

Women's experience of decision-making regarding prophylactic mastectomy

Thesis submitted in part fulfilment of the degree of

Doctorate in Clinical Psychology

(DClinPsy)

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Declaration

I confirm that this thesis (comprising of a literature review, research report and critical appraisal) is my own work. It was written and submitted in part fulfilment of the degree of Doctorate in Clinical Psychology (DClinPsy) and has not been submitted for any other academic award.

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Lauren Wright

Thesis abstract

Literature review

A systematic review of the existing literature was conducted, eliciting ten studies which met the inclusion criteria examining psychosocial predictors of prophylactic mastectomy in women with a confirmed BRCA gene alteration. Narrative synthesis identified that results coalesced around temporal, familial and other factors including conceptualisation of cancer and perceived risk. The relative scarcity of published research, and an accompanying dominant biomedical focus, highlight that further exploration of psychosocially predictive factors, particularly those which are modifiable, is needed.

Research report

Interpretative Phenomenological Analysis (IPA) was utilised to explore five women's experience and sense-making of their decision to opt for prophylactic mastectomy, and how they experienced the period between opting for preventative surgery and waiting for this to occur. Four superordinate themes were identified: 'It's a no-brainer' illuminated how women approached and made sense of their decision; 'good breast/bad breast' reflected women's experience of simultaneously holding conflicting views towards their breasts; 'big B on my shoulder' highlighted worry held in relation to geneticised identity; and 'the preciousness of life' illustrated the impact of familial and existential experience. Findings emphasised the importance of clinicians remaining mindful to experiential, emotional and systemic motivations for surgery and to recognise and support women with the potential tension they may still hold as they debate and navigate prophylactic mastectomy.

Critical appraisal

A reflective account is presented to support the consolidation of personal and professional learning points and reflections made during the research process.

Acknowledgements

Firstly, I would like to extend my sincere gratitude to the five remarkable women who volunteered to share their experiences, without whom, this research would not have been possible. I felt honoured to hear your stories and left each interview feeling truly inspired by your strength, positivity and determination. As per your wishes, I hope that others might read this thesis and gain a greater understanding and appreciation of the complexity of genomic and prophylactic decision-making and that those facing similar decisions might find reassurance, support and encouragement through shared experience.

I would also like to thank my research supervisors, Dr Noelle Robertson and Dr Gareth Morgan. Noelle, your knowledge within health psychology has been second to none and your guidance and encouragement has been invaluable in fuelling my desire to work within this area. Gareth, I have greatly appreciated your thoughtful and comprehensive feedback in the development of my empirical work and our discussions have been paramount in facilitating my understanding of new ideas and perspectives. My sincere thanks are also extended to my field supervisor¹, whose dedication to their service, and those whom it supports, has been both admirable and inspiring. I have been so very fortunate to have your enthusiasm and continued support of my research.

Finally, I would like to offer my heartfelt thanks to my wonderful family. You raised me to believe that anything is possible and this whole journey has been guided by your unconditional love, support and encouragement. Special thanks to my amazing husband, my rock, who has always believed in me, even at times when I didn't always believe in myself. I am so very grateful to you all and couldn't have done this without you.

¹ To ensure anonymity of the service, it is with regret that this supervisor could not be named.

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Part 1: Literature Review

Psychosocial predictors of prophylactic mastectomy in unaffected BRCA gene alteration carriers: A systematic review of the literature

This literature review has been prepared with consideration to
author guidelines for the Journal of Genetic Counseling

Guidelines can be found in Appendix A

Abstract

Cancer, an overarching descriptor of related diseases, is amongst the leading causes of morbidity and mortality worldwide (World Health Organisation, 2016). Extensive research over the last three decades has revealed carcinogenic risk factors, and there is growing evidence that some forms of the disease, notably breast cancer, have a strong hereditary link (NHS Choices, 2015). If individuals are found to have a gene alteration that suggests increased likelihood of developing breast cancer, preventative prophylactic surgery is a treatment option offered to mitigate risk.

The current review seeks to explore psychosocial factors that may influence the uptake of prophylactic mastectomy in women with a confirmed BRCA gene alteration, but no current diagnosis of cancer. In March 2015, August 2016 and February 2017, five databases (PsychoINFO, Medline, Scopus, Web of Science, CINAHL) were systematically searched. Studies were included and reviewed further if they were empirical, quantitative, peer reviewed, written in English and published between 2004 and 2017.

Ten observational cohort studies met the specified inclusion criteria and results were synthesised into temporal, familial and other factors. Significant predictors of prophylactic mastectomy included; being younger in age, having a first or second degree relative affected by cancer, having borne at least one child, previous illness or surgery, and having a greater perceived fear of cancer.

The relative scarcity of published research, and an accompanying dominant biomedical focus, suggests that examination of psychological predictors of prophylactic surgery is still in its infancy. It is imperative to gain a greater understanding of factors influencing women as they debate prophylactic mastectomy. Examination of predictive factors, particularly those which are modifiable, may contribute to effective, empathetic and person-centred support.

1. Introduction

1.1 Familial breast cancer

Cancer, an overarching descriptor of related diseases, is amongst the leading causes of morbidity and mortality worldwide (World Health Organisation, 2016). For women, breast cancer is the most common form of cancer in both the developed and developing world and, in the United Kingdom, a woman's lifetime risk of developing breast cancer is thought to be 1 in 8 (Breast Cancer Care, 2016; Cancer Research UK, 2016).

Research over the last three decades has increasingly identified risk factors for cancer, and although not usually inherited, there are some forms of the disease, notably breast, ovarian and prostate cancer, that can have a strong hereditary link (NHS Choices, 2015). Whilst genes can protect against cancer and work by correcting any naturally occurring damaged cells, mutations or gene alterations can prevent production of the required proteins to repair damage and, over time, these cells can build and potentially form a tumour (Gudmundsdottir & Ashworth, 2006). BRCA1 and BRCA2 (**BR**east **CA**ncer), located on chromosome 17 and 13 respectively (Sharpe & Carter, 2006) are two such identified genes. Women with an inherited BRCA1 gene alteration have a 60 to 90 *per cent* chance of developing breast cancer during their lifetime, whilst risk is estimated to be 45 to 85 *per cent* for those with the BRCA2 gene alteration (NHS Choices, 2015; The Royal Marsden NHS Foundation Trust, 2013). Together, BRCA1 and BRCA2 gene alterations account for approximately 20%-25% of hereditary breast cancers and approximately 5%-10% of all breast cancers (National Cancer Institute, 2015).

Since the discovery of the BRCA1 gene in 1994, significant medical advances have made genetic testing increasingly available to many women worldwide. Those who may consider themselves at risk, are generally eligible to have a genetic test if there is a strong history of cancer within the family or if a gene alteration has already been identified in a relative. Individuals are invited to meet with a genetic counsellor where a thorough family pedigree is collaboratively developed and reviewed. A diagnostic blood test may then be requested to provide a more conclusive indication or prediction of genetic risk. Individuals who are found to have an inherited alteration in the BRCA1 or BRCA2 gene, and who do not have a current diagnosis of breast cancer, are often referred to as unaffected or asymptomatic carriers. The identified presence of a BRCA gene alteration

alone does not mean that the individual will develop breast or ovarian cancer, however, it can be used to assist women in making further informed decisions about management.

1.2 Preventative options

Guidelines published by the National Institute for Health and Care Excellence (NICE, 2013) recommend specific treatment options for women following confirmation of a BRCA gene alteration, falling into three broad categories. The first option is surveillance which includes regular self-checking of breast tissue, annual mammograms and magnetic resonance imaging (MRI) scans, advised to achieve early detection of any potentially cancerous tissue. The second option, chemoprevention, often utilising Tamoxifen and Raloxifene (NICE, 2013), works to prevent hormone-responsive tumours (Lee *et al.*, 2008). The third option is risk-reducing surgery, which is an area of increasing public awareness and uptake. This may involve partial breast tissue removal for early evidenced cell growth (a lumpectomy), or, more frequently undertaken, prophylactic mastectomy. Healthcare professionals are advised to explicitly discuss the risks and benefits of preventative prophylactic mastectomy with women who have a known or suspected BRCA1 or BRCA2 gene alteration (NICE, 2013, p39).

The use of prophylactic mastectomy has burgeoned in the last two decades, possibly reflecting increased public awareness of familial breast cancer, and increased availability of breast reconstruction (Ghosh, 2002). Recent NICE guidelines, public health campaigns and high profile celebrity exposure, may also contribute (Evans *et al.*, 2014). Of the surgical options for mastectomy, total bilateral prophylactic mastectomy shows the greatest efficacy, potentially reducing the risk of developing breast cancer by up to 90% (National Cancer Institute, 2013; NHS Choices, 2015).

1.3 Decision-making

As awareness and uptake of prophylactic mastectomy has increased, there has been a parallel growth in research examining women's satisfaction following surgery, particularly in areas such as breast reconstruction, body image and pain. Whilst these studies are highly important to inform medical aftercare and support, less attention has been paid to the psychological considerations that may affect women's decision making regarding risk-reducing surgery.

Decision-making, particularly in healthcare, is increasingly realised to be complex. Traditional medical conceptualisations construct a shared exchange of information as a dyadic process (Charles *et al.*, 1997). However, for prophylactic surgery, decision-making is likely to extend beyond patient and clinician to include family and friends in evaluating some of the costs and benefits of proceeding with surgery. Thus, decision-making may not simply be an individual's response, but a more complex, emotionally charged process and one in which healthcare professionals' expertise may be challenged (Eddy, 1990). That this process is complex is attested to by the number of psychological theories applied to understand decision-making and predict adherence to health-protective behaviours. Amongst these is Protection Motivation Theory (Rogers, 1983) which highlights the importance of how individuals appraise threat, their sense of personal vulnerability and perceived ability to cope. Such models of decision-making and cost-benefit analysis (embodied in Becker's Health Belief Model, 1974) may have utility, although they imply rational cognition and distinct linear stages that may under privilege emotional response and iterative processes (Charles *et al.*, 1997), and potentially diminish the role of social and environmental factors (Ogden, 2012).

More recent theories have suggested that emotion and subjective experience are not in conflict with objective cognitive appraisal of risk and health related valuations but can instead facilitate decision-making (Reyna, 2015). Fuzzy Trace Theory is a dual process approach that brings together the concepts of mental representation, emotion and moral values (Reyna, 2008). The theory argues two types of mental representation: verbatim (surface information such as words, numbers or pictures) and gist (the personal meaning of information to an individual). Although individuals may encode these representations in parallel, it is suggested that people generally favour gist processing (Corbin *et al.*, 2015). For example, an individual being informed they have a 60% chance of developing cancer may engage beyond this verbatim figure to make their own subjective interpretation (gist) of what this information means to them.

In light of this complexity, greater awareness of factors which may correlate with, or predict decision-making is warranted, not least so that healthcare professionals are best able to support and facilitate potentially difficult medical decisions (Power, 2011).

1.4 Aims

The aim of the current review was thus to critically examine quantitative studies that explore psychosocial predictors of prophylactic surgery in BRCA gene alteration carriers. This was felt to be important because as more women become aware of, and choose to have, genetic testing, risk-reducing surgery referrals are likely to increase. Previous research and reviews to date have primarily examined uptake processes through referral statistics and retrospective satisfaction with surgery. This is suggestive of an area with a predominantly biomedical focus where psychological predictors of prophylactic surgery are still largely unexamined. However, greater understanding of psychosocial correlates and predictors of prophylactic surgery is of clinical importance and integral to individualised support during potentially life changing decision-making.

2. Method

2.1 Search strategy

Literature pertinent to the research question was examined with adherence to a systematic search process. Google Scholar was also initially utilised alongside contact with two prominent clinicians and authors in the field of genomics and prophylactic surgery. Academic databases: PsychoINFO, Medline, Scopus, Web of Science and CINAHL, were interrogated (with a specialist librarian who reviewed proposed search terms and databases), because of their likely focus on psychological and medical factors in decision-making. Searches were conducted in March 2015, August 2016 and February 2017. Searches initially involved broader areas of risk reducing surgery and BRCA mutations to gain greater understanding of the wider research context, before being refined specifically to prophylactic mastectomy and those with an unaffected BRCA gene alteration status. Search terms were grouped to include a combination of the main focus areas: BRCA gene alterations (BRCA*; mutation; alteration; genetic*; unaffected; breast cancer), prophylactic mastectomy (prophyla*; mastectom*; RRM; surg*; risk reduc*) and psychosocial factors (psych*; impact; decision*, predict*). Please see Appendix B for a full list of search terms.

2.2 Eligibility criteria

The trainee and supervisor screened the titles of 2642 articles identified through database searches. Only those papers meeting the following criteria were then subject to further review:

Inclusion criteria

- Peer reviewed, English language articles.
- Published from 2004 to present (February 2017) - Consistent with the National Institute for Health and Care Excellence's (NICE) first publication of familial breast cancer guidelines (CG14 - Familial breast cancer; classification and care of women at risk of familial breast cancer in primary, secondary and tertiary care, 2004).
- Quantitative methodology - Whilst initially scoping the available literature, it was evident that qualitative research within this area is still in its infancy. It was therefore decided that, in order to review a greater number of relevant studies, with larger participant samples and firmer operationalised predictors of prophylactic surgery, the current review would focus on studies adhering to a quantitative methodology.
- Adult female participants only – Aged 18 years and over (no upper age limit).
- Sample participants must have confirmation of an inherited BRCA gene alteration (BRCA1 or BRCA2).
- Focus on prophylactic mastectomy - additional exploration of oophorectomy may be included; however, the analysis must be clearly separated.
- Studies that focus on psychosocial predictors of prophylactic mastectomy.

Exclusion criteria

- Reviews, case studies or other non-empirical discursive papers.
- Articles published prior to 2004.
- Qualitative methodology.
- Male participants or female participants under 18 years of age.
- Inconclusive or unconfirmed BRCA gene alteration status.

- Participants with a current diagnosis of breast cancer – individualised risk management for those with cancer more likely to advise mastectomy, presenting a potential confound highly likely to influence decision making.
- Studies that focus on contralateral mastectomy or oophorectomy alone.
- Studies where the sole focus is on genetic testing, uptake figures, treatment adherence, surgical reconstruction or mortality rates.

After removing duplicates and applying the inclusion criteria, 205 abstracts were then read for relevance to the research question. Reference sections of these articles were also examined to identify other potentially relevant publications. After reading the abstracts, and where necessary, scanning the full articles, 165 papers were further excluded based on non-adherence to the inclusion criteria. 40 papers were obtained in full text and read thoroughly, from which 10 articles were selected as most relevant to the research question.

2.3 Data extraction and quality appraisal

Figure 1 provides a summary of the search process for the current review and outlines the number of articles identified and excluded at each stage. A data extraction form (Appendix C) was used to assist in identifying the salient features of each paper including aspects such as: study design, sample characteristics, outcome measures, statistical analysis, results and conclusions. This data extraction form was based on recommendations from the Centre for Reviews and Dissemination (2009) handbook for undertaking systematic literature reviews in healthcare.

The ten shortlisted papers were then individually appraised to provide a consistent analytical framework of quality. Previous research into quality assessment in observational studies has found heterogeneity in tools used and there has often been lack of consensus over recommendations (Sanderson, Tatt & Higgins, 2007). Kmet, Lee and Cook (2004), reviewed the strengths and limitations of various quality assessment tools and developed their own checklist, QualSyst (Appendix D) that can be utilised to review a diverse range of study designs. The QualSyst provides a consistent system for evaluating methodology in relation to study aims and therefore focuses on internal rather than external validity (Harbour & Miller, 2001) which can be pertinent to observational studies and considerations of generalisability.

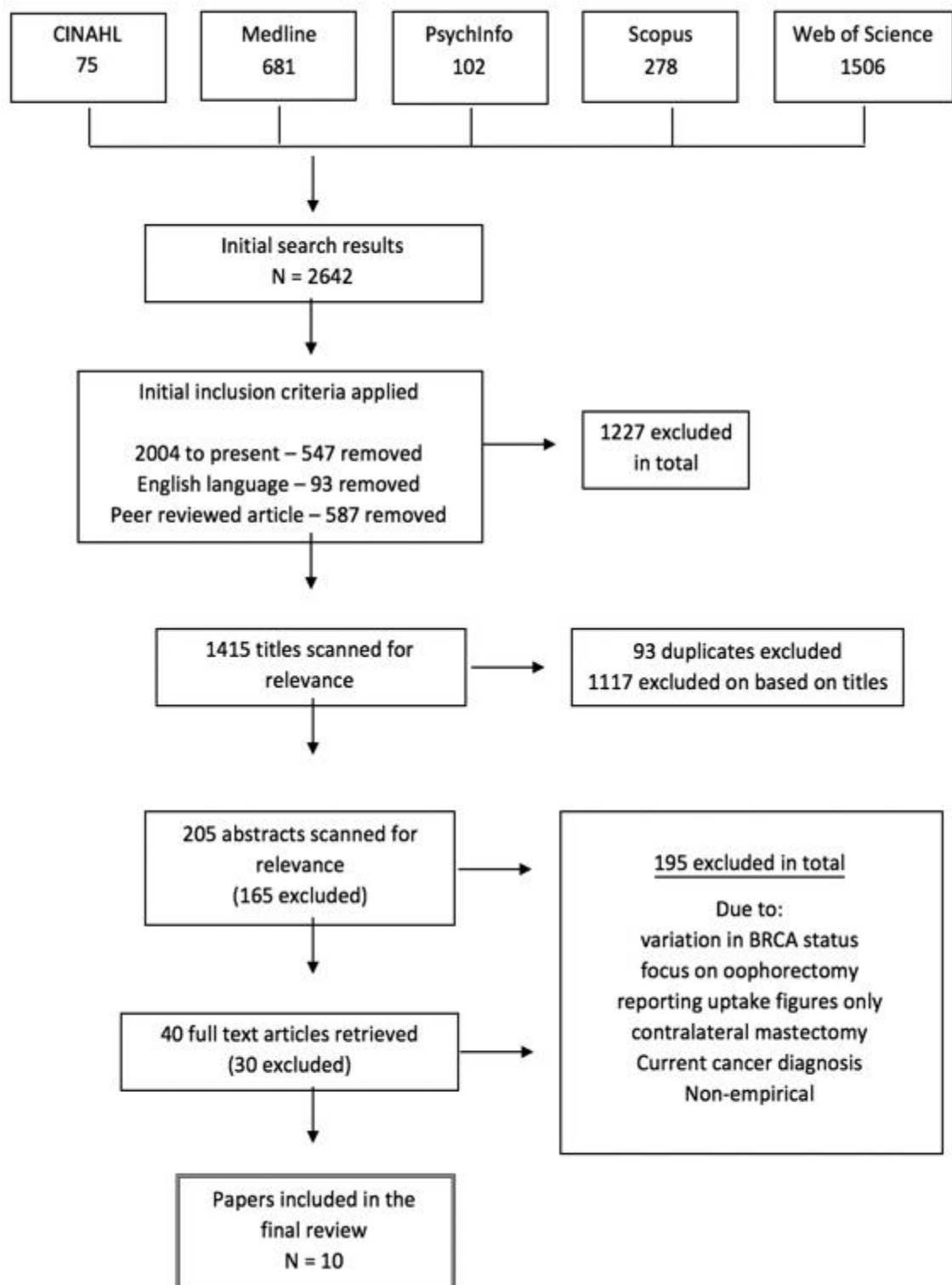


Figure 1. Systematic review process

The QualSyst includes fourteen items whereby studies are assessed as meeting (assigned a score of two), partially meeting (assigned a score of one) or not meeting (assigned a score of zero) items included on the checklist. The overall score for a study is calculated by dividing the obtained number of scores by the total number of possible scores; this provides a score that can range between 0 and 1. Items deemed non-applicable to a study design can be marked 'n/a' and excluded from the calculation summary score. The QualSyst was used to guide unbiased judgement and consistent evaluation of the papers included in the review (see Appendix E for scoring table). The criteria included in the tool has been found to demonstrate good inter-rater reliability with item agreement ranging from 73% to 100% (Kmet *et al.*, 2004). Half of the papers in this review were randomly selected and independently scored by the supervising researcher for the purposes of reliability and validity. Little discrepancy was found and further discussion achieved consensus.

