

**WOMEN'S DECISION MAKING REGARDING HEREDITARY BREAST AND  
OVARIAN CANCER RISK-REDUCING STRATEGIES**

by

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## Abstract

Women found to carry mutations in the *BRCA1* or *BRCA2* genes have up to an 88% lifetime risk of breast cancer and up to a 65% lifetime risk of ovarian cancer. Strategies to address these heightened risks include breast cancer screening, and risk-reducing (RR) surgeries (i.e., mastectomy and salpingo-oophorectomy). Some women might change their lifestyle or use complementary and alternative medicine to prevent hereditary breast and ovarian cancer (HBOC). The objectives of this research were to describe: a) the HBOC RR strategies used by women prior to receiving their genetic test results, the influence of individual and psychological factors on the uptake of these strategies, and their risk management information needs, b) how women construct the ‘right time’ to consider RR surgery decisions, and c) the process of making decisions regarding HBOC RR strategies. A survey of 143 women was conducted to address the first objective and in-depth interviews with 22 *BRCA1/2* carriers were conducted to address the remaining two objectives.

Survey respondents engaged in breast cancer screening at the time of genetic testing and a sub-group modified their lifestyle to reduce their cancer risk. Qualitative analyses revealed women’s constructions of the ‘right time’ to consider RR surgery decisions to be when: (1) decisions fit into their lives, (2) they had enough time to think about decisions, (3) they were ready emotionally, (4) all the issues and conflicts were sorted out, (5) there were better options available, and (6) the health care system was ready for them. Grounded theory analyses suggested that the overarching process of making decisions about HBOC RR strategies was one of ‘preserving the self.’ This process was shaped by the characteristics of health services, the nature of HBOC RR decisions, gendered roles, and the women’s perceived proximity to cancer. The women engaged in five decision-making styles, which were characterized by combinations of seven decision-making approaches. Findings from these three studies capture the diverse trajectories of decision making about HBOC risk management and highlight the role of personal and social context in shaping these decisions.

## Table of Contents

ABSTRACT .....	ii
TABLE OF CONTENTS .....	iii
LIST OF TABLES .....	vi
LIST OF FIGURES .....	vii
ACKNOWLEDGEMENTS .....	viii
Co-AUTHORSHIP STATEMENT .....	x
<b>CHAPTER 1: INTRODUCTION .....</b>	<b>1</b>
OVERVIEW OF THE DISSERTATION PRESENTATION .....	1
MANUSCRIPT-BASED FORMAT .....	1
RESEARCH INTRODUCTION .....	2
BACKGROUND .....	4
Risk Reduction through Cancer Prevention and Early Detection .....	4
Breast Cancer Prevention and Early Detection .....	5
Ovarian Cancer Prevention and Early Detection .....	6
HBOC RR Strategies .....	7
Health Related Decision Making .....	13
Decision Making in the Context of HBOC .....	17
The Timing of HBOC RR Decision Making .....	20
THEORETICAL LENSES .....	21
Decision Making as Gendered .....	22
Decision Making as Relational .....	23
RESEARCH PURPOSE AND OBJECTIVES .....	25
Research Objective 1 .....	25
Research Objective 2 .....	26
Research Objective 3 .....	27
RESEARCH DESIGN .....	27
REFERENCES .....	29
<b>CHAPTER 2: RISK MANAGEMENT STRATEGIES USED BY WOMEN PRIOR TO RECEIVING BRCA1/2 MUTATION TEST RESULTS: UPTAKE AND INFORMATION NEEDS .....</b>	<b>52</b>
INTRODUCTION .....	52
METHODS .....	55
Participants and Sampling Strategies .....	55
Data Collection .....	56
Data Analysis .....	57
RESULTS .....	58
Sample Characteristics .....	58
Breast Cancer Screening .....	60
Ovarian Cancer Screening .....	61
Pharmacoprevention Therapy and RR Surgery .....	63
Lifestyle Behaviour Change and CAM Use .....	64
Information Needs .....	65
DISCUSSION .....	66
CONCLUSION .....	71
REFERENCES .....	72
<b>CHAPTER 3: WOMEN'S CONSTRUCTIONS OF THE 'RIGHT TIME' TO CONSIDER DECISIONS ABOUT RISK-REDUCING MASTECTOMY AND RISK-REDUCING OOPHRECTOMY .....</b>	<b>82</b>
INTRODUCTION .....	82
BACKGROUND LITERATURE .....	82
METHODS .....	85

Study Participants .....	85
Data Collection Procedures .....	86
Data Analysis Procedures .....	87
FINDINGS .....	87
One Decision at a Time .....	88
When Decisions Fit into my Life .....	89
When I Have Taken Enough Time to Think About Decisions .....	90
When I am Ready Emotionally to Deal with Decisions .....	91
When all of the Issues and Conflicts are Sorted Out .....	92
When there are Better Options .....	93
When the System is Ready for Me .....	94
DISCUSSION .....	95
CONCLUSION .....	99
REFERENCES .....	101
<b>CHAPTER 4: PRESERVING THE SELF: THE PROCESS OF DECISION MAKING ABOUT HEREDITARY BREAST AND OVARIAN CANCER RISK REDUCTION .....</b>	<b>109</b>
INTRODUCTION .....	109
BACKGROUND LITERATURE .....	109
METHODS .....	111
Participants and Sampling Strategies .....	111
Data Collection Procedures .....	112
Data Analysis Procedures .....	112
Guiding Analytical Theories: Gender and Relational Autonomy .....	113
Sample Characteristics .....	113
FINDINGS .....	114
The Process of Preserving the Self: HBOC RR Decision Making .....	114
Contexts Shaping HBOC RR Decision Making .....	117
Decision-Making Approaches .....	120
HBOC RR Decision-Making Styles .....	126
DISCUSSION .....	131
STUDY LIMITATIONS .....	136
STUDY IMPLICATIONS .....	137
REFERENCES .....	138
<b>CHAPTER 5: CONCLUSION .....</b>	<b>148</b>
OVERVIEW .....	148
INTEGRATING THE RESEARCH FINDINGS .....	149
Contributions to Decision Making and Health Behaviour Theory .....	149
Diverse Trajectories of Decision Making .....	152
Family Matters .....	154
Psychological Considerations .....	155
Interacting with Health Care Professionals and the Health Care System .....	157
RESEARCH STRENGTHS AND LIMITATIONS .....	158
IMPLICATIONS FOR HEALTH CARE PROFESSIONALS .....	160
IMPLICATIONS FOR HEALTH POLICY .....	165
IMPLICATIONS FOR THEORIZING: RELATIONAL AUTONOMY .....	166
LOOKING TO THE FUTURE: RECOMMENDATIONS FOR RESEARCH .....	166
CONCLUSION .....	168
REFERENCES .....	170
<b>APPENDIX A : WOMEN'S DECISION MAKING ABOUT RISK-REDUCING STRATEGIES IN THE CONTEXT OF HEREDITARY BREAST AND OVARIAN CANCER .....</b>	<b>182</b>
<b>APPENDIX B : HEREDITARY BREAST AND OVARIAN CANCER RISK-REDUCING STRATEGIES QUESTIONNAIRE .....</b>	<b>219</b>
<b>APPENDIX C : ETHICS APPROVAL CERTIFICATES .....</b>	<b>225</b>
<b>APPENDIX D : CONSENT FORM .....</b>	<b>228</b>

<b>APPENDIX E : INTERVIEW GUIDE FOR <i>BRCA1/2</i> CARRIER WOMEN .....</b>	<b>231</b>
<b>APPENDIX F : DEMOGRAPHIC DATA &amp; CANCER HISTORY FORM.....</b>	<b>233</b>

## List of Tables

Table 2.1 Study 1: Characteristics of Study Sample .....	59
Table 2.2 Cancer Screening .....	62
Table 2.3 Pharmacoprevention and RR Surgery .....	63
Table 2.4 Lifestyle and CAM Cancer Prevention Strategies .....	64
Table 2.5 Information Needs about Reducing the Risk of Cancer.....	66
Table 3.1 Study 2: Characteristics of Study Sample.....	86
Table 4.1 Study 3: Characteristics of Study Sample.....	114
Table 4.2 Descriptions and Examples of Decision-Making Approaches.....	121
Table 4.3 Decision-Making Styles Characterized by Decision-Making Approaches.....	127
Table 4.4 Participants' use of Decision-Making Styles for RR Strategies .....	127
Table A.1: Quantitative Research Studies Reviewed.....	212
Table A.2: Qualitative Research Studies Reviewed.....	218

**List of Figures**

Figure 4.1: Preserving the Self through HBOC RR Decisions.....	115
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This statement of co-authorship is in accordance with the Faculty of Graduate Studies at the University of British Columbia. Acknowledgement of co-authorship addresses contributions related to identification and design of the research project, conducting the research, data analysis and interpretation, and manuscript preparation. Dissertation committee members commonly oversee all steps that lead up to the creation of the final dissertation document; however, since this dissertation consists of a series of co-authored manuscripts submitted for publication, the Faculty of Graduate Studies at the University of British Columbia requires a statement that acknowledges the significant contributions made by dissertation committee members.

Fuchsia Howard originally conceived the research project design, collected the data, performed the analysis of data, interpreted the findings from data analysis, and she was responsible for the writing of this dissertation document. While Ms. Howard was the primary contributor, her dissertation committee members were instrumental supporters of the research project process and the production of the final dissertation document.

Drs. Balneaves and Bottorff were the dissertation supervisors. They provided ongoing guidance and input throughout the process of formulating and conducting the research. Concerning the writing of the manuscripts, they were the initial reviewers of the drafts of each chapter in this dissertation. Their feedback shaped the presentation of ideas in each of the manuscripts as well as strengthened the analysis and interpretation of data. Both Drs. Balneaves and Bottorff contributed significantly to Chapters 2, 3, and 4, and, therefore, are recognized as co-authors on the submitted manuscripts.

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## **CHAPTER 1: Introduction**

### **Overview of the Dissertation Presentation**

This doctoral dissertation investigated cancer risk-reducing (RR) strategies used by women at high risk for hereditary breast and ovarian cancer (HBOC) and the processes by which these women make decisions about risk management. This research consisted of three studies. For the purposes of this doctoral dissertation, this research is presented in a manuscript-based format. The first chapter introduces the overall structure of this dissertation, highlights essential background information, and outlines the research objectives. Chapter 2 describes the risk management strategies used by women prior to receiving their *BRCA1/2* mutation test results, as well as their information needs at this time. Chapter 3 illustrates how women who carry *BRCA1/2* mutations constructed the ‘right time’ to consider decisions about RR surgeries. In Chapter 4, a theory of how *BRCA1/2* mutation carrier women make decisions about RR strategies is presented. Lastly, Chapter 5 integrates the findings from the three studies, discusses these findings in light of current research, describes the implications of this research for health care professionals, health policy, and theorizing about relational autonomy, delineates research strengths and limitations, and provides recommendations for future research.

### **Manuscript-Based Format**

The University of British Columbia Faculty of Graduate Studies and the School of Nursing have approved the use of a manuscript-based format, which is considered fitting when the dissertation research has produced one or more manuscripts suitable for journal publication. Based on guidelines from the Faculty of Graduate Studies (2009), this dissertation begins with an overarching introduction to the research and outline of the research objectives. Each research objective is linked to the subsequent findings chapters. Chapters 2 through 4 are presented as separate studies, and thus, each includes an introduction, literature review, methods section, findings section, discussion, and conclusion. There is some repetition between these chapters because each is concerned with women’s decision making about

RR strategies in the context of HBOC. These chapters consist of the content of the manuscripts, which have been published, are in-press, have been submitted for publication, or are nearly ready to submit for publication in a refereed academic journal. Other than minor formatting changes, they are not significantly altered from the published or intended-to-be published versions. Linkages are made between the three studies in the concluding chapter.

### **Research Introduction**

The lifetime risk of developing breast cancer in the general population of Canadian women is 11%, and 1.4 % for ovarian cancer (Canadian Cancer Society, 2009a). Some women are considered to be at higher risk than the general population for developing HBOC based on the history and patterns of cancers in their family, as well as genetic mutation test results. Hereditary cancers are inherited, or passed down in a family through the germ line, from a parent to a child, and account for 5% to 10% of all breast and ovarian cancer cases (Claus, Schildkraut, Thompson, & Risch 1996; Garber & Offit, 2005). The familial nature of breast and ovarian cancer has long been observed and recorded through epidemiological and clinical data (Haites & Gregory, 2002). More recently, the genetic basis of these cancers became clearer with the discovery of cancer susceptibility genes, which include *BRCA1* and *BRCA2*.

It has been anticipated that genetic information and technology will inform novel ways of predicting, identifying, diagnosing, and treating numerous diseases and conditions by means previously unimagined (Sharpe & Carter, 2006). The cloning of the *BRCA1* cancer susceptibility gene in 1994 (Miki et al., 1994) and *BRCA2* in 1995 (Wooster et al., 1995) provided the platform for the development of *BRCA1/2* mutation testing. *BRCA1/2* mutation testing offers the opportunity to learn, by DNA mutation analysis, whether an individual has a predisposition for HBOC. In Canada, eligibility for *BRCA1/2* mutation testing is based upon a woman's family and personal history of breast and ovarian cancer. For women who have these genetic mutations, their lifetime risk of developing cancer is markedly increased; up to 79.5% for breast cancer and 65% for ovarian cancer among *BRCA1* carriers, and up to 88% for breast cancer and 37% for ovarian cancer among *BRCA2* carriers (Antoniou et al., 2003; Evans et al., 2008; Ford et al., 1998). Women with a significant family history of breast cancer (i.e., two or more breast cancers under

the age of 50 years, or three or more breast cancers at any age), but who test negative for *BRCA* mutations have approximately a four-fold risk of breast cancer (Metcalf et al., 2009). As well, first-degree relatives of *BRCA1/2* mutation carriers are at 50% risk for also carrying these mutations. The majority of women who undergo genetic testing, however, receive inconclusive test results and continue to be considered at high risk for HBOC based on their family history.

Women who are at high risk for HBOC are encouraged to adopt cancer prevention and early detection strategies, including cancer screening and maintaining a healthy lifestyle. Other strategies women might choose to reduce their risk include surgery, pharmacoprevention, consulting complementary and alternative medicine (CAM) practitioners or using natural health products. However, the degree to which high-risk women engage in these RR strategies prior to receiving their *BRCA1/2* mutation test results is unknown. Women are presented with the option of risk-reducing mastectomy (RRM) when they are notified that they have a *BRCA1/2* mutation, and risk-reducing oophorectomy (RRO) is typically recommended once childbearing is complete. Yet, there is growing evidence that a sub-group of women do not make decisions about RRM or RRO immediately after receiving their genetic test results, but rather, prolong these decisions for months or years (Antill et al., 2006; Bradbury et al., 2008; Evans et al., 2009; Friebel et al., 2007). Women's experiences with RRM and RRO have begun to be described (Josephson, Wickman, & Sandelin, 2000; Lloyd et al., 2000; Meiser et al., 2000; Metcalfe, Esplen, Goel, & Narod, 2004; Metcalfe, Semple, & Narod, 2004), but their perspectives on the timing of these decisions are not well understood. Moreover, few studies have assessed high-risk women's decision-making processes related to risk management, despite recognition that these decisions are complex and hold significant consequences (McCullum, Bottorff, Kelly, Kieffer, & Balneaves, 2007). Social context factors, such as family experiences of cancer (Antill et al., 2006; d'Agincourt-Canning, 2005; Hallowell, Jacobs, Richards, Mackay, & Gore, 2001; Mellon et al., 2009), the psychological burden of being at high risk for HBOC (Coyne, Kruus, Racioppo, Calzone, & Armstrong, 2003; Fry, Rush, Busby-Earle, & Cull, 2001; Hallowell, 2000; Hurley, Miller, Costalas, Gillespie, & Daly, 2001; Kash et al., 2000; Meiser et al., 1999; Miller, Fang, Manne, Engstrom, & Daly, 1999; Stefanek, Enger, Benkendorf, Flamm Honig, &

Lerman, 1999; Trask et al., 2001; Wellisch & Lindberg, 2001), and the unique risks associated with RR surgery on self-identity, body image, and femininity (Claes et al., 2005; Lodder et al., 2002; Matloff, Barnett, & Bober, 2009) may also dramatically influence a woman's ability to make decisions.

The overall aim of this research was to investigate women's decision making related to cancer RR strategies in the context of HBOC. This research consisted of three objectives: a) to describe the HBOC RR strategies that women are engaged in prior to receiving their genetic testing results, the influence of selected factors (individual and psychological) on the use of these strategies, and their information needs related to risk management, b) to describe how women who carry *BRCA1/2* mutations construct the 'right time' to consider decisions about RR surgeries, and c) to develop an in-depth description and theory about how women make decisions regarding HBOC RR strategies.

## **Background**

### ***Risk Reduction through Cancer Prevention and Early Detection***

Canada is currently facing a "cancer crisis," with cancer being the leading cause of premature deaths in the country and incidence continuing to rise (Canadian Strategy for Cancer Control, 2006). The number of new cancer cases is expected to approach 171,000 in 2009, which represents approximately 470 Canadians diagnosed each day with some form of cancer (Canadian Cancer Society, 2009a). Accordingly, the financial burden of cancer is expected to grow, with estimates that over the next 30 years cancer will cost the health care system \$177.5 billion in direct health care costs, and will cost the Canadian economy \$543 billion in lost wages (Canadian Strategy for Cancer Control, 2006). Cancer prevention and control has become a priority focus for government, researchers, and health care professionals, with the Canadian federal government collaborating with the provinces, territories and non-governmental cancer organizations to develop the Canadian Strategy for Cancer Control (2006). Cancer prevention and early detection through screening are the most effective ways to decrease cancer incidence, morbidity, mortality, and health care costs (Canadian Strategy for Cancer Control, 2006). Prevention is the reduction of cancer mortality via decreasing the incidence of cancer, accomplished by avoiding carcinogens; pursuing lifestyle or dietary practices that modify cancer-causing factors or genetic predispositions;

undertaking RR surgery to remove tissue prior to cancer development, and/or pharmacoprevention to successfully reverse pre-neoplastic changes (National Cancer Institute, 2007). The World Health Organization predicts that up to one third of the cancer burden could be reduced by implementing cancer prevention strategies (World Health Organization, 2006), and the Canadian Cancer Society (2007) estimates that at least half of all cancers could be prevented through healthy living and policies that protect the health of Canadians. Early detection through cancer screening is intended to detect cancer, precancerous conditions, or susceptibility to cancer in asymptomatic individuals, with the goal of detecting cancer early before signs or symptoms appear, thereby improving the likelihood of successful treatment and reducing cancer mortality rates (Canadian Cancer Society, 2007; Canadian Strategy for Cancer Control: Screening Working Group, 2002).

In order to maximize benefits and limit potential harms, targeted cancer prevention and early detection (i.e., to specific age groups, individuals at high risk for particular cancers, and those exposed to carcinogenic conditions) are strongly recommended. As a whole, cancer prevention and early detection strategies can be referred to as RR strategies. Individuals who are at high risk for cancer are often faced with making complex decisions regarding the management of their risk because there are numerous cancer RR strategies, associated with both costs and benefits. Providing support to individuals faced with these difficult decisions is needed to enable efforts to reduce their risk of developing and dying from cancer, while simultaneously limiting potential psychological sequelae. Moreover, optimal decision support has been shown to increase individual's satisfaction with their choices and decrease decisional regret (O'Connor et al., 2003). In order to provide this support, an understanding of the factors that influence engagement in appropriate RR strategies is imperative. Of further importance is knowledge of *when* and *how* individuals make these decisions. This knowledge is also essential for developing and evaluating decision support interventions.

### ***Breast Cancer Prevention and Early Detection***

RR strategies for breast cancer have been a key focus in health care for the past thirty years. Recommended breast cancer prevention includes a wide array of modifiable lifestyle factors, such as

limiting alcohol consumption, maintaining a healthy weight, staying physically active, limiting fat intake, having a diet with a significant proportion of fruits and vegetables, and avoiding tobacco use (British Columbia Cancer Agency, 2009; Clarke, Purdie, & Glaser, 2006; Collishow et al., 2009; Gonzalez, 2006; Mertens, Sweeney, Shahar, Rosamond, & Folsom, 2006; Terry et al. 2006). Evidence suggests that early detection through breast cancer screening (including both mammography and clinical breast examination) could reduce breast cancer mortality by as much as 25% (International Agency for Research in Cancer, 2002), and mammography screening alone leads to a 15% relative reduction in breast cancer mortality (Gotzsche & Nielsen, 2006). Despite large-scale health promotion campaigns and widespread knowledge of breast cancer RR strategies, the uptake of screening among the general population has not reached targeted levels. This is illustrated by the fact that none of the provincially organized breast cancer programs achieved the Canadian target of 70% participation among women 50 to 69 years old, with only 35% of eligible women participating in an organized screening programs in 2002 (Canadian Cancer Society, 2007).

### ***Ovarian Cancer Prevention and Early Detection***

In contrast to breast cancer, there are no corresponding health behaviours suitable for the general population that prevent ovarian cancer. In general, women are advised to consider the factors that have been shown to be protective as well as modifiable risk factors. Ovarian cancer occurs less frequently among women who have used oral contraception for more than five years or who had a tubal ligation, both ovaries removed, or a hysterectomy. Ovarian cancer is also less common among women who have given birth and breast-fed (Center for Disease Control and Prevention, 2009; National Cancer Institute, 2009). Modifiable factors associated with an increased risk of ovarian cancer include the use of hormone replacement therapy among postmenopausal women, the use of fertility medications, frequent application of talcum powder to the perineum, and a diet high in fat and lactose (National Cancer Institute, 2009). Ovarian cancer has become known as “the silent killer” due to difficulties in early detection and the poor survival rate. Currently, there are no effective methods of screening for ovarian cancer among the general population. As such, the focus of public health agencies in Canada and the United States has been on

promoting awareness of the signs and symptoms of ovarian cancer, and advising women to consult health care professionals as soon as these signs and symptoms occur.

### ***HBOC RR Strategies***

HBOC high-risk status is based on a woman's personal cancer history as well as the pattern and prevalence of cancer in her family. Familial breast cancer is characterized by: a younger age at diagnosis than sporadic forms, a number of affected family members, an increased risk of bilateral breast cancer, and a strong association with ovarian cancer (Haites & Gregory, 2002). Familial ovarian cancer is characterized by: two first-degree relatives with a history of ovarian cancer, or the woman herself having ovarian cancer and one or more first- or second-degree relatives (Lynch & Lynch, 1992). Some women may also evaluate their high-risk status via *BRCA1/2* mutation testing, wherein a positive test result indicates they carry a genetic mutation, a negative result indicates they do not carry a mutation, and an inconclusive/uninformative result indicates that the genetic test could not rule out the possibility that they may carry a genetic mutation for HBOC.

There are a variety of RR strategies available to women at high risk for HBOC. These options can be grouped into a) screening to enhance early detection, b) pharmacoprevention, c) RR surgery, d) lifestyle changes, and e) complementary and alternative therapy use. Clinical practice guidelines regarding RR strategies for women who carry a *BRCA1/2* mutation were developed and recently published by the Canadian National Hereditary Cancer Task Force (Horsman et al., 2007). These guidelines are in use within Canadian genetic service centres to varying degrees and are described in the section below.

### ***Screening***

Strategies aimed at detecting cancer early among women known to carry a *BRCA1/2* mutation include breast and ovarian cancer screening. Breast cancer screening methods include magnetic resonance imaging (MRI), breast ultrasound, mammography, clinical breast examination (CBE), and breast self-examination (BSE). For women between the ages of 30 and 69, CBE is recommended biannually, while MRI, mammography, and breast ultrasound are recommended annually (Horsman et al., 2007).

Mammography is not recommended for women less than 30 years of age due to poor test sensitivity, and controversy exists in women ages 40 to 49 years because mortality reduction has not been demonstrated (Warner, et al., 2004). BSE is not recommended for women who carry a *BRCA1/2* mutation because, in the general population, it has not been shown to decrease mortality and it may lead to unnecessary surgical interventions (Howard & Scott-Findlay, 2006; Semiglazov et al., 1992; Thomas et al., 2002). In a Canadian study, the combined use of CBE, mammography, MRI and breast ultrasound resulted in 95% sensitivity for detecting breast cancer among *BRCA1/2* mutation carriers (Warner et al., 2004). Although breast cancer detection is high with screening, it is not a form of cancer prevention because women still develop cancer and require treatment. As well, the interpretation of ambiguous screening tests may lead to excessive follow-up, screening and testing, accompanied by significant anxiety and distress. Decisions to engage in hereditary breast cancer screening are complicated by the time intensive nature of screening and need for some women to travel long distances to access such screening (Phillips et al., 2006).

Screening methods for ovarian cancer include pelvic examination, transvaginal ultrasound, and serum CA-125. However, these screening methods are currently not recommended in Canada due to their poor sensitivity and specificity (Horsman et al., 2007). These screening methods also have high rates of false negative results that may lead to inappropriate reassurance, as well as false positive results that may lead to anxiety and unnecessary investigations and surgeries (Hensley et al., 2003). As well, large-scale studies evaluating the ability of these tests to identify early-stage tumors and reduce morbidity and mortality have reported mixed results (Fishman et al., 2005; Hermesen et al., 2007; Olivier, Lubsen-Brandsma, Verhoef, & van Beurden, 2006; Sato, Yokoyama, Sakamoto, Futagami, & Saito, 2000; van Nagell et al., 2000; van Nagell et al., 2007). Because of the sparseness of conclusive data on the effectiveness of ovarian cancer screening among high-risk women, additional studies are ongoing, the results of which may lead to future changes in recommendations. In the absence of better methods of ovarian cancer screening, these have been recommended in some genetic service centres in other countries, such as the United States (National Comprehensive Cancer Network, 2009) and Australia (National Breast Cancer Centre, 2006). Health care

professionals who interpret the evidence differently and consider ovarian cancer screening to be beneficial may also offer these tests.

### ***RR surgery***

Bilateral risk-reducing mastectomy (RRM) is the surgical removal of healthy breast tissue prior to the development of cancer, with reconstructive surgery being a possibility. RRM is not recommended to Canadian women who carry *BRCA1/2* mutations, but rather, it is framed as a woman's personal decision to be discussed with her health care professional (Horsman et al., 2007). Bilateral risk-reducing salpingo-oophorectomy (RRO) includes the surgical removal of the fallopian tubes, ovaries, and sometimes the uterus, and is recommended to *BRCA1/2* mutation carriers once childbearing is complete, usually between 40 to 45 years of age (Horsman et al., 2007). Because ovarian cancer screening is ineffective, RRO is viewed as the most effective way to reduce cancer risk among *BRCA1/2* mutation carriers (Horsman et al., 2007). The reduction in cancer risk following RR surgery is considerable; with a 90% decrease in breast cancer following bilateral RRM, and a 71% to 96% decrease in ovarian cancer and 50% decrease in breast cancer following RRO (Domchek et al., 2006; Finch et al., 2006; Kauff et al., 2008; Kauff et al., 2002; Kramer et al., 2005; Rebbeck, Kauff, & Domchek, 2009; Rebbeck et al., 2004; Rebbeck et al., 2002; van Sprundel et al., 2005). The reduction in perceived cancer risk, anxiety, and distress following RR surgery has also been reported to be of benefit to some women (Hallowell 2000; Meiser et al., 2000). Unfortunately, RR surgery is also accompanied by significant side effects. The irreversible physical effects of both RRM and RRO and the subsequent impact on women's psychological well-being, including self-identity, self-esteem, and body image, as well as intimate relationships, are significant decision-making considerations (Claes et al., 2005; Lodder et al., 2002; Matloff et al., 2009). RRO induces surgical menopause, contributing to effects such as loss of libido, hot flashes, urogenital atrophy, osteoporosis, and heart disease (Berchuck, Schildkraut, Marks, & Futreal, 1999). Women who undergo RRO are advised to take hormone replacement therapy, yet there are additional physical and emotional side effects of these medications (Brain, Gravell, France, Fiander, & Gray, 2004; Claes et al., 2005). As

well, women who undergo surgery will require time to recover, which may affect their ability to provide practical and economic support to their families (Brain et al., 2004; Hallowell et al., 2001).

### ***Pharmacoprevention***

Pharmacoprevention includes the use or discontinuation of medications to prevent cancer. Although pharmacoprevention may hold promise as a RR strategy, conclusive evidence is absent and no recommendations have been made by the Canadian National Hereditary Cancer Task Force (Horsman et al., 2007). Rather, it is recommended that the risks and benefits be evaluated on an individual basis. Tamoxifen and raloxifene prevent estrogen receptor positive cancers, and although 90% of *BRCA1* cancers are estrogen receptor negative, 60% to 80% of *BRCA2* cancers are estrogen receptor positive, suggesting promise as a future pharmacopreventive agent (Narod et al., 2000). Yet, there are also significant cardiovascular and vasomotor side effects of these medications. Controversy also exists about whether hormone replacement therapy increases a *BRCA1/2* carrier's risk of breast cancer. In the general population, there is some evidence that hormone replacement therapy increases women's risks of breast cancer (Beral et al., 2003). However, in at least one study, there was no increase in breast cancer risk among *BRCA1* mutation carriers taking hormone replacement therapy (Rebbeck et al., 2004). Further research is required on the effects of hormone replacement therapy before conclusions can be drawn. Among *BRCA1/2* mutation carriers, oral contraceptives can significantly decrease ovarian cancer risk. For example, Narod et al., (1998) found a 60% reduction in risk with six or more years of use. Yet, initial evidence also suggests that oral contraceptives increase breast cancer risk in women at high risk for HBOC (Grabrick et al., 2000; Pasanisi et al., 2009). The weighing of these speculative risks and benefits may contribute to women's uncertainty, profoundly affecting their abilities to make RR decisions with confidence.

### ***Lifestyle modification***

Due to the contradictory, yet abundant findings of research exploring environmental and lifestyle factors as potential risk factors for breast cancer among the general population, few lifestyle modifications are formally recommended for women at high risk for HBOC (personal communication, M. McCullum, April 25, 2006; Chappuis & Foulkes, 2002). Evidence concerning lifestyle modification for those with a significant family history of cancer and for *BRCA1/2* mutation carriers is just beginning to emerge. Thus far, strategies that may delay the development or reduce the incidence of *BRCA1/2* related breast cancer include caloric restriction, adherence to Canada's food guide to healthy eating, and increased vegetable and fruit diversity (Ghadirian et al., 2009; Nkondjock & Ghadirian, 2007; Nkondjock, Robidoux, Paredes, Narod, & Ghadirian, 2006). Initial evidence related to body weight suggests that weight loss early in adult life (age 18 to 30 years) protects against early onset *BRCA1/2* associated breast cancer, and that weight gain should also be avoided, particularly among *BRCA1* mutation carriers who have at least two pregnancies (Kotsopoulos et al., 2005). Although there is mixed evidence of the influence of smoking on the risk of breast cancer among *BRCA1/2* carriers (Brunet et al., 1998; Breast Cancer Family Registry et al., 2008; Colilla et al., 2006; Ghadirian et al., 2004; Ginsburg et al., 2009; Gronwald et al., 2006; Nkondjock et al., 2006), an expert panel who recently reviewed this evidence suggested that overall smoking increases risk among those with a family history of breast cancer, as well as *BRCA1/2* carriers (Collishaw et al., 2009). Where clear-cut evidence is absent, Dorval et al., (2008) argue that it is plausible to extrapolate that the modifiable risk factors of breast cancer among high-risk women are the same as those identified in the general population.

There is a lack of evidence about the effects of lifestyle on ovarian cancer risk, particularly among *BRCA1/2* mutation carriers. Accordingly, no specific lifestyle recommendations are made to mutation carriers as a means of preventing hereditary ovarian cancer. However, women may be advised to avoid risk factors for cancer in general by not smoking and avoiding tobacco smoke, eating a healthy diet, doing

physical activity every day, maintaining a healthy weight, and limiting alcohol use (Canadian Cancer Society, 2009b).

### ***Complementary and alternative medicine***

Complementary and alternative medicine (CAM) therapies include those that are not traditionally provided as part of allopathic treatment and may include biological, physical, psychological, and spiritual therapies (National Center for Complementary and Alternative Medicine, 2007). CAM practitioners may provide such therapies as acupuncture, massage, reflexology, reiki, therapeutic touch, naturopathy, homeopathy, and traditional Chinese medicine. CAM therapies also include natural health products, such as vitamins, herbs, and dietary supplements. Research indicates that a large percentage of individuals diagnosed with cancer use CAM therapies, believing this will promote their recovery from cancer and prevent a recurrence of disease (Balneaves, Bottorff, Hislop, & Herbert, 2006; Boon, Olatunde, & Zick, 2007; Eng et al., 2003). Women who are at high risk for HBOC may also perceive CAM therapies to be useful in preventing a future cancer occurrence. DiGianni and colleagues (2003) found that 53% of women at high risk for HBOC enrolled in a genetic testing program (N = 236) used some form of CAM therapy, with the majority of high-risk women in another study frequently adopting CAM therapy use in the first year following testing as part of their cancer prevention regimen (DiGianni, Rue, Emmons, & Garber, 2006). In the future, women at risk of HBOC may be encouraged to use various CAM therapies. For example, scientists have recently turned their attention to nutrigenomics, the study of the effect of nutrients on gene expression, and some have recommended that women at high risk for HBOC consider taking dietary supplements, including lycopene, selenium, diindolylmethane, and green tea polyphenols (Kotsopoulos & Narod, 2005).

Despite the availability of the above RR strategies, decisions about these strategies are complicated by the lack of conclusive scientific evidence and the significant risks and benefits associated with each. It is unclear when women consider these complex decisions or the processes through which they make these

decisions. The theory and research most relevant to decision making in the context of HBOC is reviewed in the following section to begin to understand what women's experiences might be like.

### ***Health Related Decision Making***

Theories of decision making and health behaviour, descriptive decision-making research, and frameworks developed to guide health-related decision making offer important insights into the possible processes by which individuals make health-related decisions. However, limitations and criticisms of these theories and frameworks lead one to question how relevant they are in the context of HBOC risk management, and how adequately they explain the process of making decisions about HBOC risk-reducing strategies.

The classical model of rational choice has provided the foundation for traditional theories of decision making. From this perspective, the rational person chooses what options to pursue by assessing the probability of each possible outcome, discerning the utility to be derived from each, and combining these two assessments to arrive at the option with optimal probability and utility (Gilovich, Griffen, & Kahneman, 2002). Recognizing that full rational choice was unrealistic because of limitations in information processing, most traditional theorists adopted a bounded view of rationality, wherein people reason in a rational fashion, but only within the constraints imposed by their limited search and computational capacities (Simon, 1957). One of the most influential research programs investigating bounded rationality and decision making has been Tversky and Kahneman's work on judgment under uncertainty (Kahneman & Tversky, 1982; Tversky & Kahneman, 1974). Their central tenet was that judgement under uncertainty often rests on a limited number of logical short cuts, labelled heuristics, rather than extensive algorithmic processing (Gilovich, Griffen, & Kahneman, 2002). Heuristics help in decision making when a complete and exhaustive consideration of all possibilities would be too slow or inefficient (Gigerenzer, Todd, & the ABC Research Group 1999). Heuristic reasoning analysis has been used to examine patient delay in evaluating breast cancer symptoms (Facione & Facione, 2006), and women's perceived risks of developing breast cancer (Facione, 2002; Katapodi, Facione, Humphreys, & Dodd, 2005), and provides an important framework for investigating health-related decision making.

However, it fails to explain the broader process of decision making that may include additional cognitive and emotional approaches and resources. A heuristic reasoning framework also fails to incorporate changing personal and social contexts that shape how individuals make decisions, thus limiting its relevance when decision making is prolonged or evolves over time.

Also of considerable influence has been Janis and Mann's (1977) conflict-theory model of decision making. According to this theory, decision making is a stressful event, surrounded by uncertainty, ambiguity, and insufficient resources, wherein individuals cope with stress by searching for, and evaluating information and alternatives (Balneaves & Long, 1999; Janis & Mann, 1977). Despite the widespread use of this theory to explain health-related decisions, limitations have been noted. Lazarus and Folkman (1984) contend that although Janis and Mann explain the consequences of making decisions with not enough time, they do not discuss what happens when a deadline to make a decision is distant and there is an insufficient level of arousal for a vigilant search and evaluation of information and advice. This criticism is relevant when considering the applicability of this theory to decisions that may involve longer time frames or the prevention of a possible future disease, such as HBOC. Balneaves and Long (1999) also contended that the conflict-model of decision making fails to fully elucidate the personal, transactional, and relational nature of choice behaviour. As such, they combined this model with Lazarus and Folkman's (1984) transactional framework of stress and coping to propose the Embedded Decisional Model of Stress and Coping. Despite the appeal of this hybrid model, it was designed with breast cancer treatment decision making in mind and the applicability of this model to the HBOC context, wherein women do not have a diagnosis of breast cancer and are making decisions about risk reduction, is questionable.

In contrast to the traditional, acontextual decision-making theories, naturalistic decision-making theories emphasize how decisions are complex, dynamic, and made in the "real world," as well as being personally and contextually situated (Zsombok & Klein, 1997). However, these theories typically focus on the decision making of health care professionals, rather than patients. Moreover, naturalistic decision-making theories concentrate on the ability of individuals to make clinical decisions where there is clearly

a right and a wrong decision. This normative approach does not apply to situations where there is no right answer, as is the case for HBOC risk management decisions.

Cognitive-based theories of health behaviour also provide a useful starting point to understand the possible processes by which individuals make decisions. Theories that explain and predict health behaviour can be classified into intrapersonal theories (i.e., the health belief model, the theory of reasoned action, the theory of planned behaviour, the transtheoretical model, the precaution adoption process model) and interpersonal theories (i.e., social cognitive theory, social networks and social support theory, theory of stress, coping and health behaviour, and interdependence theory) (Glanz, Rimer, & Viswanath, 2008). Intrapersonal theories focus on the behaviours of individuals incorporating cognitive and emotive aspects (Rimer, 2002). In contrast, the core assumption underlying interpersonal theories is that people's interactions with their environments and social networks are critical determinants of their health behaviour and, in turn, their health outcomes (Marcus Lewis, 2002). These intrapersonal and interpersonal health behaviour theories have been used extensively in the breast cancer screening and treatment decision-making literature. Yet, it has been noted by several authors that health behaviour theories do not provide an adequate description of the ways in which individuals make decisions (Conner & Norman, 2005; Myers, 2005). They argue that individuals do not base all of their decisions on a complex, subjective, cost/benefit analysis of the likely outcomes of different courses of action, which is the underlying assumption of the majority of health behaviour theories (Conner & Norman, 2005).

