



**RISK-REDUCING MASTECTOMY PERCEPTIONS
AND DECISION-MAKING AMONG WOMEN
AT HIGH RISK OF BREAST CANCER**

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June 2017

Submitted in partial fulfilment of the degree of Doctorate in Clinical Psychology,

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Acknowledgements

Firstly, I am especially grateful to all the people who selflessly gave their time to participate in this study and who kindly shared their stories, for without them this research would not have been possible.

I am also very thankful to my supervisors, Dr Steve Brown, Prof Peter Salmon and Dr Louise Fairburn, for sharing their knowledge and expertise, for their valuable advice and for guiding me through the course of this research.

Thank you also to my classmates for being a constant source of kindness, support and fun over the past three years. Last but not least, I am extremely grateful to my partner, my family and my friends for their never-wavering encouragement, patience and love each step of the way.

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Thesis Overview

This thesis consists of two chapters, a systematic review and an empirical paper, supplemented with additional information and supporting documents that can be found in the appendices. Both chapters are intended for publication and thus have been written in the style of the identified journal for submission, *The European Journal of Cancer Care*. The author guidelines followed can be found in Appendix A.

Recent advancements in genetic testing have led to more accurate estimations of some individuals' risks of developing breast cancer. A number of options are available for women at increased risk of this illness, defined as those who have a 30% lifetime risk of developing breast cancer due to their family history and/or to deleterious genetic mutations (National Institute of Health and Clinical Excellence, 2013). These alternatives are close surveillance using screening procedures, risk-reducing medication also known as chemoprevention, and surgical options. The latter includes risk-reducing mastectomy (RRM), the surgical removal of breast tissue with or without immediate or delayed reconstruction. There has been a notable increase in requests for RRM in recent years (Evans et al., 2015), and the knowledgebase regarding patients' decision-making continues to grow. In the United Kingdom, efforts have been made to establish guidelines for professionals working with patients who are considering RRM (British Psychological Society's Division of Clinical Psychology Faculty for Oncology and Palliative Care, 2017).

Given that the choice of undergoing RRM can be complex and influenced by wide-ranging personal and societal factors, Chapter One aims to synthesise both qualitative and quantitative literature that explored women's views of risk-reducing breast surgery and of factors affecting their decision-making regarding this procedure, specifically when they are considering at least one other risk management alternative due to being at familial high risk of developing breast cancer.

The findings of the literature review echo research within the wider literature acknowledging the multiple factors influencing patients' reasons for requesting RRM, such as risk perceptions or estimations (Fielden, Brown, Saini, Beesley, & Salmon, 2017) and psychological factors such as fear and worry about cancer (Beesley, Holcombe, Brown, & Salmon, 2013). In light of the above, Chapter Two aimed to gain a further understanding of patient's views about RRM and of how they reach their decision to opt for this procedure. Using a qualitative methodology, this study contributes to the existing research by furthering our understanding of these issues in the context of decision-making theory, highlighting that patients who are at high risk of breast cancer might reach RRM decisions heuristically.

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**The Views on Risk-Reducing Mastectomy of Women at High Familial Risk
of Breast Cancer Considering Risk Management Strategies:
A Mixed-Methods Systematic Review.¹**

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¹ To be submitted to *European Journal of Cancer Care*. Word limit: 8,000 words excluding references, figures and tables.

Abstract

Women at increased risk of breast cancer face important decisions regarding risk-reducing options and early detection strategies. The aim of this mixed-methods systematic review was to examine women's views of risk-reducing mastectomy and how they make decisions regarding this procedure when they are considering at least one other risk management alternative due to being at familial high risk of developing breast cancer. A total of 13 studies published in peer-reviewed journals were included, of which three were qualitative studies and ten were quantitative studies. Results evidenced that BRCA mutation carrier status, perceived risk of breast cancer, and cancer-related worry or fear might predict women's RRM decisions. Impact on body image and perceived confidence in risk-reducing or early detection options were highlighted as views and attitudes that might influence risk management strategy selection. However, these variables were explored by small numbers of studies, and research designs differed greatly, limiting conclusions. The great variation among participant samples also limited comparability of results. Gaps in the literature are identified with suggestions for further research directions. Future studies should explicitly incorporate theoretical models to explore how psychosocial variables and cultural differences impact women's views of risk-reducing mastectomy and their decisions.

Keywords: breast cancer, BRCA, risk-reducing mastectomy, mixed-methods systematic review.

Introduction

Accounting for 15% of new cancer diagnoses, breast cancer is the most common cancer in the United Kingdom (UK), with an estimated 1 in 8 women being diagnosed with breast cancer during their lifetime (Cancer Research UK, 2015). There are a number of ways of defining risk of developing breast cancer. In the UK, guidelines by the National Institute for Health and Care Excellence (NICE, 2013) refer to three levels of risk: general population risk, moderate risk and high risk. The latter indicates that women have a 30% or greater chance of developing the illness in their lifetime.

The identification of two breast cancer susceptibility genes, BRCA1 and BRCA2 (Miki et al., 1994; Wooster et al., 1995), has represented a significant advancement in the prognosis, care and treatment of this type of cancer. An estimated 5 – 10% of all breast cancers are caused by mutations in the BRCA1 and BRCA2 genes (Easton, Bishop, Ford, & Crockford, 1993), and individuals with a mutation in these genes have an elevated lifetime risk for breast cancer (55 – 85%) and ovarian cancer (16 – 60%) (Easton, Ford, & Bishop, 1995; Struwing et al., 1997).

Thus, genetic testing for deleterious mutations in the abovementioned genes is currently recommended for women with a strong family history of breast and ovarian cancer (Hampel et al., 2004) and with those who have received a diagnosis of breast cancer (NICE, 2013). In addition to genetic and familial susceptibility, increased risk of developing breast cancer is also linked to personal history of the disease, age, age at menarche and menopause, age at pregnancy, weight, alcohol intake, hormone replacement therapy use, use of oral contraceptives (McPherson, Steel, & Dixon, 2000) and ethnicity or ancestry, with genetic mutations occurring more frequently in Ashkenazi Jews, French Canadians, or Icelanders (National Comprehensive Cancer Network, 2008).

In the UK, those who are identified as at moderate or high risk of breast cancer are offered risk management measures in order to decrease morbidity (NICE, 2013). These evidence-based recommendations vary depending on women's ages and risk factors, and include surveillance or screening, chemoprevention, and risk-reducing surgery.

Risk-Reducing Options and Early Detection

Radiographic mammograms and/or Magnetic Resonance Imaging (MRI) screening is not preventive of cancer, but can enable early detection of breast cancer and consequently improve its treatment and prognosis (NICE, 2013). However, this type of surveillance can produce false positive and false negative results, which can cause worry and might lead to unnecessary treatment or lack thereof.

The female hormone oestrogen is associated with the development and growth of the majority of breast cancers (Jensen & Jordan, 2003). The use of Selective Estrogen Receptor Modulators (SERMs) such as tamoxifen or raloxifene, and of the aromatase inhibitor anastrozole can inhibit the development of breast cancer among women at increased risk of oestrogen-receptor positive breast cancer (NICE, 2013). A recent meta-analysis from nine randomised SERM trials reported a 38% reduction in overall breast cancer incidence and a 51% reduction in oestrogen-receptor positive tumours (Cuzick et al., 2013). The preventive effect of tamoxifen can be long-lasting (Cuzick et al., 2015), however its use has been linked to increased risk of endometrial cancer and venous thromboembolisms, whilst its side effects including menopausal symptoms (The Royal Marsden NHS Foundation Trust, 2016).

Risk-reducing mastectomy (RRM), also known as prophylactic mastectomy (PM), and risk-reducing oophorectomy (RRO) may significantly reduce the risk of developing breast and ovarian cancer for women who are carriers of mutated BRCA1 and BRCA2 genes (Burke et al., 1997). RRM is the surgical removal of breast tissue, often with immediate or

delayed breast reconstruction. RRM can be bilateral (BRRM) if performed on both breasts, or contralateral (CRRM) if performed upon the non-affected breast after a treatment mastectomy. RRM can result in an 85 – 90% decrease of cancer risk (Rebbeck, Kauff, & Domchek, 2009), however there remains a small risk of developing breast cancer after prophylactic surgery because the surgical removal of all breast tissue cannot be guaranteed (Lopez & Porter, 1996; Willemsen et al., 1998). RRM is an irreversible surgical procedure and it is not without potential risks, including those associated with anaesthesia and post-operative complications such as residual pain or discomfort in the chest area (Gahm, Wickman, & Brandberg, 2010). Furthermore, postoperative regret after undergoing RRM has been reported in the literature, for example associated with factors such as psychological distress and the unavailability of psychological and rehabilitative support (Payne et al., 2000), cosmesis and concerns about body image (Gopie et al., 2013), and sexual dysfunction (Altschuler et al., 2008; Frost et al., 2000). Nevertheless, RRM might provide psychological benefit for women who are at high risk of breast cancer and who fear developing the disease (Bebbington & Fallowfield, 2003), since it has been linked with decreased worry and distress (den Heijer et al., 2012) and a reduction in cancer-related intrusive thoughts (Bresser et al., 2007).

RRO is the surgical removal ovaries, whilst risk-reducing salpingo-oophorectomy (RRSO) is the surgical removal of fallopian tubes and ovaries, to lower the risk of developing ovarian cancer and breast cancer. These surgical procedures may reduce the risk for ovarian cancers by 85% to 90% and of breast cancer by 40% to 70% (Finch et al., 2014); however, there remains a residual risk of developing peritoneal cancer (Rebbeck et al., 2009). RRO and RRSO cause infertility, are associated with immediate surgical menopause and menopausal symptoms (Domcheck & Rebbeck, 2007), and can lead to an increased risk of

osteopenia and osteoporosis, cognitive dysfunction and cardiovascular disease (Guidozzi, 2016).

Factors Influencing RRM Decisions

The management of an increased risk of breast cancer may pose decisional dilemmas for affected women. Decision-making processes regarding breast cancer screening and risk management behaviours are influenced by women's risk perceptions. Research shows that despite genetic counselling, women with a family history of breast cancer tend to overestimate their risk (Caruso et al., 2009; Sivell et al., 2008). Additionally, holding relatively accurate beliefs about their risk level is not always predictive of women's engagement in the recommended risk management options (Keogh, McClaren, Apicella, & Hopper, 2011). Furthermore, women's intentions and decisions to undergo prophylactic mastectomies and oophorectomies are affected by psychological factors, such as perceived risks and benefits, and cancer-related worry (Antill et al., 2006; Madalinkska et al., 2007; van Dijk et al., 2003), which have been found to be predictive of more favourable attitudes towards RRM (Stefanek, Enger, Benkendorf, Flamm Honig, & Lerman, 1999).

Socio-cultural factors influence norms and attitudes about breast cancer risk perception and preventive options, including among physicians (Bouchard et al., 2004; den Heijer et al., 2013). Therefore, these factors may play a role in risk-reducing decision-making (Julian-Reynier et al., 2001; See et al., 2005) and in clinical recommendations for women at increased risk of breast and ovarian cancer (Eisinger, Geller, Burke, & Holtzman, 1999), which differ between countries. Public awareness and media attention might also influence women's choice or preference for RRM, for example shaping their perceptions of recovery and of aesthetic results (Braude, Laidsaar-Powell, Gilchrist, Kirsten, & Juraskowa., 2017).

Models for Health Prevention Decision-making

A number of models have been developed and investigated to explain factors influencing health-related decisions and engagement. Evidence strongly indicates that individuals' risk perceptions, risk preferences and decision-making processes are greatly influenced by emotional interpretations of information and inferential rules called heuristics (Lerner & Keltner, 2001; Loewenstein, 2001; Reyna, 2004). Heuristics are mental operations which can be used simultaneously when processing complex information as a means of facilitating information processing and judgement, and can lead to both valid or erroneous conclusions (Kahneman, Slovic, & Tversky, 1982). Several heuristics and affect reactions have been identified as influencing how patients construct their breast cancer risk perceptions including the availability, simulation, representativeness, affect, and perceived control heuristics, and combinations of these (Facione, 2002; Katapodi et al., 2004; Montgomery et al., 2003; Rees, Fry, & Cull, 2001).

Dual process theories, accounting for how decisions are influenced by implicit or automatic processes as well as by explicit or conscious processes, may be relevant to understand how the experiences and attitudes of women at increased risk of breast cancer shape their preference for and uptake of specific risk management options.

Petty and Cacioppo's Elaboration Likelihood Model (ELM) (Petty & Cacioppo, 1986) is a dual process theory that has been used to understand behaviours that will reduce cancer risk, such as compliance with breast cancer screening (Drossaert, Boer, & Seydel, 1996). It proposes two routes of processing stimuli, a central route and a peripheral one, with individuals being more likely to process information via one or the other depending on their motivations and their ability for critical evaluation. According to the ELM model, information perceived to be personally relevant, and therefore affecting a person's motivation, is more likely to be processed via the central route. Information processed this

way is retained longer and is more likely to lead to attitudinal change that will be enduring and predictive of behaviours (Petty & Cacioppo, 1986). Oppositely, individuals are more likely to engage in peripheral information processing if they lack motivation or capability to think critically about a topic; this might be due to their attitudes, perceived personal relevance, cognitive resources, and might rely on heuristics and social influence. When information is processed via the peripheral route, individuals are more likely to maintain their attitudes or engage in attitudinal and behavioural change which is less durable.

The aim of this review was to present a systematic synthesis of the existing quantitative and qualitative research data examining women's views of RRM when they are considering breast cancer risk management strategies due to being at familial high risk of developing breast cancer. Familial high risk of breast cancer is defined as an identified BRCA1/2 mutation or known increased familial risk due to a family history consistent with an inherited susceptibility to breast cancer. In particular, this mixed-method systematic review examines the questions:

- What influences the RRM decisions of women who are at familial high risk of breast cancer when they are considering at least one other risk management alternative?
- What are the predictors of their RRM decisions?

Method

In this mixed-method systematic review data from qualitative and quantitative published research findings are analysed and synthesised to provide a better understanding of individuals' experiences and views (Heyvaert, Hannes, Maes, & Onghena., 2013).

Search Strategy

Search terms were initially identified consulting literature reviews and articles published in peer-reviewed journals. Further search terms were incorporated after scoping searches were conducted using electronic databases. The electronic databases PsycINFO (1879-2017), Medline (1948-2017), Web of Science (1900-2017), and Scopus (1960-2017) were searched for relevant published literature using the following keywords in combination: (“Risk reducing mastectom*” OR RRM OR “prophylactic mastectom*” OR “preventative mastectom*”) AND (Perception* OR attitude* OR opinion* OR view* OR belief* OR perspective*) AND (Wom* OR patient* OR survivor*). The search strategies were based on the keywords being in titles, abstracts and keywords.

Searches were combined and duplicates were removed prior to study selection using Endnote X7. In addition, hand searches were also conducted for references from previous systematic reviews, as well as publications by key authors in the field, and the reference lists of identified publications. Search alerts were set up via the abovementioned electronic databases until May 2017, which generated weekly emails updating the search results with newly published studies.

Inclusion and Exclusion Criteria

Following the search strategy, identified publications were assessed for inclusion in this review. Studies with participants identified as at high familial risk of breast cancer, and studies where participants were BRCA1 and/or BRCA2 mutation carriers, were included. Studies that investigated participants’ views of two or more breast cancer risk management options, one of which was RRM or PM, were included.

Studies were limited to those that included adult women participant samples. Studies that investigated the opinions of health care professionals or family members, were excluded,

as were studies with participants were not identified as at increased familial risk of breast cancer. Studies that were published in English, Spanish or Portuguese languages were included. No restrictions on the date of publication were applied. For quality control, only studies published in peer-reviewed journals were included. Literature reviews and conference presentations, as well as case reports and unpublished dissertations were excluded.

Study Selection

As an initial stage, ESN screened all identified titles and abstracts of the search results for potential inclusion. Subsequently, the studies identified as potentially relevant were read by ESN in order to ascertain whether they met inclusion criteria. When it was unclear whether or not the studies should be included, a second opinion was sought (SB) and a joint decision reached.

Reference lists and exclusion criteria applied were recorded at each stage (see Appendix B). The process of study selection is presented using the Preferred Reporting Items for Systematic reviews and Meta-Analysis ([PRISMA] Moher, Liberati, Tetzlaff, & Altman, 2009) flow diagram in Figure 1. This review included 13 studies, of which ten were quantitative and three were qualitative.