The reported methodological procedures of the reviewed studies were generally fair, with scores between 0.85 and 0.95 as identified by the QualSyst. Given the paucity of research meeting the inclusion criteria, no studies were excluded based on these scores alone, however judgement of quality was considered in order to aid interpretation of results. Quality assessment did highlight minor limitations of papers, and, in two cases, further information was sought where particular aspects of recruitment and follow-up were not clear. All studies gathered data from electronic medical databases, with only three electing to add participant questionnaires, therefore the potential for further quality limitations and confounding variables were low. Although all studies were considered appropriate for statistical analysis, it was noted that none reported a-priori power calculations beforehand to determine sufficient sample sizes required.

Table 1. Characteristics of the shortlisted studies

Study details	Aims	Participants	Methodology & Measures	Analysis	Results & conclusions
Beattie, M. S., Crawford, B., Lin, F., Vittinghoff, E. & Ziegler, J. (2009). Uptake, time course and predictors of risk-reducing surgeries in BRCA carriers. <i>Genetic Testing and Molecular Biomarkers</i> , 13(1), 51–56.	To determine the uptake, time course of risk-reducing mastectomy and risk reducing oophorectomy in BRCA mutation carriers.	237 BRCA mutation carriers. No current diagnosis of cancer. No previous prophylactic surgery.	Observational cohort study – prospective. Participants recruited from 3 clinical sites in the California Risk Programme. Medical records and demographics questionnaire. Follow-up by post, over the phone and in person.	Kaplan-Meier curves estimating the cumulative proportions of those having prophylactic mastectomy. 54 had prophylactic mastectomy. Average time from genetic test to surgery = 3 months.	Three significant predictors – age below 60, prior history of breast cancer and other prophylactic surgery.
Chai, X., Friebel, T. M., Singer, C. F., Evans, D. G., Lynch, H. T., Isaacs, <i>et al.</i> , (2014). Use of risk-reducing surgeries in a prospective cohort of 1,499 BRCA1 and BRCA2 mutation carriers. <i>Breast Cancer Research and Treatment</i> , 148(2), 397–406.	To determine the usage of risk-reducing mastectomy and risk-reducing oophorectomy to obtain a better estimate of cancer preventive strategies.	1499 BRCA mutation carriers. No current diagnosis of cancer. No previous prophylactic surgery.	Observational cohort study – prospective. Participants recruited from 20 clinical sites in the PROSE consortium. Medical records database.	Kaplan-Meier curves estimating the cumulative proportions of those having prophylactic mastectomy. Log rank test to assess difference between strata. 171 had prophylactic mastectomy. 164 had prophylactic	Prophylactic mastectomy as frequent in BRCA1 and BRCA2 mutation carriers (not significant). Those with four or more children more likely to have prophylactic mastectomy (significant).

				mastectomy and oophorectomy. Mean age at surgery = 37.4 years old.	
Friebel, T. M., Domchek, S. M., Neuhausen, S. L., Wagner, T., Evans, D. G., Isaacs, C., <i>et al.</i> , (2007). Bilateral prophylactic oophorectomy and bilateral prophylactic mastectomy in a prospective cohort of unaffected BRCA1 and BRCA2 mutation carriers. <i>Clinical Breast Cancer</i> , 7(11), 875–82.	To explore factors affecting the uptake of prophylactic mastectomy and prophylactic oophorectomy in unaffected mutation carriers.	537 BRCA mutation carriers. No current diagnosis of cancer. No previous prophylactic surgery.	Observational cohort study – prospective. Participants recruited from 17 clinical sites in the PROSE consortium. Medical records and demographics questionnaire. Follow-up by post, over the phone and in person.	Logistic regression of predictors (multivariate and univariate). 35 had prophylactic mastectomy. 78 had prophylactic mastectomy and oophorectomy. Average time from genetic test to surgery = 13.6 months. Mean age at surgery = 41.3 years old.	Univariate predictors included age at genetic testing, parity and having a relative with breast cancer. Prophylactic oophorectomy performed more frequently than mastectomy (significant). Significantly higher uptake in the UK centres.
Garcia, C., Wendt, J., Lyon, L., Jones, J., Littell, R. D., Armstrong, M. A., <i>et al.</i> , (2014). Risk management options elected by women after testing positive for a BRCA mutation. <i>Gynecologic Oncology</i> , 132(2), 428–433.	To assess the uptake of risk reducing options for the management of breast and ovarian cancer risk in BRCA mutation carrier.	250 BRCA mutation carriers. No current diagnosis of cancer. No previous prophylactic surgery.	Observational cohort study – retrospective. Multiple clinical sites within the California Health System (Kaiser Permanente Northern California – KPNC). Medical records database.	Logistic regression of predictors. T-tests 110 had prophylactic mastectomy. Average time from genetic test to surgery = 6 months.	Significant predictor – those with a previous history of breast cancer. No significant difference in uptake between those with BRCA1 or BRCA2. Those who had prophylactic

					mastectomy 3.79 times more likely to have oophorectomy.
Haroun, I., Graham, T., Poll, A., Sun, P., Hill, K., Weitzner, <i>et al.</i> , (2011). Reasons for risk-reducing mastectomy versus MRI-screening in a cohort of women at high hereditary risk of breast cancer. <i>The Breast</i> , 20(3), 254–258.	To determine the reasons that motivate women in a cohort of intensive surveillance for breast cancer to undergo risk-reducing mastectomy.	246 BRCA mutation carriers. No current diagnosis of cancer.	Observational cohort study – retrospective. Multiple clinical sites – enrolled in MRI surveillance programmes. Medical records and perceived risk questionnaire.	T test for continuous variables. Chi-squared test for categorical variables. 39 had prophylactic mastectomy. Average time from genetic test to surgery = 42 months. Mean age at surgery = 42.3 years old.	50% or greater perceived risk of breast cancer more likely to have prophylactic mastectomy (significant). Women with mother or sister with breast cancer more likely to have surgery (not significant).
Hesse-Biber, S. & An, C. (2016). Genetic testing and post-testing decision making among BRCA-positive mutation women: A psychosocial approach. <i>Journal of Genetic Counseling</i> , 25(5), 978–992.	To use a social constructionist, feminist lens to understand the decision-making process, risk perception and treatment options.	303 BRCA mutation carriers. No current diagnosis of cancer.	Observational cohort study – retrospective. Online survey Impact of Cancer Scale (MICRA) and Self-Concept Scale. Both reconstructed for study but tested for validity/reliability.	Multiple regression to explore predictors. 140 had prophylactic mastectomy	Significant predictors = more distress, medical uncertainty, guilt over genetic link and involvement (life more effected by BRCA status)

<p>Julian-Reynier, C., Bouhnik, A.D., Mouret-Fourme, E., Gauthier-Villars, M., <i>et al.</i>, (2010). Time to prophylactic surgery in BRCA1/2 carriers depends on psychological and other characteristics. <i>Genetics in Medicine</i>, 12(12), 801–807.</p>	<p>To investigate the medical and psychosocial factors determining the time to prophylactic surgery of unaffected carriers of a deleterious BRCA 1/2 mutation.</p>	<p>244 BRCA mutation carriers.</p> <p>No current diagnosis of cancer.</p>	<p>Observational cohort study – prospective.</p> <p>Participants recruited from 29 clinical sites.</p> <p>Medical records and psychological impact questionnaire.</p>	<p>Kaplan-Meier curves estimating the cumulative proportions of those having prophylactic mastectomy.</p> <p>Log rank test</p> <p>8 had prophylactic mastectomy. 12 had had prophylactic mastectomy and oophorectomy. Average time from genetic test to surgery = 25 months. Mean age at surgery = 38.8 years old.</p>	<p>Significant factors = 1st degree relative with breast cancer, having a child younger than 15years old.</p> <p>Borderline significance = avoidance, intrusion and body image.</p>
<p>Metcalf, K., Foulkes, W., Kim-Sing, C., Ainsworth, P., Rosen, B., Armel, <i>et al.</i>, (2008). Family history as a predictor of uptake of cancer preventive procedures by women with a BRCA1 or BRCA2 mutation. <i>Clinical Genetics</i>, 73(5), 474-479.</p>	<p>To investigate the extent to which family history influences the uptake of cancer preventive options.</p>	<p>517 BRCA mutation carriers.</p> <p>No current diagnosis of cancer.</p>	<p>Observational cohort study – prospective.</p> <p>Participants recruited from 11 clinical sites.</p> <p>Medical records and demographics questionnaire.</p>	<p>Logistic regression to estimate risk factors.</p> <p>Chi squared t-test</p> <p>81 had prophylactic mastectomy. Mean age at surgery = 42.1 years old.</p>	<p>Those with a sister with breast cancer twice as likely to have prophylactic mastectomy (significant).</p> <p>Those with a mother also more likely but not significant.</p>

<p>Singh, K., Lester, J., Karlan, B., Bresee, C., Geva, T. & Gordon, O. (2013). Impact of family history on choosing risk-reducing surgery among BRCA mutation carriers. <i>American Journal Obstetric Gynaecology</i>, 208(4), 329.e1-6.</p>	<p>To investigate factors that might influence decision making regarding prophylactic surgeries in women with BRCA mutations.</p>	<p>136 BRCA mutation carriers.</p> <p>No current diagnosis of cancer.</p> <p>No history of cancer.</p>	<p>Observational cohort study – prospective.</p> <p>Single site only – clinic in Los Angeles.</p> <p>Medical records database.</p>	<p>Logistic regression to explore predictive factors.</p> <p>57 had prophylactic mastectomy.</p> <p>Mean age at surgery = 43.7 years old.</p>	<p>Significant predictors = Having lost a mother to breast cancer and having a first or second degree relative who passed away from cancer.</p> <p>Those with at least one child more likely to have prophylactic mastectomy.</p>
<p>Skytte, A. B., Gerdes, A. M., Andersen, M. K., Sunde, L., Brondum-Nielsen, K, <i>et al.</i>, (2010). Risk-reducing mastectomy and salpingo-oophorectomy in unaffected BRCA mutation carriers: Uptake and timing. <i>Clinical Genetics</i>, 77(4), 342–349.</p>	<p>To outline the uptake of risk reducing surgery in BRCA mutation positive women and search for factors affecting the decision.</p>	<p>306 BRCA mutation carriers.</p> <p>No current diagnosis of cancer.</p> <p>No history of cancer.</p>	<p>Observational cohort study – retrospective.</p> <p>Multiple clinical sites in Denmark.</p> <p>Medical records database.</p>	<p>Multivariate cox regression to explore predictive factors.</p> <p>Uptake of prophylactic mastectomy was 50% at 10 years.</p> <p>Average time from genetic test to surgery = 92 months.</p> <p>Mean age at surgery = 37 (BRCA1) and 38 years (BRCA2) old.</p>	<p>Younger age was associated with higher uptake of prophylactic mastectomy (significant).</p> <p>Women with children had a significantly higher uptake of prophylactic mastectomy.</p>

3. Results

3.1 Study characteristics

The main characteristics and findings of the ten shortlisted studies are shown in Table 1. All papers were longitudinal observational cohort studies, five of which utilised a prospective design (Beattie *et al.*, 2009; Chai *et al.*, 2014; Friebel *et al.*, 2007; Julian-Reynier *et al.*, 2010; Singh *et al.*, 2013) and three, a retrospective design (Haroun *et al.*, 2011; Hesse-Biber & An., 2016; Skytte *et al.*, 2010). The remaining two studies used both prospective and retrospective data collection (Garcia, *et al.*, 2014; Metcalfe *et al.*, 2008). Four studies (Beattie *et al.*, 2009, San Francisco; Garcia *et al.*, 2014, California; Hesse-Biber & An., 2016; Singh *et al.*, 2013, Los Angeles) were carried out in America, two in Canada (Haroun *et al.*, 2011; Metcalfe *et al.*, 2008) and two in Europe (Julian-Reynier *et al.*, 2010, France; Skytte *et al.*, 2010, Denmark). The remaining two (Chai *et al.*, 2014; Friebel *et al.*, 2007) included up to 20 clinics in the Prevention and Observation of Surgical Endpoints (PROSE) consortium across North America and Europe. Nine studies included participants drawn from multiple clinical sites, with only one (Singh *et al.*, 2013) utilising information from a single clinic site.

Nine studies explored psychosocial factors that may affect the uptake of both prophylactic mastectomy and prophylactic oophorectomy (the surgical removal of the ovaries). As stated in the methods section, these studies were included as the data analysis and discussion permitted examination of factors pertaining to mastectomy. Only Haroun *et al.*, (2011) explored prophylactic mastectomy alone.

3.2 Participants

The total number of women with an inherited BRCA gene alteration across the ten studies was 4275, with participant samples ranging from 136 (Singh *et al.*, 2013) to 1499 (Chai *et al.*, 2014). Due to the longitudinal nature of the ten studies, all participants were commonly followed up until; they had prophylactic surgery, received a diagnosis of breast cancer, expressed a wish to withdraw from the research or the participant died.

Only six studies specified the ages of eligible participants in their methodology section. Two reported this as an age range; Haroun *et al.*, (2011) recruited participants from the

ages of 25–65 years and Metcalfe *et al.*, (2008) from the ages of 25–80 years. Garcia *et al.*, (2014) and Julian-Reynier *et al.*, (2010) both included participants from 18 years old and upwards, however no upper age limit was specifically reported. Similarly, Skytte *et al.*, (2010) included participants from 25 years and Friebe *et al.*, (2007) from 30 years, with no upper age limit reported.

All study samples comprised a majority of Caucasian women, 96% in Hesse-Biber and An (2012) and 85% in Singh *et al.*, (2013), such that generalisability is constrained by demographics not controlled for in the participant sampling frame. Furthermore, Singh *et al.*, (2013) reported on a participant sample that included 96 (71%) women who categorised themselves as Ashkenazi Jewish, known to present with increased incidence of specific genetic alterations for breast cancer (Waller & McPherson, 2003).

Although none of the participants across the ten studies had breast cancer at the time of recruitment, there was some variation in whether participants had previously received a diagnosis of cancer. Two studies (Beattie *et al.*, 2009; Garcia *et al.*, 2014) included personal history of cancer as a predictor variable. However, Singh *et al.*, (2013) and Skytte *et al.*, (2010) state that those with a prior history of cancer were purposefully excluded as a potential confound. Furthermore, six studies specifically excluded those who had previously undergone risk-reducing surgery (mastectomy and/or oophorectomy). The remaining four studies (Haroun *et al.*, 2011; Hesse-Biber & An., 2016; Julian-Reynier *et al.*, 2010; Metcalfe *et al.* 2008) made no reference to this therefore the potential impact of prior prophylactic surgery experience is unclear.

3.3 Data collection

Demographic characteristics were reported in all studies, including; age, ethnicity, marital status, parity, level of education, occupation, religion and socioeconomic status. Some of these details were utilised alongside additional health information such as: prior cancer or surgery, BRCA status (BRCA1 or BRCA2) and having a relative affected by cancer, as predictor variables that may impinge upon decision-making.

Four studies (Chai *et al.*, 2014; Garcia *et al.*, 2014; Julian-Reynier *et al.*, 2010; Skytte *et al.*, 2010) primarily utilised electronic medical records to gather reliable and up-to-date information, participant health management and family history, and to reduce bias from

personal recall. Three studies (Beattie *et al.*, 2009; Friebe *et al.*, 2007; Metcalfe *et al.*, 2008) requested that participants complete demographic and medical history questionnaires alongside information taken from patient medical records, allowing both personal and clinical information to be collated.

Haroun *et al.*, (2011), Julian-Reynier *et al.*, (2010) and Hesse-Biber and An, (2016) gathered information from medical records but also asked participants to complete outcome measures alongside this. Methods differed: Beattie *et al.*, (2009) stated that questionnaires were either sent by post, completed over the telephone or face to face, whereas it was not clear how questionnaires were administered in the Julian-Reynier *et al.*, (2010) study. Hesse-Biber *et al.*, (2016) requested participants complete an online survey, but did not specify the geographic reach of websites. Expressed preference for sampling methods was not noted however, it is important to consider potential social desirability effects and differences in personal comfort when sharing information through different mediums (Tourangeau & Yan, 2007).

Haroun *et al.*, (2011) developed their own written questionnaires that included assessment of satisfaction with screening/surveillance options, estimates of personal cancer risk and reasons for undergoing prophylactic mastectomy, with few details offered regarding piloting, validity and reliability. Julian-Reynier *et al.*, (2010) utilised existing assessment measures; the Centre for Epidemiological Studies Depression Scale (CES-D), Impact of Life Events Scale (IES) and the Breast and Body Image Scale (BBIS) and reported internal reliability (CES-D, Cronbach's $\alpha = 0.93$; IES $\alpha = 0.91$; BBIS, Cronbach's $\alpha = 0.83$). Similarly, Hesse-Biber and An (2016) utilised two existing psychometric measures, with modifications to make them more 'statistically and theoretically meaningful' to their study. Reported reliability analyses revealed that they were moderately to highly reliable; Multidimensional Impact of Cancer Risk Assessment (MICRA, Cronbach's $\alpha = 0.5 - 0.8$) and the Self-Concept Scale (SCS, Cronbach's $\alpha = 0.7 - 0.8$).

All papers examined psychosocial predictors of prophylactic mastectomy in unaffected BRCA gene alteration carriers. Whilst the decision to undergo prophylactic surgery is an individual and unique consideration for any woman, this review revealed consistent correlates of uptake of prophylactic mastectomy. Study heterogeneity precluded meta-analysis, therefore, narrative synthesis was chosen to summarise findings. Through this

synthesis three domains were revealed; temporal factors (including age at surgery and time to surgery), familial factors (including parity and having a relative affected by cancer) and other factors (including perception of risk and previous illness or surgery).

3.4 Findings

3.4.1 Temporal factors

Age at surgery – All studies reported the mean age of those who underwent prophylactic mastectomy, with the majority of participants having had surgery between the ages of 38 and 43 years (Friebel *et al.*, 2007; Garcia *et al.*, 2014; Julian-Reynier *et al.*, 2010; Metcalfe *et al.*, 2008; Singh *et al.*, 2013; Skytte *et al.*, 2010). Three studies (Beattie *et al.*, 2009; Garcia *et al.*, 2014; Skytte *et al.*, 2010) reported age as a significant predictor of prophylactic mastectomy uptake with two of these revealing increased age as being significantly correlated with reduced uptake of elective mastectomy. Beattie *et al.*, (2009), reported women 60 years and older had the lowest uptake of prophylactic mastectomy, and Skytte *et al.*, (2010) reported younger age as being significantly associated with higher prophylactic mastectomy uptake, with the greatest number of procedures conducted in those under 40 years.

Two studies (Chai *et al.*, 2014; Skytte *et al.*, 2010) additionally explored whether the specific BRCA gene alteration (either BRCA1 or BRCA2) appeared related to the age of participants when they had surgery. Both found that participants were younger within the BRCA1 mutation category (between 30-37 years) when compared with those with the BRCA2 mutation (35-40 years).