Research investigating patients' decision making about cancer prevention (Bowen et al., 2006; Gorin, Wang, Raich, Bowen, & Hay, 2006; Myers 2005; Wackerbarth, Peters & Haist, 2008), breast cancer treatment (Balneaves & Long, 1999; Halkett, Arbon, Scutter, & Borg, 2005; O'Brien et al., 2008; Pierce, 1993), and genetic testing (Burgess & d'Agincourt-Canning, 2001; d'Agincourt-Canning & Baird, 2006; Sherwin & Simpson, 1999) has provided some insights into the complexity of these decisions and the importance of social influences on autonomous decision making. Yet, the generalizability of findings from this research to decisions about HBOC risk-reducing strategies may be inappropriate considering the distinct characteristics and challenges inherent to these contexts, as well as the HBOC context. The

limited research about decision making regarding HBOC has largely been descriptive, with only a few studies theoretically based on such theories as the Common Sense Model of Self-Regulation (van Oostrom et al., 2007), the Protection Motivation Theory (Helmes, 2002), the Expected-Utility Theory (Unic, Verhoef, Stalmeier, & van Daal, 2000), or the Self-Regulation Model of illness representations (Decruyenaer, Evers-Kiebooms, Welkenhuysen, Denayer, & Claes, 2000; Kash, Holland, Osborne, & Miller, 1995). Although the results of these studies provide a beginning understanding of decision making among women with HBOC, the findings point to limitations in existing theories in explaining the details of RR decision-making processes and the need for theory development to adequately explain women's decision-making experiences and provide direction for decision support.

The provision of decision support is not a primary focus of decision-making theories or of theories that explain and predict health behavior. To address this gap, the Ottawa Decision Support Framework (ODSF) was designed to guide the development of interventions aimed at preparing individuals and physicians for a structured process of shared decision making (Légaré, O'Connor, Graham, Saucier, Cote, Cauchon et al., 2006). In this framework, decisional conflict is identified as a key element in decision making, which has been conceptualized as a state of uncertainty about which course of action to take when the choice among competing actions involves risk, loss, regret, or a challenge to personal values (Légaré, O'Connor, Graham, Saucier, Cote, Cauchon et al., 2006). The goal of interventions based on the ODSF is to improve the quality of decision making by addressing the modifiable determinant of decisions in order to improve patient outcomes (Légaré, O'Connor, Graham, Saucier, Cote, Blais et al., 2006). The ODSF has been used to guide the development and evaluation of more than 30 patient decision aids, practitioner decision support resources, and tools to evaluate the quality and outcomes of providing decision support (Ottawa Health Research Institute, 2007). Both Metclafe et al. (2007) and Tiller et al. (2006) developed RRM decision aids theoretically guided by the ODSF, which resulted in increased knowledge levels and decreased decision conflict, yet neither improved psychological outcomes. The ODSF shows great promise for use in developing and implementing additional decision-making interventions for women considering HBOC RR strategies. However, understanding the unique decision-

making processes specific to this context is vital prior to using the ODSF to develop these additional interventions. This knowledge is also critical to improving upon existing decision-support interventions.

### ***Decision Making in the Context of HBOC***

A systematic review of the literature describing factors that influence decision making related to cancer RR strategies among women at high risk for HBOC has been published and is reprinted in Appendix A (Howard, Balneaves, & Bottorff, 2009).<sup>1</sup> The three main types of factors identified were: a) medical and physical factors, b) psychological factors, and c) social context factors. Medical and physical factors included *BRCA1/2* mutation status, a previous cancer diagnosis, age, parity, and menopausal status. Psychological factors included perceived cancer risk and cancer-related distress, anxiety and worry, while social context factors included patterns and personal experiences of cancer in the family, as well as obligations women had to their family. The research reviewed, in addition to literature describing the experiences of women at high risk for HBOC, provides evidence that existing theories, frameworks, and research may be inappropriate in the unique context of HBOC.

First, the social and family context surrounding hereditary cancer is significant. Women's knowledge of familial breast/ovarian cancer is shaped by family patterns of inheritance, personal observations and the experiences of caring for others (d'Agincourt-Canning, 2005). For example, women in one study who opted for RR surgery considered the death of a relative to be one of the most influential factors, and the importance of witnessing a relative's experience of ovarian cancer may account for the fact that 87% of the women who had RRO had first degree relatives who had died from ovarian cancer, compared with only 41% of women who chose screening (Hallowell et al., 2001).

Second, women's perceived risk of developing HBOC may be long standing and heightened as a result of family experiences with cancer. It is unclear when women begin to conceptualize themselves as high risk and contemplate decisions about RR behaviours. A number of studies provide evidence that the decision to engage in screening and proceed with RR surgery (Claes et al., 2005; Fry et al., 2001;

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<sup>1</sup> A version of Appendix A has been published: Howard, A. F., Balneaves, L. G., & Bottorff, J. L. (2009). Women's decision making about risk-reducing strategies in the context of hereditary breast and ovarian cancer: A systematic review. *Journal of Genetic Counseling*, 18 (6), 578.

Hallowell, 2000), as well as the intention to undergo future RR surgery is associated with higher levels of women's perceived risk (Antill et al., 2006; Fang et al., 2003; Hurley et al., 2001; Meiser et al., 2000; Meiser et al., 1999; Stefanek et al., 1999; Tiller et al., 2005; Unic et al., 2000).

Third, the psychological effects of being at high risk, such as anxiety and distress, may dramatically influence women's abilities to make decisions, but also the nature of the decision-making process. For example, research has demonstrated that a sub-group of women undergoing genetic testing experience distress above sub-clinical levels (Coyne et al., 2003; Mikkelsen, Sunde, Johansen & Johnsen, 2008; Mikkelsen, Sunde, Johansen & Johnsen, 2009). Some women have been reported perceiving their breasts and ovaries as "time bombs" and RR surgery represented a strategy for managing distress and anxiety about developing cancer (Brain et al., 2004).

Fourth, the varying levels of evidence about the effectiveness of certain RR strategies to prevent or detect cancer are problematic for women attempting to make decisions in the context of HBOC. While evidence exists in support of such strategies as RR surgery, there is inconclusive evidence of the effectiveness of other options, such as lifestyle behaviour changes. Making decisions with multiple risks and benefits associated with the various RR options available is undoubtedly challenging. As well, the risks and benefits take on various meanings depending on women's personal values and beliefs. Evidence suggests that the best choice for "preference-sensitive" treatment decisions depends on how patients value benefits versus harms (O'Connor et al., 2007).

Fifth, the combination of the gendered and HBOC specific context may be significant and unique. Women's roles and responsibilities in their families have been found to influence both women's uptake and rejection of screening and RR surgery (Brain et al., 2004; Hallowell et al., 2001). Women's breasts and reproductive organs are imbued with numerous social and individual meanings that possibly contribute, as mentioned previously, to concerns about the effects of RRM and RRO on self-identity, body image, and femininity (Claes et al., 2005; Fang et al., 2003; Lodder et al., 2002). Because RRO renders women infertile, whether women want to have children in the future or have completed child bearing also influences their surgery choices (Brain et al., 2004; Claes et al., 2005; Fang et al., 2003;

Hallowell et al., 2001; Lodder et al., 2002; Schmeler et al., 2006). Researchers have further found that surgically induced menopause with subsequent hormone replacement therapy is an important consideration and, at times, the decisive motive for decisions about RRO (Brain et al., 2004; Claes et al., 2005; Fry et al., 2001; Hallowell, 2001; Hallowell 2000).

Lastly, the availability of and access to genetic services, as well as the costs and lost income associated with these services and RR strategies, may be significant barriers to women's abilities to even engage in decision making. Relatively new, genetic services in Canada are offered through specialty hereditary cancer programs situated in a limited number of urban centres, making travel (often associated with significant costs) and taking time off work and away from family necessary for those women who do not reside in these centres. For example, annual MRI is increasingly recommended for women at high risk for HBOC, yet is only available through hereditary cancer services in urban centres. Inequities in accessing genetic counseling and RR strategies have arisen due to the concentration of services in these large urban centres. Moreover, due to demands on health care resources, the significant wait to access genetic counseling, at least six months (C. Kim-Sing, personal communication, December 13, 2009), as well as wait-times associated with RR surgery, may further complicate women's decision making.

Researchers have begun to describe *BRCA1/2* carriers' experiences and satisfaction with HBOC RR strategies (Josephson et al., 2000; Kenen, Shapiro, Hantsoo, Friedman, & Coyne, 2007; Lloyd et al., 2000; Metcalfe et al., 2004), yet few studies explain the preceding decision-making processes (McCullum et al., 2007). The limited existing research has largely been descriptive, incorporating such theories as the expected-utility theory (Unic et al., 2000), or the self-regulation model of illness representations (Decruyenaer et al., 2000; Kash et al., 1995). However, these theories do not address complex decision-making situations wherein multiple RR options need to be considered; nor do they take into consideration the unique factors discussed above that influence women who are at risk for HBOC. Because of the complexity of RR decision making in the context of HBOC there has been a desire to develop decision support interventions. A better understanding of women's decision-making processes and the social contexts that influence these processes is needed to guide the development of these interventions.

### ***The Timing of HBOC RR Decision Making***

The factors that influence decision making about HBOC RR strategies discussed above most likely affect the timing of when women consider these decisions as well. There is wide variation in when high-risk women consider HBOC RR strategies, with some beginning to consider risk management decisions prior to receiving their genetic test results. For example, consistently across studies, a small number of women undergo RR surgery without knowing their *BRCA1/2* status (Hallowell, 2000; Phillips et al., 2006; Ray, Loescher, & Brewer, 2005; Schmeler et al., 2006; Stefanek, Helzlsouer, Wilcox, & Houn, 1995). Yet, the degree to which high-risk women engage in screening, RRM, RRO, pharmacoprevention, lifestyle behaviour modification, and CAM use prior to receiving their genetic test results is unknown, as are the influences of individual characteristics, perceived risk, and psychological distress on the use of these RR strategies.

In Canadian genetics programs, women receive genetic counseling before undergoing *BRCA1/2* mutation testing. Some women undergo genetic testing immediately following counseling, yet, there are others who choose to wait, or have to wait (e.g., for an index result to become available in the family so that carrier testing is possible), to have blood drawn for genetic testing. The optimal time to begin providing decision support related to HBOC RR strategies is unknown, with little direction provided by existing theories of decision making, health behaviour, or decision support. The pre-test counseling session presents a teachable moment for discussions about RR strategies to begin because women have accessed the health care system and taken the initiative to investigate their *BRCA1/2* mutation status, demonstrating interest and possibly motivation to engage in health behaviours. This might also be a timely opportunity to begin health promotion discussions because women who receive indeterminate results may be especially in need of advice and support to not only understand their results but to make decisions about risk reduction because of the limited information provided about their risk for HBOC through genetic testing. A better understanding of the RR strategies that high-risk women are using prior to receiving their genetic test results, and the influence of perceived risk, psychological distress, and individual characteristics on the use of these strategies, will provide important insights that could inform

these discussions. As well, knowing women's information needs regarding RR strategies at this time is essential for assisting them to make informed decisions before and after receipt of their genetic test results. This is particularly relevant considering that the time from providing a blood sample to receipt of genetic test results can take a number of months.

There are other women who wait for years after receiving their genetic test results to make HBOC RR decisions, particularly about RRM and RRO (Antill et al., 2006; Bradbury et al., 2008; Evans et al., 2009; Friebel et al., 2007). In the study by Friebel and colleagues (2007), which included 297 women who had RRO and 113 women who had RRM, the average time from genetic testing to RR surgery was 1 year. Yet, some of these women had RRO up to 7 years later and RRM more than 8 years later. For women in another study who previously had a mastectomy to treat breast cancer, the mean time that elapsed between their primary surgery and contralateral RRM was 3.5 years (Metcalf et al., 2008). The amount of time between receipt of genetic test results and RR surgery has raised questions about why women are not acting promptly to reduce their risk, and concerns that the full benefits of genetic testing are not being realized. Research to date has not adequately captured women's perspectives on appropriate times to make decisions or factors that influence timing. A few studies have described women's decision-making processes about RR surgery as complex, dynamic, and prolonged (Hallowell et al., 2001; McCullum et al. 2007). In a case study that involved in-depth interviews with three women over one year, researchers tracked how women's thoughts and decisions about RRM changed over time, and the time they needed to achieve a level of comfort with decisions before they were able to follow through with them (McCullum et al., 2007). A better understanding of women's perspectives on the timing of decisions about RRM and RRO is needed to guide the provision of decision support as well as the development of decision support interventions that address the complex needs faced by women at high risk for HBOC.

### **Theoretical Lenses**

The two theoretical lenses used to guide this research included gender theory and relational autonomy. A gender theory lens was chosen because gender has been found to profoundly influence health behaviours (Johnson, Greaves, & Repta, 2007). As well, HBOC is a disease predominantly

affecting women and their female body parts, towards which RR strategies are aimed. Complementary to a gender lens, a relational autonomy lens (Hartrick Doane & Varcoe, 2005; Sherwin, 1998) was chosen because of its emphasis on the decision maker as contextually situated. This is particularly salient due to the multiple complex relationships within which women's experiences in making HBOC RR decisions are situated.

### ***Decision Making as Gendered***

Gender refers to the social characteristics of men and women, whereby individuals adopt masculine and feminine patterns of behavior according to social expectations and customs. These social characteristics are learned, change across the lifespan, and vary both within and between cultures (Johnson et al., 2007; Stacey, 1996; Vlassoff & Moreno 2002). Engaging in particular behaviours can be seen as a process through which people are shaped, and by which they shape themselves, in accordance with social or cultural norms. In the words of Lyons (2009), "gender is performed in daily life and through daily activities" (p. 394), rather than being an innate personal characteristic or trait (Connell & Messerschmidt, 2005). Gendered roles are the behavioural norms associated with different versions of masculinity and femininity in societies, which influence individuals' everyday actions, expectations, and experiences (Johnson et al., 2007; Lyons, 2009). Gender roles often categorize and control individuals within institutions, such as the family (Johnson et al., 2007). Performing, or enacting, gender according to gendered roles is a way of producing and reproducing gender (Butler, 1990; Connell & Messerschmidt, 2005; Lyons, 2009). Gender identity describes how individuals see themselves as women or men, affecting their emotions and behaviours. Gender identity is developed in the face of strong societal messages about "correct" gendered roles for individuals' presenting sex (Johnson et al., 2007). It is reasonable to assume that decision making occurs in a gendered context because health behaviours are profoundly influenced by the different roles, responsibilities, and activities ascribed to individuals according to their gender (Johnson et al., 2007). Moreover, health behaviour is a social practice through which gender identity and gender roles are continuously (re)constructed (Lyons, 2009; Saltonstall, 1993). Gender has been shown to figure prominently in women's decisions and experiences of cancer prevention

and treatment (Adachi, Ueno, Fujoka, Fujitomi, & Ueo, 2008; Dailey, Kasl, & Jones, 2008; Helms, O’Hea & Corso, 2008), as well as genetic testing decisions (d’Agin-court-Canning & Baird, 2006). Donchin argues that, “genetic information enters into family relations and affects family life along gender-specific paths” (2000, p.250). To add to this, disease is often conflated with gender, such that breast cancer and actions to prevent or detect it early are associated with women’s roles and stereotypical aspects of femininity (Clarke, 2004). Despite the significance of the gendered context, decision making and health behaviour theories have, thus far, failed to account for the influence of gender.

By asking questions about how gender identity shapes perceptions of femaleness in the context of HBOC, the focus is directed to understanding how various RR strategies threaten or enhance a woman’s self-identity, and how this influences decision making. A gender lens guided this research by drawing attention to how women perceived that their actions, expectations, and experiences regarding decision making around HBOC RR strategies were influenced by the diverse roles they fulfill. Concentrating on gendered roles sheds light on women’s desired and experienced roles as information seekers, decision makers, or health care consumers. A gender lens also helped elucidate how women are influenced by gendered roles in their relationships with family, friends, and health care professionals.

### ***Decision Making as Relational***

Existing decision-making models consider an autonomous individual to be an independent and rational decision maker who is inattentive to emotions, communal life, reciprocity, and the development of persons over time (Beauchamp & Childress, 2001). In this sense, both independence from controlling influences (liberty) and capacity for intentional action (agency) are essential for autonomy (Beauchamp & Childress, 2001). Using a feminist lens, Sherwin (1998) proposed an alternative conception of autonomy that she labeled relational, within which she highlighted two points of departure from traditional conceptualizations of autonomy. First, Sherwin drew attention to the development of selfhood as an ongoing process and the importance of social forces in shaping individual identity. She argued that individuals engage in activities that are constitutive of identity and autonomy (e.g., defining, questioning, revising, and pursuing projects) within a configuration of interpersonal and political relationships,

referred to as a “web of interconnected (and sometimes conflicting) relationships” (p.35). Building on Sherwin’s ideas of relational autonomy, Rodney (1997) referred to this web of interconnecting relationships as the relational matrix. The relational matrix forms a part of organizational and social contexts and highlights the significance of interpersonal relationships in shaping these contexts (Varcoe, Rodney, & McCormick, 2003). In their writing about nursing as relational inquiry, Hartrick-Doane and Varcoe (2005) contended that individuals not only are shaped by their relational interactions, but they also shape those experiences and contexts. As well, sociohistorical values, knowledge, practices, attitudes, and structures are passed on and repeated through these relational interactions.

Second, Sherwin’s emphasis on the political, particularly power and powerlessness, illuminated how forces of oppression interfere with an individual’s ability to exercise autonomy by undermining their personal sense of self as an self-directed and capable agent and depriving them of opportunities to exercise autonomy. Sherwin (1998) contended that relational autonomy is a capacity or skill that is developed and constrained by social circumstances, drawing attention to barriers that interfere with individuals’ abilities to enact their autonomy, and recognizing that autonomy is best achieved when there are amenable social conditions. Hartrick-Doane and Varcoe (2005) incorporated additional critical theories of postcolonialism and poststructuralism to understand power relations and the contexts within which choices are made and agency enacted. They drew attention to how people are contextual beings who live in relation with social, cultural, political, and historical processes and communities.

In her research on decision making about HBOC genetic testing, d’Agincourt-Canning (2006) argued that most previous research focused on genetic decision making without exploration of the deeply personal, relational, and embodied process of making decisions that are consistent with one’s morals and ethics. Findings from her research indicated that individuals’ decisions to undergo genetic testing were interconnected with their physical self (embodied self), family health and well-being (familial-relational self), and general relationships to unknown others (civic self) (d’Agincourt-Canning, 2006). Using a relational view of autonomy, Burgess and d’Agincourt-Canning (2001) also suggested that decisions concerning HBOC risk reduction might not be individual autonomous decisions, even though women

may exercise agency, such that women's roles, responsibilities and dynamics operate as barriers and facilitators during the decision-making process. Women's RR options may also be influenced by institutional policies, structures and guidelines, and legal restrictions. Furthermore, genetic service providers, who are situated in health care institutions and follow professional and institutional guidelines, may influence the decisions women make via professional standards of practice that function as barriers and facilitators to women's decisions (Sherwin, 1998). In addition, access to genetic services and RR strategies, as well as the associated financial implications, may be paramount to women's decision making.

Drawing on the work of Sherwin (1998), Rodney (1997), Varcoe et al. (2003), and Hartrick-Doane and Varcoe (2005), a relational view of autonomy was used as a guiding theoretical lens to understand the personal factors, interpersonal relationships and wider social contexts within which women made HBOC RR decisions. Important personal context factors previously mentioned include women's perceived HBOC risk as well as the multitude of psychological factors (i.e., anxiety and distress) associated with being at high risk. Using a relational autonomy lens leads to questions about how women's relationships with family, friends, and health care professionals, as well as the relational matrix within which they were situated, helped to shape their perceptions of the decision-making process. This also included consideration of how organizational, social, and political contexts shaped women's perceptions of the decision-making process.

### **Research Purpose and Objectives**

The overall purpose of this research was to describe women's decision making about RR strategies in the context of HBOC. Guiding this research were three main research objectives, each with accompanying research questions.

#### ***Research Objective 1***

The first objective of this research was to describe the HBOC RR strategies that women used prior to receiving their genetic test results, the relationship between demographic and psychological factors and RR strategies, and whether these factors predicted the uptake of RR strategies. A further objective was to

describe participants' information needs related to RR strategies. Specific research questions were as follows:

- a. What screening behaviours and/or other surgical or pharmacopreventive therapies were women using prior to receiving their genetic test results?
- b. What lifestyle behaviours were women engaged in prior to receiving their genetic test results?
- c. What complementary and alternative medicine therapies were women using prior to receiving their genetic test results?
- d. Were individual factors (age, marital status, employment level, ethnic background, level of education, annual household income, and number of children), and psychological factors (perceived risk and psychological distress) related to the uptake of HBOC RR strategies?
- e. What were the demographic and psychological predictors of women's uptake of HBOC RR strategies?
- f. What were women's information needs related to HBOC RR strategies prior to receiving their genetic test results?

### ***Research Objective 2***

The second objective was to describe how women who carry *BRCA1/2* mutations construct the 'right time' to consider decisions about RR surgeries. Specific research questions were as follows:

- a. How did women construct the 'right time' to consider decisions about RR surgeries?
- b. What factors (medical, physical, psychological, and family context) did women perceive as influencing when decisions about RR surgeries were considered?
- c. How did women perceive their gendered roles in their relationships with family and health care professionals as influencing when decisions about RR surgeries were considered?
- d. How did women perceive organizational, institutional, and structural relationships (i.e., by available and unavailable health care services and access to services) as influencing when decisions about RR surgeries were considered?

- e. How did women's perceptions of the appropriate timing of RRM and RRO decisions change over time?

### ***Research Objective 3***

The third objective of this research was to develop an in-depth description and theory about women's experiences of making decisions around HBOC RR strategies, post disclosure of *BRCA1/2* mutation test results. Specific research questions were as follows:

- a. What was the process of decision making that women engaged in as they made decisions about HBOC RR strategies?
- b. What factors (medical, physical, psychological, and family context) did women perceive as influential throughout the decision-making process about HBOC RR strategies? How did women construct the influence of these factors in shaping these decisions?
- c. How did women perceive their relationships with partners, children, other family members, and friends as shaping the decision-making process about HBOC RR strategies?
- d. How are gendered roles and gendered identities reflected in women's accounts of their decision-making experiences?
- e. How did women construct the interactions and relationships they had with health care providers regarding decisions about HBOC RR strategies? How did these interactions influence their decision making?
- f. How did women perceive that their decision making was influenced by organizational, institutional and structural relationships (i.e., by available and unavailable health care services, access to services, legal restrictions and health professional standards of practice)?

### **Research Design**

In order to address the aforementioned research objectives, three studies were carried out; (1) a descriptive quantitative survey was used to address the first research objective, (2) a qualitative thematic analysis was used to address the second research objective, and (3) a qualitative grounded theory study was used to address the third research objective. These three studies were complementary in that they all

investigated risk management decisions of women at high risk for HBOC, but the quantitative work focuses on the time prior to the receipt of *BRCA1/2* mutation test results, whereas the qualitative research focuses on the time following *BRCA1/2* mutation testing. This dissertation research cannot be classified as mixed methods research because findings from the studies did not influence each other, nor did these studies investigate the same research question (Johnson, Onwuegbuzie, & Turner, 2007; Leech & Onwuegbuzie, 2009). Nevertheless, the findings of these three studies provide important insights into women's RR decision making and will be integrated in the concluding chapter of this dissertation.

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## **CHAPTER 2: Risk Management Strategies used by Women Prior to Receiving *BRCA1/2***

### **Mutation Test Results: Uptake and Information Needs<sup>2</sup>**

#### **Introduction**

Women who present for genetic testing have an elevated risk of hereditary breast and ovarian cancer (HBOC) based on the patterns and prevalence of cancer in their families. These women are encouraged to adopt cancer prevention and early detection strategies including breast cancer screening and maintaining a healthy lifestyle. Other strategies women might choose to reduce their risk of HBOC include surgery, pharmacoprevention, consulting complementary and alternative medicine (CAM) practitioners or using natural health products. However, the degree to which high-risk women engage in these risk-reducing (RR) strategies prior to receiving their genetic test results is unknown. Furthermore, the influence of perceived risk, psychological distress, and individual characteristics on women's use of RR strategies is not understood. Accordingly, the objectives of this research were to: a) describe the HBOC RR strategies that women used prior to receiving their genetic test results, b) examine the relationship between perceived risk, psychological distress, and demographic factors, and RR strategies, and c) determine predictors of the uptake of these RR strategies. A supplementary objective was to describe participants' information needs related to RR strategies. This knowledge will be useful to health care professionals who provide information, recommendations, and counseling about HBOC risk management to women before they receive their genetic test results.

#### ***Background***

A family history of breast or ovarian cancer increases a woman's risk of developing a similar cancer. More specifically, a woman's risk of breast cancer increases progressively with the number of affected first-degree relatives: with one first-degree relative, a woman has a 1.80 relative risk, two first-degree relatives result in a 2.93 relative risk, and with three or more first-degree relatives, the risk increases to

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3.90 times (Collaborative Group on Hormonal Factors in Breast Cancer, 2001). Hereditary cancers are inherited, or passed down in a family from a parent to a child, accounting for an estimated 5% to 10% of breast and ovarian cancers (Claus, Schildkraut, Thompson, & Risch 1996; Garber & Offit, 2005). Women with a family history suggestive of HBOC may be eligible for *BRCA1* and *BRCA2* genetic mutation testing to determine whether they inherited a predisposition. For women who have a mutation in either *BRCA1* or *BRCA2*, their lifetime risk of developing cancer is markedly increased; up to 88% for breast cancer and 65% for ovarian cancer (Antoniou et al., 2003; Evans et al., 2008; Ford et al., 1998). As well, first-degree relatives of *BRCA1/2* carriers are at 50% risk for also carrying these mutations.

Women who are at high risk for HBOC are increasingly encouraged to undergo breast cancer screening. Breast cancer screening by the general population has remained limited, exemplified by national mammography rates as low as 34% (Public Health Agency of Canada, 2005). In contrast, women who carry a *BRCA1/2* mutation have mammography rates as high as 93%, with most beginning screening at some time prior to genetic testing (Metcalf et al., 2008). Other high-risk women might opt for RR mastectomy, RR oophorectomy or pharmacoprevention. Uptake rates for RR surgeries vary widely among studies of *BRCA1/2* mutation carriers, but consistently, a small number of women undergo RR surgery without knowing their *BRCA1/2* status (Hallowell, 2000; Phillips et al., 2006; Ray, Loescher, & Brewer, 2005; Schmeler et al., 2006; Stefanek, Helzlsouer, Wilcox, & Houn, 1995).

Women at high risk for HBOC are also encouraged to maintain a healthy lifestyle. Known modifiable lifestyle risk factors for breast cancer among the general population include physical activity, alcohol consumption, fat intake, and tobacco use (Clarke, Purdie, & Glaser, 2006; Gonzalez, 2006; Mertens, Sweeney, Shahar, Rosamond, & Folsom, 2006; Terry et al. 2006). Although evidence is just beginning to emerge about how such modifiable risk factors interact with breast and ovarian cancer susceptibility, it is frequently assumed that high-risk women will benefit from regular exercise, reduced alcohol consumption, low fat diets, and smoking cessation (Burke et al., 1997; Pichert, Bollinger, Buser, Pagani, & Swiss Institute for Applied Cancer Research Network for Cancer Predisposition Testing and Counseling, 2003). Limited conclusive evidence exists regarding the efficacy of receiving care from

complementary and alternative medicine practitioners, such as acupuncturists or naturopaths, or using natural health products, including vitamins or herbal supplements. Yet, women at high risk for HBOC have reported using CAM (DiGianni, Kim, Emmons, Gelman, Kalkbrenner, & Garber, 2003).

There are indications that perceived HBOC risk, psychological distress, and individual characteristics may influence women's use of these RR strategies. High-risk women often perceive their risk of cancer to be elevated because they witnessed multiple relatives affected with cancer (Evans, Blair, Greenhalgh, Hopwood & Howell, 1994; Mouchawar, Byers, Cutter, Dignan, & Michael, 1999). Heightened perceived risk has been reported by women undergoing genetic testing and HBOC screening (Claes et al., 2005; Fry, Busby-Earle, Rush, & Cull, 2001; Hallowell, 2000), and RR surgery has been associated with higher perceived risk in several studies (Antill et al., 2006; Fang et al., 2003; Hurley, Miller, Costalas, Gillespie, & Daly, 2001; Meiser et al., 1999; Meiser et al., 2000; Stefanek, Enger, Benkendorf, Flamm Honig, & Lerman, 1999; Tiller et al., 2005; Unic, Verhoef, Stalmeier, & van Daal, 2000). This heightened risk perception has also been associated with distress (Brain, Norman, Gray, & Mansel, 1999; Drossaert, Boer, & Seydel, 1996) and research has shown that women experience distress above sub-clinical levels prior to genetic testing (Coyne, Kruus, Racioppo, Calzone, & Armstrong, 2003; Mikkelsen, Sunde, Johansen & Johnsen, 2009; Mikkelsen, Sunde, Johansen & Johnsen, 2008). Moreover, researchers have reported a positive relationship between distress, anxiety, and decisions about RR strategies (Antill et al., 2006; Lodder et al., 2002; Meiser et al., 2000; Unic et al., 2000). Individual and demographic variables, such as age, parity, marital status, and socioeconomic status, have also been associated with the use of RR strategies among high-risk women in some studies, but not others (Howard, Balneaves, & Bottorff, 2009).

In Canadian genetics programs, women receive genetic counseling before undergoing *BRCA1/2* mutation testing. Some women undergo genetic testing immediately following counseling, yet, there are others who choose to wait, or have to wait (e.g., for an index result to become available in the family so that carrier testing is possible), to have blood drawn for genetic testing. The pre-test counseling session presents a timely opportunity for discussions about RR strategies. This may also represent a time to provide targeted decision support to the sub-group of women who consider RR surgery prior to receipt of

their genetic test results. A better understanding of the RR strategies that high-risk women are using prior to receiving their genetic test results, and the influence of perceived risk, psychological distress, and individual characteristics on the use of these strategies, will provide important insights that could inform these discussions. As well, knowing women's information needs regarding RR strategies is essential for assisting them to make informed decisions before and after receipt of their genetic test results. This is particularly relevant considering that the time from providing a blood sample to receipt of genetic test results can be a number of months.

### **Methods**

A descriptive, exploratory quantitative approach was used in this study. This study was conducted as part of an on-going study designed to develop a reliable and valid genetic testing psychological risk screening tool for adult onset hereditary diseases (Espen et al., 2003). A questionnaire designed to assess use of HBOC RR strategies was included with other baseline measures administered to women undergoing genetic testing for HBOC at three Canadian sites.

#### ***Participants and Sampling Strategies***

The sample included women at high risk for HBOC who recently underwent *BRCA1* and *BRCA2* genetic testing at three genetic testing centres in Canada, but had not yet received their results. Women included in this study were: 1) going through genetic testing for breast/ovarian cancer by providing a blood sample to the genetic clinics, 2) fluent in English, 3) older than 18 years, and 4) residing within 1.5 hours driving distance from the genetic testing centre. Women who indicated they had a current diagnosis of breast and/or ovarian cancer were excluded from this study because these women are likely facing treatment decisions as opposed to decisions about HBOC prevention or early detection.

Genetic service providers in each site received a description of the study. Following approval by the research ethics boards of participating genetic testing centres, a research assistant located within each of the genetic testing centres contacted women who had begun the process of genetic testing. The research assistant obtained informed consent and administered a battery of questionnaires, including the HBOC RR strategies questionnaire, shortly following genetic testing and prior to receipt of test results.

### ***Data Collection***

Self-administered questionnaires were used as the baseline measures in the primary study. These included demographic measures, the HBOC RR strategies questionnaire, measures of perceived risk, psychological distress, and information needs.

The *individual demographic data* collected included age, marital status, employment level, ethnic background, level of education, annual household income and whether women had children.

The *HBOC RR strategies questionnaire* (Appendix B) was developed for this research because no pre-existing measures existed. Initial versions of the HBOC risk-reducing strategies questionnaire were revised based on feedback provided by women from the general public with limited knowledge of HBOC, nurse colleagues, and health care professionals with expertise in genetics. Items on the questionnaire were used to assess when, if at all, women had breast cancer screening, including breast-self examination, clinical breast examination, mammography, and breast ultrasound, and ovarian cancer screening, including manual pelvic examination, transvaginal ultrasound, and serum CA125. Three items assessed women's use of hormone replacement therapy, oral contraceptives, and tamoxifen/raloxifene, whereby women indicated whether they initiated or stopped taking these medications to reduce their risk of HBOC. Items measured the uptake of mastectomy and oophorectomy as a means of HBOC risk management. Measures of the uptake of lifestyle behaviours included items about diet, physical activity, alcohol consumption, and smoking behaviour. These items were chosen based on cancer prevention information provided by the Canadian Cancer Society and were patterned after lifestyle behaviour items included in the Canadian Community Health Survey (Statistics Canada, 2000-2001). Women were asked whether they choose or avoid certain foods because they are concerned about HBOC, and if so, to specify which foods. Three items asked women whether they had changed their physical activity, their consumption of alcohol, or their smoking behaviour because they are concerned about HBOC. The uptake of CAM use was measured by asking women whether they had an appointment with a CAM provider about how to prevent HBOC or whether they had begun taking any natural health products to prevent HBOC. Three open-ended items assessed information needs regarding breast and ovarian cancer risk reduction.

Measures were included to assess women's levels of perceived risk and psychological distress.

*Perceived risk* of developing breast or ovarian cancer was assessed by asking women to rate on a Likert-type scale their personal risk in comparison to the general population risk, using categories of "mildly increased risk," "moderately increased risk", "high risk", and "very high risk" (Esplen et al., 2000).

*Psychological distress* was measured using the Impact of Event Scale (IES). The IES is a fifteen-item, Likert-type scale used to assess the experience of distress for a specific life event (Horowitz et al, 1979). The IES has good internal consistency and has been used previously to assess the stress impact among individuals at increased risk for cancer and to describe preoccupation with disease risk (Lerman et al, 1995).

Women's *information needs* were obtained using two open-ended items.

### ***Data Analysis***

The analysis of quantitative data consisted of univariate analysis of demographic factors, perceived risk, distress, and RR strategies. One missing value for age was replaced by the mean of the sample, while nine missing values for income were replaced by the median for the sample. Six missing values for risk perception, and between four and six missing values for one of the fifteen items of the IES were replaced by the sample mean (Tabachnick & Fidell, 2007). This was followed by bivariate analysis to determine relationships between demographic factors, perceived risk, distress, and RR strategies. In order to make the analysis and interpretation of relationships among variables more meaningful, the following categories were dichotomized: marital status (married vs not married), ethnicity (Ashkenazi Jewish vs not Ashkenazi Jewish), and employment status (employed vs not employed). Other factors were collapsed into three categories based on naturally occurring groups including: level of education (high school or less, part of university/college, and completed university college), household income (less than \$60,000, \$61,000 to \$100,000, and greater than \$100,000), and perceived risk (low risk, moderate risk, and high risk). The Cronbach's alpha reliability coefficient for the IES was 0.92.

Multivariate analysis using logistic regression (binomial) with the appropriate explanatory variables, as determined by previous bivariate analysis exhibiting  $p < .05$  associations, was performed to determine

the degree to which RR strategies could be predicted. Multivariate analysis was only carried out when sufficient numbers of women reported use of the RR strategy (with frequencies greater than 5 in each of the cells), and more than one predictor variable was significantly associated with the outcome variable using bivariate statistics. All statistical analyses were performed using the statistical software SPSS version 16.

Participants' responses to the open-ended questions about their information needs included in the HBOC risk-reducing questionnaire were analyzed using content analysis.

## **Results**

### ***Sample Characteristics***

The demographics of the sample, as well as the participants' scores on the measures of perceived risk and distress, are listed in Table 2.1. The mean age of participants was 45.8 years of age (range 19 to 79 years) and the majority of participants were married (74.8%) and had children (68.5%). Only 30.0% of the sample identified as being Ashkenazi Jewish. The sample was highly educated, with 68.9% having completed university or college, and the majority were employed full- or part-time (72.7%). Almost 40% had an annual household income greater than \$100,000. In terms of perceived risk, over 70% considered their risk of developing cancer to be moderately increased (41.3%), high (23.1%), or very high (7.0%) compared to the general population. The mean distress score of participants on the IES was 15.0 (range 0 to 51), a level indicative of mild distress.

Table 2.1 Characteristics of Study Sample

Characteristics	Frequency (%)
Age (years)	
19 – 39 years	49 (34.4)
40 – 49	39 (27.2)
50 – 69	51 (35.7)
70 and greater	4 (2.8)
Marital Status	
Single, never married	19 (13.3)
Married/common-law	107 (74.8)
Separated/ divorced	15 (10.5)
Widowed	2 (1.4)
Ethnicity	
Anglo-Saxon	62 (44.1)
Ashkenazi Jewish	42 (30.0)
Asian	7 (4.8)
Hispanic	4 (2.8)
Other	28 (19.3)
Education	
Part of high school	5 (3.4)
High school	18 (12.4)
Part of university/college	20 (15.2)
University/college	65 (44.8)
Graduate school	35 (24.1)
Employment	
Employed full time	81 (56.6)
Employed part time	23 (16.1)
Unemployed	8 (5.6)
Retired	12 (8.4)
Homemaker	17 (11.9)
Student	2 (1.4)
Household annual income (Canadian)	
\$0 - 20,000	9 (6.3)
\$21,000 – 40,000	11 (7.7)
\$41,000 – 60,000	25 (17.5)
\$61,000 - \$80,000	16 (11.2)
\$81,000 – 100,000	25 (17.5)
Greater than \$100,000	57 (39.9)
Children	
Have children	98 (68.5)
No children	45 (31.5)
Perceived Risk	
Mildly increased risk or less	41 (28.7)
Moderately increased risk	59 (41.3)
High or very high risk	43 (30.1)
Distress	
Sub clinical	69 (48.3)
Mild	38 (26.6)
Moderate to high	36 (25.5)

Note. N = 143

## ***Breast Cancer Screening***

### ***Bivariate analysis***

The use of breast cancer screening by participants is reported in Table 2.2. Performing BSE was significantly associated with employment status ( $\chi^2(1, N = 142) = 4.347, p = .037$ ); women were 2.33 times more likely to have performed BSE if they were employed. Having had a CBE was associated with having children ( $\chi^2(1, N = 142) = 6.094, p = .014$ ), such that women with children were 2.48 times more likely to have had a CBE within the last year. Mammography use was positively associated with older age ( $r_{pb} = .425, p < .000$ ), having children ( $\chi^2(1, N = 142) = 9.551, p = .002$ ), being Ashkenazi Jewish ( $\chi^2(1, N = 142) = 6.973, p = .008$ ), being employed ( $\chi^2(2, N = 142) = 6.999, p = .008$ ) and higher levels of education ( $\chi^2(2, N = 142) = 6.679, p = .035$ ). BSE, CBE, and mammography use were not significantly associated with household annual income, perceived risk, or distress. When broken down by age, mammography use was reported by seventeen (34.7 %) women 19 to 39 years, twenty-six (66.7%) women 40 to 49 years, forty-two (82.4%) women 50 to 69 years, and three (75.0%) women 70 and older. Having had a breast ultrasound was not significantly associated with any of the demographic variables, perceived risk, or distress.