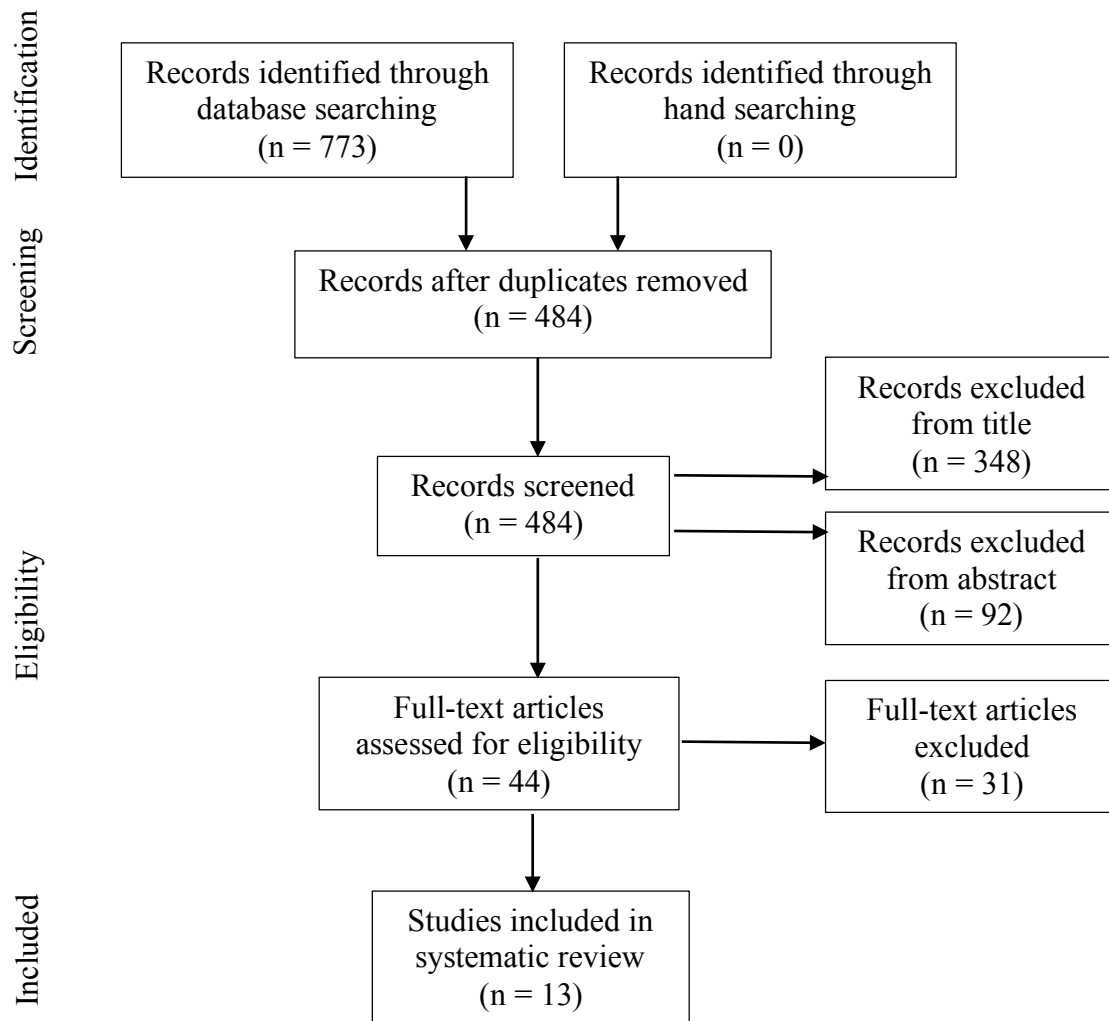


Figure 1. PRISMA flow diagram displaying the article selection process.

Quality Assessment

Quality was not an exclusion criterion, however the Quality Assessment Tool for Studies with Diverse Designs ([QATSDD] Sirriyeh, Lawton, Gardner, & Armitage, 2012) was employed to assess study quality. The 16-item QATSDD (Appendix C) has shown good reliability and validity for use in the quality assessment of studies (Sirriyeh et al., 2012) and can facilitate in-depth understanding of studies under review, including their strengths and limitations (Fenton, Lauckner, & Gilbert, 2015). The quality assessment procedure was performed by ESN and reviewed by SB. Where disagreement occurred, it was resolved by

both authors reaching a consensus. The strengths and weaknesses in study methodology were considered when synthesising the studies, and thus the final synthesis was weighted by study quality. Appendix C presents the QATSDD scoring for each study.

Results

Summary of Included Studies

An overall summary of the characteristics of the studies included in this review is presented in Table 2. The 13 eligible studies took place in nine countries: USA (n=5), UK (n=1), Canada (n=1), Italy (n=1), Belgium (n=1), Israel (n=1), Hong Kong (n=1), Netherlands (n=1), and Australia (n=1). Three studies used qualitative methods, whereas the most frequent methodological design was quantitative (n=10).

The most common quantitative data collection method was self-administered questionnaires (n=10). The sample sizes in the studies ranged from 371 to 12. A total of ten studies reported participants were identified as at high risk of breast cancer due to being BRCA1/2 mutation carriers; in five of these studies the BRCA1/2 mutation carriers constituted a subsample of participants, as the studies also included participants who were not BRCA1/2 mutation carriers. In three studies the participants were women identified as at high risk of breast cancer due to familial risk.

Results of Quantitative Studies

This review included ten studies using quantitative methodologies with varied aims, with data primarily collected by means of self-administered questionnaires using a wide range of measures, as recorded in Table 1. These studies varied also in their sample sizes (from n=62 to n=312), as well as demographic characteristics such as age range, ethnicity and BRCA

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Table 1.

Summary of Quantitative Studies

Author(s), year	Country	Sample	Sample characteristics of interest	Design	Aim(s)	Data collection method	Measures and data collected	Predictors	Summary of results of interest
Borreani et al. (2014)	Italy	C-A n=52 C-UN n=27 C-A age range: 30-70. C-UN age range: 26-57 Ethnicity: not stated.	BRCA1/2 mutation carriers, who were either cancer-affected (C-A) or cancer-unaffected (C-UN).	Prospective	Describe the impact of risk-reducing options on psychological condition of BRCA1/2 carriers. Describe the distribution of preventive strategies. Compare psychosocial variables of women selecting surveillance or risk-reducing surgery.	Self-report measures.	HADS, Breast Cancer Worry Scale, Cancer-risk perception tool, Satisfaction with risk reducing strategy questions, Digital Body Photo Test, MOS SF-12.	Psychological distress, Concerns about becoming ill with BC, Cancer-risk perception, Body image, Sociodemographic information, Oncological family history	In BRCA1/2 mutation carriers, prophylactic surgery reduced the perceived risk and worry about cancer. C-A and C-UN mutation carriers have to be considered as two separate populations.
Claes et al. (2005)	Belgium	n=68 (34 BRCA carriers, 34 non-carriers) Carriers age mean: 38.4 Carriers age range: 19 – 61	Subsample of BRCA1/2 mutation carriers.	Prospective	Describe breast and ovarian cancer surveillance practices and prophylactic surgery. Evaluate the effect of BRCA carrier status on cancer screening practices. Provide insight into factors influencing women's decisions about risk management strategies.	Semi-structured interview, Self-report questionnaires.	Sociodemographic data, disease data, health-related behaviour, uptake of risk-reducing surgery.	Sociodemographic variables, Disease-related variables, Health-related behaviour, Illness perceptions, Cancer-specific distress, Attitudes and motives towards preventive options	9% of BRCA mutation carriers underwent RRM. BRCA carriers adhered to cancer surveillance recommendations (clinical breast examination and mammography). 75% of BRCA mutation carriers eligible for RRO had this procedure.
Haroun et al. (2011)	Canada	n = 246. Age range: 26 – 66. Ethnicity: not stated.	BRCA1/2 mutation carriers without breast cancer.	Retrospective, Cross-sectional	Identify factors associated with a switch from MRI-based surveillance to RRM. Measure satisfaction with the decision.	Self-administered questionnaire developed by researchers.	Demographic data, family history of BC, satisfaction with screening, estimations of personal risk of BC, reasons to undergo RRM and satisfaction with it.	BRCA 1 or BRCA 2, First-degree relative having BC, Perceived personal BC risk, Previous BSO, Benign breast biopsy, Satisfaction with MRI screening	The women who elected RRM instead of MRI surveillance cited fear of cancer, previous cancer and concern about children as main reasons. Women who perceived BC risk to be greater than 50% were more likely to opt for RRM.

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Kram, Peretz & Sagi (2006)	Israel	n = 99 (43% BRCA mutation carriers, 57% non-carriers). Mean age: 53. Ethnicity: Ashkenazi Jews.	Subsample of BRCA mutation carriers (43%).	Retrospective, Cross-sectional	Assess the effect of genetic test results on decisions about risk-reducing surgery. Evaluate the motivating factors for decisions. Evaluate satisfaction with their decision to undergo risk-reducing surgery.	Self-administered questionnaire constructed for research.	Items about demographic data, family history, genetic test, reasons to opt for or reject RRM/RRO, attitudes about these procedures.	Genetic test results (BRCA mutation carriers v non-carriers), Demographic characteristics, Beliefs in the efficacy of surveillance strategies.	94% of BRCA carriers considered positively the option of RRO. 78% underwent this surgery. 25% of BRCA carriers considered positively the option of RRM. 19% underwent this surgery. RRO was more acceptable than RRM from an attitudinal and practical aspect.
Lerman et al. (2000)	USA	n = 216 Age: 34% 25-39 years. 66% 40 years or older. Ethnicity: Caucasian.	Subsample of BRCA mutation carriers (n=84).	Prospective	Examine prophylactic surgery and surveillance behavior one year following BRCA1/2 gene testing.	Structured telephone interviews.	Sociodemographic variables, clinical variables, Intrusion subscale of IES.	Sociodemographic variables, Clinical variables, Cancer-specific distress.	3% of unaffected BRCA mutation carriers underwent RRM. BRCA mutation carriers had significantly higher rates of mammography.
Litton et al. (2009)	USA	Total n = 312 (BC: n = 217. BRCA: n = 86). Age: not stated. BRCA carriers' ethnicity: White=75, Hispanic=7, Black=1, Asian=2, Other=1.	Subsample of BRCA mutation carriers.	Cross-sectional	Evaluate opinions about screening and surgical strategies among women at high-risk of BC.	Postal survey developed by researchers.	12 questions regarding perceptions of genetic testing, screening and risk reduction options.	BRCA mutation status, BC, Demographic characteristics, Views about preventive options	BRCA mutation carriers were more likely to believe RRM was the best way to reduce risk and worry of BC, and more likely to proceed with this intervention.
Lodder et al. (2002)	Netherlands	n = 63 (BRCA: n = 26) Age mean: BRCA+RRM=35.4 BRCA+surveillance=42.3 Non-BRCA=37.4 Ethnicity: not stated.	Subsample of BRCA mutation carriers undergoing RRM (n=14) or surveillance (n=12).	Prospective	Explore decisions of BRCA1/2 mutation carriers to undergo prophylactic surgery or regular surveillance. Examine their course of distress, body image, and sexuality.	Interviews and questionnaires.	LOT, HADS, Symptom Checklist, IES, Body image and sexuality questionnaire.	Biographical and pedigree information, Experiences with cancer in relatives, Optimism, General distress, Cancer-related distress, Body image and sexuality	BRCA mutation carriers who opted for prophylactic surgery had significantly higher distress than those who opted for surveillance. They were more often in their 30s, more often had young children, and had a longer awareness of their familial cancer risk.

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Meiser et al. (2003)	Australia	n = 371 Age median: 42. Age range: 19 – 88. Ethnicity: not stated.	Women unaffected by cancer, with unknown mutation status, from families with inherited susceptibility to BC.	Retrospective, Cross-sectional	Assessing women's attitudes towards and intention to undergo prophylactic mastectomy, prophylactic oophorectomy, or chemoprevention using tamoxifen.	Self-administered questionnaire.	Demographic characteristics, Objective BC risk, LOT, HADS, IES.	Intention to undergo RRM, Intention to undergo RRO, Intention to take tamoxifen	16% of women reported considering RRM; 33% had considered RRO; 23% reported considering tamoxifen.
O'Neill et al. (2010)	USA	Total n = 308 (RRM: n=308. RRO: n = 276). Age range: 25 – 75. Ethnicity (RRM): 53% Jewish, 47% non-Jewish. Ethnicity (RRO): 49% Jewish, 51% non-Jewish.	Women with BRCA1/2 mutation or with BRCA1/2 uninformative status.	Prospective	Examine the effect of BRCA genetic test result on perceived pros and cons of RRM, RRO and BC screening.	Telephone interview.	Sociodemographics, family cancer history, psychosocial variables, and attitudes toward risk management options (pros/cons).	Sociodemographics, Medical/Family history, Pros/Cons of risk management options, Intentions for RRM and RRO, Uptake of RRM and RRO	BRCA1/2 mutation status predicted stronger pros for RRM and RRO. Pros of surgery predicted RRM and RRO intentions in BRCA mutation carriers.
Ray et al. (2005)	USA	n=62 Age mean: 51 Age range: 25 – 80 Ethnicity: not stated.	Women at high-risk of BC who received genetic counselling, of which 37% did not have BRCA testing.	Prospective	Describe uptake of RRM or RRO in women at high risk for BC and ovarian cancer. Investigate if BRCA mutation status influences decision to undergo risk reducing surgery.	Postal self-report measure developed for the study.	Demographic and disease characteristics, Risk-reduction surgery intent, Cancer Screening and Prevention Checklist.	Demographic and disease characteristics, Risk-reducing surgery intent, Uptake of risk-reduction surgery and surgery decisions.	BRCA test result was most important influence for risk-reducing surgery decisions. Reasons for indecision included genetic testing results, concerns about surgery, timing in life, and premature menopause.

Note. USA = United States of America; n = number of participants; C-A = cancer-affected; C-UN = cancer-unaffected; BC = breast cancer; BRCA = breast cancer gene status; RRM = risk reducing mastectomy; CRRM = contralateral risk reducing mastectomy; RRO = risk reducing oophorectomy; MRI = magnetic resonance imaging; BSO = bilateral salpingo-oophorectomy; HADS = Hospital Anxiety and Depression Scale; MOS SF-12 = Medical Outcomes Study Short Form; IES = Impact of Events Scale; LOT = Life Orientation Test.

mutation status of participants, and the preventive options considered and compared by participants (RRM, RRO, chemoprevention and/or surveillance). It is notable that only one study (Ray et al., 2005) used an established theoretical model, the Precaution Adoption Process model, to guide their investigation. This highlights an important shortcoming of the overall body of research in this area.

The breast cancer risk management alternatives studied included, in addition to RRM, surveillance or screening (Borreani et al., 2014; Claes et al., 2005; Haroun et al., 2011; Lerman et al., 2000; Litton et al., 2009; Lodder et al., 2002; O'Neill et al., 2010; Ray et al., 2005) and chemoprevention using tamoxifen (Meiser et al., 2003). All studies, with the exception of Haroun et al., (2011) investigated breast and ovarian cancer preventive alternatives such as RRO and RRSO, as the BRCA positive samples were at increased high risk for both types of cancer.

Two studies reported that the low rates of RRM uptake among participants at high risk of breast cancer impeded an analysis of predictive factors (Claes et al., 2005; Lerman et al., 2000). Participants in these studies expressed a preference for or engaged in other risk management options, including surveillance practices or RRO.

Predictors of decision to undergo a RRM. A number of possible predictors have been explored and reported by the studies included in this review with varied frequencies, such as educational level (Kram, Meiser), age (O'Neill, Meiser), BRCA mutation carrier status (Kram, O'Neill), or cancer-related distress (Lodder, Meiser).

Investigating reported intent to undergo risk-reducing surgery and actual uptake, Ray et al. (2005) found that there was no statistically significant correlation between these two factors with regards to RRM. In a high-quality Australian large-sample study, Meiser et al. (2003) reported no association was found between women's perceived risk of BC and their consideration of RRM, despite it being predictive of women's intentions to take tamoxifen

and to undergo RRO. One plausible explanation offered by the authors was that body image preoccupations might represent a stronger predictor of RRM intentions than perceived risk. Furthermore, in this study of factors influencing intention to undergo RRM, there were no associations between women's intentions and psychological or family history variables considered, which in addition to perceived risk, included age, educational level, breast/ovarian cancer distress, optimism, and number of first degree relatives with breast cancer.

Other research has yielded differing results, with BRCA mutation status appearing to be a consistent predictor of RRM decisions. A small subsample of BRCA carriers chose to undergo a RRM in the study conducted by Kram et al. (2011). There were no differences in educational level nor in motherhood to young children between women who underwent the procedure and those who did not, however finding out about one's BRCA1/2 mutation carrier status impacted on women's decision-making; among BRCA1/2 mutation carriers there was a significant increase in women who considered RRM after receiving their genetic test result.

Similarly, in their study examining the perceived pros and cons of RRM, RRO and breast cancer screening, O'Neill et al. (2010) found that, compared to participants with an uninformative BRCA1/2 mutation testing results, women who tested positive reported increased perceived pros for RRM, whereas their perceived cons remained unchanged. The pros in this study corresponded with participants' perceptions of the risk-reducing benefits of surgery, such as reduction of the risk of developing breast cancer, of dying from breast cancer worry about breast cancer, worry about how their families would be affected, and physician recommendations. The cons pertained primarily to concerns about the risk of surgery, long recovery from the surgery, and changes in physical appearance. Furthermore, this study found that stronger RRM pros and weaker cons predicted RRM intentions after women received their genetic test result, but only for BRCA1/2 mutation carriers. Age was found to

also be a factor, as was ethnic origin. In this study Ashkenazi Jewish women were significantly less likely to report considering RRM as a breast cancer risk management option. Participants under the age of 50 reported stronger pros and cons and weaker intentions for all BC risk management alternatives.

More elevated anxiety and cancer-related distress were found in women with a BRCA1/2 mutation status opting for a RRM than in those who were BRCA1/2 mutation carriers opting for surveillance and in those who were non-carriers, both before and up to one year after receiving the genetic test result (Lodder et al., 2002). One study, albeit of lower methodological quality, differentiated between participants who had been affected by cancer and those who had not, reporting that surgical options were taken up more often by cancer-affected women (Borreani et al., 2014). This group of participants did not report significant cancer worry reductions after the surgery, in contrast to cancer unaffected women and despite the data showing a significant decrease in their risk perception. The authors have suggested that perceived risk of reoccurrence might be linked to perceived personal risk of breast cancer. Haroun et al. (2011) reported that women who perceived this risk to be greater than 50% were significantly more likely to opt for RRM instead of MRI surveillance than women who perceived their risk to be lower.

