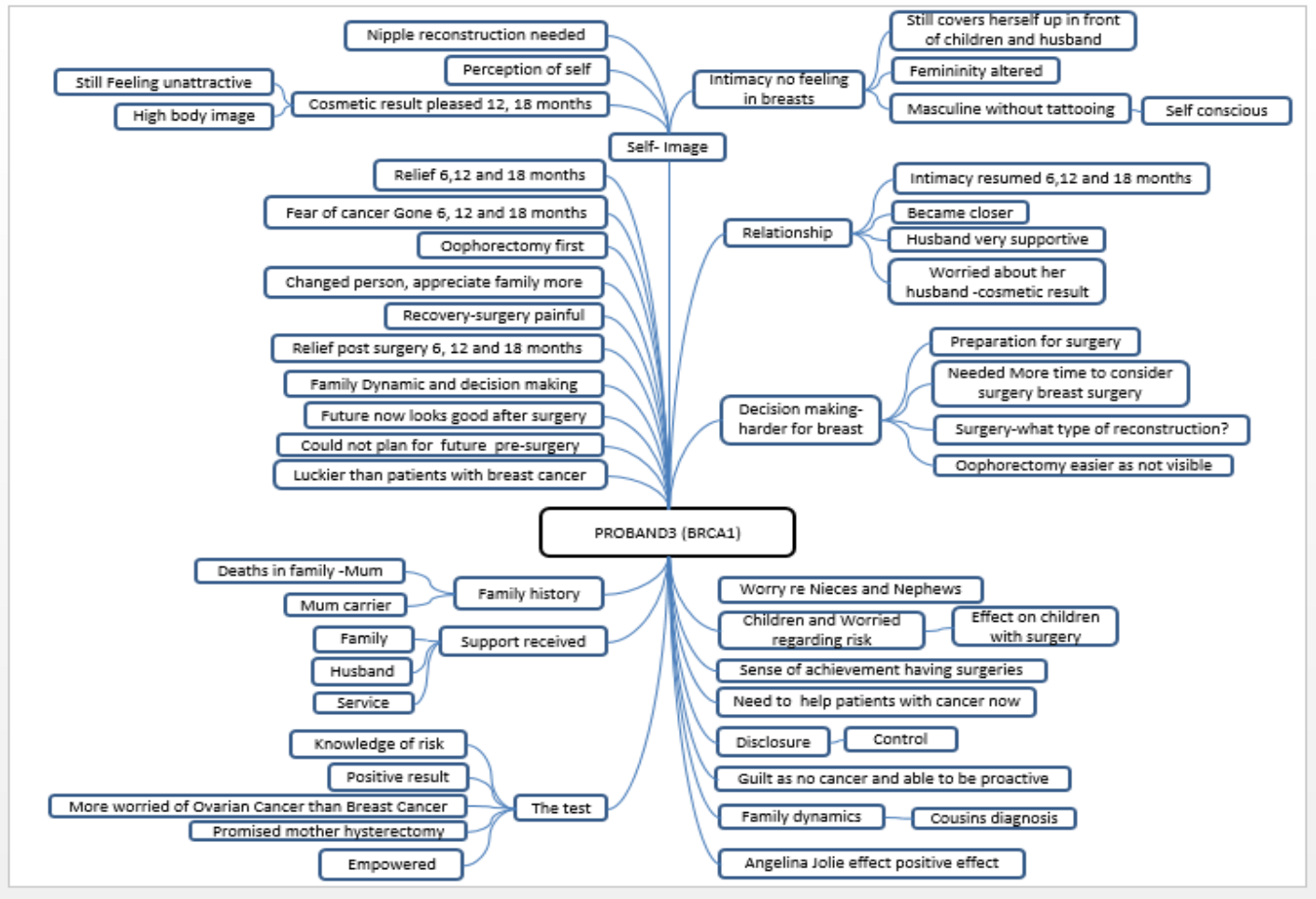
**Extract from Diary January, 2014**

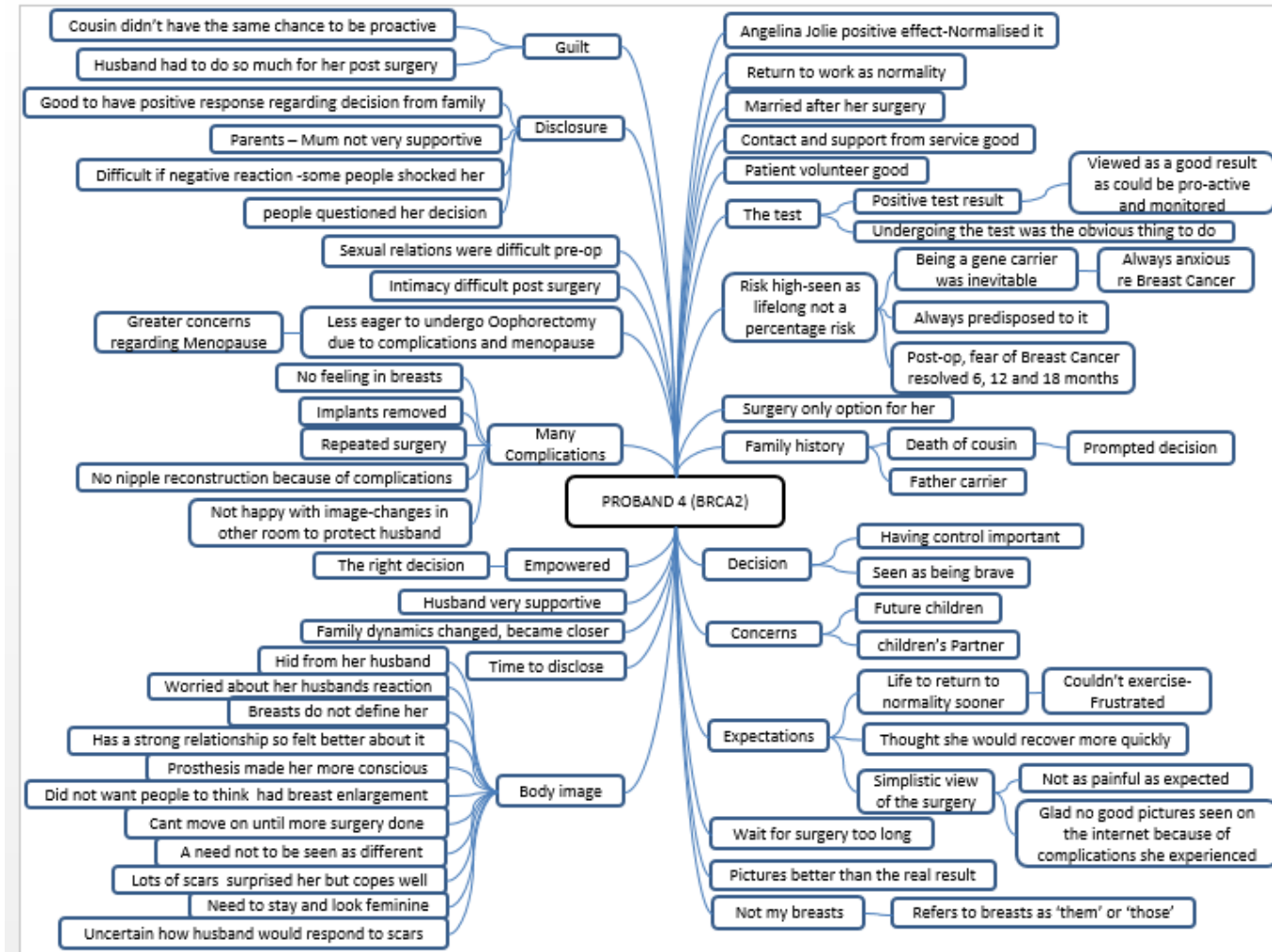
Analysis really difficult, so much that is important, need help with the transcribing. Paying professionally for transcribing. Seeing links between the whole and the parts. The issue of breast loss dominating all the interviews. No drop out from the participants and the interviews continue. Need to start writing. Practice taking over, no study time. Meeting regular with supervisors. Realise the body image scale that I intended to use will not add to the findings and the experience. Feel uncomfortable using it as it implies I am looking for body image changes and so I have decided not to include it. Does not fit with my methodology.