### ***Multivariate analysis***

The following variables were included in a backwards (likelihood ratio) stepwise logistic regression modeling mammography utilization: age (in years), children (no vs. yes), Ashkenazi Jewish background (no vs. yes), employment status (unemployed vs. employed), and level of education (high school vs. some university or college vs completed university or college). A test of the full model with all five predictors against a constant-only model was statistically significant ( $\chi^2(6, N = 142) = 33.69, p < .000$ ), indicating that the predictors, as a set, reliably distinguished between women who had a mammogram in the last year and those who did not. However, only age remained in the final model ( $\chi^2(1, N = 142) = 21.792, p < .000$ , OR = 1.09, 95% CI = 1.05, 1.13). The Hosmer-Lemeshow statistic was nonsignificant ( $p = .186$ ),

indicating that this is a good model. The variance in mammography use accounted for by age was moderate with  $R^2_{cs} = .180$  and  $R^2_N = .244$ .

### ***Ovarian Cancer Screening***

#### ***Bivariate analysis***

The use of ovarian cancer screening by participants is reported in Table 2.2. Having had a manual pelvic exam to screen for ovarian cancer within the last year was not associated with any of the demographic variables, or the variables perceived risk or distress. Having undergone a TVU within the last year was positively associated with older age ( $r_{pb} = .215, p = .011$ ), and having children ( $X^2(1, N = 138) = 6.502, p = .011$ ), with women with children 2.76 times more likely to have had a TVU. Having had a serum CA 125 within the last year was positively associated with older age ( $r_{pb} = .228, p = .006$ ) and having children ( $X^2(1, N = 108) = 7.952, p = .005$ ) (OR = 3.65). Neither TVU nor serum CA 125 were significantly associated with perceived risk or distress.

Table 2.2 Cancer Screening

Screening Test	Time of Last Test	Frequency (%)
Breast self-examination		
	Within the last 6 months	109 (76.2)
	7 months to 1 year ago	7 (4.9)
	More than 1 year ago	13 (9.1)
	Never	13 (9.1)
	N/A (no breasts)	1 (0.7)
Clinical breast examination		
	Within the last 6 months	86 (60.1)
	7 months to 1 year ago	34 (23.8)
	More than 1 year ago	19 (13.3)
	Never	3 (2.1)
	N/A (no breasts)	1 (0.7)
Mammogram		
	Within the last year	88 (61.5)
	More than 1 year ago	25 (17.5)
	Never	29 (20.3)
	N/A (no breasts)	1 (0.7)
Breast ultrasound		
	Within the last year	36 (25.2)
	More than 1 year ago	40 (28.0)
	Never	64 (44.8)
	Do not know	2 (1.4)
	N/A (no breasts)	1 (0.7)
Manual pelvic examination by a health care professional		
	Within the last year	104 (72.7)
	More than 1 year ago	29 (20.3)
	Never	5 (3.5)
	Do not know	3 (2.1)
	N/A (no ovaries)	2 (1.4)
Transvaginal ultrasound		
	Within the last year	56 (39.2)
	More than 1 year ago	36 (25.2)
	Never	46 (22.2)
	Do not know	3 (2.1)
	N/A (no ovaries)	2 (1.4)
Serum CA 125		
	Within the last year	39 (27.3)
	More than 1 year ago	7 (4.9)
	Never	62 (43.4)
	Do not know	33 (23.1)
	N/A (no ovaries)	2 (1.4)

Note. N = 143

### *Multivariate analysis*

Only age (in years) and children (no vs. yes) were included in a backwards (likelihood ratio) stepwise logistic regression modeling TVU utilization in the past year. A test of the model with both predictors

against a constant-only model was statistically significant ( $X^2(2, N = 138) = 7.570, p = .023$ ), indicating that the predictors, as a set, reliably distinguished between women who had a TVU in the last year and those who did not. However, only having children remained in the final model ( $X^2(1, N = 138) = 6.269, p = .012$ , OR = .363, 95% CI = .164, .802). The Hosmer-Lemeshow statistic was not calculated because only one categorical predictor remained. The variance in TVU use accounted for by having children was small with  $R^2_{cs} = .048$  and  $R^2_N = .064$ .

Age (in years) and children (no vs. yes) were included in a backwards (likelihood ratio) stepwise logistic regression modeling CA 125 utilization in the past year. A test of the model with both predictors against a constant-only model was statistically significant ( $X^2(2, N = 108) = 14.455, p = .001$ ), indicating that the predictors, as a set, reliably distinguished between women who had a CA 125 in the last year and those who did not. However, only age remained in the final model ( $X^2(1, N = 108) = 5.609, p = .018$ , OR = 1.048, 95% CI = 1.008, 1.090). The Hosmer-Lemeshow statistic was not significant ( $p = .767$ ), indicating that this is a good model. However, the variance in CA 125 use accounted for by age was small, with  $R^2_{cs} = .125$  and  $R^2_N = .172$ .

### ***Pharmacoprevention Therapy and RR Surgery***

The participants' use of pharmacoprevention and RR surgery are reported in Table 2.3. Because of the small numbers of participants who used pharmacoprevention or had undergone RR surgery, it was not appropriate to test whether these were associated or could be explained by demographic variables, perceived risk, or psychological distress.

Table 2.3 Pharmacoprevention and RR Surgery

<b>Preventive Strategy</b>	<b>Frequency (%)</b>
Stopped taking HRT to prevent cancer (out of 20)	6 (30)
Take oral contraceptives to prevent ovarian cancer	4 (2.8)
Stopped taking oral contraceptives to prevent breast cancer	8 (5.6)
Take tamoxifen or raloxifene to prevent cancer	1 (0.7)
Awaiting or undergone RR mastectomy (One or both breasts)	7 (4.9)
Awaiting or undergone RR oophorectomy	5 (3.5)

Note. N = 143

### *Lifestyle Behaviour Change and CAM Use*

Table 2.4 describes lifestyle behavior change and CAM used by the 143 participants to reduce their risk of HBOC. The foods most commonly chosen by participants to reduce their risk of breast cancer included vegetables, fruits, soy products, low fat foods, foods rich in anti-oxidants, high fibre foods, and organic foods. The most frequently avoided foods included red meat, dairy products, soy products, caffeine, sugar, fatty, fried, and processed foods, and foods containing preservatives or trans fats. The only significant factor associated with choosing certain foods to reduce cancer risk was older age ( $r_{pb} = .179, p = .032$ ). There were no variables significantly associated with avoiding foods. The 50 (35%) participants who changed their physical activity engaged in different types of activities, and more frequent and higher intensity exercise. Physical activity change was positively associated with distress ( $r_{pb} = .177, p = .034$ ). Thirteen (13.7%) of the 95 women who consumed alcohol decreased their consumption. A significant association existed between decreased alcohol consumption and income ( $X^2(2) = 8.625, p = .013$ ), such that women with an income less than \$60,000 were 10.5 times more likely than those making \$61,000 to \$100,00, and 3.8 times more likely than those making greater than \$100,000 to have decreased their alcohol consumption. Having children was associated with having reduced tobacco consumption ( $X^2(1) = 4.144, p = .042$ ), with women having children 2.43 times more likely to have reduced their tobacco consumption. CAM use was not associated with any of the demographic variables, or the variables perceived risk or distress.

Table 2.4 Lifestyle and CAM Cancer Prevention Strategies

<b>Preventive Strategy</b>	<b>Frequency (%)</b>
Changed diet by choosing certain foods	40 (28.0)
Changed diet by avoiding certain foods	37 (25.9)
Changed physical activity	50 (35.0)
Changed smoking behaviour (out of 29 women who smoked)	15 (51.7)
Changed consumption of alcohol (out of 95 women who drank alcohol)	13 (13.7)
Had an appointment with a CAM provider	12 (8.4)
Began taking natural health care products	36 (25.2)
Vitamins	33 (23.1)
Herbal therapies	5 (3.5)
Other	7 (4.9)

Note. N = 143

### ***Information Needs***

Information the women indicated they would like about decreasing their HBOC risk is summarized in Table 2.5. Women who wanted dietary information ( $n = 42$ ) expressed a need to know more about what specific foods might help prevent cancer, what foods they should avoid, the recommendations of professional nutritionists, and how to access nutritional counselling. Almost equal numbers of women wanted any pertinent information ( $n = 25$ ), or no information at all ( $n = 23$ ). Some women indicated that if they had any questions they could find answers online, while others did not want any information for the time being. This was evident in statements such as: “My strategic plan at this time is to wait for results [BRCA1/2 testing] before proceeding on the next course of action. I’m purposefully not researching information on what to do if you have breast cancer. Yuck! I’ll wait and see.”

Information needs related to HBOC screening included signs and symptoms of cancer, tutorials about how to perform BSE, the reliability and effectiveness of screening modalities, how to access funded screening, age appropriate screening, and recommendations of health care professionals. The women who wanted more information about RR surgery were interested in the details of RR mastectomy and oophorectomy, what to expect with surgery, possible strategies to help them cope with the consequences of surgery, and the effectiveness of RR surgery. Women also wanted information about CAM therapies that may reduce their risk of cancer, including herbs, vitamins, minerals, supplements, and meditation techniques, as well as CAM providers offering relevant advice or services. Participants indicated they wanted access to up-to-date research, news, and additional information about their cancer risk. Twelve women indicated that they did not know, nor had they received any information, about ways they could reduce their cancer risk.

Table 2.5 Information Needs about Reducing the Risk of Cancer

Desired Information	Number of women
Diet	42
Any information	25
No information	23
Screening	21
CAM	20
Research reports & risk information	14
Exercise	11
General lifestyle	8
RR surgery	7
Environmental toxins	4
Pharmacoprevention	3
Alcohol	3
Tobacco	1

### Discussion

Findings of this descriptive, exploratory study indicate that the breast cancer screening rates of this high-risk sample of unaffected women are higher than those found in the general Canadian population (Public Health Agency of Canada, 2005). Similar to previous research with other high-risk Canadian women (Dorval et al., 2008), the majority performed breast cancer screening in accordance with recommendations congruent with their family history. Yet, these screening rates were not as high as some samples of women with known *BRCA1/2* mutations (Lerman et al., 2000; Metcalfe et al., 2008; Phillips et al., 2006; Tinley et al., 2004). This suggests that even without genetic test results, high-risk women seeking genetic testing may be more motivated than the general population to undergo screening and they are informed about relevant screening recommendations. This could be attributed to higher cancer awareness among this group because of their family history or recommendations of health care professionals, including family physicians and genetic counselors. Genetic counseling appointments prior to genetic testing often include discussions about strategies for HBOC risk reduction. All participants in this study underwent genetic counseling at some point before genetic testing and it is possible that genetic counseling influenced the women's breast cancer screening uptake. Future research examining the messages about breast cancer screening that women recall from genetic counseling is warranted.

It is also noteworthy that older age was predictive of mammography, yet 43 women under the age of 50 years also obtained a mammogram within the last year. This finding may be the result of the fact that some women included in this study were from provinces where biennial mammography is recommended once women are 40 years of age (British Columbia Cancer Agency, 2009). Mammography is generally not recommended for high-risk women less than 30 years of age due to poor test sensitivity. Although it is likely that most of these women were accessing publicly funded screening services, some women indicated that they bypassed health care professionals or screening programs by paying for services they deemed necessary.

The rates of ovarian cancer screening (32% had CA 125 testing and 64 % a TVU) were high considering that these tests have not been shown to reduce morbidity or mortality and there is no proof of their efficacy in *BRCA1/2* mutation carriers (Horsman et al., 2007). It is possible that women who had pap testing to screen for cervical cancer in the previous year confused this with a manual pelvic examination to screen for ovarian cancer. These high ovarian and breast cancer screening rates could also be related to women's heightened awareness of the signs and symptoms of HBOC, their increased likelihood of consulting health care professionals about these concerns, and undergoing screening to rule out a diagnosis of cancer when signs or symptoms presented, such as a breast lump or abdominal bloating.

Consistent with previous research (Phillips et al., 2006; Ray et al., 2005; Schmeler et al., 2006), a small number of women obtained or were awaiting RR surgery prior to receiving their genetic test results. This indicates that knowledge of one's *BRCA1/2* status was superfluous to some women's RR surgery decisions and that other factors were more influential. We were unable to test whether demographic variables, perceived risk, or distress were predictive of RR surgery due to the small number of women who had RR surgery. Previous research provides some insight into the role that medical, physical, psychological, and social context factors play in influencing women's decisions about RR surgery (Howard et al., 2009), yet this question deserves attention in future research.

Study findings that the majority of women were not motivated to change their diet or level of physical activity prior to receipt of genetic test results (72% did not choose certain foods, 74% did not avoid certain foods, and 65% did not change their physical activity) were similar to those of other high-risk Canadian women undergoing *BRCA1/2* testing, wherein 63% reported having average eating habits and 71% failed to exercise at least three times per week (Dorval et al., 2008). It is possible that genetic test results might influence women's future motivation to change their diet and exercise level. In one study, most individuals (85.5%) at high risk for hereditary non-polyposis colorectal cancer indicated that if they carried a genetic mutation, they would try to adopt a healthier lifestyle, but only 60.7% would do so if their test showed them to be at low risk (Brodersen, Sutton, Goff, Hodgson & Thomas, 2004). Although women have reported improving their diet, increasing their physical activity, and attempting to quit smoking following receipt of *BRCA1/2* genetic test results (Spector, 2007), the degree to which genetic test results motivate women to modify their lifestyles remains unknown.

Interestingly, a sub-group of high-risk women in this study were motivated to modify their diet, physical activity level, alcohol, and tobacco use. That women with children were more likely to reduce their tobacco use than women without children is not all that surprising considering that some women attempt to quit smoking out of concern for the health of their children (McDermott, Dobson, & Owen, 2006). Yet, it is unclear why women with the lowest income were more than 10 times likely to decrease their alcohol consumption than women with the highest income. Perhaps they have less exposure to social situations in which alcohol is provided or expected to be consumed. Prevalent messages from public health groups and cancer organizations (i.e., Canadian Cancer Society, 2009) encouraging a diet rich in fibre, fruits and vegetables and low in fat, an active lifestyle, and quitting smoking as a means of preventing breast cancer in the general population could have been the basis upon which women made these lifestyle changes. It is also possible that these women received influential health promotion recommendations from their family physicians and genetic counselors.

Evidence concerning lifestyle modification for those with a significant family history of cancer and for *BRCA1/2* carriers is just beginning to emerge. Thus far, preventive strategies that may delay the

development or reduce the incidence of *BRCA1/2* related breast cancer include caloric restriction, adherence to Canada's food guide to healthy eating, and increased vegetable and fruit diversity (Ghadirian et al., 2009; Nkondjock & Ghadirian, 2007; Nkondjock, Robidoux, Paredes, Narod, & Ghadirian, 2006). Initial evidence related to body weight suggests that weight loss early in adult life (age 18 to 30 years) protects against early onset *BRCA*-associated breast cancer, and that weight gain should also be avoided, particularly among *BRCA1* mutation carriers who have at least two pregnancies (Kotsopoulos et al., 2005). Although there is mixed evidence of the influence of smoking on the risk of breast cancer among *BRCA1/2* carriers (Brunet et al., 1998; Breast Cancer Family Registry et al., 2008; Colilla et al., 2006; Ghadirian et al., 2004; Ginsburg et al., 2009; Gronwald et al., 2006; Nkondjock et al., 2006), an expert panel who recently reviewed this evidence suggested that overall smoking increases risk among those with a family history of breast cancer, as well as *BRCA1/2* carriers (Collishow et al., 2009). Where clear-cut evidence is absent, Dorval et al. (2008) argue that it is plausible to extrapolate that the modifiable risk factors of breast cancer among high-risk women are the same as those identified in the general population.

Findings of this study support the clinical observation that some women are interested in learning about lifestyle choices, particularly related to diet, in order to actively reduce their risk of cancer (Spector, 2007). Lifestyle changes might be less threatening to women because they have fewer negative consequences when compared to other RR strategies, such as pharmacoprevention and RR surgery. These are also actions women can begin to take while waiting for their genetic test results that might help them to gain a sense of control over their risk of HBOC. Moreover, the time of genetic counseling represents a teachable moment wherein some women are interested and motivated to make lifestyle changes. This lends strong support to the opinions of those who advocate for the promotion of preventive lifestyle behaviours to be part of genetic counseling (Biesecker, 2001). But this also raises multiple questions. What specific information and support is most appropriate? Who should develop informational resources? In what form should this information or support be delivered? And what efforts should be made to individualize relevant information and health promotion activities? On a cautionary note, almost equal

numbers of women wanted any information or no information about how to reduce their HBOC risk. This suggests that women's information and referral needs are individual and that some women might not be amenable to health promotion at the time of genetic testing. For those women who do not want information at this time, it is imperative that they be made aware of how to access information and support if and when they are ready.

Fewer women in this study reported using CAM to reduce their HBOC risk (8% consulted a CAM provider and 25% began taking natural health products) than has been reported in other research, wherein CAM use among women at high risk for HBOC ranged from 53% (DiGianni et al., 2003) to 78% (Mueller et al., 2008). One explanation for these disparate findings could be that we specifically asked women whether they were using CAM to reduce their risk of HBOC, while these other studies queried CAM use in general. In fact, a number of the women in the present study commented that they used CAM, but as a general means of health promotion. In the future, women may increasingly be encouraged to use various CAM therapies. For example, scientists have recently turned their attention to nutrigenomics, the study of the effect of nutrients on gene expression, and some have recommended that women at high risk for HBOC consider taking dietary supplements, including lycopene, selenium, diindolylmethane, and green tea polyphenols (Kotsopoulos & Narod, 2005).

The data from this study did not provide support for relationships between perceived risk and distress, and women's use of RR strategies prior to receipt of genetic test results, with the only exception being the positive association between distress and physical activity. These findings add to inconsistent evidence about the relationship between perceived risk and breast cancer screening among women with a first-degree relative with breast cancer (Martin & Lobchuck, 2003). These insignificant results could possibly be attributed to the much stronger influence of other factors related to perceived risk, but not captured with the one item question used in this study, including women's perceived vulnerability to HBOC, perceived control over disease, and beliefs about the effectiveness of RR strategies. Although at least a quarter of the women in this study were experiencing moderate to high levels of distress, this distress may have been related to the process of undergoing genetic testing more so than their pre-existing awareness

of their HBOC high risk status. Alternately, women might cope with the distress stemming from their high-risk status in very different ways; some might be motivated to use RR strategies while others might avoid engaging in behaviours that remind them of, and reinforce, their high-risk status. A reciprocal, cyclical relationship between perceived risk and distress, and women's use of RR strategies, may also exist, such that engaging in risk management affects perceived risk and distress. Regardless, the finding that some women were experiencing high levels of distress suggests that psychological support may be beneficial, particularly because the receipt of positive genetic test results could exacerbate distress.

Limitations to our study include the reliance on self-reported data regarding the use of HBOC RR strategies, lifestyle behaviour change, and CAM use. Further, we did not collect information on the reasons for using screening strategies, which may have included ruling out suspicious signs and symptoms of cancer. Moreover, it was unclear what specific information each of the women received in their genetic counseling session and the influence this had on their use of RR strategies. Another limitation of the study is a relatively small sample size, providing for limited statistical power for some analyses and lack of a comparison group. Finally, our study findings should not be considered representative of all women undergoing genetic testing since it was comprised of women who volunteered to participate in research and had accessed publicly funded health services in Canada.

### **Conclusion**

Women are accessing and using HBOC RR strategies to varying degrees prior to receiving their genetic test results. The time of pre-test genetic counseling presents an optimal time to engage women in discussions about HBOC risk reduction, especially because there may be a substantial delay between the time they go for counseling, when they go for genetic testing, and when they receive their results. This also represents a teachable moment wherein discussions about the benefits of maintaining a healthy lifestyle are appropriate. In addition to receiving information during this time, some women may require support for making decisions about some RR strategies. In order to meet the specific needs of women at high risk for HBOC, resources will need to be developed or referral systems established for those interested.

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## **CHAPTER 3: Women's Constructions of the 'Right Time' to Consider Decisions about Risk-Reducing Mastectomy and Risk-Reducing Oophorectomy<sup>3</sup>**

### **Introduction**

Genetic testing for mutations in the *BRCA1* and *BRCA2* genes has increasingly become available to individuals since the discovery of these genes over 14 years ago (Miki et al., 1994; Wooster et al., 1995). Those women found to carry *BRCA1/2* mutations are at markedly increased probability of developing hereditary breast and ovarian cancer (HBOC), with their lifetime risk of breast cancer between 45% and 88%, and their risk of ovarian cancer ranging from 11% to 65% (Antoniou et al., 2003; Evans et al., 2008; Ford et al., 1998). Accordingly, when unaffected women are notified that they have a *BRCA1/2* mutation, they are presented with a range of HBOC risk-reducing (RR) options including risk-reducing mastectomy (RRM) and risk-reducing oophorectomy (RRO). However, there is growing evidence that a sub-group of women do not make decisions about RRM or RRO immediately after genetic testing, but rather, prolong these decisions for months or years (Antill et al., 2006; Bradbury et al., 2008; Evans et al., 2009; Friebe et al., 2007). Women's experiences with RRM and RRO have begun to be described (Josephson Wickman, & Sandelin, 2000; Lloyd, Watson, Oaker, Sacks, Querci della Rovere, & Gui, 2000; Metcalfe Esplen, Goel, & Narod, 2004), but their perspectives on the timing of these decisions are not well understood. Accordingly, the purpose of this study was to describe how women who carry *BRCA1/2* mutations construct the 'right time' to consider decisions about risk-reducing surgeries.

### **Background Literature**

Women found to carry a *BRCA1/2* mutation are faced with difficult decisions about how to manage their elevated HBOC risk. Breast cancer screening is recommended to these women as a means of identifying cancers at an early stage when the prognosis of treatment is good, thus reducing the risk of dying from cancer. The most effective means of preventing breast cancer is through RRM, the surgical removal of healthy breast tissue prior to the development of cancer, with the option of reconstructive

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surgery. However, RRM is considered controversial because of the potential for psychological harm. It is, thus, generally framed as a woman's personal decision to be discussed with her health care professional (Horsman et al., 2007). As there is no evidence that ovarian cancer screening is effective in reducing mortality, it is currently assumed that RRO is the best form of risk management. As such, RRO, the surgical removal of the fallopian tubes and ovaries, is commonly recommended to women once childbearing is complete (Horsman et al., 2007).

Women are encouraged to make decisions about RRM and RRO when they receive their genetic test results. However, there is evidence that some women are delaying these decisions. In previous research, there were women who underwent RRO and/or RRM years after receiving their genetic test results (Antill et al., 2006; Bradbury et al., 2008; Evans et al., 2009; Friebel et al., 2007). In the largest of these studies, which included 297 women who had RRO and 113 women who had RRM, the average time from genetic testing to RR surgery was 1 year (Friebel et al., 2007). Yet, some of these women had RRO up to 7 years later and RRM more than 8 years later. For women in another study who previously had a mastectomy to treat breast cancer, the mean time that elapsed between their primary surgery and contralateral RRM was 3.5 years (Metcalf et al., 2008). The amount of time between receipt of genetic test results and RR surgery has raised questions about why women are not acting promptly to reduce their risk, and concerns that the full benefits of genetic testing are not being realized.

It has been suggested that medical, physical, psychological, and social context factors influence the timing of decisions and use of RR options. Women's childbearing and menopausal status have been reported to have a substantial bearing on how women frame the optimal timing of these decisions, as have women's relationships and their responsibilities to family members, friends, and employers (Bradbury et al., 2008; Brain, Gravell, France, Fiander, & Gray, 2004; Hallowell, Jacobs, Richards, Mackay, & Gore, 2001; Hamilton, Williams, Bowers, & Calzone, 2009; Howard, Balneaves, & Bottorff, 2009).

The psychological consequences of genetic testing, including anxiety, distress, and worry, may dramatically influence women's abilities to make RR decisions, as well as the nature of the decision-making process. Although these psychological symptoms decline over time for the majority of mutation

carriers (Arver, Haegermark, Platten, Lindblom, & Brandberg, 2004; Hamilton, Lobel, & Moyer, 2009; Smith, Dougall, Posluszny, Somers, Rubinstein, & Baum, 2008; van Oostrom et al., 2003), significant psychological distress can persist for others (Butow, Lobb, Meiser, Barratt, & Tucker, 2003; van Oostrom et al., 2003). Moreover, researchers have reported a positive relationship between distress, anxiety, and worry, and decisions about RR strategies (Fry, Busby-Earle, Rush, & Cull, 2001; Hurley, Miller, Costalas, Gillespie, & Daly, 2001; Lodder et al., 2002; Meiser et al., 1999; Meiser et al., 2000; Stefanek, Enger, Benkendorf, Flamm Honig, & Lerman, 1995; Unic, Verhoef, Stalmeier, & van Daal, 2000).

When positive *BRCA1/2* results are disclosed women are also faced with interpreting complex and emotion-laden information. Women have reported spending much time reviewing information and seeking additional sources of information and advice in an attempt to resolve their questions about the potential impact of RRM on their lives (McCullum, Bottorff, Kelly, Kieffer, & Balneaves, 2007). In one study, the lack of absolutes about when, or if, the women would develop HBOC led to uncertainty about the right time to make RRM and RRO decisions (Howard, Balneaves, Bottorff, & Rodney, 2009). Moreover, *BRCA1/2* carriers have expressed a need to consider relevant information in the context of their individual experiences, perceptions, and psychosocial needs (Babb, Swisher, Heller, Whelan, Mutch, Herzog, et al., 2002).

It is also important to acknowledge that once women receive their genetic test results they may encounter barriers and delays accessing health services that are central to decisions about RR surgery and receipt of risk management choices. For example, obtaining consultations with specialized health care professionals may take months and even years. Women in rural settings may experience additional barriers to treatment including costs of travel and lost time away from employment (Howard, Balneaves, Bottorff, & Rodney, 2009).

Although previous research provides important insights related to RR decision making, women's perspectives related to the timing of decisions has not been systematically studied. With few exceptions, the focus of research to date has been guided by theoretical perspectives and assumptions that have not adequately captured the complexity of women's decision making (Howard, Balneaves, & Bottorff, 2009),

or their perspectives on the importance of making timely decisions and factors that influence timing. A few studies have described women's decision-making processes about RR surgery as complex, dynamic, and prolonged (Hallowell et al., 2001; McCullum et al., 2007). In a case study that involved in-depth interviews with three women over one year, researchers tracked how women's thoughts and decisions about RRM changed over time, and the time they needed to achieve a level of comfort with decisions that they were able to follow through with (McCullum et al., 2007). It is essential that health care professionals consider women's perspectives on the 'right time' to make these decisions given the potential psychological implications of making untimely decisions, as well as the implications of delaying RRM and RRO with regard to women's morbidity and mortality. A better understanding of women's perspectives is also needed to guide the provision of RRM and RRO decision support as well as the development of decision support interventions that address the complex needs faced by women at high risk for HBOC.

### **Methods**

This study was conducted in the context of a larger grounded theory study aimed at developing an in-depth theory of women's experiences of making decisions about HBOC RR strategies (Howard, Balneaves, Bottorff, & Rodney, 2009). Qualitative methodological approaches described by Charmaz (2006), Strauss and Corbin (1998) and Sandelowski (2000) were used to develop descriptions of how women construct the 'right time' to consider decisions about RRM and RRO.

### ***Study Participants***

A behavioral research ethics board approved this research and all women participating in this study provided informed consent. Women who received positive *BRCA1/2* genetic test results, were older than 18 years of age, and English speaking, were included in this study. Women were excluded who were currently undergoing diagnostic testing for cancer or were receiving cancer treatments. Participants were recruited through a provincial hereditary cancer program, which recommends RRO to women once childbearing is complete, and offers RRM as an option to be considered.

The demographic characteristics of the 22 participants are illustrated in Table 3.1. The mean age of participants was 51 years of age (ranging from 28 to 80 years). The majority of women were Caucasian, college or university educated, and employed either full- or part-time. Most of the women were married, and just over half had children. The length of time since the women had undergone genetic testing ranged from less than a year to nine years. Most did not have a previous breast cancer history.

Table 3.1 Characteristics of Study Sample

Characteristics	Frequency (%)
Marital Status	
Married/common-law	17 (77)
Single/separated/divorced/widowed	5 (23)
Ethnicity	
Anglo-Saxon	11 (50)
Ashkenazi Jewish	6 (27)
Other	5 (23)
Education	
Some college and above	15 (68)
High school and below	7 (32)
Employment	
Employed	18 (82)
Unemployed/homemaker/retired	4 (18)
Household annual income (Canadian)	
Less than 40,000	4 (18)
\$41,000 – 80,000	8 (36)
Greater than \$80,000	10 (46)
Children	
Have children	12 (55)
No children	10 (45)
Time since genetic testing	
Less than 1 year	5 (23)
1-4 years	4 (18)
Greater than 5 years	13 (59)
Cancer history	
Previous breast cancer history	5 (23)
No previous breast cancer history	17 (77)

Note. N = 22

### ***Data Collection Procedures***

In-depth interviews, lasting 45 to 90 minutes, were conducted at a time and place convenient to the participants. All interviews were digitally recorded and transcribed verbatim. Each woman in this research was assigned a pseudonym in order to maintain her anonymity. The initial interviews began with open-ended questions to elicit participants' perspectives and were designed to draw out the underlying *process* of decision making, which also captured the timing of decisions (Charmaz, 2006). For example,

we asked the women to describe: the factors (medical, physical, psychological, and family context) they perceive as influencing when decisions about RR surgeries were considered; how they perceive their relationships with family and health care professionals as influencing when decisions about RR surgeries were considered; how they perceive organizations, institutions, or the health care system as influencing when decisions about RR surgeries were considered; and how their perceptions of the appropriate time to consider RR surgeries have changed over time. As data analysis proceeded, the questions became more specific to fill in gaps, explore important areas in greater depth, and verify emerging findings (Charmaz, 2006).

### ***Data Analysis Procedures***

Data analysis involved the constant comparison of data from different participants, and across incidents and themes (Charmaz, 2006; Strauss & Corbin, 1998). The transcripts were read numerous times and important ideas, interpretations, and themes relevant to the research questions were identified using an inductive approach (Morse & Field, 1995). Each woman's decision-making experience was captured in a narrative summary noting what RR decisions were made and when, as well as whether these decisions changed over time. In addition, interview data related to the timing of decisions was identified, and coded for retrieval and in-depth analysis using the data management software program NVivo. These data were reviewed to identify themes that recurred within the interviews and were evident in multiple women's accounts. During the next phase of analysis, exploring and describing the relationships within and between the themes helped to refine these thematic categories. This continued until all ideas, interpretations, and themes were accounted for in the final description.

### **Findings**

Decisions about RR surgery evolved over months and even years for many of the women in this study. At the time of being interviewed, all of the women were having biannual cancer screening. Thirteen of 19 women with ovaries (3 women previously had salpingo-oophorectomies to treat medical conditions) had undergone RRO and three of the 22 women with breasts had undergone RRM. The time that lapsed between receipt of genetic test results and receipt of RR surgery ranged from three months to

nine years. Of the women who had not undergone RR surgery, the majority had not ruled out considering these decisions in the future. It was important to the women for decisions about RRM and RRO to be made one at a time. Moreover, the women constructed the ‘right time’ to consider decisions about RR surgery to be when: (1) decisions fit into their lives, (2) they had enough time to think about decisions, (3) they were ready emotionally to deal with the decisions and the consequences, (4) all of the issues and conflicts were sorted out, (5) there were better options available, and (6) the health care system was ready for them.

### *One Decision at a Time*

The women in this study, with one exception, did not engage in decisions about RRM and RRO concurrently. Rather, they prioritized their decisions and then considered them one at a time. The decisions the women deemed most relevant to them, less disruptive to their lives, uncomplicated, associated with fewer negative physical and emotional consequences, and supported by others, were made first. In keeping with this, most of the women considered decisions about RRO first, and then RRM. The women also commonly made “temporary” decisions and then revisited their decisions later on, when they thought it was the ‘right time.’ Leslie, a 55 year old woman who received her genetic test results two years previously, described her approach to considering RR surgery:

Well I basically had gone from, in the summer, saying I’m going to focus on my ovaries, get that done, worry about the breast thing, and then getting really enthusiastic about maybe doing the breast stuff, going and seeing the plastic surgeon and being totally turned off, and going totally against it. And then I guess this year it came up again, when I had another mammogram in early February and then they wanted to do a bigger magnification of it. And then that started me thinking again about this. Am I making the right decision? Is this really safe to do this route, the high surveillance? But, I’ve re-examined it again, and I still feel that it’s premature to make a decision.

The women’s experiences with cancer screening and making or following through with RR surgery decisions had a significant influence on when subsequent decisions were considered. A number of women felt “protected” from cancer because they had faith in the efficacy of screening and believed that if

detected early, cancer was easily treated. The reassurance associated with screening resulted in many women feeling comfortable “sticking with screening” for the time being and postponing RRM and RRO decisions. However, anxiety, physical discomfort and experiences of finding “something suspicious” during screening prompted a revisiting of their RR surgery decisions. This was also the case when women’s negative experiences with RRO resulted in their re-evaluating the ‘right time’ to consider RRM.

### ***When Decisions Fit into my Life***

The women wanted to fit decisions about RR surgery into their current and future life plans. For some, this meant postponing decisions until important events occurred, or phases of their lives completed. Younger, single women considered the importance of their ovaries to appearing youthful and breasts to appearing attractive to potential partners. The women interested in dating, starting intimate relationships and finding a partner preferred to postpone decisions about RR surgery until they were in a serious relationship. Although most married women acknowledged that their husbands loved them for more than their breasts and ovaries, some women questioned the effects RRM and RRO would have on their relationship. It was important for these women to obtain reassurance from their partners before they seriously considered RR surgery. For some of the women who still wanted children, or who were not done childbearing, both RRM and RRO were considered premature. Christine, a 42-year-old single woman who had known her genetic test results for five years, recalled how the decision not to have children was crucial to the timing of her decision about RRO:

What I had to come to terms with mostly before I could think about the surgery [RRO] was that I wasn’t going to have kids. Once I had come to terms with that, and then basically getting to the point where I don’t want to have children, made it a lot easier for me. I never had a partner for very long and I don’t have a lot of money. Unless you can really provide for a child it doesn’t seem right.

These women felt reassured postponing their decisions about RRO because this was congruent with the hereditary cancer program recommendations to wait to make this decision until childbearing was complete. However, some women contemplated altering their life plans by looking into alternatives for having children (i.e., adoption or “freezing their eggs” and undergoing in-vitro fertilization) and engaging

in family planning discussions with their partners to determine whether changing their pre-existing plans was an option.

The women also constructed the ‘right time’ to consider decisions about RR surgery to be when these decisions aligned with their perception of who they were at particular times and phases of their lives. This included how the women saw themselves (e.g., as younger and older), their bodies (e.g., as fertile and post-menopausal), and themselves in relation to others (e.g., as single, an intimate partner, and a widow), in the present and the future. When RR surgery was seen to challenge the women’s self-concept, or would prevent them from becoming the person they wanted to be in the future, decisions about RR surgery were postponed until they could safely be incorporated into their self-concept.

### ***When I Have Taken Enough Time to Think About Decisions***

The women needed to have enough time to deliberate about RR surgery decisions so that they felt ready and able to make these decisions. Taking time to carefully consider these “big” and “complicated” decisions was not only considered by the women to be necessary, but was encouraged by health care professionals, family and friends. Maureen, a 48-year-old woman who had known her genetic test results for five years, recalled the advice given to her by her family physician:

She [physician] just said. “You know it [RRM] is irreversible, so once you’ve made that decision, there’s no turning back. So take your time, and weigh the pros and cons.” And there are just so many pros and cons and it’s huge. I mean this decision is really huge.

The women had many questions and things to think about, such as the intricate details of surgery and reconstruction, possible surgical side effects and complications, and the consequences of surgery on their feelings of femininity, body image, sexuality, and self-esteem. The ‘right time’ to consider these decisions was when the women had obtained answers to all of their questions, and thoroughly considered all facets of their possible options. This involved the use of a variety of decision-making approaches over time, including engaging with others, looking inwards, making sense of the numbers about risks, and weighing the pros and cons, as described in detail elsewhere (Howard, Balneaves, Bottorff, & Rodney, 2009).

The ‘right time’ to consider decisions about RR surgery was also when the women could concentrate their energies on making these decisions. Because some women’s lives were busy, they postponed their decisions until they could give them their full attention. Dealing with other existing health problems, such as cardiovascular disease, chronic fatigue syndrome, or fibromyalgia, took precedence over considerations about preventive surgery, as did providing care to ill family members. For example, Coral, a 51-year-old breast cancer survivor, had postponed making a decision about RRM for six years because she was too busy providing care to her ailing parents and then organizing the family affairs once they passed away. “I wasn’t ready [to decide about RRM] with all the responsibilities that I had to take care of. So now I just have myself and I don’t have anybody to worry about.”

### ***When I am Ready Emotionally to Deal with Decisions***

The women perceived that they needed time to deal with the emotional consequences of carrying a *BRCA1/2* mutation before they could make decisions about RR surgery. At times, the women were preoccupied with related anxiety, distress, and worry, which not only interfered with their everyday lives, but also prevented them from feeling capable of fully engaging in decision making. Christine, a 42-year-old single woman who had known her genetic test results for five years, recalled feeling too overwhelmed to engage in a yearly conversation about RRO with her physician:

It’s so unconscious or irrational the kind of fear that you feel, that I didn’t even show up for my appointment. And that’s so not like me and I was in trouble with Dr. [X]. She had to rebook and see me on a prostate day, which was not good. So that was one thing where I just kind of shut down and I think that day I just stayed home and cried and I was really upset. But nothing feels very conscious. It’s very emotional.

Feeling emotionally overwhelmed led some women to feel uncertain and apprehensive about their abilities to make RR decisions. Postponing these decisions gave the women the time they needed to cope with their emotions, get accustomed to the ideas of RRO, RRM and breast reconstruction, and to come to terms with the consequences of these decisions. Two women sought professional psychological support to help them cope with their anxiety and depression brought on following genetic testing. The women

constructed the ‘right time’ to consider these decisions to be when they had sufficiently addressed their anxiety, distress and worry to be able to engage in decision making without feeling overwhelmed.