Women's reasons for deciding to undergo a RRM. A number of studies, both prospective and retrospective, investigated the reasons participants ascribed to their RRM uptake decisions. Genetic testing information, and particularly genetic test results regarding BRCA mutation status, was cited as the most influential reason in the decision to have risk-reducing surgery, followed by information about cancer risk (Ray et al., 2005).

A significant proportion of BRCA1/2 mutation carriers in Litton et al. (2009) felt that they did not find mammograms to be an uncomfortable procedure, yet, compared to non-carriers, they considered that RRM was the most effective way of reducing the risk of

developing breast cancer, and the only way to reduce worry of getting breast cancer. Despite their willingness to undergo regular mammograms and perform self-directed breast examinations, the majority of women who expressed those viewpoints opted for a RRM.

Similarly, investigating factors related to women's switch from MRI-based surveillance to RRM, Haroun et al. (2011) found that dissatisfaction with the screening programme was not a motivating factor; rather, women provided other reasons, most commonly fear of cancer, fear of dying and of leaving their children behind, and wanting to avoid the experience of battling with cancer after their relative(s) had suffered this.

Women's reasons for indecision about RRM and for choosing to not undergo a RRM. In five studies of comparable good quality (Claes et al., 2005; Haroun et al., 2011; Kram et al., 2006; Lodder et al., 2002; Ray et al., 2005) the reasons for women not opting to have RRM were investigated. Women who were indecisive about risk-reducing surgery in Ray et al. (2005) cited reasons summarised in the themes of genetic testing factors (e.g. family history versus test results), concern about surgery as a procedure (e.g. fear of having surgery, major decision in life, surgery is irreversible, body alterations from surgery), timing not being right (e.g. childrearing, finishing chemotherapy, young age). The main disadvantages of RRM indicated by women who opted for breast cancer surveillance in the year after their genetic test results, were the 'negative impact on physical appearance/functioning (e.g. scars, mutilation, recovery period), 'body image' and 'negative emotional impact' (Claes et al., 2005).

Concerns about body image and fear of surgery were also reported by women who opted for MRI-based surveillance instead of RRM in the study by Haroun et al. (2011). However, they cited their satisfaction with and confidence in screening as their main reason for following the MRI surveillance programme and not undergoing RRM. Similarly, in a sample of Israeli women, the belief in the efficacy of surveillance for detecting a tumour at an

early stage was negatively correlated with their consideration and uptake of both breast and ovarian risk-reducing surgery (Kram et al., 2006). Additional reasons provided for deciding against RRM were the impact of the surgical procedure on femininity, self-image, and body-image. Similarly, participants in Lodder et al. (2002) who opted for breast surveillance instead of RRM considered they felt sufficiently safe with the surveillance programme, whereas others perceived that they either would never opt for an intervention as drastic as RRM or that they still had time to reflect on whether they wanted it in the future.

The findings reported by these studies with regards to women's reasons for deciding to undergo RRM or choosing different risk-management alternatives are echoed in the results presented by the qualitative studies included in this literature review. Common themes from the qualitative and quantitative studies include the benefits and costs of RRM as appraised by participants including body image preoccupations, the acceptability of the surgical procedure, perceptions of breast cancer risk, and cancer-related anxiety and distress.

Results of Qualitative Studies

This review included three studies which employed different qualitative methodologies (content analysis, thematic analysis, and grounded theory) and varied in their aims (exploring the impact of RRM, the meaning of the surgery, and participants' conceptualisations of risk and risk prevention options), the sample characteristics such as ethnicity and age, and the risk management options considered and compared by participants (RRM only, or RRM and RRO) (see Table 2), but which were of comparable good quality. In their cross-sectional studies, Hallowell (1998) and Salant et al. (2006) conducted interviews with women in the UK and in the USA respectively who were considering their high risk of breast cancer and the preventive options available to them. Kwon and Chu (2012) conducted their retrospective study with a sample of Chinese women who had received a contralateral RRM.

Table 2.

Summary of Qualitative Studies

Author(s), year	Country	Sample	Sample characteristics of interest	Design	Aim(s)	Data collection method	Methodology	Themes	Summary of results of interest
Hallowell (1998)	UK	n = 41 Age mean: 40. Age range: 22 – 59. Ethnicity: not stated. BRCA: not stated.	Women with a familial high risk of breast or ovarian cancer, who did not have personal history of cancer.	Cross- sectional	Provide insight into the meaning of prophylactic surgery as a risk management strategy for women with a familial risk of BC and OC.	Semi-structured interviews.	Content Analysis	Perceived benefits of prophylactic surgery Perceived costs of prophylactic surgery.	Prophylactic surgery provided women the means to fulfil their obligations to family members and to reduce and contain their fear of cancer. RRO was more acceptable than RRM, influenced by the different physical and psychosocial implications.
Kwong & Chu (2012)	Hong Kong	n = 12 Age mean: 47 Age range: 34 – 55. Ethnicity: Asian (Southern Chinese).	11 BRCA1/2 mutation carriers and 1 non- carrier. All had history of BC and had elected CRRM.	Retrospective	Explore the experience and impact of CRRM from Chinese females' subjective perspectives.	Face-to-face or telephone interview.	Thematic Analysis	Stage 1: Decision-Making Stage 2: Immediate impact of CPM Stage 3: Chronic impact of CPM.	All participants opted for CRRM due to reservations about efficacy of surveillance and worries of BC risk.
Salant et al. (2006)	USA	n = 33 Age mean: 55 Age range: 33 – 77 Ethnicity: 73% African American, 18% White, 9% Other (Hispanic, Asian).	Women without personal history of BC, at high risk of BC and/or moderate to high risk of carrying a BRCA mutation.	Cross- sectional	Examine women's conceptualisations of BC risk and views about preventive options available to them.	Semi-structured interviews.	Grounded Theory	Breast cancer risk perceptions: personal risk perceptions; meaning of “high risk”; causes of breast cancer. Breast cancer prevention: prevention choices and attitudes; gene testing.	BC screening was welcomed. Preventive options were perceived to cause problems and were only acceptable as treatment options for a disease.

Note. UK = United Kingdom; USA = United States of America; n = number of participants; BC = breast cancer; BRCA = breast cancer gene status; RRM = risk reducing mastectomy; CRRM = contralateral risk reducing mastectomy; CPM = contralateral prophylactic mastectomy; RRO = risk reducing oophorectomy.

Meaning of ‘at high risk’ and breast cancer risk perceptions. Individuals considered as ‘at high risk’ perceive their health status in diverse ways (Hallowell et al., 1998). These perceptions might be dependent on physical and emotional states. Salant et al.’s study (2006) highlighted that many women, despite their awareness of their hereditary risk, did not consider their breast cancer risk status as static or as an objective medical description, but rather as fluctuating. In particular, experiencing physical symptoms made the risk of breast cancer more salient to them. This frequently led women to misestimate risk. All participants of Kwong and Chu’s (2012) study expressed the view that the CRRM had decreased their risk of future breast cancer, but they overestimated this risk even after the procedure. The different interpretations of both the designation of ‘at high risk’ and of changes in perceived risk may, in turn, influence women’s perceptions of risk management alternatives.

Relief from fear, worry and risk. RRM afforded women relief from their breast cancer worry and fear (Hallowell, 1998; Kwong & Chu, 2012). As expressed by a participant who had underwent CPM: *“I can finally stop worrying whether the doctor would find a lump again and start living a normal life!”* (Kwong & Chu, 2012, p.2245). In contrast, for the participants in Salant et al.’s (2006) study, the relief of fear or worry was not sought from risk-reducing nor early detection strategies. Instead, participants’ understanding of risk as a physical symptom requiring medical intervention and their idiosyncratic breast cancer causality beliefs were linked to cognitive avoidance strategies, such as *“not dwelling on it”* and *“not thinking about it”*. Coping with the fear or threat of cancer was achieved by means of prayer, spiritual healing and spiritual statements of acceptance, e.g. *“what happens, happens”*.

Relationships and self. The influence and importance of women’s relationships with other people and of women’s self-perceptions are salient in two of the studies (Hallowell,

1998; Kwong & Chu, 2012). Women considering RRM or RRO (Hallowell, 1998) referred to these options in terms of their ‘benefits’ and ‘costs’ pertaining to the impact they would have upon their relationships. For example, surgery was seen by some participants as enabling them to fulfil their duties of care towards their children and families, whereas other women viewed surgery as an obstacle to being able to fulfil their social, work, and familial roles. The influence of their relationships with others in the case of Kwong and Chu’s (2012) cohort of participants extended to the decision-making for some women, who involved their families in their decision.

Issues of personal and interpersonal identities were also linked to women’s views of the acceptability of the surgical preventive options. RRM was seen as altering their body image to a greater extent than RRO and was therefore less acceptable in Hallowell’s study (1998), due to the body changes being potentially more visible and breasts being fundamental to their body image. As one 25-year-old women expressed: *“Your boobs you can see, and if my ovaries are still there or not makes no difference, because nobody else can see them”* (p.270). For the participants interviewed by Kwong and Chu’s research (2012), cosmetic concerns and impact on sexuality were not identified as important factors in their CPM decision-making. However, following the procedure, those who were in relationships were sensitive to their spouses’ reactions and attitudes, and whether the CPM affected their sexual relationships. This echoes findings in Hallowell’s (1998) paper, as some participants had expressed worry that having a RRM would impact their sexual relationships by making them appear less attractive.

Differences between risk-reducing options. Women held different views and attitudes about the risk-reducing options available to them and/or which they chose to engage in. Some women were not interested or were uncertain about taking medication such as tamoxifen, citing reasons such as reluctance to take pills, fearing harmful side effects, or the

lack of guarantee that it can inhibit breast cancer (Salant et al., 2006). Participants in Kwong and Chu's (2012) study reported that mammographic surveillance was not as effective in relieving the fear and worry that was induced by their BRCA1/2 mutation status; despite being a non-invasive option, it was perceived as a stressful recurrent reminder of their heightened risk of breast cancer: "*Surveillance was neither inconvenient nor painful but it was a periodic reminder that I was carrying a cancer bomb... physically surveillance may seem non-invasive but psychological it was difficult (to deal with)*" [sic] (p.2243). The authors highlighted that participants' prior experience of breast cancer treatment might have influenced their "pleas for a more efficacious treatment, if not closure" (p.2245). Women considering the options of RRM or RRO in Hallowell's study (1998) regarded the latter as a more acceptable alternative due to the different psychosocial and physical implications of the procedure: RRO was seen as precipitating changes which would be less visible and which would occur naturally if no surgery was had (i.e. menopause), and ovaries were not seen by participants as constituting a public representation of their femininity.

Discussion

The aim of this paper was to review the existing literature examining women's views of RRM when they are considering breast cancer risk management alternatives due to being at high risk of developing this illness. Additionally, this mixed-methods systematic review examined what are the predictors of women's RRM decisions and how they make decisions about this surgery when they are considering at least one other risk management alternative.

Overall, the findings suggest a range of factors might be relevant to, and predictive of, women's RRM uptake decisions when they are considering several breast cancer risk-reduction or early detection options. These factors include perceived risk of breast cancer or of recurrence of breast cancer, BRCA1/2 mutation status, cancer-related worry or fear, and

satisfaction with and confidence in other methods. Framed within relevant models for health prevention decision-making, the findings of this review are in line with, and advance, those reported by Howard, Balneaves, & Botorff (2009) and by Fielden et al.'s (2017) synthesis of qualitative research. The predictive value of demographic variables such as age, educational level, or ethnicity have not been adequately explored due to heterogeneity of samples and low rates of RRM uptake in several studies.

Genetic testing and, more specifically, the result of BRCA1/2 mutation status, was highlighted in some studies as the most influential factor in women's decisions to undergo risk-reducing surgery, which has been reported in other studies (Scheuer et al., 2002). Additionally, high levels of cancer-related worry, fear, or distress in women opting for RRM have been observed in previous research (Meiser et al., 2000; Stefanek, Helzlsouer, Wilcox, & Houn, 1995), as has been a longstanding awareness of being at increased risk, which decrease significantly after they undergo RRM.

Differences in people's attitudes and preferences for breast cancer prophylactic surgery or other risk-management measures have been explained by cultural differences, both for women and health care professionals (Julian-Reynier et al., 2001). The findings of individual studies included this review have not emphasized cultural or social differences as predictors or influences of women's RRM decisions. However, among the included studies there appear to be variations in participants' preferences and attitudes towards breast cancer risk management options. In line with what has been highlighted by recent research (Wainberg & Husted, 2004), it is possible that these differences could also be attributed to the variations of cultural norms, to the differences in breast cancer counselling protocols between countries, and to variation in the availability of risk-reducing options (Metcalf et al., 2008); the effects of these cultural differences on people's views of RRM are yet to be fully understood.

Additionally, women's views and attitudes about RRM relevant to their decisions when they are considering more than one breast cancer risk-reduction option are reflected on the reasons why they would or would not opt for this strategy. The findings suggest that both perceived effects on body image and sexuality, and perceived confidence in other early detection or other preventive methods were associated with women's preferences. Due to this review including studies of women considering RRM and at least one other risk management or early detection alternative, it has been possible to gain some insight into the comparisons made between these. Other possible factors, such as fear of undergoing a surgical procedure, remain to be adequately investigated in future research.

With regards to body image and sexuality, some studies proposed these factors as influencing women's views of RRM and its acceptability. There is evidence that this type of prophylactic surgery can impact individuals' body image and intimate relationships, although often these issues are not considered a priority nor addressed before the surgery; negative effects on body image and sexual or marital problems are common difficulties after the surgery (Bebbington & Fallowfield, 2003; McGaughey, 2006). With regards to confidence in other preventive or early detection measures, it could be hypothesised that women may not opt for RRM if they perceive surveillance, chemoprevention and/or RRSO as effective measures which address their perceived risk of breast cancer and the psychological consequences of being at high risk of this illness, such as fear, worry or distress.

A range of theoretical models have emphasized the role and relevance of cognitive variables in the uptake of health-related behaviours, including preventive options among individuals at increased risk of breast cancer (Cohen, 2002; Decruyenaere, Evers-Kiebooms, Welkenhuysen, Denayer, & Claes, 2000). In this review of the literature, only one study (Ray et al., 2005) applied an explicit theoretical model, namely the Precaution Adoption Process (Weinstein & Sandman, 2002), according to which the uptake or risk-reduction

surgery requires deliberate action in seven distinct stages. Other models and heuristics could have informed the studies and appear to be relevant to the findings, yet have not been applied in this specific area of research to date. For example, when considering women's views of RRM, it could be hypothesised that from the perspective of the ELM, individuals might process highly emotive, personally relevant, and complex cancer-related information via the peripheral route, thus being likely to rely on social influence and heuristics. Research indicates that breast cancer risk perceptions are influenced by several types of heuristics (Facione, 2002; Katapodi et al., 2004; Rees et al., 2001). Therefore, consideration should also be given to whether and how people's views on, and decisions about, RRM are similarly constructed relying on heuristics. It could be posited that, in addition to breast cancer risk perception, other possible RRM uptake predictors such as cancer-related distress or impact on physical appearance might be influenced by heuristics such as availability, simulation, or affect heuristics. Future investigations addressing this gap in understanding should be considered.

A study quality appraisal was conducted using a well-developed instrument (QATSDD; Sirriyeh et al., 2012). This quality assessment pointed to inconsistencies in information reporting across studies with regards to study design, sample characteristics, data collection, or psychometric properties of the measures used (Appendix C). Using this instrument, it was possible to conclude that the majority of the studies were considered to be of adequate quality but with notable shortcomings associated with study designs. This, in conjunction to the small number of studies included, demonstrates that this area of research is currently understudied, highlighting the important need to conduct further research of greater quality in order to develop the knowledgebase.

The main research designs used across studies, namely prospective studies investigating RRM uptake and retrospective reports about RRM decision-making, are not

without limitations, such as requiring long durations for follow-up (Song & Chung, 2010) or be subject to recall or hindsight biases (Guilbaut et al., 2004), to name a few. Longitudinal designs spanned a maximum of 15 months after BRCA1/2 test results were disclosed, therefore it would be of interest to conduct longer studies to explore women's views of RRM and their decisions regarding risk management options over time.

The great variability among the characteristics of study samples and numbers of participants limited the comparability across studies. The range of psychometric tools used to measure anxiety, mood, or cancer-specific distress, and the lack of information regarding the validity of these measures also limit comparability. In addition, the majority of studies relied on self-report measures, which are vulnerable to biases such as social desirability; therefore, it would be important that future studies utilise additional means of data collection and methodologies, including in-depth interviews or broader qualitative methods.

Furthermore, the majority of hypothesised predictors and factors influencing women's RRM decision-making were explored by only a small number of the included studies. Therefore, caution should be exercised in the interpretation of results and with regards to the conclusions here drawn. The possibility that views about RRM, intention to undergo RRM, and actual uptake of RRM could be better predicted by other factors or combinations of other factors should not be excluded. Thus, on the matters here investigated, the existing research offers valuable information but the results cannot be considered conclusive, for the reasons outlined above.