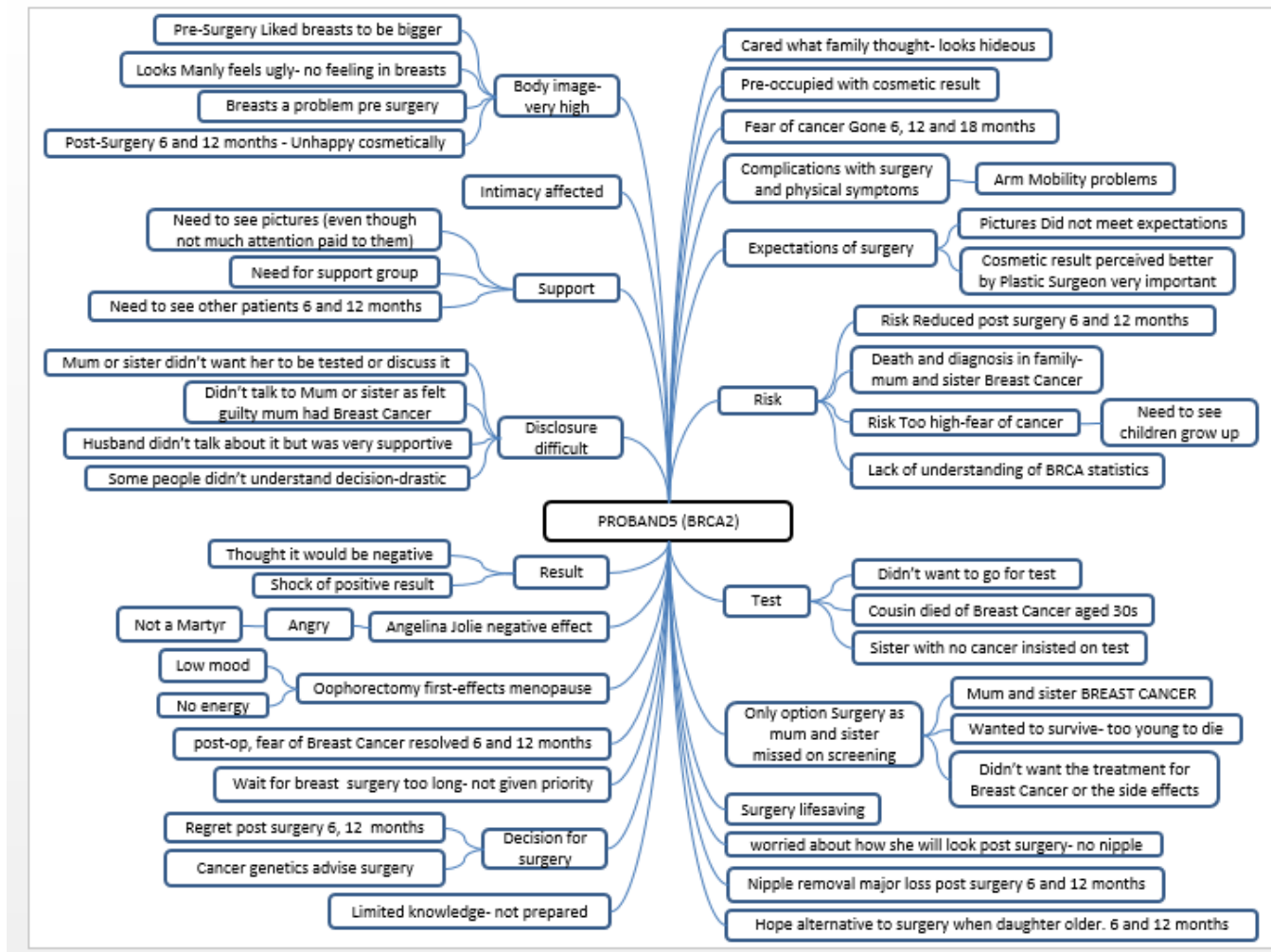
## APPENDIX SEVEN - MINDMAPS

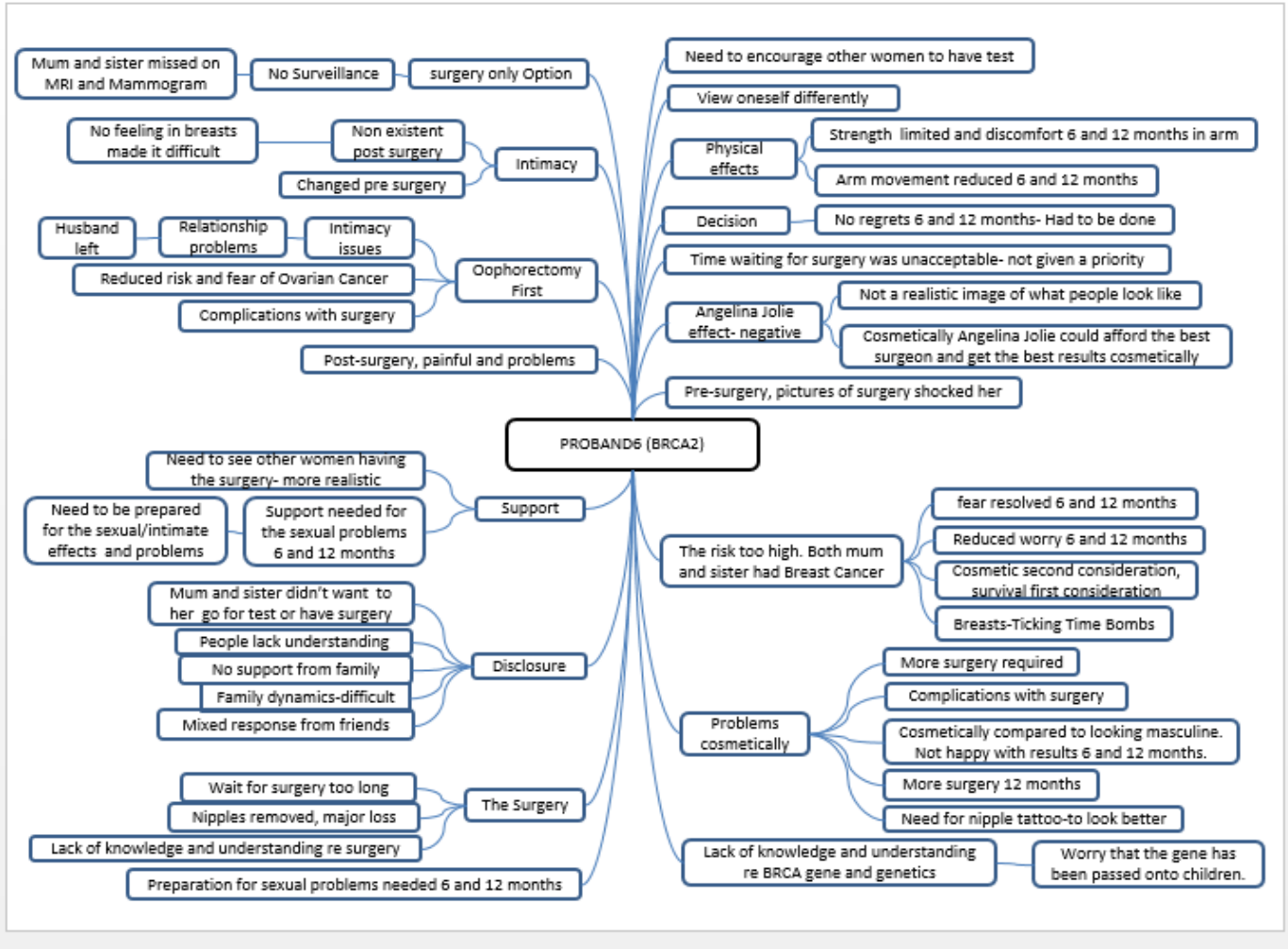




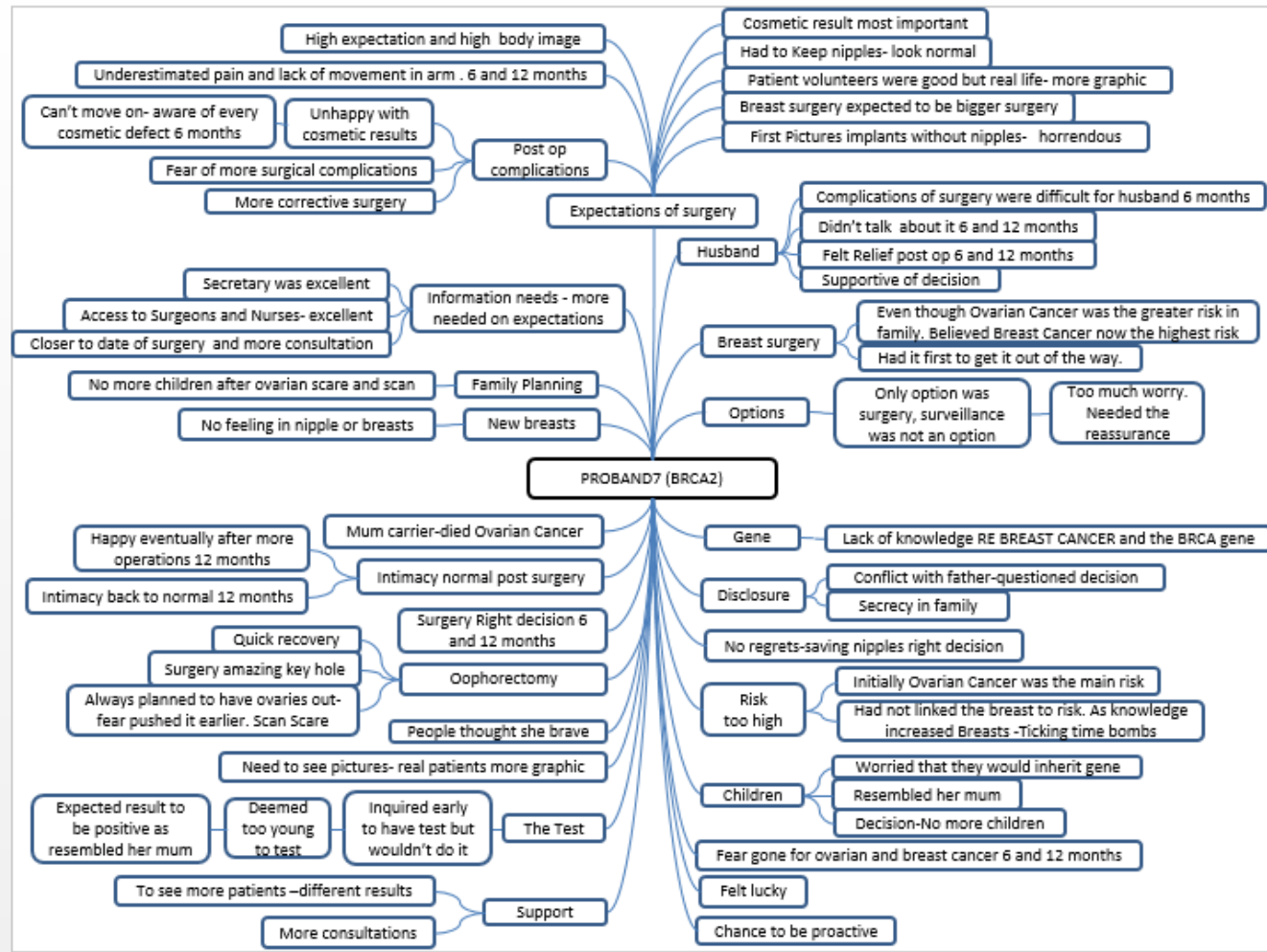


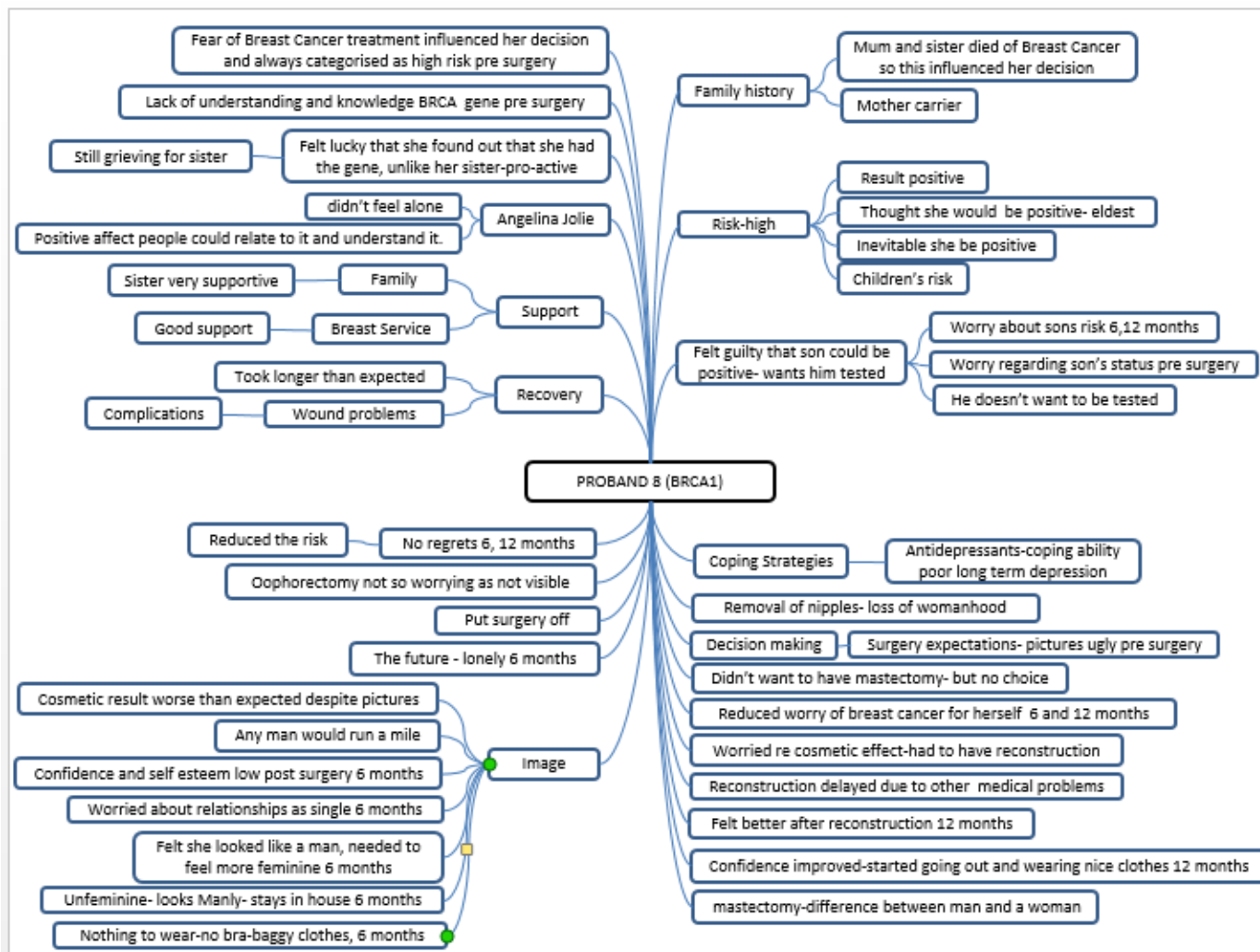


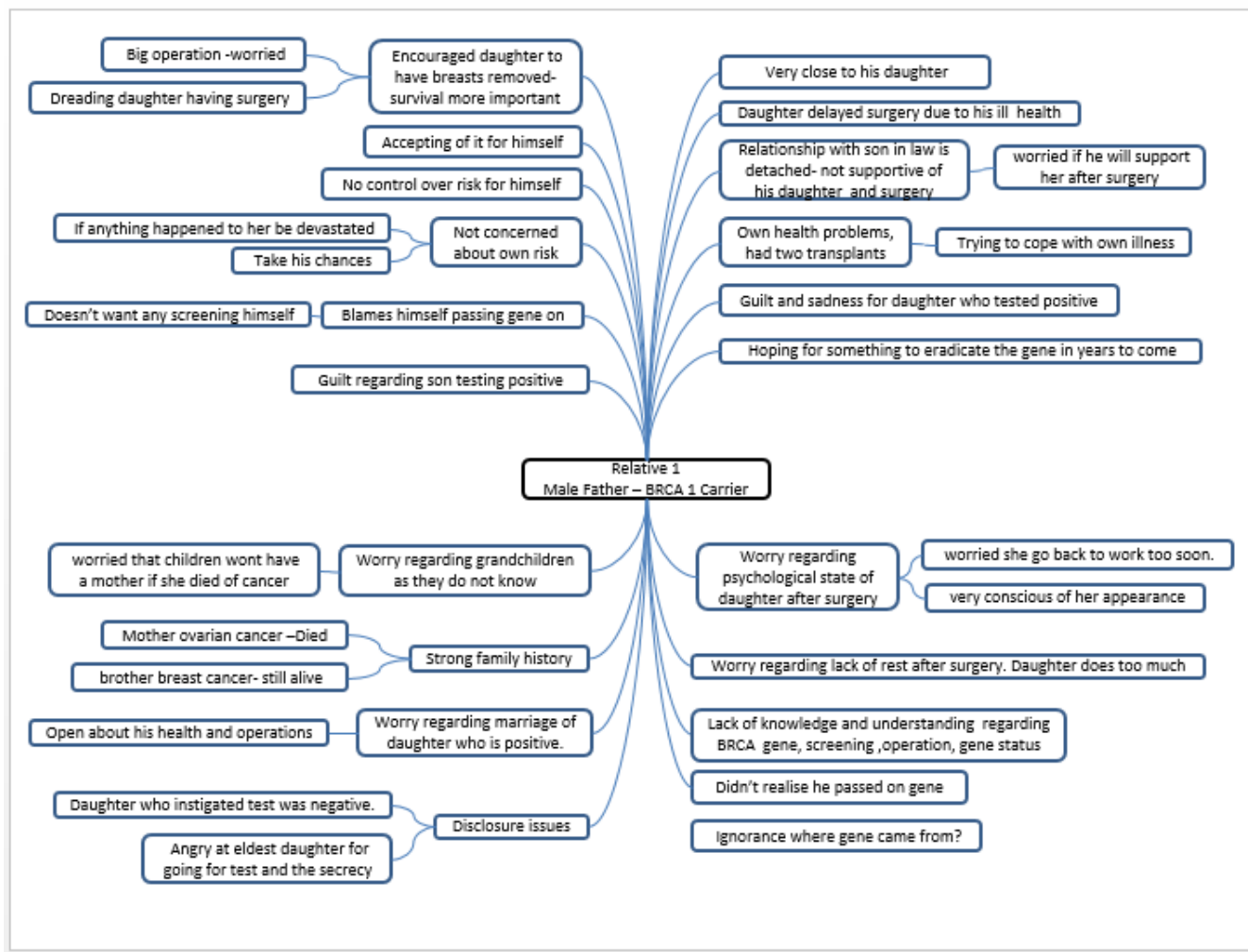


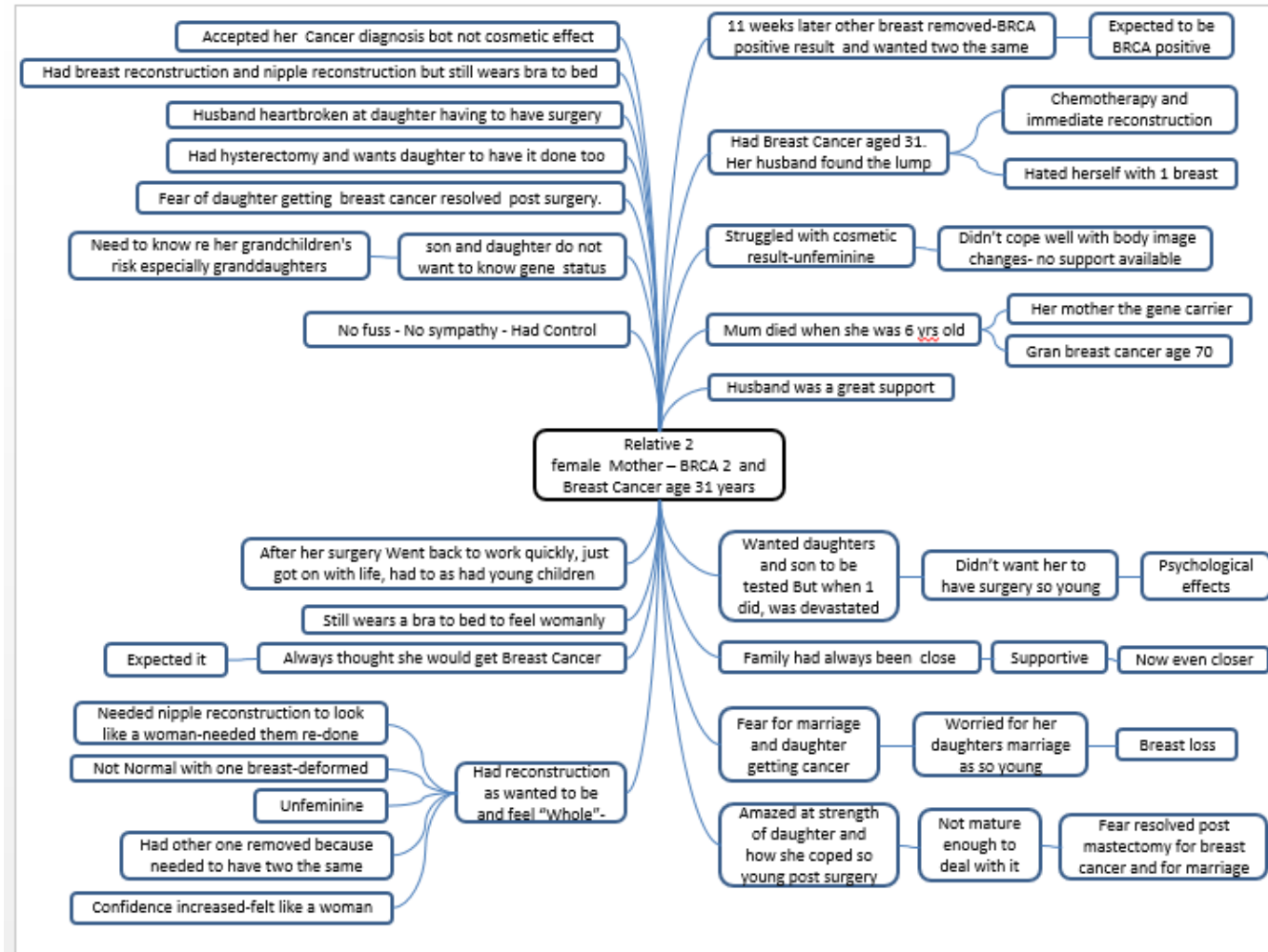


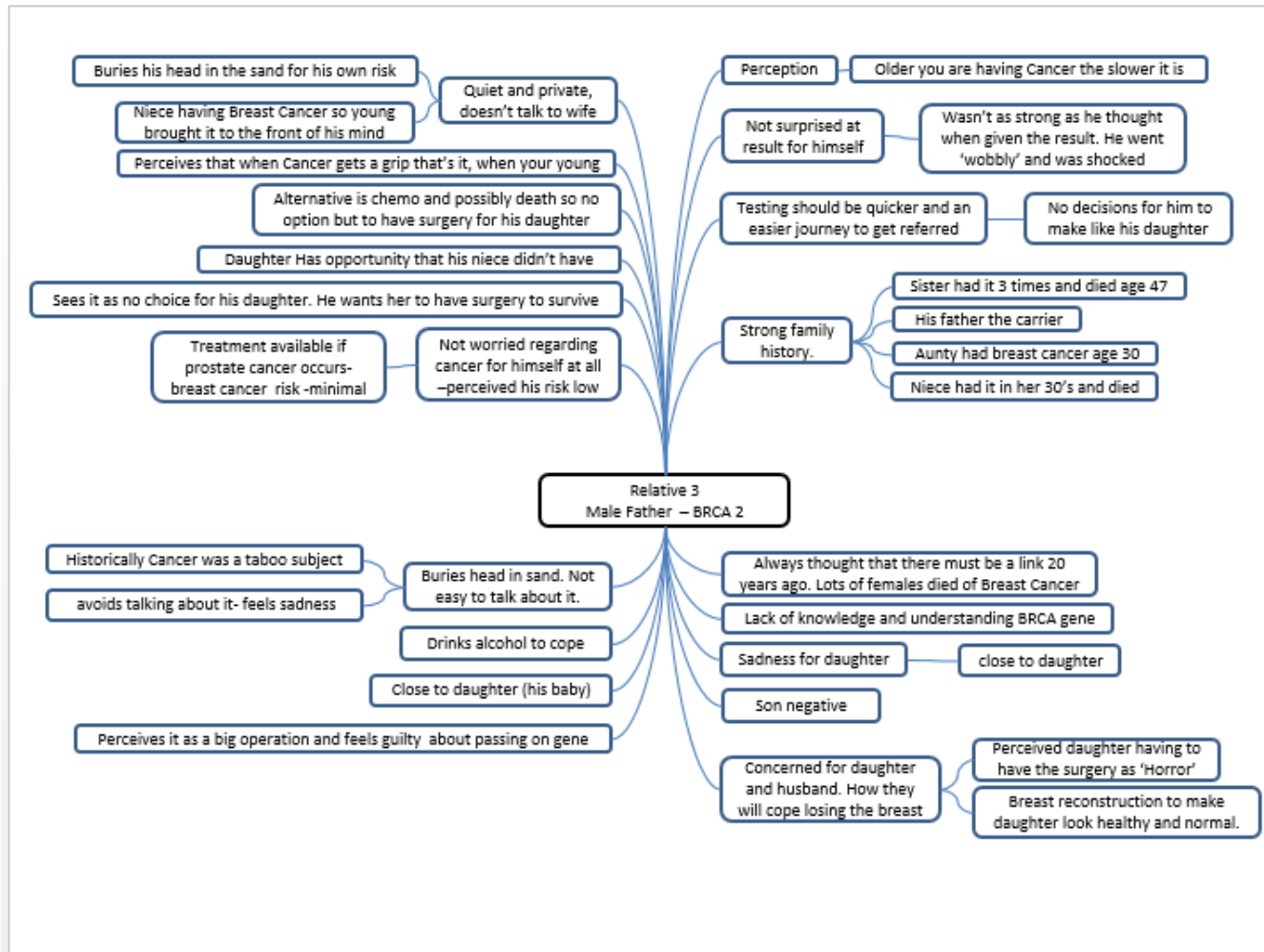


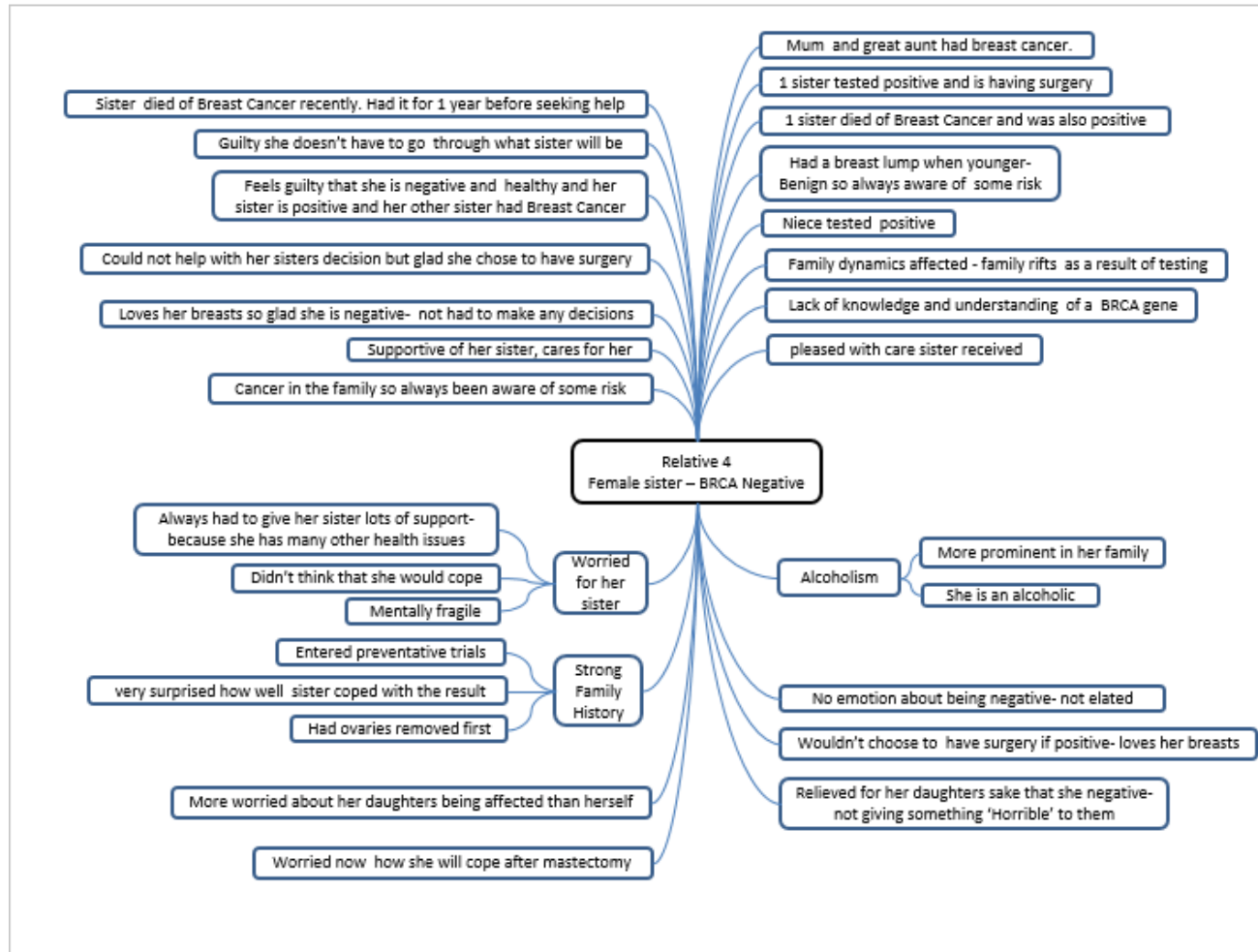


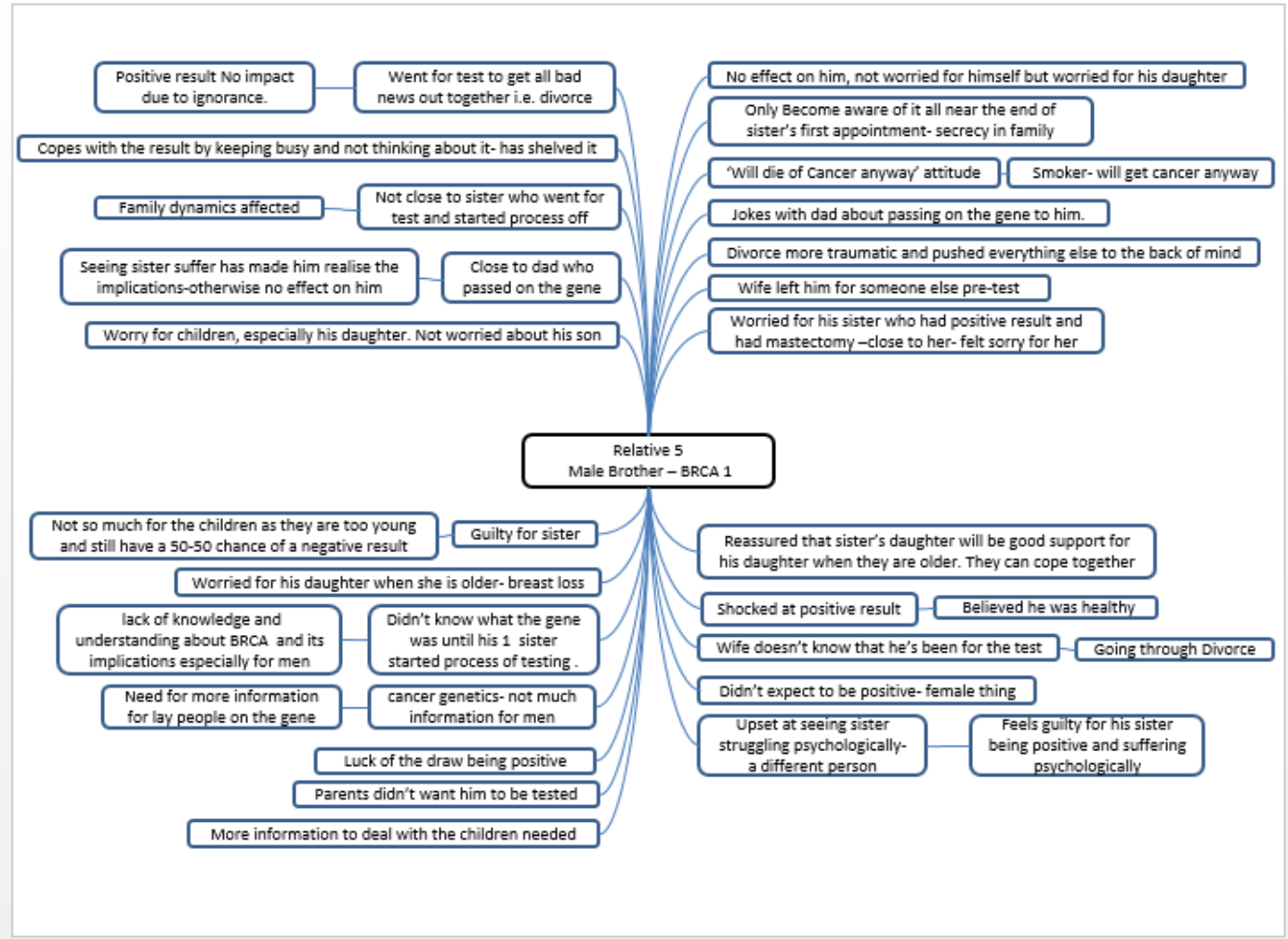


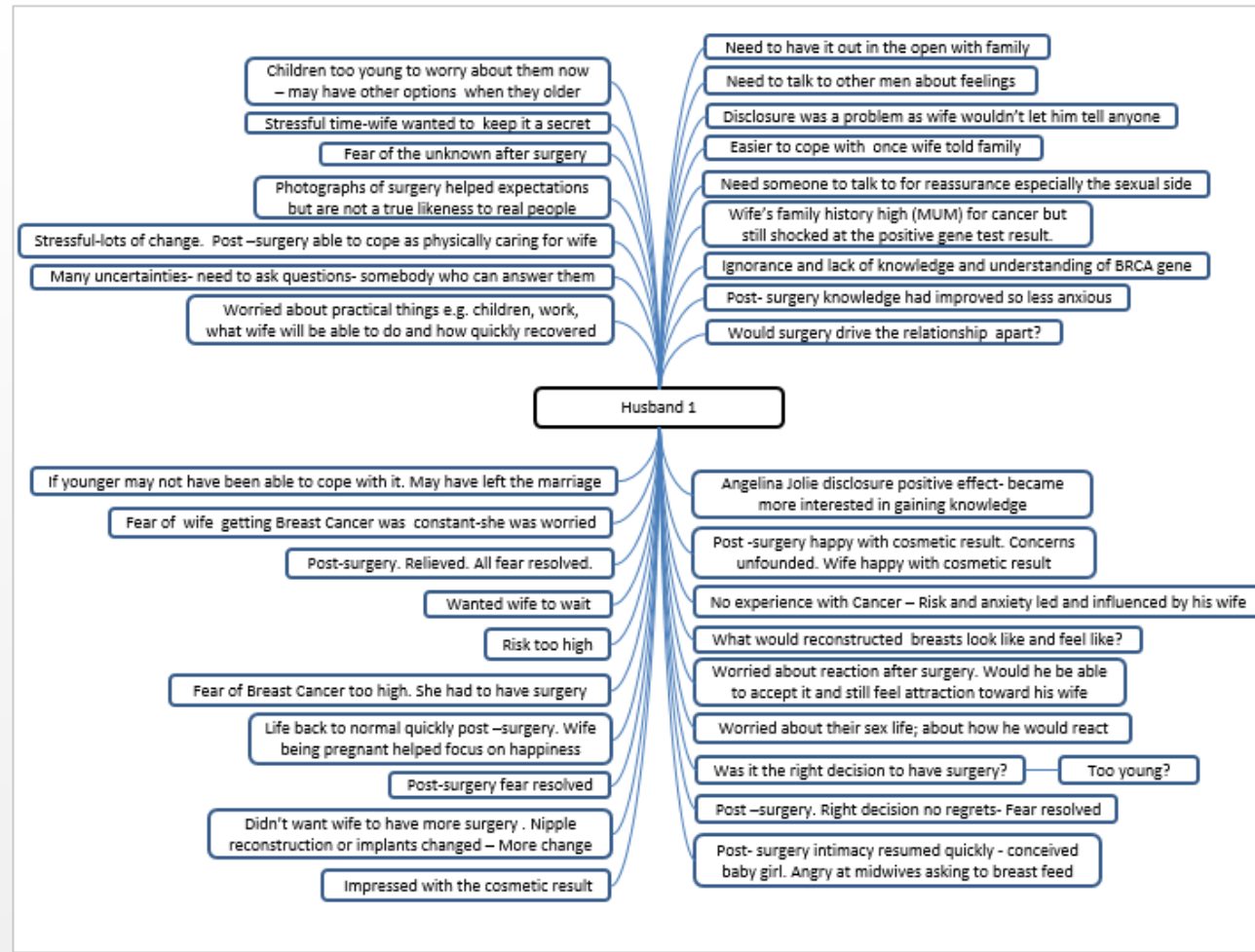




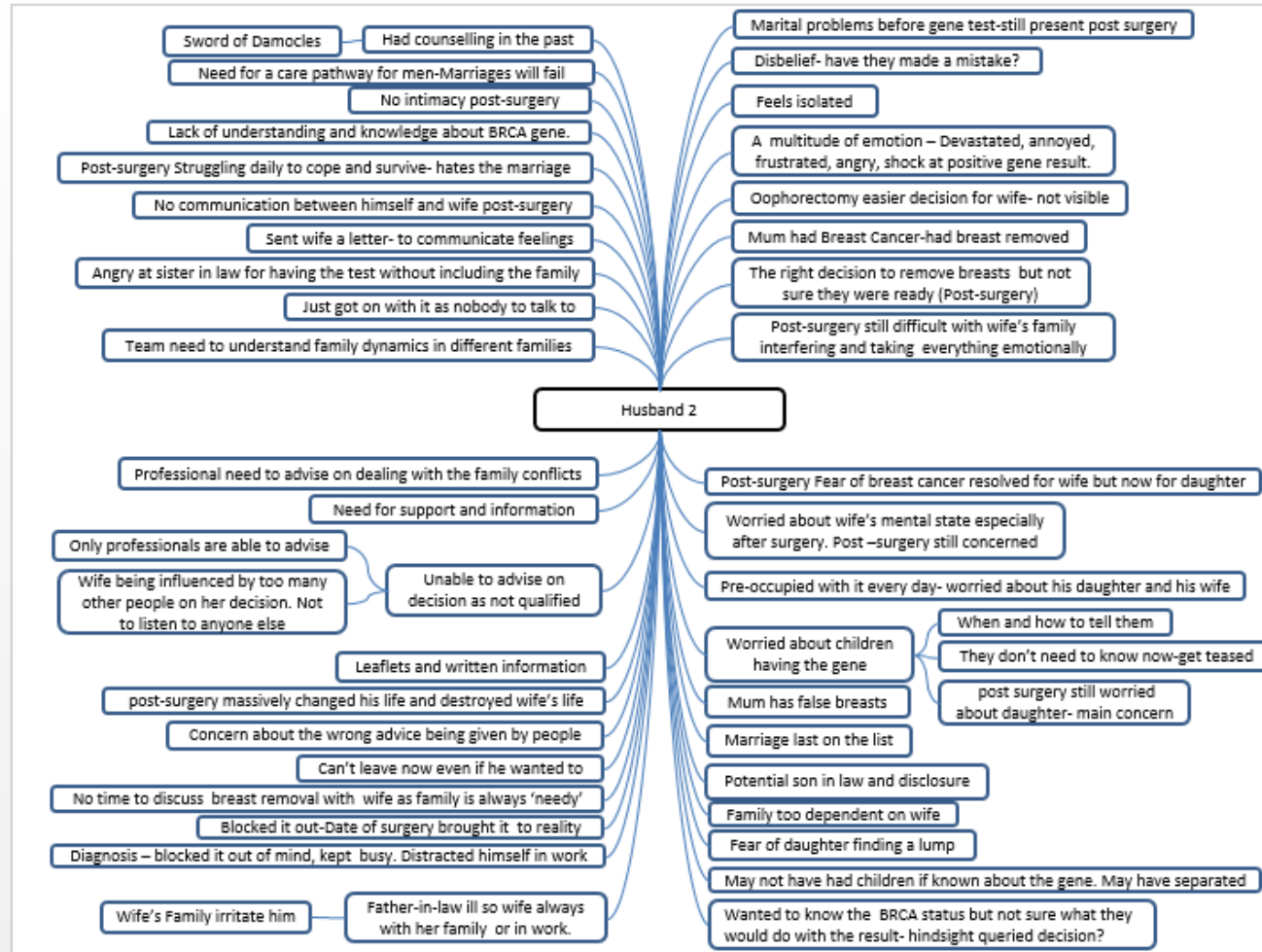


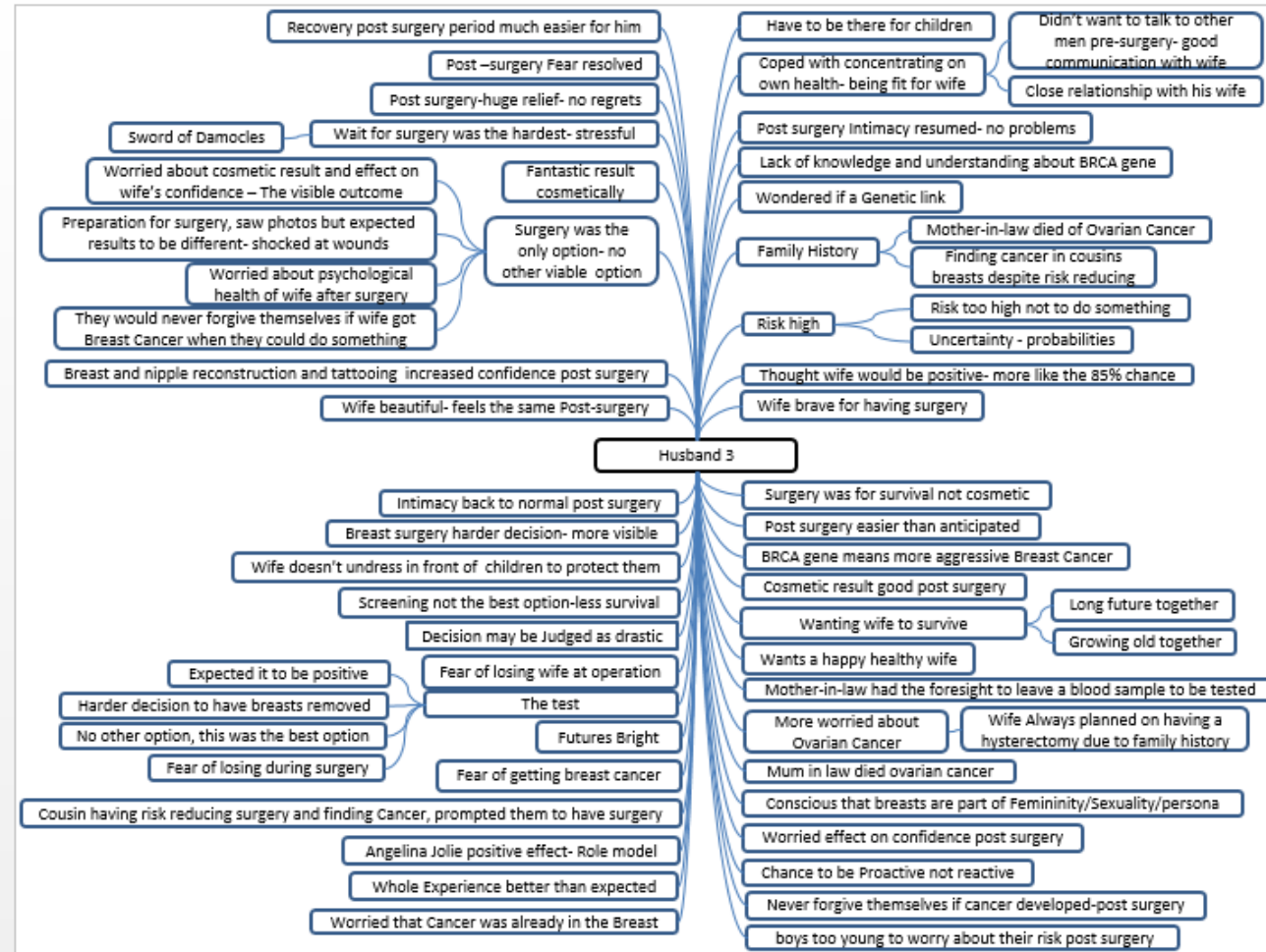
















## APPENDIX EIGHT – NHS R&D Approval



GIG  
CYMRU  
NHS  
WALES

Bwrdd Iechyd Prifysgol  
Caerdydd a'r Fro  
Cardiff and Vale  
University Health Board

Ysbyty Athrofaol Cymru  
University Hospital of Wales

Heath Park,  
Cardiff, CF14 4XW  
Phone 029 2074 7747  
Fax 029 2074 3838  
Minicom 029 2074 3632

Parc Y Mynydd Bychan,  
Caerdydd, CF14 4XW  
Ffôn 029 2074 7747  
Ffacs 029 2074 3838  
Minicom 029 2074 3632

Eich cyf/Your ref  
Ein cyf/Our ref  
Welsh Health Telephone Network 1872  
Direct line/Llinell uniongyrchol

Tel: 029 20746986  
Fax: 029 20745311  
CAV\_Research.Development@wales.nhs.uk

From: Professor JI Bisson  