***When all of the Issues and Conflicts are Sorted Out***

The women indicated that they needed to address the issues and conflicts that the decisions raised before they could finalize decisions about RR surgery. Sorting out the issues and conflicts within themselves, and with their family and health care professionals often took time. Inner conflict was the result of the women feeling uncertain about their HBOC risk, having numerous questions about RR surgery and breast reconstruction, and encountering difficulty with weighing the pros and cons of these decisions. For example, when Lauren, a 40-year-old woman, consulted a surgeon intending to have RRO, she was informed she would have to take hormone replacement therapy post-operatively - “So then the hormones would increase my chance for breast cancer, and so there was all that conflict.” Although gathering information and advice was helpful, at times this resulted in obtaining new information that needed to be considered in decisions and added further complexity to decision making. In these situations, women were often left feeling more confused and conflicted about the decisions they were trying to make. For example, Leslie, a 55 year old woman who received her genetic test results two years previously, described feeling conflicted about breast reconstruction upon learning about the details of the procedure:

But then I find out that they put it [the implant] behind your pectoralis major muscle. They don’t put it in your skin where your breast was. They put it behind your pectoralis major, between that and your chest wall. So then they have to fill this thing with water and stretch the muscle every 2 weeks. That’s what the expansion thing is. They say it’s for your skin on all the websites. No, it’s for the muscle, to accommodate this lump. And I just thought that’s ridiculous. I couldn’t believe that’s what they did. So now I don’t know what to do.

There were also issues and conflicts involving family members that the women wanted resolved before they felt comfortable finalizing their decisions. Some family members voiced their concerns about RR surgery, while others expressed their opinions about the right course of action for the women to take.

Wanting to remain sensitive to family members' concerns and opinions, some women postponed their decisions or followed through with their decisions at a later time. For example, Lilith, a 39-year-old woman, decided to have RRO and RRM a couple of months after she was found to carry a *BRCA1/2* mutation. But, she postponed RRM indefinitely and RRO for over a year because her mother was upset and disagreed with her decision. Sorting through family conflict also involved taking the time to provide explanations and information, and to try and convince family members that the decision the women were leaning towards was the best decision for them.

Sorting through issues with health care professionals was also an essential step to settling on decisions about RR surgery. Seeking second and third medical opinions in order to investigate alternative options, double check recommendations, gather more information, and obtain advice took time. Furthermore, these interactions complicated decisions when these professionals provided advice that conflicted with the recommendations or advice they previously received. Often times, disagreements and conflict with health care professionals stemmed from disparate views about what was the most important, acceptable or desirable decision. For example, at the time of the interview, Coral's decision to have RRM and reconstruction had been prolonged approximately six months because her preferences for reconstruction differed from those of her physician:

Right now we're arguing about the size [that my reconstructed breast should be]. He [physician] says well, you know with the breadth of your chest you have to be a C cup. And I'm going, "Oh please, make them smaller because you have no idea what it's like. You carry twenty pounds on your chest and then tell me that you want me to be the same size." Well I have the surgeon on my side now. She says, "It's men, you know what it's like." I mean he is the professional so I said, "Why do you think I'll look freakish? I want a B cup. I see no difference between a B or a C cup as far as my chest wall goes." So I'm just goanna stick with it.

### ***When there are Better Options***

Some women thought the time was not yet right to consider making decisions about RR surgery because not enough was known about hereditary cancer. The women considered the cancer genetics field

to be a young science characterized by new technology, numerous unanswered questions, and evolving knowledge. Reflecting on the past, the women perceived there to be more options now than ever before to reduce their risk of developing or dying from HBOC, and they expected this trend to continue. As a result, the women perceived the ‘right time’ to consider decisions about RR surgery to be when new scientific evidence emerges and less invasive, less drastic, better tolerated, and more acceptable medical and surgical options and techniques become available and accessible. Some women also relied on the guidance of experts to help them determine when better options would warrant revisiting their previous decisions. This was evident in the explanation provided by Maurice, a 65-year-old woman who received her genetic test results nine years previously:

The consensus of the cancer program and my doctor was that it [RRM] wasn’t necessary. And if it’s not necessary I’m not going to run out and do it. But if anyone thought it would really be a good idea for me, I’m prepared to do it. I have made the decision to stick with screening for now, and I’m letting that decision sit until I hear from someone who knows that there could be a better decision.

### ***When the System is Ready for Me***

The women’s constructions of the ‘right time’ to consider decisions about RR surgery were also influenced by perceptions about when the health care system could accommodate their choice. A number of women engaged in ongoing conversations about RR surgery during biannual breast cancer screening appointments. They had faith in the existing referral processes to access further services that would help them decide about RR surgery. Most women were not upset that referrals to surgeons took up to a year because this gave them time to think more carefully about RR surgery, as evident in Coral’s description:

When I saw Dr. X at the cancer agency for my check up she said, “Well, I can set you up to talk to the surgeon and read some information on it.” And if I wanted to she could put me in touch with other people who had the surgery. A lot of time went by in-between but I wasn’t ready. Waiting to have the chat, I pretty much had time to think about it. And I knew that was what I was going to do. I think my mind was made up before I even saw the surgeon.

Wanting to “get things over and done with” once they had made their decisions, some women felt disappointed and anxious when the system was not ready for them. The lack of seamless care also led some women to postpone their decisions, believing the health care system could not meet their needs, as described by Lauren, who knew her *BRCA1/2* carrier status for four years:

My decision process, and my sister’s decision process, would be a lot different if we could have the reconstruction done at the time as the mastectomy. It’s huge. My sister actually has opted to have both of her breasts removed prophylactically and to have the reconstruction later, and she’s been on a waiting list for two and a half, three years now. It just doesn’t make sense to me to do the surgeries twice. And why can’t we go with dignity and grace? You know, if I’m ever able to have reconstruction at the same time, I’d seriously consider the surgery. It would make my decision easier.

### **Discussion**

The study findings described how women who carry a *BRCA1/2* mutation construct the ‘right time’ to make decisions about RRM and RRO. It was important for the women in this study to consider decisions in the context of their lives, take enough time to deliberate, cope with their emotions, sort through issues and conflicts, wait for better medical and surgical options, and factor in the readiness of the health care system. These findings provide possible explanations for the time frames between genetic testing and RR surgeries observed in other studies (Antill et al., 2006; Bradbury et al., 2008; Evans, et al., 2009; Friebe et al., 2007).

Of note, the women in this study did not frame their decision making as “delayed,” “slow,” or taking “too long”. This differs from researchers and clinicians who are of the opinion that RR surgery is the best way to eradicate breast cancer risk in *BRCA1/2* mutation carriers and have voiced their concern that some women are waiting too long to decide about RRM and RRO (Narod, 2009, April). These conflicting perspectives appear to stem from different concerns and priorities associated with HBOC risk management. On the one hand, some women perceive prolonging or deferring decisions about RR surgery as enhancing their quality of life and helping them manage the inherent decisional conflict. On the other hand, there are also risks with delaying these decisions that cannot be ignored. These women could

develop cancer in the interim. Future research is needed to understand whether these different concerns and priorities complicate the provision of genetic services, and if so, in what capacity.

McCullum and colleagues (2007) reported that women prolonged their RRM decisions while they focused on quality of life issues, specifically the associated risk of decreased physical and emotional well-being. For the women in the present study, the physical consequences associated with RR surgery held different meanings at different phases of their lives. Consistent with the genetic testing and risk management literature (d'Agincourt-Canning, 2006; Hallowell et al., 2001; Hamilton, Williams, et al., 2009; Matloff, Barnett, & Bober, 2009; Ray, Loescher, & Brewer, 2005), younger women considered decisions about RRO and RRM inappropriate because of their life plans, such as finding a life partner, childbearing and childrearing. Yet some of the women contemplated changing these life plans or searching out alternatives, phenomena previously reported but requiring further examination (Hallowell et al., 2001). It is possible that these women were attempting to reconcile their competing desires to have RR surgery sooner than later, but also fulfill important life goals essential to their quality of life. Attending to dimensions of self other than physical health has also been found to be an important part of the decision-making process (Howard, Balneaves, Bottorff, & Rodney, 2009).

Emotional well-being also influenced when the study participants considered RR surgery decisions. These women needed time to deal with the emotional consequences of carrying a *BRCA1/2* mutation and consider the implications of RR surgery before making final decisions. Rather than rush into decisions to cope with their distress, anxiety, and worry, these women prolonged the decision-making process while they coped with their emotions and also postponed their decisions until they felt emotionally ready. Women have previously reported declining genetic testing because of their desire to protect themselves and their family from the emotional consequences (Proulx et al., 2009). In contrast, research that examined predictors of RRM and RRO found anxiety, distress and worry to be key predictors of RR surgery (Fry et al., 2001; Hurley et al., 2001; Lodder et al., 2002; Meiser et al., 1999; Meiser et al., 2000; Stefanek et al., 1995; Unic et al., 2000). Women have described their breasts and ovaries as “time bombs” and RR surgery represented a strategy for managing the associated worry and anxiety (Brain et al., 2004;

Hallowell, 2000). These disparate findings suggest that emotions of anxiety, distress and worry do not always predispose women to make particular decisions at particular times following genetic testing. Instead, emotions play a much more complex role in the timing of women's decisions related to RRM and RRO.

In the study by Kenen and colleagues (2006), women who carried a *BRCA1/2* mutation vacillated from one position to the other and took years to make up their minds about RRM. Most of these women reported spending a great deal of thought on RRM during this decision-making process. The research by Kenen et al., (2006), in combination with findings from the present study, suggests that decisions about RR surgery are fraught with decisional conflict. Decisional conflict occurs when there are: risks or scientific uncertainty about the benefits and harms, choices with large potential gains and losses, value tradeoffs in selecting a particular course of action, and potential regrets with the selected option (O'Connor & Jacobsen, 2007). Tan and colleagues (2009) found that the two most important reasons for women to postpone RRM were uncertainty about proceeding with surgery and the need for more risk information. The lack of conclusive information about the risk of HBOC and the implications of RR surgery were particularly problematic for the women in the present study, as reported in other research (Hallowell, 2000; McCullum et al., 2007; Meiser et al., 2000; Metcalfe, Liede, Hoodfar, Scott, Foulkes, & Narod, 2000). It is, therefore, unsurprising that these women struggled with uncertainty and were compelled to seek out second and third medical opinions over time. Further research is needed to understand the impact of these opinions and advice on women's uncertainty and the decision-making process, as well as how women resolve conflict when differing opinions are garnered.

The research that has focused on decisions about *BRCA1/2* genetic testing suggests that family members are intimately involved in the genetic testing process, before, during and after their relative decides to go forward with testing (Douglas, Hamilton, & Grubs, 2009). Family relationships are also affected when women are found to carry *BRCA1/2* mutations (Douglas et al., 2009; Lim et al., 2004). Researchers have reported that women who tried to discuss hereditary cancer or RR surgery with their families have been met with resistance, shock, hostility and additional negative responses (Kenen, Arden-

Jones, & Eeles, 2004a; Kenen, Arden-Jones, & Eeles, 2004b; Matloff et al., 2009). This resulted in women self-censoring by pulling back or not talking about cancer for fear of causing worry or anxiety (Kenen et al., 2004b). In a similar fashion, the women in this research further postponed making decisions about RR surgery because they wanted to remain sensitive to family member's concerns and opinions. They also attempted to resolve family conflicts and disagreements before they felt comfortable making RRM and RRO decisions. Future research exploring family dynamics in relation to decisions about RR surgery would broaden our understanding of these findings.

International differences in the uptake rates of RR surgeries have been observed and attributed to health care providers' recommendations and continuity of follow-up, as well as cultural variations (Metcalf et al., 2008). For example, in one study, 344 women who attended a cancer genetics clinic in Canada (Quebec), Britain or France were found to vary in their preferences regarding cancer prevention (Julian-Reynier, Bouchard, Evans, Eisinger, Foulkes, Kerr, et al., 2001). The authors attributed this variation to cultural differences between countries. Although it was unclear in this study whether culture was an influential factor, it was apparent that the structure of health services plays a significant role in when women consider RR surgery, as do the barriers women encounter accessing specific services that they believe would best meet their needs. Although timely and accessible health care for all Canadians is a cornerstone of publicly funded health care, patients routinely encounter significant challenges accessing and receiving timely, coordinated and comprehensive care from interprofessional teams (Health Council of Canada, 2008). It appears hereditary cancer and preventive surgical services are no exception. In the present study, the extended decision-making process was in part a by-product of the health care system in Canada, wherein the women encountered lengthy wait times for referrals as well as surgery and breast reconstruction. The finding that this time lag gave some women needed time to think more closely about RR surgery suggests that women are still engaged in the decision-making process and possibly preparing for consultations while they wait. Further inquiry into the feasibility of offering decision-making support during this time is needed.

There are limitations inherent to this research. This study included participants who were recruited from a single hereditary cancer centre in Canada. Caution must therefore be exercised when determining the relevance of the findings in other settings. As well, the perspectives of older women are underrepresented in this study since only one woman was over the age of 65. Moreover, we did not access any participants with positive *BRCA1/2* results who chose not to partake in the recommended HBOC screening. It is possible that the study participants represent a homogeneous group with ready access to screening services and beliefs in early detection. There might also be women who are considering decisions about RRM and RRO who do not have contact with health care professionals during their regular screening appointments. Genetic information, evidence about the effectiveness of HBOC risk management strategies and the availability of online and written resources about RR surgery has changed over the years. Thus, women who received their *BRCA1/2* test results and were availed of the options of RRM and RRO at different times, particularly a number of years ago, might have had different kinds of experiences than women who received their genetic test results more recently.

### **Conclusion**

In conclusion, women's perspectives about the appropriate time to consider decisions about RR surgeries have been absent from the scientific literature to date. Incorporating women's perspectives on the timing of these decisions could provide valuable direction for decision support. The above findings provide evidence that decision support ought to be accessible when women want and need the support because some women prolong or defer decisions to a later time. For those who are overwhelmed with simultaneously facing multiple decisions about HBOC risk management, it may be beneficial to help women prioritize their needs and to work through each of their decisions when they feel ready. Some women may require ongoing psychological support because they find it difficult to come to terms with their HBOC susceptibility, as well the ideas of RRM and RRO. Making this option available to those women who are interested is crucial. The findings suggest that women consider decisions about RR surgery in the context of their life plans, relationships, medical science and the health care system. This provides evidence that broader approaches to decision support, such as relational or women-centered

approaches, might complement existing services. A relational approach could assist women with mobilizing support and resources, communicating with others about HBOC risk management, and working through interpersonal issues. A women-centred approach addresses issues beyond traditional medical interventions, placing health in its broad social context and also addresses barriers to access and respects women's diversity (British Columbia Women's Hospital & Health Centre and British Columbia Centre of Excellence for Women's Health, 2004). Although RR surgery decisions are women's decisions, women should not be saddled with the burden of tackling barriers to accessing health care services. Health care professionals, health care organizations, and government must work hard to resolve these challenges.

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## **CHAPTER 4: Preserving the Self: the Process of Decision Making about Hereditary Breast and Ovarian Cancer Risk Reduction<sup>4</sup>**

### **Introduction**

A woman who receives a positive test for a genetic mutation predisposing her to breast and ovarian cancer is presented with a myriad of risks and decisions. Those with a *BRCA1/2* mutation have a lifetime risk of 45% to 88% for breast cancer and 11% to 65% for ovarian cancer (Antoniou et al., 2003; Evans et al., 2008; Ford et al., 1998). Hereditary breast and ovarian cancer (HBOC) risk-reducing (RR) options include early detection procedures, such as breast self-examination, clinical breast examination, mammography, and magnetic resonance imaging to detect breast cancer. No proven effective screening exists for ovarian cancer. The most effective breast cancer prevention strategy is risk-reducing mastectomy (RRM), generally provided with the option of reconstructive surgery. Similarly, ovarian cancer is best prevented by risk-reducing salpingo-oophorectomy (RRO). Although it is recognized that making decisions about these RR options is complex and holds significant consequences, little is known about women's decision-making processes. Accordingly, the purpose of this study was to develop a theory about women's experiences of making decisions about RR strategies following the disclosure of *BRCA1/2* genetic test results. Understanding women's decision-making processes is an essential step towards developing clinical tools to support decision making.

### **Background Literature**

Research investigating patients' decision making about cancer prevention (Bowen, Allen, Vu, Johnson, Fryer-Edwards, & Hart, 2006; Gorin, Wang, Raich, Bowen, & Hay, 2006; Myers, 2005; Wackerbarth, Peters & Haist, 2008), breast cancer treatment (Balneaves & Long, 1999), and genetic testing (Burgess & d'Agincourt-Canning, 2001; d'Agincourt-Canning & Baird, 2006; Sherwin & Simpson, 1999) has provided some insights into the complexity of these decisions and the importance of social influences on autonomous decision making. An increasing number of theorists have been

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advocating for a relational view of autonomy that characterizes the decision maker as embedded in relational networks that include family, friends, and health professionals, as well as institutional, political and social systems (Donchin, 2000; Sherwin, 1998). This stands in contrast to the individualistic, objective view of autonomy prevalent in early biomedical ethics literature and practice (Rodney, Burgess, McPherson, & Brown, 2004). Relational autonomy highlights the dynamic balance among people who are closely involved in each other's lives, with the individual dependent on close others when trying to achieve a goal (Gilbar & Gilbar, 2009). These relational networks are accompanied by social obligations, including roles and responsibilities, which provide the framework within which decisions are made (Clarke, 2003; Sherwin, 1998).

Family influences have been well documented in cancer prevention and breast cancer treatment decision making (Gilbar & Gilbar, 2009; Hilton, 1994). Several studies have shown that an individual's family responsibilities influences their genetic testing decisions because they are aware of family interests in obtaining genetic information, as well as social obligations and personal interests in maintaining these relationships (Burgess & d'Agincourt-Canning, 2001; d'Agincourt-Canning, 2006; Etchegary, Miller, deLaat, Wilson, Carroll & Cappelli, 2009; Hallowell, 2000; Hallowell, Foster, Eeles, Arden-Jones & Watson, 2003; Mellon et al., 2009). In addition, the gendered context, which shapes the different roles, responsibilities, and activities ascribed to individuals according to their gender, has been shown to figure prominently in women's experiences of cancer prevention and treatment decisions (Helms, O'Hea & Corso, 2008; Kearney, 2006; Lund-Nielsen, Muller & Adamsen, 2005; Ogden & Lindridge, 2008), as well as genetic testing decisions (d'Agincourt-Canning & Baird, 2006). Despite the significance of family and gender, decision-making theories have largely failed to account for the influence of these and other social influences.

Researchers have begun to describe *BRCA1/2* carriers' experiences and satisfaction with HBOC RR strategies (Josephson, Wickman, & Sandelin, 2000; Kenen, Shapiro, Hantsoo, Friedman, & Coyne, 2007; Lloyd, Watson, Oaker, Sacks, Querci della Rovere, & Gui, 2000; Metcalfe, Esplen, Goel, & Narod, 2004), yet few studies explain the preceding decision-making processes (McCullum, Bottorff, Kelly,

Kieffer, & Balneaves, 2007). The limited existing research has largely been descriptive, incorporating such theories of health behaviour as the expected-utility theory (Unic, Verhoef, Stalmeier, & van Daal, 2000), or the self-regulation model of illness representations (Decruyenaer, Evers-Kiebooms, Welkenhuysen, Denayer, & Claes, 2000; Kash, Holland, Osborne, & Miller, 1995). However, these theories do not address complex decision-making situations wherein multiple RR options need to be considered; nor do they take into consideration the other unique factors that influence women who are at risk for HBOC, such as family experiences of cancer (d'Agincourt-Canning, 2005; Hallowell et al., 2001; Mellon et al., 2009) and the associated heightened levels of perceived cancer risk, anxiety and distress (Antill et al., 2006; Claes et al., 2005; Coyne, Kruus, Racioppo, Calzone, & Armstrong, 2003; Coyne, Benazon, Gaba, Calzone, & Weber, 2000; Fang et al., 2003; Fry, Rush, Busby-Earle, & Cull, 2001; Hallowell, 2000; Hurley, Miller, Costalas, Gillespie, & Daly, 2001; Kash et al., 2000; Meiser et al., 1999; Meiser et al., 2000; Stefanek, Enger, Benkendorf, Flamm Honig, & Lerman, 1999; Tiller et al., 2005; Trask et al., 2001; Unic et al., 2000; Wellisch & Lindberg, 2001). Further complicating these decisions are the unique risks associated with RR surgery on self-identity, body image, and femininity (Claes et al., 2005; Fang et al., 2003; Lodder et al., 2002; Matlof, Barnett, & Bober, 2009). Because of the complexity of RR decision making in the context of HBOC there has been a desire to develop decision support interventions. A better understanding of women's decision-making processes and the social contexts that influence these processes is needed to guide the development of these interventions.

### **Methods**

A grounded theory approach (Charmaz, 2006; Strauss & Corbin, 1998) was used to gain an understanding of the HBOC RR decision-making processes of women with *BRCA1/2* mutations.

#### ***Participants and Sampling Strategies***

Women with positive *BRCA1/2* genetic test results, who were older than 18 years of age, and English speaking, were included in this study. Women currently undergoing diagnostic testing or treatment for cancer were excluded. Participants were recruited through a hereditary cancer program, which provides the following recommendations to *BRCA1/2* carriers: (1) for women between the ages of 30 and 69,

clinical breast examination is recommended biannually, while magnetic resonance imaging and mammography are recommended annually, (2) no ovarian cancer screening methods are recommended, (3) RRM is not recommended, but rather, is framed as a personal decision to be discussed with a health professional and, (4) RRO is recommended once childbearing is complete (Horsman et al., 2007).

Women were recruited into this study from December 2007 to August 2008. Open sampling was initially used. Following preliminary analysis, theoretical sampling proceeded on the basis of relevant issues, concepts and insights gained from the initial data (Charmaz, 2006). Data collection continued until no new theoretical insights emerged, categories were well developed and relationships among categories were explained (Charmaz, 2006). This was achieved following interviews with 22 women. Each woman in this research was assigned a pseudonym in order to maintain her anonymity.

#### ***Data Collection Procedures***

In-depth interviews, lasting 45 to 90 minutes, were conducted following receipt of informed consent from the women. All interviews were digitally-recorded and transcribed verbatim. The initial interviews began with open-ended questions to elicit participants' perspectives, as per a preliminary interview guide designed to query phenomena involving processes (Charmaz, 2006). As data analysis proceeded, the nature of the interviews changed with the questions becoming more specific to fill in gaps, explore important areas in greater depth, and verify emerging findings and relationships among concepts. Follow-up interviews with 4 women were conducted to invite responses to preliminary findings.

#### ***Data Analysis Procedures***

Data analysis occurred simultaneous with data collection, involving the *constant comparison* of data from different participants, incidents and categories, until a substantive theory was developed (Charmaz, 2006; Strauss & Corbin, 1998). Data analysis began by reading the interview transcripts line-by-line and highlighting passages that reflected important ideas and themes. These open codes were then condensed, sorted into categories, and entered into the qualitative data management software program NVivo, after which the interview transcripts were coded. The next phase of analysis, axial coding, focused on creating, testing and modifying linkages in the data, which was supplemented by memo writing and diagramming

the developing categories and processes. This continued until all emerging categories and subcategories were placed in a theoretical framework and the connections between categories were described (Charmaz, 2003). Finally, by refining the categories through selective coding, a central concept was identified, under which all other categories were subsumed, thereby explaining most of the variation in the data.

### ***Guiding Analytical Theories: Gender and Relational Autonomy***

The analysis of data was guided by theories of gender (Butler, 1990; Connell & Messerschmidt, 2005; Johnson, Greaves, & Repta, 2007; Lyons, 2009; Saltonstall, 1993) and relational autonomy (Hartrick Doane & Varcoe, 2005; Sherwin, 1998). As such, the influence of gender identity on perceptions of femaleness in the context of HBOC was explored, as were ways in which RR decision making threatened or enhanced self-identity. Data analysis also focused on the influence of women's gendered roles and relations with family, friends and health professionals, on their HBOC RR decision making. Using a relational autonomy lens during data analysis helped to contextualize women's experiences, and guided the exploration of the personal factors, interpersonal relationships and wider social contexts within which they made decisions. For example, when coding the transcripts, particular attention was paid to the women's descriptions of how different people, including family members, friends, health providers, and other *BRCA1/2* carriers, were involved throughout the decision-making processes.

### ***Sample Characteristics***

The demographic characteristics of the 22 participants are illustrated in Table 4.1 The mean age of participants was 51 years of age (ranging from 28 to 80 years). The women were from diverse ethnic backgrounds, with the majority having some college or university education and employed either full- or part-time. Most were married, with just over half having children. There was a wide range in the length of time since the women had undergone genetic testing, and most did not have a previous cancer history.

Table 4.1 Characteristics of Study Sample

Characteristics	Frequency (%)
Marital Status	
Married/common-law	17 (77)
Single/separated/divorced/widowed	5 (23)
Ethnicity	
Anglo-Saxon	11 (50)
Ashkenazi Jewish	6 (27)
Other	5 (23)
Education	
Some college and above	15 (68)
High school and below	7 (32)
Employment	
Employed	18 (82)
Unemployed/homemaker/retired	4 (18)
Household annual income (Canadian)	
Less than 40,000	4 (18)
\$41,000 – 80,000	8 (36)
Greater than \$80,000	10 (46)
Children	
Have children	12 (55)
No children	10 (45)
Time since genetic testing	
Less than 1 year	5 (23)
1-4 years	4 (18)
Greater than 5 years	13 (59)
Cancer history	
Previous cancer history	5 (23)
No previous cancer history	17 (77)

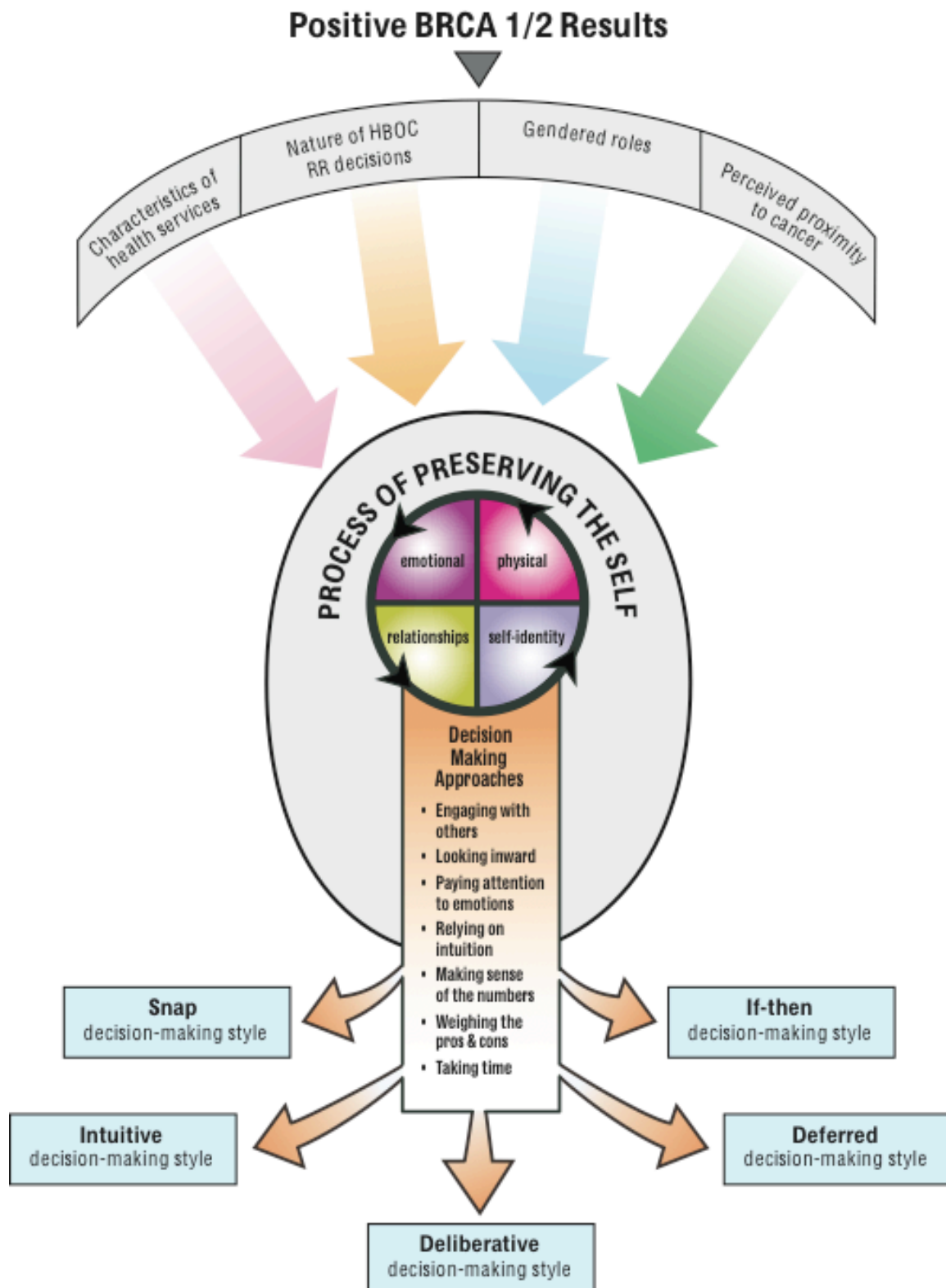
Note. N = 22

## Findings

### *The Process of Preserving the Self: HBOC RR Decision Making*

The findings from this study provide an in-depth theory of women's experiences of making decisions about HBOC RR strategies. In the following sections, the overarching process of *preserving the self* is described, as well as the contexts influencing the women's HBOC RR decisions. The decision-making approaches the women used throughout the process of preserving the self are then outlined. Lastly, we break down this process into five styles of decision making, describing each in turn (see Fig 4.1).

Figure 4.1 Preserving the Self through HBOC RR Decision Making



The overarching decision-making process related to HBOC RR strategies was directed toward preserving the self. Being at risk for HBOC and having to consider RR strategies threatened the women in four dimensions of self; their physical health, self-identity as a woman, relationships with others, and emotional well being. As Christine, a single middle-aged woman recalled, “It’s not rational but it [RRO recommendation] felt threatening, and it definitely felt like I was being threatened on some survival level.” Engaging in decision making about RR enabled women to attend to their physical health, and to protect and secure their self-identities as women, particularly as this related to their youth, beauty, sexuality, body image, and femininity. Active involvement in decision making about RR also supported women in managing and fulfilling their roles and responsibilities to family, friends and health professionals, and helped them to cope with emotions that accompanied HBOC decision making, including anxiety, worry, fear and uncertainty. Although these four dimensions of the self do not encompass all aspects of self, they represent the most salient themes expressed by the women. The four dimensions of the self were interconnected and their degree of significance shifted over time and across RR strategies.

Although the focus on preserving the self often began immediately following receipt of genetic test results, the women who already considered themselves to be at high risk for HBOC had entered into this process long before testing. Other cues for engaging in this process included receiving information and recommendations from health professionals, engaging in cancer prevention or early detection strategies, and experiencing personal life changes, such as having children. The process began with women imaging how the effects of cancer screening, risk-reducing surgery or cancer itself would challenge their physical health, self-identities, relationships, and emotional well being. When women were not ready for imagined changes or saw them as conflicting with their perceptions of self, they made decisions that reinforced their current self-identities, relationships, emotional well being, and/or physical health. Christine recalled her initial thoughts when risk-reducing surgery was recommended to her at the age of thirty-seven:

The first time I went to see Dr. X she mentioned that the preventative measure would be to have the ovaries removed and to remove the breasts as well. And I just thought well, might as well make me a penis and call me Steve at that point. You're going to remove my breasts and my ovaries? And I kind of thought, okay, well we'll talk about this later.

At other times, when women were able to accept imagined changes, they made decisions that enabled them to accommodate or prepare for expected alterations to their identity and physical, emotional, and relational self.

Preserving the self involved five decision-making styles: snap, intuitive, deliberative, deferred, and if-then. Each of these styles was characterized by a unique combination of seven decision-making approaches and differed in the amount of time devoted to decisions, who was involved, and the degree to which the decisions were revisited. The women engaged in different styles of preserving the self, depending on which dimensions of self they privileged. Of importance, the dimensions of self that were privileged and the decision-making styles used, changed over time, across RR strategies, and were influenced by the contexts within which these decisions were situated. Further details of these five decision-making styles are provided following descriptions of the contexts that shaped HBOC RR decision making and the decision-making approaches used by the women in this study.

### ***Contexts Shaping HBOC RR Decision Making***

The context in which women made RR decisions influenced the process of preserving the self. Important contextual influences included (1) the characteristics of health care service, (2) the nature of HBOC RR decisions, (3) gendered roles, and (4) the women's perceived proximity to cancer.

#### ***The characteristics of health care services***

Although access to medical care was purportedly guaranteed through government-funded health care programs, the women's efforts to make decisions were influenced by their perceptions that the health care system was "stressed," financially stretched, and characterized by scarcity. As such, the women believed they needed to be self-reliant and proactive, persistently "pushing" for consultations and timely

appointments to gain information and support for decisions that would help them protect their health. Although there was a sense of appreciation for the access they had to resource-intensive, specialized services, this access was not always guaranteed in a timely manner. For example, the inability to access breast reconstruction immediately following RRM influenced women's decisions about using RRM. Moreover, some women lived in rural areas where costs of travel, taking time off from work, and family responsibilities needed to be considered.

The research culture within the hereditary cancer program also influenced the women's decisions. For many, deciding to be part of a program closely linked to research was a moral action. Women saw themselves "doing their part," with potential future benefits to other women with *BRCA1/2* mutations and future generations. Maureen, a woman who had genetic testing five years previously, exemplified this:

I'm in the system, that's the other thing. So I think because I go, I benefit other people because I am being followed, and they have years with me... But more so, my being in the system potentially will help with the decision making for somebody down the road, which is why I think I continue to go [to the hereditary cancer program].

### ***The nature of HBOC RR decisions***

HBOC RR decisions were complex because they involved uncertainty, were often irreversible, and were situated in medical encounters where a non-directive approach was the norm. In addition to the lack of available conclusive scientific evidence to guide decisions, women found it challenging to interpret the probability of developing HBOC, as well as age-dependent patterns of both breast and ovarian cancer presentation. The lack of absolutes about when, or if, they would develop HBOC led to uncertainty about the right time to make decisions. The women hesitated because preventing the possibility of a future cancer was at odds with the more immediate personal, emotional and relational consequences of making decisions in the here and now.

Decisions about HBOC RR were framed as "big decisions" when RR surgeries were considered because of their irreversibility and profound impact on the women's sense of self. This magnified the

pressure some women felt to make the “right” decision in a way that encompassed all possible facets of the decision. Leslie recalled her conversation with her physician:

She just said. “You know it is irreversible, so once you’ve made that decision, there’s no turning back, so, take your time, and weigh the pros and cons.” And there’s just so many pros and cons and it’s huge. I mean it really is huge.

Although recommendations exist for screening and RRO for women with a *BRCA1/2* mutation, with respect to other types of RR decisions (e.g., RRM), the women received little guidance beyond encouragement to reflect on their own needs and values. Some women, especially those who previously underwent cancer treatment, expected health professionals to provide their opinions and make recommendations because this was the norm in previous interactions. Those women who wanted more direction or collaboration attempted to obtain health professionals’ opinions by asking about hypothetical situations (i.e., “if I were your daughter, what would you tell her to do?”), what they would do if in their situation, and what other high-risk women had chosen.

### ***Gendered roles***

The construction of HBOC RR decisions by hereditary cancer programs as a “woman’s issue” and, therefore, a “woman’s decision,” resulted in a few women taking on the decision-making process independently. For the majority of the women, RR decisions were influenced by important relationships in their lives. Some women recalled listening politely to unsolicited advice offered by family as a means of maintaining existing family dynamics and demonstrating respect. The women also involved family members in the RR decision-making process because they perceived that the state, capacity, and function of their bodies had implications for the lives of others. For example, when making RRO decisions, the women elicited their partner’s family planning desires. In addition, responsibility for making RR decisions was accompanied by the duty to provide, generate and communicate information with the family in a sensitive and appropriate manner (especially for family members with a previous cancer diagnosis). The women also felt an obligation to prevent cancer or detect it early to ensure they would be

able to provide care to family members in the future. At times, the needs of others were prioritized over women's own needs, resulting in the postponement of HBOC RR decisions.

### ***Proximity to Cancer***

The women's perceived proximity to cancer was also an important contextual factor shaping the process of preserving the self by influencing the women's perceived HBOC risk, their emotional reactions, and their sense of urgency to make decisions. The women who described cancer as "far away" had known no relatives with cancer and had not provided care to affected family members. These women were surprised by positive *BRCA1/2* test result but experienced few threats to the integrity of the self because of the belief that "not everyone who carries the gene will get cancer." As a result there was a lack of urgency in the way they approached decision making.

Other women described cancer as "very close." They had known a close relative with cancer, witnessed the disease from a young age, and provided care to a suffering relative. Five of the women in this study had a personal cancer history and were acutely aware of the possibility of recurrence. They perceived themselves at high risk for HBOC prior to genetic testing and expected positive test results. These women had already begun the process of making HBOC RR decisions and test results confirmed that cancer was an imminent threat to be dealt with swiftly. Between these two extremes, the women presented a continuum of perceived proximity to cancer informed by the closeness of their relationship to a stricken family member, the role they assumed in caring for relatives and their age when family members experienced cancer. This translated into varying levels of anxiety regarding the likelihood of cancer as well as variations in perceptions of threat to women's sense of self.

### ***Decision-Making Approaches***

The process of preserving the self involved the women's use of seven decision-making approaches (see Table 4.2) that provided them with important factual, personal, and relational knowledge needed to make decisions that protected their self with respect to their physical and emotional well-being, their relationships with others, and their self-identity.