Strengths and Limitations

There are several limitations to this review. Firstly, the inclusion of studies which differed greatly with regards to design and participant samples, conducted in countries with notable cultural differences, makes it difficult to draw general conclusions. Secondly,

although the inclusion criteria incorporated studies published in languages other than English, some studies might have been missed due to the data search being conducted with English keywords under the premise that studies published in other languages often include a translated title, abstract and keywords. Thirdly, there might be a risk of reporting bias, since this review focused on articles published in peer-reviewed journals and, therefore, potentially relevant research such as grey literature has not been included.

Additionally, articles retrieved via the database searches were reviewed for inclusion by one researcher (ESN); therefore, there is a possible risk of bias in the screening and selection process. However, a research supervisor (SB) was consulted multiple times during this process of screening and selection.

The inclusion of both qualitative and quantitative research constitutes a strength of this review. Nevertheless, quantitative research with a priori hypotheses about how people think may have failed to capture in-depth personal experiences (Smith, Flowers & Larkin, 2009). This shortcoming might have not been adequately supplemented due to the low number of qualitative studies included.

Clinical and Research Implications

The findings of this review, despite indicating an overall paucity of conclusive evidence, are of preliminary relevance for clinicians working with women at increased risk of breast cancer who are contemplating risk-reducing surgery. The results presented in this literature review suggest that a range of personal, social and cultural factors might influence women's views of RRM and might predict their decisions to undergo RRM, as has also been evidenced elsewhere (British Psychological Society's Division of Clinical Psychology Faculty for Oncology and Palliative Care, 2017). This suggests that clinicians' understanding

of these variables, as well as of heuristically-led decision-making processes, might help improve patients' experiences of RRM deliberation.

Nevertheless, a lot remains to be understood about the optimal management of breast cancer risk for women at high risk of this illness, and continuous evaluation of the effectiveness and psychological impact of the available risk-reducing options is necessary (Bermejo-Perez, Marquez-Calderon, & Llanos-Mendez, 2007). Equally, numerous questions remain unanswered regarding the psychosocial variables that predict and influence women's RRM decisions, for the current evidence can be considered promising but emergent. It is, therefore, recognised that a more in-depth understanding of the predictors of RRM uptake and of women's views of this surgery is required in order for future findings to inform and to be embedded in breast cancer psychosocial care and decision-making support for women at increased risk of breast cancer.

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**Exploring Risk-Reducing Mastectomy Perceptions and Decision-Making
among Women at High Risk of Breast Cancer.²**

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² To be submitted to *European Journal of Cancer Care*. Word limit: 8,000 words excluding references, figures and tables.

Abstract

Risk-Reducing Mastectomy (RRM) is the surgical removal of breast tissue to reduce breast cancer risk. However, it is non-reversible and can be associated with iatrogenic effects. Existing evidence suggests that patients at high risk of breast cancer make RRM decisions driven by fear of the illness. This study aimed to gain a further understanding of patient's views about RRM and about how they reach their decision to undergo it. Using a qualitative research design, seven participants were interviewed. Results suggest that patients made their decision to have a RRM in an immediate, non-hesitant manner. They were not able to describe those decisional processes in detail, but engaged in post-decisional reviewing processes. Participants' views of RRM as the best option for them were linked to their motivation to avoid future illness and treatment, to the sense of relief and safety that their choice provided them, and to the perception of doing something proactive to gain control. These findings are considered in the context of decision-making theory, as they highlight that patients who are at high risk of breast cancer might reach RRM decisions heuristically, guided by emotion. Strengths and limitations of this study are considered, and further avenues of investigation are proposed.

Keywords: breast cancer, risk-reducing mastectomy, decision-making, qualitative research.

Introduction

Breast cancer (BC), which affects primarily females, has been the most common cancer in the United Kingdom (UK) since 1997, and it accounts for 31% of all new cases of cancer in women (Cancer Research UK, 2015). In 2015 a total of 46,083 cases of breast cancer were registered in England, 45,764 of which were for females (Office for National Statistics, 2017). Both the disease and its treatment can result in long-term physical and psychological consequences, including adverse impacts on quality of life, body image, self-perceptions of femininity, pain or fatigue (Campora et al., 1992; Pasacrete, 1997).

Prognostic improvements in the field of cancer care mean that, increasingly, it is possible to identify whether women are at heightened risk of breast cancer (Rosenthal et al., 2017). Family history of breast cancer confers increased risk; the risk of women with an affected first-degree relative is approximately twice than that of women with no first-degree relatives (Cancer Research UK, 2014). The risk increases with larger numbers of affected first-degree relatives, or relatives with the disease under the age of 50 (Cancer Research UK, 2014). Genetic risk is mainly mediated by mutations in the BRCA1, BRCA2, P53 and CHEK2 genes, which confer increased risk of developing breast and ovarian cancer (National Institute of Health and Clinical Excellence [NICE], 2013). In the UK genetic testing for breast cancer susceptibility is available for breast cancer patients based on family history of early onset of breast or ovarian cancer, and for relatives of patients with a known inherited genetic mutation (NICE, 2013).

Early detection and risk-reducing options are available for women who are identified as at increased familial risk of breast cancer; these include lifestyle changes such as weight loss or alcohol reduction and smoking cessation, screening with MRI and mammographic surveillance, chemoprevention with selective estrogen receptor modulators, and risk-reducing breast or ovarian surgery (NICE, 2013). Risk-reducing mastectomies (RRM) are

prophylactic options that involve the surgical removal of healthy breast tissue with the aim of reducing the risk of suffering breast cancer (NICE, 2013). RRM can be performed contralaterally (CRRM) in the opposite breast to one already removed during treatment mastectomy in patients who have experienced breast cancer, or bilaterally (BRRM) in both breasts. With this procedure a residual risk of developing breast cancer remains (Lopez & Porter, 1996; Willemsem, Kaas, Pertese, & Rutgers, 1998).

Recent studies have reported an increasing trend in the uptake of RRM both for cancer affected and unaffected women (Balch & Jacobs, 2009; Tracy, Rosenberg, Dominici, & Partridge, 2013). RRM reduces the risk of breast cancer occurrence for women at high risk (Evans et al., 2013; Metcalfe et al., 2014) and improves the life expectancy of women with genetic mutations (Lostumbo, Carbine, Wallace, Ezzo, & Dickersin, 2004). Despite CRRM reducing the incidence of contralateral breast cancer by 90% (Tuttle, Habermann, Grund, Moris, & Virnig, 2007), this is not associated with an increased survival benefit unless women have a genetic mutation (Lostumbo et al., 2004). Furthermore, RRM is irreversible and can have negative outcomes, such as post-operative complications, cosmesis and concerns about body image (Altschuler et al., 2008; Geiger et al., 2006). Whilst the majority of women report satisfaction with their decision to have a RRM, evidence suggests that between 14% and 19% of women might regret their decision to undertake a RRM (Frost et al., 2000; Frost et al., 2011).

Decision-Making Regarding RRM

Current practice and policy regarding medical decision-making emphasize the importance of respecting patients' preferences and of integrating these with clinical evidence in the process of shared decision-making (Broadstock & Michie, 2000; Coulter & Collins, 2011). This requires informed consent, whereby patients understand and evaluate

information about options, probabilities, and their preferences to reach decisions. However, patients do not necessarily deliberate about health care decisions having sufficiently understood clinical issues, risks or benefits (Mendick, Young, Holcombe, & Salmon., 2010). A recent study by Beesley, Holcombe, Brown and Salmon (2013) found that women's decisions to undergo RRM were driven by a sense of vulnerability and by their awareness of an existing risk. Indeed, evidence suggests an association between distress, worry and anxiety, and higher rates of RRM uptake (Antill et al., 2006; Madalinkska et al., 2007; van Dijk et al., 2003). Furthermore, Brown et al. (2017) reported that women's decisions to seek RRM were motivated by fear of breast cancer and by feeling that they needed to do all they could to prevent it. These findings are consistent with evidence that patients seeking RRM do not necessarily consider objective risks and benefits as a primary reason for doing so (Beesley et al., 2013).

Emotion plays an important role in decision-making and in decisional processes (Loewenstein, Weber, Hsee, & Welch, 2001; Slovic, Finucane, Peters, & MacGregor., 2004). Existing evidence strongly indicates that individuals' emotional interpretations of information influence how they make health decisions, for instance by means of heuristics facilitating their information processing and judgement (Kahneman, Slovic, & Tversky, 1982; Lerner & Keltner, 2001; Pravettoni, Gorini, Bonanni, Varonesi, 2013; Reyna, 2004). Incorporating both pre- and post-decisional processes, the Differentiation and Consolidation theory proposed by Svenson (Svenson, 1992) postulates that the aim of a decision process is to reach an outcome or select an alternative that is sufficiently superior in comparison to other solutions (differentiation). Following the decision, post-decisional processes work both to consolidate the chosen alternative and to minimise regret threatening the choice (consolidation). This model incorporates the effects of affect and emotion in human decision processes.

Existing research investigating RRM uptake has focused on women's risk perceptions and their decision-making processes. However, little is known about how women perceive and think about RRM procedures, nor about what it is about RRM that qualifies it as a convincing option despite its inherent risks and/or despite other risk-reducing or surveillance alternatives being available. Furthermore, as evidenced by existing findings, for some women there might be potential disadvantages to making this health-related decision in the ways described above. This study aims to explore women's views about RRM when making RRM decisions, their decision-making process, and the factors influencing this risk-reducing procedure becoming the chosen option for women who are at high risk of breast cancer. Research in this area has the potential to help both women at risk of breast cancer and professionals providing decision support for women facing RRM decisions, by encouraging a broader consideration of factors to aid the decision-making process.

Method

Sampling and Participants

Ethical approval for the study was obtained from the National Research Ethics Service (REC: 13/NW/0421), with recruitment taking place at the Liverpool Cancer Psychology Service. Participants were recruited among women who attended a specialist psychological consultation to assess their decision-making regarding RRM and had reached a decision to have a RRM; this included recruiting women post-surgery. Patients were excluded if they were under 18 years old, if they were not fluent in English, if they were unsure about RRM, or if their care team considered they may not be able to provide informed consent.

Two recruitment strategies were followed in parallel. Firstly, initial study information was provided by a clinical psychologist who had completed a psychological assessment for RRM with the patient, including an information sheet (Appendix D); those interested in

participating consented to their contact details to be shared with the researcher, whom subsequently contacted them by telephone to further explain the study. Secondly, in order to maximize recruitment, letters with the information sheet were posted to patients who had attended the service in the previous 18 months, inviting them to take part in the research (Appendix E). A total of 15 letters were sent, with four patients responding and agreeing to take part in the study (see Appendix F for participant consent forms). A sample of seven women was recruited; this sample reflected the range of women accessing psychological assessments related to RRM procedures, with variation in type of surgery (CRRM or BRRM), personal and familial history of breast cancer, genetic mutation status, and sociodemographic characteristics. Participants were interviewed by the researcher at a convenient date and location. Further study details are resented in Appendix G.

Data Collection

Semi-structured face-to-face interviews (median duration: 49 minutes; range 41 – 67 minutes) were conducted by ESN. Participants completed a brief demographic information sheet (Appendix H) in order to characterise the sample.

Interviews were conversational, with open ended questions, prompts and reflections to facilitate the exploration of women's views. The researcher adopted a non-directive, non-judgmental approach. The interviews were semi-structured following a topic guide, which was progressively refined as the analysis proceeded (Appendix I). The initial topic guide was developed drawing from previous findings from the wider study (Brown et al., 2017).

The study had a data protection protocol (Appendix J) and a protocol to be followed if women experienced distress (Appendix K); its use was not required. Participants had the option of receiving a summary of findings, with 6 women requesting this information (Appendix L). Interviews were digitally audio-recorded, transcribed verbatim, and

anonymised. The researcher transcribed the initial two interviews with the aim of becoming familiar with the data. A university approved transcriber was employed for the subsequent interviews, with the researchers providing clear guidelines (Appendix M) and checking the transcripts for accuracy.

Data Analysis

In order to engage critically and creatively with the data, and to overcome the limitations of flexibility associated with ‘branded’ methodological approaches, whilst maintaining creativity and rigour, a pragmatic data analysis approach was followed. Similar approaches have been advocated by qualitative researchers recently (Frost et al., 2011; Gwyn, 2002; Wright, Holcombe, & Salmon, 2004). Data were analysed using a pluralist qualitative methodology, whereby the researchers adopted a flexible stance to the methods and epistemological positions, as required by the data and by the emerging analysis (see Appendix L for further information).

Data analysis took place in parallel with data collection. As part of the iterative process, interview transcripts were read and re-read sequentially by the research team, and discussed in regular research meetings. Brief narratives of the salient features of every interview were developed to aid remembering and reflection (Appendix O). The aforementioned discussions informed the ongoing analysis and data collection. Findings were further refined and tested in the research meetings by ‘cycling’ between the emerging analysis and new data.

The iterative data analysis commenced at a descriptive level, as the research team developed and defined broad analytic categories illustrating recurring features of women’s accounts that were relevant to the study objectives. These categories were explored in further depth in a subsequent interpretive level of analysis drawing from interpretive analysis and discourse approaches in order to describe and understand the content, meaning and function

of the data in the context of the whole interview, of other interviews, and of the wider social settings where women engaged in their RRM decision-making. Although analysis was shaped by commonalities between participants and across the data, attention was also paid to heterogeneity and divergence. In this respect, contradictions to the emerging analysis and pivotal cases were recurrently discussed. Disagreements between researchers during the data analysis were encouraged and resolved through discussions. With the aim of producing a detailed and reflective description of the analytic process, summaries and an analytic record of the steps taken were produced (see Appendices P and Q). The quality of the developing analysis was assessed against standards of validity, including theoretical and catalytic validity (Kincheloe & McLaren, 2000) and trustworthiness (Guba, 1981; Shenton, 2004), as explained in Appendix L.

Results

Sample Characteristics

Participant's demographic information is presented in Table 1. Participants ranged in age from 31-70 years and had varied educational backgrounds, with the lowest education level being GCSE or equivalent, and the highest being postgraduate degrees. Two participants were retired and seven were working part-time. Two of the three women who opted for BRRM did not have a personal history of breast cancer, whilst the four participants who opted for CRRM had personal histories of breast cancer. Five participants were interviewed prior to the surgical procedure, and they were on a waiting list for RRM. Two participants (Participant 4 and Participant 6) were interviewed after having RRM, and both expressed being satisfied with the outcome. Five participants had a family history of breast cancer, and five had a family history of other cancers. Two women had a BRCA1/2 genetic

mutation, one had a CHEK2 genetic mutation, two participants were BRCA1/2 negative, and the genetic mutation status was unknown for two participants.

Table 1

Participant Demographic Information

ID	Age	BC history	Time since BC diagnosis	Other cancer history	Family BC history	Family history of other cancers	Genetic testing status	RRM	Inter-viewed pre or post RRM	Highest education level	Work status
1	46-50	Yes	20 months	No	Yes	Yes	Negative	CRRM	Pre	GCSE or equivalent	Part-time
2	56-60	Yes	108 months	No	Yes	No	Un-known	BRRM	Pre	A-levels or equivalent	Retired
3	56-60	No	N/A	No	Yes	Yes	BRCA1/2 Positive	BRRM	Pre	Postgraduate	Retired
4	66-70	Yes	48 months	No	No	Yes	Negative	CRRM	Post	GCSE or equivalent	Part-time
5	46-50	Yes	29 months	Yes	No	No	CHEK2 positive	CRRM	Pre	Postgraduate	Part-time
6	31-35	No	N/A	No	Yes	Yes	BRCA1/2 Positive	BRRM	Post	Postgraduate	Part-time
7	46-50	Yes	12 months	No	Yes	Yes	Un-known	CRRM	Pre	Bachelor's degree	Part-time

Note. ID = Participant identification number; BC = Breast cancer; RRM = Risk Reducing Mastectomy; CRRM = Contralateral Risk Reducing Mastectomy; BRRM = Bilateral Risk Reducing Mastectomy; BRCA1/2 and CHEK2 = Breast cancer susceptibility genes; GCSE = General Certificate of Secondary Education; A-Level = General Certificate of Education Advanced Level (secondary school qualification).

Overview of Findings

Women's views about RRM and the ways in which they made their decision were remarkably homogenous in important respects, despite the wide-ranging ways in which they first become aware of RRM and regardless of their diverse personal histories. Women made

their decision to undergo RRM immediately and generally without prior consideration, but when asked were not able to describe their decision-making in detail. Their views of RRM were associated with a sense of safety and relief and using the decision to gain control over their lives. A posteriori, they continuously elaborated on these and other reasons for their choice, in a process of post-decisional reviewing and justification.

Table 2

Summary of Findings

Main categories	Sub-categories
Before the decision	<p>“I don’t want to go through any of that (again)”</p> <p>Finding out about RRM and initial reactions</p>
The decision	<p>“So it was just immediate that I made that decision”</p>
After the decision – Reasons for RRM	<p>Relief and safety</p> <p>“This is my doing something”: Taking action, taking control</p>
After the decision – Reviewing	<p>Post-decisional reviewing and processing</p>

Before the decision

Participants spoke of personal or familial experiences of cancer that had affected them. They also detailed how they had first learned about RRM. Most participants knew about RRM before being in the position of making their decision.

