Motivations for Males Affected by HBOC to Disclose Genetic Health Information to Family Members and Health Care Providers

THESIS

Presented in Partial Fulfillment of the Requirements for the Degree Master of Science in the Graduate School of The Ohio State University

By
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Abstract

Introduction: Hereditary breast and ovarian cancer syndrome (HBOC) is a cancer-predisposition syndrome that affects both men and women, with more significant cancer risk elevations in women. Because there are well-established guidelines for cancer risk reduction and prevention in HBOC, it is critical that health care professionals understand information-sharing patterns among patients to facilitate communication processes and identify at-risk family members. Dissemination of familial genetic risk information in females with HBOC is well defined, but knowledge about how males share this information is limited. The aims of this study include: to describe participants’ feelings and opinions about HBOC; to ascertain participants’ extent of information sharing with family and medical personnel; and to describe the needs of participants for information and resources provided by genetic counselors and other health care providers.

Methods: We interviewed 21 primarily Ashkenazi Jewish men who were accrued through Facing Our Risk of Cancer Empowered (FORCE). Interviews focused on family cancer history, experiences with cancer and genetic testing, motivations to pursue genetic testing and subsequently disclose genetic test results, information sharing patterns, healthcare provider response, and participants’ emotional support systems. The interviews were transcribed in their entirety, coded, and analyzed based on grounded theory.
Results: Eighteen transcripts were used for the analysis. Results can be classified into 5 main themes. Participants (n=8) were most concerned about cancer risk for their children and female family members, and most (n=11) mentioned HBOC provides them increased personal awareness, but has a negligible impact on their life overall (n=9). Men (n=11) were interested in a male focused support group to discuss HBOC and gain knowledge and information. Participants (n=9) took on active and open communication roles with family members and health care providers. The majority of participants (n=14) discussed the need for knowledge and awareness among the health care community and general population regarding male HBOC risks.

Conclusion: This study serves as a pilot study and provides important and novel insights into psychosocial impacts, communication patterns, encounters with health care practitioners, and expressed needs of males with HBOC.
Acknowledgments

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Leigha Senter, my thesis advisor, Robert Pilarski and Dr. Doreen Agnese, members of
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member population, and the men that participated in this study.
Vita

May 2010 .................................................. Mountain Vista High School

May 2014 ................................................. B.A. Ecology and Evolutionary Biology,
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Fields of Study

Major Field: Genetic Counseling
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Chapter 1: Background

Hereditary breast and ovarian cancer syndrome (HBOC) is a cancer-predisposition syndrome that is caused by germline mutations in \textit{BRCA1} and \textit{BRCA2}. \textit{BRCA1} and \textit{BRCA2} gene mutations are inherited in an autosomal dominant manner and both male and female first-degree relatives of mutation carriers have a 50% risk of having the same mutation (Liede et al., 2000). Females with a \textit{BRCA} gene mutation have a lifetime risk of breast cancer of up to 85%, and a lifetime risk of ovarian cancer of up to 40% (Antoniou et al., 2003). Males with a \textit{BRCA} gene mutation have an increased risk of developing breast and prostate cancers with lifetime risks of 7-9% and 20%, respectively (Liede, Karlan, & Narod, 2004). Male breast cancer is typically diagnosed later and with a worse prognosis than female breast cancer (Liede et al., 2004). Some studies show that both males and females also face an increased risk of cancers of the colon, pancreas and melanoma, with higher risks observed in \textit{BRCA2} mutation carriers than \textit{BRCA1} mutation carriers (Liede et al., 2004).

When an individual is found to have a \textit{BRCA} gene mutation, it is critical that family members are notified, given their potential for elevated cancer risk and implications of these risks for their health management. Due to privacy laws and the fact that it is unusual for a given provider to have direct contact with their patients’ relatives, health
care professionals must often rely on probands, or the first individual in a family to undergo genetic testing, to disseminate information about his or her result, and the genetic disorder running in the family, to other family members and in many cases, also to other health care professionals. Given the importance of this responsibility, it’s worthwhile for providers to understand the factors that impact communication following the receipt of genetic testing results.

In a review of 33 studies of familial communication of genetic health information that included 24 studies of families that have HBOC, Weiseman et al. identified four main reasons that participants describe as motivation to communicate genetic health information to family members: to discharge personal responsibility for informing the family, to obtain support or advice, to obtain further information from family members, and to prevent illness in family members (Wiseman, Dancyger, & Michie, 2010). Follow through on these motivations or lack thereof was influenced primarily by six observed factors: perceived responsibility to tell, relationship type and quality, difficulty deciding who to tell, anticipation of relatives’ reactions, mutation status, and personal feelings. Overall, counselees felt that they were responsible for communicating genetic information and that their family members had a right to know such information, most often in order to prevent disease or facilitate healthcare decision-making. Counselees communicated information most often to family members they felt physically and emotionally close to, and most often informed those that they felt would benefit from the information, have a direct interest in the information, and/or take action on the
information. Study participants felt no obligation to share information with family members that they were estranged from or had poor relationships with, and dissemination of information was more difficult in families with poor communication history and negative emotions surrounding the discussion of cancer. Guilt, anxiety, and feelings of vulnerability to family members inhibited the dissemination of genetic information (Wiseman et al., 2010).

Similarly, Dancyger et al. conducted semi-structured interviews regarding how females in the United Kingdom communicate genetic health information, approximately one month after probands affected with breast or ovarian cancer received a positive BRCA result (Dancyger et al., 2011). The study also included interviews with at least two relatives of each of these females. Like in the previous study, three major themes that were positively associated with dissemination of genetic test results emerged from the interviews, including the responsibility to communicate results to relatives, perceived emotional and developmental readiness of the individual receiving results, and communicating results in the context of existing family culture and dynamics. Interestingly, though, some participants were unaware that BRCA affects men at all (Dancyger et al., 2011).