Table 4.2 Descriptions and Examples of Decision-Making Approaches

Decision making approach	Description and Example
<b>Engaging with others</b>	Included receiving medical guidance, involving, checking-in with and protecting family members, and learning from the first-hand experiences of others.  “My mom could talk it through [RRO decision], and my sister could listen and talk it through, and my girlfriends could do that, but not him [her husband]. I would tell him more what I thought I was going to do.” Maureen
<b>Looking inwards</b>	Involved introspective reflection in which women considered and clarified aspects of self-identity that could be threatened by HBOC RR decisions.  “I mean you can fill the clothes with pads and stuff like that, but I don’t know, I think that [RRM] would really change how I felt about my femininity.” Christine
<b>Paying attention to emotions</b>	Involved taking note of, discerning and monitoring emotions. This also involved considering the effects of RR decisions on emotions, followed by tending to these emotions.  “I just made the decision [to have RRO] because I thought that that was my best opportunity to not worry, and the CA125 was not reliable and even though I was having that, I didn’t really even think it was worth having.” Maureen
<b>Relying on intuition</b>	Involved acknowledging, trusting, and following initial gut reactions and beliefs.  “I don’t remember ever swaying far from my gut feeling, you know, considering, I think I’ve basically stuck with my gut feeling.” Rose
<b>Making sense of the numbers</b>	Involved interpreting and personalizing probabilities, percentages, and statistics about the risks of developing cancer and the efficacy of RR strategies to detect cancer early or prevent cancer.  “I mean you’re sort of looking at all the statistics. You know, this gene has this percent and that gene has that percent, and evidently having both one and two doesn’t increase my risk.” Helen
<b>Weighing pros and cons</b>	Involved weighing and balancing out the positives, benefits, and advantages of RR decisions with the negatives, risks, and disadvantages of RR decisions.  “I have actually spent a few moments trying to weigh out the pros of breast feeding versus, you know, the percentage of risk that might occur over the next few years while we’re waiting to have the next kid.” Angela
<b>Taking time</b>	Involved fitting RR decisions into personal timelines by putting the decisions on hold until a more appropriate or desirable time, or until decision could be made with full knowledge and consideration of benefits and risks.  “You’re going to remove my breasts and my ovaries, and I kind of thought, okay, well we’ll talk about this later, when I’m ready” Christine

### ***Engaging with Others***

As indicated in Table 4.2, the women in this study used the decision-making approach of engaging with others to attend to their self-identities as women, their relationships, emotional well being and physical self. The women obtained advice and medical information crucial for protecting their physical self. They relied on family, friends and health professionals - not just for interpretations of the scientific evidence, but also for teasing out what aspects of the evidence were most relevant to them. Women also sought out other women who had undergone RRO, RRM, and breast reconstruction to learn about their experiences, in efforts to determine the best way to protect their self-identities as women while at the same time protect themselves from cancer. Discussions centered on the physical and psychological effects of risk-reducing surgery on feelings of youth, beauty, sexuality, body image and femininity.

Engagement with others also reflected women's attention to their emotional well being and relationships. In order to ensure significant others were prepared to provide support and help make decisions, some women brought them into their decision making by having them come to appointments, and discuss with them the implications of surgery. When involving others as sounding boards, the women "checked-in" as they talked through the complexities underpinning their decisions. Other women communicated only the most necessary details to the people in their lives who were "overwhelmed," as a means of keeping them in the loop while simultaneously protecting them from fear, anxiety and worry. Engaging with others came with risks for some women, particularly those who "shocked" others by seriously considering RR surgery, as exemplified by Angela:

But she [family member] actually thought that it [RRM] was a stupid decision and I had a few reactions like that, "Oh that's stupid, you're being stupid, why would you do that?" I've had to defend myself by saying, 'Well why not [have RRM]? What are you keeping exactly [by not having RRM]?' You're keeping risky material [breast tissue].

Because of the unpleasant reactions of others, some women avoided contact with unsupportive individuals and limited the information they shared.

### ***Looking inwards***

Looking inwards was a decision-making approach that involved reflecting on and clarifying how HBOC RR decisions would influence their self-identities as women. The women reflected on the multiple functions and meanings of their breasts and ovaries, as evident in Lauren's description:

It's [breasts] part of my body. It fed my daughter, it's part of me. I have a lot of fears and concerns with having the surgery. I know most of them are just silly or in my head. I think the biggest obstacle would be myself. Feeling that I'm not a woman, I'm not attractive. Will my husband still want me?

The women questioned their ability to adapt to the functional consequences of RRM, including not being able to breastfeed and loss of breast sensitivity and related pleasure, and the surgically induced menopause and infertility associated with RRO. In looking inwards, women realized their breasts and ovaries held very personal meanings, the loss of which was envisioned by some as disfigurement, getting old early, and becoming a man, an "old hag or wicked witch." However, some women were embarrassed about attaching significant meaning to their breasts or ovaries, questioning whether this was vain or petty in light of the threat of cancer. In contrast, other women concluded their breasts or ovaries were trivial to their womanhood. When offered a "breast lift" while consulting a surgeon about RRM, Maureen responded that, "my self-esteem is not wrapped up with how perky my breasts are."

### ***Paying attention to emotions***

The decision-making approach of paying attention to emotions was a principal means by which the women protected their emotional well being. Emotional reactions to genetic test results and medical guidance occurred automatically, ranged from indifference to distress, and subsequently guided information processing and decision making. The women considered the effects of these emotional responses on their current and future lives. For example, Jane, a retired woman, explained:

Seeing what my sister went through and not wanting to do that. I mean she was very good about the whole thing but I've had two years of worry with her and I just can't go there. I've just got to get this [RRM] done.

When emotional responses were overwhelming, some women disengaged from the decision-making process until they could reflect more clearly on their emotional responses. For other women, worry and distress were cues to immediately engage in decision making as a means of keeping these emotions in check.

### ***Relying on intuition***

Intuition was an automatic, visceral, instinctual, gut reaction that many of the women attended to as they considered RR strategies. Although similar to paying attention to emotions in that cognitive reasoning was not overtly employed, relying on intuition involved what women described as an automatic knowing, understanding or believing. When faced with RR decisions, the women honoured their intuitive beliefs about whether or not they would develop HBOC and the best course of action to protect them physically from cancer. Relying on intuition also served the women's emotional self because their intuition reinforced the women's beliefs in their own abilities to cope with emotional threats.

### ***Making sense of the numbers***

This approach involved efforts to interpret the probabilities, percentages and statistics provided by health professionals. This took considerable time and energy as women went over and over the information and sought additional information to help them understand what the numbers meant. The women tried to integrate the numerical information they were given into their pre-existing perceptions of HBOC risk in a way that made sense, provided a sound basis for decision making, or gave them reassurance that the decisions they already made were the right ones. Rose, a younger woman aware of her genetic test results for over five years, exemplified this:

When I do look at ovarian cancer in the family tree it seems like it was all women who were in their seventies, eighties. So I don't see any statistics that make me think that my 20% is going to happen before that. So just believing in the fact that I'm doing what I can to stay healthy and that statistically

it could happen earlier but my family evidence is that it shouldn't happen so early... So things like that make me feel comfortable with not going in for surgery.

### ***Weighing pros and cons***

Weighing pros and cons was a decision-making strategy whereby the women attempted to fully comprehend the impact of decisions on their sense of self. The women were inclined to try to “balance the whole thing out,” weighing the advantages and disadvantages of a decision on all of the dimensions of the self. As Jane explained,

I definitely weigh pros and cons. And with the ovarian thing, there was nothing con. Even the risks of the surgery were minor compared to the risks involved with cancer. There was no way I was going to keep those things [ovaries] in my body.

Some women performed a cursory inventory of those dimensions of self they perceived to be most threatened, while for others, the process extended over years, wherein the women continually incorporated new pros and cons into the equation, as they became known. These women experienced a constant internal dialogue as they went back and forth over various options and consequences. For some, it seemed as if they were going in circles, constantly losing the balance they thought they had achieved as they reviewed options.

### ***Taking time***

Taking time involved women slowing the decision-making process down to give them time to establish what and when decisions would be made, and ensure their sense of self was not unduly threatened by the decisions they faced. The women were confronted with making decisions about HBOC RR strategies while they were healthy and in the context of their busy lives. For some, this was “just not the right time,” or unexpected, as evident in the quote by Scarlet:

Then I phoned Dr. [name], and I said maybe I should go and at least get in the wait list for this breast stuff. So she did a referral, and low and behold, within a week I get this phone call that I can go and

see this plastic surgeon. And I thought, “What?” I thought it was going to be a 2-year waitlist, which would give me enough time.

By taking time, the women fit these serious decisions into personal timelines that acknowledged their responsibilities to family members. Other women needed time to employ additional decision-making approaches that served their self-identities, emotional well being, or physical health.

### ***HBOC RR Decision-Making Styles***

The seven decision-making approaches described above were combined in five distinct styles of decision making that served to preserve the women’s sense of self (see Table 4.3). These included the snap, intuitive, deliberative, deferred and if-then decision-making styles. Table 4.4 lists the number of women who used the decision-making styles for HBOC screening, RRO and RRM decisions. Of note, a number of the women used more than one decision-making style throughout the process of deciding about each of the RR strategies. The use of these decision-making styles was also influenced by the RR approach under consideration, and the previously described contexts within which these decisions were situated.

Table 4.3 Decision-Making Styles Characterized by Decision-Making Approaches

Decision-Making Styles	Privileged dimensions of preserving the self	Use of Decision-Making Approaches						
		Looking inwards	Paying attention to emotions	Taking time	Relying on intuition	Engaging with others	Making sense of the numbers	Weighing the pros and cons
<b>Snap Decision Making*</b>	The dimension of self that was initially most threatened	X	X			X	X	
<b>Intuitive Decision Making</b>	The dimension of self that was intuitively threatened	X	X		X			
<b>If-then Decision Making</b>	All dimensions of self	X	X	X	X		X	X
<b>Deliberative Decision Making</b>	All dimensions of self	X	X	X		X	X	X
<b>Deferred Decision Making</b>	Self-identity, relationships, and emotional well being	X	X	X				

\* The women used only one of the four possible decision-making approaches when making a snap decision.

Table 4.4 Participants' use of Decision-Making Styles for RR Strategies

Decision-making Style	RR Strategy		
	Screening	RRO	RRM
Snap	18	9	4
Intuitive	3	2	1
If-then	0	0	10
Deliberative	1	12	9
Deferred	2	5	10

### *The snap decision-making style*

The snap decision-making style was the most commonly used by the women to make decisions about RR strategies. This style was used when the women perceived the best option for them as a “no brainer” and was not pondered. These decisions were made with confidence and a sense that women could not be

dissuaded or drawn into considering other options – even when presented with new information. This style typically involved a short time frame, limited information, instant processing and the involvement of few other people because a detailed analysis of options was considered unnecessary and women did not revisit their decisions. For example, Iris, an older woman who previously had breast cancer, shared that upon the offering of RRO, “I thought I’m healthy, I don’t need that. It was just an automatic, no I don’t think so.” As evident in Iris’s quote, the women’s perceived closeness to cancer influenced the use of the snap decision-making style. When cancer was perceived to be “far away”, decisions were automatically dismissed or postponed. In contrast, when cancer was seen as being “very close”, snap decisions were used in response to the urgency these women felt to reduce their HBOC risk. Women who made snap decisions focused on informing their family of their decisions in ways that were sensitive to their family’s needs and well being. The women’s decisions were reinforced when family members were in agreement. But when there were disagreements the women attempted to defend their decisions whilst also maintaining these relationships. Because of the rapid nature of the snap decision-making style, the women relied on only one of the four decision-making approaches that were characteristic of this style and helped preserved the dimensions of self most threatened.

### ***The intuitive decision-making style***

The intuitive decision-making style focused on “gut feelings.” Compared to the snap decision-making style, the women took their time in making intuitive decisions and relied heavily on their knowledge and beliefs. Intuition carried more weight than trying to gather information and advice or understand the risk numbers. This style, used by only three women, helped to preserve the dimensions of self perceived to be most threatened, namely their self-identities as women, their emotional well being, and their physical health. In addition to relying on intuition, the women also used the decision-making approaches of looking inwards and paying attention to emotions. These women held tightly to their beliefs about if or when they would develop HBOC, which were shaped by their perceived proximity to cancer, even in the face of contrary information. This allowed the women to maintain their identities and experience little

emotional upheaval. However, health professionals struggled with this style and questioned the credibility of such decisions, which resulted in them offering women unsolicited information and advice. In these instances, the women made efforts to maintain their relationships with health professionals despite disparate views about HBOC risk reduction.

### ***The if-then decision-making style***

This style of decision making was conditional, where the women worked through possible future decisions under various conditions in order to increase their comfort with their current decisions. The women identified hypothetical situations that would threaten their self-identities as women (e.g., if RRM is recommended and timely reconstruction is unavailable), their relationships (e.g., if family members require care and the women feel an obligation to remain cancer free), emotional well being (e.g., if screening becomes too distressing) or physical health (e.g., if diagnosed with breast or ovarian cancer). This often occurred in the context of women's relationships with others, as Gwen, an older woman working part-time, shared:

If somebody like my husband got sick and needed me to look after him for a long time I wouldn't want to risk getting cancer. And I think maybe if I got grandchildren I would want to see them. So I might consider it [RRM] then to reduce the risks.

Rather than make decisions based on uncertain probabilities of developing HBOC, this was a process of conceiving of situations that would pose significant enough threats to the self that the women would feel compelled to make different decisions than they had previously. The decision-making approaches used in this style were all directed towards anticipated future threats to the self.

### ***The deliberative decision-making style***

The deliberative decision-making style was an iterative technique that occurred over a number of months or years, wherein the women deliberated repeatedly about the RR decisions that confronted them. Owing to the irreversible nature of surgical RR decisions and the profound impact on women's lives,

some women considered the deliberative style the most reasonable because it allowed them to consider all possible facets of the decision. The women made tentative decisions and then contrasted them by imagining different HBOC RR decisions and incorporating old and new information, advice, values and beliefs. Deeply valued were the opinions of close family members who the women anticipated would be affected by these decisions. In the deliberative style, the women attended to all of the threats that RR strategies posed to their self-identities, emotional well being, relationships and physical health. However, this complicated and prolonged the process of making a decision because when the women privileged one aspect of the self, a different aspect of the self was sometimes threatened. Attending to all four dimensions of the self was only possible through the use of numerous decision-making approaches, as exemplified by Coral, a woman with a history of breast cancer:

I did go on the Internet and read, get some literature and books, talk to the doctors, I mean the doctors are great...I think I made the right decision and I think I made that decision based on what I think I know. Do you know what I mean? What I've read, what I think I know about myself, what I think I know about the statistics, what I know about just wanting to forget about things maybe and just say this is it, the last bit, I'm over and done and just carry on.

Moreover, as contexts changed over time (i.e., the women's status related to relationships, childbearing, and employment, and beliefs about their proximity to HBOC), the dimension of self most threatened by HBOC RR decisions also changed. The hallmark of this style was the continual weighing and incorporating of pros and cons, associated with the four dimensions of self, which required women to take time with their decisions.

### ***The deferred decision-making style***

This style involved intentionally "putting off" HBOC RR decisions until a later, unspecified, and "right" time. The women used this style without hesitation, judging RR decisions to be premature and suitably pushed into the future, although not put off indefinitely. Angela, who considered cancer to be her "genetic destiny," described her reactions to being offered RRO:

Gee, that's really scary, let's just talk about that [RRO] when it becomes more of an issue. I have a lot of time still and everything will change within that time and it's just something for me to process within this period.

Putting RR decisions on the “backburner” was perceived as the best technique because the women saw no advantages of making decisions right now and by waiting, options might change, become available or more accessible. This style enabled them to carry on with their busy lives, attend to more pressing matters, cope with overwhelming emotions, or continue to reflect on the decisions they faced over extended time periods. Concentrating on familial responsibilities, including providing care to children and ailing parents, was considered more of a priority for the time being and was reinforced by the perception that HBOC was “far away.” Women who used the deferred decision-making style were more concerned about their self-identities, relationships and emotional well being than they were about the immediate physical threat of cancer. Accordingly, the decision-making approaches used in this style included taking time, looking inwards, and paying attention to emotions.

### **Discussion**

The results of this study shed important new light on the process of decision making about HBOC screening, RRM and RRO. A theoretical model is described that focuses on preserving the self. In this model, engaging in decision making is conceptualized as a way for women to attend to their physical health and protect themselves from cancer as well as protect their self-identity as a woman, emotional well being, and relationships. As a means of protecting these four aspects of the self, seven different decision-making approaches are used, which were reflected in five distinct decision-making styles. Over time, women use more than one decision-making style when deciding about each of the RR strategies. The findings further indicated that the process of preserving the self is influenced by four main contexts; the characteristics of health services, the nature of HBOC RR decisions, gendered roles, and the women's perceived proximity to cancer.

The overarching aim of the preserving the self theory with regard to protecting women's four main dimensions of the self is supported by self-schema theory. Used by Esplen and colleagues (2009) to

explain the changes experienced by *BRCAl/2* carriers in their perceptions of themselves, self-schemas are mental representations of the self that might shift over time based on a woman's conception of who she is in the present, as well as memories of who she was in the past, plus visions of who she might be in the future. These future oriented ideas of the self, referred to as 'possible selves,' are versions of the self one expects (e.g., healthy), fears (e.g., a victim of cancer) or wishes to be (e.g., a mother) (Markus & Nurius, 1986). The women in this study appeared to be experiencing challenges to their current self-schema while making decisions about HBOC RR strategies. Furthermore, some women struggled to hold on to their current self-schema while imagining a future self-schema.

Two noteworthy aspects of the women's self-schema that were threatened by HBOC RR decisions centered on womanhood and relationships with family. Although threats to the 'self as a woman' and the 'self in relation to others' schemas are evident in research investigating genetic testing decisions (Etchegary et al., 2009; Hallowell, 1998), this study is the first to describe how these threats influence decisions about RR strategies. Many of the women in this study felt their 'self as a woman' schema was threatened because RR surgery had the potential to disrupt the function and meaning they attached to their breast and ovaries. The women's 'self in relation to others' schema encompassed the women's relational networks, which included their roles and responsibilities to others, as mothers, wives, partners, sisters, and daughters. This self-schema was also threatened because RR strategies were envisioned as potentially disruptive to the women's relational networks by interfering with their abilities to fulfill their roles and responsibilities to others. When considering RR strategies, the women reflected on their willingness, readiness or ability to face these challenges now and in the future. At times, the women resisted shifts to their self-schema by engaging in decision making that reinforced their current perceptions of themselves (e.g., a woman with two breasts). Yet, at other times, the decision-making process facilitated becoming a future self (e.g., a woman protecting her children by reducing her risk through surgery). In other words, accommodating shifts in self-schema, particularly related to feelings of womanhood and relationships with others, was an integral part of the decision-making process.

The preserving the self theory complements health behaviour theories that acknowledge the importance of context for health behaviour, including the preventive health model (Myers, 2005), the social cognitive theory (Bandura, 1986), and the theory of stress, appraisal and coping (Lazarus & Folkman, 1984). However, the theory of preserving the self extends these explanations by describing the social and personal contexts salient to decision making about HBOC RR. This theory further illustrates how women actively interpret and engage with these personal, social and structural contexts and the inherent relational networks, which in turn, raises some important questions worthy of research. For example, understanding the specific decision-making needs of women living in remote communities related to HBOC RR strategies or the role of altruism in motivating women's RR decisions may provide important insights that will extend the preserving the self theory and inform future decision support interventions.

Study findings suggest that the decision-making process was complicated for some women because of the nature of consultations with health professionals who took a nondirective approach to offering RR options, a philosophy of care common in genetic counseling (Sharpe & Carter, 2006). Of note, some researchers consider this nondirective approach as being inadequate, and have advocated for shared decision making wherein health care providers make recommendations while the patient retains a crucial role in making the final decision (Elwyn, Gray, & Clarke, 2000; Smets, van Zwieten, & Michie, 2007). Shared decision making can complement a nondirective approach by providing guidance to health care professionals as to how to engage patients in determining or negotiating the degree of desired directiveness (Elwyn et al., 2000). Shared decision making has been strongly supported in cancer care (Stacey, Samant, & Bennett, 2008) and deserves further investigation as a model for decision making about HBOC risk management.

The theory of preserving the self also highlights how women make decisions in the context of their gendered roles within their families. Although many family members provided practical, informational, and emotional support to women in making their decisions, other family members complicated the decision-making process by negatively reacting to women's RR decisions. Similar to other research

involving *BRCA1/2* carriers (Etchegary et al., 2009; Hallowell, 1998), some of the women in this study also attempted to balance the needs of others with their own needs, and occasionally, the needs of others constrained their decision making. These findings are consistent with theories, such as the Preventive Health Model (Myers, 2005), which considers the social support and influence of significant others to be central to decision making about cancer prevention and control behaviours. These findings are also congruent with the theory of Stress, Appraisal and Coping (Lazarus & Folkman, 1984) wherein positive and negative support is considered to be the result of social systems that create both resources and demands on individuals. The preserving the self theory extends these theories by illustrating how the family context created resources and demands for using particular RR decision-making approaches and ultimately shaped the nature of the HBOC RR decision-making process. For the women in this study, these resources and demands were in part the product of gendered roles and responsibilities that the women felt compelled to fulfill throughout the decision-making process. This resulted in the women listening politely to unsolicited advice as a means of maintaining existing family dynamics, and communicating information and making RR decisions in a manner sensitive to the needs of family members. This is consistent with the ideas of Hartrick Doane and Varcoe (2005), that “the way that gender roles and inequities are played out in families are continuous with the wider fabric of society. So, gendered family roles and experiences are not merely ‘choices’ made by individuals, but rather are enactments of wider social values, expectations, and ideologies” (p. 67). Further, through the illustration of how the family context created resources and demands for using particular RR decision-making approaches and ultimately shaped the nature of the HBOC RR decision-making process, this study enriches the concept of relational autonomy by furnishing fine grained details about how it operates in practice. Future research examining how diverse social and cultural contexts shape gendered family roles and how these, in turn, influence women’s HBOC RR decisions will be an important next step. Such research could also contribute significantly to ongoing empirical and theoretical work exploring how ethical concepts such as autonomy and consent are operationalized in cancer care.

The theory of preserving the self included both cognitive and emotional approaches of decision making that were heavily influenced by the women's perceived proximity to cancer, which appears to be akin to the concept of perceived risk. In other research, a woman's perceived cancer risk has been well established as a factor influencing the uptake of, preference for, or intention to seek RRM and RRO (Antill et al., Claes et al., 2005; 2006; Fang et al., 2003; Fry et al., 2001; Hallowell, 1998; Hallowell, 2000; Hallowell et al., 2001; Meiser et al., 1999; Stefanek et al., 1995; Tiller et al., 2005; Unic et al., 2000; van Dijk et al., 2003; van Dijk, van Roosmalen, Otten, & Stalmeier, 2008).

Specific cognitive approaches, such as making sense of the numbers and weighing the pros and cons, as well the central role of emotions in decision making and information processing has been supported by existing theories (Balneaves & Long, 1999; Lazarus & Folkman, 1984; Leventhal, Brissette, & Leventhal, 2003; Miller, Shoda, & Hurley, 1996; Myers, 2005; Reyna, 2008). For example, in the Cognitive-Social Health Information Processing model (Miller et al., 1996) women's decisions about RRO are determined by how they cognitively and emotionally process information about their cancer risk. For many of the women in this study, distress and worry were long standing emotions because of personal experiences of a cancer diagnosis and treatments, having witnessed family members with cancer, or caring for affected relatives. As in other research (d'Agincourt-Canning, 2005), this experiential knowledge, both empathetic and embodied, ultimately shaped how decisions were made. For example, the women used the decision-making approaches of paying attention to the emotions that accompanied being at risk for HBOC and taking the time needed to protect one's emotional well being. The women's emotional well being also influenced the women's sense of urgency to make decisions, and even functioned as a motivator to both engage and disengage from decision making. It must be noted that RR decision making was so overwhelming for some that their decisions were predominantly directed towards coping with these emotions more so than preventing or detecting cancer.

The cognitive and emotional processes used in the theory of preserving the self often functioned interactively, in parallel, and to varying degrees in the different decision-making styles. These findings lend support to the dual-process theories of information processing, wherein decisions involve the

interaction of two overlapping systems of reasoning (Gilovich, Griffin, & Kahneman, 2002). One is deliberative and analytical, while the other is intuitive, automatic, non-reflective, associative, and experiential. The experiential system involves the use of heuristics; logical shortcuts that people use when processing information (Kahneman & Tversky, 1982). One main heuristic used in the experiential system is the affect heuristic wherein positive or negative feelings are experienced as conscious or unconscious (Slovic, Peters, Finucane, & MacGregor, 2005). Although subsumed under the experiential category, in the theory of preserving the self, the decision-making approaches of paying attention to emotions and relying on intuition were distinct. Relying on intuition, one's gut reaction, has also been used by women seeking genetic testing (D'Agincourt-Canning, 2006) and by individuals making other medical decisions (Lam, Fielding, Chan, Chow, & Or, 2005; Reyna, 2008). The distinction between emotion and intuition in the preserving the self theory might be reflections of different forms or expressions of the affect heuristic. Alternatively, intuitive decision making might be informed by emotions but also incorporate an automatic knowing, understanding, or believing based on previous experiences, values, or beliefs. Analyses of heuristic reasoning has shed light on patient delays in seeking evaluation of breast cancer symptoms (Facione & Facione, 2006), as well as women's perceived risk for breast cancer (Facione, 2002; Katapodi, Facione, Humphreys & Dodd, 2005) and could guide a fine grained analysis of decision making about HBOC RR that would complement the theory of preserving the self.

### **Study Limitations**

Although this study offers the first theory describing *BRCA1/2* carrier women's decision-making processes about HBOC RR strategies, several limitations must be noted. First, the participants were all recruited through one provincial Canadian cancer program and the experiences of women likely differ nationally and internationally because of variations in the provision of genetic services. There is much variability in the recommendations and options made available to *BRCA1/2* carriers and international differences in the rates of RRM uptake have been noted (Metcalf, Lubinski, Ghadirian, Lynch, Kim-Sing, Friedman, et al., 2008). Moreover, we did not access any participants with positive *BRCA1/2* results who chose not to partake in HBOC screening. It is possible that the participants represent a homogeneous

group with ready access to services and beliefs in conventional cancer prevention and early detection. Because we restricted the sample to women who were not undergoing diagnostic testing or treatment for breast cancer, it precludes the application of the theory to women with cancer and faced with cancer treatment decisions and risk-reducing contralateral mastectomy. Furthermore, although insights from this study might spark ideas for how individuals make health decisions following other types of genetic tests, such as HNPCC genetic testing for colon cancer, further research is required to understand these specific contexts.

### **Study Implications**

The theory of preserving the self provides direction for the provision of HBOC RR decision support and care. Foremost, because women's decisions go beyond their physical health, which is generally the focus of health professionals, decision support should include attention to a woman's self-identity, her emotional well being, and her relationships with others. For example, strategies to help women reflect on their feelings about the effects of RR options on femininity, body image and sexuality at a time that is right for them, and in a manner that takes in to account the potential emotional consequences may be helpful. As well, based on the findings, educational, information-based approaches are less likely to be successful than those incorporating psychosocial interventions aimed at emotional coping and adjustment. Making available different avenues by which women can obtain information and social support, such as online or in-person support groups or networking forums, might be valuable to women who learn from the first hand experiences of others. Finally, incorporating a relational framework that focuses on women's relational networks along with their roles and responsibilities would be suitable considering the complexities of HBOC RR decision making. Future research might examine the benefits of these approaches in supporting HBOC RR decision making.

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## CHAPTER 5: Conclusion

### Overview

Individuals are faced with difficult decisions as new genetic knowledge develops and strategies to reduce the risk of disease become available. As such, attention and effort must be directed towards developing interventions to support these individuals in coping with the complexities of these decisions that enhance not only physical, but also psychological well-being. The discovery of the cancer susceptibility genes, *BRCA1* and *BRCA2*, and the subsequent development of technology to test whether individuals carry mutations in these genes, has provided a means of translating these technologies into improvements in health, including the use of hereditary breast and ovarian cancer (HBOC) risk-reducing (RR) strategies. Although efforts have been made to develop decision support resources for women considering options to reduce their chances of developing HBOC, these have not been informed by a good understanding of women's perspectives on the importance of making timely decisions about RR strategies or the factors that influence the timing of their decisions. As well, the complex process of how women make decisions about RR strategies has not been described. If the goal of offering genetic counseling and testing is to improve health and well-being, then making recommendations and offering medical options regarding hereditary breast cancer screening and RR surgeries is but a starting point. Knowledge of women's decision-making processes and the driving forces behind decisions made in the context of HBOC is needed to guide the provision of decision-making support.

The overall aim of this research was to investigate women's decision making related to cancer RR strategies in the context of HBOC. Survey data resulted in descriptions of the HBOC RR strategies that high-risk women used prior to receiving their genetic test results, the predictors of the uptake of these RR strategies, and participants' information needs regarding HBOC risk reduction (Chapter 2). Interview data revealed how women who carry *BRCA1/2* mutations construct the 'right time' to consider decisions about RR surgeries (Chapter 3), and grounded theory research resulted in a theory of how these women make

decisions about RR strategies (Chapter 4). This final chapter integrates the findings from the three studies and discusses these findings in light of current research in the field. Implications this research has for health care professionals, health policy, and theorizing about relational autonomy are delineated, followed by a discussion of the research strengths and limitations. This chapter concludes with recommendations for research.

### **Integrating the Research Findings**

In this section, the most noteworthy findings are highlighted and discussed in relation to current research. The contributions the preserving the self theory makes to decision making and health behaviour theory are outlined first, followed by a discussion of the diverse trajectories of decision making evident in this research. Next, the social and psychological factors found to shape the timing and process of HBOC RR decision making are described. Last, there is a brief summary of how interactions with health care professionals and the health care system influence these decisions.

#### ***Contributions to Decision Making and Health Behaviour Theory***

The most substantial contributions to the field of decision making and health behaviour theory were provided by the preserving the self theory, which include the acknowledgment and description of the decision maker as multidimensional, the influence of social context, and the use of cognitive and emotional approaches to decision making. This theory also provides details specific to decision making regarding HBOC RR strategies that have not been described elsewhere, and that complement more general theories.

In the preserving the self theory, the decision maker is multidimensional, in that four dimensions of the self are considered to be salient to women throughout the process of making decisions about HBOC RR strategies. In this theory, engaging in decision making is conceptualized as a way for women to attend to their physical health and their risk of cancer, as well as protect their self-identity as a woman, emotional well being, and relationships. The preserving the self theory is the first to detail the significant role of each of these dimensions in the process of making HBOC risk management decisions and to bring these dimensions together in a coherent theory. Previous research has focused primarily on women's

perceived risk of cancer and their related emotions (Antill et al., 2006; Lerman et al., 2000; Madalinska et al., 2007; McInerney-Leo, Hadley, Kase, Giambarrresi, Struewing, & Biesecker, 2006), with little attention paid to the profound relational and self-identity implications. Moreover, existing decision making and health behaviour theories that focus predominantly on beliefs, attitudes, and perceptions about disease and disease risk, such as the Health Belief Model (Champion & Sugg Skinner, 2008), the Transtheoretical Model of Behavior Change (Prochaska, 2008; Prochaska, Redding, & Evers, 2008), and the Theory of Reasoned Action and the Theory of Planned Behaviour (Fishbein and Ajzen, 1975; Ajzen & Fishbein, 1980), are a useful place to begin theorizing, but are insufficient on their own because they fail to account for the multidimensional nature of the decision maker and the complexity that is inherent to decisions in the context of HBOC.

The preserving the self theory complements other theories of decision making and health behaviour that have been used to predict or explain cancer prevention or treatment decision making, and that emphasize the importance of environmental and interpersonal factors. Existing theories, such as the Preventive Health Model (Myers, 2005), the Social Cognitive Theory (Bandura, 1986), and the Theory of Stress, Appraisal and Coping (Lazarus & Folkman, 1984), provide broad descriptions of environmental and interpersonal factors that influence health behaviours. The preserving the self theory extends these explanations by describing social and personal contexts most relevant to making decisions about HBOC risk reduction, including the characteristics of health services, the nature of HBOC RR decisions, gendered roles, and the women's perceived proximity to cancer. This theory further illustrates how women actively interpret and engage with these contexts and their inherent relational networks, which create both resources and demands on individuals that function as facilitators and barriers to decision making and influence choices made.

The cognitive and emotional approaches identified in the preserving the self theory are also consistent with existing theories (Anderson, 2007; Balneaves & Long, 1999; Decruyenaere, Evers-Kiebooms, Welkenhuyse, Denayer, & Claes, 2000; Lazarus & Folkman, 1984; Leventhal, Brissette, & Leventhal, 2003; Miller, Fang, Manne, Engstrom, & Daly, 1999; Myers, 2005; Pierce, 1993; Reyna, 2008). Although

numerous theories have acknowledged that affect plays a role in the decision-making process, descriptions of affective approaches remain under-developed and under-emphasized. The preserving the self theory describes the most significant emotions experienced by women faced with making decisions about HBOC RR strategies, as well as how women use emotional and intuitive approaches. These appear to be similar to approaches described by others, such as the affect heuristic (Slovic, Peters, Finucane, & MacGregor, 1995), emotional coping (Lazarus & Folkman, 1984), intuitive decision making (Lam, Fielding, Chan, Chow, & Or, 2005), and gist processing (Reyna, 2008). Of note, these cognitive and emotional approaches were often found to function interactively, in parallel, and to varying degrees. These findings lend support to the dual-process theories of information processing, wherein decisions involve the interaction of two overlapping systems of reasoning (Decruyenaere et al., 2000; Gilovich, Griffin, & Kahneman, 2002). One is deliberative and analytical, while the other is intuitive, automatic, non-reflective, associative, and experiential. In the present research, different combinations of cognitive and emotional approaches resulted in different decision-making styles.

The theory of preserving the self easily lends itself to integration with more general theories and frameworks of decision making and health behaviour. One such theory is the Embedded Decisional Model of Stress and Coping (Balneaves & Long, 1999). The preserving the self theory complements this embedded model by identifying specific contexts and decision-making approaches that function as causal antecedents to the decision-making process, probable mediating processes, and possible immediate and long-term effects of considering decisions about HBOC RR strategies. Moreover, the embedded model classifies decision making as a dynamic process that reflects changes within the person-environment relationship and the preserving the self model describes *how* social and personal contexts shape HBOC RR decision making. A second complementary theory is the Ottawa Decision Support Framework (ODSF), which was designed to guide the development of interventions aimed at preparing individuals and physicians for a structured process of shared decision making when these decisions involve uncertainty and conflict (Légaré et al., 2006). The ODSF guides the assessment of the needs or determinates of a decision, the provision of decision support, and the evaluation of decision making

(O'Connor & Jacobsen, 2007). The preserving the self theory complements the ODSF by identifying specific knowledge, expectations and values relating to the physical risks and benefits associated with different HBOC RR decisions. However, there are also substantial differences, which have implications for the provision of decision support. First, the ODSF was not developed specifically for the context of women's RR decision making in relation to HBOC and the risks and benefits inherent to these decisions, regarding a woman's emotional well-being, her relationships with others, and her self-identity are domains that are absent from the ODSF. Second, the ODSF appears to be based on an individual model of decision making, whereas the preserving the self theory acknowledges the significance of family, friends, health care professionals, and other *BRCA1/2* mutation carriers, to the decision-making process. As such, decision support ought to take into account the relational elements of decision making by assisting women to assess and resolve sources of interpersonal conflict, for example. Third, the ODSF seems to fit most closely with the deliberative decision-making style rather than outline the full range of decision-making styles evident in the preserving the self theory. Clearly, means of supporting women when they use a wide array of decision-making styles need to be explored.

### ***Diverse Trajectories of Decision Making***

Overall, the three studies in this dissertation capture the wide variation in RR decision-making trajectories in the context of HBOC. Some women consider these decisions prior to genetic testing, others consider these decisions shortly after receiving their genetic test results, and still others consider these decisions many years later. As well, the process of decision making is quick and condensed for some women while it is prolonged and dynamic for others. These prolonged and dynamic decisions could be a reflection of how women's thoughts and feelings about their *BRCA1/2* positive status and the appropriate risk management options change over time, evolving experiences within the health care system (Lim, Macluran, Price, Bennett, Butow, and the kConFab Psychosocial Group, 2004), and a lack of systematic efforts to support women's decision making. The trajectory of these decisions also varies according to the RR strategies under contemplation and there is a general order of when decisions are considered; screening first, RR oophorectomy (RRO) second, and RR mastectomy (RRM) last. Survey evidence

indicates that the majority of high-risk women are aware of their heightened risk for breast cancer prior to genetic testing and a large percent undergo screening accordingly. Following genetic testing, most women make snap decisions to partake in HBOC screening, with little variation in the timing of this decision. In contrast, the time to consider decisions about RRM and RRO varies considerably. These findings suggest that the trajectory of decision making is very individual and will shift within the context of women's social roles and responsibilities.

There are different perspectives on when the appropriate time is to consider decisions about RRM and RRO. For the women in this research, it was important to consider decisions in the context of their lives, take enough time to deliberate, cope with their emotions, sort through issues and conflicts, wait for better medical and surgical options, and factor in the readiness of the health care system. These findings provide possible explanations for the time frames between genetic testing and RR surgeries observed in other studies (Antill et al., 2006; Bradbury et al., 2008; Evans et al., 2009; Friebe et al., 2007). Of importance, these participants did not frame their decision making as “delayed,” “slow” or taking “too long”. This is contrary to researchers and clinicians who are of the opinion that RR surgery is the best way to eradicate breast cancer risk in *BRCA1/2* mutation carriers and have voiced their concern that some women are waiting too long to decide about RRM and RRO (Narod, 2009, April). These conflicting perspectives appear to stem from different concerns and priorities associated with HBOC risk management. On the one hand, some women perceive prolonged or deferred decisions about RR surgery as enhancing their quality of life and helping them manage the inherent decisional conflict. On the other hand, there are risks with delaying these decisions that cannot be ignored - some of these women could develop cancer in the interim. Thus, the struggle for women is finding a balance between their needs related to RR surgical decisions and the cancer risk they live with.

There are also tensions that arise from these differing perspectives on the appropriate time to consider RRM and RRO decisions that can magnify women's decisional uncertainty. Although the uncertainty and ambiguity surrounding RR surgery has been documented (Brain, Gravell, France, Fiander, & Gray, 2004; Kenen, Shapiro, Friedman, & Coyne, 2007; McCullum, Bottorff, Kelly, Kieffer, & Balneaves, 2007), this

research extends our understanding of how interactions with others can add to this uncertainty and ambiguity. These differing perspectives can also create conflict between women and family members, as well as health care professionals. Although previous research provides evidence that discussions about HBOC and genetic test results can create family conflict (Kenen, Arden-Jones, & Eeles, 2004a; Kenen, Arden-Jones, & Eeles, 2004b; Matloff, Barnett, & Bober, 2009), this research is the first to describe women's need to manage and resolve conflicts and disagreements before they feel comfortable making RRM and RRO decisions. Both uncertainty and conflicts with others can complicate and prolong the decision-making process.