“I don’t want to go through any of that (again)”. All women had experienced cancer up close; some had personal recent experience of the illness and its treatment, whilst others had witnessed its impact having cared for relatives or friends who had developed it: *“And I’ve*

lost my mum and my brother to cancer. And it's, it's just too raw, so I wanted to do everything in my power... to survive" (Participant 1)³. Their recollections of the experiences of those who had passed away from breast cancer or other cancers were salient in their accounts of their decisions to have a RRM: *"(...) she [friend] actually died in November, so watching her deteriorate just made me even more determined to not be in that situation really"* (Participant 3). Some women described the increased awareness that they had about the pervasiveness and morbidity of cancer due to their contact with people with it, be it through their personal lives or through their jobs: *"(...) my granddad got lung cancer and died (...) and my auntie had lung cancer and she died at 47, so I might have like a hyperawareness of cancer and I'm hyper-frightened"* (Participant 6).

Those who had undergone treatment for cancer, such as chemotherapy or radiotherapy, also recollected this experience as difficult and undesirable, and one which they wished to avoid: *"So I went through everything and then as I got better and got over it and I thought - I don't want to do that again. I don't want to go for chemotherapy, I don't want to lose my hair"* (Participant 4). For some participants this extended to the effect that their illness and treatment had had on their families as well as on themselves: *"The thought of me having more chemotherapy, because I know my kidneys wouldn't hold out for anymore because they're not great at the moment anyway, and the fact that, you know, I wouldn't like to put my family through all that again"* (Participant 5). As highlighted in this quote, their motivation to avoid future disease and treatment constituted a strong reason why they decided to opt for RRM, and for many was related to their desire to protect their loved ones from that too.

Finding out about RRM and initial reactions. Participants recounted different ways of learning about RRM. This variability pertained to both the means by which they first found out about RRM and to variations in how well remembered these occasions were, from a

³ Ellipses denote pauses in speech. Bracketed ellipses indicate text has been omitted.

vague sense of having known about it to a salient memorable moment. The latter was the case of Participant 4 when she was about to undergo surgery for breast cancer: *“The morning I was going in for my surgery there was a lady came in and, as you do, you sit and you talk, and she was having both breasts off and I said to her ‘why are you doing that?’ so she said ‘well, it’s only in one’ but she said ‘I’m not taking the chance’”*.

Several women found out about the procedure from other breast cancer patients, some of whom were family members: *“We knew that she [aunt] was going to have obviously the other one [breast] removed, and from there really (Participant 6). Other participants recalled hearing about RRM via the media, their responses to which were variable, including praising the widely publicised decision of a celebrity: “Through the media probably, and Angelina Jolie, and her having it, that was the first one that I’d heard [about] (...) It’s just that, you know, she spoke about it publicly, and that she was quite brave and positive”*. Participant 2 also reported: *“It was just something I read, absorbed it, put it at the back of my mind, and didn’t really discuss it any further at that point”*, reflecting the little relevance that RRM had to participants at the time when they first found out about it.

The decision

Decisions were instantaneous in all cases and participants had little insight into how they made them, although one woman (Participant 3) had considered the decision hypothetically.

“So it was just immediate that I made that decision”. In their reflections about their election to have a RRM, almost all women described an apparently instantaneous decision that required no deliberation, as is evidenced in Participant 1’s account: *“It wasn’t a hard decision. As soon as I was diagnosed I knew, while the consultant was talking to me, what I was going to do (...) It was really strange, my sister is next to me crying her eyes out, and I*

was really straight-faced, just staring at the consultant while she's explaining to me erm and all I kept thinking about was 'take the other one off, take the other one off', so it was just immediate that I made that decision". Accounts such as those of Participant 6, who knew that she wanted to have a BRRM at the time of finding out about her genetic high risk of breast cancer, illustrate the sense of urgency with which the decision was reached: *"She [genetics doctor] said 'your risk is 80%', at which point I said 'just get them off as fast as you can'. There was no... there was no 'but I feel sad'. Because I just thought, I'm... just... quick. Just as quick as you can"*.

An exception was Participant 3, who had thought about RRM because through her job as a radiographer working with breast cancer patients she had considered that decision in hypothetical terms in the past: *"It's always been in the back of my mind, all these years when I've been [working with] ladies who've had breast cancer and I've always thought that if I was in that situation I would want to be rid of my whole breast, get rid of the lot. I wouldn't want a lumpectomy or anything. So I've always had that feeling in the back of my mind. So then when I got into, not the same situation obviously, but a way of preventing me being in that situation, it was fairly straightforward for me. It's always been my frame of mind anyway."* (Participant 3).

Women's accounts denoted their certainty about their decision, and they recalled the moment when they reached it as well as how certain they were about their choice, yet attempts to expand on that particular moment through means of interview questions were not successful:

- Participant 5: *"When I found out I did have breast cancer and I knew I was going to have surgery on this left breast I did say to the consultant 'Can you just take the two of them off?' and he said 'It's not as easy that. We need to concentrate on the side that's got the cancer first and then obviously after that we'll chat about...' , so*

I sort of researched it myself. When I say researched, just had a little look about different forums on the internet and things people had chosen to do really”

- (...)
 - Q: *“And do you remember at the time, so obviously, you know, before you spoke with the consultant and everything, do you remember what you thought about it [RRM]?”*
 - Participant 5: *“I’d made me mind up before I’d spoken to him.”*
 - Q: *“Had you, yeah?”*
 - Participant 5: *“Definitely ... I definitely wanted the other breast off. I definitely wanted a risk-reducing mastectomy.”*
 - [...]
 - Q: *“Why do you think it was such, you know, such an easy decision for you?”*
 - Participant 5: *“I don’t know. I just thought to myself - I’ve made my mind up and that’s what I want doing”*

Despite interviewing efforts to explore how they made their decision to opt for RRM, women were not able to expand on this in detail, more often describing their main motives for it or their memory of when they reached their decision instead:

- Q: *I just wondered how, if you can explain to me more - it might be difficult to put it in to words - but how did you come up with that decision, if that makes sense?*
- Participant 7: *Because I’m a cautious person.*
 - [...]
 - Q: *Do you remember how it became like your decision?*
 - Participant 7: *Well after they told ... the first ... when they told me and they did the tests but they hadn’t said it’s cancer yet, but I was having those tests ... yeah ... and I had to wait 10 days and I went back home and we sat and I was talking to my husband*

and I said to him, I said 'I don't want them to do a lumpectomy. I just want it taken away now', and it was then. It was before I found out it was cancer.

After the decision – Reasons for RRM

Women described that with their RRM decision they gained relief and perceptions of safety and control. These were advanced as reasons for their RRM choice, but women did not report thinking about these when they made their initial decisions.

Relief and Safety. Women experienced concerns and uncertainty associated with their increased risk of breast cancer, particularly about the possibility of developing the disease in the future: *"It's just at the back of your mind. It just sits there quietly but it's there. So hopefully it will disappear after that"* (Participant 3). Accompanying their election to undergo a RRM was a wish to reduce their risk of breast cancer, as well as to alleviate their fears of developing cancer, and they achieved a sense of relief by doing so: *"Even if he [surgeon] said 'we can't really do anything other than take your boobs off', that's fine, I've more or less accepted, I'm not looking to have a perky pair, I'm not really bothered, you know, as long as I get rid of the worry and stress"* (Participant 2). Participants often emphasised that this was of more importance than cosmetic outcomes.

Some participants perceived RRM as providing them safety from fear in the face of the recurrent uncertainty surrounding scans and tests, imprinted in their memory via their personal experience or via a vicarious awareness: *"I'm thinking I can have yearly mammograms and have that thought in my mind all the time, or every time I go for my mammogram I'm going to be worrying - are they going to find something? - or I can have a mastectomy and I'm never going to have to worry about that again, am I?"* (Participant 5).

It appeared that for most of the participants interviewed, the notion of eradicating the tissue that can harbour the risk was strikingly convincing in comparison to the perceived

uncertainty of the effectiveness of other risk reducing options, which participants did not view as sufficient to reduce their risk nor to assuage their worry of developing breast cancer:

“Well, from what I can tell my options are a healthy lifestyle or the surgery, and really the healthy lifestyle isn’t enough for me. I would like to know that the tissue is gone, really”
(Participant 3).

Furthermore, some women, such as Participants 1 and 2, saw RRM as an option that would enable them to stop experiencing worry and to gain closure instead, which would have not been achieved by other risk reducing or early detection interventions:

- Q: *“Why did you prefer this RRM option over the mammograms and yearly checks?”*
- Participant 1: *“Peace of mind, it’s just peace of mind all the time for me. I just don’t want to live in fear. I want to sort of put it to bed and let me move forward with my life”*
- Q: *“Yeah, so would you feel that the mammograms wouldn’t allow you to do that?”*
- Participant 1: *“No, they probably would, but it’s every year, do you know what I mean? It’s something you’re waiting for. And I want to know now that I’ve done the best I can, and I can move forward”*

This was echoed by Participant 2: *“It’s just that it’s quite final, once it’s done, it’s done. And it means that you don’t have to keep coming back for more lumpectomies, or radiotherapy, all that’s gone and dusted”*.

In contrast with the sense of urgency that they conveyed when they first reached their decision to have RRM, after the option was discussed and agreed with their surgeons, several participants felt sufficiently safe from worry so as to request to postpone the surgical procedure in order to time it conveniently around important life events and commitments such

as weddings or holidays, or starting a new job role: *“She [surgeon] would have done it within weeks but I had holidays, hen dos, two weddings, and her words were “well, when can you fit me in?”, so that’s what she said to me.”* (Participant 4). This postponement initiated by women did not affect their views of RRM or their certainty about their decision.

‘This is my doing something’: Taking action, taking control. Women’s choice to undergo RRM appeared to stem from, or to reflect, their need to obtain a sense of control against the disempowerment they felt due to the uncertainty characterising many aspects of the disease and of its treatment: *“The chemotherapy, the radiotherapy and, you know, the “oh my god, what if” and, like I say, the day I thought I was dying. There’s no control over that. It comes in and it’s sitting looking at you and then you’ve got to go - hang on a minute, you know, I need to do something positive”* (Participant 4).

Women’s proactive motivations for electing to have a RRM were expressed in their perceptions of needing to do ‘all that they can’, a motivation that was often grounded in their strong wish to survive for their children and loved ones: *“I can’t just sit back and do nothing because to sit back and do nothing is to give up and I won’t do that. I want to do something because I want to be here for my children and that’s the main thing so I’m ... I need to do something and this is my doing something”* (Participant 7).

Seemingly due to its attribute of being an active response to breast cancer risk, RRM was also viewed as a protection against regret in the future: *“I had 4 tumours but they were at the back of my breast bone so I would never have felt them, and this is a fear factor of mine, I don’t want to put myself in that situation where I can’t feel them and it’s too late”* (Participant 1). Other women, such as Participant 7, reflected the belief that a possible future diagnosis of breast cancer due to residual risk after RRM would be more bearable and preferable than the regret of not opting for the procedure that was available to her: *“If I do it and I - I understand I could still get cancer - and I still get breast cancer bizarrely, but say I do, I would sit there*

and think ‘but I did everything I could at that’ ... and that’s the way I view it. To not do something when you can do something? No. Do it.”

After the decision - Reviewing

After their RRM decision, women reviewing it by means of consulting other people’s views and by finding additional information about RRM.

Post-decisional reviewing and processing. Following their choice, and prior to the surgical procedure taking place, women engaged in a post-decisional reviewing process of an iterative nature:

- Q: *“So through that time, did your confidence in that decision waver, or did you change your mind at any point?”*
- Participant 6: *“It wavered very slightly, but only very slightly, and I think that was me being sure because I was so ‘yeah I’ll just have it done, I’m not bothered’, I didn’t want to look silly by then breaking down and having some sort of big breakdown after it and then somebody saying ‘well, you shouldn’t have been such a knob and said you can cope with it if you clearly can’t’ - so I was trying to get mentally prepared really, and the only time I thought ‘should I?’ is when I was just double-checking in my mind really. I was just double-checking.”*

For women in this study, active reviewing of their decision also took place, for example, through conversations with other people including about loved ones’ views regarding RRM:

“So it wasn’t a moment of doubt for me, it was just a moment of doubt from somebody else that made me think ‘well, am I doing the right thing?” (Participant 2). One participant, whose husband worried about the risks of the procedure, described how the possibility of surgical complications concerned her but ultimately did not make her change her mind: *“Well then the doubts come in and I did have a couple of sleepless nights when I thought - my God, you*

know, I might die - and then all of a sudden I thought 'I know I'm not going to die', you know, and I don't feel I'm going to die" (Participant 4).

Participants sought additional information about the procedure, many primarily online, and described being selective about the information accessed: *"It's always interesting to see what people have been through and what they're talking about and some of it you think 'oh I'll just ignore that one' but a lot of them are quite interesting"* (Participant 3). None of the women used the information to question their choice, but it did serve them to ratify their decision and their confidence in RRM as the best option for them:

- Q: *"What was the effect of researching that information on your decision?"*
- Participant 3: *Confirming really.*
- Q: *Confirming, yeah?*
- Participant 3: *Yeah. I don't think there was anything that made it less likely. I don't think anything put me off really. Most of the stuff ... well, most of the stuff that I found was quite positive.*

Only one contributor, Participant 7, described doubting her decision to have a RRM after having reached it with determination; this occurred in the context of her emotional reaction to a diagnosis of breast cancer and feelings of loss of control during its aftermath: *"That's when I broke down and I thought ... I lost even more control. I wasn't me. I realized I wasn't me and I thought 'I cannot make another decision again. I don't want to make another decision again' and then my sister stepped back and she said 'just go with what you did at the start. Your own decision'".* This period of hesitation about her choice to undergo RRM, precipitated by a perceived loss of control, was resolved by a return to a sense of certainty about her decision: *"And I almost took a deep breath and thought 'Yeah. Yeah'. I just needed her to tell me, someone to tell me that"* (Participant 7).

Discussion

This qualitative study explored women's views of RRM and their decisions to undergo it. Participants viewed breast cancer and its treatment as something they strongly wished to avoid, and made their decision to undergo RRM in an immediate and intuitive manner, regardless of how they had learned about this prophylactic option. Participants reported that a RRM was an option that gave them a sense of safety and relief in the face of the worry and fear about breast cancer that they experienced. Their decision was also a way to engage in a proactive action, which appeared to serve as protection from possible future regret. Participants did not report considering these reasons before the decision, however they reported experiencing fear, worry and loss of control. Subsequent to their decision, participants engaged in post-decisional processes that sustained their views of RRM being the preferred risk-reducing alternative for them.

The results of this research are consistent with the findings reported by Brown et al. (2017) as well as with those described in the wider literature (Beesley et al., 2013; Fielden, Brown, Saini, Beesley, & Salmon, 2017; Simard et al., 2013). The decision to undergo a RRM originated in response to the sense of menace or fear. This fear was related to participants' experiences of cancer, whether through personal diagnosis and treatment, or due to family and/or friends developing cancer. As reported by Covelli et al. (2015), cancer risk perceptions and decision-making are shaped by the experience of affected family members, as opposed to statistical probabilities, and might be a motivating factor in prophylactic surgery (Singh et al., 2013). Participants in this study communicated their experiential knowledge of the illness and its treatment, for example recounting salient stories of breast cancer recurrence in family members or other cancer affected patients, and expressed their determination to avoid both the disease and its treatment. Fear of recurrence has been found

to be a principal concern among women with breast cancer (Fischer, Dolbeault, Sultan, & Bredart, 2014).

Participants formed their risk-management decision in an apparently immediate manner, for example voicing their preference to have a RRM when informed about their breast cancer diagnosis, or even prior to finding out the outcomes of their genetic mutation tests. Furthermore, the stability of their choice was evident given that participants reported not changing their minds about their RRM decision. This echoes van Dijk, Roosmalen, Oteen and Stalmeier's (2008) findings that women had formed their risk-management preference before genetic test disclosure, and this preference did not change after a positive test result.

Participants were not able to convey details of how their immediate decision was formed. Despite not matching our initial expectations at the start of this research, this failure to expand upon the decision-making moment is informative. It appears to indicate that, although women's motivations for and views of the surgery are accessible to them, the manner in which they make their minds up might not be so, due to the immediate and instinctive attribute of the decision-making.

The need to obtain a sense of control and to be proactive was often described in women's accounts of 'doing all that they could' to prevent future breast cancer. These imperative stances were associated with the disempowerment and vulnerability they experienced due to having breast cancer and undergoing treatment, as well as with their drive to survive for their loved ones. Similar findings have been reported elsewhere (Haroun et al., 2011). Women's anticipated emotions, such as anticipated feelings of regret in case of a negative outcome, are seemingly bound to their proactive stance and motivation to do all that they can in order to prevent future harm. As argued by van Dijk et al. (2008), selecting the option of RRM due to perceiving it to be the safest option to avoid potential future self-blame

and regret is an understandable reason for choosing prophylactic surgery. Nevertheless, evidence indicates that when making decisions associated with a heavy emotional burden people might act more inflexibly and might disregard trade-offs between costs and benefits (van Dijk et al., 2008).

Affect and emotions play an influential role in decision-making (Zikmund-Fisher, Fagerlin, & Ubel, 2010), and recent research by Beesley et al. (2013) has found that psychological reasons such as the reduction of patients' cancer worry influenced their decision to undergo CRRM. Furthermore, Rendle, Halley, May and Frosch (2015) have found that an important decision-making factor for women opting to have a CRRM was avoidance of future breast cancer surveillance and of associated worry. The findings of the present study reflect similar themes, with participants viewing RRM as an alternative that would enable them to stop experiencing cancer-related worry, and to gain a sense of relief from their concerns and of safety from their breast cancer risk.