Time to surgery – Eight studies reported data on the average time taken from receiving confirmation of a BRCA gene alteration to having prophylactic surgery. Seven studies reported that the majority of participants had prophylactic surgery within two years of genetic testing. By contrast, Skytte *et al.*, (2010) noted a lengthier overall median time between testing and prophylactic mastectomy of 92 months (7.6 years). The researchers did however note a reduced median time to surgery of 67 months (5.5 years) in those below the age of 40, perhaps suggesting that younger participants elected to have surgery sooner.

Also related to temporal considerations, Hesse-Biber and An (2016) reported that being older in age at the time of genetic testing was significantly related to having prophylactic surgery sooner. Furthermore, Julian-Reynier *et al.*, (2010), reported that time to surgery was significantly shorter when breast cancer had been diagnosed in a first degree relative before the age of 50 years.

3.4.2 Familial factors

Relative affected by cancer – Five studies explored whether having a relative with a diagnosis of cancer was associated with uptake of prophylactic mastectomy in unaffected BRCA gene alteration carriers. Friebel *et al.*, (2007), Julian-Reynier *et al.*, (2010) and Singh *et al.*, (2003) reported that women with a first and/or second degree relative had a significantly higher uptake of prophylactic mastectomy. Singh *et al.*, (2003) utilised regression analysis and found that losing a mother to breast cancer was a significant predictor of prophylactic mastectomy. Metcalfe *et al.*, (2008) however found the most significant predictor of prophylactic surgery was having a sister who had been diagnosed with breast cancer, and that those with an affected sister were twice as likely to opt for prophylactic mastectomy than those whose sister was unaffected.

Parity – Eight studies explored whether parity (having borne children) was associated with prophylactic surgery in unaffected BRCA gene alteration carriers. Chai *et al.*, (2014) reported that women with four or more children had a significantly higher uptake of prophylactic mastectomy, than women with one, two or three children. Singh *et al.*, (2013) utilised regression analysis and found that having borne at least one child was a significant predictor of surgery, and that women who declined surgery were less parous or nulliparous. In addition, Hesse-Biber and An (2016) reported a significant difference in uptake rates between those who had daughters and those who had only sons, the former opting for mastectomy more frequently. Julian-Reynier *et al.*, (2010) also found that women who had children below the age of 15 had a significantly higher uptake of prophylactic mastectomy.

3.4.3 Other factors

Perception of risk – Although most studies considered risk perception in their discussion, only two specifically explored this within their analysis. Haroun *et al.*, (2011) reported that those who perceived their risk of developing breast cancer to be 50% or greater were significantly more likely to have prophylactic mastectomy. Julian-Reynier *et al.*, (2010) asked participants to rate their perceived risk of cancer prior to genetic testing and found that 57% believed that their lifetime risk was ‘high’. Higher perceived risk was positively correlated with uptake of prophylactic mastectomy.

Hesse-Biber and An (2016) found that reported guilt (of passing on the BRCA gene alteration), negative rumination and fear were all significant predictors of having prophylactic mastectomy. Distress and uncertainty, as determined by the Multidimensional Impact of Cancer Risk Assessment (MICRA), were however found to be significant negative predictors of prophylactic mastectomy, suggesting that the more distressed and uncertain women were regarding their decision, the less likely they were to proceed with prophylactic surgery.

Previous illness or surgery – Two studies included previous diagnosis of cancer as a variable potentially affecting the decision to undergo prophylactic mastectomy. Beattie *et al.*, (2009) reported participants with a history of breast cancer prior to genetic testing had a significantly higher uptake of prophylactic surgery. Similarly, Garcia *et al.*, (2014) utilising logistic regression, found that participants receiving a prior diagnosis of breast cancer were 2.96 times more likely to choose prophylactic mastectomy than women with no such personal history.

Four studies also explored whether prior prophylactic oophorectomy (the surgical removal of the ovaries) was related to the decision to have further prophylactic surgery. Beattie *et al.*, (2009), Garcia *et al.*, (2014) and Singh *et al.*, (2013) all found that those who underwent prophylactic oophorectomy also had a significantly higher uptake of prophylactic mastectomy.

4. Discussion

4.1 Summary of findings

This systematic review examined psychosocial predictors associated with elective prophylactic mastectomy in women with a BRCA gene alteration. The three domains that results coalesced around were temporal, familial and other factors including risk and illness perception.

Reviewed studies suggested the majority of women underwent surgery within two years of genetic testing, and that those opting for prophylactic mastectomy were younger in age, with fewer women having surgery over the age of 60. Those with a BRCA gene alteration are known to have a greater risk of developing breast cancer at a younger age, compared to the general population (Royal Marsden NHS Foundation Trust, 2013) and the effectiveness of screening in younger women is controversial (Rees *et al.*, 2004). It is anticipated that age related guidance would have been discussed with participants through individualised risk management, where such clinical recommendations may have also contributed towards timeframes for surgery.

The lower uptake of prophylactic surgery in those over the age of 60 may also suggest differing motivations for genetic testing, where prioritisation may be given to alerting children and grandchildren to hereditary risk. However, this may be artefactual. The studies utilised in this review appear to include fewer participants over the age of 60 (likely due to convenience and volunteer sampling effects as well as upper age exclusion limits - Haroun *et al.*, (2011)) and findings should be interpreted cautiously.

Familial factors were also found to be related to decision-making. Notably witnessing a first and/or second degree relative with cancer, led to a significantly higher uptake of prophylactic surgery. Gender seemed very salient since this was more pronounced when an individual had lost their mother or sister to breast cancer. Witnessing diagnosis, treatment and life limitation of a loved one may increase perceived risk through identification or empathy and increase motivation to take preventative action. Such experiences within a family are likely to shape illness appraisals that are understood to influence emotional and behavioural responses to risk (Wellisch *et al.*, 1996; Rees *et al.*, 2004). This also supports the Fuzzy Trace Theory of decision-making which recognises personal interpretation and emotional experience (gist) alongside verbatim information

and medical recommendations (Reyna, 2008; Corbin et al., 2015). In this instance, gist representations may involve emotional underpinnings and observations such as bereavement, anger towards cancer and fear of going through the same experience.

Parity was also found to be significant factor in women's uptake of prophylactic mastectomy. Julian-Reynier *et al.*, (2010) and Hesse-Biber and An, (2016) reported that women with younger children were more likely to opt for risk reducing surgery. Surgery may be perceived as a means to reduce disease likelihood and threats to function as a nurturer of the next, dependent generation. It was also found that those with a daughter were more likely to have elective surgery than those who had only sons. Whilst men do carry the BRCA gene, their lifetime risk of developing breast and/or prostate cancer is considerably lower than women (NHS Choices 2015; Royal Marsden NHS Foundation Trust, 2013). Whilst this may highlight some of the putative mechanisms by which decision-making might occur, these correlations do not reveal the qualitative narratives involved. For example, whether identification with a fellow woman increases salience and anxiety or that those with a daughter could experience greater fear of their child having the BRCA gene alteration and may wish to provide a positive role model for preventative surgery should their daughter face the same decision in the future.

These findings highlight that, alongside personal considerations, there is also a sense of external awareness, threat and distress in such decision-making. This supports the suggestion put forward by Eddy (1990) that decisions are seldom viewed in isolation but are instead the combination of subjective and systemic influences. For example, this may include the contemplation of how the decision will affect others and, as a result, potentially experiencing feelings of guilt, which can be a common emotion in genetic disease (Stotland & Stewart, 2001). Acknowledging the systemic nature of shared decision-making, the researcher was surprised that no studies explicitly examined relationship status and whether this was predictive of prophylactic surgery.

Greater distress, perceived risk and rumination were also found to be associated with higher uptake of prophylactic mastectomy (Haroun *et al.*, 2011; Hesse-Biber & An, 2016; Julian-Reynier *et al.*, 2010). These factors are not mutually exclusive and are likely to interact and exacerbate one another. This supports other studies (Hopwood *et al.*, 2001; Watson *et al.*, 1999) where risk perception has been positively associated with cancer

distress. Loewenstein and Lerner (2003) suggest that as emotions intensify, they exert an ever-increasing influence on behaviour.

Protection Motivation Theory (Rogers, 1983) and the Self-Regulatory Model (Leventhal *et al*, 1997) can also be used to further understand findings from the studies. Both models argue the importance of appraised severity, perceived threat and self-efficacy as being influential to health behaviour. In this instance, women may have been more inclined to take action if they believed their risk of developing breast cancer was high and if they felt confident that a preventative mastectomy would offer the greatest overall risk reduction.

4.2 Strengths and limitations of reviewed studies

A strength across all studies reviewed was the longitudinal nature of follow up. This is particularly pertinent to observational cohort studies, allowing a more comprehensive and inclusive timeframe for data collection. To carry out such research over a shorter period of time would inevitably omit data from those who may have taken longer to reach a decision or those who elected to have surgery at a point in the future. It is however also acknowledged that longitudinal timeframes could also allow increased opportunity for confounding variables such as medical and surgical advances, media awareness and changing public attitudes and trends towards risk reduction over time.

In nine of the ten studies, data were gathered from multiple sites which increases the potential for variation in clinical practice and treatment recommendations across the participant sample. Skytte *et al.*, (2010) highlighted that chemoprevention was not available in Denmark in 2010, which would have also reduced alternative options available to women participating in the study. Furthermore, there is inevitable variation in the cost of healthcare provision around the world. Such differences in the availability and affordability of treatment could impact an individual's choice regarding preventative action, particularly affecting those from less affluent socioeconomic areas and those without health insurance. It was therefore surprising to find that none of the studies specifically explored the availability and affordability of genetic testing and prophylactic surgery as potential variables that could impinge upon decision-making.

Regarding potential selection bias, all research respondents had previously accessed a healthcare centre for genetic testing, suggesting these may be women with an existing awareness of genetic risk who have initiated steps to ascertain this, may already be more inclined to act on genetic test results, and thus be more open to preventative action. Several studies included in the review were also unable to account for any further surveillance or surgical procedures that participants may have sought elsewhere. This presents an unknown variable that could have led to under-representation of preventative options during the follow-up period.

4.3 Limitations of the current review

The rigorous review procedure revealed relatively few studies (ten) specifically investigating psychosocial factors predictive of prophylactic mastectomy and suggests a research field in its infancy. The dominant discourse around preventative surgery appeared biomedical with proportionately less attention paid to potential contributory psychological and social factors.

Whilst common correlates were evidenced across the ten papers however, synthesis was constrained by heterogeneity in design methodologies, particularly variation in: the participant sample (those with/without a prior diagnosis of cancer or experience of prophylactic surgery), multisite recruitment (differing guidelines, physician recommendations and treatment availability) and data collection (postal, online, telephone and face to face). Generalisability is also constrained by samples comprising predominantly Caucasian women, and an under-representation of ethnicities who may be at more risk.

4.4 Clinical significance and future implications

With regard to how this information can be drawn together to inform avenues for future research and best practice, it is clear that this is an area of increasing interest and uptake that still requires and deserves more attention worldwide. It is important to further understand why some women choose prophylactic mastectomy but also why so many women still decline this option despite its established efficacy.

There is also a recognised need for further qualitative research in this area, where priority is given to women's own voices in sharing their understanding of factors most important to them in making decisions about prophylactic mastectomy.

In this review, findings were synthesised into three areas; temporal, familial and other factors. Whilst temporal and familial factors can offer important predictive value, it is recognised that they are already determined. It may therefore be prudent to examine other factors influential to decision-making, particularly considering the scarcity of research examining modifiable factors for women offered prophylactic mastectomy. This may include areas such as: fear, perceived risk, locus of control, self-efficacy and motivation to engage in preventative behaviour. The majority of studies in this review gathered data from medical records, with only three making use of participant questionnaires. However, in order to further understand psychological considerations in perceived risk and decision-making, it could be beneficial to utilise appropriate psychometric measures in research, such as: The Perceived Illness Perception Scale (Moss-Morris, 2002), the General Health Questionnaire (Goldberg & Williams, 1998) or the Perceived Personal Control questionnaire (Berkenstadt *et al.*, 1999). Such information could help to identify causal factors most amenable to psychological support and intervention.

Expanding upon the limited evidence base of predictive and modifiable factors, and applying developing theory to clinical practice, would provide a greater understanding of what is important to women when considering prophylactic mastectomy. This knowledge is integral to facilitating more effective and person-centred decision-making to ensure the greatest overall breast cancer reduction.

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Part 2: Research report

Women's experience of decision-making regarding prophylactic mastectomy

Abstract

Introduction

Approximately 20-25% of hereditary breast cancers are attributed to BRCA1 and BRCA2 gene alterations (National Cancer Institute, 2015). For women with an inherited alteration, but no diagnosis of breast cancer, preventative surgery is a treatment option offered to mitigate risk (NICE, 2013). This has been an area of largely biomedical dominance, with recent research reflecting an increase in quantitative studies examining factors predictive of uptake and satisfaction with surgery. Less attention however, has been paid to the psychological and experiential issues that may affect women's decision-making prior to mastectomy.

Method

Interpretative Phenomenological Analysis (IPA) was utilised to explore five women's experience and sense-making of their decision to opt for prophylactic mastectomy in the context of receiving results confirming they carried a BRCA gene alteration. The study also sought to explore the experiences of women in the period between opting for the preventative surgery and waiting for this to occur.

Results

Four superordinate themes were identified: 'It's a no-brainer' illuminated how women approached and understood their decision; 'good breast/bad breast' reflected women's experience of holding conflicting views towards their breasts; 'big B on my shoulder' highlighted worry held in relation to geneticised identity; and 'the preciousness of life' illustrated the impact of familial and existential experience.

Conclusion

The distinction between making the decision to have prophylactic mastectomy, and living with the decision, highlighted the importance of holistic and multi-disciplinary support throughout the process, where clinicians remain mindful to experiential, emotional and systemic motivations. By identifying and 'naming' the tension individuals could hold towards their decision and the imminent loss of their breasts, women may feel more confident in sharing their concerns, which will also assist services to gain a greater understanding of what is important to women as they debate and navigate prophylactic mastectomy.

1. Introduction

1.1 Familial breast cancer

Breast cancer is the most prevalent form of cancer in women worldwide (World Health Organisation, 2016) and, in the UK, a woman's lifetime risk of developing breast cancer is estimated at 12% (Breast Cancer Care, 2016; Cancer Research UK, 2016). Research over the past two decades has highlighted increasing evidence of the strong inheritability of breast cancer, with approximately 5-10% of all cancers being attributed to inherited gene alterations (Honrado *et al.*, 2004). Two genes most commonly associated with breast cancer are BRCA1 and BRCA2 (**BR**east **C**Ancer). These genes instruct the provision of tumour suppressing proteins that work to repair damaged cells (National Cancer Institute, 2015). However, a mutation or alteration to the gene can prevent its normal functioning, triggering cells to grow and divide uncontrollably. Research has suggested that women with an inherited BRCA1 gene alteration have a 60 to 90 *per cent* chance of developing breast cancer during their lifetime, whilst risk is estimated to be 45 to 85 *per cent* for those with the BRCA2 alteration (Royal Marsden NHS Foundation Trust, 2013). Together, BRCA1 and BRCA2 account for approximately 20-25% of all hereditary breast cancers (National Cancer Institute, 2015).

Women with an inherited BRCA gene alteration, but no diagnosis of cancer (unaffected carriers), can use genetic test results to make informed decisions about risk reduction. Of the treatment options available, total bilateral prophylactic mastectomy shows the greatest efficacy, reducing the risk of developing breast cancer by up to 90% (National Cancer Institute, 2015). Traditionally, medical attention has emphasised the treatment of disease, however, preventative approaches have burgeoned alongside recent genomic advances, allowing individuals to take a proactive approach to risk management. This raises issues about the positioning of people with BRCA gene alterations within the healthcare system: Although unaffected carriers have an increased risk of breast cancer that can be reduced through medical intervention, the women are typically healthy and free from disease. A given carrier's action may be influenced by how they position themselves on this healthy-unhealthy continuum. Social identity theory (Tajfel, 1979) states that a person's sense of self can be influenced by identification with a particular group, and that confusion can occur when groups are seen as overlapping. For example, women with an inherited BRCA

gene alteration may fall between two groups: Those with cancer and those without cancer. Reyna and Brainerd (1985) referred to this as ‘inclusion illusion’ and argued its potential impact on reasoning, probability judgement and risk assessment.

1.2 Theories of decision-making

Early models of decision-making drew upon subjective expected utility theory (Edwards, 1954) and computational accounts (Simon, 1956) that posited decisions were made following rational information processing and cost-benefit analyses. Whilst such models may have utility, they neglect emotional and iterative processes (Charles *et al.*, 1997). Recent research in health psychology has highlighted that knowledge alone does not necessarily predict behaviour change and that greater attention should be paid to understanding how individuals think about their health and behaviour (Ogden, 2012). In a review of relevant research, Zikmund-Fisher *et al.*, (2010) concluded that emotions were often more influential than knowledge when people were making decisions about cancer treatments.

Decisions that involve elements of probability and uncertainty, such as those relating to the risk of cancer from the BRCA gene alteration, are also thought to be particularly complex due to the difficulty in forecasting accurate cost-benefit analyses. Individuals are often averse to uncertainty and predominantly favour a sense of certainty (Tversky & Kahneman, 1986). This notion fits with attribution theory (Kelley, 1967) and the self-regulatory model (Leventhal *et al.*, 1992), which both posit that individuals are motivated to assure their world is as predictable and controllable as feasible. When individuals are faced with a difficulty, they interpret the problem and identify suitable coping strategies to solve the problem and re-establish a state of equilibrium (Ogden, 2012). This may involve autonomous and intrinsic motivation but also extrinsic motivation, where behaviour may be related to pleasing or protecting significant others.

Mindful of these influences, protection motivation theory sought to expand upon early computational models to include more emotional components to the understanding of health behaviour. The model posited severity, susceptibility and fear as being related to threat appraisal, and that response effectiveness and self-efficacy were related to coping appraisals (Rogers, 1983). Two sources of information were also suggested as being

influential to these appraisals: environmental factors (verbal persuasion, observational learning) and intrapersonal factors (past experience). This view is also put forward through fuzzy trace theory (Reyna, 2008) which suggests a dual process approach to decision-making. This includes verbatim representations (including words, numbers and pictures) and gist representations (interpretation and meaning to the individual). Both models serve to highlight the complexity of decision-making and suggest a combination of both rational and emotional considerations, whereby information is acknowledged and used for purposes of reasoning, but that decisions may also be shaped through affect, interpretation, self-efficacy and experience.

1.3 Rationale and aims

Whilst there has been an increase in quantitative research exploring factors predictive of uptake and satisfaction with preventative mastectomy, this has predominantly involved examination of breast reconstruction, body image and pain (Bebbington Hatcher & Fallowfield, 2003; Claes *et al.*, 2005; Schwartz *et al.*, 2012). However, in this largely biomedical domain, significantly less attention has been paid to the psychological issues that may affect women's decision-making prior to mastectomy or how women might experience the wait for surgery. In addition to these constraints, there have been relatively few studies worldwide specifically exploring these considerations in women with an inherited BRCA gene alteration but no diagnosis of cancer.

In order to understand women's experience of decision-making and navigating the process of prophylactic surgery, researchers must first understand some of the most important and influential factors involved in this process. It has been argued that how people conceptualise and make sense of genetic risk information determines how they respond to this information (Marteau & Senior, 1997). This in-depth and psychologically informed understanding is integral to the provision of more effective and empathetic support offered to women throughout this potentially life changing process.