### ***Family Matters***

Of significance, the findings of this research suggest that women's gendered familial networks shape their decisions about HBOC RR strategies. The women did not construct the self as independent, whereby their choices affect only themselves. Rather, the self was constructed as interdependent (Kenen et al., 2004a; Kenen et al., 2004b) and in relation to others (Hallowell, Jacons, Richards, Mackary, & Gore, 2001). These decisions were perceived by the women in this research to have important implications for family planning and family dynamics. Moreover, family members often influenced the decision-making process. The implications RR decisions have on a woman's ability to find a partner, have children, and fulfill her roles and responsibilities to her family members were of primary consideration. As families changed (e.g., when children were born or family passed away), so did the timing and process of HBOC RR decision making. In other studies, women contemplating *BRCA1/2* mutation testing and RR surgery have expressed similar concerns (Brain et al., 2004; Claes et al., 2005; d'Agincourt-Canning, 2006; Fang et al., 2003; Hallowell et al., 2001; Hamilton, Williams, Bowers, & Calzone, 2009; Hamilton, Williams, Skirton, & Bowers, 2009; Lodder et al., 2002; Matloff et al., 2009; Ray, Loescher, & Brewer, 2005; Schmeler et al., 2006).

Women in previous research also reported that family relationships are affected when they are found to carry a *BRCA1/2* mutation (Douglas et al., 2009; Lim et al., 2004), and that some are met with resistance, shock, hostility, and additional negative responses when attempting to discuss HBOC or RR

surgery with their families (Kenen et al., 2004a; Kenen et al., 2004b; Matloff et al., 2009). The findings of this research indicate that family members provide invaluable practical, informational, and emotional support to women in making their decisions. Yet, other family members complicate the decision-making process by reacting negatively to women's efforts to consider and make decisions about RR strategies. Social expectations about what should be important to women regarding her roles and responsibilities in the family, femininity, and sexuality was privileged by some family members and contributed to their lack of support for women considering surgery options. Wanting to remain sensitive to the concerns, opinions, and needs of family members, the women direct their energies towards maintaining family dynamics or minimizing relationship change (Hamilton, Williams, Skirton, & Bowers, 2009). Akin to other research (Etchegary, Miller, deLaat, Wilson, Carroll, & Cappelli, 2009; Kenen et al., 2004a; Kenen et al., 2004b), some of the women also attempted to balance the needs of others with their own needs, and occasionally, the needs of others constrained their decision making. This research illustrates how the family context creates resources and demands that are, in part, the product of gendered roles and responsibilities, and that women are compelled to fulfill these responsibilities while making their decisions.

### *Psychological Considerations*

This research contributes to our understanding of the role that psychological factors, particularly distress and perceived risk, play in women's decision making regarding HBOC RR strategies. Consistent with previous research (Coyne, Kruus, Racioppo, Calzone, & Armstrong, 2003; Hamilton, Lobel, & Moyer, 2009), at least half of the women surveyed prior to receiving their genetic test results experienced heightened levels of distress (26.6% mild distress and 25.5% moderate to high distress), and perceived themselves to be at moderate (41.3%), or high to very high (30.1%) risk of developing HBOC. However, findings do not provide evidence of relationships between perceived risk and distress, and women's use of RR strategies prior to receipt of genetic test results, with the only exception being the positive association between distress and physical activity. This adds to the already inconsistent evidence about the relationship between perceived risk, distress, and cancer screening among high-risk women (Martin & Lobchuck, 2003).

In contrast, the two qualitative studies provide evidence that women's perceived proximity to cancer and distress play an important role in HBOC RR decision making following notification of positive *BRCA1/2* mutation status. These psychological factors do not predispose women to make particular decisions at particular times, in any particular manner. Instead, they play a much more complex role in the timing and process of women's decision making. For many of the women in this research, feeling "close" to cancer and psychological distress were long standing dimensions of their lives because of personal experiences of a cancer diagnosis and treatments, having witnessed family members with cancer, or caring for affected relatives. As in other research (d'Agincourt-Canning, 2005), this experiential knowledge ultimately shapes when and how decisions are made. The women's emotional well being influenced their sense of urgency to make decisions. Motivated by distress, some women made snap decisions about HBOC RR strategies as a means of coping. Other women prolonged the decision-making process while they coped with their emotions and postponed their decisions until they felt emotionally ready. This is similar to women who previously reported declining genetic testing because of their desire to protect themselves and their family from the emotional consequences (Proulx et al., 2009). The complex role of distress in women's decision making described in this research may reflect women's diverse means of coping with their heightened risk and distress. This provides possible explanations for inconsistent findings among studies that have examined whether or not distress predicts the use of breast and ovarian cancer screening, RRM, and RRO (Claes et al., 2005; Fang et al., 2003; Fry et al., 2001; Howard, Balneaves, & Bottorff, 2009; Hurley, Miller, Costalas, Gillespie, & Daly, 2001; Isaacs, Peshkin, Schwartz, Demarco, Main, & Lerman, 2002; Lerman et al., 2000; Lodder et al., 2002; McInerney-Leo et al., 2006; Meiser, Butow, Barratt, Friedlander, Gattas, Krik, et al., 1999; Meiser, Butow, Friedlander, Schnieden, Gattas, Kirk, et al., 2000; Unic, Verhoef, Stalmeier, & van Daal, 2000; van Dijk, Otten, Zoetewij, Timmermans, van Asperen, Breunin, et al., 2003). Findings of this research also provide additional evidence that the relationship between emotions and decision-making is bi-directional and transactional (Balneaves & Long, 1999; Lazarus & Folkman, 1984), whereby emotions may shift as the process of decision making unfolds and vice versa.

### ***Interacting with Health Care Professionals and the Health Care System***

International differences in the uptake rates of RR strategies have been observed and attributed to health care providers' recommendations and continuity of follow-up (Metcalf et al., 2008). In this research, women made decisions about HBOC RR strategies while interacting with health care professionals and the larger health care system. Prior to receiving their genetic test results, many of the women obtained cancer screening or used other strategies to prevent cancer. These women were clearly in contact with health care professionals regarding breast and ovarian cancer risk management and were able to access services. Once informed that they were *BRCA1/2* mutation carriers, the women in this research were encouraged by health care professionals to undergo regular breast cancer screening and RRO once done childbearing, and RRM was offered as an option. They expressed gratitude for the resource-intensive, specialized genetic services they received, particularly because they perceived the health care system as under strain, underfunded, and characterized by scarcity in resources. The women's perceptions of the 'right time' to consider decisions about RRM and RRO were framed by accessible health care services, as well as the barriers they encountered accessing specific services that they believed would best meet their needs. The extended decision-making process was, in part, a by-product of the lengthy wait times for obtaining referrals as well as surgery and breast reconstruction. The finding that this time lag gave some women needed time to think more about RR surgery suggests that women are still engaged in the decision-making process and possibly preparing for consultations while they wait. Women who wanted to proceed quickly with surgery once they had made their decisions felt frustrated, disappointed, and anxious when the system was not ready for them. Delays at this point may have further threatened the women's sense of self. Delays in accessing these services have rarely been reported in the literature and may be more common in countries, such as Canada, where these services are provided within public health systems. In other countries with different health care systems, other barriers, such as financial costs, may be more relevant.

### Research Strengths and Limitations

The main strengths and limitations of this dissertation research stem from the research design, the guiding theoretical lenses, the research samples, and the methods of data collection and analysis. The use of three studies to investigate women's decision making regarding HBOC risk management allowed for different aspects of the decision-making process to be explored. In particular, these studies facilitated the investigation of the trajectory of decision making over time. The quantitative study focused on the RR strategies used by women prior to receipt of their genetic test results and the qualitative studies explored decision making once women were informed they carried a *BRCA1/2* mutation. An additional strength of this research was the complementary nature of the quantitative and qualitative data, which provided helpful insights for interpreting the findings of each of the studies. For example, the questionnaire data suggests that distress does not predict the uptake of RR strategies prior to women receiving their genetic test results. The qualitative data provides evidence that emotions, such as distress, play a much more complex role in shaping decision making, rather than predisposing women to make particular decisions.

The influence of social context on decision making was captured, in part, because this research was informed by theories of gender and relational autonomy. Although a strength of this research, there are likely additional contexts, such as culture or socioeconomic status, that significantly shaped women's decision making about HBOC risk management, which could be elucidated by conducting research that examines these contexts in greater detail.

The samples for the three studies were restricted to Canadian women who volunteered to participate in research and accessed genetic services offered through the public health care system. Caution must therefore be exercised when determining the relevance of the findings in other settings and contexts. Because we excluded women who were undergoing diagnostic testing for cancer or who were currently undergoing treatment, it also precludes the application of findings to women with cancer who are faced with treatment decisions as well as RR decisions. Research that focuses specifically on how these women make cancer treatment and RR decisions is warranted. In the qualitative research, efforts were made to capture diverse experiences through purposive and theoretical sampling. This was accomplished by

including women of diverse ages and ethnicities, who differed in their marital and childbearing status, and who varied in the number of years they had known their *BRCA1/2* positive status. Yet, women of lower education and income level were underrepresented. Moreover, we did not access any participants with positive *BRCA1/2* results who chose not to partake in the recommended HBOC screening. It is possible that the study participants represent a homogeneous group with ready access to services and beliefs in early detection.

The data collected for this research was self-reported and included both quantitative questionnaire data and qualitative data transcribed from in-depth interviews. In the questionnaire we did not collect information on the women's reasons for using breast and ovarian screening, which may have included ruling out suspicious signs and symptoms of cancer. Moreover, it was unclear what specific information or advice each of the women received from their primary care provider or their genetic counselor and the influence this had on their use of RR strategies. Future research should account for women's reasons for using RR strategies and the messages women receive from health care professionals before they receive their genetic test results. The survey data was also only collected at one time point, prior to women receiving their genetic test results. It would be worthwhile in future research to follow women longitudinally to investigate how women's use of HBOC RR strategies evolves following receipt of their test results and thereafter.

The strength of the research process for both qualitative studies was supported by the systematic approaches to data collection and analysis, facilitated by the use of a software program, and the range of expertise represented within the dissertation committee. The author conducted all the in-depth interviews enhancing the ability to compare and contrast emerging themes and tailor questions in the subsequent interviews accordingly. As a means of ensuring resonance, a criteria contributing to rigor (Charmaz, 2006), preliminary findings were shared with participants in succeeding interviews to obtain their comments on how the preserving the self theory fit with their decision-making experiences. While the theory was not an exact representation of each individual's experience, the participants were able to recognize themselves in the descriptions of decision making implied within the theory. The credibility of

each of the qualitative studies was enhanced through vivid descriptions of the findings, with references to representative quotations and experiences from the data. Insights from this research might spark ideas for when and how individuals make health decisions following other types of genetic tests, such as for hereditary colon cancer, yet further research is required to understand these specific contexts.

### **Implications for Health Care Professionals**

This research provides a window into women's decision making experiences and points to the challenges and complexities of the process. As such the findings reinforce the need for decision support, and offers insights that could inform the provision of decision support regarding HBOC risk management both prior to and following receipt of positive genetic test results. Health care professionals who have opportunities to support women throughout the decision-making process include nurses, social workers, genetic counselors, psychologists, and physicians. In this section, the implications outlined pertain to: (1) discussing cancer screening and health promotion, (2) providing support throughout the decision-making trajectory, (3) addressing uncertainty, (4) focusing on a woman's self-identity, emotional well being and relationships with others, (5) exploring alternate models of supporting women, and (6) questioning expectations about decisions and assumptions about decision making.

The pre-test genetic counseling session presents a timely opportunity for discussions about RR strategies to begin. Prior to receiving their genetic test results, a large number of women surveyed indicated they underwent breast cancer screening. Although this is reassuring, women who have not undergone breast cancer screening ought to be informed by health care professionals of available screening options and decision support provided accordingly. A small number of women in this research were using pharmacoprevention or underwent RR surgery prior to receiving their genetic test results, and qualitative findings indicate that some women who already perceive themselves at high risk for HBOC enter into the RR decision-making process long before they are aware they carry a *BRCA1/2* mutation. This reinforces the common practice of engaging women in discussions about RR strategies at the pre-test genetic counseling appointment as a way of preparing women to make informed decisions when they are ready. It also suggests that some women may be ready to make a decision at that time and health care

professionals need to take the time and provide relevant decision support. In contrast, women who are not ready to discuss HBOC screening or RR surgery may benefit from discussions about additional options available to reduce their risk. This may also be a time to assess for emotional needs that require additional support.

Only a subgroup of women in this study modified their lifestyle in order to decrease their HBOC risk. Yet, findings of this study are in line with the clinical observation that some women are interested in learning about lifestyle choices in order to actively reduce their risk of cancer (Spector, 2007). For younger women who want to wait until after significant life events have occurred to consider RR surgery, it may be beneficial to begin breast cancer screening and health promotion. This lends support for offering health promotion services as a part of genetic counseling (Biesecker, 2001). Alternatively, women could be referred to health professionals or programs focused on these lifestyle behaviours through hereditary cancer services. This appears to be an opportune time wherein some women are interested and motivated to begin health promotion activities. On a cautionary note, almost equal numbers of participants wanted any information or no information about how to reduce their HBOC risk. This suggests that women's information needs are individual, and that decision support at this time should be tailored accordingly. Yet, if women do not want information, then health care professionals may feel conflicted because of their sense of duty to promote health and prevent disease, as well as to provide support in the manner women see most fitting. For those women who do not want information at this time, it is imperative that they be made aware of how to access information and support if and when they are ready.

Considering how uncertainty shapes decisions about HBOC RR and can prolong and complicate the decision-making process, efforts to address this uncertainty are likely to be beneficial. Strategies to address uncertainty may include various means of communicating different types of information, individualizing this information where possible and assisting women to explore their preferences and values associated with the risks and benefits. However, not all uncertainty can be reduced because women are faced with broad ranges of probabilities about their likelihood of developing HBOC and there are

numerous unanswered questions about how and when HBOC might develop. Thus, it is imperative that women are assisted to manage this inherent uncertainty. Providing psychological support and interventions aimed at coping with uncertainty might also be a crucial step in helping women to prepare for the decision-making process.

Based on the qualitative research findings, decision supports are also required that focus on a woman's self-identity, her emotional well being, and her relationships with others. An integral part of the decision-making process was accommodating shifts in self-identity, or self-schema, particularly related to feelings of womanhood. Tools to assess the degree to which a woman's self-identity is threatened by these decisions include the recently developed self-concept scale (Esplen et al., 2009). Strategies to help women reflect on their feelings about the effects of RR options on femininity, body image and sexuality at a time that is right for them, and in a manner that takes in to account the potential emotional consequences, may be helpful. Concordant with others (Brain et al, 2004; McCullum et al., 2007; Schwartz, Peshkin, Tercyak, Taylor, & Valdimarsdottir, 2005), incorporating standard psychological assessments and supports into genetic counseling is warranted to detect and manage psychological distress, to plan psychological support services accordingly, and to help women considering RR surgery come to an informed decision. Women need to be assessed for key psychological issues that could influence their well-being and their decisions. Assessment tools to screen for psychological distress among *BRCA1/2* carriers are currently under development and testing, and include the distress thermometer and problem list (van Dooren et al., 2009), and the Psychosocial Genetic Screening Tool (Esplen et al., 2003). In the meantime, offering psychological consultations to women who are trying to make decisions about RR surgery options may be helpful. In the study by Patenaude and colleagues (2008), all women considering RRM believed psychological consultation would aid their decision making. Tan and colleagues (2009) also reported that 70 out of 73 women accepted an optional psychological consultation prior to RR surgery, and that additional psychological support was given to 31% of participants prior to and 14% after RRM. This indicates a high level of acceptability of psychological consultations. Yet, this may be a controversial recommendation among health care

professionals who are concerned that incorporating routine psychological assessments and consultations into decision support is paternalistic.

This research provides evidence that alternate models of supporting women could complement existing services. Incorporating a relational framework that acknowledges gender influences in RR decisions is warranted considering the complexities and gendered nature of HBOC RR decision making. Such an approach could help women explore how their own relational networks, along with their gendered roles and responsibilities, factor into decision making. Tools to assist women with mobilizing support and resources, communicating with others about HBOC risk management, and working through family conflict may be useful (Etchegary et al., 2009). Family interventions may also be important for some, and specialist staff with expertise in family dynamics may be required for such interventions. Family members might benefit from educational and psychosocial support that involves the provision of information, clarification of misconceptions, exploration of the impact of RR strategies on the family, and the promotion of family coping and adjustment to the decision-making process (Patenaude et al., 2008).

This research generated evidence that health care professionals play an integral role in when and how women make decisions about HBOC RR strategies. Although most of the women expressed gratitude and satisfaction with the care they received, some encountered difficulties associated with the nondirective approach used by health care professionals to provide information and support. The underlying assumption of this approach is that by providing patients with the appropriate information in a nondirective manner, they will be able to reach a decision that reflects their own values and is consistent with their preferences (Schwartz et al., 2005). A shared decision making approach has been strongly supported in cancer care, has recently received attention in the area of genetic counseling (Elwyn, Gray, & Clarke, 2000; Stalmeier, Unic, Verhoef, & Van Daal, 1999; Stacey, Samant, & Bennett, 2008), and deserves further investigation as a model for decision making about HBOC risk management. Shared decision making typically involves at least two participants (the health care professional and the patient), the participation of both parties in the process of decision making, the sharing of information by both parties, and arriving at and agreeing to a decision together (Charles, Gafni, & Whelan, 1997). With this

approach, it is common for health care providers to make recommendations while the patient retains a crucial role in making the final decision (Elwyn et al., 2000; Smets, van Zwieteren, & Michie, 2007). Shared decision making can complement a nondirective approach by providing guidance to health care professionals as to how to engage patients in determining or negotiating the degree of desired directiveness (Elwyn et al., 2000). If shared decision making is integrated into the provision of decision support, it will be important to assess each woman's preferred level of involvement and her desire for family involvement.

The findings of this research direct health care professionals to examine their expectations about women's decisions and assumptions about what is "good" decision making. There ought to be an expectation that not all women will make the same decisions, in the same amount of time, and that some will decide not to have RR surgery and choose other strategies to manage their risk. Women's decisions need to be respected and recognized as the best decision for women at that point in time. If women choose to revisit their decisions at a later time, then decision support must be available when it is wanted and needed. Traditionally, good decision making was considered to be informed, rational, problem-based, consistent with the patient's preferences, and free of emotion and outside influence (Anderson, 2007; Janis & Mann, 1977). In contrast, the decision-making processes for the women in this research were shaped by their personal and social contexts, consisted of cognitive, emotional and intuitive approaches, involved and accounted for others, and took the form of unique decision-making styles. This challenges health care professionals to move beyond the current emphasis on cognitive processing of probabilities, risks, and benefits as the primary focus when delivering decision support. It also highlights the need for holistic, women-centered decision support that takes into account women's lives, and the dynamic and ongoing nature of RR decision making. A women-centered approach addresses barriers to access and respects women's diversity, providing for their health needs in the social and cultural contexts of their experiences (British Columbia Women's Hospital & Health Centre and British Columbia Centre of Excellence for Women's Health, 2004). A women-centred approach also addresses issues beyond medical interventions, placing health in its broad social context.

### **Implications for Health Policy**

The qualitative research findings suggest that the time women take to consider, make, and enact decisions about HBOC RR strategies is influenced by health care system factors, such as wait times to get specialist consultation, delays in getting RR surgeries, and the limited availability of reconstructive breast surgery. As well, the centralization of hereditary services in select urban centers has created regional access inequities such that women living far from services may face significant barriers associated with managing their HBOC risks. These barriers to health services and the lack of timely surgical and reconstructive options must be addressed in order to support women and to take the focus off women as solely responsible for the timing of decisions. Recent health care reform initiatives in Canada have focused on decreasing wait times for specific hospital and medical services, as well as attempting to reduce regional and other inequities in access to timely and appropriate health care (Commission on the Future of Health Care in Canada, 2002; Health Council of Canada, 2008). Although inroads have been made for priority surgeries, clearly more needs to be done at the provincial and federal level to reduce delays in access to HBOC services and RR surgeries. Strategies to eliminate the financial burden of HBOC risk management must also be addressed. Creating partnerships between hereditary cancer services and health care professionals in different geographic locations could curb travel for some women if these health care professionals participated in health promotion, the provision of hereditary breast cancer screening, and decision support. Capitalizing on technology, such as videoconferencing, could enhance these partnerships. Coverage of the financial costs incurred while engaging in HBOC risk management also needs to be closely evaluated. Furthermore, there is a wide gap between the organization and delivery of hereditary cancer services and surgical services. Efforts to promote seamless, coordinated care between these two services at the provincial level would likely be beneficial. Individual health care professionals must also continue to advocate on behalf of women for timely, accessible, and coordinated care. As new evidence about HBOC risk management emerges, appropriate best practice guidelines will need to be developed.

### **Implications for Theorizing: Relational Autonomy**

This qualitative research provides an example of how individuals engage in activities, in this case HBOC risk management decision making, which are constitutive of identity. In accordance with Sherwin's (1998) conceptualization of relational autonomy that draws attention to the development of selfhood as an ongoing process, the women in this research reflected on, and at times re-worked, their self-identities. Yet, when women were not ready for imagined changes or saw them as conflicting with their perceptions of self, they made decisions that reinforced their current self-identities. This research enriches the concept of relational autonomy by furnishing fine-grained details about how it operates in practice and illustrating how personal and social contexts shape the enactment of autonomy throughout the decision-making process. The significance of women's gendered roles within families raises further questions about how diverse social and cultural contexts shape gendered family roles and how these, in turn, influence women's HBOC RR decisions.

### **Looking to the Future: Recommendations for Research**

The findings from this research raise a number of questions worthy of investigation and provide a foundation for future research. Recommendations for future research can be grouped into two main areas: (1) developing interventions to support decision making about cancer RR strategies, and (2) investigating the influence of making HBOC risk management decisions on family members and family dynamics.

Researchers have only recently started to develop and test interventions to support decision making about HBOC RR strategies (Esplen et al., 2004; McCullum et al., 2007; McKinnon, Naud, Ashikaga, Colletti, & Wood, 2007; Metcalfe et al., 2007; Tiller et al., 2006). Both Metcalfe et al., (2007) and Tiller et al., (2006) developed RR surgery decision aids theoretically guided by the ODSF that included educational information and values clarification. Both of these interventions have been shown to increase knowledge levels and decrease decision conflict, but neither improved psychological outcomes. Means of providing psychosocial support in the context of HBOC are currently under development. One intervention that shows promise as a feasible and effective intervention for improving psychosocial functioning among *BRCA1/2* mutation carriers is supportive-expressive group therapy (Esplen et al.,

2004). Led by health professionals and including six to ten women per group, this intervention involves facilitation of mutual support, creation of a sense of normalization through shared experience, encouragement of emotional expressiveness, promotion of family and social support, enhancement of an expanded repertoire of coping skills, vicarious learning through others, detoxification of being at risk, and the integration of new meaning (Esplen et al., 2004). A large randomized trial is warranted at this time to evaluate this intervention. Other interventions that enhance social support, such as online or in-person peer support or networking groups, have provided valuable forums wherein women learn from the first hand experiences of others (Brain et al., 2004; Kenen et al., 2007). In a recent study examining peer support for decisions about RRO and hormone replacement therapy, women sought resources, emotional support, and specific experiential knowledge from each other and generated a unique sense of community and a high level of trust by participating in the Facing Our Risk of Cancer Empowered web-based message board (Kenen et al., 2007). However, support groups may not be the best fit for everyone, as both Di Prospero et al., (2001) and Dorval et al., (2001) report a relatively low level of interest in support group participation by *BRCA1/2* mutation carriers. Research is currently needed that evaluates existing interventions designed to facilitate decision making regarding HBOC RR strategies. Moreover, efforts towards developing novel decision support interventions that provide educational materials, assist with values clarification, address psychological functioning, and assist with mobilizing social support are a crucial next step. The findings of the present research also suggest that developing and testing interventions that take into account women's experiences of decision making, women's needs, and changes that occur over time and across different personal and social contexts would be a useful research direction.

The findings that women's decisions about HBOC RR strategies have important implications for family dynamics and that family members often influence the decision-making process also raise questions worthy of future research. Researchers have begun to investigate the impact of genetic testing information on family members (Hughes et al., 2002; McInerney-Leo et al., 2005; Nycum, Avard, & Knoppers, 2009), yet remarkably few have focused on the impact of risk-management decisions on

individual family members, family communication, or family relationships. Future investigations that incorporate the perspectives of family members and focus on how risk management decisions affect the psychosocial well-being of family members, including husbands, siblings, parents, and children, would be valuable. Hughes et al., (2002) assessed the communication of *BRCA1/2* test results to relatives of women who had undergone testing. Relaying genetic information was the primary reported reason for communicating results, but women also reported wanting to share this information to gain emotional support and advice. Women in the present study also obtained support and advice from family regarding decisions about HBOC RR strategies, yet this complicated their relationships and the decision-making process at times. An important next step will be to explore how women discuss HBOC risk management with different family members or disclose their decisions to family, as well as their intentions and experiences of doing so. This will be important for assisting women to communicate with their families about risk management options and manage the psychological and emotional issues that such communication may bring. Another line of inquiry could be to explore the feasibility and desirability of incorporating family members into a shared decision-making process.

### **Conclusion**

If we are to realize the full benefits of technological advances in science, then as our understanding of biological processes evolves, so too must our understanding of human behaviour related to those advances. In other words, to maximize health outcomes, not only must we personalize health care services based on patients' genetic profiles, but we must also personalize genetic health care services based on patients' psychosocial profiles. Genetic services that bridge the gap between technology and personalized medicine will be those that create partnerships between health care professionals and patients by acknowledging patient perspectives and addressing their priorities. We must also recognize that supporting individuals to make decisions based on genetic information that indicates a predisposition to conditions, like HBOC, is a unique endeavor. Applying theory specific to the context of HBOC to craft and evaluate decision support interventions will likely result in more powerful interventions and improved patient outcomes. Whether concepts and processes central to decisions about HBOC are

relevant to decisions facing individuals at risk for other genetically linked conditions, such as colon cancer, hemochromatosis, or neurological disorders, remains to be seen. Moving beyond the era of ‘new genetics,’ the challenge for health care professionals is to translate genetic technologies into improved health and well-being.

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## Appendix A: Women's Decision Making about Risk-Reducing Strategies in the Context of Hereditary Breast and Ovarian Cancer<sup>5</sup>

### Introduction

Some women are at higher risk than the general population for developing hereditary breast and ovarian cancer (HBOC) based on their family history of cancer and identified through genetic mutation testing. Although categorization of high-risk and probability of carrying a genetic mutation vary according to different models, general characteristics of HBOC include personal or family breast cancer prior to age 40; epithelial ovarian cancer; multiple cases of breast and/or ovarian cancer in the same individual or close blood relatives, either paternal or maternal; close male relatives with breast cancer; a family member with a known mutation in a breast or ovarian cancer susceptibility gene; a clustering of breast cancer with other cancers; or specific ethnicity, such as Ashkenazi Jewish, Icelandic, Swedish, Hungarian, and French-Canadian, associated with higher genetic mutation frequency (National Comprehensive Cancer Network, 2008). HBOCs account for 5% to 10% of all breast and ovarian cancers, and 80% to 90% of those cases of HBOC are caused by mutations in *BRCA1* and *BRCA2* (Ford, Easton, & Stratton, 1998). The discovery of those genes (Ford et al., 1998; Miki, Swensen, Shattuck-Eidens, Futreal, Harshman, Tavtigian, et al., 1994; Wooster, Bignell, Lancaster, Swift, Seal, Mangion, et al., 1995) has led to revolutionary clinical tests for mutations in *BRCA1* and *BRCA2*, carriage of which presents lifetime risks of 45% to 87% for breast cancer and 15% to 40% for ovarian cancer (Antiniou, Pharoah, & Narod, 2003; Easton, Ford, & Bishop, 1995; Ford et al., 1998; Whittemore, 1997).

Various options are available to susceptible women to reduce their risk of HBOC. Strategies include breast cancer screening (clinical breast exam, breast self exam, mammography, MRI, breast ultrasound),

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ovarian cancer screening (transvaginal ultrasound, pelvic ultrasound, manual pelvic examination, blood testing for CA125), risk-reducing mastectomy (RRM) and salpingo-oophorectomy (RRO), and chemoprevention (tamoxifen or raloxifene). Medical recommendations about these risk-reducing strategies vary nationally and internationally (American Cancer Society, 2008; Horsman, Wilson, Avar, Meschino, Kim-Sing, Plante, et al., 2007; National Comprehensive Cancer Network, 2008). For example, semiannual clinical breast exam (CBE) and annual mammogram and breast MRI are now commonly recommended, yet recommendations vary for breast self-exam (BSE) and breast ultrasound (American Cancer Society, 2008; Horsman et al., 2007; National Comprehensive Cancer Network, 2008). RRM is generally not recommended but rather, may be presented as an option and a personal decision to be discussed. Consensus on the recommendation for RRO following childbearing has become more common, but the effectiveness of ovarian cancer screening techniques as well as chemoprevention remains controversial resulting in diverse recommendations and availability. Moreover, access to genetic counseling and testing, as well as the uptake of risk-reducing recommendations vary considerably (Metcalf, Birenbaum-Carmeli, Lubinski, Gronwald, Lynch, Moller, et al., 2008a).

Decisions about these risk-reducing strategies are complex owing to the multiple associated risks and benefits. Breast cancer screening has relatively few side effects and increases the likelihood of detecting cancer early, but it does not prevent cancer. Ovarian cancer screening has not proven to be effective. In contrast, prophylactic surgeries are effective means of preventing cancer among *BRCA1/2* carriers. RRM reduces the risk of breast cancer by approximately 95% (Rebbeck, Friebel, Lynch, Neuhausen, van't Veer, Garber, et al., 2004), and RRO confers an 80% reduction in ovarian/fallopian tube cancer risk and a 50% reduction in breast cancer risk (Rebbeck, Kauff & Domchek, 2009). However, these prophylactic surgeries have the potential to markedly influence a woman's sense of self-identity, quality of life and psychological health (Hallowell, 1998; McCullum, Bottorff, Kelly, Keiffer, & Balneaves, 2007; Patenaude, 2005). In addition, RRO results in infertility and surgically induced menopause, which also raises a woman's risk for other health conditions such as osteoporosis and heart disease (Berchuck, Schildkraut, Marks, & Futreal, 1999; Kritz-Silverstein, Barrett-Connor, & Wingard, 1997). Although

chemoprevention may hold promise, conclusive evidence about the risks and benefits associated with use are unclear at this time (Horsman et al., 2007).

Understanding what influences women's decisions about these risk-reducing strategies is important to health professionals providing decision support. This systematic review synthesizes the current research literature regarding the factors that influence high-risk women's decision making around risk-reducing strategies related to HBOC. Furthermore, we explore how research methodology has shaped the current body of evidence and make recommendations for future research.

### **Methods**

The databases PubMed, CINAHL, and PsychInfo, as well as the search engines Web of Science and Google Scholar, were searched to locate relevant literature published between 1995 and 2008. Keywords entered into these searches included "decision making," "hereditary breast cancer," "hereditary ovarian cancer," "*BRCA1*," "*BRCA2*," "risk-reducing surgery," "risk-reducing mastectomy," "risk-reducing oophorectomy," "prophylactic mastectomy," "prophylactic oophorectomy," "mammography," "mammogram," "clinical breast exam," "breast self exam," "screening," "surveillance," "chemoprevention," "tamoxifen," "prevention," "early detection," and "risk-reduction." To be included in this systematic review, articles had to be written in English, include participants who were women identified as being at high-risk for HBOC based on their family history or genetic testing (with test results that were positive, negative and inconclusive/uninformative), and examine factors that may influence women's decisions about risk-reducing strategies. Uptake or selection of an HBOC risk-reducing strategy presumes decision making, and so, research articles examining factors found to influence women's uptake of such strategies were included in this review. Research assessing intent or hypothetical decision making was also included, as were studies with high-risk women who had completed treatment for a previous breast or ovarian cancer diagnosis considering additional risk reduction. However, studies that included women who were undergoing diagnostic testing or active treatment for breast or ovarian cancer (i.e., surgery, chemotherapy, and radiation) at study enrollment were excluded because decisions regarding risk reduction may be influenced by the knowledge that one has cancer as well as by the process of selecting

and undergoing cancer treatments. Studies that only reported the rate of risk-reducing behaviour uptake, articles describing decisions about whether or not to have genetic testing, and review articles were also excluded.

## **Findings**

Forty-three research studies met the inclusion criteria, including thirty-four quantitative studies (Table A.1) and nine qualitative studies (Table A.2). The most commonly cited factors identified in the studies reviewed were categorized as medical and physical, psychological, and social context factors. Only four studies described factors related to chemoprevention and they are discussed separately. Of note, results of bivariate analyses within the studies were not reported in this review when further multivariate analysis failed to reach significance.

### ***Medical and physical factors***

Medical and physical factors influencing women's decisions about HBOC risk-reducing strategies included *BRCA1/2* status, a previous cancer diagnosis, age, parity and menopausal status.

#### ***BRCA1/2 status***

In studies investigating the influence of *BRCA1/2* status, the decision to undergo RRO was more common than RRM (Phillips, Jenkins, Lindeman, McLachlan, McKinley, Weideman, et al., 2006; Ray, Loescher, & Brewer, 2005; Scheuer, Kauff, Robson, Kelly, Barakat, Satagopan, et al., 2002). With the exception of two studies (Phillips et al., 2006; Uyei, Peterson, Erlichman, Broglio, Schmeler, Lu, et al., 2006) the rates of RRM uptake were too low (0% to 9%) to examine statistical associations with *BRCA1/2* test results (Antill, Reynolds, Young, Kirk, Tucker, Bogtstra, et al., 2006; Botkin, Smith, Croyle, Baty, Wylie, Duston, et al., 2003; Claes, Evers-Kiebooms, Decruyenaere, Denayer, Boogaerts, Philippe, et al., 2005; Lerman, Hughes, Croyle, Main, Durham, Snyder, et al., 2000), possibly due to short follow-up times of 12 to 24 months in some studies. Nevertheless, Phillips and colleagues (2006) found that Australian and New Zealander women who knew their mutation positive status were at least seven times more likely than women who did not know their status to undergo risk-reducing surgery, including RRM. As well, in a review of medical records of 554 American women, Uyei et al. (2006) found positive

*BRCA1/2* status to be associated with risk-reducing surgery, including RRM (OR = 5.96, 95% CI = 2.74, 12.97).

Uptake rates for RRO varied considerably among studies (5.4% to 78%), with the majority of studies finding *BRCA1/2* mutation carrier status to be associated with decisions to undergo RRO (Antill et al., 2006; Botkin et al., 2003; Claes et al., 2005; Fry, Rush, Busby-Earle, & Cull, 2001; Lerman et al., 2000; Phillips et al., 2006; Tiller, Meiser, Gould, Tucker, Dudding, Franklin, et al., 2005; Uyei et al., 2006). However, in a study by Ray and colleagues (2005), the presence of seven women with negative *BRCA1/2* testing results choosing risk-reducing surgery suggests that additional factors influence the decision to undergo surgery. Findings reported by van Dijk and colleagues (2008) do not suggest that disclosure of *BRCA1/2* status greatly influences RRM decision. Women who did not undergo genetic testing, who were awaiting their genetic test results and who decided not to receive test results were also found to undergo RRM and RRO in other studies (Hallowell, 2000; Phillips et al., 2006; Ray et al., 2005; Schmeler, Sun, Bodurka, White, Soliman, Uyie, et al., 2006; Stefanek, Helzlsouer, Wilcox, & Houn, 1995).

Six studies involving *BRCA1/2* mutation carriers demonstrated an increase in uptake of screening, including mammography, clinical breast exam, ovarian ultrasound, and serum CA125 (Botkin et al., 2003; Claes et al., 2005; Lerman et al., 2000; Peshkin, Schwartz, Isaacs, Hughes, Main, & Lerman, 2002; Phillips et al., 2006; van Dijk, Otten, Timmermans, van Asperen, Meijers-Heijboer, Tibben, et al., 2005), and three did not (Botkin et al., 2003; McInerney-Leo, Hadley, Kase, Giambaresi, Struewing, & Biesecker, 2006; Uyei et al., 2006). In a prospective, observational study in the United States (US), *BRCA1/2* carrier status was independently associated with mammography, and within a year of receiving genetic test results, 59% of carriers reported having a mammogram, and 95% a CBE (Peshkin et al., 2002). However, comparing clinical screening of patients with positive *BRCA1/2* mutation testing to those with true negative or uninformative results may be misleading. In at least two studies, the difference in HBOC screening between patients with positive and true negative tests was related to a decrease in inappropriate screening of the negative patients rather than an increase in appropriate screening of positive patients (Lerman et al., 2000; van Dijk et al., 2005). Interestingly, in a comparison of *BRCA1/2*

mutation carriers and non-carriers two years following receipt of test results, no significant difference was found in increased rates of BSE, mammograms or CBE (Botkin et al., 2003). Noteworthy is the number of high-risk women who did not engage in recommended cancer screening. Phillips et al. (2006) found that among women who knew their positive mutation status, 7% reported no mammography, 20% no CBE, 21% no transvaginal ultrasound (TVU), and 75% no serum CA125. Similar results have been identified by Botkin et al. (2003) and Lerman et al. (2000).

In summary, the studies reviewed did not provide conclusive evidence of the influence of *BRCA1/2* mutation status on RRM decisions. Although the studies reviewed provided compelling evidence that positive *BRCA1/2* mutation status is associated with RRO, additional factors appear to be important. Genetic test results may influence decisions about HBOC screening, but further research is required to understand the nature of this relationship. Because *BRCA1* and *BRCA2* mutations confer different patterns of risk for breast cancer and prevalence risks of ovarian cancer, differences in risk-reducing decisions between *BRCA1* and *BRCA2* mutation carriers have begun to be examined (Friebel, Domshek, Neuhausen, Wagner, Evans, Isaacs, et al., 2007; Metcalfe, Foulkes, Kim-Sing, Ainsworth, Rosen, Armel, et al., 2008b). For example, Canadian women with a *BRCA2* mutation were less likely to have a RRO than those with a *BRCA1* mutation (OR = 0.49,  $p = 0.0004$ ) (Metcalfe et al., 2008b). Friebel et al. (2007) found differences between *BRCA1* and *BRCA2* carriers such that parity and ovarian cancer family history predicted RRM among *BRCA1* carriers, while smoking and ovarian cancer family history predicted RRM among *BRCA2* carriers. Moreover, age greater than 40 years and parity predicted RRO among *BRCA1* carriers, but age greater than 40 years and ovarian cancer family history predicted RRO among *BRCA2* carriers.

