

THE UNIVERSITY OF HULL

**Women's experiences of family planning and prophylactic mastectomy following
BRCA gene diagnosis**

Being a thesis submitted in partial fulfilment of the requirements for
the degree of Doctor of Clinical Psychology
in the University of Hull

By

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Overview

This portfolio thesis is comprised of three parts: a systematic literature review, an empirical report and supporting appendices.

Part one is a systematic literature review which explores experiences women have of making family planning decisions following BRCA gene diagnosis. A systematic search of five databases found five papers suitable for review. Themes and subthemes are identified and discussed, along with a review of the methodological quality of the reviewed papers. Implications for clinical practice and future research are discussed.

Part two is an empirical report of an original piece of research exploring women's experiences of undergoing prophylactic (preventative) mastectomies following BRCA gene diagnosis. Data was collected via semi-structured interviews with each of the three participants. Interpretative Phenomenological Analysis (IPA) was carried out, with four themes identified and discussed in relation to each individual participant interview. Implications for future research within this population are discussed, as well as implications for clinical practice.

Part three is a collection of combined appendices for both the literature review and empirical report, including an epistemological statement and a reflective statement from the researcher.

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Part One

Systematic Literature Review

**Women's experiences of family planning after BRCA gene diagnosis: A systematic
literature review**

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Abstract

Purpose: To review the literature exploring how women diagnosed with the BRCA genes navigate and make decisions regarding the process of family planning.

Methods: A systematic search of the relevant literature was conducted using EBSCOhost search engine to search several databases. The relevant literature was searched and selected based upon inclusion and exclusion criteria. Papers included for review were assessed for quality and analysed using a narrative synthesis approach to identify themes, similarities and differences amongst the papers.

Results: Four overarching themes were identified within the papers: Priorities, Pressure and Time, Legacy and Hope. These themes were then broken down into the following subthemes: Cancer Management vs Family Planning, External Pressures, Urgency and the Ticking Clock, (Un)certainty, Family History, A Sense of Responsibility, Optimism for the Future and Being more than BRCA.

Conclusions: Women are greatly impacted upon by their family histories of cancer and their sense of pressure and urgency to make life-altering decisions about family planning. More research is needed to explore what this experience is like for couples, as well as individuals. This review has implications for future research in this area as well as implications for the development of support services for women navigating these difficult decisions in a limited timeframe.

Introduction

The presence of the breast and ovarian cancer susceptibility genes, BRCA1 and BRCA2, puts carriers at a 56-87% lifetime risk of developing breast cancer and a 10-60% lifetime risk of developing ovarian cancer (Heshka et al, 2008). Genetic testing for these genes is beneficial for identifying heightened risk (Lerman, Croyle, Tercyak & Hamann, 2002; Heshka et al, 2008), and 95% of women in families with hereditary breast cancer will seek genetic testing. Women's perceived risk of cancer (Struewing et al, 1995; Lipkus et al, 1999) and anxiety relating to the development of cancer (Andrews et al 2004, Foster et al 2004, Lerman et al 1997) have shown to influence test uptake. Women already affected with breast or ovarian cancer are also more likely to get tested for the susceptibility genes (Andrews et al 2004, Julian-Reynier et al 2000, Lee et al 2002, Lerman et al 1996, Jacobsen et al, 1997), showing that family factors can motivate testing, making women with children (Meijers-Heijboer et al 2000) and women with first degree relatives affected by cancer (Lerman et al 1996) more likely to seek testing. The testing process, as well as the outcome and results, have been found to increase distress and anxiety in some patients (Heshka et al, 2008), which could be due in part to the impact a positive test result can have on people's lives. One aspect of this impact is the decisions women have to make regarding the management of this diagnosis. Options offered to carriers of the gene to reduce cancer risk include prophylactic (preventative) mastectomy, which can unilaterally involve one breast, or bilaterally both breasts, and oophorectomy, the removal of the ovaries. Research suggests that 12 months after testing, between 0 and 51% of women have mastectomies and 13-65% have oophorectomies (Heshka et al, 2008), lowering the lifetime risk of breast and ovarian cancer by up to 90% (Frost et al, 2000; Hatcher, Fallowfield & A'Hern, 2001).

Not only does BRCA gene diagnosis leave women required to make decisions regarding health management and cancer prevention, it also leaves women with questions about their futures, which for some may entail decisions to be made regarding family planning. Carrying the BRCA gene can have implications for fertility amongst carriers (Finch et al, 2013; Lin et al, 2013; Wang et al, 2014), and although difficult decisions to pursue oophorectomy can reduce cancer risk by up to 80% (Friedman et al, 2012), it means women are then unable to conceive biological children. Aside from issues relating to fertility, women with preserved fertility are still required to make decisions regarding family planning which are inevitably impacted upon by their diagnosis, as being a carrier creates a 50% chance of passing on the gene to any biological children (Vadaparampil et al, 2009). These difficulties relating to navigating decisions about childbearing can lead some women to decide not to pursue having biological children at all (Smith et al, 2004). Meanwhile, others may pursue alternative options such as adoption or pre-implantation genetic diagnosis (PGD), which offers women the opportunity to have biological children who are not carriers of the BRCA gene (Fortuny et al, 2009).

Although previous literature reviews have investigated issues relating to things such as communication about BRCA in families (Young et al, 2017) and the impact of BRCA diagnosis on reproductive function (Jegu et al, 2015; Daum, Peretz & Laufer, 2018) and fertility (Daum, Peretz & Laufer, 2018), at the time of writing the researcher was not able to identify any literature reviews focused upon qualitative research and data relating to women's experiences of making family planning decisions after BRCA diagnosis. However, a review of that nature seemed an important addition to the research landscape as it relates to the BRCA population, as no paper had yet combined the qualitative findings to review women's experiences in their own words. This review therefore aimed to understand women's experiences of making family planning

decisions after BRCA diagnosis by reviewing qualitative research papers relating to the subject. The research question for this review was:

“What are the experiences of family planning for women diagnosed with the BRCA gene?”

Method

The process of this systematic review was carried out using specific guidelines and following a stepwise process to complete the review (Popay et al, 2006). This process of systematic review included the following steps:

1. Searching and mapping relevant evidence to develop a relevant scope for the review and to assess its need.
2. Specifying the review question, ensuring it is both relevant and answerable.
3. Identifying studies for inclusion in the review based on specific selection criteria.
4. Extracting detailed data from the papers included carrying out individual assessments of methodological quality.
5. Synthesising data through narrative synthesis to draw conclusions about the reviews findings.

Search Strategy

The review question developed from the mapping of relevant literature was as follows:

What influences family planning decisions of female BRCA carriers?

A search of the relevant literature was conducted between January and February 2019.

The literature search service EBSCOhost was used to search the following databases:

PsycINFO, PsycARTICLES, MEDLINE, Academic Search Premier and the Cumulative

Index to Nursing and Allied Health Literature (CINAHL Complete). These databases were selected to ensure that literature published under domains relating to both physical health and psychology were captured, as well as literature published on the topic within journals from other fields. The search terms used were as follows:

Women OR female**

AND

BRCA

AND

family planning OR family-planning OR child OR bab* OR reproduc* OR pregnan**

These search terms were selected as they remained broad enough to reduce the likelihood of eliminating literature due to the use of narrow terminology. Words such as “fertility” were not included in the search terms, as during pilot searches this generated vast amounts of literature relating to fertility treatments and the biological underpinnings of fertility in cancer populations. The * was used to broaden search terms and ensure plurals or variations of words were captured in the searches.

Selection Strategy

The search terms used returned 951 papers from the following databases: MEDLINE (436); Academic Search Premier (301); CINAHL Complete (169); PsycINFO (43); PsycARTICLES (2). After duplicates were removed, 768 papers were left for review. Papers were reviewed based upon their title, and 39 papers were identified as suitable for further review. These papers were then assessed based upon their abstracts, and papers were excluded at this stage based upon the following criteria: the paper focused on medical issues relating to fertility; participants all had cancer; the papers were

reviews themselves; the papers had a focus on how individuals share family planning information with family; did not meet the inclusion criteria. A flowchart to depict this process of selection can be seen in Appendix B.

The inclusion criteria for studies were as follows: a) The majority of participants must be women with a BRCA diagnosis b) Methodology and data must be qualitative c) Data must be gathered via interviews. The exclusion criteria were as follows: a) The majority of participants must not have been previously or currently diagnosed with cancer b) The paper must not be a review itself to ensure women's experiences were captured, therefore reducing the inclusion of secondary data and reviewer's interpretations of primary data. These inclusion and exclusion criteria were designed to ensure that the papers included would be appropriate to answer the research question, which focused upon women's experiences of family planning following BRCA diagnosis, and which focused only upon the impact of genetic diagnosis on this and not upon the impact of cancer diagnosis. Only studies which gathered qualitative data via interviews were considered for inclusion as this review aimed to look only at research which allowed women to discuss their experiences in their own words, without the confines of a questionnaire or other qualitative methodology.

The reference lists of the selected papers were hand-searched for further literature to be included in the review, however no suitable papers were found. Key words were entered into the Google Scholar search engine to capture any literature not captured via EBSCOHost searches. These key words included "family planning" and "BRCA", however these searches did not return any further papers not already included in the review. Overall, five papers were included in the review.

Quality Assessment

Data was extracted from the papers using a bespoke data extraction tool designed by the lead researcher (See Table 1). This tool was designed to collect and organise demographic and methodological information relating to each study in one place and to enable comparisons to be made. This tool was limited in its ability to serve as an analysis tool or as a method for assessing quality but served to bring information together in a way that made later methodological quality analysis easier to conduct without the need for repeated references back to each individual study. Two quality assessment tools, the Critical Appraisal Skills Programme (CASP) Qualitative Checklist (CASP, 2018) (see Appendix C) and NICE Qualitative Checklist (NICE, 2012) (see Appendix D) were used to assess the quality of the papers included for review. These measures were each used to generate a quality rating score for each paper, which was converted into a percentage. These two scores were then combined to make an overall percentage quality rating score, which was then used as the overall quality assessment score for each individual paper. The decision was made to use two quality assessment tools which looked at similar factors to ensure validity in the quality assessment process, in part because the NICE assessment tool is more thorough than the CASP. Although both tools address similar issues, such as whether or not the research methodology is appropriate to meet aims, the NICE tool consistently offers more in-depth prompts and questions for each criteria. For example where the CASP tool asked “Is a qualitative methodology appropriate?”, the NICE tool asks several questions relating to appropriateness of qualitative approach, clarity of what the study seeks to do, and the defensibility and rigor of the design and methodology. The NICE tool also addresses criteria not addressed by the CASP tool, such as whether or not the data is rich, and whether or not the research context is clearly described. Using two tools of assessment helped to provide confidence in the scores reached by clarifying that the same strengths and weaknesses were recognised in each study regardless of assessment

tool. Certain percentage scores awarded for methodological quality from the CASP tool were consistently awarded alongside certain scores awarded by the NICE tool. For example, a 70% score on the CASP tool always occurred alongside a 78% score on the NICE tool. The maximum difference between two percentage scores awarded by the two tools was 14%, and the minimum 5%.

This assessment process was repeated by a peer reviewer to assess the quality of 50% of the included papers, who arrived at scores within 5% of the scores concluded by the primary researcher. The quality rating scores assigned to each of the included papers were not used to eliminate papers from review, but provided a basis for assessing the quality of the paper's results and to offer a method of comparing the quality of papers based upon common factors.

Data Analysis and Synthesis

A process of narrative synthesis (Popay et al, 2006) was used to analyse the data in this review. This involved a process of carrying out initial synthesis of the data within each paper, which involved organising data into themes and subthemes. The next step was to assess the similarities and differences between the papers included in review and previous research, and finally, to assess the quality of the synthesised data and draw conclusions.

Results

Characteristics of Papers

Within the papers reviewed, studies took place between 2008 and 2017. Overall, 111 participants were included across all five of the included studies, with sample sizes ranging from 20 to 23. Two studies (Werner-Lin, 2008; Werner-Lin, 2010) shared the same sample of 23 participants. Both were included as they presented different results

and analysis of the data. The majority of participants were women with only one study including a single male participant (Dekeuwer & Bateman, 2013). All of the papers collected qualitative data through semi-structured interview, and all employed methods of qualitative analysis. Further demographic information of participants and methodological characteristics of the reviewed papers can be seen in Table 1.

Table 1. Completed data extraction tool showing demographic and methodological information of each paper included in review

Study (Year)	Objective	Sample Demographics and Information	Recruitment Method	Data Collection Method	Analysis Method	Conclusions	Implications Identified	Assigned Quality Rating Score
Dean & Rauscher (2017)	To investigate how women who test positive for a BRCA mutation but have not been diagnosed with cancer make	20 women Criteria: - BRCA diagnosis before completing family - Aged 18+ - Have	Recruited through distribution of flyers at cancer conference	Semi- structured interviews via telephone 24-68 minutes long Audio-	Constant comparison method	Women engaged in logical or emotional decision making. Logical prioritised personal cancer management whereas emotional prioritised family planning and having children.	Research: Research should look at why women undergo preventative surgery to reduce personal risk but do not engage in	83%

	decisions regarding family planning	committed partner - Have talked about family planning with partner		recorded		Women experienced urgency to make decisions and felt guilt about passing the gene on to future generations. They coped with this guilt by having hope for the future of medical advancements.	screening for the BRCA mutation when conceiving a child. Research should also look at providers perceptions of surgery and family planning. Clinical: Genetic counsellors and professionals	
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							should facilitate family planning discussions to explore the issues raised in this research.	
Dekeuwer & Bateman (2013)	To investigate the way in which carriers of a mutation on the BRCA1 or the BRCA2 gene make their reproductive	19 women, 1 man 30-62 years old 16 married or living with partner, 1 single, 3 divorced (2 partners were	Recruited from hospitals offering genetic testing	Semi-structured interviews Audio-recorded	Not specified	Family experiences are crucial to making family planning decisions. Participants worried about transmitting the gene to their children and also worried about their	No explicit research or clinical implications were identified or highlighted.	58%

	decisions.	included in interviews) 15 had children 5 had no children 8 had had breast cancer 1 had had ovarian cancer				children's experiences of parental or familial loss. Some participants worried about finding a partner to navigate these decisions with.		
Donnelly, Watson, Moynihan, Bancroft,	To investigate how young women identified as carrying a	25 women 18-45 years old Diagnosed prior to having children and up		Semi- structured interviews 30-120 minutes	Thematic analysis	Family history had a significant impact on family planning. Participants felt responsibility not to	Research: Further research should focus on experiences of couples, the	75%

Evans, Eeles, Lavery & Ormondroyd (2013)	BRCA gene mutation before they had children approach reproductive decision-making	to 5 years before study Over 2+ years since cancer diagnosis (if diagnosed) 8 had children since diagnosis		Audio-recorded		pass on the gene. Women struggled to balance surgery and family planning and often felt pressured to children early. They reflected on the positive aspects of their own lives and had hope for the future of medical technology.	impact of social class, and how family planning information is given by professionals. Clinical: Family planning should be addressed in genetic counselling.	
Werner-Lin (2008)	To investigate how their family	23 women 22-35 years old Criteria:	Recruited from online non-profit	Open-ended, semi-structured	The Listening Guide	The importance of time in relation to familial loss,	Research: Research should investigate the	75%

histories with cancer and their gene status inform meaning construction around cancer risk and family development for young women with elevated risk of developing hereditary breast or	-Genetic testing was 6+ months before interview -English speaking -Aged 21-35 -Not pregnant -Not diagnosed with cancer All had college education All Caucasian	support organisation and cancer support clinic at hospital	narrative interviews Telephone screening prior Interviews approx.2 hours long conducted at participants' homes Compensated		exposure to cancer and perceived time to achieve family planning goals. Participants prioritised family planning over personal cancer risk management. Women with and without partners or children had different experiences of making family planning decisions.	importance of timing of genetic testing within the life cycle. Clinical: Social workers may be able to use findings to assist with family planning decisions due to opportunities for prolonged	
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	ovarian cancer. .			with \$25 gift card			contact.	
Werner-Lin (2010)	This investigate and identify the influences of family medical histories and genetic testing on reproductive choices and examine the meanings of family	23 women 22-35 years old Criteria: -Genetic testing was 6+ months before interview -English speaking -Aged 21-35 -Not pregnant	Recruited from online non-profit support organisation and cancer support clinic at hospital	Open-ended, semi- structured narrative interviews Telephone screening prior Interviews approx.2 hours long	The Listening Guide	Family planning needed to be balanced with cancer risk management. Family history of cancer played an important role in family planning, as did the pressure felt from medical recommendations. Participants worried about the quality of	Research: Research needs to be carried out to compare the experiences of women with family history of cancer and women without. Research also needs to look at how couples jointly navigate	83%

	<p>planning and parenting in the context of genetic medicine.</p>	<p>-Not diagnosed with cancer</p> <p>All had college education</p> <p>All Caucasian</p>		<p>conducted at participants' homes</p> <p>Compensated with \$25 gift card</p>		<p>family experiences that their children would have.</p>	<p>family planning after a BRCA diagnosis.</p> <p>Clinical: Genetic counsellors and social workers should attend to childhood cancer-related grief of women when helping them to navigate family</p>	
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							planning decisions. Interventions targeting specific BRCA dilemmas need to be developed.	
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Methodological Quality

The papers included in this review share some common limitations. Several of the included papers (Werner-Lin, 2008; Dekeuwer & Bateman, 2013) did not clearly state the aims of their research. None of the included studies explored in detail the potential ethical issues that could arise during their research, nor did they reflect upon how these would be managed. Similarly, none of the included papers reflected upon the role of the research, their potential bias, how this would be managed, and what impact the researcher may have upon the research itself. In contrast, the reviewed papers shared similar strengths. All of the papers which stated their research aims showed a methodological approach which was consistent with them. When aims were not explicitly identified, the methodological approach was appropriate for the area of investigation. One paper (Donnelly et al, 2013) however not did explicitly describe the location of recruitment, which was a relative weakness of that paper. The analysis process was considered to be rigorous in four of the five papers (Werner-Lin, 2008; Werner-Lin, 2010; Donnelly et al, 2013; Dean & Rauscher, 2017), with detailed descriptions of the analysis process provided within the paper. However, one paper (Dekeuwer & Bateman, 2013) failed to provide such details, suggesting a less rigorous and robust analysis process. The data of all papers were assessed as being rich data, with all five papers providing a clear statement of findings.