R&D Director  
R&D Office, 2<sup>nd</sup> Floor TB2  
University Hospital of Wales  
Cardiff  
CF14 4XW

14 May 2012

Mrs Nicola West  
Consultant Nurse/Lecturer  
Llandough Breast Centre  
University Llandough Hospital  
Penlan Road  
Cardiff  
CF64 2XX

Dear Mrs West

**Cardiff and Vale UHB Ref : 11/CLC/5265 : The Long Term Experience of BRCA 1/2 (Gene Mutation Carriers) Women Undergoing Bilateral Risk Reducing Mastectomy**

**NISCHR PCU Ref: 89313**

The above project was forwarded to Cardiff and Vale University Health Board R&D Office by the NISCHR Permissions Coordinating Unit. A Governance Review has now been completed on the project.

Documents approved for use in this study are:

Document	Version	Date
NHS R&D Form	3.3	Received 1 November 2011
SSI Form	3.3	Received 1 November 2011
Protocol	1.0	2 September 2011
Participant Information Sheet	4.0	1 January 2012
Participant Information Sheet: Relative	4.0	1 January 2012
Participant Consent Form: Relative	2.0	1 January 2012
Participant Consent Form	2.0	1 January 2012

I am pleased to inform you that the UHB has no objection to your proposal.

Page 1 of 2

Version 1.0. 09.06.10