### ***Previous cancer diagnosis***

Four quantitative studies (Ray et al., 2005; Scheuer et al., 2002; Schmeler et al., 2006; van Dijk, van Roosmalen, Otten, & Stalmeier, 2008) and one qualitative (Frost, Venne, Cunningham, & Gerritsen-McKane, 2004) found a previous cancer diagnosis to be associated with a preference or decision to undergo risk-reducing surgery. In a prospective Dutch study of 304 women at high risk for HBOC, a

personal history of breast cancer was associated with a preference for RRM prior to the disclosure of *BRCA1/2* genetic test results ( $OR = 3.59, p < 0.0001$ ) (van Dijk et al., 2008). In another study of 169 *BRCA1/2* mutation carriers, those who intended to have RRM were more likely to be breast cancer survivors (24% and 12%,  $p = 0.03$ ) (Hoogerbrugge, Kamm, Bult, Landsbergen, Bongers, Brunner, et al., 2008). Selection of RRM has also been found to be predicted by the type of previous surgical procedure ( $\beta = 0.22, p = 0.04$ ), with women who had undergone a mastectomy being more inclined to have RRM of the contralateral breast than women who had breast conserving therapy (van Dijk, Otten, Zoetewij, Timmermans, van Asperen, Breuning, et al., 2003). Similarly, women with a personal history of breast cancer or surgery were more likely to undergo contralateral RRM (Ray et al., 2005), as well as RRO (Scheuer et al., 2002). Other studies, however, did not find a previous cancer diagnosis to significantly influence women's preference for risk-reducing surgery (Botkin et al., 2003; Fry et al., 2001; Scheuer et al., 2002; Tiller et al., 2005; van Dijk et al., 2003; van Dijk et al., 2008).

With regard to the influence that a previous cancer diagnosis has on breast and ovarian cancer screening, evidence was limited to one American study, in which a personal breast or ovarian cancer history was not found to conclusively contribute to mammography, CBE or breast ultrasound adherence (Tinley, Houfek, Watson, Wenzel, Clark, Coughlin, et al., 2004).

Overall, the evidence regarding the influence of a previous cancer diagnosis on women's decisions about HBOC risk-reducing strategies is contradictory. These results may reflect the diverse nature of women's cancer treatments and experiences, and the impact of these experiences on women and subsequent decisions about additional risk-reducing strategies. It is also worth noting that other concurrent medical conditions or circumstances, such as breast fibroadenomas, fibrocystic breasts, endometriosis, ovarian cysts, or dysmenorrhea might be influential. For example, in the study by Stefanek et al. (1995) women who underwent RRM were more likely to have had prior breast biopsies, and in the study by Hallowell (1998), women who decided to have RRO (plus hysterectomy) considered the relief of their pre-existing gynecological problems to be a positive benefit from surgery.

### *Age*

The influence of age was explored in a number of studies. In a small Australian study, seven of eight women who had RRM were over the age of 40 (Antill et al., 2006). In contrast, Scheuer et al. (2002) reported that among a sample of 194 American *BRCA1/2* mutation carriers, the mean age of the 29 women opting for RRM was significantly lower than those who decided not to have the surgery (43.0 v 46.8,  $p = 0.015$ ). A Dutch study including 63 women found mutation carriers opting for RRM to be more often in their 30s, while women opting for surveillance were either 30 years or younger, or older than 43 years (Lodder, Frets, Trijsburg, Meijers-Heijboer, Klijin, Seynaeve, et al., 2002). This curvilinear relationship with age was also apparent among 193 Australian women reporting intention to undergo RRM (Meiser, Butow, Friedlander, Schnieden, Gattas, Kirk, et al., 2000). Given that women's risk of developing HBOC is highest under 50 years of age, with the risk of HBOC decreasing thereafter, this curvilinear relationship may reflect women's and health care professionals' interpretations of this evidence.

Given that RRO is commonly recommended to women between the ages of 35 - 40 years of age, once childbearing is complete, it is not surprising that older age was found to be associated with RRO (Antill et al., 2006; Bradbury, Comfort, Dignam, Cummings, Verp, White, et al., 2008; Claes et al., 2005; Friebe et al., 2007; Meijers-Heijboer, Brekelmans, Menke-Pluymers, Seynaeve, Baalbergen, Burger, et al., 2000; Phillips et al., 2006; Scheuer et al., 2002; Schmeler et al., 2006). Scheuer et al. (2002) found that of the 179 American women with intact ovaries, those electing for RRO were older than those opting for surveillance (mean 47.3 v 41.6 years,  $p < 0.001$ ). In an international study involving 537 *BRCA1/2* carriers, RRO was more common among women 40 years of age or greater compared to women less than 40 years of age (68% v 43%,  $p < 0.001$ ) (Friebe et al., 2007). In qualitative research, older women discussed the impact of age on their decision to undergo RRO, sharing their perception that their ovaries were "equipment," no longer required and best removed (Brain, Gravel, France, Fiander, & Gray, 2004). Although Dutch women who opted for RRO 12 months following the first consultation for ovarian cancer prevention were significantly older than those who chose screening (mean 48.3 v 45.3,  $p = 0.037$ ), older

age failed to predict RRO in multivariate analyses (Madalinska, van Beurden, Bleiker, Valdimarsdottir, Lubsen-Brandsma, Massuger, et al., 2007).

Screening uptake has been shown to be associated with age in five studies (Claes et al., 2005; Lerman et al., 2000; McInerney-Leo et al., 2006; Peshkin et al., 2002; Phillips et al., 2006). However, the direction of this relationship is unclear, with one US study demonstrating a curvilinear relationship between age and mammogram uptake (Isaacs, Peshkin, Schwartz, DeMarco, Main, & Lerman, 2002), and one other study reporting younger non-carrier Belgian women engaged in screening activities not recommended for their age group (Claes et al., 2005). This over-screening may be a reflection of increased breast cancer awareness or psychosocial factors, such as heightened anxiety, related to a family cancer history or family members undergoing genetic testing.

Overall, the evidence regarding the influence of age on the decision to undergo RRM and screening is contradictory and no definite conclusion can be drawn. Further research is needed that examines the personal and social context underlying age as an influencing factor, as well as the impact of age-specific guidelines on women's decisions related to risk-reducing strategies.

### ***Parity***

Five studies investigating the influence of parity on decisions about RRM showed that women with children opted for RRM more often than those without (Friebel et al., 2007; Lodder et al., 2002; Meijers-Heijboer et al., 2003; Unic, Verhoef, Stalmeier, & van Daal, 2000; van Dijk et al., 2008). In a study involving North American and European women, parity significantly predicted RRM among 339 *BRCA1* mutation carriers (OR = 2.7, 95%CI = 1.01, 7.14) (Friebel et al., 2007). van Dijk and colleagues (2008) found that among 304 Dutch women who underwent genetic testing, having children 13 years of age or younger was associated with a preference for RRM (OR = 3.44,  $p < 0.0001$ ). Dutch women who had children have commented that they wanted to live at least until their children grew up and, therefore, chose RRM (Unic et al., 2000).

The results of seven qualitative and quantitative studies from North America and Europe showed that the choice of RRO, with its resultant infertility, was related to a woman's parity (Brain et al., 2004; Claes

et al., 2005; Friebe et al., 2007; Hallowell, Jacobs, Richards, Mackay, & Gore, 2001; Schmeler et al., 2006). Reproductive decisions, including loss of fertility and the desire to have more children, have been indicated by women as being motives for choosing surveillance over RRO (Claes et al., 2005; Hallowell et al., 2001). Contradictory findings, in which parity had no relationship with decisions related to RRO (Fang, Miller, Malick, Babb, Hurley, Engstrom, et al., 2003; Madalinska et al., 2007), may be explained by the fact that the majority of study participants had completed childbearing or a willingness to alter childbearing plans to accommodate surgery needs. To date, no research has been conducted on the influence of parity on women's decisions related to screening behaviour.

Overall, the quantitative and qualitative evidence suggests that women's decisions regarding risk-reducing surgery are indeed influenced by current childbearing status, as well as future family planning. Further research that includes women in their childbearing years and investigates the influence of parity on screening decisions is warranted.

### ***Menopausal status***

Research into the influence of menopausal status on women's decisions regarding HBOC risk-reducing strategies was limited to decisions about RRO. Three quantitative studies did not find menopausal status to influence women's decisions about RRO (Hurley, Miller, Costalas, Gillespie, & Daly, 2001; Madalinska et al., 2007; Tiller et al., 2005). However, among participants in American and British studies, frequently cited concerns about RRO included surgically induced menopause, the need for hormone replacement therapy, perceived acceleration of the ageing process, and psychological side effects of menopause (Claes et al., 2005; Brain et al., 2004; Hallowell, 1998; Hallowell et al., 2001; McCullum et al., 2007; Swisher, Babb, Whelan, Mutch, & Rader, 2001). Yet, in two qualitative studies in the UK, many women were unaware that the removal of their ovaries would result in immediate menopause and that they would require hormone replacement therapy (Hallowell, 2000; Hallowell et al., 2001). Despite these contradictory findings, menopausal status and surgically induced menopause are considered by some women in making decisions about RRO, but women appear to differ considerably in

how they balance the impact of surgically-induced menopause with the reduction in breast and ovarian cancer risk.

In summary, the research exploring the relationships between risk-reducing strategies and genetic testing, previous cancer history, age, parity and menopausal status are the first steps in understanding women's decisions about surgery and participation in screening. How these medical and physical factors influence the decision-making process regarding risk-reducing strategies remains unknown. Clearly, none of these medical and physical factors are independent or act in isolation. Rather, these factors might reflect larger concepts, such as the stages of life women are in or their beliefs about cancer. Further research in this area would help tease apart the relationships among these factors, describe the larger concepts that might be involved, and clarify how women weigh medical and physical factors when making decisions about risk-reducing strategies.

### ***Psychological factors***

Among the articles reviewed, there was evidence of the influence of psychological factors, including perceived risk and cancer related distress, anxiety and worry, on women's decision making regarding RRM, RRO and HBOC screening.

#### ***Perceived cancer risk***

Eight studies suggested that heightened perceived risk of developing HBOC was associated with uptake of, preference for, or intention to seek RRM, despite variations in the measurement of perceived risk (Antill et al., 2006; Fang et al., 2003; Hallowell, 1998; Meiser, Butow, Barratt, Friedlander, Gattas, Kirk, et al., 2000; Stefanek et al., 1995; Unic et al., 2000; van Dijk et al., 2008; van Dijk et al., 2003). In only one study was perceived risk found not to be associated with RRM (Meiser, Butow, Price, Bennett, Berry, Tucker, et al., 2003).

Eight studies also provided evidence that perceived risk influenced decisions about RRO (Claes et al., 2005; Fang et al., 2003; Fry et al., 2001; Hallowell, 1998; Hallowell, 2000; Hallowell et al., 2001; Meiser et al., 2003; Tiller et al., 2005). Quantitative studies have found that perceived risk was positively

associated with consideration of, or intention to undergo, RRO (Fang et al., 2003; Meiser et al., 2003; Tiller et al., 2005) and “reducing the risk of ovarian cancer” was the most important decision-making factor regarding RRO (Fry et al., 2001). However, Hurley et al. (2001) suggested that perceived risk might be influential only in current treatment decision making and not in future intentions related to surgery. Contradictory findings have also been found in qualitative research, with one study reporting that British women who believed they would develop ovarian cancer chose both surgery and screening (Hallowell et al., 2001). Only one Dutch study, which included 160 *BRCA1/2* mutation carriers, did not find higher levels of perceived risk to be predictive of RRO (Madalinska et al., 2007).

The influence of perceived risk on breast and ovarian cancer screening was investigated in four studies, one of which found significant associations (Claes et al., 2005; Isaacs et al., 2002; McInerney-Leo et al., 2006; Tinley et al., 2004). In a retrospective study by Isaacs et al. (2002), American women with elevated perceived cancer risk were not found to be more likely to have CBE, but they were more likely to have obtained a CA125 test (OR = 4.4, 95% CI = 1.5, 13.2) and a transvaginal or pelvic ultrasound (OR = 2.9, 95% CI = 1.4, 6.2).

This body of research provides evidence that perceived risk influences high-risk women when making decisions regarding risk-reducing surgery. How different levels and types of perceived risk influence women throughout the decision-making process remains unexplored.

### ***Cancer-related distress, anxiety and worry***

Cancer-related distress, anxiety and worry were the psychological factors most commonly assessed among the studies reviewed. The majority of researchers found a positive relationship between distress, anxiety (Antill et al., 2006; Lodder et al., 2002; Meiser et al., 2000; Unic et al., 2000), and worry (Stefanek et al., 1995; van Dijk et al., 2003), and actual or hypothetical decisions about RRM. Despite not finding an association between the uptake of RRM and anxiety among 168 Australian women, Antill et al. (2006) did find that women who would consider future RRM were more likely to have higher levels of anxiety.

Fry et al. (2001) found the best predictor of RRO to be the importance women attributed to reducing cancer worry, while women with high breast/ovarian cancer anxiety were more likely to consider RRO in the study by Meiser and colleagues (1999). Similarly, “reduction in anxiety/uncertainty” was found to be the strongest predictor of current interest in RRO, independent of objective risk classification, perceived risk, severity of cancer anxiety, and intrusive ideation (Hurley et al., 2001). In qualitative findings, women have described their ovaries as “time bombs” and RRO represented a strategy for managing the associated worry and anxiety (Brain et al., 2004; Hallowell, 2000). In contrast, three research studies (Claes et al., 2005; Fang et al., 2003; Madalinska et al., 2007) failed to find higher levels of distress to be predictive of the decision to undergo RRO among *BRCA1/2* mutation carriers and high-risk women with a family history of ovarian cancer.

There is limited evidence of an association between cancer-related distress, anxiety and worry and HBOC screening (Isaacs et al., 2002; Lerman et al., 2000; McInerney-Leo et al., 2006). Only ovarian cancer worry has been found to be predictive of the decision to undergo pelvic ultrasound (McInerney-Leo et al., 2006).

Similar to the literature on perceived risk, cancer-related distress, anxiety and worry appear influential in women’s decisions about risk-reducing surgery, although how these psychological factors affect women’s perceptions and cognitions across the decision-making processes remains unexplored. For example, there may be a small sub-group of women who experience such severe cancer-related distress that their ability to attend to treatment decisions is seriously compromised. There is also insufficient evidence on which to base any conclusions regarding the relationship between cancer-related distress, anxiety and worry and HBOC screening. Further research is required that teases out the conceptual differences among cancer specific versus generalized distress, anxiety and worry and the role of these concepts in decisions about HBOC risk reduction. Expansion of this body of research to explore other psychological factors associated with risk-reducing surgery, such as body image, self-identity, self-esteem and feelings of femininity, is also warranted given preliminary findings suggesting these factors play a

role in women's risk-reduction decisions (Claes et al., 2005; Fang et al., 2003; Hallowell, 1998; Kenen, Shapiro, Hantsoo, Friedman & Coyne, 2007; Lodder et al., 2002).

### ***Social context factors***

The social context factors influencing women's decisions about risk-reducing strategies include patterns and personal experiences of cancer in the family, as well as obligations women had to their family.

#### ***Patterns and personal experiences of cancers in the family***

The most common measure of the influence of family on women's decision making about HBOC risk-reducing strategies was the presence and number of first and second-degree relatives affected by breast and/or ovarian cancer. Five studies found the presence and number of relatives with breast or ovarian cancer influenced decisions about risk-reducing surgery (Antill et al., 2006; Bradbury et al., 2008; Friebe et al., 2007; Metcalfe et al., 2008b; van Dijk et al., 2006). For example, in the largest study of 537 North American and European mutation carriers, a family history of breast or ovarian cancer predicted RRM among both *BRCA1* (OR = 0.41, 95%CI = 0.21, 0.78) and *BRCA2* carriers (OR = 2.26, 95%CI = 0.09, 0.84), and RRO among *BRCA2* carriers (OR = 3.61, 95%CI = 1.41, 9.23) (Friebe et al., 2007). However, contradictory evidence has emerged suggesting no significant relationship between family history and risk-reducing surgery, more so with regard to RRO (Antill et al., 2006; Botkin et al., 2003; Fang et al., 2003; Hurley et al., 2001; Meiser et al., 1999; Schmeler et al., 2006; Tiller et al., 2005; Madalinska et al., 2007) than RRM (Meiser et al., 2000; Unic et al., 2000). In addition, one US study suggested that women with three or more relatives with breast cancer were less likely to have RRO than women with less than three relatives with breast cancer (Bradbury et al., 2008).

With regards to mammography, women with three or more relatives affected with breast cancer have been found to be more likely to have had a mammogram in the previous year (OR = 2.6, 95% CI=1.3, 5.1) than women with a less significant family history (Issacs et al., 2002). While it is not clear how significant a family history (i.e. number of first- and second degree relatives with ovarian cancer) is

needed to influence ovarian cancer screening decisions, two studies have found a positive association between family history and uptake of ovarian cancer screening (Issacs et al., 2002; Meiser et al., 1999). For example, Isaacs et al. (2002) found women with one or more relatives with ovarian cancer more likely to have obtained a CA125 test (OR = 12.3, 95% CI 4.6, 33.0) and a transvaginal or pelvic ultrasound (OR = 4.9, 95% CI = 2.4, 10.1).

Understanding how family history influences women's decisions about HBOC risk-reducing strategies can be drawn from two qualitative studies (Hallowell et al., 2001; McCullum et al., 2007). In one study of 23 women who had RRO and 26 women who chose surveillance, the influence of nursing a dying relative and experiencing the death of a relative were the most influential factors in their decision (Hallowell et al., 2001). In McCullum et al.'s (2007) study, Canadian women heavily relied upon the HBOC screening and risk-reducing surgical experiences of their siblings in making their own decisions about HBOC risk-reducing strategies.

While the evidence regarding the relationship between family history and the decision to undergo RRM is quite strong, there is less certainty regarding how family history impacts women's decisions about RRO and ovarian cancer screening. In addition, understanding the meaning of witnessing relatives who live through, or possibly die, from breast or ovarian cancer and how it influences women's HBOC risk-reducing decisions is important to better support women throughout the decision-making process.

### ***Family obligations***

A limited number of studies investigated the influence of family obligations on women's risk-reducing decisions. All of the 41 women in the study by Hallowell (1998) discussed their obligations to support and care for family, by establishing their cancer risk and taking action to address this risk. For some, prophylactic surgery provided the means by which they could guarantee their survival and enable them to fulfill their obligation of care. Women in this study, as well as some of the 49 women in the study by Hallowell et al. (2001), justified their desire to undergo RRO in order to prevent their family from seeing them suffer or becoming caregivers in the event of a cancer diagnosis. The seven women who decided not to undergo surgery mentioned the long period of convalescence following surgery and the

effect that it would have on their ability to provide practical and economic support for their family (Hallowell et al., 2001). In a qualitative study by Brain et al. (2004), the greatest perceived obstacle to RRO was time off work, and several women were concerned about being able to care for family members. When discussing RRM, several women in the qualitative study by Kenen et al. (2007) expressed their desire to protect their families from the stigmatizing effects of RRM. Women were also concerned for their children's future, particularly their daughters, and hoped that participating in a screening program would benefit family members.

In summary, the literature describing the family context factors that influence women's uptake of HBOC risk-reducing strategies is in its infancy. Although patterns and experiences of cancer in a woman's life and family obligations may be important, future descriptive research is required.

### ***Pharmacoprevention***

Four studies investigated high-risk women's decisions about tamoxifen use as a HBOC risk-reducing strategy (Altschuer & Somkin, 2005; Bober, Hoke, Duda, Regan, & Tung, 2004; Meiser, Butow, Price, Bennett, Berry, Tucker, et al., 2003; Metcalfe, Snyder, Seidel, Hanna, & Lynch, 2005). Reasons Canadian women provided for not taking tamoxifen included worry about side effects, receiving no recommendation, previous RRM and/or RRO, hormone replacement therapy, pregnancy planning, doubt about tamoxifen's efficacy, aversion to pills, and limited drug coverage (Metcalfe et al., 2005). Bober et al. (2004) found women with a physician's recommendation, a history of abnormal breast biopsy, heightened cancer-related anxiety and breast cancer worry, and greater perceived risk more likely to opt for tamoxifen use. Meiser and colleagues (2003) also found perceived risk predictive of intention to take tamoxifen. Altschuler and Somkin (2005) suggested that women were less likely to use tamoxifen if they had concerns about co-morbidities, such as heart disease, and if they had passed the age at which a first-degree relative developed breast cancer.

Overall, the factors found to influence women's use of risk-reducing tamoxifen were similar to those found to influence women's decisions regarding other risk-reducing strategies, with the exceptions of side

effects associated with its use and physicians' recommendations. Additional research investigating decision making about tamoxifen use is warranted at this time given its recent approval in numerous countries as a preventive therapy for breast cancer in high-risk women.

### **Methodological Influences**

Although the literature reviewed provides some insights into high-risk women's decisions about HBOC risk-reducing strategies, these need to be considered in relation to the methodological strengths and limitations of this body of research. Of the forty-three studies reviewed, only two teams of researchers (Kenen et al., 2007; Ray et al., 2005) used a conceptual framework to guide their research. The application of decision-making theory, or other theories of health behaviour, to this growing field of research would advance theory, as well as allow new conceptual understandings to be developed, and provide an evidence-based framework from which to explore women's decisions about risk reducing strategies. In addition, research focused on generating unique decision-making theories specific to this context may prove to be a fruitful area of development.

The geographical location of studies included in this review and the sampling strategies employed also have implications for how the findings are interpreted. The studies took place in a number of countries and the international differences in health care systems and availability of genetic services may have influenced results. For example, women's decisions about screening or risk-reducing surgery may be influenced by the costs of procedures in different countries as well as differences in health insurance coverage. In addition, the majority of study participants were recruited through established hereditary cancer programs. Women who are enrolled in these programs may represent a subset of women who have ready access to risk-reducing options and their decisions regarding risk-reducing strategies may be largely influenced by the recommendations provided by health professionals within these programs. As well, these women may represent a motivated group because they have already sought out hereditary cancer services. Although the international scope of this research is a strength, direct comparisons are difficult to make.

Also influencing the findings was the length of time allocated for outcome follow-up. In a number of studies, follow-up occurred within 12 months of genetic counseling or post disclosure of *BRCA1/2* genetic test results (Bober et al., 2004; Brain et al., 2004; Claes et al., 2005; Fang et al., 2003; Lerman et al., 2000; Lodder et al., 2002; McCullum et al., 2007; McInerney-Leo et al., 2006; Peshkin et al., 2002; Ray et al., 2005; van Dijk et al., 2005). This follow-up time is based on the assumption that women make their decisions about HBOC risk-reducing strategies within one year. There is evidence, however, that some risk-reduction decisions take more time and may change over time (McCullum et al., 2007). Longitudinal studies with extended follow-up could provide important insights.

The definitions and measurement of risk-reducing strategies also varied across the studies. For example, screening uptake was defined as adherence to recommendations by hereditary cancer clinics in some studies (Claes et al., 2005; Lerman et al., 2000; Peshkin et al., 2002; Schmeler et al., 2006), yet in others it was defined as the uptake of individual breast and ovarian cancer screening strategies (Botkin et al., 2003; McInerney-Leo et al., 2006; Phillips et al., 2006; Scheuer et al., 2002). Other studies appear to categorize, by default, women who did not undergo risk-reducing surgery as undergoing surveillance (Fry et al., 2001; Lodder et al., 2002; Uyei et al., 2006). It is not clear what types of screening, if any, these women were actually engaged in. Also influencing the interpretation of results were the ten studies wherein hypothetical decision making or future intention to engage in HBOC screening or risk-reducing surgery was the outcome variable (Brain et al., 2004; Fang et al., 2003; Hurley et al., 2001; Meiser et al., 1999; Meiser et al., 2000; Ray et al., 2005; Tiller et al., 2005; Unic et al., 2000; van Dijk et al., 2005; van Dijk et al., 2003). Some evidence suggests that hypothetical measures are in fact predictive of future uptake (Tiller et al., 2005); however, if we are to truly understand how women make decisions regarding HBOC risk-reducing strategies, women's perspectives and experiences of actual decision making should be examined.

### **Conclusion**

This review summarizes evidence regarding the medical and physical, psychological and social context factors that influence women's decisions regarding RRM, RRO, HBOC screening, and

chemoprevention. Evidence garnered from moderate sized studies in several countries suggests that *BRCA1/2* mutation status and parity influences women's decisions about RRO. The psychological factors of perceived cancer risk, and cancer related distress, anxiety and worry also appear to be influential in risk-reducing surgery decisions, as well as the presence and number of relatives affected with cancer. Research findings related to other factors influencing risk reduction decisions are mixed. In addition, evidence regarding decisions about chemoprevention is only just emerging.

Additional factors that may influence decisions that have not been examined include prevailing social and cultural norms and values, and health care system factors. International differences in the uptake of RRO and RRM have been documented, accompanied by suggestions that social and cultural factors may be responsible. Access to health services, costs associated with treatment and screening, and limited health insurance might also be significant. Moreover, the specific recommendations of health professionals regarding risk-reducing strategies and the way this information is communicated to women have historically varied and likely play a significant role in women's decisions. Where the efficacy of various risk-reducing strategies has been demonstrated, clinical recommendations are being established (Horsman et al., 2007). It will be important in future research to understand the impact of such guidelines as well as how different modes of communicating this information influence women's decision-making processes. Furthermore, as new and existing screening methods, surgical techniques, and lifestyle factors are evaluated through research, factors influencing decision making may shift accordingly. Comparative studies to examine the influence of variations in health services and access to these services on risk-reduction decisions are needed, as well as studies examining decisions related to reducing risk through life-style choices, such as smoking cessation, exercise, dietary changes, and the use of complementary therapies (e.g., natural health products). Importantly, broadening future research to investigate additional social context factors, such as community attitudes, social networks, ethnicity, gender roles, relationships with health care providers, and socioeconomic status, is essential to understanding how these contexts shape women's decisions and also for situating women's decisions in the world within which they live. Factors that influence women's decision making about HBOC risk-reducing strategies function in concert

and may change throughout the process of decision making. How factors operate in women's lives over time would best be elucidated through prospective, longitudinal research, as well as qualitative research.

The use of existing theoretical frameworks to guide research or the development of frameworks specific to decision making about HBOC risk-reducing strategies will also advance the field. Particularly relevant are existing theoretical frameworks that take into account not only medical and physical factors, but also psychological factors, examples of which include the Cognitive-Social Health Information Processing model (Miller, Fang, Manne, Engstrom, & Daly, 1999) and the Embedded Decisional Model of Stress and Coping (Balneaves & Long, 1999). Other useful theoretical frameworks include those that incorporate the influence of social contexts within which decisions are made, such as the Preventive Health Model (Myers, 2005), as well as those developed specifically for use in situations involving difficult decisions and decisional conflict, such as the Ottawa Decision Support Framework (O'Connor, 2006). In addition, recent research has highlighted the role that "gut feelings," intuition, and underlying affect play in decisions about genetic testing (D'Agincourt-Canning, 2006) and other medical decisions (Reyna, 2008). Research investigating how intuition and affect influence HBOC risk-reducing decisions would shed light on this complex process.

This research, in conjunction with existing knowledge about influential factors identified in this review, could provide valuable insight and inform the provision of decision-making support. Decisional support may be improved by assessing how medical and physical, psychological and social context factors are influencing women's decision making, discussing the factors most significant to individuals, and helping them to clarify their related values and beliefs. Moreover, the development and tailoring of information and decision aids to women at high-risk for HBOC incorporating evidence of influential factors may result in highly effective and useful tools that will reduce women's decisional conflict and improve satisfaction with the decision-making process.

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Table A.1: Quantitative Research Studies Reviewed

Author and Country	Study Sample	Data Collection Method, and Follow-up time of Measurement	Risk-Reducing Strategy Investigated	Factors Investigated
Antill et al. (2006), Australia	182 women at moderate and high risk for HBOC (21 were <i>BRCA1/2</i> mutation carriers)	Self-report questionnaire and medical record data  Mean 3.73 years following initial risk assessment	Uptake and consideration of future RRM and RRO	<i>Demographics:</i> age, employment status, household income, level of education, marital status, ethnicity and number of offspring <i>Medical history:</i> <i>BRCA1/2</i> mutation status, breast and ovarian cancer, risk assessment, and previous mastectomy or oophorectomy <i>Family history:</i> breast and ovarian cancer <i>Perceived cancer risk</i> <i>Psychological factors:</i> distress
Bober et al. (2004) USA	129 women eligible to take tamoxifen	Self-report questionnaire and telephone interviews  2 and 4 months following counseling	<i>Tamoxifen uptake and decision satisfaction</i>	<i>Demographics:</i> age, employment status, level of education, marital status, and ethnicity <i>Medical history:</i> personal history of cancers and health conditions affecting eligibility to take tamoxifen, CBE history, mammography history, and number of breast biopsies <i>Family history:</i> breast and ovarian cancer <i>Physician recommendation re: tamoxifen</i> <i>Concerns about side effects</i> <i>Perceived cancer risk</i> – breast/ovarian <i>Psychological factors:</i> distress, depression, breast cancer worry and optimism
Botkin et al. (2003) USA	37 <i>BRCA1</i> mutation carriers and 92 non-carriers	Computer assisted telephone interviews  Baseline, 1-2 weeks, 4-6 months, 12, and 24 months post disclosure of <i>BRCA1</i> test results	<i>Breast cancer screening:</i> BSE, CBE, mammography, breast ultrasound <i>Ovarian cancer screening:</i> TVU and CA125 <i>RRM and RRO uptake</i>	<i>Demographics:</i> age, level of education, household income and marital status <i>Medical history:</i> personal cancer history, previous mastectomy or oophorectomy <i>Family history:</i> number of 1 <sup>st</sup> and 2 <sup>nd</sup> degree relatives with a breast and ovarian cancer history <i>Psychological factors:</i> distress
Bradbury et al. (2008) USA	88 <i>BRCA1/2</i> mutation carriers	Retrospective medical records review (Jan 1996 to Dec 2003)	<i>RRO uptake and timing of RRO</i>	<i>Demographics:</i> age, marital status, ethnicity and number of children <i>Medical history:</i> genetic test date, test disclosure date, <i>BRCA1/2</i> mutation status, personal history of breast cancer and mastectomy <i>Family history:</i> number of 1 <sup>st</sup> and 2 <sup>nd</sup> degree relatives with a breast and ovarian cancer history
Claes et al. (2005) Belgium	34 <i>BRCA1/2</i> mutation carriers and 34 non-carriers	Self-report questionnaire and semi-structured interviews  Baseline at pretest counseling session and 12 months post disclosure of <i>BRCA1/2</i> test results	<i>Breast cancer screening:</i> BSE, CBE, mammography (or breast ultrasound) <i>Ovarian cancer screening:</i> Ovarian ultrasound <i>RRM and RRO uptake</i>	<i>Demographics:</i> <i>BRCA1/2</i> test result, age, level of education, marital status and number of children <i>Family history:</i> breast and ovarian cancer <i>Perceived cancer risk, perceived serious of illness and perceived control</i> <i>Psychological:</i> distress
Fang et al. (2003) USA	76 women enrolled in a familial cancer risk assessment program	Self-report questionnaire  Following familial cancer education session and risk counseling	<i>RRO intention in following 12 months</i>	<i>Demographics:</i> age, level of education and marital status <i>Family history:</i> number of 1 <sup>st</sup> and 2 <sup>nd</sup> degree relatives with a breast and ovarian cancer history <i>Perceived cancer risk</i> <i>Perceived benefits and limitations of RRO</i> <i>Psychological factors:</i> distress

Author and Country	Study Sample	Data Collection Method, and Follow-up time of Measurement	Risk-Reducing Strategy Investigated	Factors Investigated
Friebel et al. (2007) North America and Europe	537 <i>BRCA1/2</i> mutation carriers	Self-report questionnaire and medical record review  More than 6 months post disclosure of <i>BRCA1/2</i> test results  Mean follow-up time was 39.6 months (median 36 months)	<i>RRM and RRO uptake</i>	<i>Demographics:</i> age and parity <i>Medical history:</i> <i>BRCA1</i> or 2 status, ever/never use of hormonal birth control and ever/never alcohol use <i>Family history:</i> 1 <sup>st</sup> degree relative with a breast and ovarian cancer history
Fry et al. (2001) UK	30 women who had undergone RRO and 28 women at high risk for HBOC who attended a screening program	Self-report questionnaire  Registered with the familial cancer clinic within the previous 5 years	<i>RRO uptake versus ovarian cancer screening</i>	<i>Demographics:</i> age, level of education and marital status <i>Medical history:</i> time since registration with the familial cancer clinic and time since surgery <i>Psychological factors:</i> reducing cancer worry and worries about effectiveness of ovarian screening, <i>Additional factors:</i> reducing ovarian cancer risk, risks of surgery, need for hormone replacement therapy, symptoms that would be helped by hysterectomy, other family members choosing risk-reducing surgery, partner's attitude, recovery time, desire to have children, fear of menopausal symptoms, need to feel like a woman, dislike of ovarian cancer screening methods, loss of "periods," and inconvenience of attending clinic
Hoogerbrugge et al. (2008) The Netherlands	196 <i>BRCA1/2</i> mutation carriers	Not reported  Median follow-up was 2 years (range 1-9 years)	<i>RRM intention Breast cancer screening intention:</i> CBE, mammography and MRI	<i>Demographics:</i> age, daughters and age of youngest child <i>Medical history:</i> <i>BRCA1</i> or <i>BRCA2</i> status, breast cancer history, age at previous breast cancer diagnosis and menopausal status <i>Family history:</i> maternal inheritance, mother with breast and ovarian cancer history, mother's age at breast and ovarian cancer diagnosis, sister with breast cancer, age of youngest sister with breast cancer and age of youngest relative with breast cancer and ovarian cancer
Hurley et al. (2001) USA	94 women with varying degrees of familial risk for ovarian cancer who had not had <i>BRCA1/2</i> testing	Questionnaire completed during telephone interviews	<i>RRO interest (current and future)</i>	<i>Demographics:</i> age, level of education, marital status, number of children, ethnicity and menopausal status <i>Objective cancer risk</i> <i>Perceived cancer risk</i> <i>Psychological factors:</i> severity of cancer anxiety and stress-related ideation <i>RRO pros and cons</i> <i>RRO Efficacy</i> <i>Desire for childbearing</i>
Isaacs et al. (2002) USA	216 high-risk women in a hereditary cancer program	Telephone interviews  Baseline prior to genetic testing	<i>Breast cancer screening:</i> CBE and mammography <i>Ovarian cancer screening:</i> ultrasound and CA125	<i>Demographics:</i> age, level of education, marital status, income and ethnicity <i>Medical history:</i> <i>BRCA1/2</i> mutation status, breast biopsy history and year of study entry <i>Family history:</i> number of relatives with breast and ovarian cancer <i>Perceived cancer risk</i> <i>Psychological factors:</i> distress and cancer worries

Author and Country	Study Sample	Data Collection Method, and Follow-up time of Measurement	Risk-Reducing Strategy Investigated	Factors Investigated
Lerman et al. (2000) USA	216 women in HBOC families (84 <i>BRCA1/2</i> mutation carriers, 83 non-carriers and 49 <i>BRCA1/2</i> test decliners)	Structured telephone interviews  Baseline, 1, 6 and 12 months post disclosure of <i>BRCA1/2</i> test results	<i>Breast cancer screening</i> : mammography <i>Ovarian cancer screening</i> : TVU and CA125 <i>RRM and RRO uptake</i>	<i>Demographics</i> : age, education level, marital status and health insurance status <i>Medical history</i> : <i>BRCA1/2</i> status, whether subjects participated in previous genetic studies and mode of genetic counseling <i>Psychological factors</i> : distress
Lodder et al. (2002) The Netherlands	63 women who underwent <i>BRCA1/2</i> mutation testing (26 <i>BRCA1/2</i> mutation carriers and 37 non-carriers)	Interview and self-report mailed questionnaire  1-3 weeks, 6 and 12 months post disclosure of <i>BRCA1/2</i> testing results	<i>Breast cancer screening</i> : BSE, CBE, mammogram and MRI <i>Ovarian cancer screening</i> : gynecologic exam, TVU and CA125 <i>RRM uptake</i>	<i>Demographics</i> : age, level of education, marital status and number, gender and age of children <i>Medical history</i> : <i>BRCA1/2</i> carrier status and oophorectomy history <i>Family history</i> : age women confronted with breast and ovarian cancer in relative, aware hereditary nature disease in family, number of affected relatives, outcome and age relatives diagnosed <i>Psychological factors</i> : optimism, distress, body image and sexuality, specific physical and psychological implications of RRM and RRO
Madalinska et al. (2007) The Netherlands	160 <i>BRCA1/2</i> mutation carriers	Self-report questionnaire and medical record review  Baseline, 3 months and 9 months after RRO, or 6 and 12 months after baseline for women in the screening group	<i>Ovarian cancer screening</i> : TVU and CA125 <i>RRO uptake</i>	<i>Demographics</i> : age, level of education, employment status, marital status and parity <i>Medical history</i> : menopausal status, mutation type, breast cancer history, comorbidities and duration of being aware of high risk status <i>Family history</i> : number of 1 <sup>st</sup> degree relatives with breast and ovarian cancer <i>Perceived cancer risk</i> <i>Psychosocial factors</i> : health perceptions, generic mental health, quality of life and distress <i>Perceived benefits and barriers to RRO</i>
McInerney-Leo et al. (2006) USA	138 high risk women from HBOC families	Self-report questionnaire  6 and 9 months disclosure of <i>BRCA1/2</i> test results	<i>Breast cancer screening</i> : BSE, CBE and mammography <i>Ovarian cancer screening</i> : CA125 and pelvic ultrasound	<i>Demographics</i> : age, employment status, marital status, income and religious affiliation <i>Medical history</i> : <i>BRCA1/2</i> carrier status and cancer history <i>Perceived cancer risk</i> <i>Psychological factors</i> : cancer related distress, breast cancer worry and ovarian cancer worry
Meijers-Heijboer et al. (2003) The Netherlands	275 unaffected women with a 50% risk and 136 women with a 25% risk of carrying a mutation	Not reported  9 months, 1 year, and 2 years post disclosure of <i>BRCA1/2</i> test results	<i>Breast cancer screening</i> : not specified <i>Ovarian cancer screening</i> : not specified <i>RRM and RRO Uptake</i>	<i>Demographics</i> : age and parity <i>Medical history</i> : <i>BRCA1/2</i> carrier status
Meiser et al. (1999) Australia	95 high-risk women who approached familial cancer clinics	Self-report questionnaire  Prior to attendance at a familial cancer clinic	<i>Ovarian cancer screening</i> : ovarian ultrasound and CA125 <i>RRO intention</i>	<i>Demographics</i> : age, level of education, marital status and number of children <i>Family history</i> : number of 1 <sup>st</sup> and 2 <sup>nd</sup> degree relatives with a breast and ovarian cancer history <i>Objective ovarian cancer risk</i> <i>Psychological factors</i> : anxiety and cancer burden
Meiser et al. (2000) Australia	333 women awaiting appointments for risk assessment at familial cancer clinics	Self-report questionnaire  Prior to attendance at a familial cancer clinic	<i>RRM intention</i>	<i>Demographics</i> : age, level of education, marital status and number of children <i>Objective cancer risk</i> <i>Perceived cancer risk</i> <i>Psychological factors</i> : anxiety, distress and stress