Synthesis of Findings

Throughout all the reviewed papers, given the context of the reviewed research and the nature of its content, overarching themes of cancer, surgery, worry and the future were all present. These overarching themes transgressed all boundaries between the themes and subthemes outlined in the analysis of this review. The vastness of these themes and their presence across all other themes and subthemes within these papers led to the

decision to focus upon other, less vast themes and subthemes for the purposes of analysis. The data was therefore broken down into four themes for the purposes of review: Priorities, Pressure and Time, Legacy and Hope. These themes were then broken down further into subthemes, which will be discussed with reference to the five papers included in this review. These themes and their corresponding subthemes can also be seen in Table 2.

Theme 1: Priorities

Cancer Management vs Family Planning

The majority of the included papers cited that women struggled to navigate the dilemma of planning for families and surgeries at the same time, and struggled to know what to prioritise and when with regards to managing their cancer risk and deciding when, or whether, to have children (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). Some women felt a fear of diagnosis alongside a desire for a family (Dean & Rauscher, 2017) and struggled to manage caring for their personal health whilst simultaneously wanting to have children (Dekeuwer Bateman 2013).

These decisions and resulting plans, although very different in nature, were entwined for some women who felt a need to develop logical timelines to enable them to achieve both an avoidance of cancer diagnosis and their dreams for a family with biological children (Dean & Rauscher, 2017). In four of the five studies reviewed, some women prioritised their family planning decisions and desire for a family above their own health and cancer prevention measures (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). One study found that older women in committed relationships prioritised having as many children as possible before having preventative surgery, therefore prioritising having children over their personal cancer risk management, at times pushing back surgeries in order to have

babies (Dean & Rauscher, 2017). Other studies echoed this idea that women were prepared to limit their preventative options in order to have children (Werner-Lin, 2010) and would prioritise having children even when it contradicted medical advice (Werner-Lin, 2008). One study highlighted how for some a desire to proceed with family planning decisions outweighed fears for the women's personal health, and would lead women to pursue these decisions regardless of the risk it posed to themselves (Dekeuwer & Bateman, 2013). For some women, the desire to plan and have a family outweighed not only the concern they had for their own health, but their concerns with regards to passing the BRCA gene on to their children (Dean & Rauscher, 2017).

In three of the four reviewed papers, in contrast to the aforementioned, other women prioritised decisions about cancer prevention, including preventative surgeries, above their family planning goals or their desire to have children (Werner-Lin, 2008; Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). This decision to prioritise health and surgery planning was prevalent across samples, however motivated by different ideas for different women. Some cited reducing their personal cancer risk as primary priority and therefore decided to prioritise surgeries and make decisions regarding family planning based around when these surgeries happened (Dean & Rauscher, 2017). Other women were motivated by their desire to stop the BRCA gene being passed down to the next generation, and at times opted not to have children to prevent this from happening, as this was their primary priority (Werner-Lin, 2008; Dekeuwer Bateman 2013; Dean & Rauscher, 2017). For some women who were already mothers to children, their decision to prioritise surgery was motivated by their sense of owing their existing children care and a desire to remain alive and able to care for them (Werner-Lin, 2008; Dekeuwer Bateman 2013).

One paper (Dean & Rauscher, 2017) cited an age difference in regards to prioritisation, with older women more typically in favour of prioritising their family planning goals

and decisions, compared to younger women, who were more likely to prioritise their personal cancer management plans and preventative surgeries.

Theme 2: Pressure and Time

External Pressures

In all five of the reviewed papers, participants spoke of a feeling of pressure in regards to what family planning decisions they should make, what other related decisions they should make, and when they should make them (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Some women felt this pressure from their physicians and medical professionals (Werner-Lin, 2008; Werner-Lin, 2010; Dean & Rauscher, 2017), whilst others felt it from their families (Werner-Lin, 2010; Dekeuwer & Bateman, 2013). For some, this pressure came in the form of a pressure to have cancer preventative surgeries as soon as possible to facilitate future childbearing and the living out of family plans (Werner-Lin, 2008; Werner-Lin, 2010; Dean & Rauscher, 2017). For other women, this pressure was felt more as a pressure to carry out family planning and childbearing as soon as possible to be able to facilitate future surgeries with biological children already accounted for (Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). This pressure to make decisions about family planning and related medical procedures caused some women to make decisions about children or in some cases have children before they felt prepared, or before they would have chosen to prior to their genetic diagnosis (Werner-Lin 2010; Donnelly et al, 2013; Dean & Rauscher, 2017). This pressure resulted in some women feeling that their family planning decisions were dictated by others within a timeframe decided by others, which was inflicted with a sense of pressure and force (Werner-Lin, 2010; Werner-Lin, 2008; Dekeuwer & Bateman, 2013).

Urgency and a Ticking Clock

In all five of the reviewed studies, participants discussed a sense of urgency to make family planning decisions and carry out these plans. This was due to a feeling that they only had a limited time to do so and a sense of living with a ticking clock which made them feel that, one day, the opportunity to plan for and have a family would cease to be (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Some women described this as a “race against time” (Werner-Lin, 2008, p. 429; (Werner-Lin 2010, p. 21) and others felt the burden of a “biological clock” (Dean & Rauscher, 2017, p. 1306) or a general feeling that they were running out of time (Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). For some women, this was felt as an urgency to have preventative surgeries before they ran out of time to plan for and have a family (Werner-Lin, 2010) and a sense that there was a limited timeframe within which to make take these preventative medical actions (Werner-Lin 2010; Dean & Rauscher, 2017). Other women described a sense of urgency to have children quickly to ensure that they did not run out of time to do so before they would need to make medical decisions and begin having surgeries (Werner-Lin, 2008; Werner-Lin, 2010; Dean & Rauscher, 2017). Whether women felt the urgency to have surgeries or children first, the idea that there exists a ticking countdown for when it is too late to do one, or either, of those things was present and prominent across the reviewed literature.

For some women, the urgency they felt related more so to the idea that there was a ticking clock counting down to a diagnosis of cancer. These women worried about making family planning decisions before falling ill and then being unable to have more, or any, biological children (Werner-Lin 2010), and some worried about making decisions about surgery before this point as well (Donnelly et al, 2013). Across four of the five studies included in review, a sense of certainty in relation to cancer

development prevailed and underpinned many family planning decisions. Women shared the opinion that the development of cancer was inevitable for them given their BRCA carrier status (Werner-Lin, 2008; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). For some this motivated them to commence surgery as soon as possible to enable biological childbearing in the future (Donnelly et al, 2013; Dean & Rauscher, 2017) with a belief that if they didn't, they would die (Donnelly et al, 2013), perhaps leaving children without parents, or leaving themselves without the opportunity to ever make or pursue family plans of their own. This certainty of cancer development, coupled with the uncertainty of when it would occur (Dean & Rauscher, 2017), had a large impact on the ways that many women navigated family planning.

In two of the reviewed papers, which shared a participant population, women expressed an urgency that they felt to find life-partners as soon as possible whom they could make family planning decisions with, and felt that there was a ticking clock and limited timeframe within which to achieve this before it would be too late (Werner-Lin, 2008; Werner-Lin 2010). These women felt an urgency to find a partner who accepts “a distinctive vision of family planning” (Werner-Lin, 2008, p. 432) and who would be able to “take care of [them] when [they] get cancer” (Werner-Lin, 2008, p. 424).

Theme 3: Legacy

Family History

The influence of a family history of cancer and BRCA diagnosis on family planning decisions was a prominent theme within all five of the reviewed papers (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Many women used the illness trajectory of a close family member as a blueprint for their decision making, viewing diagnosis and anniversaries of loved ones' deaths as deadlines to work towards when it came to making decisions about family

planning, including decisions about when to have surgeries (Werner-Lin, 2010; Donnelly et al, 2013; Dean & Rauscher, 2017). For some, a family history of cancer influenced whether they decided to have surgery, and when (Werner-Lin, 2010; Dekeuwer & Bateman, 2013), based upon a desire to change the narrative of cancer within a family from one of loss and grief to one of survivorship (Werner-Lin, 2010). For others, their family history influenced their decisions about whether or not to have children, and when (Donnelly et al, 2013).

For some, a family history of cancer generated an idea that cancer is inevitable (Werner-Lin, 2008; Donnelly et al, 2013) and women felt they would be able in part to predict their diagnosis and death based upon their lived experiences of when and how they lost their mothers, or other family members (Werner-Lin, 2008). This caused worries about family planning, such as worries about leaving children without a mother and how this would impact them (Werner-Lin, 2010). As one participant shared, “It’s not the stranger that might pull up in a car, it’s losing family members [...]. Those are the things that I talk about with them.” (Werner-Lin, 2010, p. 21). Most certainly, family history impacted the way that women from families with a history of cancer made decisions about family planning compared to their peers (Werner-Lin, 2008), with a family history dictating the extent to which cancer was seen as manageable (Donnelly et al, 2013). For those fortunate enough to have come from a family where cancer is an illness that is survived, they felt that they did not see it “as a reason not to bring a child into the world” (Donnelly et al, 2013, p. 1008). Nevertheless, family history of cancer diagnosis, death or survivorship impacted the way that women across all studies navigated family planning decision making.

A Sense of Responsibility

A sense of responsibility for their family planning decisions and the impact on their existing or potential children was a common theme throughout all five of the reviewed papers (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Many women felt a responsibility not to pass on the BRCA gene to their children, which impacted decisions they made around having more, or any, children. They worried about the impact that the gene would have on them, their lives, their health and their family planning decisions once the time came (Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Some felt that there was not a justification for them to pass on the BRCA gene with the knowledge that they were carriers, deeming it both a parental and moral duty in some cases not to do so (Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). In three of the studies, this sense of responsibility was coupled with a feeling of guilt for having taken the risk of passing on the gene, and questions about the selfishness of this decision and what it meant with regards to the responsibility they held as parents or potential parents to protect their children from harm (Werner-Lin 2010; Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). However, for some, these worries about gene transition did not outweigh the desire for biological children, and did not halt or hinder family planning decisions (Werner-Lin, 2010).

In four of the five reviewed studies, women felt a responsibility for protecting their children from familial loss as a result of cancer diagnosis and death, most predominantly parental loss (Werner-Lin, 2008; Werner-Lin 2010; Donnelly et al, 2013). This impacted the family planning decisions of some who decided to delay having a family until they had managed their personal cancer risk through surgery (Werner-Lin, 2010). For others, it motivated them to continue to have more children as soon as possible to protect their existing children from managing potential parental loss without the support of a sibling (Dekeuwer & Bateman, 2013; Donnelly et al, 2013).

This sense of responsibility to protect children from familial and parental loss motivated some women to have surgeries and make personal cancer management decisions quickly to facilitate their family planning decisions (Werner-Lin, 2008; Werner-Lin, 2010; Donnelly et al, 2013; Dean & Rauscher, 2017).

Theme 4: Hope

Optimism for the Future

In four of the five reviewed studies, optimism for the future was a prominent theme (Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Many women felt optimistic about future medical advancements and the positive impact that they would have on any children they decided to have despite their BRCA diagnosis. They believed that cancer would become easier to treat (Werner-Lin, 2010; Donnelly et al, 2013; Dean & Rauscher, 2017), that medical technology in general is advancing (Werner-Lin 2010; Donnelly et al, 2013; Dean & Rauscher, 2017), and that these advancements will provide their children, despite BRCA carrier status, with better options and choices when the time comes for them to make family planning or cancer management decisions (Werner-Lin 2010; Dean & Rauscher, 2017). For some, this optimism helped to manage the guilt they felt from potentially passing on the gene to their children (Dean & Rauscher, 2017). In one study conducted in France, women hoped that the legalisation of pre-implantation genetic diagnosis would also help their future children to navigate their BRCA status and associated family planning dilemmas with more ease than they had been able to (Dekeuwer & Bateman, 2013). For some women, the theme of optimism lay also in the way women approached their own sense of cancer risk. They reserved an optimism that because their own cancer risk was not 100%, the potential for harm as a result of deciding to have children would be uncertain also, and counterbalanced by the benefit of having a family and the

possibilities of new options in the future. This optimism helped women to make family planning decisions with more hope and a view of cancer and the future as manageable and full of potential for positive change (Donnelly et al, 2013; Dekeuwer & Bateman, 2013).

Being more than BRCA

In four of the five reviewed papers (Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017) the theme of hope was expressed through a further subtheme relating to the idea women held about themselves as being more than their BRCA diagnosis, which helped them to make family planning decisions with more optimism and less guilt. Women viewed themselves as multi-faceted (Werner-Lin 2010), and held in mind the idea that everyone has something potentially harmful that they could pass on to their children, and that this made family planning decisions for them less dictated by their diagnosis (Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). In two of the reviewed papers, women highlighted how pleasant their lives had been despite their carrier status, and an understanding that had their mothers made alternative family planning decisions with awareness that they were carriers, the women would not exist today. This helped them to make family planning decisions regardless of diagnosis and with a positive view of both themselves and their future children, as well as the lives that they would lead, just as they had, as carriers (Dekeuwer & Bateman, 2013; Donnelly et al, 2013). As one participant said, “One must make an effort to accept life with its imperfections” (Dekeuwer & Bateman, 2013, p. 242).

Discussion

Overall, the papers included in the review highlighted the huge impact of family history, most specifically relating to family history of cancer and familial loss, on women’s

decision making with regards to family planning (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Most participants felt a sense of urgency to make family planning and surgery related decisions as soon as possible and felt the burden of a ticking clock not dictated by themselves, which would determine when they would supposedly run out of time to make choices (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Women struggled to balance their desire to have children with their need to engage in preventative measures as a method of personal cancer management, such as having prophylactic surgeries (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). For some, the guilt they felt about passing on their BRCA gene to their children made a huge impact on their family planning decisions (Werner-Lin 2010; Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017), with some women questioning the morality of making such a decision with full awareness of the risk (Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017). Many women expressed that, despite the difficulties of being a BRCA carrier, they viewed themselves as much more than that, and held a sense of optimism that they would be able to provide the same happy lives to their children that their mothers had provided to them (Dekeuwer & Bateman, 2013; Donnelly et al, 2013). Some felt optimistic that, should they engage with the risk of passing on their BRCA gene, medical advancements would create a safer, better future for their children where the BRCA diagnosis would not impact upon them, their lives and their reproductive decisions so much (Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017). Overall, participants seemed to prioritise their desire for biological children above their personal cancer management (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Dean & Rauscher, 2017).

The reviewed papers cited several implications of their research in regards to further research as well as clinical practice, which are supported by the findings and synthesis within this review. Suggestions were made regarding further research investigating how the timing of genetic testing within the life cycle affects family planning decisions (Werner-Lin, 2008). Other suggestions include research into why women opt for genetic testing and preventative surgery yet do not engage with Pre-implantation Genetic Diagnosis (PGD) – a method of conceiving children using embryos confirmed not to be carrying the BRCA gene - when it comes to family planning (Dean & Rauscher, 2017). Multiple papers stressed the importance of research looking at the perspectives of professionals on family planning within BRCA populations (Donnelley et al, 2013; Dean & Rauscher, 2017) and the experiences of couples jointly (Werner-Lin, 2010; Donnelley et al, 2013; Dean & Rauscher, 2017). Further research comparing women with family histories of cancer to women without and their family planning decisions was also suggested (Werner-Lin, 2010). With regards to clinical implications, the reviewed papers suggested a need for discussions around family planning to be better facilitated by professionals during the genetic testing process (Donnelley et al, 2013; Dean & Rauscher, 2017). It was also suggested that interventions focused on dilemmas specific to the BRCA population, such as family planning decisions, be developed, and that grief from childhood cancer-related loss be approached and discussed during genetic counselling (Werner-Lin, 2010).

Limitations

A limitation of this review was the small number of papers included. This is due in part to the narrow body of research available which fit the inclusion criteria of this review. However, although there may be little research currently available relating to the family planning decisions of BRCA carriers, the research that does exist lends itself towards a continuous narrative of common themes in women's experiences.

The research papers included in review shared a number of limitations, such as a failure to account for ethics (Werner-Lin, 2008; Dekeuwer & Bateman, 2013), a failure to consider the impact of the researcher upon research (Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer & Bateman, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017) and a failure to explicitly identify the aims of the research (Werner-Lin, 2008; Dekeuwer & Bateman, 2013). It is arguable that the recruitment processes adopted by the researchers created an unavoidably biased sample of volunteers due to the narrow field of research which requires such specific participant inclusion criteria. Despite this there was some element of heterogeneity within the samples of the included studies who ranged in age and other demographic variables. This range in participant demographics may serve as a limitation of this review and may affect the generalisability of its findings. However, this too was arguably unavoidable due to the current landscape of research within this area, and provides a level of insight into this population which will contain a natural range in demographic variables amongst its members. This review therefore provides an initial exploration of the current research, which would benefit from repetition in later years when there is more research available.