You have informed us that Cardiff University is willing to act as Sponsor under the Research Governance Framework for Health and Social Care.

Please accept this letter as confirmation of permission for the project to begin within this UHB.

May I take this opportunity to wish you success with the project and remind you that as Principal Investigator you are required to:

- Inform NISCHR PCU and the UHB R&D Office if any external or additional funding is awarded for this project in the future
- Submit any substantial amendments relating to the study to NISCHR PCU in order that they can be reviewed and approved prior to implementation
- Ensure NISCHR PCU is notified of the study's closure
- Ensure that the study is conducted in accordance with all relevant policies, procedures and legislation
- Provide information on the project to the UHB R&D Office as requested from time to time, to include participant recruitment figures

Yours sincerely,



**Professor Jonathan I Bisson**  
R&D Director

**CC R&D Lead Dr Kate Craig**  
Chris Shaw, RACD, Cardiff University

## APPENDIX NINE – REC Approval

Part of the research infrastructure for Wales funded by the National Institute for Social Care and Health Research, Welsh Government.  
Yn rhan o seilwaith ymchwil Cymru a ariannir gan y Sefydliad Cenedlaethol ar gyfer Ymchwil Gofal Cymdeithasol ac Iechyd, Llywodraeth Cymru



Uned  
Cyflynu  
Caniatâd | **PCU** | Permissions  
Co-ordinating  
Unit

South East Wales Research Ethics Committee  
Sixth Floor, Churchill House  
17 Churchill Way  
Cardiff CF10 2TW

Telephone : 029 2037 6823  
E-mail : [jagit.sidhu@wales.nhs.uk](mailto:jagit.sidhu@wales.nhs.uk)

Website : [www.nres.nhs.uk](http://www.nres.nhs.uk)

### South East Wales Research Ethics Committee - Panel D

25 January 2012

Mrs Nicola West  
Consultant Nurse Breast Care/Lecturer  
Cardiff and Vale University Health Board  
Llandough Hospital Penlan Road Penarth  
Cardiff  
CF64 2XX

Dear Mrs West

**Study title:** The long term experience of BRCA1/2(gene mutation carriers) Women undergoing bilateral risk reducing mastectomy.  
**REC reference:** 12/WA/0003  
**Protocol number:** spon 1027-11

Thank you for your letter of 20 January 2012, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair, Dr K Craig.

#### Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation [as revised], subject to the conditions specified below.

#### Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Cynhelir Cydweithrediad Gwyddor Iechyd Academaidd y Sefydliad Cenedlaethol ar gyfer Ymchwil Gofal Cymdeithasol ac Iechyd gan Fwrdd Addysgu Iechyd Powys

The National Institute for Social Care and Health Research Academic Health Science  
Collaboration is hosted by Powys Teaching Health Board

Non-NHS sites

### Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

*Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.*

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

*Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.*

*For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.*

*Sponsors are not required to notify the Committee of approvals from host organisations*

**It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).**

### Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Interview Schedules/Topic Guides		
Letter from Sponsor	Cardiff University	10 October 2011
Other: CV	H Sweetland	01 January 2012
Other: CV	A Murray	01 January 2012
Other: CV	N West	01 January 2012
Other: CV	K Featherstone	01 August 2011
Participant Consent Form ✓	2	01 January 2012
Participant Consent Form	2	01 January 2012
Participant Information Sheet	4	01 January 2012
Participant Information Sheet: Relative	4	01 January 2012
Protocol	1.0	02 September 2011
REC application	IRAS 3.3	09 December 2011
Response to Request for Further Information		20 January 2012

### Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.



## After ethical review

### Reporting requirements

The attached document "*After ethical review – guidance for researchers*" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

### Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

**12/WA/0003**

**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project

Yours sincerely

**Dr K J Craig**  
**Chair**

Email: [jagit.sidhu@bsc.wales.nhs.uk](mailto:jagit.sidhu@bsc.wales.nhs.uk)

*Enclosures:* "After ethical review – guidance for researchers" [SL-AR2]

*Copy to:* Professor Jonathan Bisson, Cardiff and Vale UHB

## APPENDIX TEN – REC Provisional Approval



South East Wales Research Ethics Committee

Panel D

Sixth Floor, Churchill House

17 Churchill Way

Cardiff CF10 2TW

12 January 2012

Mrs Nicola West

Consultant Nurse Breast Care/Lecturer

Cardiff and Vale University Health Board

Llandough Hospital Penlan Road Penarth

Cardiff

CF64 2XX

Dear Mrs West

**Study Title:** The long term experience of BRCA1/2(gene mutation carriers)  
Women undergoing bilateral risk-reducing mastectomy.

**REC reference:** 12/WA/0003

**Protocol number:** spon 1027-11

The South East Wales Research Ethics Committee - Panel D reviewed the above application at their meeting held on the 12<sup>th</sup> January 2012.

Thank you for attending to discuss the study, the clarification that you provided was most helpful.

### Documents reviewed

The documents reviewed at the meeting were:

<i>Document</i>	<i>Version</i>	<i>Date</i>	
Investigator CV	N West		

Investigator CV	K Featherstone		
Letter from Sponsor	Cardiff University	10 October 2011	
Participant Consent Form	1	01 December 2011	
Participant Information Sheet: Patient Information Sheet inc Initial Contact Consent	3	01 December 2011	
Protocol	1.0	02 September 2011	
REC application	IRAS 3.3	09 December 2011	

### Provisional opinion

The Committee noted that this was a single site study with the main objective being to explore the long term experiences of women who have inherited the breast cancer gene and who decide to undergo bilateral risk-reducing mastectomy with or without breast reconstruction.

The Committee noted that the study was being carried out for an educational qualification.

The Committee noted that potential participants would initially be identified and approached by healthcare professionals involved in their care.

The Committee noted that potential participants will be provided with written information about the purpose of the study, why they have been invited to participate, who is conducting the research, how the data would be used and what participation will require of them. They will also be given the opportunity to ask any questions about the study. Written consent will be obtained prior to participation in the study. It will be made clear throughout the study that participation is entirely voluntary and that they can withdraw at any point for any reason.

The Committee also noted that once participants have consented they will be given information to pass on to their partners/others to consider taking part.

The Committee noted that no intervention or procedure, which would normally be considered a part of routine care, would be withheld from participants.

The Committee noted that the Cardiff University would be acting as sponsor for the above study in accordance with the Research Governance Framework.

The Committee noted that the sponsor had signed the declaration in the application form to confirm that an appropriate process of scientific critique had demonstrated that the research proposal is worthwhile and of high scientific quality and that the necessary insurance or indemnity arrangements will be in place prior to commencement of the research.

The Committee noted from Q (A43) of the application form that study data would be stored for over 3 years after the end of the study. The Committee asked that you ensure that all data is stored and destroyed in line with the Data Protection Act (1998).

The Committee noted your reassurance that some data would be stored on laptop computers and that all data would be stored in line with University policy on such matters and that a suitable encryption programme would be used to ensure the security and confidentiality of the data.

The Committee noted that the start date on the application was incorrect and asked that the study does not commence until all the relevant approvals are in place.

The Committee agreed that they would be content to give a favourable ethical opinion of the research, subject to receiving a complete response to the request for further information set out below.

The Committee delegated authority to confirm its final opinion on the application to the Chair.

### **Further information or clarification required**

1. The patient information sheet should explain that you will be approaching the patient's partner, with the patient's knowledge and consent, to participate in the research.
2. The information sheet section: 'What are the possible benefits of taking part?' The Committee agreed that it was important not to exaggerate the possible benefits to the particular patient during the course of the study. Where there is no intended clinical benefit to the patient from taking part in the study this should be stated clearly.
3. The information sheet; 'How and where will the research be undertaken?' The Committee asked that the third sentence be revised to read 'They will be take place either in your own home, or if you prefer at a location of your choice at your convenience'.
4. The Committee noted that you intended to contact the patient's partner in order to recruit them into the study. The Committee were of the view that it is important that the patient has knowledge of this and has given permission for you to contact their partner/relative. The Committee were of the view that the most pragmatic method of doing this would be to provide the patient with a letter explaining the nature of the study and providing your contact details, which could be passed by the patient to their partner if they wish to do so. The partner can then contact you if they wish to participate.
5. The Committee noted that the information sheet referred in places to 'I' and other places to 'we' this should be consistent.
6. The Committee noted that information sheet stated that 'The tapes will be locked in a secure cabinet within the University'. You should state for how long and explain when they will be destroyed.
7. The Committee noted that the page layout of the information sheet was random and asked that you look at this.
8. The Committee made reference to the consent form 'point 4' and asked that you explain in the consent form who the 'involved organisations' are.
9. The Committee asked that you provide a topic guide or interview schedule for the patients and their partners.
10. The Committee asked that a separate partner information sheet and consent form be provided.
11. The Committee noted that your CVs were not dated as required by the Standard Operating Procedures for Research Ethics Committees . The CVs must therefore be revised and resubmitted.

**If you would find it helpful to discuss any of the matters raised above or seek further clarification from a member of the Committee, you are welcome to contact Mrs J Sidhu, Co-ordinator on 02920 376822.**

When submitting your response to the Committee, please send revised documentation where appropriate underlining or otherwise highlighting the changes you have made and giving revised version numbers and dates.

If the committee has asked for clarification or changes to any answers given in the application form, please do not submit a revised copy of the application form; these can be addressed in a covering letter to the REC.

The Committee will confirm the final ethical opinion within a maximum of 60 days from the date of initial receipt of the application, excluding the time taken by you to respond fully to the above points. A response should be submitted by no later than 11 May 2012.

#### **Membership of the Committee**

The members of the Committee who were present at the meeting are listed on the attached sheet.

#### **Statement of compliance**

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

<b>12/WA/0003</b>	<b>Please quote this number on all correspondence</b>
-------------------	---

Yours sincerely

**Dr K J Craig**  
**Chair**

Email: jagit.sidhu@bsc.wales.nhs.uk

*Enclosures: List of names and professions of members who were present at the meeting and those who submitted written comments.*

*Copy to: Professor Jonathan Bisson, Cardiff and Vale UHB*

## **APPENDIX ELEVEN – Potential Interview Topics**

Potential Topics / Information to be discussed in one to one interviews.

Questions and areas discussed pre-surgery with patients will cover:

- What has it been like living with the Risk and BRCA1/2 status?
- What if any, were the disclosure problems to friends and family members?
- What are your fears and expectations?
- What is your past medical history of physical and psychological problems? Body image/ Sexuality problems
- How do you cope with stressful situations, what are your Coping strategies?

Information needs - were they met and how can they be improved.

Areas discussed with the patient post-surgery will cover:

- Recovery.
- Satisfaction, regret?
- Body image and sexuality.
- Anxiety and fear.
- Problems experienced.
- Effects on the family.
- The impact of the experience.

- Coping.
- The need for psychological help?
- Areas of questions for the partners/ relatives.
- How has the results of the gene testing affected you?
- What are your fears /expectations?
- What are your information and psychological needs?
- How can the breast care team help you?
- What has the whole experience of the results and what your relative has been through been like for you?
- Are there any specific problems that you think we could help with?

## APPENDIX TWELVE - Patient Consent Form Version 2



GIG  
CYMRU  
NHS  
WALES

Bwrdd Iechyd Prifysgol  
Caerdydd a'r Fro  
Cardiff and Vale  
University Health Board

### Consent Form Version 2, January 2012

#### Study Title: The long term experience of BRCA1/2(gene mutation carriers) women undergoing bilateral risk-reducing mastectomy

Please initial box

1. I confirm that I have read the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.

3. I understand that my interview will be recorded on a digital recording device. I give permission for this.

4. I understand that data collected may be looked at by responsible representatives from Cardiff University and Cardiff and Vale Health Board for the purposes of monitoring and auditing the conduct of the research. I give permission for this.

5. I understand that data collected will not be transferred to any commercial organisation.

6. I understand that if I lose the capacity to consent, that I will be withdrawn from the study. I understand that any information collected from interviews/observations before the loss of consent will still be included in the study.

7. I agree to take part in the above study.

Name of Person taking consent  
Date  
Signature



## APPENDIX THIRTEEN - Patient Information Sheet



### **Patient Participant Information Sheet (Relative) Version 4 January 2012**

#### **Information about the Research**

##### **Study title: The long term experience of BRCA1/2(gene mutation carriers) women undergoing bilateral risk-reducing mastectomy.**

You are invited to take part in the above research study because your relative is planning to undergo bilateral risk-reducing mastectomy in order to prevent breast cancer. Before you decide if you would like to take part in this research, please read the following information and feel free to discuss it with others. My name is Nicola West, I am a Consultant Nurse working within the Cardiff breast centre and this research is my PhD. For this research, I am based at the school of nursing and midwifery, Cardiff University. I am exploring the long term experiences and impacts of this surgery on women who carry the BRCA1/2 gene and their partners and family. This is important as this research will inform local services, so that they can be tailored to meet both women's physical needs and the emotional needs of the family.

#### **What is the research about?**

The research is about developing an understanding of the long term experiences of women who carry the gene and decide to undergo bilateral mastectomy with or without breast reconstruction. The research will also look at the wider impact on partners and family, which has been rarely examined.

#### **Why is the research being undertaken?**

At present, NHS services focus on patients with breast cancer, thus here is limited service provision (follow up and ongoing care) supporting women at high risk of breast cancer who undergo risk-reducing surgery. There is little evidence of the long term physical and psychological impacts of such surgery or the effects on partners and significant others. The findings from this research will inform the development of appropriate care pathways so that women and their families can receive the support and help they require.

#### **How and where will the research be undertaken?**

If you agree, you will be asked to take part in a number of interviews with me. There will be three interviews, which will be conducted at different time points such as pre-surgery, 6 months after the surgery and 12-18 months later.

They will take place either in your own home, or a location at your convenience or choice. Interviews will be audio recorded and will usually take 1-1.5 hours. All information is completely confidential and you will not be recognised as taking part in the study unless you want to.

**Do I have to take part?**

You do not have to take part in the study, and your care will not be affected in any way.

**What are the potential benefits of the research?**

You will be able to talk openly and honestly about your experiences and this will provide you with an opportunity to contribute to future services. It will also mean that there is a contact for you. This may be the first time you have been able to express your feelings and experiences. In addition, it will provide health professionals involved with women and families with a high risk of breast cancer with a much better understanding of the longer term effects of such surgery on the family and be able to offer appropriate support and care.