2. Method

2.1 Design

A qualitative methodology was used to explore women's experience and sense-making of their decision to opt for a prophylactic mastectomy in the context of receiving results confirming they carried a BRCA gene alteration. The study also sought to explore the experiences of women in the period between opting for the preventative surgery and waiting for this to occur. Qualitative research is integral to the investigation of exploratory areas such as this by facilitating in-depth understanding of lived experiences, beliefs, motivations and social issues (Corbin & Strauss, 2008). Qualitative research is also less susceptible to reductionism and Al-Busaidi (2008) suggests that, when this method is incorporated into health research, it can provide rich and detailed information that may otherwise be lost through quantitative means alone.

Interpretative Phenomenological Analysis (IPA) was selected as the preferred methodology due to its psychological focus upon personal experience and how people make sense of, and apply meaning to their lived experiences (Smith, 2011). IPA is informed by three conceptual touchstones of philosophical thought: phenomenology hermeneutics; and idiography (Smith *et al.*, 2009). Firstly, the approach is phenomenological in that it seeks to understand how individuals experience themselves and the world in which they live. IPA is also hermeneutic as it involves the interpretation and 'meaning making' of experience and it is suggested that this cannot occur without existing conceptions and prejudices. This study involved a double hermeneutic where the researcher sought to make sense of the participant's own sense making. Thirdly, IPA is idiographic and committed to the in-depth understanding of how particular experiential phenomena are understood from the individual perspective, which implies focus on the particular rather than the general (Larkin & Thompson, 2011).

The epistemological position of contextual constructionism was adopted by the author for the current study (Appendix F). This supports the endeavour towards understanding the 'reality' of experiences whilst also acknowledging the potential influences which may shape an individual's interpretation of an experience. As such, a contextual constructionist epidemiology is of particular relevance to IPA, where the researcher and participant are both recognised as conscious beings interpreting and acting on the world

around them (Giorgi, 1995). This subjectivity in the production of knowledge, and the way information is perceived in the social world, can depend upon an individual's beliefs, experiences and expectations (Bunge as cited in Madill *et al.*, 2000).

2.2 Ethical considerations

The research proposal was submitted to the Health Research Authority (HRA) and local Research Ethics Committee (REC). A favourable opinion (Appendix G) was granted following minor amendments to the participant information leaflet and consent form. Approval was also granted from the local NHS Research and Development (R&D) department (Appendix H).

Details contained within the participant information leaflet advised that participants could find topics within the interview to be sensitive in nature and potentially upsetting. Participants were therefore offered the opportunity to talk more informally with the researcher before and after the interview, where more general support was offered if required. All participants were current service users of a clinical genetics service; therefore, the contact details of an agreed staff member were also provided to participants should they have wished to speak with someone in their clinical team.

2.3 Participants

A sample of five participants were recruited from a regional clinical genetics centre in the UK. The recommended sample size for a doctoral level IPA study is between three and six participants (Smith *et al.*, 2009), where importance is placed upon the quality and richness of the data, rather than the number of participants (Larkin & Thompson, 2011). A circumscribed number of participants is encouraged due to IPA's orientation towards in-depth experiential analysis and idiography. In a similar vein, representativeness is not considered important nor desirable. Instead, IPA privileges homogeneity within a sample such that experiences and meaning-making can be considered in relation to a small group of people who do not differ too much with regards to factors that could impact upon experience of the phenomenon under investigation.

Individuals were invited to participate if they met the following inclusion criteria:

- Women aged between 25 and 49 – The specific age range was chosen to support homogeneity by increasing the potential for more similar life events and experiences amongst participants.
- Confirmation of an inherited BRCA gene alteration – All participants had undergone testing within a clinical genetics centre where the presence of either a BRCA1 or BRCA2 gene alteration had been identified.
- No current diagnosis of cancer – At the time of interviewing, participants had no diagnosis of cancer (unaffected) as this would have presented heterogeneity meaning that the phenomenon under investigation would have been different.
- Elected to have prophylactic mastectomy – The rationale for this was to minimise any influence that participation could have upon an individual's independent decision-making.
- Awaiting surgery – Participants to be interviewed prior to having prophylactic mastectomy.

The final sample comprised five participants aged between 39 and 49, thus providing good homogeneity in terms of age. Four participants were married and four had given birth to at least one child. No further demographic information is provided in order to maintain participant anonymity. In addition, participants were given pseudonyms, which are used throughout the current study. The aforementioned demographic details can be found in Table 2.

Table 2. Participant demographics

	Age	Partnership status	Had children	Decision to have BRCA genetic testing
Anna	39	Married	Yes	Decision made after relative tested positive
Beth	41	Single	No	Self-initiated decision to have BRCA genetic test
Claire	45	Married	Yes	Self-initiated decision to have BRCA genetic test
Donna	49	Married	Yes	Decision made after relative tested positive
Emily	47	Married	Yes	Initiated both familial and self-testing

2.4 Procedure

2.4.1 Recruitment

A pack containing the introductory invite (Appendix I), participant information letter (Appendix J) and participant response slip (Appendix K) was given out by the clinical genetics service to those who met the inclusion criteria. Individuals were then asked to confirm their interest to participate by either contacting the researcher directly or by returning the response slip in the post. Once an expression of interest was received, the researcher made contact with the individual through their preferred method of communication in order to answer any outstanding questions and to arrange a convenient date for the interview.

Recruitment spanned April 2016 to February 2017. Through regular communication between the researcher and clinical site collaborator, it was agreed that information packs be sent to eleven prospective participants who met the inclusion criteria. From these eleven individuals, four made no further contact, one was diagnosed with breast cancer and one had their surgery brought forward.

2.4.2 Data collection

Participants were offered the choice of location for the interview: the local hospital; University; or their own home. Due to the sensitive nature of the study, participants were advised to choose a location where they felt most comfortable and where privacy could be guaranteed.

Data were gathered via semi-structured interviews that ranged from 58 minutes to 92 minutes in duration. Semi-structured interviews were chosen as the exemplary data collection method for IPA research (Osborn & Smith, 1998). A written consent form (Appendix L) was completed prior to each interview.

Interviews began by inviting participants to provide contextual information relating to their pathway to genetic testing and what stage they were at with regards their decision for surgery at the time of the interview. The researcher aimed to sensitively support participants to recount the experiences and meaning-making that they thought was pertinent to their decision to opt for prophylactic mastectomies, rather than to bias the interview to the issues the researcher thought were key. Thus, although an interview schedule (Appendix M) had been developed, this was utilised in a non-rigid manner to provide prompts and re-orientate the interview if content moved too far off topic. The researcher utilised exploratory phrases and open-ended questions with the aim of providing rich data relating to participants' experiences and meaning-making.

2.4.3 Data analysis

The transcription of audio recordings was completed by the researcher to facilitate greater familiarisation and immersion with the data. Smith *et al.*, (2009) suggest that IPA analysis can be characterised by a set of common stages (Appendix N) however advise that such guidelines should be used flexibly and iteratively. The process involved line by line coding of the descriptive, linguistic and conceptual understandings for each participant. Emergent patterns (themes) noted within the exploratory coding were then identified, highlighting areas of convergence and divergence, commonality and nuance. Themes were further refined and prioritised with consideration to the research question and by the identification and interpretation of 'objects of concern' to the participant (Larkin & Thompson, 2011). Examples of these analytical steps are included in Appendix O. This strategy was applied to individual cases before moving on to explore themes across cases.

Techniques such as abstraction and subsumption were used to identify and cluster emergent themes to develop superordinate or higher-level themes.

2.5 Quality issues

Quality issues in qualitative research are distinct from those in quantitative research where individuals conducting research within health or medical psychology can find themselves evaluated by criteria that may not be applicable to their methodology (Stern, 1997). Qualitative researchers typically formulate using observation, interpretation and theory but do not however claim generalisability to the wider population (Stiles, 1993). Instead in IPA, because a particular experience is identified for one person, it could then be suggested that this type of experience, or meaning-making, could also be possible for another person in similar circumstances.

Yardley (2000) states that good qualitative research should be rigorous, coherent and transparent so that it may be useful and understood by others. It is also suggested that the most appropriate framework for assessing the quality of qualitative methods varies with the epistemological position taken (Smith, 2011). Whilst the role of subjectivity is acknowledged, quality assessment within a contextual constructionist framework also supports that the researcher considers how their own perspective could have influenced the way they approached the research. In order to enrich theme development and reflexivity within the research, the analysis of each individual case was discussed between the researcher and research supervisor, where personal and alternative reflections and interpretations were considered. In addition, emergent and superordinate themes across cases were discussed within a peer supervision group with colleagues familiar with IPA methodology. This provided further support to ensure that interpretations remained grounded in the participant data and also for the researcher to reflect on their own position regarding the area of research. In this case, the researcher was a female who had no lived experience of genetic disease or personal experience of prophylactic surgery. The researcher did however know individuals who had both accepted and declined mastectomy and reflected that, should they have found themselves in a position similar to those interviewed, they too would have been likely to opt for prophylactic mastectomy.

Please see Appendix R for a full chronology of the research process.

3. Results

Figure 1 below illustrates the four superordinate themes and ten sub-themes generated from the participant² interviews. Further information on the frequency with which each participant contributed to the themes can be found in Appendix R.

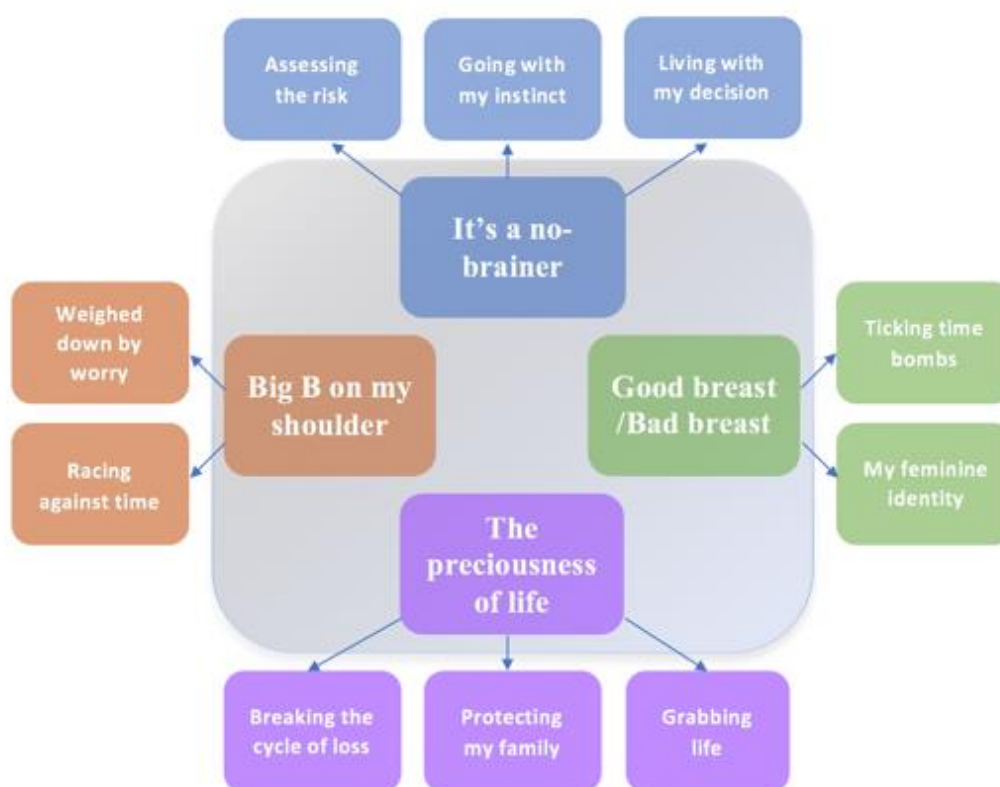


Figure 1. Superordinate themes and sub-themes

3.1 It's a no-brainer

The first superordinate theme reflects women's relationship to their decision to undergo prophylactic mastectomy and how they made sense of the decision-making process. During the interviews, three women specifically referred to this decision as a '*no-brainer*', a colloquial term used to describe a choice requiring little or no cognitive effort. For all women, the decision itself was viewed as the obvious choice when faced with the potentially fatal alternative, and women struggled to consider why one would not elect to have the surgery.

² To maintain participants' anonymity, pseudonyms have been given and these are used throughout the thesis

If you're diagnosed with breast cancer you don't have that luxury of time to think things over (...) I mean, I'm sure if you have breast cancer it's still a choice, but is it really a choice, it's gonna kill you, who wants that to happen to them (Anna)

I would rather have something preventive now rather than having to go through like chemo or whatever for any kind of cancer (...) it seemed like the only sensible option really (Donna)

Participant accounts also indicated that the women had already decided prior to receiving their genetic test results, that they would opt for surgery in the event of a positive diagnosis for the BRCA gene alteration.

I was matter of fact about it because I'd already prepared myself that it would be a positive result, um, because everybody else had tested positive for it, um, so I immediately had a plan in mind (Anna)

This further supports the idea that participants did not really perceive surgery as something they could refuse. Differences were however noted in how the women approached and made sense of this decision. As such, this superordinate theme comprises three sub-themes: The first considers how participants drew upon medical information and statistics to justify their decision and the second concerns examples of participants' emotional intuition and instinct. Whilst these sub-themes have been separated for the purpose of discussion, these were not mutually exclusive approaches and may have been privileged at different times. The third sub-theme relates to the distinction between making, and living with, the decision.

3.1.1 Assessing the risk

Respondents made repeated reference to their cancer risk as a statistical probability. Some women may have found it helpful to view their personal risk objectively, which could have accompanied use of emotional detachment as a protective strategy. As well as using probability-based statistics to justify decisions for surgery, women and their significant

others, seemed to return to these figures to provide powerful motivational reminders concerning why they were opting for intervention.

If you do it in like a calculated mathematical way my reasoning would be, or the justification would be... ok, I could have a normal risk of say is it 12% or something and then it increases up to 70-80%, to me it's like a no-brainer really (Donna)

At the end of the day the surgery, I consider it necessary because I want to take that risk down to 2% or the national average (Anna)

Deference to medical knowledge and advice, or '*bowing to the expert's opinion*' also seemed evident in some post-hoc justifications. Such an approach to medical authority may have reduced anxiety about making 'correct' choices by deferring decision-making responsibilities to medics. Donna, for example, spoke of her need to take the process one '*stage*' at a time and actively sought medical opinion before proceeding further. By following the prescribed stages, focusing on the present, and giving over responsibility to professional judgement, Donna experienced a sense of containment whereby she could '*blur the near distant future*' until advised what to do next.

I assume the people who do the surgery are the experts, that's what I have to assume, (...) that these people are experts and they know what they're doing and it will be... (...) you have to focus on the positive... and that's what I'm doing, that's all I'm thinking about (Donna)

Medical consultation and opinion was also recognised as influential to Anna's post-hoc justification of her decision. Linking with the next sub-theme, Anna described herself as understanding that she wouldn't get breast cancer, and perhaps embraced this intuition to justify her decision not to seek surgery during an earlier life-stage when her genetic status had been confirmed. Anna however experienced contradiction when provided with statistical information and medical advice contrary to her belief and began to consider that she did not want to take unnecessary risk by ignoring it.

Because it was there written in black and white that really made me think, well you know, the doctors told me, they know more than me, I probably don't feel like I'm gonna get sick (...) if there is this chance then I should take steps, take precautions (Anna)

3.1.2 Going with my instinct

The second sub-theme reflects some of the more emotionally intuitive and spiritual influences to decision-making. As the previous quote indicated, Anna had a sense that she 'knew' she would be very unlikely to get cancer. In contrast, other women reported an intuition that they would definitely develop cancer.

I didn't do a whole list of pros and cons (...) I just knew, I just had this intuition, I just knew... that I am gonna get it... (sighs), I just know, I just know and ... that's not based on, see you've got the science (...) and then there's this sort of intuition, this spiritual bit, and I'm like... I know I'm gonna get this (Beth)

This intuition was often linked to the appraisal of high risk and a strong fear of developing breast cancer. It has also been considered that 'instinctive' beliefs may have emerged from personal or family narratives regarding perceptions of illness and risk-reducing behaviours. For Beth and Claire, familial experience may have shaped a perceived sense of inevitability and foreboding which contributed to the demonization of breast cancer. Beth viewed breast cancer as a 'curse' within the family and personified the disease as having a personal vendetta. As such, prophylactic surgery was viewed as the only option to alleviate the anxiety experienced by carrying the BRCA gene alteration.

I check myself, um... never found anything... but I just think... no, it's there, it's just waiting, that's what it feels like (...) it's there, it's just lying dormant (...) so, yeah. I just knew... I had to do it (Beth)

I believe that if I don't have this surgery I will get breast cancer... and that's not trying to be dramatic... I feel that it's there somewhere in the future... I actually think that for me my instinct tells me that my path is breast cancer (Emily)

In addition to personal intuition and emotional experience, spiritual influence also appeared important to some women in the decision-making process. This was particularly recognised in Beth and Claire, who had both lost loved ones to breast cancer.

They're with me... and it was my mum who pushed me to do this, I know it was... I strongly believe that (Claire)

I went to see a psychic last year and (...) the first thing she says is oh I've got your mum here and your mum says you've got four years (long pause) that's what she said (upset)... she just said, you've got four years, you know what you need to do and I was just like oh my god... (sighs).... so I knew then, that's when I made my decision (Beth)

3.1.3 Living with the decision

Whilst all women had already elected to undergo prophylactic mastectomy, interviews also revealed a distinction between making the decision, and living with, or accepting the decision and its associated repercussions. Many women had arrived at their decision through collaborative discussion with their families, potentially leaving some feeling uncomfortable or reluctant to share concerns revealed whilst awaiting surgery. Potential feelings of doubt and uncertainty may have also been difficult for women to accept given the decision seemed to be a 'no-brainer' when available medical information focussed solely on statistics.

Pragmatically... I knew what I was gonna do when I got the result, it's completely different when suddenly someone tells you you've got it because suddenly you think... oh... actually now I've gotta action what I said I was gonna do all along (Claire)

There was an interesting split between those who viewed themselves as feeling ‘empowered’ and in control of their decision, through having knowledge and choice, and those who recognised or experienced a sense of helplessness regarding their genetic status or being pulled along by the process.

I have actually found relying on everybody else, you know relying on doctors, relying on tests, relying on... the hospitals and the waiting lists... I've found that quite difficult (Emily)

This may have been related to women’s sense of agency in relation to their decision; supporting previous subthemes indicating that some participants didn’t necessarily experience their course of action as choice, but as a path they had to pursue. Whilst women perceived the decision as a ‘no-brainer’, there was considerable anxiety around navigating the various decisions involved in progressing towards surgery and its anticipated outcomes.

[My husband] thought because we'd decided what we were going to do it was it was all okay and I was like just because the decisions been made it doesn't make it any easier to live with (Claire)

3.2 Good breast/bad breast

Women reported heightened vigilance regarding their breasts after making the decision to undergo prophylactic mastectomy. Some experienced anxiety about losing their breasts and struggled to make sense of this in the context of not having previously regarded them as important to their identity. At the same time, women reported that they wanted to ‘get rid’ of their breasts as quickly as possible because they recognised the risk associated with them. This second superordinate theme reflects the emotional conflict women experienced by simultaneously holding positive and negative attitudes towards their breasts and the strategies used to cope with this tension whilst waiting for surgery. The theme title alludes to object relations theory, although psychodynamic interpretation was not specifically applied.

3.2.1 Ticking time bombs

The first sub-theme reflects the shift that women experienced in their relationship with their breasts and encompasses the view of breasts as a threat and how this fuelled a sense of urgency for surgery. Whilst some women appeared to locate their sense of threat within their genes (Anna, Claire), all women spoke of having an altered perception of their breasts.