Conclusions

The review highlights the importance of family cancer history on family planning decision making with the BRCA population, as well as the accompanying guilt women feel when navigating these decisions with an awareness of their risk. The importance of exploring this family history during the process of genetic diagnosis is apparent. The review also highlights the pressure women feel to make the correct decisions at the correct time, and the sense of a lack of agency that can at times accompany this feeling of a ticking clock, which counts down the time left to make life-altering and personal

decisions. However, there is an overarching sense of hope and optimism for BRCA carriers, who despite struggling to balance risk management with the right to typical family planning processes, manage to maintain a belief that everything will be okay. More research is needed to explore this experience of family planning from a couples perspective, and the findings of this review provide a good foundation for understanding the importance of family planning information and discussion during the BRCA journey to help women to feel empowered and less rushed to navigate such a difficult decision making process as family planning.

Conflict of Interest Statement

No potential conflict of interest was reported by the authors.

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Table 2. Table showing themes, subthemes and the corresponding papers in which they are present.

Theme	Subtheme	Papers
<i>Priorities</i>	<i>Cancer Management vs Family Planning</i>	Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer et al, 2013; Dean & Rauscher, 2017
<i>Pressure and Time</i>	<i>External Pressures</i>	Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer et al, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017
	<i>Urgency and the Ticking Clock</i>	Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer et al, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017
<i>Legacy</i>	<i>Family History</i>	Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer et al, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017
	<i>A Sense of Responsibility</i>	Werner-Lin, 2008; Werner-Lin, 2010; Dekeuwer et al, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017
<i>Hope</i>	<i>Optimism for the Future</i>	Werner-Lin, 2010; Dekeuwer et al, 2013; Donnelly et al, 2013; Dean & Rauscher, 2017

Being more than BRCA

Werner-Lin, 2010; Dekeuwer et al, 2013; Donnelly et al, 2013; Dean &
Rauscher, 2017

Part Two: Empirical Report

**“You just get on with it”: Experiences of women who undergo prophylactic
mastectomy following BRCA gene diagnosis**

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Abstract

Objectives: The aim of this study was to understand the experiences of women who undergo prophylactic (preventative) mastectomies following BRCA gene diagnosis.

Design: This study employed a qualitative methodology and opportunistic sampling. Recruitment was carried out within two NHS trusts within England. Participants had an average age of 37.6 years and were on average 3.3 years post-surgery. Each was interviewed for between 20-60 minutes about their experience of surgery.

Methods: Semi-structured interviews were audio-recorded, transcribed and analysed using Interpretative Phenomenological Analysis (IPA). Themes were identified across each interview, and explored in relation to the similarities and differences between each participants' experience.

Results: Four themes were identified: 'The Concept of Choice', 'Sense of Reality', 'Wanting to get back to normal' and 'Getting on with it'. These themes related to the conflicting feelings of surgery as obligatory and optional, the difficulty with surgery feeling somewhat surreal, a desire to get back to normal and leave the sick role, and a sense of needing to get on with things both before, during and after surgery.

Conclusions: This research provides insight into women's experiences of prophylactic mastectomy and their shared experiences of several emerging themes, linked closely to different parts of the surgery experience. This research provides insight into the potential role of psychological services for this population post-surgery, which is currently minimal within most healthcare contexts within the UK. Further research into the experiences of couples as well as research into women's experiences of support and help-seeking post-surgery would be beneficial.

Introduction

Breast cancer is the most common cancer amongst women worldwide (World Health Organisation, 2014), with susceptibility genes BRCA1 and BRCA2 putting women at a 56-87% lifetime risk of developing breast cancer (Heshka et al, 2008). Following genetic diagnosis, up to 51% opt to have a prophylactic (preventative) mastectomy (Botkin et al, 2000; Heshka et al, 2008) to reduce cancer risk. Research suggests surgery can have psychosocial effects on body image satisfaction, feelings of femininity, self-esteem, and relationships (Frost et al, 2000; Hopwood et al, 2000). For some, this leads to regrets relating to surgery being traumatic or emotionally difficult, feeling mutilated and a lack of psychological support following surgery (Payne et al, 2000). Other research has suggested minimal psychosocial difficulties (Frost et al, 2000; Hatcher et al., 2001; Stefanek et al., 1995), however methodological issues may have impacted the validity and generalisability of results, such as failing to assess sexual histories pre-surgery to provide a baseline when considering post-surgery sexual functioning (Monteiro-Grillo, Marques-Vidal & Jorge, 2005). Much data is collected on average 14.5 years post-surgery (Frost et al, 2000) with women with an average age of 45.5, which may skew data in favour of positive outcome (Borgen et al, 1998), as research shows older women tend to be more satisfied with their surgery (Metcalf et al, 2004). Previous studies have failed to explore preventative mastectomy as an experience, instead focusing on quantitatively measuring psychosocial outcomes, most often “cancer related distress”. This distress is therefore reduced or absent following surgery as prophylactic mastectomy drastically reduces cancer risk.

Literature relating to psychological cancer adjustment is useful to consider when investigating experiences of women at high risk of developing cancer, who are encouraged to make cancer-management decisions. This literature is relative as it considers cancer-specific factors, which separate BRCA populations from the general

population, and unite BRCA and cancer populations due to shared experiences of cancer-specific threat. Greer and Watson (1987) outlined five adjustment styles to cancer, which can be considered when investigating experiences of women who have undergone prophylactic mastectomy: 'Fighting spirit', 'Avoidance and denial', 'Fatalism', 'Hopelessness and helplessness' and 'Anxious preoccupation'.

Lloyd et al (2000) used a grounded theory approach to investigate women's experience of prophylactic mastectomy which led to the development of a process model of prophylactic mastectomy. Deciding to have surgery is the first step, influenced by family cancer experiences, genetic information, cancer fears, and feeling responsible for reducing cancer risk. Telling family members is the second step, followed by surgery itself, surgery recovery and an element referred to as 'maintaining womanliness'. This involves experiencing loss and sadness, viewing surgery as threatening womanliness and reduced body-image satisfaction post-surgery linked with self-blame. Other elements include processing loss, moving on, focusing on the future and a sense of partners 'riding it through'. Isolation and support run throughout this process, with distance from specialist services and a lack of formal support making women feel isolated.

The aim of this research is to answer the following research question: "What are the experiences of women who undergo prophylactic mastectomies following BRCA gene diagnosis?" This research will utilise qualitative research methods to explore experiences rather than quantitatively measuring psychosocial outcomes. There is currently minimal psychological support for women following prophylactic mastectomies, therefore the results of this research may help to understand experiences and the potential need for psychological support post-surgery.

Method

Design

The study design involved semi-structured interviews to gather qualitative data relating to women's experiences of having prophylactic mastectomies to be analysed using interpretative phenomenological analysis (IPA).

Ethics

Ethical approval was granted by the NHS Health Research Authority, Yorkshire and The Humber Sheffield Research Ethics Committee (see Appendix E for letters).

Recruitment

Participants were recruited using opportunistic sampling from NHS trusts local to the researcher. Clinicians were contacted, who identified potential participants based upon inclusion and exclusion criteria. The inclusion criteria were as follows: a) female b) aged over 18 c) diagnosed with BRCA d) had a prophylactic mastectomy within the last five years d) surgery was over three months ago. The exclusion criteria were a) previous or current cancer diagnosis b) undergoing cancer treatment. These criteria were designed to capture experiences soon after surgery and separate this research from previous research, and to ensure women had enough time post-surgery to have medically recovered. Excluding women currently or previously diagnosed with cancer ensured the research focused upon the BRCA population and their unique experiences. Potential participants were contacted by clinicians and consented to be contacted by the researcher. Telephone calls were made to arrange interviews. Paperwork completed by participants can be found in Appendix F, G, and H. Participants gave informed consented to audio-recorded interviews and signed consent forms (see Appendix I).

Participants

Across the NHS trusts involved in recruitment, 19 women were identified by clinicians. Three declined to take part, two did not meet criteria, two could not be reached and six did not return messages. Of the six who agreed to take part, when screened again before interview, only three met criteria. These women were interviewed.

Lindsay is 39 years old and had her surgery one and a half years ago. Chloe is 30 years old and had her surgery five years ago. Renna is 44 years old and had her surgery three and a half years ago. Their average age was therefore 37.6 and average time since surgery was 3.3 years.

Data Collection

Three semi-structured interviews were conducted lasting between 20-60 minutes at the researcher's university or participants' homes. The research schedule contained broad questions used to allow participants to discuss their experiences in their own words without being guided by the researcher. All interviews began with "Could you tell me what your experience of preventative mastectomy was like?" An example of the general interview schedule can be found in Appendix J. These interviews were audio-recorded and transcribed by the researcher for analysis. Following interviews, participants were given debrief sheets (see Appendix K). Letters were sent to participants' GPs with their consent to inform them of their involvement (see Appendix L).

Data Analysis

Interview transcripts were analysed using Interpretative Phenomenological Analysis (IPA), a qualitative analysis method involving idiographic analysis of each piece of data and exploration of emerging themes across the entire data set (Smith, Flowers & Larkin, 2012). This analysis process began with several readings the transcripts, followed by

exploratory note annotations of each transcript individually (Smith, Flowers & Larkin, 2012). An example can be found in Appendix M. These notes were then used to identify sub-ordinate themes, which were grouped into super-ordinate themes within each transcript. These super-ordinate themes were then analysed together to develop general super-ordinate themes present across the entire data set (Smith, Flowers & Larkin, 2012).

Researcher's Position

The primary researcher had experiences relating to elective surgery, and held an assumption that for each woman their experiences of surgery, healthcare services, pain and adjustment are different. The researcher used regular supervision to reflect upon her position, her personal experiences, and the potential interaction between these factors and the data analysis. The researcher maintained an impartial approach to the data analysis as much as possible and took steps to ensure a separateness between her own experiences, and the experiences of the participants in the present study. This was achieved through the use of a reflective research journal, and the discussion of issues with her research supervisor throughout the research process.

Results

The data analysis resulted in four identified emerging themes across the data set: 'The Concept of Choice', 'Facing the Reality', 'Wanting to get back to normal' and 'Getting on with it'.

Theme One: The Concept of Choice

When discussing their experiences, participants implied an active and purposeful choice.

“I decided quite quickly that...as soon as I found out I had the gene...that that’s what I wanted to do” – Renna

“The decision for surgery was...common sense” – Lindsay

“It was a no brainer for me really” – Chloe

Renna’s use of “I wanted to” implied choice, with both Lindsay and Chloe echoing similar feelings. However, their motivations were rooted in family cancer histories, ideas of cancer as inevitable, and an implied obligation to prolong their lives for their families. When discussing her journey to surgery, Lindsay talked about her sister’s cancer experience and their BRCA diagnoses. Lindsay made reference to them having a strong bond, implying a sense of solidarity in their experiences of being carriers.

“We’ve always had a sister thing”, she said, and of her BRCA gene, a duplicate of her sister’s; *“it’s nice to share”*.

The implicit suggestion they were sharing something, perhaps an experience, gave insight into her motivations for surgery. Her sister’s experience and opinion was integral to her decision.

“She very much just thought it’s the right thing to do, so...and that was, that was the decision done”

Lindsay’s surgery was a decision made by two sisters with a *“sister thing”*. Her repeated use of “we” when discussing the decision contrasted other comments suggesting an active pursuit of surgery individually.

“We had the thought process of everything [she] had gone through and we’d talked about it, we’d obviously discussed what would happen if it was me as well”

The influence of Lindsay's sister implied surgery was perhaps the only choice, at least a choice her sister wanted. There was an implicit guilt in Lindsay's account of her genetic testing as an "*opportunity*", and her implied solidarity with her sister perhaps suggested the same guilt applied when making decisions about surgery.

Lindsay said the decision to undergo surgery had not felt like a choice. The experience of a "sister thing" and of sharing something seemed important. The role of her sister's cancer seemed central, perhaps leading her to experience the decision as out of her control.

"It didn't really feel like a decision it was like right when's the surgery? It was, it was...just...ok yep I've got the gene as well...right so...right when's the surgery?"

Lindsay's experience of her time in hospital created an imagery of being imprisoned, implying she was a passive recipient of surgery.

"I thought if I cause myself that pain they're gonna keep me in"

Her description of the pain as something she would "cause [her]self" was an interesting insight into her ongoing confliction between feeling both in and out of control. For Renna, surgery had not felt much like a choice either.

"You just have to do it...just have to do it"

"It was for the best, and I needed it"

Like Lindsay, Renna's decision to undergo surgery was closely tied to her family experiences of cancer, and her sense of duty to be there for them.

"When you've got a family and that you just want to do anything you can to reduce that risk of getting cancer"

This sense of not having made a decision felt important to Renna, and she worried this would be misunderstood. Her differentiation between herself and those pursuing cosmetic surgery when, unlike her, they didn't "have to", was clear.

"I just think why would you put...and I know people have different reasons but I just think why would you want to put yourself through something you don't have to when it's so painful...to have implants put in...it's not...and I I know mine will have been different, because they took everything away and it was really painful...but I just don't know why anyone would choose to do that"

Renna's separation of her surgery as "different" from others highlighted her sense of not having made a choice. When reflecting on her surgery in interview, Renna's passive voice and description of surgery as something done to her further implied these feelings.

"I had everything taken away"

"They'd taken everything away"

Renna's repetition of having "everything taken away" suggested feelings of loss and a passive experience of what "they" had done to her. Chloe shared this feeling of not having made a choice about her surgery, and feeling it was something she "had" to do.

"For me, I didn't feel like I had a choice. For me I knew it had to be done"

"Obviously it's not what you choose to have done in life but, I had to, and that was that"

Chloe's description of surgery as "not what you choose" implied she felt there was no other choice and suggested the alternative to surgery would be even less desirable. For Chloe, the alternative seemed clear.

"I always thought prevention was better than the cure so I just did it"

All participants implied that their family histories suggested cancer was inevitable, therefore avoiding surgery would result in illness or death. This correlation between surgery and death was evident for all three participants, who each shared their fears of not waking up from surgery or having something go wrong. These fears were perhaps projections of a greater fear of death, which was perhaps more tangible and manageable when projected onto surgery itself. Linday, Renna and Chloe seemed somewhat forced by a sense of family duty and fear of their own mortality to make a choice, the alternative to which was perhaps worse, implying it was perhaps no choice at all.

Theme Two: Sense of Reality

The theme of reality was both explicit and implicit throughout the data. Each participant had a part of their experience after which it began to feel real, suggesting that during other parts of their experiences, things had felt surreal.

“I was fine, right up until I got the date. And then when I got the date it was really real” – Chloe

There was a time in one of the appointments, I’d just met the surgeon, erm...I think she was like, I think she’d given me a possible provisional few dates or something...and erm...[my partner] was with me and I, and...I just started crying, and I was like aw god...I just remember thinking aw my god”- Lindsay

“My partner took me to the hospital the day I had it done, and I was absolutely fine you know, sat reading, and this nurse came and she said you’re so brave, you’re so young to go through something like this and I just burst out crying. I thought why you would say that to me? I didn’t even think about it!”- Renna

Renna’s lack of thought before this moment implied she had avoided the reality of her circumstances. This avoidance, and the implication by all participants that there was

time surgery felt real, and time it did not, suggested difficulty coming to terms with the reality of surgery. This was discussed by Lindsay, beginning before her surgery with the avoidance of literature relating to the reality of what was to come.

“I just didn’t want to overly concern myself about...things that I didn’t need to think about”

Lindsay’s use of “didn’t need to” and later on “*shouldn’t need to*” imply a feeling she had of shock, injustice and perhaps therefore difficulty with accepting the reality of surgery happening. She discussed this avoidance more explicitly when explaining her time in hospital.

“If I didn’t move I could pretend oh yeah I’m just lying here it’s all fine, but then as soon as I moved I had these horrible drains to contend with and that made it a bit more realistic”

This avoidance seemed to continue after her return home, where she implied an effort to continue to avoid the reality of her circumstances, by thinking “*yeah I’m just watching DVDs all day...just having a bit of a rest*”. This feeling of reality and unreality fluctuated for all participants, with the actuality of surgery and painful aftermath seeming to serve as a grounding experience after which things began to feel more real. For Renna, this sense of things feeling surreal extended to feelings about her breasts post-surgery, which she said “*didn’t feel real*”.

For Lindsay there was not only difficulty accepting the reality of surgery happening, but an implicit sense that it was not as she expected.

“You expect a grand hall theatre don’t you? (laughs) It was like a...cubicle...(laughs) Yeah I was, I was not wowed by my theatre at all”

Lindsay's reference to a "grand hall theatre" creates an imagery of performance, providing insight into Lindsay's confrontation with the reality of surgery as not a performance, therefore un-real, but a real experience. Lindsay's first memories of waking up from surgery imply a similar experience of things feeling somewhat un-real, and suspended not only from her usual sense of reality, but from her ideas about what the reality of surgery would be.

"[I] saw my surgeon in the first bit where...recovery room? Where you first wake up, I saw her there and wh...she was clocking off for the day she's like I'm off into town!"

Lindsay's surgeon leaving to continue normal life seemed an important and prominent memory. Lindsay later explained her journey from her first thoughts about surgery as being straightforward were implied, along with her experience of the true reality of surgery as different from her expectations. She made links between these experiences of the reality of surgery and her perception of her own need for support.