Participants' post-decisional processing can be understood in light of Svenson's Differentiation and Consolidation theory (Svenson, 1992). The findings of this study suggest that the attractiveness of RRM might depend on the importance women place upon its salient attributes; namely, that it offers the possibility of maximizing risk reduction and perceived control whilst minimizing fear, worry, and future regret to a greater degree than the other risk-reduction alternatives available. This might be the case even for participants for whom other risk-reduction alternatives such as chemoprevention were not suitable or those for whom surveillance would not guarantee early detection of breast cancer, as arguably an alternative to RRM would be to opt to decline it. Following the decision to undergo a RRM, post-decisional processes enabled women to both to consolidate their decision and to minimise possible regret about making a wrong decision. This refers to a process of justification reported elsewhere (Brown et al., 2017), which included accessing information

selectively and drawing on their family and friends to support their decisions, which appeared to be psychologically helpful for participants.

Both patients' appraisal of the attributes of RRM as the most convincing and suitable risk-reduction alternative and the subsequent consolidation processes appeared to be facilitated by heuristics. An example of this might be the influence of availability and simulation heuristics upon individuals' perceptions of their risk of breast cancer, since participants had either personal or familial experiences of this disease. By means of the affect heuristic, it is also possible that participants' conscious and unconscious emotional states might have influenced their information processing and thus decision-making. For instance, it could be hypothesized that the positive feelings of relief and safety from worry associated with the prospect of reducing breast cancer risk, and the positive affect resulting from perceiving a gain of control by taking action, lead to participants' view that RRM was the most convincing alternative and 'the right thing to do'.

Furthermore, this theoretical understanding might account for participants' initial decision to have RRM remaining stable over time. Differentiation and Consolidation theory postulates that the probability of a preliminary chosen alternative being the final choice is greater than the probability of it being changed for another option, even if its attributes are equally attractive (Svenson, 1992). In this study none of the participants' risk-reducing preferences oscillated between RRM and other options, indicating that their preliminary choice of undergoing a RRM remained the leading one throughout the differentiation and consolidation process. Nevertheless, caution should be exercised when considering the above, due to the inherent selection bias of interviewing participants who had reached their decision to have a RRM and who did not intend to revisit it. Further research is required to expand the findings of this study, and to include the views of women for whom the decision-

making process was experienced differently, and of those who did not regard RRM as the most convincing alternative to manage their risk of breast cancer.

Strengths and Limitations

Participant recruitment was dependent upon the rate of patient referrals to the psychology service. This precluded obtaining a larger sample of participants and therefore data saturation has not been reached, potentially limiting the generalisability of findings. Nevertheless, participant inclusion criteria for this study were purposefully broad in order to recruit a heterogeneous sample, representative of the population of women opting to have a RRM at the breast unit. Thus, this study examined RRM views and decision-making processes in women with a wide-range of personal experiences, including personal or familial history of breast cancer, time since breast cancer diagnosis, genetic mutation status, and demographic information, some of whom were interviewed pre-operatively and others who had already undergone RRM. However, the heterogeneity of the sample might affect the integration of the results into a unified picture as well as limit the generalisability of findings.

The use of a qualitative methodology whereby a pragmatic approach to data analysis was adopted enabled researchers to continuously revise the ongoing investigation in light of the developing findings as new data emerged, allowing for the analysis and interpretation of participants' experiences without a preemptive reduction of the data. The use of face-to-face semi-structured interviews provided in-depth information about individuals' views and beliefs (Ho, 2006); however, the use of this method of data collection might not adequately capture aspects of participants' opinions nor of their decision-making process, such as those which are implicit, not accessible, or which participants are unwilling to share with the researcher. For example, stressful experiences might affect memory processes and the ability to recall details of past events (Schwabe, Joëls, Roozendaal, Wolf, & Oitzl, 2012).

Furthermore, some participants indicated they had in the past considered, in hypothetical terms, what they would do regarding the option of RRM; these accounts might be an indication of earlier decisional processing that would merit further investigation.

Key aspects of the research quality and validity have been carefully considered through this research, and are presented in depth in Appendix L.

Clinical Implications and Future Research

Advances in the understanding of women's views of RRM and of the ways in which they make RRM choices are of immediate relevance to clinicians working with patients facing RRM. This study adds to a growing body of literature demonstrating that patients' decisions about RRM are shaped by emotions; these decisions were reached in an immediate and decisive manner, and were followed by a process of post-decisional deliberation whereby the appraisal of the attributes and anticipated outcomes of RRM as the most convincing and suitable prophylactic option were sustained. The role of emotions and of inferential rules such as heuristics in decision-making can have both positive and negative outcomes; indeed, De Vries, Fagerlin, Witteman and Scherer (2013) posit that emotion-based decision-making can be advantageous and lead to better decisions. Thus, as has been argued by Brown et al. (2017), understanding the option to undergo a RRM as a means of reducing negative affect can be respected as an autonomous choice if risks and benefits have been taken into account by patients.

Additionally, it is important for clinicians to understand and acknowledge the emotions and negative affect experienced by women at high risk of breast cancer, such as fear, worry or the motivation to do all they can to avoid future regret, and which are at the center of their decision-making process. In this respect, it could be argued that some patients might benefit from psychological interventions aimed specifically at reducing these difficult emotions.

Furthermore, most women appeared to decide about RRM at an early stage of their illness or genetic counselling, before the risk-reducing option was discussed with them by health care professionals. Therefore, it might be of benefit for comprehensive discussions about risk-management preferences and about coping with breast cancer risk to be held between clinicians and patients at an early stage.

Future research to expand upon the findings of this study can be considered. Participants were interviewed at one point in time after they were assessed as having reached their decision to undergo RRM. Future investigations could adopt a longitudinal approach by interviewing women at different time points, including between the RRM decision being initially considered and after the RRM procedure has or has not taken place; this would be of particular relevance to investigate the experiences and views of patients who delay formalizing their decision to have an RRM, who subsequently change their minds about risk-reduction strategies, or opt to not have a RRM.

Conclusion

Women at increased risk of breast cancer face complex decisions to manage this risk, with one of the options available being RRM. The results from this study add to our understanding of the decision-making process regarding RRM, providing valuable insights into patients' perspectives about their choice to undergo RRM and about the surgical procedure itself. This research complements existing studies highlighting that patients might make RRM-related decisions guided by their emotions. Additional research is needed to explore these findings in larger samples and among women who have had different experiences of RRM decision-making.

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APPENDIX A

AUTHOR GUIDELINES: EUROPEAN JOURNAL OF CANCER CARE



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Impact Factor: 1.794

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APPENDIX B

LITERATURE SEARCHES AND SELECTION

Table 1

Total combined search by database

Database	Results
PsycINFO	
MEDLINE	194
CINMAHL Plus	
Web of Science	294
PubMed	15
Scopus	270
Total	773

Table 2

Reasons for exclusion

Reason	Examples
Sample	Carer Child Current BC Male Not BC/Not increased risk of BC Professional sample Other
Topic	Not women's experience Not specific to BC/RRM Other
Methodology	Single case study
Type	Review Poster Book Book critique Editorial/position paper Other
Duplicate not previously identified	

Table 3

Exclusion from abstract

Reason	Number
Sample	6
Topic	55
Methodology	0
Type	31
Duplicates not previously identified	0
Total	92

Table 4

Exclusion from full text

Reason	Number
Sample	6
Topic	24
Methodology	0
Type	0
Duplicates not previously identified	1
Total	31

APPENDIX C
QATSDD TABLE

Criteria	Hallowell (1998)	Kwong & Chu (2012)	Salant et al. (2006)	Borreani et al. (2104)	Claes et al. (2005)	Haroun et al. (2011)	Kram et al. (2006)	Lerman et al. (2000)	Litton et al. (2009)	Lodder et al. (2002)	Meiser et al. (2003)	O'Neill et al. (2010)	Ray et al. (2005)
Explicit theoretical framework	3	3	2	1	2	1	2	1	3	3	3	2	3
Statement of aims/objectives in main body of report	3	3	3	3	3	3	3	3	3	2	3	3	3
Clear description of research setting	3	3	2	3	3	3	3	3	3	3	3	3	3
Evidence of sample size considered in terms of analysis	0	0	0	0	3	0	0	0	0	2	0	0	0
Representative sample or target group of a reasonable size	2	2	3	3	2	3	2	3	2	3	3	3	2
Description of procedure for data collection	3	3	2	1	3	3	3	3	3	1	3	3	3
Rationale for choice of data collection tool(s)	2	2	3	0	1	1	1	3	1	0	2	2	2
Detailed recruitment data	3	3	3	3	1	3	3	3	3	3	3	3	3
Statistical assessment of reliability and validity of measurement tool(s) (Quantitative only)	-	-	-	0	0	0	0	0	0	2	2	0	0
Fit between research stated research question and method of data collection (Quantitative only)	-	-	-	2	3	3	3	3	2	3	3	3	3
Fit between stated research question and format and content of data collection tool (Qualitative only)	3	3	3	-	-	-	-	-	-	-	-	-	-
Fit between research question and method of analysis	3	3	3	3	3	3	3	3	3	2	3	3	3

RRM PERCEPTIONS AND DECISION-MAKING AMONG WOMEN AT HIGH RISK OF BC

Good justification for analytic method selected	2	2	2	0	2	1	2	2	1	3	2	1	2
Assessment of reliability of analytic process (Qualitative only)	1	1	1	-	-	-	-	-	-	-	-	-	-
Evidence of service user involvement in design	0	0	0	0	0	0	0	0	0	0	0	0	0
Strengths and limitations critically discussed	3	3	3	3	3	3	3	3	3	3	3	3	3
Total Score	31	31	30	22	29	27	28	30	27	30	33	28	30
Percentage Score	73.8	73.8	71.42	52.38	69.04	64.28	66.66	71.42	64.28	71.42	78.57	66.66	71.42

Note. 0 = Not at all; 1 = Very slightly; 2 = Moderately; 3 = Complete.

Summary

The 16 items of the QATSDD were rated on a 4-point scale from “not at all” (0) to “complete” (3). Comprehensive scoring guidance notes for all criteria can be found in Sirriyeh et al. (2012). Percentage scores were calculated using the actual score and the maximum total score of 42. In order to categorise the quality ratings of the papers included in this review, studies scoring over 75% were considered to be of “high” quality, those between 50% and 75% were ranked as “good”, those between 25%–50% as “moderate”, and studies with a rating below 25% were considered to be of “poor” quality.

An overview of the quality ratings as appraised by means of the QATSDD indicates that studies included in this review can be considered of overall good quality, with QATSDD ratings ranging from 78.57% to 52.38%, with an average score of 68.85%. However, an examination of the individual criteria items of the QATSDD evidences some notable shortcomings in the methodological quality of the studies included in this

review. The most common areas of concern include the lack of justification of sample sizes, lack of references to reliability or validity of measures used, poor justifications for analysis methods, and complete absence of service user involvement in study designs.

Reference

Sirriyeh, R., Lawton, R., Gardner, P., & Armitage, G. (2012). Reviewing studies with diverse designs: the development and evaluation of a new tool. *Journal of Evaluation in Clinical Practice*, 18, 746–752. doi:10.1111/j.1365-2753.2011.01662.x.

APPENDIX D
PARTICIPANT INFORMATION FORM

**Patient and surgeon decision-making in risk-reducing mastectomy:
an ethical and empirical analysis**

The Royal Liverpool and 
Broadgreen University Hospitals
NHS Trust



You are being invited to take part in a research study. Before you decide to take part or not, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear, or if you would like more information. Take time to decide whether or not you wish to take part.

Thank you for reading this.

What is the purpose of the study?

We are interested to learn how women make decisions about whether to have a risk-reducing mastectomy or not. We are also interested in how clinical staff help them to make their decisions and how they see their role in the decision process. We hope that our findings will help us to understand how clinicians can best help women to make decisions in future.

Why have I been chosen?

You have been asked to take part in the study because we understand that you have considered having a risk-reducing mastectomy at the Breast Unit of the Linda McCartney Centre.

Do I have to take part?

You do not have to take part in this study. It is up to you to decide whether or not to take part. If you decide to take part, you will be given this Information Sheet to keep and will be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time and without giving a reason. Whether you participate or not will not affect the care you receive. If you were to participate but then withdraw, this would not affect the care you receive either.

What will happen to me if I take part?

If you choose to take part, you will be given a copy of this Information Sheet and a signed Consent Form to keep.

We would like to interview you about how you have thought about the possibility of having this operation and how you decided about it. In particular, we are interested in how you thought and felt about the risk of cancer and risk-reducing mastectomy and how you and your clinician came to decide about what is best to do. You can choose whether you would prefer to be interviewed at the Breast Unit, at the University of Liverpool or at your own home. If you choose to be interviewed at the Breast Unit or the University of Liverpool, your travel expenses will be paid.

- Your interview will be audio-recorded if you agree to this. The length of the interview will vary, depending on how much you wish to talk about. However, the interview is likely to last between 20 minutes and 60 minutes.
- The audio-recordings of the interview will be typed up by the researcher. All information which might identify you will be removed and replaced by a code so that any personal information (such as names, addresses, doctors' names etc) will not be included in the research.
- The interview will focus on the following topics: how you viewed the risk of breast cancer (re)occurrence, how you first became aware of the possibility of having a risk-reducing mastectomy, the factors that contributed to your preferences to have the risk-reducing mastectomy or not and how you and the clinical staff interacted to make the decision.
- With your permission, we would also like to interview clinical staff who have been involved in this decision. None of the interviewees will be told what any other interviewee has told us.

What are the possible disadvantages of taking part?

The interview may involve talking about information that was or is upsetting for you. However, you do not have to talk about anything you do not wish to. If you feel that you need help with any of the matters that we have discussed one of the research team will be happy to speak to you to give you advice on further sources of support.

What are the possible benefits of taking part?

You will not personally benefit from taking part in the study. However, any information that you give us can help us find out more about how people make this decision and about the roles that clinical staff should play. We hope that our findings will help clinicians to be more effective in helping women in future.

What if something goes wrong?

If you wish to complain, or have any concerns about any aspect of the way you have been approached or treated during the course of this study, the normal National Health Service complaints mechanisms will be available to you. You can complain to:

Customer Relations Team
Royal Liverpool and Broadgreen University Hospitals NHS Trust
Prescot Street
Liverpool
L7 8XP
Tel: 0151 706 4903
complaints@rlbuht.nhs.uk

Liverpool Women's Hospital Patient Quality team 0151 702 4416

Will my taking part in this study be kept confidential?

All data collected for this study will be kept safely and securely on computer and on transcribed paper records. Dr. Stephen Brown will be the custodian of all study data. An audio-recording will be made of the interview and transcribed onto paper. After we have analysed the data the audio-recording will be destroyed. All information about you will be confidential. Any information which identifies you (for example, your name, names of family, friends, and doctors, addresses, names of hospitals, telephone numbers, date of birth, and employment) will be removed from transcriptions and replaced by a code.

What will happen when the study ends?

After all identifying details have been removed from the transcribed records of consultations and interviews, these will be analysed by the study team. The results will be published in reports and scientific journals, but it will not be possible to identify any individuals from these reports. Any quotation used in the writing up of the final study will be kept anonymous.

With your permission, transcripts of audio recordings of interviews and consultations will be stored at the University of Liverpool for 10 years after the end of this study for possible use in future studies. If you consent to it, a printed transcript of the interview will be offered to UK Data

Archive where it will be made available to future researchers. All information that might enable you to be recognised will be removed from these records

What will happen to the results of the research study?

The clinical psychology student who is part of the team will write up the results of the study for the Degree of Doctor of Clinical Psychology. This thesis will be kept in the library of the Division of Clinical Psychology, University of Liverpool. A copy of this thesis will also be kept in the University's Library. After the interview, you can ask if you would like a summary of these results, which will then be available after October 2017. You will not be identified in these results.

We will also write the results up to publish them in academic journals. After the interview, you can also ask if you would like a copy of these publications. You will not be identified in any publications.

Who is organizing and funding the research?

The chief investigator is Dr Stephen Brown, a senior lecturer at Liverpool University. He is collaborating with Professor Peter Salmon and Dr Louise Fairburn. Ms Emma Shaw Núñez is a student in Clinical Psychology who is conducting the research as part of her qualification.

The Economic and Social Research Council is funding the research.

Who has reviewed the study?

The study has been reviewed by:

- The Department of Psychological Sciences at Liverpool University
- The Economic and Social Research Committee
- Liverpool Local Research Ethics Committee.

Contact information

Stephen Brown, Chief investigator
Department of Psychological Sciences
University of Liverpool
Liverpool L693GB

Tel: 0151 794 5526
Email: slbrown@liverpool.ac.uk

For independent advice please contact:

Patient Advice and Liaison Service
Latham Court
Bridgemere Close
Liverpool L7 0LS

Tel: 0800 073 1106
Email: PALS@liverpoolpct.nhs.uk

APPENDIX E
STUDY PARTICIPATION LETTER



Dear _____,

I am contacting you as in the past year you have attended an appointment with the Liverpool Cancer Psychology Service to discuss your decision to have risk reducing breast surgery. You will have met with Dr Jan Ablett, Dr Louise Fairburn, or Dr Lesley Doyle, Clinical Psychologists.

In collaboration with the University of Liverpool and the Royal Liverpool University Hospital, we are currently recruiting for a research study entitled '*Patient and Surgeon Decision-Making in Risk Reducing Surgery*' which is interested in learning how women make decisions about whether to have risk reducing mastectomy or not. As you have recently made this decision, we are writing to ask if you would like to take part in the study. I have enclosed an information sheet with details about the study.