**Is there any risk of harm from the research?**

There is no predicted harm from participating in this study, although it is possible that discussing some aspects of undertaking your treatment and its effect on your life and family may cause some distress. If this happens, the researcher is a qualified counsellor and is able to offer support.

**Will my taking part in the study be kept confidential?**

Yes. I will follow ethical and legal practice and all information about you will be handled in confidence. You will not be identified by name and all procedures for handling storage and destruction of the data match the Caldecott principles. I will collect the data from the interviews which I will do by taping the interviews and the information will be stored on my computer within the university with an encrypted password. After 5 years of keeping the data, Cardiff University will archive and then destroy the information. The recordings will be locked in a secure cabinet within the University until transcribed and verified and then destroyed.

**What do I do now?**

If you are interested in taking part, please complete the attached form and return it in the pre-paid envelope .I will then contact you to organise a time convenient to meet with you.

**Who to contact?**

If you have any questions or queries, please feel free to contact me:

Nicola West. Room 702A.School of Nursing and Midwifery Studies, Cardiff University East Gate House .35 – 43 Newport Road.CF24 0AB  
On 078119595830 or [westn@cf.ac.uk](mailto:westn@cf.ac.uk)

Lynne Rollins Secretary on 02920716823  
Dr Katie Featherstone at featherstonek@cf.ac.uk

Alternatively you could contact Dr Mark Rogers or Dr Alexandra Murray at  
The All Wales Cancer Genetics Service on 02920744058.

## **Additional Information about the Research**

### **Study title: The long term experience of BRCA1/2 (gene mutation carriers) women undergoing bilateral risk-reducing mastectomy**

All the information collected will be treated confidentially, your name and address will not be attached in any way. All data will be safely stored. The tapes used to record the interviews will be safely stored, and the tapes will be destroyed when the interviews have been transcribed. The data collected will only be shared with my supervisors, but may be reviewed by Cardiff and Vale University Health Board to monitor the conduct of the research.

### **What will happen with the results of the research?**

The findings of the study will be discussed with the healthcare team at the University Hospital of Wales and Breast care team in Cardiff. The findings will also be submitted for publication in various Journals for healthcare professionals. The results will be disseminated across Wales so that all patients at high risk have access to appropriate care.

### **Who is funding the study?**

I work as a Consultant nurse within the Breast Centre at Llandough hospital and also as a lecturer at Cardiff University. Cardiff University and The Health Board are funding my study.

### **Who has approved the study?**

The study has been approved by the Research Ethics Committee for South East Wales, and has undergone scientific review by the Joint University Health Board and Cardiff University review service (Cardiff Research Review Service). Research and Development approval has been issued following this review by the Cardiff and Vale University Health Board Research and Development office.

### **What if something goes wrong?**

I do not expect any harm to come to you from taking part in this study. However, if you are not happy about any aspect of the study, please feel free to contact me on the details above. Alternatively, you can contact any member of the team at the Cardiff Breast unit on 02920715722 or the All Wales Cancer Genetics Team about any aspect of the research. If you wish to make a more formal complaint, the Cardiff and Vale University Health Board has a complaints service, and they can be contacted on 029 2074 2202.

**Other considerations**

If you lost capacity to consent during the study you would be excluded and no new information would be collected from you. However, previous information collected during interviews would still be used – the researcher will explain this in full if you decide to take part in the study.

**Patient Initial Contact Consent****Consent for Initial Contact.**

Nicola West  
School of Nursing and Midwifery  
Cardiff University  
Room 702 Eastgate House  
Newport Road, Cardiff CF24 0AB  
Mobile 07811959583

I confirm that I have read the information leaflet about your research project and I am happy for you to contact me to arrange a meeting.

I understand that my participation is entirely voluntarily and that I am free to withdraw at any time without giving a reason.

Name.

Address.

Signature.

Telephone Number.

Preferred method of contact.

Relationship to Patient

## APPENDIX FOURTEEN - Patient Information Sheet Final



### Patient Participant Information Sheet

Version 4 January 2012

### Information about the Research

#### **Study title: The long term experience of BRCA1/2(gene mutation carriers) women undergoing bilateral risk-reducing mastectomy.**

You are invited to take part in the above research study because you are planning to undergo bilateral risk-reducing mastectomy in order to prevent breast cancer. With your consent, your partner will also be approached to take part in the study. Before you decide if you would like to take part in this research, please read the following information and feel free to discuss it with others. My name is Nicola West, I am a Consultant Nurse working within the Cardiff breast centre and this research is my PhD. For this research, I am based at the school of nursing and midwifery, Cardiff University. I am exploring the long term experiences and impacts of this surgery on women who carry the BRCA1/2 gene and their partners and family. This is important as this research will inform local services, so that they can be tailored to meet women's physical and emotional needs.

#### **What is the research about?**

The research is about developing an understanding of the long term experiences of women who carry the gene and decide to undergo bilateral mastectomy with or without breast reconstruction. The research will also look at the wider impact on partners and family, which has been rarely examined.

#### **Why is the research being undertaken?**

At present, NHS services focus on patients with breast cancer, thus here is limited service provision (follow up and ongoing care) supporting women at high risk of breast cancer who undergo risk-reducing surgery. There is little evidence of the long term physical and psychological impacts of such surgery or the effects on partners and significant others. The findings from this research will inform the development of appropriate care pathways so that women and their families can receive the support and help they require.

**How and where will the research be undertaken?**

If you agree, you will be asked to take part in a number of interviews with me. There will be three interviews, which will be conducted at different time points such as while you are waiting for the surgery, 6 months after the surgery and 12-18 months later. They will take place either in your own home, or if you prefer at a location at your convenience or choice. Interviews will be audio recorded and will usually take 1-1.5 hours. In addition, you will be asked to complete a simple self-assessment scale to assess body image. All information is completely confidential and you will not be recognised as taking part in the study unless you want to.

**Do I have to take part?**

You do not have to take part in the study, and your care will not be affected in any way.

**What are the potential benefits of the research?**

You will be able to talk openly and honestly about your experiences and this will provide you with an opportunity to contribute to future services. It will also mean that there is a contact for you. Partners and other family members will benefit because it may be the first time they have been able to express their feelings and experiences. In addition, from a clinical perspective, it will provide health professionals involved with women with a high risk of breast cancer with a much better understanding of the longer term effects of such surgery and be able to offer appropriate support and care.

**Is there any risk of harm from the research?**

There is no predicted harm from participating in this study, although it is possible that discussing some aspects of undertaking your treatment and its effect on your life and family may cause some distress. If this happens, the researcher is a qualified counsellor and is able to offer support.

**Will my taking part in the study be kept confidential?**

Yes. I will follow ethical and legal practice and all information about you will be handled in confidence. You will not be identified by name and all procedures for handling storage and destruction of the data match the Caldecott principles. I will collect the data from the interviews which I will do by taping the interviews and the information will be stored on my computer within the university with an encrypted password. After 5 years of keeping the data, Cardiff University will archive and then destroy the information. The recordings will be locked in a secure cabinet within the University until transcribed and verified and then destroyed.

**What do I do now?**

If you are interested in taking part, please complete the attached form and return it in the pre-paid envelope. I will then contact you to organise a time convenient to meet with you. Could you please also pass on my details to your partner if you are happy for them to be approached?

**Who to contact?**

If you have any questions or queries, please feel free to contact me:  
Nicola West. Room 702A. School of Nursing and Midwifery Studies, Cardiff University East Gate House .35 – 43 Newport Road. CF24 0AB  
On 078119595830 or [westn@cf.ac.uk](mailto:westn@cf.ac.uk)  
Lynne Rollins Secretary on 02920716823  
Dr Katie Featherstone at [featherstonek@cf.ac.uk](mailto:featherstonek@cf.ac.uk)

Alternatively you could contact Dr Mark Rogers or Dr Alexandra Murray at The AI Wales Cancer Genetics Service on 02920744058.

**Additional Information about the Research****Study title: The long term experience of BRCA1/2 (gene mutation carriers) women undergoing bilateral risk-reducing mastectomy**

All the information collected will be treated confidentially, your name and address will not be attached in any way. All data will be safely stored. The tapes used to record the interviews will be safely stored, and the tapes will be destroyed when the interviews have been transcribed. The data collected will only be shared with my supervisors, but may be reviewed by Cardiff and Vale University Health Board to monitor the conduct of the research.

**What will happen with the results of the research?**

The findings of the study will be discussed with the healthcare team at the University Hospital of Wales and Breast care team in Cardiff. The findings will also be submitted for publication in various Journals for healthcare professionals. The results will be disseminated across Wales so that all patients at high risk have access to appropriate care

**Who is funding the study?**

I work as a Consultant nurse within the Breast Centre at Llandough hospital and also as a lecturer at Cardiff University. Cardiff University and The Health Board are funding my study.

**Who has approved the study?**

The study has been approved by the Research Ethics Committee for South East Wales, and has undergone scientific review by the Joint University Health Board and Cardiff University review service (Cardiff Research Review Service). Research and Development approval has been issued following this review by the Cardiff and Vale University Health Board Research and Development office.

**What if something goes wrong?**

I do not expect any harm to come to you from taking part in this study. However, if you are not happy about any aspect of the study, please feel free to contact me on the details above. Alternatively, you can contact any member of the team at the Cardiff Breast unit on 02920715722 or the All Wales Cancer Genetics Team about any aspect of the research. If you wish to make a more formal complaint, the Cardiff and Vale University Health Board has a complaints service, and they can be contacted on 029 2074 2202.

**Other considerations**

If you lost capacity to consent during the study you would be excluded and no new information would be collected from you. However, previous information collected during interviews would still be used – the researcher will explain this in full if you decide to take part in the study.

**Patient Initial Contact Consent****Consent for Initial Contact.**

Nicola West  
School of Nursing and Midwifery  
Cardiff University  
Room 702 Eastgate House  
Newport Road  
Cardiff  
CF24 OAB  
Mobile 07811959583

I confirm that I have read the information leaflet about your research project and I am happy for you to contact me to arrange a meeting.

I understand that my participation is entirely voluntarily and that I am free to withdraw at any time without giving a reason.

Name.

Address.

Signature.

Telephone Number.

Preferred method of contact.



## APPENDIX FIVETEEN – Partner Consent Form Version 2



GIG  
CYMRU  
NHS  
WALES

Bwrdd Iechyd Prifysgol  
Caerdydd a'r Fro  
Cardiff and Vale  
University Health Board

### Consent Form Version 2, January 2012 (Partner/Relative)

**Study Title: The long term experience of BRCA1/2 (gene mutation carriers) women undergoing bilateral risk-reducing mastectomy**

Please initial box

1. I confirm that I have read the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.

3. I understand that my interview will be recorded on a digital recording device. I give permission for this.

4. I understand that data collected may be looked at by responsible representatives from Cardiff University and Cardiff and Vale Health Board for the purposes of monitoring and auditing the conduct of the research. I give permission for this.

5. I understand that data collected will not be transferred to any commercial organisation.

6. I understand that if I lose the capacity to consent, that I will be withdrawn from the study. I understand that any information collected from interviews/observations before the loss of consent will still be included in the study.

7. I agree to take part in the above study.

Name of Participant  
Date  
Signature

Name of Person taking consent  
Date  
Signature

## APPENDIX SIXTEEN – FN Award Letter



# The Florence Nightingale Foundation

34 Grosvenor Gardens, First Floor Front, London SW1W 0DH

Tel: 020 7730 3030

Fax: 020 7730 6262

Email: [admin@florence-nightingale-foundation.org.uk](mailto:admin@florence-nightingale-foundation.org.uk) [www.florence-nightingale-foundation.org.uk](http://www.florence-nightingale-foundation.org.uk)

14 July 2014

Mrs Nicola West

Roslyn  
167c Marshfield Road  
Marshfield  
Cardiff  
Email: [agentors@btinternet.com](mailto:agentors@btinternet.com)

[Nicola.west@wales.nhs.uk](mailto:Nicola.west@wales.nhs.uk)

Dear Nicola

The Selection Panel were delighted to have the opportunity to meet you at your recent skype interview.