"I would have said I don't need to see that counsellor, completely would have not wanted to see...I only did it because I was told to. I wouldn't have volunteered to go. I wouldn't have done, at all, I was just really like yeah that's fine, let's do it, chop em off, don't need em, might kill me, get rid. You know, bish bosh, done. And er...and whether that's tuning out of the emotion side of it, I don't know. But as soon as you've had the surgery, you can't switch your mind off, you're always thinking...y-y-your outlook's changed after that, it is, it does...it does chan...you think 'aw yeah it's just this', and it isn't. It isn't."

Lindsay's awareness of her expectations of the reality and true experience of reality are evident most when she explains "you think 'aw yeah it's just this', and it isn't. It isn't". Lindsay explained that, in time, "you're more accepting of what's happened" implying

not only the unavoidability of the reality eventually, but the process each participant had from expectation of reality, to realisation of reality and finally, to acceptance of reality.

Theme Three: Wanting to get back to normal

Each participant described changes in their feelings of ‘normality’ after surgery. The painful experience of surgery had been a catalyst for facing the reality of their circumstances and that something was no longer ‘normal’.

“I couldn’t even sit up in bed, because it felt like there was concrete boulders there” - Renna

“I couldn’t even lift my arms up to wash my own hair” – Chloe

Each expressed desire to get back to ‘normal’, implicitly linked to pain and feelings of illness. This rejection of the sick role in an effort to return to ‘normal’ was perhaps linked to beliefs about what ‘normal’ patients look like. The ‘normal’ trajectory of being healthy, then unwell, then undergoing treatment to return to ‘normal’ had not been the same for the participants, whose experiences had been of going into hospital ‘normal’ and leaving somewhat different. Striving to get back ‘normality’ was therefore tied to wanting to get home from hospital, and with previous feelings of health. This was most explicitly expressed by Chloe, who said *“I wanted to feel normal again, I didn’t wanna feel ill anymore”*. For Lindsay, this desire to get back to ‘normal’ seemed to be somewhat suffocating, implied by her description of the experience as stifling and her difficult articulating it.

“I remember thinking I can’t be in here another night cause I’ll flip out. I couldn’t...I couldn’t, I needed to get the window...I felt...really...claustrophobic...almost...not claustrophobic cause I wasn’t in a small area but I just needed fresh air. I felt really stifled”

The desire they shared to get back to ‘normal’ was further implied through their expressed concern that something would go wrong, such as infection, perhaps as it would prolong the time spent feeling abnormal. A desire for ‘normality’ and an awareness that things were in some ways no longer ‘normal’ were implied when discussing the changed body after surgery. Renna and Chloe talked about their bodies as different not only from their previous personal ‘normal’, but from the wider ‘normal’ within society. Renna “*didn’t want to look ridiculous*”, and Chloe reflected on her changed body and its effect on her experiences of motherhood.

“Your boobs don’t grow as you put weight on...so it’s a bit weird, like when I was pregnant I had tiny boobs, but I was pregnant, and most people their boobs grow with their belly, but yeah. Couldn’t breast feed...not, I didn’t breast feed the first time but...I might have given it a go this, the second time”

This awareness of her body as not ‘normal’ like “*most people*” was shared by Renna. Both of them seemed to accept their new ‘normal’ whilst maintaining an understanding that it was different. Both referenced their partners when discussing their changed bodies, perhaps implying the importance of having a partner transition from old to new ‘normal’, and implying an understanding of their bodies as different in wider society and the implications of this on desirability.

“It might be different if I wasn’t so settled. I’ve been with my husband for fifteen years, so, it’s, was never a problem for either of us. Might have been different if I was single I suppose” - Chloe

“I’ve been with my partner for years and years, so, he...whatever surgery I had, he was behind me” – Renna

Renna expressed her concerns that other people may notice her difference and implied desire to be unnoticed, suggesting a desire to be ‘normal’ in order to achieve that.

*“My partner had booked a...a holiday and I was thinking will people see me?
Will they know that I’ve had this op?”*

For Lindsay, there was an implied struggle with not only wanting to get back to ‘normal’, but with accepting her body as no longer ‘normal’. She had avoided looking at it after surgery, implying difficulty accepting the change.

“I just didn’t wanna...no...no...no I couldn’t face it. I literally just cleaned myself without looking and...yeah...I just I just couldn’t”

When she eventually had looked, she had found this difficult and confronting.

“I was in the shower, and...I don’t know...just...it just...I think it just hit me...like oh my god, I’ve got a weird body...this isn’t mine anymore, this is...what’s happened? And then, yeah...bang”
“It’s all a bit alien”

Lindsay’s description of her body as no longer belonging to her raises the question of who she felt it belonged to, and her descriptions of her body as “weird” and “alien” are implicative of it no longer being ‘normal’. Her use of the word “bang” is suggestive of abrupt change and an abrupt realisation that there was now a new ‘normal’ that she would need to adjust to in time.

Unlike Lindsay, Renna and Chloe had found it easier to cope with the ways in which their lives and bodies no longer felt ‘normal’, and to adjust to their new ‘normality’. For Lindsay, this had been more difficult, and her focus on fast recovery was implicative of her difficulties with her feelings of difference and with the need to establish a sense of ‘normality’, even if it was different from before.

“I ended up being more practical, and just thinking about post-surgery, what I’d wear, what I’d be comfy in, urm...and the practicalities of recovery. That’s what I kind of threw myself into”

She described her need for reassurance that even in recovery, she was ‘normal’.

“I’ve had this pain is that normal? What’s this...is this normal? Cause you just don’t know, you haven’t a clue.”

Lindsay described feeling “*relieved*” to be reassured she was “*fine*” by her surgeon at follow-up, implying that to adjust to the change in her previous ideas of what it meant to be ‘normal’, she needed to establish a new sense of ‘normality’ in which she fit.

Theme Four: Getting on with it

The theme of getting on with it was both explicit and implicit throughout all interviews, and seemed a prominent theme throughout participants’ experiences, beginning with surgery itself and extending through to life after surgery. During interviews, the idea of needing to get on with things was explicitly discussed several times.

“I’m quite strong and like, I just get on with it” – Chloe

“You just get on with it...you just, crack on” – Lindsay

This sense of needing to get on with surgery for Lindsay linked with guilt about not wanting to hassle people with what she felt were “silly questions”.

“I didn’t want to be a bother, I remember thinking that a lot”

Each seemed to focus on getting on with surgery in part by implying it was not a significant event, which perhaps links back to previous themes of feeling like the

decision was not theirs, and feeling the situation did not quite feel real. This understated view of surgery was present with her use of the word ‘just’ followed by a graphic description of surgery, implying surgery is something for which ‘getting on with it’ is simple.

The theme of getting on with things was implicit and present at other times. When discussing surgery, Renna often coupled negative descriptions of surgery with expressions of satisfaction.

“It was really good, but I could fee-...cause there’s wire mesh in there, to hold them in place, at this point, and I could feel the scalpel grating on the wire. So it was, a bizarre thing, but I was in and out in the same day so it was over and done with”

Renna’s pairing of graphic imagery and reports of severe pain with expressions of being pleased with her surgery implied a feeling she had that being positive and getting on with things was something she needed to do. This implied sense of obligation was present in conversation with Chloe, also.

“I worry about stupid things like flying. But big, life changing things like that, I don’t seem to...when it’s me I don’t seem to be bo-, yeah it’s okay”

Chloe describes her surgery as a “life changing thing”, and begins to describe how she is less concerned with these major events when they are happening to her, implying a sense of obligation to get on with things and perhaps be grateful for her surgery. This implied sense of obligatory gratitude was present for Lindsay, too, who implied a sense of guilt around her difficulties with being able to get on with things.

“I didn’t wanna have a big cry. Not at that point. It felt silly. I didn’t have cancer, I’d just dodged it hopefully”

“urm....kind of...angry at myself for being like that. But then, because I’ve I’ve had the pleasure, if you can call it a pleasure, of knowing that I could possibly have this...cancer come and get me, and I’ve taken that away...so why on earth should I feel sad about it?”

This acknowledgement of surgery as a negative experience, whilst holding in mind a sense of having “*the pleasure*”, implied a difficulty with getting on with it, but a feeling of needing to, in part due to a feeling of owing it to those not able to undergo the preventative treatment she had, however painful it had been.

“You don’t think about, everyday life you’re just going around, suddenly, and then suddenly, major surgery, you’re like, floored. You’re suddenly weak, you...you don’t wanna eat much, and your whole, your whole outlook is like oh my god, yeah we’re immortal – not! You...you’ve got that luck suddenly...it’s good to be alive, enjoying life, some people haven’t got this... and...some people are dying of cancer, so you your thought process is all that, plus the physical side of it...and they’re linked in, cause you can’t have a shower without thinking about it, cause you’re washing yourself, so...yeah...so it’s...very much...just tuning out eventually, that you’ve had the surgery and healing and...yeah...time does heal”

For Chloe and Renna, getting on with life after surgery was easier as they felt relieved and less worried about cancer. However, overall, descriptions of surgery remained neutral or negative, with the positives found perhaps to make getting on with it easier. Getting on with it perhaps seemed the only option, and one expected by society.

Lindsay’s descriptions of surgery included:

“It’s really fucking awful, but you get through it, and it’s done...and your life’s still going on...hopefully, cancer free, cause you’ve eliminated one...possible source”

“It’s been hell and I didn’t enjoy any of it obviously, it’s done, and...I made the decision for me that...I’ve taken hopefully that...cancer away from...eating that part of my body”

Chloe’s description of her approach to getting on with life after surgery provided a further example of the implied obligation to get on with things.

“I’m not gonna live my life worrying about that cause I’ve got a couple of scars, so, that’s how I saw it. At least I could be there for my kids”

This implied need to get on with things and accept the physical impact of surgery for the sake of children was perhaps linked to common societal narratives about women’s bodies, motherhood and childbirth, and the idea that women sacrifice their bodies during pregnancy and birth, and that this is an opportunity they should be grateful for.

Despite the implied sense of obligation to get on with things, the truth seemed to be that they were, and are, getting on with it. As Lindsay said, *“life just carries on”*.

Discussion

Summary

The findings of this research suggest shared emerging themes across the data set, suggesting similarities in participants’ experiences of prophylactic mastectomy. There were shared conflicting feelings of choice versus obligation regarding surgery and experiences of surgery feeling real and surreal. Each participant expressed a strong desire to return to ‘normal’, perhaps influenced by a sense of obligation to be grateful and a denial of the sick role. This sense of obligation influenced the decision to have surgery, the pursuit of ‘normality’ afterwards, and a focus on getting on with things before, during and after surgery. The four emergent themes were therefore interconnected, each interacting with one another. A diagram depicting this can be

found in Appendix N. Each participant's experience was uniquely different, and differed in its implications of such themes, but shared them in common, highlighting similarities across experiences.

Strengths and Limitations

The predominant strength of this research is the insight offered into experiences of prophylactic mastectomy through a qualitative methodology and IPA analysis. To the best of the researcher's knowledge, no study like this had been conducted with women within the first five years post-surgery. This research, whilst unique in its approach, suggests results in-keeping with other research findings, including the important influence of family history on preventative decision making and the impact of prophylactic mastectomy on body image (Frost et al, 2000; Hopwood et al, 2000; Meijers-Heijboer et al, 2000). It is important to consider that all participants in the present study had undergone breast reconstructions, as opposed to having breast tissue removed and no implants inserted, as previous research has highlighted the potential positive impact of reconstructive surgery on emotional outcomes (Metcalf et al, 2004; Monteiro-Grillo, Marques-Vidal & Jorge, 2005).

Greer and Watson's (1987) adjustment styles to cancer seemed somewhat evident in the present study, providing insight into the application of cancer literature to BRCA populations and the cross-overs in their experiences. The 'Fighting spirit' seemed evident throughout, specifically relating to the emergent theme of 'Getting on with it'. 'Avoidance and denial' and 'Anxious preoccupation' seemed especially fitting for Lindsay whose experience was perhaps more challenging. 'Fatalism' and 'Hopelessness and helplessness' seemed evident across the data with fears of death, feelings of inevitability and feelings of pressure relating to the choice of surgery implicit and explicit for all participants. These adjustment styles suggest a method of coping based

upon minimisation of experiences, which may impact upon support-seeking behaviour. It is therefore important to consider the role of support services in this coping strategy and whether the lack of formal post-surgery psychological support promotes minimisation-based coping and adjustment styles.

This research differed from Lloyd et al's (2000) research most predominantly with regards to methodology, as the present study employed IPA analysis whilst Lloyd et al (2000) employed grounded theory analysis to a larger sample of 10 women and eight of their partners. The present research offers new insights into Lloyd et al's (2000) process model, presenting areas for development. Decisions within the present study to have surgery were influenced by family cancer experiences, genetic information, cancer fear, and a sense of responsibility to reduce risk. Issues relating to a process of 'maintaining womanliness' involving a view of surgery as threatening womanliness, loss, sadness and an unhappiness with body image post-surgery linked with a sense of self-blame seemed less prevalent. Although these themes of loss, sadness and body image were at times implicit within the data, their existence was minimal and un-related to feelings of womanliness. However, the remaining elements of Lloyd et al's (2000) model were evident, including processing the loss and moving on, focusing on the future and a sense of partners 'riding it through'. The lack of formal support suggested by Lloyd et al (2000) was experienced by Lindsay most specifically. The current research therefore presents experiences which vary from Lloyd et al's (2000) model, suggesting the potential for the development of a new model to accommodate the variation in experiences.

The small sample size of this study could potentially serve as a limitation. However, multiple publications highlight the benefit and importance of a small sample size in IPA, due to IPA's focus on idiographic information (Smith, Flowers & Larkin, 2012). The aim of IPA research is not to produce generalisable findings but to investigate a

homogenous sample in detail by gathering rich data and conducting in-depth analysis (Smith, Flowers & Larkin, 2012). The current study's sample was homogenous not only in terms of meeting the necessary inclusion criteria, in that the women were all BRCA carriers who had undergone mastectomies within the last five years. In addition, all women were within a simple life stage, aged between 30 and 44. Each had similar jobs, lived in a similar area and were of similar socioeconomic status. Several had children, and all were in long-term relationships, if not married. The sample size of this study although smaller than suggested by some publications (Turpin et al, 1997) is therefore satisfactory to meet the requirements of IPA and achieve the aims of this study, and is in-keeping with the more recent encouragement of small sample sizes suggested by other literature (Pietkiewicz & Smith, 2012; Smith & Osborn, 2003). The present sample size is also suggested to be ideal for a first-time IPA researcher (Smith & Osborn, 2003; Smith, Flowers & Larkin, 2012). This study provides unique findings based upon its design and creates a platform for future research.

Implications

This research provides interesting insight into potential avenues for future research. Research into experiences of couples would be of great interest given the importance each participant assigned to the role of their partner as a source of support and important opinion. This would accompany the findings of this research and expand upon them as they relate to women's feelings of desirability and the role of mastectomy at different life-stages.

Research into women's experiences of support from professionals following prophylactic mastectomy through interview would also be relevant. During interview, Lindsay expressed feelings of dissatisfaction with the support offered post-surgery, most specifically related to emotional support and literature resources (see Appendix O

for quotes). It would be interesting to explore these issues further and gain a wider perspective. The development or adaptation of psychological or process models of women's experiences following BRCA diagnosis would also be useful theoretically and clinically.

Recruitment difficulties in future research should be considered. Future research into BRCA populations may perhaps take a longitudinal approach to data collection. The conduction of research within the parameters of healthcare services in which women are already patients would be ideal to allow multiple opportunities for recruitment during the surgery and post-surgery process.

This research presents several implications for clinical development. The British Psychological Society (BPS) guidelines (BPS, 2018) relating to the role of psychologists in risk-reducing breast surgery highlight the importance of thorough pre-surgery assessment and availability of post-surgery support. However, there are currently no specific or specialist services offering the latter. Although introducing mandatory psychology follow-up is perhaps not a clinical possibility, developing more rigorous follow-up procedures informed by the findings of this research may be beneficial. The provision of specialist psychological support services for this population would perhaps be beneficial, including peer support groups, one-to-one or family work often offered within other areas of health psychology. The findings of this research suggest that this provision may help to challenge the minimisation of these experiences and encourage women to talk through the difficult parts of this experience, by acknowledging that they exist, if they so wish. The referral of these women to mental health services for support with adjustment to health crises seems unsuitable, as does the inclusion of these services within existing psychological medicine or psycho-oncology services due to the absence of illness. Instead, the development of new psychological services, perhaps led by clinical psychologists and centred around

supporting adjustment and promoting coping skills, perhaps with input from experienced nurses in the field, could be beneficial. This work could perhaps use a Compassion Focused Therapy (CFT) approach to address the implicit feelings of guilt and apparent difficulties with self-compassion with regards to minimising the experience. Further resources, such as literature addressing the more practical elements of surgery and recovery, would be welcomed to enable women to feel more empowered in their recovery and prepared for what to expect, and could be incorporated into this structured approach to support. Therefore the offering of perhaps not a follow-up appointment, but a package of post-surgery psychological support to be opted in or out of, seems an appropriate clinical development.

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Part three: Appendices

Appendix A - British Journal of Health Psychology instructions for contributors

British Journal of Health Psychology:

Author Guidelines

The aim of the British Journal of Health Psychology is to provide a forum for high quality research, including Registered Reports, relating to health and illness. The scope of the journal includes all areas of health psychology as outlined in the Journal Overview.