If you are interested in participating, or would like to discuss anything about the study, then please contact the psychology service on 0151 706 3126 by the end of March 2017 and I will be happy to discuss this with you. If you would like to participate, the interviews would last approximately 30 - 60 minutes and can take place either at your home or at the Linda McCartney Centre, whichever you prefer.

Yours sincerely,

Dr Louise Fairburn

Macmillan Principal Clinical Psychologist

APPENDIX F

PARTICIPANT CONSENT FORM



The Royal Liverpool and 
Broadgreen University Hospitals
NHS Trust

Centre Number:

Participant Identification Number for this study:

Patient Consent Form

Title of Project:

Patient and surgeon decision-making in risk-reducing mastectomy: an ethical and empirical analysis

Name of researcher: _____

Please initial box

1. I confirm that I have read and understand the information sheet dated (version) for the above study. I have had the opportunity of consider the information, ask questions and have these answered satisfactorily.		
2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my medical care or legal rights being affected.		
3. I understand that audio recordings will be made as part of this study, and that brief quotations from some interviews or consultations may be included in study reports. I understand nobody will be able to identify me in these reports.		
4. I agree to anonymous records of my interview being stored at the University of Liverpool for up to 10 years after the end of this study.	Yes	No
5. I agree to one or more staff who have participated in my decision being interviewed. I understand that no details of what I have said will be made known to them.	Yes	No
6. I agree to an anonymous transcript of my interview being held by the UK Data Archive and this being available to other researchers registered with UK Data Archive.	Yes	No
7. I would like to receive a summary of the findings at the end of the study.	Yes	No
8. I agree to take part in the above study.		

Name of participant

Date

Signature

Name of researcher

Date

Signature

APPENDIX G

ADDITIONAL STUDY DETAILS

Wider Study Information

This research was part of a wider study, ‘Patient and surgeon decision-making in risk-reducing mastectomy: An ethical and empirical analysis’, developed and conducted by researchers at the University of Liverpool and an NHS breast unit. The aims of this wider study are as follows:

1. To identify the outcomes that women consider when they form CRRM and BRRM preferences, whether and how they consider the concept of risk and how risk and distress affect their decision processes.
2. To understand how surgeons’ perceptions of patients’ best interests are formed, how the surgeons introduce and respond to requests for RRM, the role of distress in justifying or avoiding RRM, and how surgeons respond to requests that they may feel are ill-made.
3. To better understand how patients’ and surgeons’ joint decisions are negotiated, including any modifications that they may make to previous positions and the reasons for these.
4. To place these findings in relation to current thinking in medical decision-making and ethics with a view to determining better modes of practice in making RRM decisions.

Initial findings from this wider study suggested that women’s decisions to undergo RRM was driven by fear of breast cancer and the belief that they should do all they can to reduce risk, not by a utility-based comparison of risks of cancer and side effects of RRM. Women reported engaging in deliberative decision-making strategies, but these were seen as justifications for a decision that had already been taken. These results have been published elsewhere (Brown et al., 2017). Due to these interesting initial findings, there was an

opportunity for further investigation to be carried out with regards to patients' views of RRM and their decision-making, in line with the wider study aims and as a means of developing further the emerging outcomes. The researcher (ESN) was involved in planning the focus of the research here presented, in order for it to complement the ongoing wider study whilst being sufficiently different from previous research, so as to add novel evidence to the literature. Additionally, ESN led the reviewing and refining of the interview guide, as well as all aspects of data collection and analysis for this study. Data analysis was jointly conducted with the study supervisors, who were researchers in the wider project. The data and findings of this study will form part of the aforementioned wider study, which continues to be ongoing.

Ethics and Procedure

The wider study received ethical approval by the local NHS Research Ethics committee (13/NW/0421) and sponsorship from the University of Liverpool (UoL000957). The trainee clinical psychologist also received approval to collaborate in the study, and the NHS Trust granted the trainee a letter of access to conduct research within their organisation.

Patients were first informed about the study by clinical psychologists working at the breast unit, either in person or via written correspondence (see Appendix E). The purpose of this recruitment strategy was two-fold: on one hand, to ensure only the inclusion of patients who had indicated they had reached a firm decision regarding RRM and did not plan to revisit this decision and, on the other hand, to ensure that patients were able to provide informed consent to participate.

Clinical psychologists aiding with recruitment were asked to not approach women who had been interviewed in the initial phase of the wider study, nor those who had previously declined to participate. Patients who indicated to the Clinical Psychologists an interest in participating in the study were provided a Participant Information Sheet (Appendix D) and

were requested permission for the researcher (ESN) to contact them. The researcher established contact with the potentially interested patients by telephone in order to explain the study in further detail and to answer any questions raised by patients. To ensure adequate time to consider participation, telephone contact was made again after several days. All patients contacted this way indicated their interest in participating in the study, and interviews were arranged at a convenient time and location. Six participants were interviewed in a private room in a hospital setting and one was interviewed in their own home. To ensure the safety of the researcher, the University of Liverpool's lone working policy was adhered to. Prior to obtaining written consent and to the interviews commencing, the study information was verbally reiterated to patients in person in accessible language, and there was an opportunity to discuss any queries. The researcher reiterated to patients that their participation was completely voluntary and would not impact on their care.

Patients who consented to participate were informed that all information provided would be anonymous unless there were any concerns for their safety or that of others, as stipulated in the Dealing with Distress Protocol (Appendix K). During the conduct of this study, there were no cases that required breaches of confidentiality. Participants were also informed of their right to withdraw or remove their data without negative consequences. No participants in this study opted to do so. All participants were offered £6 to cover transport and/or parking costs. Four participants received this amount, and three participants declined it.

The Data Protection Protocol (Appendix J) was followed for the management of data throughout the study. Data were recorded using a digital audio recording device and was subsequently transferred to a secure computer file store at the University of Liverpool at the earliest opportunity, following which it was removed from the audio recorder. The researcher transcribed the initial two interviews, and a transcriber approved by the University

of Liverpool transcribed subsequent interviews. The transcriber was provided a template and specific guidance (see Appendix M) to ensure the confidentiality and anonymity of participants were protected. Following consistency checks and the removal of any potential identifying information, audio recordings were deleted. Anonymised interview transcripts were shared via email with the research supervisors for analysis. Anonymised demographic information was linked to the interviews by a participant identification number. Participants were informed that anonymised interview transcripts would be stored at the University of Liverpool for up to ten years, after which it would be destroyed. Signed consent forms were kept for the duration of the study and stored in a locked cabinet at the University of Liverpool. The data custodian (principal investigator) was responsible for ensuring data and identifiable information will be destroyed after the adequate time period.

Reference

Brown, S. L., Whiting, D., Fielden, H. G., Saini, P., Beesley, H., Holcombe, C., . . . Salmon, P. (2017). Qualitative analysis of how patients decide that they want risk-reducing mastectomy, and the implications for surgeons in responding to emotionally-motivated patient requests. *PloS One*, 12(5), e0178392. doi:10.1371/journal.pone.0178392

APPENDIX H

PARTICIPANT DEMOGRAPHIC FORM

Participant ID: _____

Age (please tick):	16-20 <input type="checkbox"/> 21-25 <input type="checkbox"/> 26-30 <input type="checkbox"/> 31-35 <input type="checkbox"/> 36-40 <input type="checkbox"/> 41-45 <input type="checkbox"/> 46-50 <input type="checkbox"/> 51-55 <input type="checkbox"/> 56-60 <input type="checkbox"/> 61-65 <input type="checkbox"/> 66-70 <input type="checkbox"/> 70+ <input type="checkbox"/>
Education (please tick highest qualification):	GCSEs or equivalent <input type="checkbox"/> NVQ <input type="checkbox"/> A-Levels or equivalent <input type="checkbox"/> Bachelor's degree <input type="checkbox"/> Post-graduate degree <input type="checkbox"/>
Employment status (please tick):	Unemployed <input type="checkbox"/> Long-term sick <input type="checkbox"/> Part-time <input type="checkbox"/> Full-time <input type="checkbox"/> Retired <input type="checkbox"/>
Previous breast cancer (please tick):	Yes <input type="checkbox"/> No <input type="checkbox"/>
Time since diagnosis of breast cancer (if applicable):	_____
Time since end of treatment for breast cancer (if applicable):	_____
BRCA 1/2 status	Positive <input type="checkbox"/> Negative <input type="checkbox"/> Unknown <input type="checkbox"/>
Previous other cancer (please tick):	Yes <input type="checkbox"/> No <input type="checkbox"/>
Family history of breast cancer (please tick):	Yes <input type="checkbox"/> No <input type="checkbox"/>
Family history of other cancer (please tick):	Yes <input type="checkbox"/> No <input type="checkbox"/>
Risk-reducing surgery (please tick):	Contralateral <input type="checkbox"/> Bilateral <input type="checkbox"/>

APPENDIX I

INTERVIEW GUIDE WITH REFINEMENTS

Please note: Italics denote refinements.

Interviews will be at a time convenient for the patient. The interview will be conducted at the hospital, at the patient's home or by telephone as the participant prefers. For consistency, and to ensure that the research questions are addressed, a semi-structured approach will be used, with participants prompted to address specific topics in each interview.

Closed questions will be avoided as much as possible as these constrain the information gained from the participants. For this reason, interruptions from the interviewer will be kept to a minimum with interviewer dialogue limited to reflecting, prompting and summarising, with open or closed questions and probing where necessary. To avoid generalised responses, participants will be encouraged to speak about their specific experiences as much as possible.

Before the interview commences it is essential to ensure that the patient has read the information sheet, and that they have made a decision and do not plan to revisit it.

Questions and prompts below are resources on which the interviewer will draw and only relevant questions should be asked. Questions should not be imposed to disrupt conversational style.

1. Introduction.
2. Reassurance of confidentiality (including reassurance that their doctors and nurses and other clinicians will not be told what the participant has said and that their treatment will not be affected by what they say, unless they say something that indicates risk).
3. Clarification of research aims.

4. Elicit and answer questions about the interview process and the Patient Information Sheet.
5. The interview questions will be guided by the structure below. The questions are illustrative and the format and sequencing will be guided by the patient's responses.

1. Finding out about RRM:

- When did you first hear about RRM?
 - Ask whether before diagnosis/illness.
- How did you first hear about RRM?
 - Explore with further questioning if aware of RRM due to family or friends, media, own research, professionals, etc.
 - *What did you draw from this information? How did you use it?*
- If it was before experience of cancer: At that time, what did you think about RRM?
What was your reaction?

2. RRM as an option:

- When did you first consider RRM as an option for you? / Did you ever consider RRM as an option for you?
- Who thought about the option of RRM first?
 - Enquire about specific times and people, e.g. where were you?
 - What was on your mind at the time? / what did you think?
- *What did it change, when the RRM decision was made?*
- *Enquire further if their decision led to a sense of relief.*
- *What do you mean by 'taking back control'?*

3. RRM decision-making:

- Tell me about how you came to the decision [prompting any specific topics]

- *How did you make that decision?*
- *What did making the decision/the decision itself do?*
- *If worry/peace of mind is mentioned, ask what would the worry be about?*
- What factors were going on in your life at the time that influenced whether you wanted/not wanted to have RRM?
- Who did you talk to about it? What did you tell them about it?
- Where other preventative options discussed with you? Did you consider other preventative options?
 - *Enquire about specific times and people, e.g. where were you?*
 - *What did you think at that time?*
- If they did consider other preventative options but changed their mind: Could you describe what/who made you change your mind? *Any reasons if decided against them?*
- After your decision, were there any moments of doubt? e.g. anything you learned/someone said that made you question your decision? Tell me about it.

4. Views of RRM & other preventative methods:

- Have you encountered anything in your life that may have similarities with your decision about RRM?
- How have you explained what RRM is to other people/to people who were not familiar with it?
- What was it about RRM specifically that made you decide to have it/not to have it?
- Is there anything unique or different about RRM compared to other options?
- Has your view on that changed over time?
- What was it about [other preventative option] specifically that made you decide to have RRM/not to have RRM?

- *Depending on response: What was it about other methods that were not as suitable or as convincing for you?*
- Did you seek the opinion of other people about RRM? Why?
 - If sought other people's views: Whose and what did they say? How did this influence your opinion of RRM?
- If they say it was 'the obvious thing to do'/'a no-brainer': Why did you consider RRM as 'the obvious thing to do'/'a no-brainer'? What lead you to consider RRM as 'the obvious thing to do'/'a no-brainer'?
- Imagine you had not been able to have RM, what would this mean for you?
- Has your view/opinion of RRM changed at any point, either before or after you made your decision? If so, what person/event influenced that?
- *Enquire about fear of future regret or worry of future regret influencing decision-making or RRM perception.*
- *Ask about risk perception, e.g. what risk is left to them after the operation?*

5. Final questions:

- What do you think was the biggest influence in reaching your decision? Why?
- How easy or difficult was it to make the decision? Why?

6. Ending:

- Does the participant have any questions or any concerns about anything that has been talked about?
- Thank participant for taking part.

APPENDIX J

DATA PROTECTION PROTOCOL

Demographic information will be pseudo-anonymised (to link with interview transcript) and all potentially identifying information will be removed/ replaced with a code. Information will be stored electronically at The University of Liverpool on a secure password protected computer system. Research team members only will have access to the transcripts during analysis. Following transfer to the computer, raw data (paper files e.g. demographic information) will be stored in the D.Clin.Psychol. office.

Interviews will be recorded on a digital dictaphone; audio data will be transferred on to a university password protected computer as soon as possible. Following transfer to computer, audio data stored on the dictaphone will be deleted.

A university approved transcription service will be responsible for most transcriptions. Emma Shaw Núñez (Trainee Clinical Psychologist) will be responsible for ensuring that the transcription service employed during the investigation has deleted all electronic copies of the data from their computer hard drives/portable disk drives etc. sent by Emma Shaw Núñez (Trainee Clinical Psychologist). When data is part of the wider study and sent for transcription by a member of the wider research team, responsibility for data deletion will fall to the data custodian, Dr Steve Brown (Chief Investigator/ supervisor).

Transcriptions will be pseudo-anonymised with respect to all names of people and places and other potentially identifying information. Transcripts will be stored electronically at The University of Liverpool on a secure password protected computer system. Research team members only will have access to the transcripts during analysis.

Emma Shaw Núñez (Trainee Clinical Psychologist) will retain an electronic copy of all transcribed files for her own records until the point of the viva voce examination. Up-to-date copies of all electronic files (relevant to the thesis) will be stored on researcher's allocated

workspace on the University computer network. Transcribed files will not be kept on any home PC.

Following the viva voce examination, Emma Shaw Núñez (Trainee Clinical Psychologist) will submit hard data to the D.Clin.Psychol. office for secure destruction by the University Records Management Service.

All the electronic data relating to the thesis will be archived on a CD. The CD containing the electronic data and any remaining copies of the raw hard data, such as participant consent forms, will be given to the data custodian, Dr Steve Brown (Chief Investigator/ supervisor), who will be responsible for data storage. Transcripts will be archived with the Economic and Social Data Service for access by other researchers (data sharing procedures are explained in ESDS (2012) Managing and Sharing Data (p.23) <http://www.data-archive.ac.uk/media/2894/managingsharing.pdf>). This will be explained to patients and they will be asked to consent to it.

It is the responsibility of the data custodian to ensure that all hard and electronic copies of any data files that relate to the major research project at the end of the data retention period have been securely destroyed.

APPENDIX K

DEALING WITH DISTRESS PROTOCOL

Within the information sheet, participants are informed that they do not have to take part in the study and are free to withdraw at any point without negative consequences for the participant. The researcher will reiterate this information as regularly as is felt necessary.

If a participant becomes distressed during the course of the interview (visually appears distressed or informs the researcher that this is the case), the interview will be paused and the participant will be asked if she wishes to continue, or if she wishes to interrupt or stop the interview and/or if she wishes to discuss her concerns with the researcher.

If the researcher is concerned about the degree of distress, the participant should be encouraged to contact their GP or mental health provider or, with the participant's consent, the researcher will ask a member of the research team to do so. The researcher will also provide the participant with contact details and information about MacMillan Cancer support (0808 808 00 00 Mon-Fri 9am – 8pm, or online community), Breast Cancer Care (0808 800 6000 Mon-Fri 9am-5pm, Weds 9am-7pm, Sat 9am-1pm) and/or Samaritans (0151 708 8888 Liverpool and Merseyside; or 116 123). With consent, the researcher will inform the Clinical Psychologist from the Liverpool Cancer Psychology Service of the distress experienced.

If the researcher is concerned that the participant is at risk of harm, or that someone else is at risk, she will seek her consent to refer these concerns to an appropriate clinician. However, the duty of care would mean that, even if consent were not provided, the researcher would need to make this referral as appropriate.

If the participant feels able and the researcher agrees, the interview will be resumed. With consent, the participant will be contacted the following day. The participant will be encouraged to contact the research team if she experiences increased distress in the hours/days following the interview. Contact numbers for the research team are supplied on the

participant information sheet.

If a participant becomes distressed the researcher will record the action taken.

If the participant feels unhappy with the interview process and wishes to complain, the participant will be directed to the contact details for the Customer Relations Team on the participant information sheet.

APPENDIX L
LAY SUMMARY OF RESULTS FOR PARTICIPANTS



Dear _____,

I would like to thank you again for taking part in the research project

**Patient and surgeon decision-making in risk-reducing mastectomy:
an ethical and empirical analysis**

As part of this project, and of my Doctorate in Clinical Psychology research, we carried out interviews in 2016 and 2017 with women who had opted to have a risk-reducing mastectomy (RRM). I am writing to provide you with a summary of the results. I hope you find this summary and the results of interest:

Why did we do this study?