Through the generosity of The Stephanie Thompson Memorial Trust we are pleased to offer you an award of £1,500.00 to assist you in undertaking the 4<sup>th</sup> year of your PhD in Nursing September 2014 to April 2015. The title of your study is: What are the experiences of BRCA1/2 woman, their partners and relatives following diagnosis and up to 18 months post bilateral riskreducing mastectomy? I am aware that this is less than you requested but we believe should still enable you to achieve your stated objectives. This money will be paid either to the University where you are undertaking your course or directly to you, as required. In either case, we would need to have sight of an invoice or receipt as appropriate.

Royal Patron: HRH Princess Alexandra, the Hon. Lady Ogilvy, KG GCVO

President: the Baroness Emerton, DBE DL Chairman: Mr Bryan Sanderson, CBE Chief Executive: Professor Elizabeth Robb Administrator: Stephanie Dawes

A company limited by guarantee Registration No. 518623 Registered in England and Wales

Charity Registration No. 229229 and in Scotland Charity Registration No. SC044341

Would you thank Mr Jeff Thompson, on behalf of The Stephanie Thompson Memorial Trust at the email below.

jct\_jct@yahoo.com

The terms and conditions of acceptance of this scholarship are:

- The Foundation require a short six month review report by the end of April 2015 to update us on your progress. This needs to be a brief update of no more than 500 words which can be sent to the administrator at [admin@florence-nightingale-foundation.org.uk](mailto:admin@florence-nightingale-foundation.org.uk) as an attachment in a word document.
- I attach some notes for guidance on report writing plus a front cover for your report. A full report on your research (two hard copies and one on CD or stick) should reach The Foundation on completion of your studies covered by this award. Your report should include an acknowledgement of your sponsor and The Florence Nightingale Foundation, and be presented in a clear plastic cover.
- You should also send a copy of your report to your sponsor at the end of your scholarship.
- Your final report should be submitted within 3 months of completion of your year of study but no later than 18 December 2015.
- Your sponsor may also ask you to give a formal presentation on the outcome of your studies at a future meeting; we will liaise with you on this nearer the time.
- If asked, you should be willing to present your work at a Foundation conference.
- We also ask you publish something as a result of your scholarship. By accepting this award you agree to the Foundation sharing intellectual property rights generated as a result of this scholarship.
- By accepting this award we also hope that you will be happy to become an Alumni of the Foundation to help support future scholars within your area of expertise. Unless you inform us otherwise we will keep your electronic personal data on file and contact you with relevant information and opportunities to collaborate with The Foundation.

- If your home address or employment details change before you have submitted your report, you are required to keep the Foundation up-to-date with your contact details at all times.
- If you have a significant period of sickness or any event that may impact on the completion and/or submission of your final report, you are required to inform the Foundation as soon as possible to negotiate a new submission date if required.
- In the event that you do not undertake the study and produce an acceptable report for which this award is being made, you will return to The Foundation the money you have received, in order that it may be used to fund another Scholar.

If you accept the award in accordance with the conditions set out above in this letter, ***please sign one copy and return it to me as soon as possible, but by 24/7/14 at the latest*** keeping the second copy for future reference.

If you require any further information, please do not hesitate to contact this office.

With best wishes and congratulations on this award.

Yours sincerely



Professor Elizabeth Robb

Chief Executive

I accept this award on the conditions set out above and undertake to refund the money if circumstances outlined above apply.

.....  
 Name Date

Royal Patron: HRH Princess Alexandra, the Hon. Lady Ogilvy, KG GCVO  
 President: the Baroness Emerton, DBE DL Chairman: Mr Bryan Sanderson,  
 CBE Chief Executive: Professor Elizabeth Robb  
 Administrator: Stephanie Dawes  
 A company limited by guarantee Registration No. 518623 Registered in England and Wales  
 Charity Registration No. 229229 and in Scotland Charity Registration No. SC044341

## APPENDIX SEVENTEEN – FN Award Letter 2013/14



# The Florence Nightingale Foundation

34 Grosvenor Gardens, First Floor Front, London SW1W 0DH

Tel: 020 7730 3030

Fax: 020 7730 6262

Email: [admin@florence-nightingale-foundation.org.uk](mailto:admin@florence-nightingale-foundation.org.uk)

[www.florence-nightingale-foundation.org.uk](http://www.florence-nightingale-foundation.org.uk)

5 July 2013

Nicola West  
Roslyn  
167c Marshfield Road  
Cardiff CF3 2TU  
Email: [WestN@cardiff.ac.uk](mailto:WestN@cardiff.ac.uk) [argentors@btinternet.com](mailto:argentors@btinternet.com)

Dear Nicola

### **THE FLORENCE NIGHTINGALE FOUNDATION RESEARCH SCHOLARSHIPS 2013-2014: GARFIELD WESTON FOUNDATION RESEARCH SCHOLARSHIP**

The Selection Panel were delighted to have the opportunity to meet you at your recent Skype interview.

Through the generosity of Garfield Weston Foundation we are pleased to offer you an award of £1,914.00 to assist you in undertaking the 3<sup>rd</sup> year of her PhD in Nursing. The title of her study is: What are the experiences of BRCA1/2 woman, their partners and relatives following diagnosis and up to 18 months post bilateral risk-reducing mastectomy? This money will be paid either to the University where you are undertaking your course or directly to you, as required. In either case, we would need to have sight of an invoice or receipt as appropriate.

Would you thank Clare Wilkinson, on behalf of Garfield Weston Foundation. Her contact details are:

Clare Wilkinson  
Grants Manager  
Garfield Weston Foundation  
Weston Centre  
10 Grosvenor Street  
London W1K 4QY

Patron: HRH Princess Alexandra. The Hon. Lady Ogilvy, KG GCVO

President: the Baroness Emerton, DBE DL Chairman: Mr Bryan Sanderson, CBE Chief Executive:

Professor Elizabeth Robb

Administrator: Stephanie Dawes

The terms and conditions of acceptance of this scholarship are:

- The Foundation require a short six month review report by the end of March 2014 to update us on your progress.
- I attach some notes for guidance on report writing plus front covers for your report. A full report on your research (two hard copies and one on CD or stick) should reach The Foundation on completion of your studies covered by this award. Your report should include an acknowledgement of the Garfield Weston Foundation and The Florence Nightingale Foundation, and be presented in a clear plastic cover.
- You should also send a copy of the report to your sponsor at the end of your scholarship.
- Your final report should be submitted within 3 months of completion of your year of study i.e. no later than 29 November 2014.
- Your sponsor may also ask you to give a formal presentation on the outcome of your studies at a future meeting; we will liaise with you on this nearer the time.
- If asked, you should be willing to present your work at a Foundation conference.
- We also ask you publish something as a result of your scholarship. By accepting this award you agree to the Foundation sharing intellectual property rights generated as a result of this scholarship.
- By accepting this award we also hope that you will be happy to become an Alumni of The Foundation to help support future scholars within your area of expertise. Unless you inform us otherwise we will keep your electronic personal data on file and contact you with relevant information and opportunities to collaborate with The Foundation.
- If your home address or employment details change before you have submitted your report, you are required to keep the Foundation up-to-date with your contact details at all times.
- In the event that you do not undertake the study and produce an acceptable report for which this award is being made, you will return to The Foundation the money you have received, in order that it may be used to fund another Scholar.

If you accept the award in accordance with the conditions set out above in this letter, ***please sign one copy and return it to me as soon as possible, but by 23/7/13 at the latest*** keeping the second copy for future reference.

If you require any further information, please do not hesitate to contact this office.

With best wishes and congratulations on this award.

Yours sincerely



Professor Elizabeth Robb  
Chief Executive

---

I accept this award on the conditions set out above and undertake to refund the money if circumstances outlined above apply.

.....

.....

Name

Date

## APPENDIX EIGHTEEN – FN Award Letter 2012/13



# The Florence Nightingale Foundation

34 Grosvenor Gardens, First Floor Front, London SW1W 0DH

Tel: 020 7730 3030

Fax: 020 7730 6262

Email: [admin@florence-nightingale-foundation.org.uk](mailto:admin@florence-nightingale-foundation.org.uk)

[www.florence-nightingale-foundation.org.uk](http://www.florence-nightingale-foundation.org.uk)

13 July 2012

Mrs Nicola West  
Roslyn  
167c Marchfield Road  
Cardiff CF3 2TU

Dear Nicola

### **THE FLORENCE NIGHTINGALE FOUNDATION RESEARCH SCHOLARSHIPS 2012-2013: THE BAND TRUST RESEARCH SCHOLARSHIP 2012-2013**

The Selection Panel were delighted to have the opportunity to meet you at your recent interview.

Through the generosity of The Band Trust we are pleased to offer you an award of £1,766.00 to assist you in undertaking your 2<sup>nd</sup> year PhD. This money will be paid either to the University where you are undertaking your course or directly to you, as required. In either case, we would need to have sight of an invoice or receipt as appropriate.

Would you please write and thank The Hon Mrs Nicholas Wallop, Chairman at The Band Trust.

Her address is:

The Hon Mrs Nicholas Wallop  
The Band Trust  
4 Lexham Mews  
London W8 6JW

The terms and conditions of acceptance of this scholarship are:

- The Foundation require a short six month review report by the end of March 2013 to update us on your progress.
- I enclose some notes for guidance on report writing plus front covers for your report. A full report on your research (two hard copies and one on CD or stick) should reach The Foundation on completion of your studies covered by this award. Your report should include an acknowledgement of The Band Trust and The Florence Nightingale Foundation, and be presented in a clear plastic cover.
- You should also send a copy of the Abstract to your sponsor at the end of your scholarship.
- Your final report should be submitted within 3 months of completion of your year of study i.e. before Christmas 2013.

Patron: HRH Princess Alexandra, The Hon. Lady Ogilvy, KG, GCVO  
President: The Baroness Emerton, DBE DL Chairman: Mr Bryan Sanderson, CBE Chief Executive: Professor Elizabeth Robb  
Administrator: Stephanie Dawes

A company limited by guarantee Registration No 518623 England Charity Registration No 229229




- Your sponsor may also ask you to give a formal presentation on the outcome of your studies at a future meeting; we will liaise with you on this nearer the time.
- If asked, you should be willing to present your work at a Foundation conference.
- We also ask you publish something as a result of your scholarship. By accepting this award you agree to the Foundation sharing intellectual property rights generated as a result of this scholarship.
- By accepting this award we also hope that you will be happy to become an Alumni of The Foundation to help support future scholars within your area of expertise. Unless you inform us otherwise we will keep your electronic personal data on file and contact you with relevant information and opportunities to collaborate with The Foundation.
- If your home address or employment details change before you have submitted your report, you are required to keep the Foundation up-to-date with your contact details at all times.
- In the event that you do not undertake the study and produce an acceptable report for which this award is being made, you will return to The Foundation the money you have received, in order that it may be used to fund another Scholar.

If you accept the award in accordance with the conditions set out above in this letter, ***please sign one copy and return it to me as soon as possible, but by 31/7/12 at the latest*** keeping the second copy for future reference.

If you require any further information, please do not hesitate to contact this office.

With best wishes and congratulations on this award.

Yours sincerely



Professor Elizabeth Robb  
Chief Executive

---

I accept this award on the conditions set out above and undertake to refund the money if circumstances outlined above apply.

.....  
Name

.....  
Date