The types of paper invited are:

- papers reporting original empirical investigations, using either quantitative or qualitative methods, including reports of interventions in clinical and non-clinical populations;
- theoretical papers which report analyses on established theories in health psychology;
- we particularly welcome review papers, which should aim to provide systematic overviews, evaluations and interpretations of research in a given field of health psychology (narrative reviews will only be considered for editorials or important theoretical discourses); and
- methodological papers dealing with methodological issues of particular relevance to health psychology.

Authors who are interested in submitting papers that do not fit into these categories are advised to contact the editors who would be very happy to discuss the potential submission.

All papers published in The British Journal of Health Psychology are eligible for Panel A: Psychology, Psychiatry and Neuroscience in the Research Excellence Framework (REF).

1. Circulation

The circulation of the Journal is worldwide. Papers are invited and encouraged from authors throughout the world.

2. Length

Papers describing quantitative research (including reviews with quantitative analyses) should be no more than 5000 words (excluding the abstract, reference list, tables and figures). Papers describing qualitative research (including reviews with qualitative analyses) should be no more than 6000 words (including quotes, whether in the text or in tables, but excluding the abstract, tables, figures and references). In exceptional cases the Editor retains discretion to publish papers beyond this length where the clear and concise expression of the scientific content requires greater length (e.g., explanation of a

new theory or a substantially new method). Authors must contact the Editor prior to submission in such a case.

3. Editorial policy

The Journal receives a large volume of papers to review each year, and in order to make the process as efficient as possible for authors and editors alike, all papers are initially examined by the Editors to ascertain whether the article is suitable for full peer review. In order to qualify for full review, papers must meet the following criteria:

- the content of the paper falls within the scope of the Journal
- the methods and/or sample size are appropriate for the questions being addressed
- research with student populations is appropriately justified
- the word count is within the stated limit for the Journal (i.e. 5000 words, or 6,000 words for qualitative papers)

4. Submission and reviewing

All manuscripts must be submitted via Editorial Manager. The Journal operates a policy of anonymous (double blind) peer review. We also operate a triage process in which submissions that are out of scope or otherwise inappropriate will be rejected by the editors without external peer review to avoid unnecessary delays. Before submitting, please read the terms and conditions of submission and the declaration of competing interests. You may also like to use the Submission Checklist to help you prepare your paper.

By submitting a manuscript to or reviewing for this publication, your name, email address, and affiliation, and other contact details the publication might require, will be used for the regular operations of the publication, including, when necessary, sharing with the publisher (Wiley) and partners for production and publication. The publication and the publisher recognize the importance of protecting the personal information collected from users in the operation of these services, and have practices in place to ensure that steps are taken to maintain the security, integrity, and privacy of the personal data collected and processed. You can learn more at <https://authorservices.wiley.com/statements/data-protection-policy.html>.

5. Manuscript requirements

- Contributions must be typed in double spacing with wide margins. All sheets must be numbered.
- Manuscripts should be preceded by a title page which includes a full list of authors and their affiliations, as well as the corresponding author's contact details. You may like to use this template. When entering the author names into Editorial Manager, the corresponding author will be asked to provide a CRediT contributor role to classify the

role that each author played in creating the manuscript. Please see the [Project CRediT](#) website for a list of roles.

- For articles containing original scientific research, a structured abstract of up to 250 words should be included with the headings: Objectives, Design, Methods, Results, Conclusions. Review articles should use these headings: Purpose, Methods, Results, Conclusions. As the abstract is often the most widely visible part of your paper, it is important that it conveys succinctly all the most important features of your study. You can save words by writing short, direct sentences. Helpful hints about writing the conclusions to abstracts can be found [here](#).
- Statement of Contribution: All authors are required to provide a clear summary of ‘what is already known on this subject?’ and ‘what does this study add?’. Authors should identify existing research knowledge relating to the specific research question and give a summary of the new knowledge added by your study. Under each of these headings, please provide 2-3 (maximum) clear outcome statements (not process statements of what the paper does); the statements for ‘what does this study add?’ should be presented as bullet points of no more than 100 characters each. The Statement of Contribution should be a separate file.
- Conflict of interest statement: We are now including a brief conflict of interest statement at the end of each accepted manuscript. You will be asked to provide information to generate this statement during the submission process.
- The main document must be anonymous. Please do not mention the authors’ names or affiliations (including in the Method section) and always refer to any previous work in the third person.
- Tables should be typed in double spacing, each on a separate page with a self-explanatory title. Tables should be comprehensible without reference to the text. They should be placed at the end of the manuscript but they must be mentioned in the text.
- Figures can be included at the end of the document or attached as separate files, carefully labelled in initial capital/lower case lettering with symbols in a form consistent with text use. Unnecessary background patterns, lines and shading should be avoided. Captions should be listed on a separate sheet. The resolution of digital images must be at least 300 dpi. All figures must be mentioned in the text.
- For reference citations, please use APA style. Particular care should be taken to ensure that references are accurate and complete. Give all journal titles in full and provide doi numbers where possible for journal articles. For example:

Author, A., Author, B., & Author, C. (1995). Title of book. City, Country: Publisher.
Author, A. (2013). Title of journal article. Name of journal, 1, 1-16. doi:
10.1111/bjep.12031
- SI units must be used for all measurements, rounded off to practical values if appropriate, with the imperial equivalent in parentheses.

- In normal circumstances, effect size should be incorporated.
- Authors are requested to avoid the use of sexist language.
- Authors are responsible for acquiring written permission to publish lengthy quotations, illustrations, etc. for which they do not own copyright. For guidelines on editorial style, please consult the APA Publication Manual published by the American Psychological Association.
- Manuscripts describing clinical trials are encouraged to submit in accordance with the CONSORT statement on reporting randomised controlled trials.
- Manuscripts reporting systematic reviews and meta-analyses are encouraged to submit in accordance with the PRISMA statement.
- Manuscripts reporting interventions are encouraged to describe them in accordance with the TIDieR checklist.

If you need more information about submitting your manuscript for publication, please email Hannah Wakley, Managing Editor (bjhp@wiley.com) or phone +44 (0) 116 252 9504.

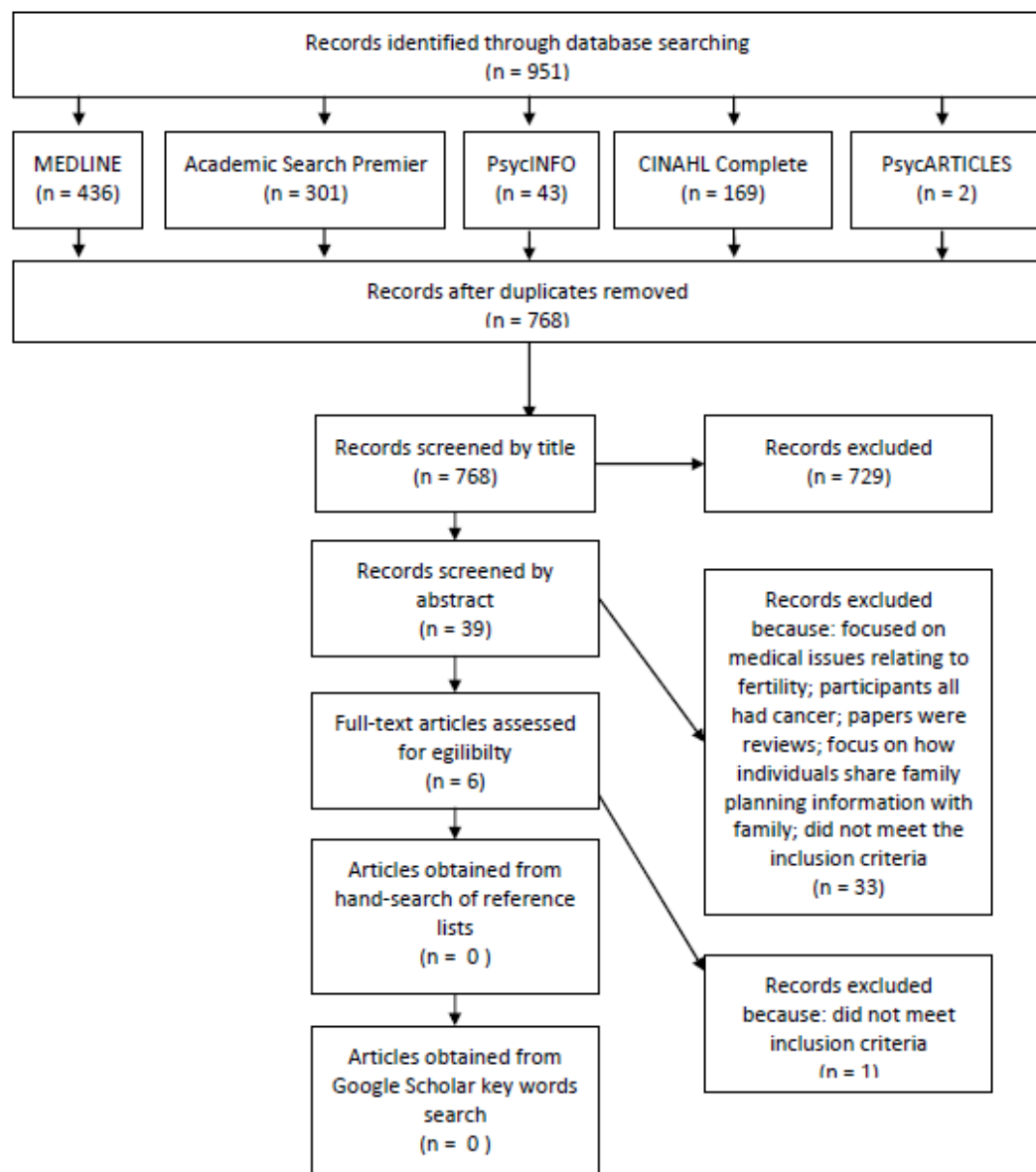
6. Supporting information

We strongly encourage submission of protocol papers or trial registration documents, where these are in the public domain, to allow reviewers to assess deviations from these protocols. This will result in reviewers being unblinded to author identity.

Supporting Information can be a useful way for an author to include important but ancillary information with the online version of an article. Examples of Supporting Information include appendices, additional tables, data sets, figures, movie files, audio clips, and other related nonessential multimedia files. Supporting Information should be cited within the article text, and a descriptive legend should be included. Please indicate clearly on submission which material is for online only publication. It is published as supplied by the author, and a proof is not made available prior to publication; for these reasons, authors should provide any Supporting Information in the desired final format.

For further information on recommended file types and requirements for submission, please visit the Supporting Information page on Author Services.

Appendix B - Flowchart to show search strategy and paper selection.



Appendix C – CASP appraisal form



CASP Checklist: 10 questions to help you make sense of a **Qualitative** research

How to use this appraisal tool: Three broad issues need to be considered when appraising a qualitative study:

- ▶ Are the results of the study valid? (Section A)
- ▶ What are the results? (Section B)
- ▶ Will the results help locally? (Section C)

The 10 questions on the following pages are designed to help you think about these issues systematically. The first two questions are screening questions and can be answered quickly. If the answer to both is "yes", it is worth proceeding with the remaining questions. There is some degree of overlap between the questions, you are asked to record a "yes", "no" or "can't tell" to most of the questions. A number of italicised prompts are given after each question. These are designed to remind you why the question is important. Record your reasons for your answers in the spaces provided.

About: These checklists were designed to be used as educational pedagogic tools, as part of a workshop setting, therefore we do not suggest a scoring system. The core CASP checklists (randomised controlled trial & systematic review) were based on JAMA 'Users' guides to the medical literature 1994 (adapted from Guyatt GH, Sackett DL, and Cook DJ), and piloted with health care practitioners.

For each new checklist, a group of experts were assembled to develop and pilot the checklist and the workshop format with which it would be used. Over the years overall adjustments have been made to the format, but a recent survey of checklist users reiterated that the basic format continues to be useful and appropriate.

Referencing: we recommend using the Harvard style citation, i.e.: *Critical Appraisal Skills Programme (2018). CASP (insert name of checklist i.e. Qualitative) Checklist. [online] Available at: URL. Accessed: Date Accessed.*

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Critical Appraisal Skills Programme (CASP) part of Oxford Centre for Triple Value Healthcare Ltd www.casp-uk.net

Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes
Can't Tell
No

- HINT: Consider
- what was the goal of the research
 - why it was thought important
 - its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes
Can't Tell
No

- HINT: Consider
- If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
 - Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes
Can't Tell
No

- HINT: Consider
- if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

Comments:

4. Was the recruitment strategy appropriate to the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the researcher has explained how the participants were selected
 - If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study
 - If there are any discussions around recruitment (e.g. why some people chose not to take part)

Comments:

5. Was the data collected in a way that addressed the research issue?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the setting for the data collection was justified
 - If it is clear how data were collected (e.g. focus group, semi-structured interview etc.)
 - If the researcher has justified the methods chosen
 - If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide)
 - If methods were modified during the study. If so, has the researcher explained how and why
 - If the form of data is clear (e.g. tape recordings, video material, notes etc.)
 - If the researcher has discussed saturation of data

Comments:

6. Has the relationship between researcher and participants been adequately considered?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location
- How the researcher responded to events during the study and whether they considered the implications of any changes in the research design

Comments:

Section B: What are the results?

7. Have ethical issues been taken into consideration?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained
- If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the study on the participants during and after the study)
- If approval has been sought from the ethics committee

Comments:

8. Was the data analysis sufficiently rigorous?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If there is an in-depth description of the analysis process
- If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
- Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
- If sufficient data are presented to support the findings
 - To what extent contradictory data are taken into account
- Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

Comments:

9. Is there a clear statement of findings?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider whether

- If the findings are explicit
- If there is adequate discussion of the evidence both for and against the researcher's arguments
- If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
- If the findings are discussed in relation to the original research question

Comments:

Section C: Will the results help locally?

10. How valuable is the research?

HINT: Consider

- If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature
- If they identify new areas where research is necessary
- If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Comments:

Appendix D – NICE appraisal form

Checklist

Study identification: Include author, title, reference, year of publication		
Guidance topic:	Key research question/aim:	
Checklist completed by:		
Theoretical approach		
1. Is a qualitative approach appropriate? For example: <ul style="list-style-type: none"> Does the research question seek to understand processes or structures, or illuminate subjective experiences or meanings? Could a quantitative approach better have addressed the research question? 	Appropriate Inappropriate Not sure	Comments:
2. Is the study clear in what it seeks to do? For example: <ul style="list-style-type: none"> Is the purpose of the study discussed – aims/objectives/research question/s? Is there adequate/appropriate reference to the literature? Are underpinning values/assumptions/theory discussed? 	Clear Unclear Mixed	Comments:
Study design		
3. How defensible/rigorous is the research design/methodology? For example:	Defensible Indefensible	Comments:

<ul style="list-style-type: none"> • Is the design appropriate to the research question? • Is a rationale given for using a qualitative approach? • Are there clear accounts of the rationale/justification for the sampling, data collection and data analysis techniques used? • Is the selection of cases/sampling strategy theoretically justified? 	Not sure	
Data collection		
4. How well was the data collection carried out? For example: <ul style="list-style-type: none"> • Are the data collection methods clearly described? • Were the appropriate data collected to address the research question? • Was the data collection and record keeping systematic? 	Appropriately Inappropriately Not sure/inadequately reported	Comments:
Trustworthiness		
5. Is the role of the researcher clearly described? For example: <ul style="list-style-type: none"> • Has the relationship between the researcher and the participants been adequately considered? • Does the paper describe how the research was explained and presented to the participants? 	Clearly described Unclear Not described	Comments:
6. Is the context clearly described? For example: <ul style="list-style-type: none"> • Are the characteristics of the participants and 	Clear Unclear	Comments:

settings clearly defined? <ul style="list-style-type: none"> • Were observations made in a sufficient variety of circumstances • Was context bias considered 	Not sure	
7. Were the methods reliable? For example: <ul style="list-style-type: none"> • Was data collected by more than 1 method? • Is there justification for triangulation, or for not triangulating? • Do the methods investigate what they claim to? 	Reliable Unreliable Not sure	Comments:
Analysis		
8. Is the data analysis sufficiently rigorous? For example: <ul style="list-style-type: none"> • Is the procedure explicit – i.e. is it clear how the data was analysed to arrive at the results? • How systematic is the analysis, is the procedure reliable/dependable? • Is it clear how the themes and concepts were derived from the data? 	Rigorous Not rigorous Not sure/not reported	Comments:
9. Is the data 'rich'? For example: <ul style="list-style-type: none"> • How well are the contexts of the data described? • Has the diversity of perspective and content been explored? • How well has the detail and depth been demonstrated? • Are responses compared and contrasted across 	Rich Poor Not sure/not reported	Comments:

groups/sites?		
10. Is the analysis reliable? For example: <ul style="list-style-type: none"> • Did more than 1 researcher theme and code transcripts/data? • If so, how were differences resolved? • Did participants feed back on the transcripts/data if possible and relevant? • Were negative/discrepant results addressed or ignored? 	Reliable Unreliable Not sure/not reported	Comments:
11. Are the findings convincing? For example: <ul style="list-style-type: none"> • Are the findings clearly presented? • Are the findings internally coherent? • Are extracts from the original data included? • Are the data appropriately referenced? • Is the reporting clear and coherent? 	Convincing Not convincing Not sure	Comments:
12. Are the findings relevant to the aims of the study?	Relevant Irrelevant Partially relevant	Comments:
13. Conclusions For example: <ul style="list-style-type: none"> • How clear are the links between data, interpretation and conclusions? • Are the conclusions plausible and coherent? • Have alternative explanations been explored 	Adequate Inadequate Not sure	Comments:

<p>and discounted?</p> <ul style="list-style-type: none"> • Does this enhance understanding of the research topic? • Are the implications of the research clearly defined? <p>Is there adequate discussion of any limitations encountered?</p>		
Ethics		
<p>14. How clear and coherent is the reporting of ethics?</p> <p>For example:</p> <ul style="list-style-type: none"> • Have ethical issues been taken into consideration? • Are they adequately discussed e.g. do they address consent and anonymity? • Have the consequences of the research been considered i.e. raising expectations, changing behaviour? • Was the study approved by an ethics committee? 	<p>Appropriate</p> <p>Inappropriate</p> <p>Not sure/not reported</p>	<p>Comments:</p>
Overall assessment		
<p>As far as can be ascertained from the paper, how well was the study conducted? (see guidance notes)</p>	<p>++</p> <p>+</p> <p>—</p>	<p>Comments:</p>

Appendix E - Letters of ethical approval



**Health Research
Authority**

Yorkshire & The Humber - Sheffield Research Ethics Committee

NHSBT Newcastle Blood Donor Centre
Holland Drive
Newcastle upon Tyne
NE2 4NQ

Tel: 0207 104 8082

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

24 July 2018

Miss Emily Rawding
Doctorate in Clinical Psychology
Aire Building, University of Hull
Cottingham
HU6 7RX

Dear Miss Rawding

Study title:	Experiences of women who undergo prophylactic mastectomies following BRCA 1 or 2 gene diagnosis.
REC reference:	18/YH/0203
Protocol number:	N/A
IRAS project ID:	241594

Thank you for your letter of 13 July 2018, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

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

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

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

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

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

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System, at www.hra.nhs.uk or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

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

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

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

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Approved documents

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

<i>Document</i>	<i>Version</i>	<i>Date</i>
Costing template (commercial projects) [Costs estimate]		25 April 2018
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [University of Hull Indemnity Policy]		25 April 2018
GP/consultant information sheets or letters	Version 2	20 July 2018
IRAS Application Form [IRAS_Form_15052018]		15 May 2018
IRAS Checklist XML [Checklist_21052018]		21 May 2018
IRAS Checklist XML [Checklist_13072018]		13 July 2018
Other [REC response letter]	1	
Participant consent form [Consent to be contacted form]	1	25 April 2018
Participant consent form	Version 3	20 July 2018
Participant information sheet (PIS) [Debrief and source of support sheet]	1	25 April 2018
Participant information sheet (PIS) [Personal details form]		
Participant information sheet (PIS)	Version 3	20 July 2018

Research protocol or project proposal [Final research proposal]	2	25 April 2018
Summary CV for Chief Investigator (CI) [Chief Investigator CV]		25 April 2018
Summary CV for supervisor (student research) [Dr Emma Lewis CV]		25 April 2018

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document “*After ethical review – guidance for researchers*” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

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

User Feedback

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

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

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at <http://www.hra.nhs.uk/hra-training/>

18/YH/0203

Please quote this number on all correspondence

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

Yours sincerely

pp



Dr Amaka Offiah Chair

Email: nrescommittee.yorkandhumber-sheffield@nhs.net

Enclosures: "After ethical review – guidance for researchers"

Copy to: Mr Stephen Walker, Humber NHS Teaching Trust



Ymchwil Iechyd
a Gofal Cymru
Health and Care
Research Wales



Miss Emily Rawding

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HU6 7RX

Email: hra.approval@nhs.net
Research-permissions@wales.nhs.uk

25 July 2018

Dear Miss Rawding

HRA and Health and Care

Research Wales (HCRW)

Approval Letter

Study title:	Experiences of women who undergo prophylactic mastectomies following BRCA 1 or 2 gene diagnosis.
IRAS project ID:	241594
REC reference:	18/YH/0203
Sponsor	Humber NHS Teaching Trust

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

How should I continue to work with participating NHS organisations in England and Wales?

You should now provide a copy of this letter to all participating NHS organisations in England and Wales, as well as any documentation that has been updated as a result of the assessment.

Following the arranging of capacity and capability, participating NHS organisations should **formally confirm** their capacity and capability to undertake the study. How this will be confirmed is detailed in the “*summary of assessment*” section towards the end of this letter.

You should provide, if you have not already done so, detailed instructions to each organisation as to how you will notify them that research activities may commence at site following their confirmation of capacity and capability (e.g. provision by you of a 'green light' email, formal notification following a site initiation visit, activities may commence immediately following confirmation by participating organisation, etc.).

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed [here](#).

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How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?

HRA and HCRW Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) has been sent to the coordinating centre of each participating nation. You should work with the relevant national coordinating functions to ensure any nation specific checks are complete, and with each site so that they are able to give management permission for the study to begin.

Please see [IRAS Help](#) for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?

HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your nonNHS organisations to [obtain local agreement](#) in accordance with their procedures.

What are my notification responsibilities during the study?

The document *"After Ethical Review – guidance for sponsors and investigators"*, issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including: ☐ Registration of research

- Notifying amendments
- Notifying the end of the study

The [HRA website](#) also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

I am a participating NHS organisation in England or Wales. What should I do once I receive this letter?

You should work with the applicant and sponsor to complete any outstanding arrangements so you are able to confirm capacity and capability in line with the information provided in this letter.

The sponsor contact for this application is as follows:

Name: Mr Stephen Walker

Tel: 01482301723

Email: stephen.walker7@nhs.net

Who should I contact for further information?

Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **241594**. Please quote this on all correspondence.

Yours sincerely

Kevin Ahmed

Assessor

Telephone: 0207 104 8171

Email: hra.approval@nhs.net

Copy to: Mr Stephen Walker, R&D Contact, Humber NHS Teaching Trust **List of Documents**

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	Version	Date
Costing template (commercial projects) [Costs estimate]		25 April 2018

Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [University of Hull Indemnity Policy]		25 April 2018
GP/consultant information sheets or letters	Version 2	20 July 2018
HRA Schedule of Events	1.0	21 June 2018
HRA Statement of Activities	1.0	21 June 2018
IRAS Application Form [IRAS_Form_15052018]		15 May 2018
Other [REC response letter]	1	
Participant consent form	Version 3	20 July 2018
Participant consent form [Consent to be contacted form]	1	25 April 2018
Participant information sheet (PIS) [Personal details form]		
Participant information sheet (PIS)	Version 3	20 July 2018
Participant information sheet (PIS) [Debrief and source of support sheet]	1	25 April 2018
Research protocol or project proposal [Final research proposal]	2	25 April 2018
Summary CV for Chief Investigator (CI) [Chief Investigator CV]		25 April 2018
Summary CV for supervisor (student research) [Dr Emma Lewis CV]		25 April 2018

Summary of assessment

The following information provides assurance to you, the sponsor and the NHS in England and Wales that the study, as assessed for HRA and HCRW Approval, is compliant with relevant standards. It also provides information and clarification, where appropriate, to participating NHS organisations in England and Wales to assist in assessing, arranging and confirming capacity and capability.

Assessment criteria

Section	Assessment Criteria	Compliant with Standards	Comments
1.1	IRAS application completed correctly	Yes	No comments
2.1	Participant information/consent documents and consent process	Yes	No comments
3.1	Protocol assessment	Yes	No comments

4.1	Allocation of responsibilities and rights are agreed and documented	Yes	The sponsor has submitted the HRA Statement of Activities and intends for this to form the agreement between the sponsor and study sites. The sponsor is not requesting, and does not require any additional contracts with study sites.
4.2	Insurance/indemnity arrangements assessed	Yes	No comments
4.3	Financial arrangements assessed	Yes	No study funding will be provided to sites, as detailed at Schedule 1 of the Statement of Activities.
5.1	Compliance with the Data Protection Act and data security issues assessed	Yes	No comments
5.2	CTIMPS – Arrangements for compliance with the Clinical Trials Regulations assessed	Not Applicable	No comments
Section	Assessment Criteria	Compliant with Standards	Comments
5.3	Compliance with any applicable laws or regulations	Yes	No comments
6.1	NHS Research Ethics Committee favourable opinion received for applicable studies	Yes	No comments
6.2	CTIMPS – Clinical Trials Authorisation (CTA) letter received	Not Applicable	No comments
6.3	Devices – MHRA notice of no objection received	Not Applicable	No comments
6.4	Other regulatory approvals and authorisations received	Not Applicable	No comments

Participating NHS Organisations in England

This provides detail on the types of participating NHS organisations in the study and a statement as to whether the activities at all organisations are the same or different.

The Chief Investigator or sponsor should share relevant study documents with participating NHS organisations in England and Wales in order to put arrangements in place to deliver the study. The documents should be sent to both the local study team, where applicable, and the office providing the research management function at the participating organisation. Where applicable, the local LCRN contact should also be copied into this correspondence.

If chief investigators, sponsors or principal investigators are asked to complete site level forms for participating NHS organisations in England and Wales which are not provided in IRAS, the HRA or HCRW websites, the chief investigator, sponsor or principal investigator should notify the HRA immediately at hra.approval@nhs.net or HCRW at Research-permissions@wales.nhs.uk. We will work with these organisations to achieve a consistent approach to information provision.

Principal Investigator Suitability

This confirms whether the sponsor position on whether a PI, LC or neither should be in place is correct for each type of participating NHS organisation in England and the minimum expectations for education, training and experience that PIs should meet (where applicable).

The Chief Investigator will be responsible for all research activities performed at study sites.

GCP training is not a generic training expectation, in line with the [HRA statement on training expectations](#).

HR Good Practice Resource Pack Expectations

This confirms the HR Good Practice Resource Pack expectations for the study and the pre-engagement checks that should and should not be undertaken

Where arrangements are not already in place, network staff (or similar) undertaking any of the research activities listed in A18 of the IRAS form would be expected to obtain a Letter of Access based on standard DBS checks and occupational health clearance would be appropriate.

Other Information to Aid Study Set-up

This details any other information that may be helpful to sponsors and participating NHS organisations in England to aid study set-up.

The applicant has indicated that they do not intend to apply for inclusion on the NIHR CRN Portfolio.

Appendix F - Consent to be contacted form

CONSENT FORM

Title of Project: Experiences of women who undergo prophylactic mastectomies following BRCA 1 or 2 gene diagnosis

Name of Researcher: Emily Rawding

Please initial boxes

1. I confirm that I give consent for my personal details, including my name and contact details such as telephone number and email address, to be provided to the named researcher

☐

2. I confirm that I give consent for the named researcher to contact me regarding participation in this research project

☐

Name of participant

Date

Signature

—

—

—

Name of person taking
consent

Date

Signature

—

—

—

Appendix G - Personal details form

PERSONAL DETAILS FORM

Title of Project: Experiences of women who undergo prophylactic mastectomies following BRCA 1 or 2 gene diagnosis

Name of Researcher: Emily Rawding

NAME:

DATE OF BIRTH:

DATE OF SURGERY:

TELEPHONE NUMBER:

EMAIL ADDRESS:

I WOULD PREFER TO BE CONTACTED VIA THE FOLLOW METHOD:

☐

TELEPHONE

EMAIL

☐

Name of participant

Date

Signature

Name of person taking
information

Date

Signature

Appendix H - Participant information sheet

Participant information sheet

PROJECT TITLE

Experiences of women who undergo prophylactic mastectomies following BRCA 1 or 2 gene diagnosis.

INVITATION

I would like to invite you take part in a research study on women's experiences of undergoing a prophylactic (preventative) mastectomy (surgical removal of one or both breasts) following diagnosis of the BRCA 1 or 2 gene (breast and ovarian cancer susceptibility genes). Please read the following information to see if taking part in this research would be of interest to you.

PURPOSE OF STUDY

We aim to find out what women's experiences of having this surgery are like, and how they feel about this surgery as very little is known about this currently.

WHY HAVE YOU BEEN INVITED TO PARTICIPATE?

This study requires women over the age of 18 who have undergone a prophylactic mastectomy within the past 3 years and who is diagnosed with the BRCA 1 or 2 gene. You have therefore been invited to take part as you meet this specific criteria.

DO I HAVE TO TAKE PART?

No. Taking part in this research is completely voluntary. If you do decide to take part and then change your mind, your data can be removed from this study any time up until the point where the data is being analysed. After this data is anonymised and it would be difficult to remove one person's data. Up until this point you can withdraw without having to give a reason for this, and this will not affect your medical or legal rights.

WHAT WILL PARTICIPATING INVOLVE?

Taking part in this study will involve meeting with the researcher for a one-to-one interview, which will typically last between one and two hours. This will involve discussing your experience of having a prophylactic mastectomy and will be recorded (audio only) and then transcribed. This interview will then be anonymised and used in the write-up of this research. Direct quotes will be used but your name and personal information will not be disclosed.

WHERE WILL THE RESEARCH TAKE PLACE?

These interviews will take place at your local NHS hospital when possible, however if this is not possible due to availability of rooms or logistical issues, interviews can be carried out at the University of Hull.

EXPENSES AND PAYMENTS

Your participation in this study is voluntary. However, you will be reimbursed for any travel expenses if you visit a hospital or the University of Hull to take part.

WHAT ARE THE BENEFITS AND RISKS OF TAKING PART?

This study involves little risk. However, it is possible that you may experience some distress and upset discussing your surgery if this is an emotional subject for you. There are no identifiable medical benefits to taking part in this research, however it is hoped that the results of this study will help with the development of services for women in the future.

ANONYMITY AND CONFIDENTIALITY

Any personal information obtained in the study will be accessible only by the researcher and their research supervisor. Your GP will be informed of your participation but will not have access to any information that you share. Any personal data will only be used for the purposes of this research project. All information is stored securely for 10 years and will then be destroyed. Information is collected by myself only and all information will be anonymised and participants will not be identified by name at any point in the write-up of this research. Personal information will be shared if there is a duty of care to the participant in the event that the researcher deems there to be a risk to your safety or the safety of others. In these circumstances, information will be shared with third parties which may include your GP or the police. Any action taken in the event of this will be discussed with you beforehand. We will follow ethical and legal practice and all information about you will be handled in confidence.

WHAT WILL HAPPEN WITH THE RESULTS OF THE STUDY?

The results of this study will be presented in a doctoral thesis, submitted for publication in an academic journal and may be presented at conferences. No individual participant details will be identified in the presentation of data.

WHO IS ORGANISING THIS STUDY?

This research is carried out as part of a doctorate level training program in clinical psychology with approval of Humber NHS foundation trust.

WHAT IF THERE IS A PROBLEM?

If you have concerns about any aspects of this study you can contact Dr Tim Alexander at the University of Hull (T.Alexander@Hull.ac.uk/ 01482 464008). You can also contact the local NHS Patient and Advice and Liaison Service (PALS) on telephone number 01482 303 966 or via email: pals@humber.nhs.uk.

WHAT SHOULD I DO NEXT?

If you wish to take part please inform the member of staff, they will then be able to advise you about what to do next.

FOR FURTHER INFORMATION

Miss Emily Rawding and Dr Emma Lewis will be happy to answer any questions about this study at any time:

Email: e.rawding@2012.hull.ac.uk/e.lewis@hull.ac.uk

Address: Miss Emily Rawding/Dr Emma Lewis, Doctorate in Clinical Psychology, Aire Building, University of Hull, Cottingham Road, Hull, HU6 7RX

Thank you for taking the time to read this letter!

Yours Sincerely

**Emily Rawding
Trainee Clinical Psychologist**

Supervised by

**Dr Emma Lewis
Clinical Psychologist**

Appendix I - Consent form

CONSENT FORM

Title of Project: Experiences of women who undergo prophylactic mastectomies following BRCA 1 or 2 gene diagnosis

Name of Researcher: Emily Rawding

Please initial boxes

1. I confirm that I have read and understand the information sheet dated for the above study. I have had the opportunity to consider the information. If I had any questions, they have been answered satisfactorily.

☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason up to the point of data analysis and transcription, without my medical care or legal rights being affected.

☐

3. I confirm that direct quotes from the interview may be used in future publications.

☐

4. I understand that any use of information or direct quotes from my interviews used in publications will be anonymised.

☐

5. I understand that data collected during the study may be looked at by individuals from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.

☐

6. I consent to my interview being audio recorded.

☐

7. I agree to take part in the interview part of the study.

☐

Name of participant	Date	Signature
_____	_____	_____
_____	_____	_____

Name of person taking consent	Date	Signature
_____	_____	_____
_____	_____	_____

Appendix J - General interview schedule

Example general interview schedule/questions

- Could you tell me what your experience of having a preventative mastectomy was like?
- How did you come to have the surgery?
- What happened before that?
- What happened after that?
- How did you feel about the surgery?
- What was that like?
- Could you tell me more about that?
- How did you feel then?

Appendix K - Debrief/sources of support form

Experiences of women who have undergone prophylactic mastectomy following BRCA 1 or 2 gene diagnosis

Debrief Sheet

Dear Participant,

Thank you for giving your time to take part in this research, your input is greatly appreciated.

The research you have taken part in is interested in understanding the experiences of women like yourself who undergo prophylactic mastectomies following BRCA 1 or 2 gene diagnosis. The hope is that in understanding the experiences of these women, and the impact that this surgery process may have on women's psychological well-being, services will be better equipped to support them in the future.