Whereas my breasts, I was just like... well I do love them, I really like mine (...) but I don't love them enough to... I've got a different association with breasts, ya know, as wonderful as they are.. and they feed babies, and look good, da de daa da... but there's always that underlying connotation of, yeah but they can kill you (Beth)

Here, Beth described that whilst she valued her breasts as both enhancing appearance and nurturing, she had come to experience her breasts as toxic and as the source of cancer threat. Beth stated, '*they can kill you*' reflecting negative personification of her own breasts. Claire too framed her breasts as external, protectively acknowledging her detachment from their threat and imminent removal.

I didn't want to look at them, and I don't know whether it was a detachment, whether it's like, ok well I'm not gonna see... have them in a couple of month's time so just don't look at them... I'm a bit better now but for the first two weeks I just couldn't, I couldn't look at them (Claire)

Due to the loss of her mother to breast cancer, Claire had always considered herself to be very '*breast aware*' and checked regularly for any visual or tactile change. Perhaps this vigilance was linked to an existing awareness for Claire whereby she already recognised the potential threat she believed her breasts might pose. Following confirmation of her genetic status however, Claire avoided looking at, or touching, her breasts, which was experienced as challenging, particularly considering their external position on the body.

Differing and fluid attitudes towards breasts were also apparent in their variable description through an interview. For example, Donna referred to '*boobs*' when discussing cosmetics, '*breasts*' when framing medical/surgical interactions and

'them/those' and 'organs' suggestive of emotional detachment and threat. These descriptions capture an example of the fluidity from more frivolous or informal terms, to technical communication and externalisation.

The externalisation and objectification of breasts in terms of their practical utility may have also been a useful strategy to protect against the anxiety of losing them. This may have made it harder for women to continue to experience their breasts as part of themselves or as integral sources of esteem or sexual pleasure. Perhaps related to an artefact of homogeneity, Claire, Donna and Emily appeared to view their breasts as 'objects' that had served their practical function, in terms of having a partner and having already raised their children. It was therefore perhaps easier for these women to objectify their breasts as being surplus to requirement.

*I was just like oh just get them off, I've breastfed my babies, get them off
(Claire)*

It is suggested that this consideration may have also contributed to why Anna decided to delay surgery when she was younger, if she believed that her breasts had not yet served their purpose in relation to her stage of life. Whereas Donna described having reached a stage in her life where she felt comfortable in her marriage and had completed her family. Therefore, when privileging the roles of wife and mother, Donna viewed her breasts as surplus to requirement, which may have supported an emotional distancing from any underlying concerns of how it felt to lose her breasts as a woman.

*I suppose as a... wife... you think oh God am I going to be... it's not even am
I going to be attractive is it, am I going to miss that kind of thing, that sexual
thing (...) but they're not something I think I'm proud of or whatever, they...
they now are almost like a ... a wound or something that can injure me... so
they, they're not doing me any good (Donna)*

3.2.2 My feminine identity

Despite the acknowledgment of objective risk and interpretation of the breasts as potentially dangerous, the second sub-theme reflects the tension and emotional impact of mastectomy to identity. During the interviews, this tension was mentioned almost

apologetically and described as 'silly'; perhaps suggesting women believed they ought not feel that way when faced with a pragmatic medical decision. As such, some found it difficult to understand and verbalise their emotional apprehension towards impending surgery.

I still cannot totally explain why I'm so bothered about the surgery... it's almost something deep deep within that you can't tap in to... I... I don't know whether it's body image, I don't know whether it's... like say knowing that you've had it done because you're quite scarred, erm I don't know whether it's to do with kind of like losing your femininity, I don't know what it is but there's something about this surgery which is deeply personal at the deepest level... and yet I can't explain what it is (Emily)

It might have felt difficult for Emily to make this connection if, as a woman who self-identified as strong and sensible, she favoured rejection of societal objectification and sexualisation of women's breasts. Emily might have believed she shouldn't feel concerned with appearance and, when faced with the impending loss of her breasts, she found it difficult to verbalise her concerns because she also needed to be appreciative of her fortunate position at being offered 'life-saving' surgery. For some, worrying about appearance was akin to vanity and was experienced as a confusing contradiction for those who identified themselves as sensible and pragmatic women who hadn't thought of themselves as being concerned about appearance.

I was quite surprised at myself because I've always... respected myself almost to be really pragmatic (...) and then suddenly I was behaving like this and I thought where the hell has that come from because I've never imagined myself to think like that so it threw me (Claire)

Three women also spoke of their unease and frustration that others could misconstrue their motivation for surgery as being 'vain', cosmetic or like a 'boob job' and how surgery would never have been considered without presence of the BRCA gene alteration. Until recently, none of the women had needed to consider life without their breasts, but since making the decision, breasts and their imminent loss had become extremely prominent.

I think for something that's so invasive and so... um, ya know... really gets to the core of who you are as a woman, aesthetically (Beth)

Perhaps women were concerned that, without their breasts, they would feel different as women. Beth in particular, struggled with her sense that she was about to 'mutilate' her breasts having worked to get to a point where she was comfortable with her body. Beth was aware of the objectification of breasts and felt particularly concerned with how others would perceive her post-surgery body but also felt strongly that women should not lose the right to take pride in their body following mastectomy.

I don't want to be with somebody before surgery and see my body before surgery and then remember it after surgery, I ... I need to meet someone as a new me... bizarrely, even though I'm gonna be the same but I'm not gonna be the same and that's quite important (Beth)

3.3 Big B on my shoulder

This superordinate theme reflects some of the ways in which women experienced their genetic status and its impact on their perceived control, anxiety and wellbeing. The theme title incorporates a quote by Claire in which 'Big B' (referring to the BRCA gene alteration) is a play on 'Big C', a colloquial term for cancer. This appeared to describe the experience of looming worry but also captured how women experienced a sudden awareness of themselves and their families as geneticised beings which, for some, provoked independent request of further assessments to explore other facets of their genome.

There's a 50% chance of passing it on you know, 50% chance, it's not the same as passing on your hair colour, your skin tone your eye colour (...) it's a big thing to pass on and it's not a good thing (Anna)

Genetics are not reversible, they're not and you can keep the wolf from the door through what I've done... plant based diet, exercise and no drinking... for so long, but it's gonna come through, it will... I'm sorry but your genetics do eventually prevail don't they, it's just the way it is (Claire)

This superordinate theme comprises two sub-themes; the first reflecting some of the worries women experienced in relation to BRCA and how they attempted to limit and manage this worry and the second relating to anxiety experienced through waiting for surgery.

3.3.1 Weighed down by worry

Whilst all women spoke of feeling grateful and fortunate to be in the position of knowing their genetic status, for some, this knowledge could be experienced as overwhelming and consuming at times. The genetic legacy of BRCA was also perceived by women as potentiating a ripple effect: worry was seldom isolated to the individual but had potential to negatively impact upon loved ones.

It's a big responsibility for me to talk to people [relatives] ... you know, just because this is what I'm choosing to do it doesn't mean that's what they've got to have done (...) like I said before, do what you want with me, just leave everyone I love and care about out of it (Claire)

Women generally reported increased worry since learning of their genetic status. This was often a magnification of existing anxieties, and whilst gaining risk knowledge was often viewed as a pragmatic preventative step, for those women who had witnessed a loved one affected by cancer, they had already carried fear of cancer for many years.

I'm dragging round a sack of... rocks (...) that I'm pushing through everyday... and it's a really big worry... a big... rock... you know, a really big heavy worry on my back in a big rucksack at the moment and it's weighing me down... you know and it fills my chest... and at times up to the point of which I couldn't fit anymore in (Emily)

Furthermore, whilst having prophylactic surgery was primarily viewed as a means to reduce risk, this was also interwoven with an anticipated reduction in anxiety post-surgery.

I hope I feel like I did before I had the news... because erm... the worry is constantly there (...) it's like having a radio on low in the background, it's always there... and I need to switch it off (Emily)

Given the pervasive dread attributed to the ever-present threat of the BRCA gene alteration, and attendant sense of anxiety and helplessness, some women sought both formal and informal support to assist them to feel more 'in control' (Anna), 'informed' (Beth) and 'supported' (Emily). Beth, for example, spoke of how she had found it helpful to make contact with other women who had also chosen prophylactic mastectomy. Alongside practical advice from genetic counsellors and surgeons, Beth, Claire and Emily appeared to value information and emotional advice from 'real people'. In this instance, women who had already had preventative surgery were viewed as having expertise.

It feels like you get insider information (...) because a lot of the information is probably not something you could put on an information sheet (...) so I feel a lot more clued up about things (...) but ya know who's gonna tell you that (...) the women have already been through that (Beth)

Having informal support from other women was also viewed as an opportunity to share concerns as well as questions. Emily in particular, recognised the emotional support this could offer, and this may have been linked to her experience of not always feeling able to share her concerns with her family or the medical team.

It would've been helpful for me to have talked to someone who'd got children as that was my biggest guilt and remains my biggest guilt... you know it would have been nice to actually ... to get somebody else's perspective I think that's what I've missed (...) cause I've... I have felt quite alone (Emily)

3.3.2 Racing against the clock

With the exception of Anna, all women described the experience of waiting for surgery as feeling particularly difficult. When this was raised during interviews, it was often expressed as a race against time whereby watchful waiting was inadequate and action to beat BRCA was needed.

I could have a scan or a mammogram next week and be fine but if something grows there, a fast growing something and if I'm checked again in a year's time it might be too late to do anything (Donna)

Women have been in... erm... for preventative surgery... but when it's come to it they have actually developed cancer you know there is this almost feeling that the clock's ticking... and you've got to you've got to beat the clock (Emily)

Whilst the 'good breast/bad breast' theme highlighted how some women located the source of cancer threat to their breasts ('ticking time bombs'), some women also appeared to locate this within their genes. Although all women had elected to have prophylactic mastectomy, it has been considered whether perceived attribution of risk could have also influenced women's sense of self-efficacy regarding their preventative action.

I've always been quite pragmatic about it, but since knowing that I've got BRCA1 I'm like oh God just get them off (...) get it done because now I feel like I've got is ticking time bomb inside me, it's like any minute now it could, it could all kick-off (Claire)

Alongside fear and hypervigilance to carcinogenic symptoms, some women experienced both intrinsic and extrinsic anxieties and motivations to have surgery within a particular timeframe.

My mum was diagnosed with breast cancer when she was 39 (...) and people will tell you who've lost their mums when they've been very young that you hit milestones, and the milestone for me was when she was first diagnosed and then... she died when she was forty four, which... is only three years away (Beth)

It is possible that some women may have also wished to have surgery as soon as possible to reduce the potential opportunity of being able to go back on their decision. Perhaps this may have been related to why Anna delayed her decision to have preventative surgery, if she was still initially experiencing an underlying belief that surgery was unnecessary.

3.4 The preciousness of life

The final superordinate theme summarises some of the existential considerations women experienced when contemplating prophylactic mastectomy. Participants described an acute awareness of life as time limited and precarious, strongly contributing to women's desire to seek surgery. These considerations also appeared to link back to past experiences of loss, the prioritisation of significant relationships and women's hopes for the future after surgery. The three sub-themes below map onto these domains.

3.4.1 Breaking the cycle of loss

Four women recounted historical experiences of witnessing a loved one suffering or dying from cancer. Narratives were emotionally charged and reflected a powerful incentive for women to have preventative surgery, particularly for those who had lost their mother to breast cancer.

It might've been different if I hadn't seen someone suffer with breast cancer and that that was my mum, and there is something about that mother daughter link and that bond, and that legacy that she's left me (Beth)

Cancer steals from people, its steals valuable years, precious years, you know when you're young you just take for granted that you're gonna have your mum there and then suddenly (...) she's not, it's cruel (Claire)

Some women still felt raw, bereaved and angry towards cancer for causing pain within their family. This experience and awareness of the impact of loss instilled in women a determination that they would not impose the same loss on their children. In fact, this was described as one of the strongest motivating forces behind women's decision to have prophylactic mastectomy.

I'm just happy that I've got this opportunity, I'm grateful because I've stopped that cycle, my grandmother never saw my sister being born, my mother never saw... myself get married and have my babies and so I'm damned if I'm not going to be here to see my babies for what I can control (Claire)

This was also evident in Beth's interview when she was thinking about the impact her potential death might have on children she considered having in the future:

I think the impact of [my mother's] death on me and the choices that I've, I was making and I kind of realised that actually not wanting to have a baby on my own wasn't just about not wanting to have a baby on my own... I didn't want to have a baby and leave her...and... leave her.... oh god, um... (upset).... and not die (Beth)

For those who had experienced familial cancer, there was also a sense that they wished to respect and honour the memory of loved ones by taking preventative opportunities that were not available to them. Due to the heritability of the BRCA gene alteration, this may have involved a sense of survivor guilt facilitating an emotional obligation to pursue preventative surgery.

I've been given the chance to stay alive... and my mum never had that, my aunt never had that, my cousins never had it (Claire)

I sat there and I just thought you know how blessed I am really to have this warning (...) to have this incredible chance to do something about it (...) I know my mum has said to me (...) she would've definitely had all the surgery and she wouldn't have been the situation where (...) she was told she had got cancer (Emily)

For Anna, the loss of her mother also contributed towards a significant turning point in her life where she began to consider surgery a greater priority. As previously mentioned, Anna had perceived her own risk of developing breast cancer as low and, as such, had planned to follow her own life trajectory and wait for the 'right time' to have surgery. However, when her mother suddenly died, her first experience of close family loss, Anna seemed to question her own mortality and her perceptions of actuarial risk and medical opinion appeared to change: she decided to 'prepare for the worst' by taking steps towards surgery just in case cancer was a stronger possibility than she had previously thought.

After my mum died (...) that kind of hit me at that point that there is this risk and, and it is a reality and I need to do something about it, (...) I don't think I really considered that before that point that I had an 80% risk, more or less, of getting breast cancer, but because, I hadn't really experienced it in my family, I think that that's probably made a difference (Anna)

3.4.2 Protecting my family

Following on from the previous sub-theme, 'protecting my family' reflects how all women described the need to protect their family from loss and to manage the process as effectively as possible, which was often prioritised over their own wellbeing. Within each interview, women described how they sought to protect loved ones, particularly children, in order to avoid upsetting or overwhelming them with health concerns. As such, some women experienced a responsibility to 'stay strong' and to recover as quickly as possible to avoid placing further strain on the family. For Claire, she viewed her wellbeing as linked to her family's and this emphasised how important Claire's family were to her in influencing the decisions she made about surgery.

I'd rather do it in one go... it's less impact on my family (...) to do that potentially three times over the next six months would be really harrowing I think, for me, watching everyone struggling around me (Claire)

In addition, some women spoke of the need to maintain a sense of normality and equilibrium within the family home, not only to protect others, but also to assist them to feel more calm, confident and prepared for surgery. This involved an element of women trying to present themselves, or perhaps perform, that everything was ok.

There was always this feeling that this was news that was affecting everybody and I almost had to suck it all back in... to protect everybody else, so everybody else was ok (Emily)

A powerful part of this performance was also related to how women experienced a sense of responsibility to be a positive role model for their children, particularly daughters, should they be faced with similar decisions in the future. Claire reflected that she had

developed this from her own mother, whom she admired as being ‘*incredible*’ throughout her breast cancer diagnosis and treatment.

I also feel that it is my responsibility to go through this... well... in front of them, to cope with it well to, show them that it is ok, because if they then choose to do anything about it in the future they can say well my mum did it and she was fine... she was okay, she did it, you know (Claire)

It's about reducing this worry... but it's about doing it in a way that doesn't worry everybody else, you know, I want to be a good role model for my daughter in case she ever needs to have the same thing... I want her to know that... you know you can get through it (Emily)

3.4.3 Grabbing life

The third sub-theme reflects an anticipation of life after surgery. This was something that women held in mind throughout the process and was described by Donna as ‘*thinking of the bigger picture*’. For some women, the possibility and probability of having a future, was viewed as significantly increased through their preventative action. This anticipation often included a sense of positivity and hope at the prospect of being able to put aside longstanding worry and ‘*closing the door*’ on breast cancer. The theme of empowerment was raised by most women during interviews, with this predominantly being described as a sense of feeling empowered through having knowledge and choice regarding preventative options.

It feels like a bit of a taboo but actually it's not, there's so much to celebrate about it, it's taking control, it's empowering and its healthy women taking control (...) I haven't got cancer, I'm healthy, I'm gonna have surgery (Beth)

I like to see it as the best of both worlds because I've had the time to think about it a lot and er, think about all the different options (...) I consider it a luxury, to be able to really think about the right thing for me and my family (Anna)

Although the decision-making journey was acknowledged as ‘scary’ and ‘intense’ at times, overall, women appeared confident in having made their decision and the opportunity for post-traumatic growth was evident. Emily however also acknowledged her concerns for the potential of ongoing anxiety after surgery, not just for herself, but also for her family.

It may be solved for me but it's not solved for my children you know, it will rumble on and on... but if I'm in a better place after my surgery then I'm in a better place to help them (Emily)

It's a visible reminder of what you've had done, I think you're walking around with a visible reminder of the fact you have this gene (...) I will know I've had breast surgery because I had an 85% chance of getting breast cancer and although I'll be forever grateful (...) it's almost like being branded I think... you know, it's always there (Emily)

When experiencing such thoughts, many women appeared to defend against this and found reassurance through positive self-affirmations and by reflecting on the distinction between those who had breast cancer and those who carried the BRCA gene alteration. Beth for example, experienced herself as being somewhere within the middle; not having cancer but also acknowledging she was at a higher risk than the general population. All women however appeared to view this distinction favourably and felt fortunate to be in a position where they were able to take preventative action.

As horrible as it is, it's actually ok and you're much luckier than a lot of people who never get that information, never get that choice to make that decision (Claire)

Emily described how she was looking forward to having a ‘lightness of spirit’ following surgery and letting go of the ‘heavy rucksack of worry’ she had been carrying for so long. This newfound appreciation of life also involved an admiration of those who had fought cancer, whilst also holding onto the hope of living a life less tainted by carcinogenic worry following surgery.

It makes it very real and it had never been that real before to me... erm and it makes life feel more fragile and more precious... more to be... grabbed with both hands and you know to make sure that you don't waste it, to make sure you're doing things that you love, that you're with people you enjoy... erm... I've always enjoyed life but I've very much taken it for granted cause I've had no reason not to (Emily)

4. Discussion

4.1 Summary of research findings

The aim of the current research was to illuminate women's experience of decision-making regarding prophylactic mastectomy. This included the exploration of meaning-making for women with an inherited BRCA gene alteration and how they navigated the period since opting to have preventative surgery, and waiting for the surgery. Utilising an interpretative-phenomenological approach, four superordinate themes were generated: 'it's a no-brainer'; 'good breast/bad breast'; 'Big B on my shoulder'; and 'the preciousness of life'. Findings will be discussed below with reference to extant theory and research to provide a lens with which to advance clinical understanding of lived experience (Smith *et al.*, 2009). The section will conclude by considering study limitations and clinical implications.

4.1.1 Making sense of the decision

For all women, the decision to have prophylactic mastectomy was viewed as an obvious choice when faced with the potentially fatal alternative. What did however emerge from the participant interviews was a distinction between making the decision and living with the decision, with the latter being described as more challenging and emotionally complex. Four women in the current study explained they had made the decision to have surgery either before receiving their genetic test results, or, as an automatic reaction within the clinic room. For these women, the process of cost-benefit analysis, implicit in early models of decision-making, were less apparent. Similar findings were highlighted by Dijk *et al.*, (2008) where women were found to have already reached their decision

regarding risk management preferences very early in the process. It has however also been considered that women may still have experienced the gradual integration of information over time, particularly those who had historically considered their carcinogenic risk prior to genetic testing. It is therefore difficult to disentangle precisely when the decision-making process may have consciously or unconsciously begun.