- Breast cancer is the most common cancer in the UK. Cancer Research UK estimate that 1 in 8 women are diagnosed with breast cancer in their lifetime.
- There is research suggesting that many factors can play a role in how women decide to have a RRM, including psychological factors. For example, decisions can be influenced by perceptions of risk of developing breast cancer or by cancer-related worry.
- During the first part of this study (carried out in 2013) we found that patients decided to choose RRM because they felt vulnerable to breast cancer, and that many patients made immediate decisions about wanting to have a RRM. This was because they wanted to do all they could to eliminate risk.

We aimed to find out more information about what women thought about RRM as a convincing option, and about how they reached their decision to have a RRM.

How did we do this study?

- Each participant was interviewed about their own experiences of deciding to have a RRM. All participants had in common that they were over 18 years old, they could speak English fluently, they had reached a decision to have a RRM, and they had attended a consultation with a clinical psychologist about this.
- To date we have interviewed 7 participants; with the data that we have collected we have found some helpful results. We will continue to interview more women if possible, which will help us to confirm the results we have found.

What were the results of the study?

In total, 2 participants had already had a RRM and 5 were waiting for it. Three of the participants had a known genetic mutation (BRCA1/2 or CHEK2). Five had had breast cancer and two had not. All participants had either close family members or friends who had suffered cancer.

Analysing the content of the interviews, we found that:

- Participants found out about RRM in many different ways, including through family members, through media, or through other patients. Most participants had made their decision that they wanted a RRM before the option had been suggested by clinical staff.
- Although participants had different personal and family experiences of cancer, and although they had first learned about RRM in different ways, how they made their decision to have a RRM was similar because they decided in an immediate and determined way.
- Participants described cancer and the treatment of the illness (e.g. chemotherapy, radiotherapy) as something they understandably wanted to avoid. Wanting to avoid future breast cancer and future treatment was a strong reason to opt for RRM.
- RRM was an option that gave participants a sense of relief and of safety from fear, because it reduced their worry of developing cancer and it freed them from the uncertainty surrounding scans and tests.

- RRM was an option that enabled participants to take control, because they had felt disempowered by the illness and treatment. It was also an option that protected participants from regretting not having done all they could in the future. This was related to wanting to survive to be there for their loved ones.
- After making their minds up to have a RRM, women had to wait a period of time for it to take place (as part of the usual clinical pathway), and this involved discussing the decision with clinical staff and with other people in their lives. During this time, participants reviewed their decision, but did not change their minds that RRM was the best option for them.

Why is this study important?

It is important that health care staff working with women who are at risk of breast cancer and opt to have a RRM understand how and why these decisions are made. This includes understanding what women think about RRM and why they perceive it to be a convincing option. It also encompasses understanding that the RRM decision is often immediate, and that this might be related to psychological factors such as emotions.

This is a small study still in progress, but nevertheless it adds new information to the growing body of evidence in this field of research.

**We would once again like to express our gratitude for giving your time to take part
in this research – thank you!**

Contact us:

If you would like any further information about this study, please feel free to contact us.

- Before 30th September 2017: please contact Emma Shaw Núñez (Trainee Clinical Psychologist, D.Clin.Psychol. Programme, The University of Liverpool) via email on emmasn@liverpool.ac.uk
- After 30th September 2017: please contact Dr Steve Brown (Senior Lecturer, The University of Liverpool) via email on slbrown@liverpool.ac.uk

APPENDIX M

INSTRUCTIONS FOR TRANSCRIBER

TRANSCRIPTION GUIDANCE

Thank you for doing interview transcriptions for this research project. The key points below are guidelines to help you whilst doing this. It will be important to follow these in order to comply with ethical standards. If you have any queries, please do not hesitate to contact me on emmasn@liv.ac.uk

- ✓ The interviews in this research are with women who have reached a decision about whether to have a risk reducing mastectomy or not. On occasions they might discuss topics which might be upsetting to hear, including the consequences of suffering cancer or the reasons why they made their decision. If you do find an interview upsetting, please remember that you can stop transcribing if you wish to do so. You can also contact me via the email address above if you want to discuss this any further.
- ✓ It is likely that you might encounter some terms or words which are specific to the area of research of this project. Some of them might include:
 - *RRM or risk reducing mastectomy*: Surgery to remove the tissue of one or both breasts.
 - *BRCA1 and BRCA2* [pronounced "bracka"]: Gene mutation that might increase the risk of developing breast cancer.
 - *CHEK2*: Gene mutation that might increase the risk of developing different types of cancer.
 - *Tamoxifen*: a drug used to treat breast cancer.
 - *Oophorectomy*: the surgical removal of the ovaries.
 - *Lumpectomy*: the surgical removal of a suspected cancerous tumour from a woman's breast.
- ✓ Please use the transcription template provided. It is a word document formatted in advance for your convenience and to ensure that all transcriptions follow the same standard format. A copy of it will be saved in the encrypted memory stick.
- ✓ In order to protect participants' confidentiality, it is important to not transcribe any personal identifiable information, including names, addresses, locations, very unique professions, etc. When transcribing the interview, please substitute these terms with generic ones. For example:

- John = [name]
 - Dr Doe = [doctor] or [staff]
 - London = [city]
 - Template University Teaching Hospital = [hospital]
-
- ✓ If in doubt, please do not transcribe the information. Instead, substitute it with a generic term and highlight it and noting the minutes of the audio file, such as [profession] 0:34:11. This will allow the researchers to locate it in the text in order to decide whether to include it in the transcript or not.
 - ✓ Similarly, if there is a section of the audio recording which is unintelligible or inaudible, please mark this in the text by highlighting it and noting the minutes of the audio file, for example [inaudible?] 00:34:11
 - ✓ Please do not save the audio recordings or transcripts in any other computer, drive or external memory device. They should only be saved on the encrypted memory stick provided.
 - ✓ Please keep the password for the encryption software in a safe place, do not share it with anybody else and do not store it with the memory stick.
 - ✓ Once you have transcribed an interview, please let me know the number of hours that it has taken you to do this work. I will confirm that the transcript is saved in the encrypted memory stick and then email the finance department with a payment memo for your work. I will aim to do so within the same day.

Thank you again for your help transcribing interviews for this research project.

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APPENDIX N

METHODOLOGY

The aims of the study required a primarily inductive approach, for to date there have been scarce theoretical studies examining the influence of affect and emotions on how patients view RRM and decide about its uptake; thus, a qualitative method was followed. The qualitative approach to analysis and interpretation is explained in the main body of the thesis, supplemented with information below. This method of qualitative analysis is based on previous research conducted by the study investigators (Wright, Holcombe & Salmon, 2004; Salmon, Mendick & Young, 2011; Brown et al., 2017).

Epistemology and Data Analysis

Qualitative pluralism proposes the combination of multiple epistemological and/or methodological frameworks for the understanding of experiences, as required by the data and emerging analysis (Frost, 2011). The use of pluralist qualitative methodologies is increasing, with guidance about how to conduct pluralist qualitative research ensuring rigour and validity increasingly available (e.g. Frost, 2011). A pragmatic, constant comparative approach was followed throughout the analysis, whereby analytic categories were developed both descriptively, in relation to the content of patients' interviews, and theoretically, addressing the meaning and function of speech in context.

The study aimed to understand patients' views of RRM and gain further insights about their decision-making. Data analysis began descriptively, an approach that can be situated within a largely positivist position, initially drawing from a thematic analysis approach in order to identify recurrent subjects and foci in participants' accounts. An example of this was the identification of the expressions pertaining to 'losing and taking control', 'taking action', and 'doing all they can', which led to this recurrent feature to be investigated further in

subsequent interviews and informed the finding that RRM was seen as a proactive choice in the face of the loss of control experienced due to breast cancer and its treatment.

The analysis became more interpretative as the themes or analytic categories were developed, which enabled the data to be considered in the context of the interview as a whole, in the context of previous interviews, within the wider study, and within broader social contexts. Drawing from interpretative analytic approaches, adopting a social constructionist position, enabled the data analysis to progress further. The content, meaning and function of participants' accounts were, therefore, carefully considered and guided the analysis, as did the awareness of the limitations of interviews for data collection. It was recognised that interviews carry the danger that past events, views and processes might be misremembered or reconstructed for congruence with current circumstances and perspectives. Furthermore, the potential tendency of interviews about emotive topics to elicit justifications rather than explanations for behaviour was acknowledged. In this respect, interviews were not regarded as necessarily providing direct access to participants' experiences and intentions, and the interpretative approach shaped the analysis. An example of the above was the hypothesis that RRM, in that it removes most of the tissue which can harbour the risk of breast cancer, was viewed as a more reassuring procedure than other risk-reducing options, e.g. healthy lifestyle changes, the risk-reducing outcomes of which were conceptually not as straightforward.

Whilst the analysis was developing and new interviews were carried out, previous interviews were regularly revisited, 'cycling' between the emerging findings and the data to test, refine and elaborate the framework of findings; this was achieved by examining commonalities across the data whilst attending to heterogeneity and divergence too. This approach enabled the identification of pivotal cases or outliers, which were highlighted and

discussed in the analysis; a pertinent example was the account of one participant who described a period of doubt about her decision to opt for RRM.

Data analysis was planned to finalise when theoretical saturation was reached. However, the rate of participant recruitment limited the number of interviews conducted and there remains the question whether theoretical saturation has been confidently reached. For this reason, participant recruitment and data analysis is planned to continue beyond the write up of the findings presented in this thesis.

Reflexivity

Personal and epistemological reflexivity are emphasised as important features of qualitative research. The researchers acknowledge the impossibility of remaining ‘outside of’ the research being conducted, holding an awareness of how their own values, experiences or beliefs have shaped the research, and of how the study and methods used might have defined and limited the data and the findings (Willig, 2008).

In this respect, prior to the commencement of the study, the researcher did not have any personal or professional experience with breast cancer. However, when the analysis was being completed, the researcher worked as a trainee clinical psychologist in a psycho-oncology service and conducted RRM consultations with patients requesting this procedure. It is possible that the researcher’s stances of positive regard and of respectful curiosity towards people affected by cancer may have impacted upon the interactions with participants. Additionally, the interviewing researcher being a woman might have also influenced participants’ interactions during interviews. Another example pertains to the limitations of the methods of data collection, which have been discussed previously.

Validity

Standards by which the developing analysis was assessed included theoretical and catalytic validity, i.e., potential utility for practice and research (Kincheloe & McLaren, 2000). This was achieved by considering the possible implications of the findings for clinical practice and for further research.

Trustworthiness

Issues of quality in qualitative research have been approached with a focus on achieving trustworthiness through the conduct of this study, with particular attention paid to the following four criteria, as suggested by Guba (1981) and Shenton (2004):

Dependability. In order to make the research more dependable, the processes followed have been reported in detail. This includes the research design and its implementation, how the data was gathered, and how it was analysed. Records of data analysis meetings have been kept, as have documents which were refined throughout the study, providing an audit trail that is available for review. Examples can be found in Appendices P and Q.

Confirmability. To ensure as far as possible that the study's findings were derived from the data rather than correspond with the preferences of the researchers, care has been taken to explicitly recognise potential biases. This study was preceded by another investigation which was part of the wider research project (Brown et al., 2017). In light of the potential for the current study to pursue a biased convergence with these initial findings instead of expanding the conclusions, the researchers engaged in continuous efforts to falsify and corroborate the emerging analysis. Additionally, the researchers have provided an in-depth methodological description to allow the integrity of the research results to be scrutinised.

Credibility. The following provisions have been made to increase the credibility of the study and of the findings:

- a. The procedures followed for this research, including the method of data analysis, were derived from those previously and successfully used in comparable studies.
- b. The researcher aimed to establish rapport with participants from the beginning. Participants were encouraged to be open and honest, for example by being assured that there were no right answers to the questions and that the researcher was interested in their particular experiences.
- c. Peer scrutiny of the study: the researcher presented and discussed the project to colleagues in academic settings, which encouraged the inclusion of a detailed explanation of the research design and of the method of data analysis.
- d. Background and experience of the investigators: the credibility of the researcher and of the wider research team is important. All researchers had prior experience of conducting research in clinical settings. In particular, the trainee clinical psychologist was experienced in the conduct of semi-structured interviews and had extensive experience of working individually with people to gather information about sensitive topics in a respectful manner.
- e. Previous research findings and published literature have been examined, in order to relate the results of this study to the growing body of knowledge.

Transferability. The generalisability or transferability of the findings to other settings has been attempted through the description of relevant characteristics of study, including information about the data collection, about the participants, as well as about the wider context in which the findings are situated.

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APPENDIX O

EXAMPLE BRIEF NARRATIVE OF INTERVIEW

For further details see data CD.

Interview 1:

This participant was a middle-aged woman, mother of three teenagers, in a relationship with a supportive husband, and was a carer for her sister. She had previous history of breast cancer, and her diagnosis of breast cancer two years ago had been due to a series of fortuitous events, including a fall that required medical attention. She has nursed and lost several family members to cancer in the past, including her mother, her brother and her aunt. She reported not hesitating to have a contralateral RRM because she wanted to know that she had done all she can to get rid of the risk of cancer and to be there for her children for as long as she can, describing her children as her 'driving force'.

APPENDIX P

EXAMPLE OF INITIAL ANALYSIS FOR AN EMERGING CATEGORY

April 2017

<p>Category: Safety from worry</p> <p><u>Possible Subthemes</u></p>	<p>Quotes: <u>Safety from worry</u></p>
<ul style="list-style-type: none"> • Being at risk of breast cancer leads to heightened worry. • Feeling at risk leads to wanting to do something to feel safe. • Other risk management alternatives would not get rid of worry. • Consideration about other risk managements alternatives was very brief. • Consideration about other risk managements alternatives might happen after RRM decision. • Eliminating uncertainty only achieved with RRM. 	<p>ID06: when I went to the genetic counselling woman and she said there's chemo like preventative chemotherapy and high risk screening that very quickly for me I thought well then you're living that same shit every single year – pardon my French – you're going to go through that every year. Are they going to find anything this year? So that ... I never even thought about it.</p> <p>---</p> <p>ID05: Well exactly because even though it would make a little difference - it might only be a slight difference - that's a slight difference less chance of me getting breast cancer again.</p> <p>---</p> <p>ID05: I'm thinking I can have yearly mammograms and have that thought in my mind all the time or every time I go for my mammogram I'm going to be worrying are they going to find something or I can have a mastectomy and I'm never going to have to worry about that again am I?</p> <p>---</p> <p>ID05: I just think I need to do ... I just need to do it for my own peace of mind as well as everybody else's as well.</p> <p>---</p> <p>ID04: I can't have things hanging over me, and particularly the fact that it could come back. I couldn't live with that ... and not do anything about it.</p> <p>---</p> <p>ID04: Because if you get over the first one and you've still got a boob and the chances are that maybe 5 years down the line it's going to pop it's head up again, like I say, you don't know at what stage where it is in your body and for me that risk is not worth taking so if I was talking to you I would say to you get well, get over it and consider it because the other option is just horrendous.</p> <p>---</p> <p>ID04: There's women presenting over 70 with breast cancer and I thought I'm not waiting to go down for a mammogram again and have a recall. I've been through all that most of my life and I thought I don't want that again so let's have that out the way.</p> <p>---</p> <p>ID03: so I was about 57 when I got the results. She [nurse] felt that I didn't need it but then the genetics result was a 30–50% chance of developing cancer, which I thought was still quite high. And of our family there are actually only 4 ladies in our family who've got/developed breast cancer. Two were young and two were my age so I don't feel I'm out of the risk region.</p> <p>---</p> <p>ID02: And there was a period of quite a lengthy spell from when I found the lump to getting any treatment or having it acknowledged, and I thought that uncertainty for me was more worrying than the op, I could've dealt with the op quite quickly, get it all over and done with</p> <p>---</p>

For further details see data CD.

APPENDIX Q

EXAMPLE OF CONCEPTUAL CATEGORIES IN DATA ANALYSIS

For further details see data CD.

1. **“So it was just immediate that I made that decision”**

- Descriptive finding, explaining context of how the decision was reached.
- Homogeneity in the decision immediacy and the degree of certainty reported by participants.
- RRM is simple conceptually, e.g. breast tissue removed = breast cancer cannot occur.
- Decision-making moments were difficult to access in detail, i.e. how, not why, the decision was reached in that particular moment.
- This might explain the decision being immediate or intuitive.
- Theoretically it could be that the timeline would divide into 2 – weighing things up before the decision, and justifying it after, but pre-decision timeline is very short → Continue to try to expand the first part of the timeline in future interviews.

2. **Relief and safety**

- RRM perceived as a way to terminate the immediate worry they are living with.
- Worry related to the illness returning. Safety related to not having this worry always accompanying them.
- Worry related to not having done ‘all they can’ to protect themselves and their loved ones.
- Women did not express worry after RRM about cancer metastases.
- Once RRM decision is agreed with surgeon, this results in relief/feelings of safety.
- If women knew that they could have RRM, the worry decreased and it was sufficiently tolerable so as to postpone the operation without an increase of worry or sense of vulnerability.
- Relief/Safety related to the elimination of uncertainty (e.g. about risk of developing BC, about difficulties in diagnosing it, etc.).
- RRM perceived as a more certain option than other alternatives.
- Other risk management strategies would not get rid of the worry to the same degree.