Please see the attached document which contains information about sources of support for individuals who have undergone a prophylactic mastectomy and who are diagnosed with the BRCA 1 or 2 gene. The document also contains information regarding local support for any difficulties with psychological well-being. Additionally you are reminded that you can speak to your General Practitioner, or the member of staff who has helped facilitate your involvement in this research, if you require further support.

If you have any further comments or concerns, please contact Dr Emma Lewis at e.lewis@hull.ac.uk.

Many thanks,

Emily Rawding
Trainee Clinical Psychologist

Sources of support and information regarding Prophylactic Mastectomy

Macmillan Cancer Support offer information and advice regarding prophylactic mastectomy and the emotional impact of this surgery, as well as forums for support:

<https://www.macmillan.org.uk/>

Confidential helplines: 0808 808 00 00

Monday-Friday, 9am-8pm

Breast Cancer Care offer advice and information regarding mastectomy and the BRCA genes:

<https://www.breastcancercare.org.uk>

Speak to a breast care nurse: 0808 800 6000

Let's Talk Hull offer help and support for anxiety, stress and depression:

<http://www.letstalkhull.co.uk>

To discuss talking therapies, call 01482 247111 or Text TALK to 61825

Should you have any specific issues regarding your treatment that taking part in this study has raised then you can contact the **Researcher** at:

e.rawding@2012.hull.ac.uk

You can also seek advice from your GP

Appendix L - GP Letter

Doctorate in Clinical Psychology Course

School of Health and Social Care
Faculty of Health Sciences

Aire Building

University of Hull

(Date)

HU6 7RX

(Address of GP)

PRIVATE AND CONFIDENTIAL

Dear (name of GP),

RE: (name of patient)

DOB: (DOB of patient)

I am writing to inform you of your patient's involvement in a research study being conducted by myself as part of my training on the University of Hull Doctorate in Clinical Psychology Course.

The study aims to understand the experiences of women who undergo prophylactic mastectomies for the prevention of breast cancer following diagnosis of the BRCA 1 or BRCA 2 breast and ovarian cancer susceptibility genes. Participation in the study involves taking part in a 1-2 hour long interview, which is audio recorded and then transcribed. This information is then anonymised and will be used in the write-up of a qualitative report. The research aims to identify how these women describe their experiences, from the decision to have surgery to the adjustment to life after surgery, and how this affects their psychological wellbeing and/or levels of distress at all.

All participants will be provided with information on how to seek further support if necessary following their involvement in the study.

If you have any questions or queries regarding this, please contact me at e.rawding@nhs.net. The research project is being supervised by Dr Emma Lewis, Clinical Psychologist. You are also able to contact her at emma.lewis7@nhs.net.

Yours sincerely,

Emily Rawding
Trainee Clinical Psychology.

Appendix M - Example of annotated transcript

Sub-ordinate theme	Transcript	Exploratory comments
	E: So when you had the surgery what was it like?	
Expectations vs Reality	L: Urm...you expect a grand hall theatre don't you? (laughs) It was like a...cubicle... (laughs) Yeah I was, I was not wowed by my theatre at all (laughs) They were lovely, they were really really nice, urm...I remember feeling just like hmm...making me feel a bit dizzy this and then that was it, gone, urm...I felt really really sick when I woke up, urm, and vomited really quickly, bright green, it was pleasant, yeah, really nice...(laugh) urm...and then yeah just.....yeah.....didn't really feel pain...when I first woke up of any kind, just...I knew where I was, it was fine, Steve came to see me...urm...saw my surgeon in the first bit where...recovery room? Where you first wake up, I saw her there and	Suggestive of surgery as performance Reality of surgery not being as she expected
Normality	uh...she was clocking off for the day	Surgeon goes back to
Things being different	she's like I'm off into town! (laugh) I	normal, things are

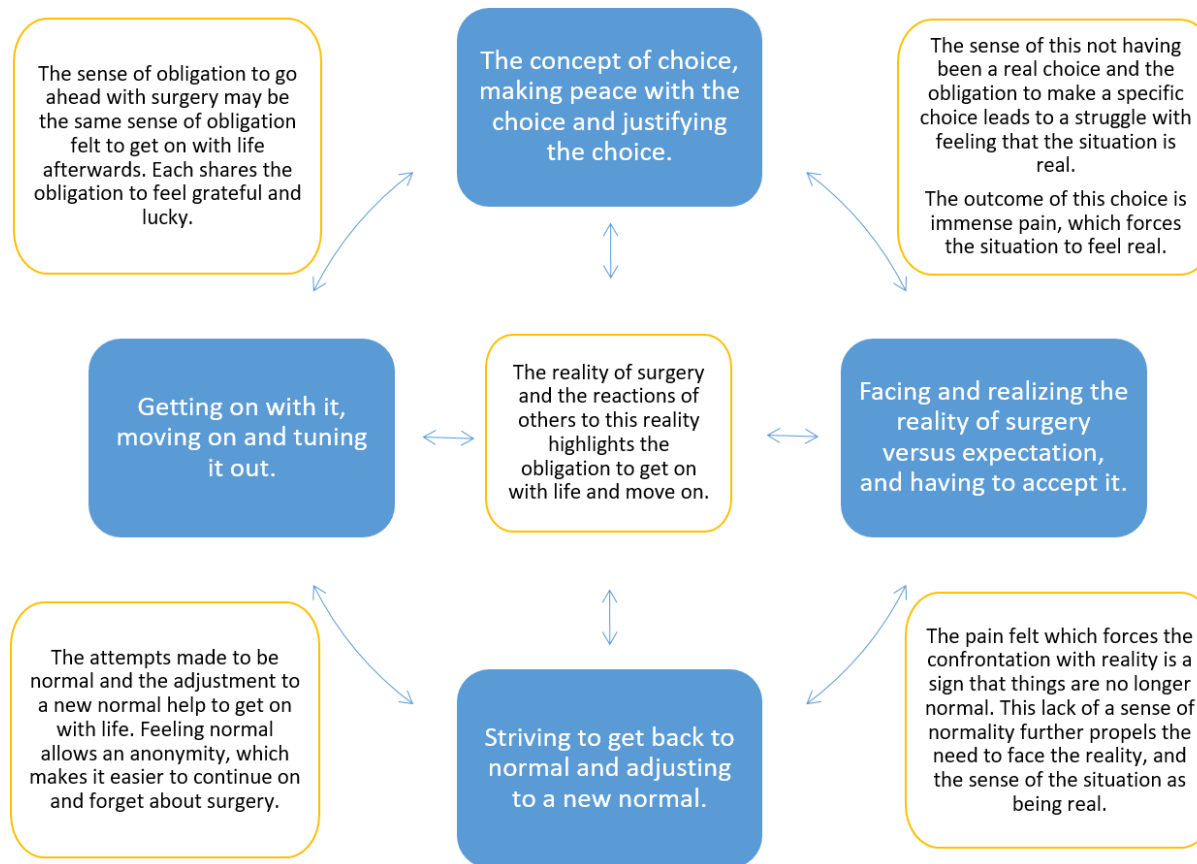
	remember all that, ringing Steve to say	not normal for
	I was awake and it had all gone alright.	Lindsay
	Urm...but then yeah, got moved to the ward and that was it. Hell night after	'Hell' gives image of worst situation
Wanting things over with	that. There was this woman on the ward that just...was a nut job...so I	imaginable, strong language
Wanting to get back to normal	didn't get any rest, and...I just couldn't wait to get out of there. It was horrible.	Wants to go home,
Denial of the sick role	If I could have just gone home instantly I would have done. She was just...she was just a nightmare patient.	perhaps where she isn't a patient, back to normal
Pain	But I made a really good friend on the ward, the lady opposite me, urm, she'd had a double mastectomy the day	
The physical impact and reality of surgery	before...and she'd actually been due to go home, urm, but she'd yanked on one of her drains...and...it's excruciating...and urm...to the point where she nearly passed out and they kept her in another night. But that extra night made me meet her, and we've kept in touch ever since. Regularly meet up and stuff...urm...urm, different story to mine completely urm, but yeah, it was really good so we had	Graphic memories of pain and surgery, a prominent experience. The reality of surgery as painful and difficult
Wanting to get back	each other, and this nut job (laughs)	

to normal	but yeah, it was a long night, and I	
	couldn't wait to get home and I had	
	really high blood pressure, and I	
	remember the nurse – the head nurse	Wanting to go home
	on the ward – she didn't want me to go	
Denial and avoidance	home...urm...because of my high	
of the reality	blood pressure. And I remember	
Struggling with the	thinking I can't be in here another	
reality	night cause I'll flip out. I couldn't...I	
	couldn't, I needed to get the	Creates imagery of
	window...I felt	feeling suffocated
	really...claustrophobic.....almost....not	
	claustrophobic cause I wasn't in a	
Feeling overwhelmed	small area but I just needed fresh air. I	
	felt really stifled. I was nearest the	
	window and I kept going up to the	'Stifled' suggests
	window and...I avoided going to the	being overwhelmed
	loo as much as I could. Like I'd really	by the reality
	try and stop myself because obviously	
Denial of reality	as soon as I moved, I had my drains,	
	and if I didn't move I could pretend oh	
	yeah I'm just lying here it's all fine,	
	but then as soon as I moved I had these	Wanting to pretend
	horrible drains to contend with and that	things weren't
	made it a bit more realistic. And	happening
	because I hadn't slept, it just felt really	

	long weird time...urm...had those	Avoiding the reality
	horrible stocking things on...uh,	Physical impact
	god!....urm....and then the nurse kept	makes it feel real
	coming in and then the nurse kept	
	coming in and making up medication,	
	and, it was just really noisy and yeah, it	
	just didn't happen. I just didn't sleep. I	
	must have had about an hour. I had	
	music on but then she, nightmare	
	woman, went to sleep and snored. I had	
	music on, I tried to like, put myself	
	into a nice relaxed mode to sleep, just	
	didn't happen. I don't know whether it	
	was the medication or...or I was too	
	alert, I don't know, but it was a long	
	night. It really was. Urm...	
	E: You talked about avoiding the toilet	
	because of drains and if you did go it	
	made it more real, can you tell me	
	more about what that was like?	
	L: Well I obviously knew why I was	'They' – othering of
	there, urm...I was still a bit worried	medical staff,
	that I'd go woozy...and if I feel pain, I	imagery of being
Being passive	might go woozy. And I really wanted	'done to' and being a
recipient	to go home (laughs) and I really	passive recipient
Choice/agency	thought again if I faint on them they're	Ideas of being 'kept'

	gonna keep me here	cause I knew what as if imprisoned
	had happened to my friend cause she	
	stayed another night and I was really	
	anxious then if I moved and wasn't	
	quite familiar with where they were yet	
	and where things were attached...I	
	thought if I cause myself that pain	Suggest blame/fault
Getting on with it	they're gonna keep me in. so I was	lies with her
Being a good patient	trying to be good. Slow movements,	Trying to be good'
	and, and things like that but...yeah...I	suggest a sense of
	did have to go to the loo eventually	there being a correct
	(laughs) and it was fine. But then	way to behave, being
	it...it...you got all sorts of new	the perfect patient,
	feelings and pains, so then it made it	obligation to be
Denial and avoidance	more real that, oh my god, yeah, this	'good'
of reality	has happened. This is different. And	Wanting to ignore the
Things are no longer 'normal'	then its learning about the drains, and	reality, struggling to
	where they were attached and	face it
	movements you can make and	
	shouldn't make and...yeah...totally	
	different. Totally different weird	
	feeling.	

Appendix N - Diagram to show interactions and links between themes



Appendix O. Lindsay's quotes regarding post-surgery support

“There’s nothing for BRCA...there is, there’s on Facebook...I don’t wanna join a group in Facebook cause anyone can join a group on Facebook and see that you’re in the group and it’s a small bloomin world when you’re, you know...so I – I can’t join any of those groups cause I don’t want...I wanna be anonymous”

“I don’t know if there’s a random nurse out there that can (laugh) have a mobile phone waiting for random people like me to say “yeah this shooting pain feels like flashing lightening, is that normal? Are you getting that?” (laugh) but yeah just...cause you just don’t know”

“So there was that literature and then this exercise literature, but there want anything else. She explained about the drains to us in person, and when Steve was there as well, thank god, ‘cause I don’t think I would have taken it in. Urm...urm....but yeah there want anything else. Some sort of extra leaflet saying you might experience shooting pains, or throbbing, or...hmm...you might make sudden movement and have a pain – all these are normal. Summat like that would be...yeah...’cause I had soooo much stuff given to me beforehand, it was unreal. I’ve still got it all as well. Yeah...booklets on family history of cancer, booklets on mastectomy, booklets on implants, booklets on...surgery and different types of surgery and...yeah....all that information before, but suddenly it’s...yeah...pretty cold turkey afterwards...”

“The other thing I think, if you can do it for me...I really do think, urm...you sh-...uh...there was the option, don’t get me wrong, I could have rung up and said I wanna speak to that counsellor again...urm...but I didn’t...but I think you should be made to...’cause I think it would’ve helped. Cause your emotions are all over the place, your body’s changing, cause you’re swollen and you’re losing it...and your more accepting of what’s happened and what you look like...urm...but yeah, I, I, definitely experienced

...just being...sad...probably...so I had my surgery in November, and around January, February, whether it just hit me or not, what I'd been through, but yeah...I did...have a big cry. Around about that time. So whether it would have helped, someone enforcing me right go and speak to this person...Four months five months six months after...that would have been probably beneficial...but yo-you just get on with it”

“Whether I should have made that phone call myself, I don't know, to speak to somebody, erm, but...I sort of always expected, I was still having appointments with the surgeon...so...I could have said to her would you mind booking me in...but I never did...urm...and I sort of always expected something...afterwards anyway. My friend got like, urm...she went to exercise classes and stuff like that, and I never did...urm...and it was because she'd had her lymph nodes out and I didn't, cause I was just having...them chopped off...so, I didn't need any of that. But she'd had cancer cells, so they took her lymph nodes out, so she had a different...she had more pain...so I never, she went to all these groups, walking groups and all sorts of different things...and then, she met people who had been through her same journey that she sort of gone of, and...she talked to me about it...so it was quite good, but then I – I – I wondered if I should go on any of these things, or I'd missed a letter or, I'd been missed out...and...since Steve had spoke up and said how come she's not gone on anything and she just said oh it's just not necessary. So...fine, that's fine, but...if I wouldn't have known, I'd have been none the wiser if it want for me friends, but...yeah...I don't know...maybe just another open day for everybody. Do a group, a class of...urm...exercises and...I don't know”

Appendix P - Reflective statement

The research topic

When I began exploring ideas for my thesis, I found it difficult to select just one area of interest from the many things I was interested in to focus on. I decided it made sense to be logical, and to make my decision based not only on my interests and passions – because there were so many – but on the experience of the research supervisors available. I wanted to root my thesis subject firmly in the expertise of a supervisor, to ensure that I would have the necessary knowledge and support available to me during what felt then like such a daunting process. Health psychology quickly seemed to be the perfect mix of passion and logic. I enjoyed the cross over between psychology and medicine, and had identified my now supervisor Emma as someone that I felt I would work well with, and who would provide the necessary expertise I needed. My gravitation towards a research topic which focused on women's health issues, as a woman, admittedly seemed to happen without much conscious thought. During the process of first developing thesis ideas and meeting with supervisors, I was drawn in by Emma's interest in researching the female BRCA population. I knew very little about the gene, its impact and the process that women went through from diagnosis onwards, but it caught my attention quickly. Emma explained how minimal the research was in this area and how little support these women were offered after making life altering decisions, such as the decision to remove reproductive organs or breasts to prevent disease, with no certainty that these measures are life-saving. The idea of what these women were going through, and the idea that very few people were talking to them about it, was something that I struggled to ignore, and so began my research journey.

During a free-writing exercise in a research study group, we were asked to write without thought about our research topic. The question of "Why have you chosen this topic?"

was one many of my cohort were struggling to answer thoroughly, so the task was put to us. Although I knew that my emotional investment in women's issues generally was a large motivating factor for my research topic, I knew that there must be more to it. Why was I drawn to these women and their experiences? Why was I so invested in giving them a voice? It was within that task that I began to reflect on what now seems to me an obvious link between myself and these women: my endometriosis diagnosis. Despite the differences in our diagnoses, and the heightened significance and risk of theirs as it compares to mine, it seems this population and I share some similar experiences: the increased threat of gynaecological cancer, potentially reduced fertility, the need to pursue elective surgeries. In the early stages of my thesis process, I took a month off to have an elective surgery and realised then why women's surgery, most specifically as it relates to prevention and an invisible cause, mattered so much to me.

Recruitment

When I began my thesis, I expected that recruitment would be a reasonably simple and straight-forward part of the process. My supervisor Emma and I had identified hospitals and clinicians to aid recruitment and expected that finding 6-12 participants to meet the ideal sample size for IPA analysis would be easy to do. The recruitment process was slowed somewhat by the ethics process, and by the process of gaining access to the trusts used for recruitment. This took longer than I expected, and navigating communication with several different people with conflicting work schedules was harder than I anticipated it would be. I am somewhat regretful that I didn't begin the process of applying for letters of access earlier. The process of recruitment for this study has been heavily reliant upon help and support from clinicians, which was unavoidable due to the population I needed to recruit. Accessing women via medical professionals who had been involved in their care during the surgery process was the only way to access women whilst upholding data protection boundaries, and so this process took a

long time. Many emails were exchanged, and things posted back and forth. Less women than we anticipated were able to be identified and eligible for participation. I found recruitment, and whilst writing this am still finding recruitment, the most frustrating part of my empirical research process. It has been a long process, but one that I have remained committed to. I understand that it perhaps would have been easier if I had explored other avenues of recruitment, such as social media and online forums, but I was reluctant to do this from the beginning of my thesis. I worried it would create a bias sample of women who were already actively engaging in sharing their stories and whose stories were therefore likely to be extreme, either positively or negatively. I was far more interested in recruiting women through hospitals who were local and perhaps less biased, especially as local services would be the services with which I felt I would have most luck engaging with post-research to share my findings in the hopes they may be implemented in the development of psychological services, if necessary. Recruiting through NHS trusts also provided me with some security as provided an easily accessible avenue for support if my interviews had been a cause of any distress for participants, or if any clinical risks or psychological wellbeing issues had been raised. I therefore continued in my efforts to recruit via my originally planned method. I am glad that I made this decision, despite all of the ways that it felt it hindered my project's progress.