As put forward by protection motivation theory (Rogers, 1983), sense-making and justificational accounts of decisions, were found to incorporate many different factors. Whilst some women in the current study appeared to display preferences, or perhaps defences, in their approach to decision-making, for all women, a broad range of actuarial, experiential and emotional considerations were drawn upon. This supports fuzzy trace theory (Reyna, 2008) which posits the importance of personal interpretation and emotional experience (gist) combined with verbatim information and medical recommendations. Alongside models of decision-making, the researcher also noted resonances with dialectical behaviour therapy (DBT), which proposes different mind states and processing systems. The ‘rational mind’ is associated with logical thinking based on empirical facts and the ‘emotional mind’ reflects intense and sometimes impulsive affect (McKay *et al.*, 2007). The third state, the ‘wise mind’ is described as the integration of both the rational and the emotional mind, with the addition of intuition. This description felt particularly relevant to how women in the study approached, and made sense of, their decision-making regarding prophylactic mastectomy, where aspects of the ‘wise mind’ were recognised in all women through the acknowledgement of logical, emotional and intuitive thinking at different times throughout the process.

4.1.2 Altered perceptions of identity

Perhaps related to the complexity and fluidity of sense-making, women in the current study also experienced a shift in their relationship to their bodies and, in particular, their breasts. Despite the acknowledgement of objective risk and interpretation of breasts as potentially dangerous, there was a recognised tension and increased awareness of the breasts to identity. As one of the most visibly defining features associated with the conception of womanhood, the loss of one’s breasts may challenge a woman’s feminine identity (Kasper, 1995). Whilst this may be dependent upon a women’s independent sense of self, such perceptions can also be influenced by the societal objectification of breasts

in relation to sexuality, femininity and eroticism (Yalom, 1997; Young, 1980). As such, women might view their breasts as objectified and trivialised by others which could prevent them from realising their breasts as something personally important to them. Women might also experience pressure to conform to accepted norms of beauty and could experience low self-esteem when this is felt as jeopardised. Supporting this, Sischo and Martin (2015), found that women contemplating mastectomy experienced decreased self-esteem and a heightened awareness of their breasts, prompting them to consider breasts differently. Women in the current study, described a similar experience, with Anna and Beth appearing concerned about how their bodies might be negatively judged after surgery. For some, this was experienced as uncomfortable because to be concerned with appearance was akin to vanity and the potential for negative misconceptions by others. During the interviews, some women also found it difficult to comprehend, and to verbalise, their apprehension towards the impending loss of their breasts. This appeared to link with efforts to remain pragmatic and emotionally distant from their breasts by considering their functionality and served purpose. By splitting themselves as separate from their breasts, this objective appraisal served as a protective defence against the anxiety of losing them.

An additional aspect of altered identity raised by participants was that all women experienced an increased awareness of themselves, and their family, as geneticised beings. In a study by Klitzman (2009), individuals were interviewed regarding their self-concept since receiving confirmation of an inherited gene alteration. Findings suggested that many experienced an internal conflict of whether genetic risk was viewed as negative or neutral and some had also begun to question how, and to what extent, they were now the product of their genes.

In the current study, women made frequent distinctions between those with an inherited BRCA gene alteration and those diagnosed with breast cancer. As suggested by Reyna and Brainerd (1995), an 'inclusion illusion' is thought to occur when individuals view themselves as falling between two groups: in this case, those with cancer and those without. Senior *et al.*, (2002) found that an individual's perception of risk could be minimised to some extent by downward social comparison of those perceived to be at greater risk. Although all who participated in this study viewed this distinction favourably and spoke of feeling fortunate to have not been diagnosed with breast cancer, it highlights the importance of how individuals might perceive, and respond to, such differences.

Whilst it could be reassuring and grounding for women to make such distinctions, it is also possible that these could contribute to personal and societal narratives that women should predominantly see themselves as fortunate and grateful to be offered prophylactic surgery and may not feel able to share any fears or doubts because they also recognise that they are thankful to be alive. This could also be related to dichotomous narratives where women feel they must be either the warrior or the victim.

4.1.3 Thinking of the wider influences

Due to the familial nature of inherited gene alterations, all those who took part in the study had witnessed a loved one affected by cancer, with three women having lost their mothers. This appeared to have instilled in women an acute awareness of the temporality and fragility of life which was found to be an emotive and powerful incentive for preventative surgery. The impact of previous experience on decision-making has been more widely recognised in protection motivation theory (Rogers, 1983) and fuzzy trace theory (Reyna, 2008), however previous research has also highlighted the influence of familial experience of illness to medical decision-making (Friebel *et al.*, 2007; Julian-Reynier *et al.*, 2010; Metcalfe *et al.*, 2008). As put forward by Ogden (2012), when individuals are faced with a difficulty, they often interpret the problem and identify suitable coping strategies to solve the problem and re-establish a state of equilibrium.

In the current study, family, particularly children and partners, were at the forefront of decision-making and women often felt the need to reduce their risk and, subordinate the self, in order to avoid imposing worry or loss on loved ones. Previous research by Hesse-Biber and An (2016), found that those with a daughter experienced a higher sense of responsibility to be a positive role model to support their children in the eventuality that they made be faced with a similar decision in the future. Such systemic considerations again contrast earlier models, which argued decision-making to be a predominantly individual process. More recently however, Luketina *et al.*, (2012) conducted a review of the literature and found that family, and the opportunity for social support, played a significant role in the treatment decision-making of women diagnosed with ovarian cancer.

All women spoke of their anticipation of life after surgery, particularly in relation to putting aside longstanding worry, and were eager for their lives to regain a sense of

normality. Descriptions appeared reflective of post-traumatic growth which theorises that positive change can occur through the experience of trauma and adversity (O’Leary & Ickovics, 1995). Women with an inherited BRCA gene alteration may have faced multiple traumatic experiences, including receiving confirmation of their genetic status, debating surgery and the loss of their breasts. Janoff-Bulman (1992) discussed how the experience of trauma can have a shattering effect on people’s assumptive world, where previously held schemas of the self and the world, are challenged.

Previous authors (Caver, 1998; Tedeschi & Calhoun, 1995) have suggested key areas of potential growth following trauma which included: perceived changes in self; closer family relationships; and changed perspective on life. Furthermore, in a review of the qualitative literature on post-traumatic growth following life threatening illness, Hefferson *et al.*, (2009) suggested the additional areas of: re-appraisal of life purpose and priorities, reflection on mortality and existential re-evaluation. Yalom (1980) suggests that existential conflict can arise when there is a tension between one’s awareness of the inevitability of death and one’s wish to continue to be. Whilst death is recognised as unavoidable and one of the fundamental inevitabilities in life, sudden confrontation or ‘turning towards’ this, can lead to feelings of dread, existential anxiety and uncertainty. An awareness of mortality however can also ground a person in the appreciation of their life. In a qualitative study by William and Jeanetta (2015), survivors of breast cancer described a realisation of how ‘precious life is’ and not to take each day for granted. This also appeared to support women’s experience in this study and was thought to be particularly powerful in those who had experienced loss and subsequently questioned their own sense of mortality and life since learning of their genetic status.

4.2 Limitations and recommendations for future research

The current research attended to the gap in the literature by exploring women’s experience, and sense-making, of the decision to have prophylactic mastectomy. As per IPA methodology, this was undertaken with a relatively homogenous sample to facilitate insight into a particular experience of a specific group (Smith *et al.*, 2009). This means that findings are unlikely to be generalisable to the wider population as IPA methodology is purposefully idiographic rather than nomothetic. However, because a particular experience is identified for one person, it could then be suggested that this type of

experience, or meaning-making, could also be possible for another person in similar circumstances.

Participants were intentionally sampled to ensure women had already reached their decision regarding prophylactic mastectomy, but prior to surgery itself. It has been considered however, that purposive and homogenous sampling is likely to have orientated findings towards those who may have similar views towards prophylactic mastectomy. For example, all women had sought genetic testing, highlighting an existing awareness of genetic risk and self-initiated steps to ascertain this. Therefore, it is possible that individuals may have been more inclined to act upon their test results, and thus be more open to preventative action. It may therefore be instrumental for future research to also explore the decision-making experience of women who decline prophylactic mastectomy. Whilst it is imperative to gain a greater understanding of factors influencing women as they debate prophylactic mastectomy, there is also a need to understand why many women still decline this option despite its established efficacy.

The current study had a relatively inclusive sampling frame with regard to age, however, the age range of those who agreed to participate, was between 39-49 years, thus creating a more homogeneous group than initially anticipated. This was found to be a particular strength in terms of methodology, and facilitated increased opportunity for women to have experienced similar life events associated with their stage of life. Previous research (Beattie *et al.*, 2009; Garcia *et al.*, 2014; Skytte *et al.*, 2010) suggests that many women choose prophylactic surgery at a younger age, where differing life stages may contribute towards women having a different relationship towards their breasts. Therefore, it may be prudent for future research to also explore the experiences of those who received confirmation of genetic status at a younger age. Utilising findings from this study, it may be hypothesised that younger women might feel more acutely aware of reproductive and marital considerations in their decision-making. Linked to this, and given the paucity of existing research, it may also be helpful for greater emphasis to be placed on systemic influences to decision-making. By encouraging women to specifically consider the wider systems around them, researchers may be able to develop more contemporary and novel theories of decision-making to inform practice. For example, by utilising approaches such as Bronfenbrenner's (1979) ecological systems, there may be increased potential to

explore individual considerations such as gender and age, but also to highlight the importance of family, friends, healthcare systems, culture and society in decision-making.

With regard to potential bias, it has been considered that some women may have felt inclined to frame their experience in a more positive light due to the researcher's role within the NHS. Whilst it was made clear in the participant information leaflet that participation, or non-participation, would have no bearing upon normal care, it has been considered that participants may have still felt inclined to provide a more measured response. During the interviews, women also spoke of valuing the opportunity to discuss their experience with other women with an inherited gene alteration, and whilst the researcher made every effort to assist participants to feel as comfortable as possible, limited relatability may have constrained discussions. Considering this, an avenue for potential future research may be to have greater service user involvement in similar research, and perhaps, even the interview process. It is possible that this may facilitate a more open dialogue and create an invaluable opportunity for women to make supportive connections.

4.3 Clinical implications

A prominent view within the area of genomics is that individuals should be supported to make their own informed choices, with health professionals being non-directive in this (Harper, 1988). This is laudable given the distinct nature of an area where otherwise healthy individuals may be alerted to risk and subsequently offered preventative interventions. The current study however highlighted that, for some women, a more directive approach might be valued as a means of avoiding making anxiety-provoking decisions (effectively 'choosing not to choose'). A person-centred approach should therefore be utilised to acknowledge and discuss women's preferences regarding decision-making. This could be supported by clinical staff and perhaps through the early use of appropriate psychometric measures to gain an initial understanding of how an individual might be conceptualising the experience.

Based upon findings from the current study, some women placed great value on being able to speak with other women with lived experience of the BRCA gene alteration. This was found to be particularly helpful when women were at a similar stage of life in terms of age, parity and marital status. This provided the opportunity for women to be able to

ask more informal questions, providing the opportunity for normalising reactions that can't be spoken about or that they had perhaps been unable to raise through their medical appointments. Women offering peer support to other women appeared to be particularly empowering and provided the additional benefit of individuals feeling they could be of use to others by sharing their knowledge and experience. In fact, this was a strong motivator for the women who took part in the current study. Services may therefore be able to act as a conduit to connect women through signposting to support groups and online forums where they may also benefit from more informal support throughout the process.

The importance of a holistic and multi-disciplinary approach to supporting women with an inherited BRCA gene alteration is evident, as is effective communication between disciplines involved in an individual's care so that any preferences or concerns might be shared. This could also facilitate clinicians to view the woman as a 'whole' and not as respective parts associated with their particular discipline. For example, it may be helpful for medical and surgical staff to remain mindful to the potential for women to still be experiencing doubt or existential shock regarding their decision, despite perhaps presenting with confidence and certainty in clinic. By identifying and 'naming' the tension individuals may hold towards their decision and the imminent loss of their breasts, this could support women to feel more confident in raising their concerns and any outstanding questions they may have.

It may also be helpful for clinical psychologists to further support women who might be experiencing difficulty in making sense of their concerns and to model a safe therapeutic space where they feel able to explore conceptions of decision-making and to perhaps consider the wider familial and societal discourses involved. For example, dominant societal practices that sexually objectify women's breasts might make it hard for women to voice concerns about the impact of a mastectomy on their female identity, as these dominant discourses might make the woman and others consider such concerns as 'cosmetic' and trivial. Giddens (1990) suggested that supporting individuals to share such narratives can facilitate sense-making that transcends beyond the dualism of bio-medical and social constructions of the body. Exploration however should be approached respectfully and tentatively with an awareness and appreciation that, for many, defence

mechanisms such as detachment and a woman's seeming rejection of her breasts pre-surgery, may also serve a helpful strategy to protect from unwanted anxiety.

By expanding upon the limited evidence base of research within this area, and applying developing theory to practice, clinicians may gain a greater understanding of what is important to women as they debate and navigate prophylactic mastectomy. This knowledge is integral to facilitating more effective and person centred decision-making support and to ensure the greatest overall breast cancer reduction.

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Part 3: Critical appraisal

**Personal and professional reflections of
undertaking the research**

Critical appraisal

1. Overview

The following critical appraisal has been presented with the aim of supporting and consolidating personal and professional reflections made during the research process. Information contained within this critical appraisal was aided by the use of a reflective journal and field notes, which supported the identification of salient thoughts and learning points throughout the experience.

2. Developing a research topic

Although I came to doctoral training with a broad range of clinical experience and curiosity, I had always experienced a strong interest in health psychology and, in particular, how factors such as attribution, self-efficacy and resilience might impact upon the conceptualisation of illness and behaviour. Since starting clinical training, I have been fortunate to have had placements within medical and paediatric psychology and this served to further fuel my interest within the burgeoning area of hereditary disease. Working within the NHS also increased my understanding and appreciation of how the wider systems might best support the individual and their families through potentially difficult treatment decisions. The aforementioned experiences undoubtedly influenced the development of my research and I was enthusiastic to contribute towards the limited psychological evidence base in genetic research and for this to be of use to clinical services and service users.

In May 2013, a month before the National Institute for Health and Care Excellence published guidelines on familial breast cancer (NICE, 2013), the New York Times published an article entitled 'My Medical Choice' (Jolie, 2013). This was an open letter by the American actress and humanitarian Angelina Jolie, where she candidly shared her decision to have prophylactic mastectomy following confirmation of an inherited BRCA gene alteration. This ground-breaking disclosure received unprecedented media coverage and debate around the world. In addition, the longstanding publicity this generated is thought to have raised public awareness of familial cancer and increased referral rates for genetic testing and risk-reducing surgery (Bhatti & Redelmeier, 2015; Evan *et al.*, 2014).

I was fascinated by the public reaction to this article and recalled feeling particularly struck when conversations of genetic risk and prophylactic surgery became more prominent in everyday discourse. Perhaps linked to this, two people that I knew shared that they too had an inherited BRCA gene alteration. Interestingly, one had chosen to have a prophylactic mastectomy, and the other, decided not to. This led me to contemplate what some of the contributing factors may have been to these contrasting decisions, and how women might make sense of their genetic risk and the prospect of preventative surgery.

3. Choosing a methodology

Whilst the literature review revealed the paucity of research exploring decision-making in unaffected women with an inherited BRCA gene alteration, it also highlighted the lack of qualitative research exploring this. Perhaps related to biomedical dominance within this area, existing research was primarily focussed upon quantitative predictors in the uptake of prophylactic surgery (Evan *et al.*, 2009; Petrie *et al.*, 2014) and women's retrospective satisfaction following surgery (Den Heijer *et al.*, 2012; Gopie *et al.*, 2013). Reflecting upon this, and the research aims of the current study, a qualitative methodology was agreed to be the most appropriate approach.

Research supervision was used to explore different qualitative methodologies and to discuss their respective strengths and limitations to the epistemological stance of the research. Due to their noted similarities, initial consideration was given to IPA and grounded theory as the most potentially suitable approaches to explore decision-making and navigation of the prophylactic process. Although advantages were discussed, practical concerns regarding grounded theory, such as the researcher having no preconceptions and theoretical sampling yielding sufficient data for saturation effect, were discussed during research supervision. Grounded theory focuses on the development of theory that emerges through the data and posits that the researcher should not pre-empt participant experience or understanding based on existing literature. The researcher must enter with an open mind and allow the data to guide them until theoretical saturation is achieved (Glaser & Strauss, 1967). Although the point of saturation can be affected by the research question and the sensitivity of the phenomena being investigated, Thomson (2011) suggest that the average sample size suggested for sufficient theoretical saturation

in grounded theory research is between 25-30 participants. However, after meeting with the field supervisor, there was some initial concern that the rigorous inclusion criteria of the study suggested that recruitment may yield a relatively small number of participants. It was however believed that this would be sufficient for thorough analysis, akin to IPA, where importance is placed upon the quality and richness of the data, rather than the number of participants (Larkin & Thompson, 2011). IPA was also chosen as the most appropriate approach, primarily due to its strong philosophical and psychological emphasis upon personal experience and how people make sense of, and apply meaning to their lived experiences (Smith *et al.*, 2009).

4. Navigating the ethical process

Through discussion with research supervisors and the clinical team at the recruitment site, a research proposal was developed and submitted to the university for peer review. I felt reassured when this received positive feedback from academic staff and the Service User Reference Group (SURG) without significant amendments being requested. The process of applying for ethical approval first required the completion of the online Integrated Research Application System (IRAS). Having had no prior experience with IRAS, and through hearing comments from others who had found the system daunting, I felt quite apprehensive about this stage. This did transpire to be quite a complicated and lengthy process but I believe this was predominantly due to unfortunate timing as significant changes were simultaneously occurring within the research governing authority. As such, I often sought consultation with peers, academic staff and other professionals involved in NHS research, to navigate the process correctly.

On completion and submission of the IRAS form, I was invited to attend a Research Ethics Committee (REC) meeting where my research was discussed by a panel who then had the opportunity to ask questions. Whilst I recognised and respected the value of the strict monitoring in NHS research, this felt particularly anxiety provoking at the time. I was however very relieved when only minor recommendations were suggested.

5. Participant recruitment and data collection

Following confirmation of ethical approval, I was eager to start the recruitment phase of the research and fortunately, within two weeks, I felt both relieved and excited to receive

the first participant response slip. Recruitment was enthusiastically facilitated by the field supervisor and through the positive support of staff at the research site. As such, the first three interviews were carried out in relatively quick succession, however this was followed by a period where no further contact was received from prospective participants. During this time, I attended clinical team meetings and a regional conference in genomics to update others on my research progress but also to politely encourage clinicians to hold my research in mind and to further support recruitment. Following this, responses from two further participants were received. Although I had initially hoped to recruit six participants, I felt confident that data from the five interviews was sufficient for a thorough qualitative analysis and I felt reassured that a sample size of between three and six participants is suggested as being appropriate for a doctoral level IPA study (Smith *et al.*, 2009).

During the interviews, I recognised an underlying inclination to respond to participants in more of a therapeutic manner, and later reflected that this was because the role of clinician felt more familiar to me. I became aware, particularly during the first two interviews, that my interview style still incorporated the use of therapeutic techniques such as summarising and paraphrasing. Whilst this was helpful in terms of developing rapport and checking back understanding, it was acknowledged that this also increased the potential for closed responses and monosyllabic agreement from participants. Being aware of this earlier in the data collection stage enabled me to become more mindful of my interactional style in subsequent interviews and I felt able to further develop my confidence in the role of neutral researcher.