Author and Country	Study Sample	Data Collection Method, and Follow-up time of Measurement	Risk-Reducing Strategy Investigated	Factors Investigated
Metcalf et al. (2005) Canada	81 <i>BRCA1/2</i> mutation carriers	Self-report questionnaire  2 to 110 months post disclosure of <i>BRCA1/2</i> test results	<i>Tamoxifen or raloxifene use</i>	<i>Demographics:</i> age and level of education <i>Medical history:</i> unspecified and previous RRM or RRO <i>Knowledge of risks and benefits</i> <i>Desire for additional information</i>
Metcalf et al. (2008b) Canada	517 <i>BRCA1/2</i> mutation carriers (326 <i>BRCA1</i> mutation carriers and 186 <i>BRCA2</i> mutation carriers)	Telephone and mailed questionnaires  Baseline at the time of genetic testing and follow-up mean of 4.5 years (range 1.6 to 10.1 years)	<i>RRM and RRO uptake</i>	<i>Demographics:</i> age (at baseline and follow-up) <i>Medical history:</i> <i>BRCA1/2</i> mutation status, cancer history, tamoxifen use <i>Family history:</i> number of relatives with cancer (age, diagnoses and mortality) and side of family from which mutation was inherited
Peshkin et al. (2002) USA	107 high-risk women (41 <i>BRCA1/2</i> mutation carriers and 66 non-carriers)	Telephone interviews  Baseline prior to genetic counseling and 1, 6 and 12 months post disclosure of <i>BRCA1/2</i> test results	<i>Breast cancer screening:</i> BSE, CBE and mammography	<i>Demographics:</i> age, level of education, marital status, income and ethnicity <i>Perceived cancer risk</i> <i>Psychological factors:</i> distress
Philips et al. (2006) Australia and New Zealand	142 <i>BRCA1/2</i> mutation carriers	Self-report questionnaire and medical records  3 years following enrollment in a familial breast cancer research program	<i>Breast cancer screening:</i> BSE, CBE, mammography, breast ultrasound and MRI <i>Ovarian cancer screening:</i> TVU and CA125 <i>RRM and RRO uptake</i>	<i>Demographics:</i> age, level of education, marital status, ethnicity, parity, country of birth and residence <i>Medical history:</i> <i>BRCA1/2</i> mutation status, awareness of <i>BRCA1/2</i> mutation status, and prior mastectomy and oophorectomy <i>Family history:</i> breast cancer
Ray et al. (2005) USA	62 high-risk women who received genetic counseling	Self-report questionnaire  3 months post genetic counseling	<i>RRM and RRO uptake</i> <i>RRM and RRO intent</i>	<i>Demographics:</i> age, level of education, income, ethnicity and occupation <i>Medical history:</i> <i>BRCA 1/2</i> mutation status, breast or ovarian cancer history, previous mastectomy and oophorectomy <i>Information women considered influential in their decisions and reasons for indecision</i>
Scheuer et al. (2002) USA	233 <i>BRCA1/2</i> mutations carriers	Standardized questionnaires, chart reviews, contact with primary physicians  Mean 24.8 months (mean 1.6 to 66.0 months) following genetic counseling	<i>Breast cancer screening:</i> BSE, CBE, mammography and MRI <i>Ovarian cancer screening:</i> TVU and CA125 <i>RRM and RRO uptake</i>	<i>Demographics:</i> age <i>Medical history:</i> <i>BRCA1/2</i> mutation status, breast and ovarian cancer history, and prior mastectomy and oophorectomy <i>Family history:</i> number of 1 <sup>st</sup> and 2 <sup>nd</sup> degree relatives with a breast and ovarian cancer history
Schmeler et al. (2006) USA	106 <i>BRCA1/2</i> mutation carriers	Medical record review	<i>Ovarian cancer surveillance:</i> pelvic examination, TVU and CA125 <i>RRO uptake</i>	<i>Demographics:</i> age, marital status, parity and ethnicity <i>Medical history:</i> <i>BRCA1/2</i> mutation status and breast cancer history <i>Family history:</i> breast and ovarian cancer

Author and Country	Study Sample	Data Collection Method, and Follow-up time of Measurement	Risk-Reducing Strategy Investigated	Factors Investigated
Stefanek et al. (1995) USA	164 high-risk women	Self-report questionnaire  Prior to attendance at a familial breast cancer clinic	<i>RRM interest and uptake</i>	<i>Demographics:</i> age <i>Medical history:</i> history of breast biopsies <i>Family history:</i> number of 1 <sup>st</sup> degree relatives with a breast cancer history <i>Perceived cancer risk</i> <i>Psychological factors:</i> depression, breast cancer worry <i>Beliefs about breast cancer prevention and early detection</i> <i>Recent breast cancer related life events</i>
Tiller et al. (2005) Australia	129 high-risk women	Self-report questionnaire  Following attendance at a familial cancer clinic	<i>Intended decision:</i> watchful waiting, oral contraceptives, ovarian cancer screening (not specified) and RRO	<i>Demographics:</i> age, level of education, marital status and children <i>Family history:</i> <i>BRCA1/2</i> mutation status, breast and ovarian cancer history, and number of 1 <sup>st</sup> and 2 <sup>nd</sup> degree relatives with ovarian cancer <i>Information preference style</i> <i>Medical decision making preference style</i> <i>Knowledge of ovarian cancer risk management options</i>
Tinley et al. (2004) USA	112 women (33 <i>BRCA1/2</i> mutation carriers and 79 women at 50% risk of carrying a mutation)	Self-report questionnaires  Baseline, at least 2 years post disclosure of <i>BRCA1/2</i> test results	<i>Breast cancer screening:</i> BSE, CBE and mammography <i>Ovarian cancer screening:</i> ovarian ultrasound and CA125	<i>Demographics:</i> age, level of education, employment status, household income and marital status <i>Medical history:</i> breast and ovarian cancer history <i>Family history:</i> breast and ovarian cancer <i>Perceived cancer risk</i> <i>Psychological factors:</i> distress <i>Barriers to screening</i> <i>Adherence determinants for preventing cancer</i>
Unic et al. (2000) The Netherlands	54 high-risk women	Questionnaire (mode of delivery not specified)  After genetic testing and before receiving <i>BRCA1/2</i> test results	<i>Breast cancer screening:</i> not specified <i>RRM uptake (actual and hypothetical)</i>	<i>Demographics:</i> age, level of education, marital status, employment status and children <i>Family history:</i> number of relatives with a breast cancer history, minimum age of breast cancer occurrence in the family and number of relatives who died of breast cancer <i>Medical recommendation about RRM or screening</i> <i>Psychological factors:</i> breast cancer concern, desire to participate, shared decision-making satisfaction, emotional reaction to information material, decisional stress and risk knowledge
Uyei et al. (2006) USA	554 women who underwent <i>BRCA1/2</i> genetic testing (142 <i>BRCA1/2</i> mutation carriers and 410 non-carriers)	Medical records review  Women had undergone genetic testing between January 2001 and December 2005	<i>Breast and ovarian cancer screening:</i> not defined <i>RRM and RRO uptake</i>	<i>Demographic data:</i> ethnicity <i>Medical history:</i> breast and ovarian cancer history, oral contraceptive use, hormone replacement therapy use and history of breast biopsy <i>Family history:</i> breast and ovarian cancer
van Dijk et al. (2003) The Netherlands	241 high-risk women	Self-report questionnaire  Before and immediately following first genetic counseling appointment	<i>RRM intention</i>	<i>Demographics:</i> age, level of education, marital status and number of children <i>Medical history:</i> previous breast cancer history and previous surgery to treat breast cancer <i>Family history:</i> breast cancer history and <i>BRCA1/2</i> detected in family <i>Objective cancer risk</i> <i>Perceived cancer risk</i> <i>Psychological factors:</i> breast cancer worry

Author and Country	Study Sample	Data Collection Method, and Follow-up time of Measurement	Risk-Reducing Strategy Investigated	Factors Investigated
van Dijk et al. (2005) The Netherlands	183 women with uninformative <i>BRCA1/2</i> test result (41 <i>BRCA1/2</i> mutation carriers, 49 <i>BRCA1/2</i> non-carriers and 183 women with uninformative results)	Self-report questionnaire  Before and 1 month post disclosure of <i>BRCA1/2</i> test results	<i>Mammography intention</i> <i>RRM intention</i>	<i>Demographics:</i> age, level of education, marital status and number of children <i>Medical history:</i> <i>BRCA1/2</i> status, breast cancer history, lifetime breast cancer risk and access to breast surveillance <i>Perceived likelihood of having inherited a BRCA1/2 mutation</i>
van Dijk et al. (2008) The Netherlands	338 women underwent <i>BRCA1/2</i> testing (80 <i>BRCA1/2</i> mutation carriers and 258 non-carriers)	Self-report questionnaire  Baseline, 1 week post <i>BRCA1/2</i> testing, 4 weeks post <i>BRCA1/2</i> testing, 2 weeks, 3 months and 9 months post disclosure of <i>BRCA1/2</i> test results	<i>Screening preference</i> <i>RRM preference and uptake</i>	<i>Demographics:</i> age, level of education, marital status, presence and age of children <i>Medical history:</i> breast and ovarian cancer history, time since diagnosis, breast cancer treatment if applicable, having had biopsies and having undergone RRM <i>Perceived cancer risk</i> <i>Psychological factors:</i> anxiety <i>Anticipatory regret</i>

Note. BSE: breast self-examination  
CBE: clinical breast examination  
TVU: transvaginal ultrasound  
RRM: risk-reducing mastectomy  
RRO: risk-reducing oophorectomy

Table A.2: Qualitative Research Studies Reviewed

Author, Date, & Country	Study Sample	Methodology & Data Collection Method	Purpose
Altschuler & Somkin (2005) USA	28 high risk women who took chemoprevention and 23 high risk women who declined chemoprevention	Qualitative, grounded theory  In-depth, semi-structured interviews	Explore why women at similarly high calculated risk made different choices about whether or not to join a clinical trial of raloxifene or tamoxifen for five years to reduce their likelihood of developing breast cancer.
Brain et al. (2004) UK	10 women newly identified as being high-risk for ovarian cancer	Qualitative  In-depth & semi-structured interviews	To explore women's perceptions of the information received regarding risk management options and attitudes towards both ovarian screening and RRO.
Frost et al. (2004) USA	4 <i>BRCA1/2</i> mutation carriers, 5 non-carriers and 6 women with <i>BRCA1/2</i> variants of uncertain significance	Qualitative, grounded theory  Focus groups, open-ended in-depth interviews	To explore how uncertain genetic test results influenced women's decision making about subsequent medical management.
Hallowell (1998) UK	41 high-risk women who had genetic counselling	Qualitative  In-depth & semi-structured interviews	To describe women's perceptions of the meaning, acceptability, and the potential costs and benefits of prophylactic surgery.
Hallowell (2000) UK	23 high-risk women who had undergone RRO	Qualitative  In-depth interviews	To determine the information needs of high-risk premenopausal women who are faced with ovarian cancer risk management decisions.
Hallowell et al. (2001) UK	23 high-risk women who had undergone RRO and 26 high-risk who remained on a screening program	Qualitative  In-depth interviews	To describe the factors that influence high-risk premenopausal women's decisions about RRO.
Kenen et al. (2007) USA	21 high-risk women who posted on the Facing Our Risk of Cancer Empowered website	Qualitative  Online postings that were part of a thread about genetic testing, genetic counseling and family history	To examine what women considered to be the physical, psychological, and social consequences of RRM during the decision-making, recuperation and reconstruction process.
McCullum et al. (2007) Canada	3 <i>BRCA1/2</i> mutation carriers	Qualitative, case series  Open-ended and semi-structured interviews	To explore women's decision-making experiences related to the option of RRM.
Swisher et al. (2001) USA	30 high-risk women undergoing RRO and 30 high-risk women undergoing ovarian cancer surveillance	Qualitative  In-depth telephone interviews and open-ended questions	To evaluate decision making, information gathering, satisfaction and regret in women at increased risk of ovarian cancer who had undergone RRO or ovarian cancer surveillance.

## Appendix B: Hereditary Breast and Ovarian Cancer Risk-Reducing Strategies

### Questionnaire

# Hereditary Breast and Ovarian Cancer Risk Reducing Strategies Questionnaire

The last sections are only for women who are at risk for hereditary breast and ovarian cancer.

If you have a family history of breast and ovarian cancer, please answer the questions in the next section and continue to the end of the survey.

In this questionnaire, the information you are being asked for will help us understand the strategies you may presently be using, or have used in the past, that influence your risk of developing breast and ovarian cancer. **Please read each statement and question carefully and where necessary, answer by placing a firm checkmark in the most appropriate space; otherwise, please write your answer in the space provided.**

1. I have undergone the following in order to screen for breast cancer so that if found, it can be treated early:

a. Breast self examination

- ☐ within the last 6 months
- ☐ 7 months to 1 year ago
- ☐ more than 1 year ago
- ☐ I have never performed a breast self examination
- ☐ do not know

b. Physical breast examination by a health care professional (clinical breast examination)

- ☐ within the last 6 months
- ☐ 7 months to 1 year ago
- ☐ more than 1 year ago
- ☐ I have never had a clinical breast examination
- ☐ do not know

c. Mammogram (breast x-ray)

- ☐ within the last year
- ☐ more than 1 year ago
- ☐ I have never had a mammogram
- ☐ do not know

d. Breast ultrasound (a small hand held unit is passed gently over the breast to make a picture of the inside of the breast)

- ☐ within the last year
- ☐ more than 1 year ago
- ☐ I have never had a breast ultrasound
- ☐ do not know

2. I have undergone the following in order to screen for ovarian cancer so that if found, it can be treated early:

- 
- a. Manual pelvic examination performed by a health care professional is a procedure where two fingers are inserted into your vagina and one hand placed on your abdomen to examine your ovaries. This is often done at the same time as a pap test.

I have had a manual pelvic examination by a health care professional:

☐ within the last year      ☐ more than 1 year ago      ☐ never      ☐ do not know

- 
- b. A transvaginal ultrasound is a test where a device is gently inserted into your vagina to make a picture of your female organs, including your ovaries.

I have had a transvaginal ultrasound:

☐ within the last year      ☐ more than 1 year ago      ☐ never      ☐ do not know

- 
- c. CA 125 is a protein in the blood that may be elevated in people with certain types of cancers, including ovarian cancer. The level of CA 125 can be checked by a blood test.

I have had a blood test for CA 125:

☐ within the last year      ☐ more than 1 year ago      ☐ never      ☐ do not know

3. Regarding the use of hormone replacement therapy (estrogen and/or progestin):

☐ I have never taken hormone replacement therapy

☐ I currently take hormone replacement therapy

☐ I took hormone replacement therapy, but have now stopped taking this medication

The main reason I stopped taking hormone replacement therapy was because (**please CHECK the most important reason**):

☐ I didn't need it any longer

☐ I didn't like the side effects of the medication

☐ I wanted to reduce my risk of breast or ovarian cancer

☐ My doctor told me to stop taking it

☐ Other (specify)

---

4. Regarding the use of oral contraceptives (the birth control pill):

- ☐ I do not take oral contraceptives
- ☐ I currently take oral contraceptives as a form of birth control
- ☐ I currently take oral contraceptives to prevent ovarian cancer
- ☐ I took oral contraceptives, but have now stopped taking this medication

The main reason I stopped taking oral contraceptives was because (**please CHECK the most important reason**):

- ☐ I didn't need it any longer
  - ☐ I didn't like the side effects of the medication
  - ☐ I wanted to reduce my risk of breast cancer
  - ☐ My doctor told me to stop taking it
  - ☐ Other (specify)
- 

5. Regarding the use of tamoxifen or raloxifene (medications):

- ☐ I do not take tamoxifen or raloxifene
- ☐ I currently take tamoxifen or raloxifene as part of my treatment for cancer
- ☐ I currently take tamoxifen or raloxifene to prevent breast or ovarian cancer
- ☐ I took tamoxifen or raloxifene, but have now stopped taking this medication

The main reason I stopped taking tamoxifen or raloxifene was because (**please CHECK the most important reason**):

- ☐ I didn't need it any longer
  - ☐ I didn't like the side effects of the medication
  - ☐ My doctor told me to stop taking it
  - ☐ Other (specify)
- 

6. Regarding the surgical removal of my breast(s) to **prevent** breast cancer (prophylactic mastectomy):

- ☐ I have not had my breasts surgically removed to prevent breast cancer.
- ☐ I have had one breast removed to prevent breast cancer
- ☐ I have had both breasts removed to prevent breast cancer
- ☐ I am awaiting breast surgery to prevent breast cancer

7. Regarding the surgical removal of my breast(s) to **treat** breast cancer (mastectomy):

- ☐ I have not had my breasts surgically removed to treat breast cancer
- ☐ I have had one breast removed to treat breast cancer
- ☐ I have had both breasts removed to treat breast cancer
- ☐ I am awaiting breast surgery to treat breast cancer

8. Regarding the surgical removal of my ovaries to **prevent** ovarian cancer (prophylactic oophorectomy):

- ☐ I have not had my ovaries surgically removed to prevent ovarian cancer
- ☐ I have had my ovaries surgically removed to prevent ovarian cancer
- ☐ I am awaiting surgery to remove my ovaries to prevent ovarian cancer
- ☐ I have had my ovaries surgically removed for other medical reasons

9. Regarding the surgical removal of my ovaries to **treat** ovarian cancer (oophorectomy):

- ( ) I have not had my ovaries surgically removed to treat ovarian cancer  
 ( ) I have had my ovaries surgically removed to treat ovarian cancer  
 ( ) I am awaiting surgery to remove my ovaries to treat ovarian cancer  
 ( ) I have had my ovaries surgically removed for other medical reasons

10. I choose certain foods to eat because I am concerned about breast and ovarian cancer.

- ( ) No ( ) Yes

**If yes, please specify what foods**

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11. I avoid certain foods because I am concerned about breast and ovarian cancer.

- ( ) No ( ) Yes

**If yes, please specify what foods**

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12. Since I became aware of my risk of developing breast and ovarian cancer I have attempted to change my physical activity.

- ( ) No ( ) Yes

**If yes, describe changes in your physical activity**

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13. Since I became aware of my risk of developing breast and ovarian cancer I have attempted to change my consumption of alcohol.

- ( ) No ( ) Yes

( ) Not applicable because I do not drink alcohol

**If yes, I have (please CHECK all that apply):**

- ( ) Increased the amount of alcohol I consume  
 ( ) Decreased the amount of alcohol I consume  
 ( ) Quit drinking alcohol  
 ( ) Changed the type of alcohol I consume  
 ( ) Other, please specify

---

14. Since I became aware of my risk of developing breast and ovarian cancer I have changed my smoking behaviour.

- ( ) No ( ) Yes  
( ) Not applicable because I do not smoke

**If yes, I have (please CHECK all that apply):**

- ( ) Increased the amount I smoke  
( ) Decreased the amount I smoke  
( ) Quit smoking  
( ) Changed the brand of cigarette I smoke  
( ) Other, please specify
- 

15. Since I became aware of my risk of developing breast and ovarian cancer, I have had an appointment in person with an alternative/complementary health care provider (for example, a naturopath, homeopath, traditional Chinese medicine physician) about how to prevent breast and ovarian cancer.

- ( ) No ( ) Yes

**If yes, this included (please CHECK all that apply):**

- |  |  |
|--|--|
| ( ) Massage therapist                        | ( ) Acupuncturist                      |
| ( ) Homeopath                                | ( ) Naturopath                         |
| ( ) Traditional Chinese Medical Practitioner | ( ) Meditation or Relaxation therapist |
| ( ) Biofeedback teacher                      | ( ) Herbalist                          |
| ( ) Reflexologist                            | ( ) Spiritual healer                   |
| ( ) Reiki Master                             | ( ) Therapeutic/Healing Touch          |
| Practitioner                                 |  |
| ( ) Other (specify)                          |  |
- 

16. Since I became aware of my risk of developing breast and ovarian cancer, I have begun taking natural health products (e.g., vitamins, minerals, herbal therapies, and dietary supplements) to prevent breast and ovarian cancer.

- ( ) No ( ) Yes

**If yes, this included (please CHECK all that apply):**

- ( ) Herbal therapies  
(specify) \_\_\_\_\_  
( ) Vitamins/minerals  
(specify) \_\_\_\_\_  
( ) Other dietary supplements (specify) \_\_\_\_\_
-

17. In addition to the information you have already received, what other information would you like about decreasing your risk for developing breast cancer?

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18. In addition to the information you have already received, what other information would you like about decreasing your risk for developing ovarian cancer?

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Additional comments about risk-reducing strategies for breast and ovarian cancer:

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08/12/09 11:43 AM



The University of British Columbia  
Office of Research Services  
**Behavioural Research Ethics Board**  
Suite 102, 6190 Agronomy Road, Vancouver, B.C. V6T 1Z3

## CERTIFICATE OF APPROVAL - FULL BOARD

<b>PRINCIPAL INVESTIGATOR:</b> Joan L. Bottorff		<b>INSTITUTION / DEPARTMENT:</b> UBC/UBCO VP Academic (Academic Units)/UBCO Academic		<b>UBC BREC NUMBER:</b> H07-02281	
<b>INSTITUTION(S) WHERE RESEARCH WILL BE CARRIED OUT:</b>					
<b>Institution</b> UBC BC Cancer Agency Other locations where the research will be conducted: N/A			<b>Site</b> Vancouver (excludes UBC Hospital) Vancouver BCCA		
<b>CO-INVESTIGATOR(S):</b> Charmaine Kim-Sing Patricia A. Rodney Lynde G. Balneaves Amanda Fuchsia Howard					
<b>SPONSORING AGENCIES:</b> N/A					
<b>PROJECT TITLE:</b> Women's Decision Making Related to Cancer Risk-Reducing Strategies in the Context of Hereditary Breast and Ovarian Cancer					
<b>REB MEETING DATE:</b> November 22, 2007		<b>CERTIFICATE EXPIRY DATE:</b> November 22, 2008			
<b>DOCUMENTS INCLUDED IN THIS APPROVAL:</b>				<b>DATE APPROVED:</b> December 17, 2007	
<b>Document Name</b>	<b>Version</b>	<b>Date</b>			
<b>Consent Forms:</b>					
Consent Form High Risk Women	N/A	December 12, 2007			
Consent Form: Expert Consultation Meeting - Health Care Prof	N/A	October 27, 2007			
Consent Form: Expert Consultation Meeting - High Risk Women	N/A	October 27, 2007			
<b>Questionnaire, Questionnaire Cover Letter, Tests:</b>					
Demographic & Family Cancer History Form - High Risk Women	N/A	October 27, 2007			
Interview Guide - High Risk Women	N/A	October 27, 2007			
<b>Letter of Initial Contact:</b>					
HCP Letter of Invitation	N/A	December 12, 2007			
Letter of Invitation - Export Consultation Meeting	N/A	October 27, 2007			
<b>Other Documents:</b>					
BCCA REB Email Correspondence	N/A	December 3, 2007			
BCCA/HCP Letter of Support	N/A	November 20, 2007			
The application for ethical review and the document(s) listed above have been reviewed and the procedures were found to be acceptable on ethical grounds for research involving human subjects.					
Approval is issued on behalf of the Behavioural Research Ethics Board and signed electronically by one of the following:					
Dr. M. Judith Lynam, Chair Dr. Jim Rupert, Associate Chair Dr. Laurie Ford, Associate Chair					



The University of British Columbia  
Office of Research Services  
**Behavioural Research Ethics Board**  
Suite 102, 6190 Agronomy Road, Vancouver, B.C. V6T 1Z3

## CERTIFICATE OF APPROVAL- MINIMAL RISK RENEWAL

<b>PRINCIPAL INVESTIGATOR:</b> Joan L. Bottorff	<b>DEPARTMENT:</b> UBC/UBCO VP Academic (Academic Units)/UBCO Academic	<b>UBC BREB NUMBER:</b> H07-02281
<b>INSTITUTION(S) WHERE RESEARCH WILL BE CARRIED OUT:</b>		
<b>Institution</b> UBC BC Cancer Agency Other locations where the research will be conducted: N/A	<b>Site</b> Vancouver (excludes UBC Hospital) Vancouver BCCA	
<b>CO-INVESTIGATOR(S):</b> Charmaine Kim-Sing Patricia A. Rodney Lynda G. Balneaves Amanda Fuchsia Howard		
<b>SPONSORING AGENCIES:</b> N/A		
<b>PROJECT TITLE:</b> Women's Decision Making Related to Cancer Risk-Reducing Strategies in the Context of Hereditary Breast and Ovarian Cancer		
<b>EXPIRY DATE OF THIS APPROVAL:</b> November 13, 2009		
<b>APPROVAL DATE:</b> November 13, 2008		
The Annual Renewal for Study have been reviewed and the procedures were found to be acceptable on ethical grounds for research involving human subjects.		
<p style="text-align: center;"><b>Approval is issued on behalf of the Behavioural Research Ethics Boardand</b></p> <p style="text-align: center;">             Dr. M. Judith Lynam, Chair              Dr. Ken Craig, Chair              Dr. Jim Rupert, Associate Chair              Dr. Laurie Ford, Associate Chair              Dr. Daniel Salhani, Associate Chair              Dr. Anita Ho, Associate Chair           </p>		



The University of British Columbia  
Office of Research Services  
**Behavioural Research Ethics Board**  
Suite 102, 6190 Agronomy Road, Vancouver, B.C. V6T 1Z3

## CERTIFICATE OF APPROVAL- MINIMAL RISK RENEWAL

<b>PRINCIPAL INVESTIGATOR:</b> Joan L. Bottorff	<b>DEPARTMENT:</b> UBC/UBCO Health & Social Development/UBCO Nursing	<b>UBC BREB NUMBER:</b> H07-02281
<b>INSTITUTION(S) WHERE RESEARCH WILL BE CARRIED OUT:</b>		
<b>Institution</b> UBC BC Cancer Agency Other locations where the research will be conducted: N/A	<b>Site</b> Vancouver (excludes UBC Hospital) Vancouver BCCA	
<b>CO-INVESTIGATOR(S):</b> Charmaine Kim-Sing Patricia A. Rodney Lynda G. Balneaves Amanda Fuchsia Howard		
<b>SPONSORING AGENCIES:</b> N/A		
<b>PROJECT TITLE:</b> Women's Decision Making Related to Cancer Risk-Reducing Strategies in the Context of Hereditary Breast and Ovarian Cancer		
<b>EXPIRY DATE OF THIS APPROVAL:</b> October 23, 2010		
<b>APPROVAL DATE:</b> October 23, 2009		
The Annual Renewal for Study have been reviewed and the procedures were found to be acceptable on ethical grounds for research involving human subjects.		
<p style="text-align: center;"><b>Approval is issued on behalf of the Behavioural Research Ethics Board</b></p> <p style="text-align: center;">             Dr. M. Judith Lynam, Chair              Dr. Ken Craig, Chair              Dr. Jim Rupert, Associate Chair              Dr. Laurie Ford, Associate Chair              Dr. Anita Ho, Associate Chair           </p>		

## Appendix D: Consent Form

December 12, 2007

**School of Nursing**  
 302-6190 Agronomy Road  
 Vancouver, B.C., Canada V6T 1Z3  
 Tel: (604) 822-5081 Fax: (604) 822-7869  
[www.nursing.ubc.ca](http://www.nursing.ubc.ca)

### CONSENT FORM – HIGH RISK WOMEN

**Title of Study:** Women's Decision Making Related to Cancer Risk-Reducing Strategies in the Context of Hereditary Breast and Ovarian Cancer

**Principal Investigator:**

Dr. Joan Bottorff, Professor, Dean of Faculty of Health & Social Development, UBC-O  
 250-807-9901

**Co-Investigators:**

Ms. Fuchsia Howard, PhD Candidate, School of Nursing, UBC	604-822-5081
Dr. Lynda Balneaves, Assistant Professor, School of Nursing, UBC	604-822-7679
Dr. Paddy Rodney, Associate Professor, School of Nursing, UBC	604-822-7507
Dr. Charmaine Kim-Sing, Director, Hereditary Cancer Program BCCA	604-877-6000 ext 2672

This study is being conducted in partial fulfillment of Ms. Fuchsia Howards' doctoral dissertation at the University of British Columbia, School of Nursing. Data collected for this study will be accessible by the principal and co-investigators only. The findings of this study will be published in the form of a doctoral dissertation and written research reports. Your name will NOT be included in any of the published findings.

**Purpose:**

We are doing this study to understand how women, who are at high risk for hereditary breast and ovarian cancer (HBOC), make decisions about ways they may decrease their risk of developing cancer. These risk-reducing strategies may include cancer screening, prophylactic surgery, and drug therapy. Women may also perceive lifestyle behaviour changes and complementary and alternative therapy use as being risk-reducing strategies. The findings from this study will give guidance for providing decision support to women at high risk for HBOC. You are being asked to participate in this study because you are at high risk for HBOC.

**Study Procedures:**

As a participant in this study, you will take part in an interview with one of the researchers. During this interview, you will be asked about your experiences of cancer in your family. Questions will also be asked about the ways you know of to decrease your risk of developing HBOC and the strategies you have previously, or are currently, considering to prevent HBOC. You will be asked about what it was, or is, like for you to make these decisions, as well as the factors that have influenced your decision making. The interview will last up to one hour, will occur at a time and place of your choice, will be tape recorded, and later transcribed. You will also be asked to complete a short questionnaire that includes demographic questions and questions about your family history of cancer.

Lastly, as a participant in this study, you may be invited to attend a consultation meeting to talk about the implications of the findings of this study.

**Risk and Potential Benefits:**

There are no expected risks (problems) related to taking part in this study. Because this may be a sensitive topic for you to discuss, however, participation in this study may bring about strong emotions. Should this occur, we will ensure there are appropriate resources made available to help you. Your decision whether or not to take part in this research will not influence the health care you receive.

**Confidentiality:**

We will keep your name and information you provide strictly confidential. We will not use your name in the research reports, nor will we include your name or other personal information in the transcribed interviews. Instead, you will be provided a numerical identifier. A list linking your name to the numerical identifier will be kept separate from the interviews. All study information will be stored in a locked file cabinet and computer files will be password protected. The information collected in this study may be used for teaching purposes without revealing any information that would identify you.

**Consent:**

It is entirely up to you whether you take part in this study or not. You can change your mind and withdraw from this study at any time. By signing this consent form, you are agreeing to participate in this study and acknowledge that you have received a copy of this consent form for your own records. By signing this consent form, you do not waive any of your legal rights.

If you have any questions about this study, now or later, please contact Ms. Howard (604-822-5081). If you have any questions about your rights as a research participant, you can contact the Research Subject Information Line in the UBC Office of Research Services at 604-822-8598.

\*\*\*\*\*

*I have read the above information and understand what I have to do and what will happen if I take part in this study. I freely choose to take part in this study and I have a copy of the consent form.*

\_\_\_\_\_  
Please print name

\_\_\_\_\_  
Signature of Participant

\_\_\_\_\_  
Date

\*\*\*\*\*

Would you be willing to be contacted in the future for other studies related to hereditary breast and ovarian cancer?

- ☐ Yes, I would like to receive information about future studies  
☐ No, I would not like to receive information about future studies

\_\_\_\_\_  
Signature of Participant

\_\_\_\_\_  
Phone Number

\_\_\_\_\_  
Date

\*\*\*\*\*

Please provide us with the following information. PLEASE PRINT

Name: \_\_\_\_\_

Address: \_\_\_\_\_

City: \_\_\_\_\_

Postal Code: \_\_\_\_\_

Telephone Number: (\_\_\_\_) \_\_\_\_\_

PLEASE MAIL THE SIGNED CONSENT FORM TO THE RESEARCH TEAM:

FUCHSIA HOWARD  
 SCHOOL OF NURSING  
 302 – 6190 AGRONOMY ROAD  
 UNIVERSITY OF BRITISH COLUMBIA  
 VANCOUVER, B.C.  
 V6T 1Z3

OR, FAX TO (604) 822-7869

## **Appendix E: Interview Guide for *BRCA1/2* Carrier Women**

### **Section A: Being at High Risk for HBOC**

1. When, if at all, did you first experience, or come to know about, cancer in your family?
  - a) What was it like?
  - b) What did you think then?
  - c) Did you discuss this with anyone (i.e., family, friends, health care professionals)? If so, can you recall a conversation you had?
2. Tell me about how you came to think that you were at high risk for HBOC?
  - a) Tell me about your thoughts and feelings when you learned about HBOC.
  - b) Tell me about how you learned to handle being at high risk for HBOC.
3. How, if at all, have your thoughts and feelings about being at high risk for cancer changed since then?
4. What positive changes, if any, have occurred in your life since becoming aware of HBOC?
5. What negative changes, if any, have occurred in your life since becoming aware of HBOC?

### **Section B: Decision Making about Risk-reducing Strategies**

6. When you first learned you were at high risk for HBOC, what, if anything, did you know about ways you could decrease your risk of developing or preventing cancer?
  - a) Tell me about how you came to know about these.
  - b) Did you discuss this with anyone (i.e., family, friends, health care professionals)? If so, can you recall a conversation you had?
7. Now I'd like to get a picture of the types of strategies you may have previously used or are currently using to detect or prevent HBOC and how you came to decide to use these.  
What, if anything, have you done to detect or prevent HBOC?  
(For each risk-reducing strategy described by participants, ask the following questions):
  - a) What first lead you to consider this?
  - b) How did you go about making a decision to \_\_\_\_\_(use participants language)?
  - c) Looking back, are there any reasons or events that stand out in your mind that convinced you to \_\_\_\_\_ (use participants language)?
    - i. Please describe these?
  - d) What, if anything, made it easier for you to decide to \_\_\_\_\_(use participants language)?
    - i. How so? What would be an example?
  - e) What, if anything, made it harder for you to decide to \_\_\_\_\_(use participants language)?
    - i. How so? What would be an example?
  - f) Who has been the most helpful to you while making the decision to \_\_\_\_\_(use participants language)?
    - i. Give me an example of how he/she has been helpful.
  - g) Tell me whom you have talked to about your decision to \_\_\_\_\_(use participants language)?

- i. If I were in the room, what conversations would I have heard?
- h) Who, if anyone, influenced your decision to \_\_\_\_\_(use participants language)?
  - i. When was that?
  - ii. How were they involved?
- i) What types of information, if any, did you use to help you make your decision to \_\_\_\_\_(use participants language)?
  - i. What type of evidence did you look for? What was most persuasive?
  - ii. Did you feel equally confident about the information you receive from different sources, or did you value certain opinions over others?
  - iii. Has this changed (use participants language)?
- j) Has any organization been helpful?
  - i. What did \_\_\_\_\_ help you with?
  - ii. How has it been helpful?
- k) Has any organization made it more difficult to make a decision to \_\_\_\_\_(use participants language)?
  - i. What did \_\_\_\_\_ make more difficult?
  - ii. How has it been more difficult?
- l) Have you ever felt you were restricted in making your decision to \_\_\_\_\_(use participants language)?
  - i. What did \_\_\_\_\_ make more difficult?
  - ii. How has it been more difficult?
- m) How was this decision different than the way you make other types of decisions?
- n) How was this decision similar to the way you make other types of decisions?
- 8. How have your thoughts about ways of detecting cancer early or preventing cancer changed since you first became aware you were at high risk?
  - a) Tell me about what is different now?
  - b) Tell me about what is the same.
- 9. What intentions, if any, do you have of using strategies to detect or prevent cancer in the future?

### **Section C: Summary & Future Directions**

- 10. After having these experiences, what advice would you give to a woman who is trying to make decisions about HBOC risk-reducing strategies?
- 11. What recommendations do you have for health care professionals working with women who are trying to make decisions about HBOC risk-reducing strategies?
- 12. Is there anything that you might not have thought about before that occurred to you during this interview?
- 13. Is there anything else you think I should know or understand better?

***This has been very helpful. These are all the questions that I have. Thank you for taking part in this interview.***

## Appendix F: Demographic Data & Cancer History Form

**Date:** \_\_\_\_\_  
dd/mm/yyyy

ID \_\_\_\_\_

<p>1. Age in years: _____</p>	<p>2. Current Marital Status:</p> <p>___ (1) Single, never married</p> <p>___ (2) Married/common-law</p> <p>___ (3) Separated, divorced</p> <p>___ (4) Widowed</p>
<p>3. Current Employment Status: (select the best response)</p> <p>___ (1) Employed full-time</p> <p>___ (2) Employed part time by choice</p> <p>___ (3) Employed part-time due to illness</p> <p>___ (4) Unemployed due to illness</p> <p>___ (5) Unemployed</p> <p>___ (6) Retired</p> <p>___ (7) Homemaker</p> <p>___ (8) Student</p>	<p>4. Ethnic Background:</p> <p>___ (1) Anglo-Saxon</p> <p>___ (2) African-Canadian</p> <p>___ (3) Ashkenazi Jewish</p> <p>___ (4) Asian</p> <p>___ (5) Hispanic</p> <p>___ (6) Other</p>
<p>5. Level of Education:</p> <p>___ (1) Elementary school</p> <p>___ (2) Part of high school</p> <p>___ (3) High school</p> <p>___ (4) Part of university/college</p> <p>___ (5) University/college</p> <p>___ (6) Graduate school</p>	<p>6. Annual Income Level: (household)</p> <p>___ (1) \$00,000 - 20,000</p> <p>___ (2) \$21,000 - 40,000</p> <p>___ (3) \$41,000 - 60,000</p> <p>___ (4) \$61,000 - 80,000</p> <p>___ (5) \$81,000 - 100,000</p> <p>___ (6) \$ greater than \$100,000</p>
<p>7. Do you have children?    ___ (1) Yes    ___ (2) No</p> <p>8. Number of Children: _____</p> <p>9. Number of children in each age category</p> <p>___ under 1 year – 5 years old</p> <p>___ 6 – 12 years old</p> <p>___ 13 – 19 years old</p> <p>___ 20 – 30 years old</p> <p>___ 31 or older</p>	

10. Have you previously had cancer?

- ☐ No  
☐ Yes

If Yes, please specify:

The type of cancer you had. \_\_\_\_\_

The age at which you were diagnosed. \_\_\_\_\_

The year you were diagnosed. \_\_\_\_\_

The treatments you received. \_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

11. Has anyone in your family had cancer?

- ☐ No  
☐ Yes

If Yes, please specify (for each family member who has had cancer):

The type of cancer they had. \_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

The age at which they were diagnosed. \_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

The year they were diagnosed. \_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

The treatments they received. \_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

12. When did you go for genetic testing? \_\_\_\_\_