It feels important to highlight that this study was never intended to have only three participants. It had been planned that there would be between 6-12 participants, and up until two months before submitting this thesis, that felt possible. Several things went wrong, were under or over-estimated, and things didn't work out as we had hoped. The decision to submit this thesis with only three participants was a difficult one. So much time and effort, from my both myself and the many clinicians and trainee clinicians who helped me, went into recruiting just those three. Their stories were similar and

contrasting in ways that felt not only interesting, but in-keeping with the requirements of IPA analysis. I sought advice from experienced researcher, Dr Emma Wolverson, who suggested I speak to experienced IPA researchers who may be able to offer some more specific advice relating to the small sample size. I contacted Dr Rachel Shaw at London's Aston University. Like most people I have reached out to during my thesis journey, she was kind and helpful. She told me that an IPA study would suit a small sample size of three participants due to its focus on idiographic information as well as more general themes. I have included my correspondence with Rachel in the Appendices of this thesis, for reference (see Appendix R). My sample size is not what I planned, but finding my three women was difficult and at times exhausting for myself and I am sure others involved. I feel that my analysis is strong, that the data was rich, and that I have produced a good piece of first-time IPA research, and I am proud of it. Just as proud as I would have been with the sample size of my dreams. There are different decisions I could have made during recruitment, but they may have compromised the homogeneity of my sample and the security of my participants should anything have upset or distressed them. I could have recruited from other local trusts, without this problem, but I believed that I had sufficient participants up until a week before beginning interviews, and was worried about over-recruitment. This research experience has taught me a lot of lessons, most of which for me relate to recruitment and how long the process takes. I would perhaps do things differently next time, but regardless am proud of my work and feel that my small sample size resulted in very little compromise when it comes to what my research can offer, especially as IPA – and therefore my research – has never been about making sweeping statements. In a field of research which remains small, providing what I understand to be the first piece of IPA research on the experiences women have of prophylactic mastectomy following BRCA

diagnosis, I believe this research can contribute a great foundation of understanding and insight upon which much more research can be built.

Data Collection and Analysis

I was, as I imagine many are, nervous to begin my data collection. I was worried about conducting interviews because of the pressure I felt to absorb everything; I was worried that if I didn't, my final empirical paper would lack an ability to truly reflect my participants' experiences the way I wanted it do. I am grateful for the kindness and co-operation of the women who took part in my interviews. Each of them was welcoming and this helped tremendously with easing my nerves. I thoroughly enjoyed conducting my interviews and gathering data. Sitting with my participants and hearing about their experiences first-hand continues to feel like a privilege.

The data analysis was something else that made me nervous about my thesis. I was worried about getting it 'wrong', and so I studied books on IPA and qualitative methods throughout my thesis process, even before analysis had begun. I sought advice from experienced IPA researchers, and shared my themes with both them and fellow trainees to help me to take a step back from my data. By the time it came to completing my analysis, I felt somewhat more confident; I felt that I understood IPA and accepted the advice that it was less about getting things 'right' or 'wrong', and more about being a 'good enough researcher'. I enjoyed my data analysis, despite finding it difficult at times to see my themes. It took several attempts to be able to separate myself from making procedural notes about the data, and truly focus on themes and language rather than explicit content. I think this difficulty with the 'interpretative' element of IPA is common, and it helped to talk about it with other researchers. However challenging it felt at times, I enjoyed finding emerging themes and pouring over my data for long periods of time, most likely because I had been so engrossed in the interviews, and

therefore getting closer to the data, so to speak, was interesting. When I finally settled on my themes and understood what they were, it all seemed to come together, and the hours spent feeling confused started to make sense too as a part of the process.

Write up and Journal Selection

I have always enjoyed writing, both in and out of academic contexts, and so I took great pleasure in writing up my thesis. It was the part of the thesis process, from conception to completion, which I felt the most confident and comfortable with. The most difficult part about this process was writing my empirical paperwork within the word limit constraints of the British Journal of Health Psychology. I had settled on writing for the British Journal of Health Psychology before I collected my data because it seemed a good fit and would mean that my research reached its intended audience; professionals working within health psychology in the UK. This seemed to be the audience for whom my work would have the greatest use and application. I had decided against writing for an oncological journal, as it felt important to me to separate the BRCA population from cancer populations. After conducting my interviews and reaching this stage, I remain happy with that decision. My participants seemed to draw a differentiation between themselves as women diagnosed with BRCA and cancer populations, and so it seems in-keeping with their sense of identity to write for a journal aimed more generally at health psychology, rather than psycho-oncology.

Systematic Literature Review

Upon reflection, I think that I was drawn to my literature review topic for the same reasons that I was drawn to the BRCA population as a whole. In the lead up to carrying out my final data collection and analysis for the review, I carried out many preliminary, pilot searches of various terms. I was determined to ensure my SLR focused upon a topic closely linked to my empirical research, and focused upon the same population of

women. I was hesitant to venture into cancer populations, in part because so much research exists on cancer-diagnosed populations when compared with the un-diagnosed BRCA population. I explored several research topics within this population and struggled to find the right question which both satisfied my desires as a researcher and provided enough literature for review. The experiences that women diagnosed with BRCA have of family planning interested me immediately and felt an important topic to explore. It remained difficult to gather a lot of literature for review, because the population is so under-researched, but I felt very strongly that the literature I did find was important for what it communicated about women living with BRCA and their experiences of planning for and having families. I was happy that my data became very saturated very quickly, and so the small number of papers included in my review became less of a concern as I commenced the data extraction and analysis process. I found my SLR enjoyable to research and write up, which I had expected that I would as this type of work has always felt somewhat more natural to me than carrying out empirical research of my own.

Personal values and experiences

As much as my personal experiences of health, illness, surgery and healthcare in some ways motivated my passion for this thesis topic, I felt capable of refraining from developing too many assumptions of what my data would show. My personal experiences, and thus views and perspective, are varied and I therefore respected that this range would likely exist within my participant population, too. I reflected on my thoughts and feelings and understood very much that diagnosis, surgery, illness and pain are perhaps not inherently positive or negative; that there exists a unique and intricate interaction between all of these things, how they make us feel, and how they affect – or don't affect – us. Being a woman with some experience of surgery felt helpful when understanding the medicalised language of my participants but the great

distance between my experience and theirs felt necessary to be as impartial as possible. IPA analysis acknowledges the researcher as a research tool and the idiosyncrasies that come with interpretation from one researcher to another, and so I made sure to reflect on my experiences throughout the process of interpretation.

Final Reflections

When I began my doctorate, there were elements of the programme which appealed more to me than others, and parts that I felt would come more naturally to me. I was motivated most by the opportunities it would provide me to work clinically and to help others, and secondly by my deep love for academic work and essay writing. The research element of the doctorate was, admittedly, the part that least sparked my enthusiasm. I think I under-estimated its importance not only in regards to how big of an undertaking it would be, but with regards to how important it is within the field to contribute to the shaping of service development and ongoing scientific or psychological knowledge in this way. I have enjoyed this process of research more than I anticipated I would. It has been stressful, tiring and at times overwhelming but it has also expanded my confidence in myself not only as an academic, but as a clinician. I feel more embedded in and a-part-of the scientific, psychological community because of this work. Research – both as a researcher and as a beneficiary of other people’s research – means more to me now than it has done before. The help I was provided by other professionals during my thesis research is something that I will carry forward with me in my clinical career as a “how-to” guide of sorts. Each of them greeted me with enthusiasm, going beyond what would ever be expected of them to help me carry out this research. I only hope that I will get the opportunity to aid someone else’s research journey in the future the way that others have aided mine.

I am excited for my journey as a clinician, and hope that I can carry out further research in the future. I understand now that my job is not just about my clinical work, but about the contributions I can make to the way we understand what is needed within that clinical work through research. I feel incredibly proud to be submitting this thesis at all, and humbled by the endurance and resilience I have needed to develop to maintain my stamina during the difficult parts of my doctorate journey. It has been an expanding process from start to finish and I feel indebted to the university and to my employing trust for the opportunity.

Appendix Q. Epistemological Statement

As research is based upon the production of knowledge which one can believe to be valid (Green & Thorogood, 2014), there are many theoretical approaches to both epistemology and ontology in the conduction of research. It is therefore important to consider the theoretical underpinnings of this thesis, including its epistemological stance and ontological assumptions. This epistemological statement is therefore intended to provide transparency regarding the assumptions behind this work, the philosophical position of the researcher, and the influence of these factors on this research.

Ontology is the study of what exists; therefore, the study of truth (Effingham, 2013). Epistemology is an area of philosophy concerned with the theory of knowledge. It is concerned with understanding not only what we know about the world, but what justifies the beliefs we hold about the truth, and what evidence we should use when seeking to know the truth, and how we have come to have belief in that knowledge and its validity (Audi, 2010; Green & Thorogood, 2014). Therefore one is concerned with what is true, and the other is concerned with how we know that, and what leads us to believe it.

A positivist approach suggests a stable reality, or truth, and that the existence of this does not rely upon being understood; it will exist whether it is looked at or not.

Positivism therefore creates realist approaches to research, which assume the existence of a reality which is separate from our understanding and this belief serves as a starting point, and motivator, for research (Green & Thorogood, 2014). Research is therefore motivated by the idea that the more knowledge gained about objective truths through research, the better we will understand these truths. This type of research focuses on empiricism, studying only observable phenomena, and a value-free inquiry, imploring science and truth to be held as separate from society and thus the researcher and their

subjective viewpoints. In positivism, the truth exists objectively and will remain a truth regardless of time, place and other subjective factors (Green & Thorogood, 2014). This approach to research is common within quantitative research, but has been questioned and challenged over time, and is often not well-suited to qualitative research (Green & Thorogood, 2014). The complexity and unpredictability of human behaviour is therefore difficult for some to consider researching with a positivist perspective. Many feel that the attainment of objective truth is an unattainable goal for research of this kind, as human beings are sense-making creatures with subjective views on research and researchers (Green & Thorogood, 2014). This anti-positivist approach to research is therefore concerned not with explaining and quantifying human behaviour and experience, but with understanding it. This leads to an interpretative approach, concerned not with the idea of reality, but with how human beings interpret this reality (Green & Thorogood, 2014).

Phenomenology suggests that everything is subject to perception, in keeping with interpretative ideas (Green & Thorogood, 2014). It suggests that life's objects are not passively understood, but perceived through subjective experiences, and therefore to understand phenomena we must understand how a 'life-world' is experienced (Green & Thorogood, 2014). This 'life-world' itself is created through behaviour and interactions. This methodological stance therefore sets apart assumptions about reality and the world, as in positivism, and instead focuses upon the idea that phenomenon are 'real' because they are experienced and treated as real, regardless of their objective 'truth' (Green & Thorogood, 2014).

The method of analysis employed in the empirical research of this thesis was Interpretative Phenomenological Analysis (IPA). IPA is concerned with understanding how individuals view and experience the world. It aims to gain insight into their thoughts as they relate to the phenomenon being researched (Willig, 2013) and to focus

upon understanding individuals' experiences of this phenomena, as well as their perceptions and views (Smith, Flowers & Larkin, 2012). IPA therefore explores the interpretation of meaning and assumes that data can tell us about people's involvement in the world and how they make sense of it. IPA focuses on how individuals make sense of significant life experiences and aims to engage with the reflections individuals have on this significance (Smith, Flowers & Larkin, 2012).

In this case, the phenomena being researched was the experience of undergoing a prophylactic mastectomy following BRCA gene diagnosis. IPA aims to gain knowledge relating to these experiences and is concerned with the subjective rather than objective experiences of others, not as truths or falsities, but rather as experiences mediated by thoughts and feelings. IPA adopts relativist ontology in this regard, and takes a symbolic interactionist perspective to experiences, understanding that meaning whilst somewhat idiosyncratic in nature is mediated and influenced by social interactions (Willig, 2013). IPA suggests that thoughts and feelings provide an attributed meaning, which creates experience. IPA recognises the importance of a researcher's own views and beliefs and their influence upon research findings, however does not incorporate or account for them within analysis. It therefore adopts a reflexive researcher approach, understanding that insights gained from research are a form of interpretation. IPA recognises researcher's beliefs not as biases, but as necessary for being able to understand the experiences of others (Willig, 2013).

IPA is concerned with how a specific phenomenon is experienced by a specific person in a specific circumstance. This is influenced with idiography, which is concerned with the particular (Smith, Flowers & Larkin, 2012). Idiography therefore takes an approach to establishing generalisations based on locating them within an individual and their particular contexts and experiences, therefore developing them more cautiously than positivist approaches before making general claims (Smith, Flowers & Larkin, 2012).

IPA is therefore a combination of phenomenological and interpretative approaches, coupling an attempt to get close to a personal experience of an individual with the understanding that this will become an interpretative endeavour as it understands both participants and researchers as sense-making beings (Smith, Flowers & Larkin, 2012).

This research was approached with a belief that human experiences are shaped by our interactions with others and can only be understood by an attempt to align ourselves with another person's 'life-world'. This will help to gain an understanding of how an individual has experienced a specific phenomenon, and what beliefs or perspectives they hold that led to this phenomenon to be experienced as 'an experience'. I understand the potential impact of my own beliefs and experiences as a sense-making being on this research, and consider these not as influences to be considered closely within this work, but as causes of a somewhat subjective interpretation of the data. I therefore understand this research to be an interpretative process. With regards to this research specifically and its concern with understanding the experiences of individuals, I am motivated not to find the 'truth' as defined by an objective entity which exists without understanding of it, but as a subjective entity which exists only with an individual's reflection upon and personal understanding of it. I understand the experiences of others to be both idiographic and shaped by interactions with the world and with other people, as well as subjective thoughts and feelings about phenomena which provide them meaning, and lead them to become 'experiences'. I acknowledge the limitations of any epistemological or ontological claims or truths. A positivist approach to this research with the aim of developing generalisable 'truths' was neither appropriate nor necessary. This research will therefore have use and contribution within this field of research not because of its ability to make generalizable statements about the 'truth', but because of its exploration of individual experiences.

References

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Smith, J.A., Flowers, P. & Larkin, M. (2012) *Interpretative Phenomenological Analysis*. London: Sage.

Willig, C. (Eds.) (2013) *Introducing Qualitative Research in Psychology*. UK: Open University Press.

Appendix R. Email exchange with Dr Rachel Shaw

From: EMILY RAWDING
Sent: Friday, May 10, 2019 3:30 PM
To: Shaw, Rachel
Subject: Re: IPA study

Hi Rachel,

Thank you for your permission, and for your advice and help today. It has been really useful and I really appreciate it.

Kind regards,

Emily

From: Shaw, Rachel <r.l.shaw@aston.ac.uk>
Sent: Friday, May 10, 2019 2:32:44 PM
To: EMILY RAWDING
Subject: Re: IPA study

Hi Emily,
Yes, I'd be happy for you to use our exchange.

Best wishes with your research.
Rachel.

From: EMILY RAWDING <E.Rawding@2012.hull.ac.uk>
Date: Friday, 10 May 2019 at 14:23
To: Rachel Shaw <r.l.shaw@aston.ac.uk>
Subject: Re: IPA study

Hi Rachel,

Thank you so much for your fast response and advice. That is really useful and having your paper to hand is useful too.

I wondered if you would be ok with me including this email exchange in my appendices, just so that I can make clear that I sought advice from experienced IPA researchers on this topic.

Kind regards,

Emily

On 10 May 2019, at 12:31, Shaw, Rachel <r.l.shaw@aston.ac.uk> wrote:

Hi Emily,

Having a sample of 3 isn't a problem for an IPA study. As you know, IPA is an idiographic study which means you do an individual analysis of each person before you make any comparisons between them.

One of my papers only has 3 participants & it works really quite well! I hope it's useful.

Best wishes,
Rachel.

From: EMILY RAWDING <E.Rawding@2012.hull.ac.uk>

Date: Friday, 10 May 2019 at 09:19

To: Rachel Shaw <r.l.shaw@aston.ac.uk>

Subject: IPA study

Dear Rachel,

My name is Emily and I am a final year student on the University of Hull's Doctorate in Clinical Psychology course.

I am currently completing my thesis which is an IPA study into experiences of women who undergo preventative mastectomies following BRCA gene diagnosis. I have 3 participants at present and am struggling to recruit more. I spoke to Dr Emma Wolverson about it to seek advice, and she suggested that I email you regarding this.

I wondered if you could offer any advice regarding completing an IPA study with so few participants. I wondered if you could perhaps point me in the direction of previous work people have completed or published with sample sizes as small as 3.

Any advice or insight you can offer would be helpful.

Kind regards,

Emily
<Shaw 2011 JCASP.pdf>