Whilst I was able to use the reflective journal to consider my role as both clinician and neutral researcher, I also took the time to consider my response as a woman. Although I did not have an inherited gene alteration, I experienced strong feelings of empathy, admiration and genuine warmth towards the women who participated in the study. Through the use of research supervision, I was also able to recognise that I had perhaps felt more fondly towards those whom I believed shared similar values to my own. For example, women who placed high priority on the importance of family and a strong wish to help other women through charity endeavours and support groups. Some women also spoke of their externalised anger towards cancer and the negative impact it had had on their families. From a personal perspective, I could also relate to this feeling as I have

also experienced loss at the hands of breast and ovarian cancer within my family and this alerted me to my own prejudices and pre-understandings. It therefore felt very pertinent to hear women speak of their determination to beat their risk of cancer, which I personally experienced as both admirable and inspiring.

Following one of the interviews, I was asked by a participant what I would do if I had an inherited BRCA gene alteration. Whilst this was something that I had previously given thought to, it was the first time I had been asked by a participant. I was able to draw on some of my existing clinical skills, but reflected that I had felt quite unprepared to formulate an appropriate response to the question whilst not wanting to appear dismissive or invalidating of their interest. I later considered whether women would find it reassuring to hear that other women might come to the same, albeit hypothetical, conclusion regarding prophylactic surgery. This also led me to wonder how it might have felt for the participant if I had offered a response that was not in-keeping with their own decision or values. Acknowledging how any response to this question may have influenced rapport, I felt fortunate that this was raised after the interview.

6. Transcription and analysis

Early in the research planning, I decided that I wanted to transcribe the interviews myself, recognising this to be an inherent part of the interpretive process (Pietkiewicz & Smith, 2012). Whilst this transpired to be a more time-consuming task than I had perhaps initially anticipated, it did feel like a welcome break at times from the more challenging and demanding aspects of research. Transcribing the interviews also enabled further engagement with the data and allowed closer scrutiny of more subtle aspects such as nuanced tone and hesitations in speech that might have gone unnoticed during the interview. Each interview was transcribed shortly after it occurred and this also provided the opportunity to reflect on, and refine, interview technique for subsequent interviews. For example, by listening back to the interview, I was able to notice instances where I had perhaps missed the potential to further explore themes raised by the participant. This was a useful exercise and I found that as my confidence and awareness increased over time, I was able to feel more mindful during the interviews and to respond more readily to observations in the moment.

The analytic phase of the research was found to be highly absorbing, thought-provoking and enjoyable. As recommended by Smith *et al.*, (2009), data were initially coded with particular attention to semantic, linguistic and conceptual features. Research supervision was also utilised to share examples of coded transcripts and it was helpful to refine my early coding based upon discussion and feedback. For example, this highlighted the need to further develop my conceptual observations where many of my initial interpretations were equivocal and rhetorical. With an awareness of this, I then went back and tried to answer some of my questions and expand upon the interpretations. Reflecting on this, I believe that I had initially experienced some apprehension that my own interpretations might lead the research away from the participant's own account. To support my confidence and understanding, I attended an IPA workshop facilitated by Dr Michael Larkin where I was able to gain further advice and feedback on my coding. This supported my confidence and engagement with the double hermeneutic when trying to make sense of the participant's sense making (Smith, 2011). Peer supervision was also helpful in expanding my consideration of alternative perspectives and interpretations in the data.

Every effort was made to adhere to the idiographic epistemology of IPA by analysing one transcript at a time, however, it sometimes felt difficult to fully disregard similarities noted from previous cases. This was found to be increasingly challenging as the research progressed but I found it helpful to write a brief summary page for each participant and to focus myself by re-reading an individual's summary before I returned to their analysis. On a personal note, I also recognised this phase of the research as particularly poignant as someone close to me was diagnosed with cancer. Whilst at times, it felt difficult to turn towards the research, after a few weeks away from data analysis, I felt able to channel this into my determination to share the voices of the women who participated and to think of the wider impact that my research might have.

Following the completion of case by case analysis, I was enthusiastic to be able to consider themes across cases. However, due to the sheer number of early themes generated, I initially experienced this stage as quite overwhelming. To help manage this, I found it helpful to utilise different coloured post-it notes to aid the visual detection of themes that were shared across cases and was able to move these around as necessary. In addition, as recommended by Smith *et al.*, (2009), I tried not to hold onto emerging themes too tightly but to allow reflexivity in the approach to participant themes.

Techniques such as abstraction and subsumption were used to identify and cluster themes, where prioritisation was given to those more closely linked to the research question and interpreted as ‘objects of concern’ to the participant (Larkin & Thompson, 2011).

As I started to write the research report I was struck by the magnitude of quotes that I wanted to include to illustrate each theme and was initially concerned that the analysis might lose some of the idiosyncrasies of individual respondents. As such, I found it helpful to revisit the data and to make a list of the primary quotes that I believed best represented the research and those which I definitely did not want to lose from the results. This required many re-writes and felt particularly intense and time consuming, but I felt a sense of responsibility to take my time and do it justice as it was important to share the experience and overall message from the women who participated.

7. Dissemination

Given the scarcity of qualitative research exploring prophylactic decision-making in women with a BRCA gene alteration, it feels particularly important that research findings be disseminated appropriately. Firstly, this will include passing on a copy of the completed research report to those participants who expressed a wish to be kept informed. A meeting will also be arranged with the field supervisor and clinical team at the research site where findings from the study will be discussed with the hope that specific recommendations might be incorporated into service delivery going forward.

I intend to publish the research and this has been discussed with the field supervisor who expressed their interest to support this and remain involved throughout the wider dissemination process. In addition, an overview of the research, including a poster presentation, will be shared at a research conference to be held in September 2017.

8. Reflections on professional and personal development

Overall, I found the research process to be a challenging, yet rewarding, learning experience. Managing the multiple demands of research, alongside clinical training and placements, has often felt like it required a whole new level of organisational and multi-tasking skills. I have however frequently become aware of the need to maintain a greater work life balance and recognised in myself the need to take a step back at times. The phrase that I have most commonly used throughout the whole process has been ‘keep it

in perspective' and I genuinely believe this helped me to not feel preoccupied by the all-encompassing details but to frequently remind myself of the bigger picture. Peer support has also been invaluable, particularly in terms of validation, empathy and encouraging each other to take time out and to be a little kinder to ourselves.

Throughout the research journey, I feel I have further developed my confidence, and ability, in designing and undertaking clinically meaningful research. Navigating the process of research within the NHS has also increased my awareness of the practical issues this entails. Prior to training, I had not had the opportunity to undertake qualitative research, but had always been interested in this methodology. As an undergraduate, I elected to complete a module in ethnography and valued the strong emphasis this placed on understanding human behaviour and social phenomenon through the exploration of lived experience (Scott, 2008). I therefore feel particularly fortunate to have had the opportunity to undertake IPA research through doctoral training and this has encouraged me to develop my research skills in new areas. I also recognise how such methodologies can be used to promote the 'patient voice' in healthcare and to explore thoughts and potential barriers to change (Al-Busaidi, 2008). I believe this to be paramount to the encouragement and utilisation of service user involvement in service development and this is something that I will take with me as I embark upon my career as a qualified clinical psychologist working within the NHS. I feel I have undoubtedly grown as a researcher and I look forward to continuing this journey, where the knowledge and experience I have gained to date, will be invaluable to my future endeavours.

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Appendices

Appendix A: Author guidelines – Journal of Genetic Counseling

Guidelines for the Journal of Genetic Counseling retrieved January 2017 from:
<http://www.springer.com/biomed/human+genetics/journal/10897>

General

Manuscripts should be checked for content and style (American English spelling, punctuation, and grammar; accuracy and consistency in the citation of figures, tables, and references; stylistic uniformity of entries in the References section; etc.)

Comments section: Authors should detail in the comments section of the submission that the manuscript is submitted solely to this journal and was not published elsewhere, and disclose details of any previous or anticipated publication history related to the manuscript's content. Submission is a representation that the manuscript has not been published previously and is not currently under consideration for publication elsewhere.

Manuscript Preparation

1. Type double-spaced and include all illustrations and tables. Original research articles should be no longer than 25 double-spaced typed pages and qualitative research no longer than 40 double-spaced typed pages.

2. Title page: A title page is to be provided and should include the title of the article, authors name (no degrees), authors affiliation, and suggested running head. The affiliation should comprise the department, institution (usually university or company), city, and state (or nation) and should be typed as a numbered footnote to the author's name. The suggested running head should be less than 80 characters (including spaces) and should comprise the article title or an abbreviated version thereof. The title page should also include the complete mailing address, telephone number, fax number, and e-mail address of the one author designated to review proofs.

3. Abstract: An unstructured abstract is to be provided, approximately 200 words.

4. Key words: A list of 3-10 key words is to be provided directly below the abstract. Key words should express the precise content of the manuscript, as they are used for indexing purposes.

5. Section headings: All major sections should carry section headings (such as Introduction, Methods, Results, Discussion, Conclusions, etc.) type centered. Side headings in Methods section should include, as appropriate: Participants, Instrumentation, Procedures, and Data Analysis. Side headings in Discussion should include: Study Limitations, Practice Implications, and Research Recommendations. All Acknowledgements (including those for grant and financial support) should be typed in one paragraph (so-headed) on a separate page that directly precedes the References section.

6. Reference list: The journal follows the reference and citation style recommendations of the Publication Manual of the American Psychological Association (APA style). See also: <http://apastyle.apa.org/>

List references alphabetically at the end of the paper. References should include (in this order): last name and initials of authors, year published, title of article, name of publication, volume number, and inclusive pages. Where there are seven or more authors, abbreviate the seventh and subsequent authors as et al. Refer to the references in the text by name and year in parentheses. Multiple citations should be listed alphabetically by author's last name.

7. Illustrations: Illustrations (photographs, drawings, diagrams, and charts) are to be numbered in one consecutive series of Arabic numerals. The captions for illustrations should be provided. Photographs and drawings should show high contrast. Electronic should be in TIFF or EPS format (1200 dpi for line and 300 dpi for half-tones and gray-scale art). Color art should be in the CMYK color space. A hard copy of photographs or illustrations may be requested prior to publication.

8. Tables: Tables should be numbered (with Roman numerals) and referred to by number in the text. Each table should be on a separate sheet of paper at the end of the submission. Center the title above the table, and type explanatory footnotes (indicated by superscript lowercase letters) below the table.

9. Footnotes: Footnotes should be avoided. When their use is absolutely necessary, footnotes should be numbered consecutively using Arabic numerals and should be typed at the bottom of the page to which they refer. Place a line above the footnote, so it is set off from the text. Use the appropriate superscript numeral for citation in the text.

10. Pedigrees: Pedigrees should follow the recommendations for standardized nomenclature accepted by the National Society of Genetic Counselors. Authors should consult the following references for these recommendations:

Bennett, R. L. , Steinhaus, K. A., Uhrich, S. B., O' Sullivan, C. K., Resta, R. G. , Lochner-Doyle, D., Markel, D. S., Vincent, V., & Hamanishi, J. (1995). Recommendations for Standardized Human Pedigree Nomenclature. *Journal of Genetic Counseling*, 4, 267-279.

Bennett, R. L., Steinhaus French, K., Resta, R. G., & Lochner Doyle, D. (2008). Standardized Human Pedigree Nomenclature: Update and Assessment of the Recommendations of the National Society of Genetic Counselors. *Journal of Genetic Counseling*, 17, 424-433.

11. Conflict of Interest: Conflict of interest statements should be present on every manuscript before the References section. The statement should mention each author separately by name. Recommended wording is as follows:

Author X declares that he has no conflict of interest.

Author Y has received research grants from Drug Company A.

Author Z has received a speaker honorarium from Drug Company B and owns stock in Drug Company C. If multiple authors declare no conflict, this can be done in one sentence:

Author X, Author Y and Author Z declare that they have no conflict of interest.

12. Human Studies and Informed Consent: For studies with human subjects, please include the following statement before the References section:

'All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000 (5). Informed consent was obtained from all patients for being included in the study.'

If any identifying information about patients is included in the article, the following sentence should also be included: 'Additional informed consent was obtained from all patients for which identifying information is included in this article.'

13. Animal Studies: For studies with animals, include the following sentence in the manuscript before the References section: 'All institutional and national guidelines for the care and use of laboratory animals were followed.'

If the authors did not carry out animal studies as part of their article they must include the following statement in the manuscript before the References section:

'No animal studies were carried out by the authors for this article'

Appendix B: Database searches

<u>Research question</u>	<u>Database</u>	<u>Search terms utilised and combined</u>
<p>Psychosocial predictors of prophylactic mastectomy in unaffected BRCA mutation carriers</p>	<ul style="list-style-type: none"> • PsychInfo • Medline • Scopus • Web of Science • CINAHL 	<p>BRCA* Mutation alterations 'BRCA mutation' "BRCA alteration" gene genetic*</p> <p>mastectomy mastectom* 'prophylactic mastectomy' prophyla* surgery surg* risk 'risk reduc*' 'RRM'</p> <p>psychosocial psych* predict* decision 'decision making'</p> <p>unaffected asymptomatic breast 'breast cancer'</p>

Appendix C: Data extraction form

Title	
Primary author	
Publication Date	
Country	
Journal details	
Aims:	
Participants: <i>(total number of participants, age, gender, recruitment, inclusion/exclusion criteria)</i>	
Study Type: <i>(design, qualitative/quantitative, prospective/retrospective)</i>	
Measures: <i>(validity, reliability, questionnaire, survey, semi structured interview)</i>	
Results: <i>(power calculation, statistical method)</i>	
Findings and conclusions:	
Limitations: <i>(generalizability, bias, withdrawals. Exclusions – acknowledged by the author)</i>	
Additional Comments:	

Appendix D: Quality appraisal checklist (QualSyst)

Criteria for quantitative studies (2 = fully met, 1 = partially met, 0 = not met)

- 1 Question/objective sufficiently described?
 - 2 Study design evident and appropriate?
 - 3 Method of subject/comparison group selection or source of information/input variables described and appropriate?
 - 4 Subject (and comparison group, if applicable) characteristics sufficiently described?
 - 5 If interventional and random allocation was possible, was it described?
 - 6 If interventional and blinding of investigators was possible, was it reported?
 - 7 If interventional and blinding of subjects was possible, was it reported?
 - 8 Outcome and (if applicable) exposure measure(s) well defined and robust to measurement/misclassification bias? Means of assessment reported?
 - 9 Sample size appropriate?
 - 10 Analytic methods described/justified and appropriate?
 - 11 Some estimate of variance is reported for the main results?
 - 12 Controlled for confounding?
 - 13 Results reported in sufficient detail?
 - 14 Conclusions supported by results?
-

Criteria for qualitative studies (2 = fully met, 1 = partially met, 0 = not met)

- 1 Question/objective sufficiently described?
 - 2 Study design evident and appropriate?
 - 3 Context for the study clear?
 - 4 Connection to a theoretical framework/wider body of knowledge?
 - 5 Sampling strategy described, relevant and justified?
 - 6 Data collection methods clearly described and systematic?
 - 7 Data analysis clearly described and systematic?
 - 8 Use of verification procedure(s) to establish credibility?
 - 9 Conclusions supported by the results?
 - 10 Reflexivity of the account?
-

Reference:

Kmet L, Lee R, Cook L. Standard Quality Assessment Criteria for Evaluating Primary Research Papers From a Variety of Fields. 2004. Edmonton: Alberta Heritage Foundation for Medical Research. 2011

Appendix E: Quality appraisal scores (QualSyst) of studies included in the review

	Beattie <i>et al.</i> (2009)	Chai <i>et al.</i> (2014)	Friebel <i>et al.</i> (2007)	Garcia <i>et al.</i> (2014)	Haroun <i>et al.</i> (2011)	Hesse-Biber (2016)	Julian-Reynier (2010)	Metcalfe <i>et al.</i> (2010)	Singh (2013)	Skytte (2010)
1. Question or objective sufficiently described?	2	1	1	1	2	2	2	1	2	2
2. Design evident and appropriate to answer study question?	2	2	2	2	2	2	2	2	2	2
3. Method of subject selection (and comparison group if applicable) or source of information/input variables is described and appropriate?	2	2	2	2	2	1	2	2	2	2
4. Subject (and comparison group if applicable) characteristics or input variables/information sufficiently described?	2	2	2	2	2	2	2	2	2	2
5. If interventional and random allocation was possible, was it described?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
6. If interventional and blinding of investigators was possible, was it reported?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
7. If interventional and blinding of subjects was possible, was it reported?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
8. Outcome (if applicable) exposure measure(s) well defined & robust to measurement/misclassification bias? Means of assessment reported?	2	2	2	2	2	2	2	2	2	2
9. Sample size appropriate?	1	1	1	1	1	1	1	1	1	1
10. Analytic methods described/justified and appropriate?	2	2	2	2	2	2	2	2	2	2
11. Some estimate of variance is reported for the main results?	2	2	2	2	1	2	2	1	2	2
12. Controlled for confounding?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
13. Results reported in sufficient detail?	2	2	2	2	2	2	2	2	2	2
14. Conclusions supported by results?	2	2	2	2	2	2	2	2	2	2
Summary score (total sum/total possible sum)	0.95	0.90	0.90	0.90	0.90	0.90	0.95	0.85	0.95	0.95

Appendix F: Statement of epistemological position

Interpretative Phenomenological Analysis (IPA) was chosen due to its psychological focus upon personal experience and how people make sense of, and apply meaning to their lived experiences (Smith, 2011). IPA is informed by three conceptual touchstones of philosophical thought, these are: phenomenology: hermeneutics: and idiography (Smith *et al.*, 2009). Firstly, the approach is phenomenological meaning that it seeks to understand how individuals experience themselves and the world they live in. IPA is also hermeneutic as it involves the interpretation and ‘meaning making’ of experience and it is suggested that this cannot occur without existing conceptions and prejudices. This study involved a double hermeneutic where the researcher sought to make sense of the participant’s own sense making. Thirdly, IPA is idiographic and committed to the in-depth understanding of how particular experiential phenomena are understood from the individual perspective, which implies focus on the particular rather than the general (Larkin & Thompson, 2011). The idiographic and hermeneutic nature of IPA that enables a researcher to interpret the lived experiences of their participants, is also congruent with contextual constructionism (Smith *et al.*, 2009).

During this study, the researcher adopted a contextual constructionist epistemological stance which, in contrast with realist frameworks, does not construe reality as something ‘true’ which can be revealed through rigorous adherence to the method of enquiry (Madill, 2000). This supports the endeavour towards understanding the ‘reality’ of experiences whilst also acknowledging the potential influences which may shape an individual’s interpretation of an experience. As such, a contextual constructionist epidemiology is of particular relevance to IPA, where the researcher and participant are both recognised as conscious beings interpreting and acting on the world around them (Giorgi, 1995). This subjectivity in the production of knowledge, and the way information is perceived in the social world, can depend upon an individual’s beliefs, experiences and expectations (Bunge as cited in Madill *et al.*, 2000).

Ideas from social constructionism were also drawn into the discussion to consider how societal discourses might impact upon how women made sense of gender (namely feminism and breasts) and illness.

References

Giorgi, A. (1995). Phenomenological psychology. In J.A. Smith, R.Harre, & L.Van Langenhove (Eds.) *Rethinking psychology*. London: Sage.

Larkin, M. & Thompson, A.R. (2012). Interpretative phenomenological analysis. In A. Thompson & D. Harper (Eds.) *Qualitative research methods in mental health and psychotherapy: A guide for students and researchers*. Oxford: Wiley.

Madill, A., Jordan, A. & Shirley, C. (2000). Objectivity and reliability in qualitative analysis: Realist, contextualist and radical constructionist epistemologies. *British Journal of Psychology*, 91, 1-20.

Smith, J.A. (2011). Evaluating the contribution of interpretative phenomenological analysis. *Health Psychology Review*, 5(1), 9-27.

Smith, J.A., Flowers, P. & Larkin, M. (2009). *Interpretative phenomenological analysis: Theory, method and research*. Sage: London.

Appendix G: Correspondence from Research Ethics Committee



Health Research Authority

[REDACTED] Research Ethics Committee
[REDACTED]
[REDACTED]
[REDACTED]

17 March 2016

Mrs Lauren Wright
Trainee Clinical Psychologist
[REDACTED]
[REDACTED]
[REDACTED]
[REDACTED]
[REDACTED]

Dear Mrs Wright

Study title:	Women's experience of decision making regarding prophylactic mastectomy
REC reference:	16/EM/0098
Protocol number:	UNOLE 0560
IRAS project ID:	197568

Thank you for your letter of 15 March 2016, 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.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact the REC Manager, [REDACTED].

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.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the

study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for NHS permission for research is available in the Integrated Research Application System, www.hra.nhs.uk 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 management permissions from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to contest the need for registration they should contact [REDACTED] the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

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).

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).

Non-NHS sites

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Indemnity letter UoL]	N/A	04 February 2016
Interview schedules or topic guides for participants [Interview guide discussion prompts]	1	22 February 2016
Interview schedules or topic guides for participants [Interview guide discussion prompts]	1	22 February 2016
Letters of invitation to participant [Participant invite]	2	29 January 2016
Letters of invitation to participant [Participant Invite (signed)]	3	10 March 2016
Letters of invitation to participant [Participant Invite (signed)]	3 (with highlighted changes)	14 March 2016
Other [Participant response form]	2	29 January 2016
Other [Service User feedback]	N/A	
Other [Research costs]	1	20 January 2016
Other [NHS Letter of Access]	N/A	
Other [GCP certificate - Lauren Wright]	1	16 December 2015
Other [Consent certificate - Lauren Wright]	N/A	10 February 2016
Other [Amendments Summary - Provisional REC]	1	15 March 2016
Other [Signed Participant Invite]	3	10 March 2016
Participant consent form [Participant consent]	2	29 January 2016
Participant consent form [Participant Consent]	3	10 March 2016
Participant consent form [Participant Consent]	3 (with highlighted changes)	14 March 2016
Participant information sheet (PIS) [Participant information]	2	29 January 2016
Participant information sheet (PIS) [Participant Information]	3	10 March 2016
Participant information sheet (PIS) [Participant Information]	3 (with highlighted changes)	14 March 2016
REC Application Form [REC_Form_18022016]		18 February 2016
Referee's report or other scientific critique report [Peer review]	N/A	10 January 2016
Research protocol or project proposal [Research Proposal]	5	10 March 2016
Research protocol or project proposal [Research Proposal]	5 (with highlighted changes)	14 March 2016
Summary CV for Chief Investigator (CI) [Chief Investigator (LW) CV]	N/A	13 January 2016
Summary CV for supervisor (student research) [Academic Supervisor (SB) CV]	N/A	01 January 2016
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Research summary]	1	10 January 2016

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 HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:

<http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at

<http://www.hra.nhs.uk/hra-training/>

16/EM/0098

Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

[Redacted signature]

[Redacted name]

Chair

Email:

[Redacted email address]

Enclosures:

"After ethical review – guidance for researchers"

Copy to:

[Redacted copy to list]

Appendix H: Correspondence from Research and Development

Wright Lauren

From: [REDACTED] - Head of Research Operations [REDACTED]
Sent: 24 March 2016 09:43
To: Wright, Lauren
Cc: [REDACTED] - Research Support Officer
Subject: [REDACTED] - R&I Authorisation confirmation

Dear Lauren

I am pleased to confirm that with effect from the date of this email, the above study has Trust Research & Innovation authorisation to commence at [REDACTED]. The research must be conducted in line with the Protocol and fulfil any contractual obligations agreed. If you identify any issues during the course of your research that are likely to affect these obligations you must contact the R&I Office as soon as possible.

In order for the [REDACTED] Trust to comply with targets set by the Department of Health through the 'Plan for Growth', there is an expectation that the first participant will be recruited within 70 days of receipt of a Valid Application. The date that a Valid application was received was **23rd March 2016**. You therefore have **69 REMAINING DAYS** to recruit your first participant. It is essential that you notify the [REDACTED] Data Management Team as soon as you have recruited your first participant to the study, and ensure that the date is recorded on the [REDACTED] Database by your local [REDACTED] User. The [REDACTED] Data Management team can be contacted on [REDACTED] or by phone [REDACTED].

If we have not heard from you within the specified time period we will contact you not only to collect the data, but also to record any issues that may have arisen to prevent you from achieving this target. It is essential that you get in touch with us if there is likely to be a problem in achieving this target so that we can discuss potential solutions. The Trust is contractually obliged to meet the 70 day target and if an adequate reason acceptable to the NIHR has not been submitted to explain the issues preventing the recruitment of your first participant, the Trust will be financially penalised. In addition, we are required to publish the Title, REC Reference number, local target recruitment and actual recruitment as well as 70 days data for this study on a quarterly basis on the [REDACTED] public accessed website.

Undertaking research in the NHS comes with a range of regulatory responsibilities. Please ensure that you and your research team are familiar with, and understand the roles and responsibilities both collectively and individually.

Documents listing the roles and responsibilities for all individuals involved in research can be found on the R&I pages of the Public Website. It is important that you familiarise yourself with the Standard Operating Procedures, Policies and all other relevant documents which can be located by visiting [REDACTED].

The R&I Office is keen to support and facilitate research where ever possible. If you have any questions regarding this or other research you wish to undertake in the Trust, please contact this office. Our contact details are provided on the attached sheet.

Please note that a letter confirming authorisation will not be sent. Please retain a copy of this email in your site file.

We wish you every success with your research.

With Best Wishes
[REDACTED]

Appendix I: Participant invite



Women's experience of decision making regarding prophylactic mastectomy

Dear Madam,

We are writing to invite you to take part in a research study which is being undertaken by a Doctoral student in Clinical Psychology at the University of Leicester. The title of the study is: Women's experience of decision making regarding prophylactic mastectomy. According to our recent records you have been identified as someone who may be able to offer a valuable perspective relevant to this research.

The study is interested in the experiences of women who have an inherited BRCA gene alteration, who have made the decision to have a prophylactic mastectomy. The aim is to understand more about how women may arrive at their decision to have surgery and use these findings to inform how we may support others in the future. Your participation would involve taking part in an interview discussion to share your own experience of this.

The enclosed information sheet provides further details regarding the study and what to expect if you wished to participate. Please read through this information and contact the researcher should you have any further questions or queries. If you decide that you would like to participate in the study, then please fill in the reply slip and return it in the stamped addressed envelope provided. The main researcher, Lauren Wright, will then contact you to discuss this further and arrange a convenient time for you both to meet. Thank you for taking the time to consider participating in the research study.

Yours sincerely,

A black rectangular redaction box covering the signature area.

A small black rectangular redaction box covering a line of text.

A long black rectangular redaction box covering a line of text.

Appendix J: Participant information letter



Women's experience of decision making regarding prophylactic mastectomy

What is the purpose of the study?

Over the last decade the number of women having genetic testing has significantly increased. There has also been a growing number of women who then go on to have their breast or breast tissue removed to reduce the risk of cancer (prophylactic mastectomy). Whilst there has been some recent research exploring women's satisfaction following risk reducing surgery, very little is known about how women may reach their decision to have surgery.

Why have you been invited to take part?

You have been invited to take part in the study because [REDACTED] [REDACTED] have identified you as someone who has recently decided to proceed with surgery and be able to offer a valuable perspective relevant to this research. The study is being undertaken as part of an educational qualification and the [REDACTED] have not passed on any of your personal information or details to the University. They have however agreed to support the study by sending invitation/information letters to women who may be able to assist by taking part. It should be noted that a decision not to take part in the study would not impact on your usual care.

What would be involved in taking part?

If you wish to take part, you will be invited to meet with the researcher for an interview to discuss your own experience. Interviews usually take approximately one hour. There is some flexibility on where the meeting could take place (e.g. hospital, university, your home) depending on where you feel most comfortable to talk. If travel is required, your expenses will be reimbursed by the University of Leicester (e.g. fuel, car parking).

During the interview, the researcher will have some brief open ended questions however discussion will also be guided by yourself and what you feel comfortable to talk about in relation to the topic. The interview will be audio recorded so that the researcher is able to type up the meeting afterwards.

What would you be asked about?

During the interview you would be asked about your personal experience of being faced with the option of prophylactic surgery. The research is interested in how you may have made sense of this experience and some of the key considerations that may have affected and/or influenced the decision-making process.

What will happen with your personal information?

All of the information that you provide will be kept strictly confidential at all times. No personally identifiable data will be held electronically and your audio recording will be destroyed immediately after being typed up (transcribed). All written transcripts of interviews will be anonymised and stored securely on an NHS password protected memory stick. Your name would never be attached to your transcribed interview; instead a unique code will be used and will be known only by the researcher. While some quotations from interviews will be used to illustrate the findings, no information that could identify you would be included within the final written report or in any potential publication of the research study.

Are there any risks in taking part?

There are no specific risks involved in taking part in the study, however during the interview you will be asked to talk about your experiences of genetic testing and prophylactic mastectomy. It is appreciated that this may be a difficult topic to discuss and could raise some emotions for you. You will not be pressured to talk about anything you feel uncomfortable discussing and you will be in control of how much information you choose to share. Should you wish to have further support or talk to a member of the clinical care team following the interview, you would be provided with contact details for [REDACTED]

What if I am harmed by the study?

It is very unlikely that you would be harmed by taking part in this type of research study. However, if you wish to complain or have any concerns about the way you have been approached or treated in connection with the study you should as to speak to [REDACTED]

[REDACTED]. If you remain unhappy and wish to address your concerns or complaints on a formal basis, you should contact [REDACTED]

[REDACTED] In the event that something does go wrong and you are harmed during the research, and this is due to the event of someone's negligence, then you may have grounds for a legal action for compensation against the University of Leicester but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you (if appropriate).

What are the advantages of taking part?

Some people can find it a helpful experience to discuss matters that are personal and important to them. Initial feedback from women who have utilised the service and been involved in advising the study was that it was deemed a particularly relevant and important topic for women to discuss and share. It is hoped that findings from this study could be utilised to inform and support other women considering prophylactic surgery. If we can gain a greater understanding of women's experiences when faced with such choices, this could inform how to improve support and information offered by healthcare services.

How will the findings of the study be used?

The findings from the study will be written up and presented in a research report format as part of a Doctoral thesis. This is anticipated to be completed by Autumn 2017 – if you would be interested in receiving a summary of the findings please notify the researcher. A summary of the study will also be presented in poster format and shared with local clinicians. It is planned that the finished study will be published in a relevant academic journal.

What if I want to withdraw from the study?

Should you decide that you no longer wish to take part in the study, please make contact with the lead researcher to confirm this. Your decision would be fully respected and you would not have to indicate a reason for doing so. Any audio recorded or written data that you provided for the purpose of the study would be destroyed immediately and would no longer be included in the research. It is requested however that your wish to withdraw from the study is confirmed with the lead researcher before 1st January 2017. After this point, it would be difficult for your contribution to be removed without invalidating the study itself.

Who is funding and reviewing the research?

The research is being funded by the University of Leicester (Doctorate in Clinical Psychology) and supported by [REDACTED]. The study has been reviewed and approved by several members of the clinical staff team at the University of Leicester and by the [REDACTED] Research Ethics Committee [REDACTED].

Further information and contact details

If you would like to receive further information or to discuss aspects of the study, please make contact with the lead researcher, Lauren Wright, [REDACTED]

[REDACTED] Additional points of contact include [REDACTED]
[REDACTED]
[REDACTED]

What to do next

If you would like to take part in the research study, please complete the enclosed response slip. This should then be returned in the stamped address envelope provided. Once the researcher has received this, they will make contact with you via your preferred details/method where you can arrange a suitable date and time for you both to meet. Alternatively, you may prefer to contact the researcher directly (telephone or email) to express your interest in taking part and to arrange a meeting.

Appendix K: Participant response form



Title: Women's experience of decision making regarding prophylactic mastectomy

Name of Researcher: Lauren Wright, Trainee Clinical Psychologist, University of Leicester

To express your interest in taking part in this research, please return this response slip in the enclosed stamped addressed envelope.

Alternatively, you can contact the researcher directly via email [REDACTED] or by telephone [REDACTED]

Please circle your desired response.

I am interested in taking part: YES / NO

Your name:.....

Phone/mobile:.....

Email:.....

How would you prefer to be contacted?

.....

Appendix L: Participant consent form



Title: Women's experience of decision making regarding prophylactic mastectomy
Name of Researcher: Lauren Wright, Trainee Clinical Psychologist, University of Leicester

If you would like to participate in the research study and are happy to proceed further please could you read and complete (initial and sign) the consent information below.

- I confirm that I have read and understood the participant information letter (Version2 - 29/01/16) and have had the opportunity to ask further questions regarding the above study and these have been answered satisfactorily. ☐
- I understand that my participation is voluntary and I have the right to withdraw from the study at any time. ☐
- I understand that I will be interviewed by the Researcher and that this interview will be audio recorded and typed up. ☐
- I understand that data collected during the study may be looked at by individuals from regulatory authorities, the NHS Trust or the Sponsor, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data. ☐
- I understand that data from the interview will be kept securely on an NHS encrypted memory stick, stored at the University of Leicester and that the recording will be deleted once it has been typed up. ☐
- I understand that my data will be completely anonymised and my identity will be protected throughout all aspects of the study. ☐
- I understand that some anonymised quotes from the interview will be included as part of a Doctoral thesis and that results may be published in a relevant academic journal. ☐
- I agree to take part in this research study. ☐

Name of Participant

Signature of Participant

Date

Name of Researcher

Signature of Researcher

Date

Appendix M: Interview schedule and discussion prompts

The researcher used some of the following prompts to support participants to recount the experiences and meaning-making that they thought was pertinent to their decision to opt for prophylactic mastectomies. This was used as a guide, rather than to bias the interview to the issues the researcher thought were key. The following topic questions were utilised in a non-rigid manner to provide prompts and re-orientate the interview if content moved too far off topic. The researcher utilised exploratory phrases and open ended questions with the aim of providing rich data relating to participants' experiences and meaning-making.

- Can you tell me about your journey to genetic testing?
- How did you make sense of your genetic test results?
- How did you feel regarding preventive options?
- What was your experience of your decision-making?
- How would you describe your decision-making to have prophylactic mastectomy?
- What were some of the considerations for you in choosing to have surgery?
- Were you aware of anything that may have supported or hindered your decision-making? (e.g. personality, previous experience)
- Were any others involved in your decision-making? (e.g. family, friends)
- Were there any other considerations that may have influenced your decision? (e.g. public/media perceptions of prophylactic mastectomy)
- How do you feel now you have reached your decision to have surgery?
- Is there any further information and/or support that could have helped you throughout the decision-making process?

Appendix N: Stages of Interpretative Phenomenological Analysis (IPA)

Smith *et al.*, (2009) advocate a number of stages in the process of Interpretative Phenomenological Analysis (IPA) and these were drawn upon to inform analysis in the current study. These stages, as described below, were used flexibly and iteratively by the researcher, as recommended by Larkin and Thompson (2011).

- The first stage involved listening back to the audio-recorded interview and then repeated reading of the transcript to support immersion with the data.
- Maintaining an idiographic stance, free textual analysis was then conducted on the transcribed interview. This involved the researcher maintaining an open stance and making note of any initial interest and observations within the transcript.
- Developing this coding, particular attention was then paid to the following features:
 - Descriptive comments – Highlighting what mattered to the participant at ‘face value’, for example, key words, objects of concern or explanation.
 - Linguistic comments – Focussing on the functional aspects of language and how the transcript reflects how content and meaning were presented.
 - Conceptual comments – Involving the researcher’s sense-making and interpretations of the participant’s sense-making. Whilst this inevitably draws upon the researcher’s experiential knowledge, Smith *et al.*, (2009) highlight the importance of interpretation deriving from the participant’s account, rather than being externally imported.
- The next stage of analysis involved the development of emerging themes. Pietkiewicz and Smith (2012) describe this as formulating a concise phrase, at a slightly higher level of abstraction or conceptualisation, that remains grounded in the participant’s account.

- The above steps were then replicated for each transcript. The final stage involved the exploration of patterns and connections across themes, which was assisted through the mapping of themes to consider how the researcher believed they might reflect convergence and divergence within participant's accounts. To maintain reflexivity, and to minimise the potential for researcher bias, the researcher had regular supervision with an academic who was conversant in IPA methodology.

References

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Pietkiewicz, I. & Smith, J.A. (2012). A practical guide to using interpretative phenomenological analysis in qualitative research psychology. *Psychological Journal*, 18(2), 361-369.

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Appendix O: Example of coded transcript (Anna)

[illegible]

Appendix P: Emerging theme development (Anna)

Emerging themes	Line	Quote	Exploratory/interpretative comments
Centring life/centring surgery	383	I did have to kind of, after a couple of years together say, start to go into panic mode, I'm getting on and you're still young but I need to get married soon so I can have children before I'm too old	Life trajectory – getting married & having children – knowledge of positive test results could have jeopardised both – Taking control and proceeding with plans by putting BRCA to one side (mum delayed testing to – protection?)
	354	I thought well I've already had children so why risk passing that gene on to a third child. I've probably already risked it with the two, because I didn't know, although I did know before I was pregnant that it was a possibility that I carried the gene, and, um, I very happily went off and got pregnant, and pregnant again, without considering that	
Changing my life trajectory	25	She [mum] put off being tested until after the wedding, um, because she didn't want any result hanging over my wedding plans	Self describes as a planner/organiser - likes to be in control and prepare for the worst (knowledge is power – better to know) – contradiction to how testing/surgery was approached (delayed) – was this different because it went against original life plans (also pressure to get married have children due to age) - I won't let this change my life (overarching life plan)
	584	I guess I'd already made the plans before receiving the actual diagnosis, before I'd had the blood test I'd kind of made plans, that's just how I am though (laughs) jump ahead to the future	
Importance of planning	302	Once you know you can't go back and not know, which is a big thing	Life decisions/plans resentfully revolving around preparing for & having the surgery more recently - starting on a path that excludes other things to the detriment of original life plan/trajectory?
	376	I think a benefit of having the test first is that it gives you time so if you don't have a family, you have that time to think that now might be the time to have a family, it, it speeds things up but it also allows you the time to make plans or change plan	
	390	You don't know what's gonna happen next and you might want to do certain things and have to change your plans, all of your plans, um, to revolve around having this surgery and getting your risk right down	
	58	At that time I put off taking any steps towards having surgery	

Appendix Q: Examples of superordinate theme development



Appendix R: Frequency of emergent themes

		Anna	Beth	Claire	Donna	Emily
It's a no-brainer	Assessing the risk	x	x	x	x	x
	Going with my instinct	x	x	x		x
	Living with my decision	x	x	x	x	x
Good breast/bad breast	Ticking time bombs		x	x	x	x
	My feminine identity	x	x	x	x	x
Big B on my shoulder	Weighed down with worry	x	x	x		x
	Racing against the clock		x	x	x	x
The preciousness of life	Breaking the cycle of loss	x	x	x		x
	Protecting my family	x	x	x	x	x
	Grabbing life	x	x	x	x	x

Appendix S: Chronology of the research process