Councellees in the 33 studies (with primarily female participants) that were reviewed by Weiseman et al. also demonstrated specific patterns of communication, and consistently communicated with female first-degree relatives first (Wiseman et al., 2010). They
communicated less often with second-degree or more distant relatives, male relatives, and young children. Siblings and adult children of counselees were often informed first, with parents following. Counselees that were found to be mutation positive had similar patterns of dissemination to those found to be mutation negative, though those that were mutation negative or had inconclusive results tended to communicate with fewer relatives (Wiseman et al., 2010). To date, information regarding the communication of genetic information surrounding HBOC has been largely focused on women (Hallowell et al., 2005). Few studies pertain to communication in males, and it is not widely understood to what extent and to whom men discuss family history of HBOC or BRCA genetic test results.

Though none of the 33 studies reviewed by Weisman et al. directly investigated the difference in communication according to gender, families often had a “pivotal person,” usually a woman, who took on the responsibility to disseminate results to key family members, both inside and outside of the core family unit (Wiseman et al., 2010). Moreover, communication of genetic test results was found to be a “gendered activity” with women assuming the majority of the burden for disseminating the results, and male counselees reporting information to fewer relatives, and with less emphasis on “supportive functions,” like receiving emotional support from other family members (Wiseman et al., 2010).
d'Agincourt-Canning explored how gender relates to the moral duty to disclose genetic information and included a subset of interviews with 36 participants—31 women and 5 men (d'Agincourt-Canning, 2001). The author argues that general responsibility for family health falls to women in Western society, and that women often “negotiate” health care for their families and subsequently communicate health information (d'Agincourt-Canning, 2001). Another study found that female family members inhabit the primary role in all aspects of genetics, taking the lead in communicating information, even when it originates on the paternal side of the family (d'Agincourt-Canning, 2001; Richards & Marteau, 1996).

Data suggest that women readily disseminate *BRCA* gene test results to a variety of family members. In a study of 1103 high-risk women, Cheung et al. reported the association of demographic variables on family communication and uptake of cascade genetic testing (the testing of family members for a known familial gene mutation) (Cheung, Olson, Yu, Han, & Beattie, 2010). Rates of communication among women treated at a tertiary referral cancer center, as well as those treated at a public county hospital, were consistently high, with 97.5% of participants reporting communication of genetic test results to at least one blood relative. However, women with higher socioeconomic status may be more inclined to share test results, as hospital type was independently associated with communication of test results, and lower communication rates occurred at the public county hospital (Cheung et al., 2010). In addition to high rates of disclosure with family members, females do not limit communication to family
alone, as many women share their test results with friends, coworkers, and other acquaintances (d'Agincourt-Canning, 2001).

Interestingly, women also tend to share genetic health information regardless of their mutation status, i.e. a positive, negative, or uncertain genetic test result, and the majority of women do so within a few weeks of results disclosure (Cheung et al., 2010; Gadzicki et al., 2006; Hughes et al., 2002; Patenaude et al., 2006). In a prospective study of 77 female breast cancer patients, Vadaparampil et al. described the frequency of communication of genetic test results and determined dissemination of genetic information to family members and health care providers at 5 months post results-disclosure (Vadaparampil, Malo, de la Cruz, & Christie, 2012). As echoed in the literature, most probands communicated test results with family members regardless of mutation status, though the frequency with which they shared, and with whom they shared, varied by mutation status. Women with positive results shared with 100% of sisters and parents and with about half of brothers (Vadaparampil et al., 2012).

Though women’s rates of disclosure are high, women do not disseminate information equally within the family. A study of 273 BRCA mutation-positive women indicated that women were more likely to inform sisters of their genetic testing results than brothers (Patenaude et al., 2006). Moreover, test results were communicated to female family members more than male family members in all categories, i.e. mothers were told more than fathers, sisters more than brothers, daughters more than sons (Patenaude et al.,
Females were more likely to share with their father if he had a diagnosis of cancer, possibly because of increased communication regarding cancer, or perception that one’s father is more interested in cancer-related topics. Participants were more likely to share the test result with their brothers if the mutation was paternally inherited (Patenaude et al., 2006). This observation of more sharing with female family members is prevalent in the literature despite male relatives carrying the same risk for carrying a mutation, and is likely due to the increased cancer risks for female family members (Cheung et al., 2010; Finlay et al., 2008; Gadzicki et al., 2006; Patenaude et al., 2006; Vadaparampil et al., 2012). Although the feeling of responsibility to share genetic information remains, women may feel less urgency if they only have sons or when considering their male relatives without children (Dancyger et al., 2011). Males are also less likely to be included in family discussions about HBOC, even when they are identified as mutation carriers (Liede et al., 2000).

Other factors that influence the communication of genetic health information among women include whether women had a positive or negative experience with genetic testing and their degree of information retention (Vadaparampil et al., 2012). Satisfaction with a participant’s decision to undergo genetic testing and greater knowledge of screening and risk-reduction recommendations are correlated with increased communication of test results (Cheung et al., 2010). Furthermore, existing family culture and dynamics influence the decision to disclose results to family members, as individuals from families with a more open communication style are more likely to share this information than
families with more secretive or closed communication styles (Dancyger et al., 2011). Of note, cancer status of the individual communicating results is found to be unrelated to dissemination of result information (Cheung et al., 2010; Patenaude et al., 2006; Tercyak, Peshkin, DeMarco, Brogan, & Lerman, 2002).

Researchers have also investigated the barriers to sharing genetic information. Probands may withhold genetic risk information to relatives due to the relative’s perceived emotional well-being, ability to cope, and current life stage (Dancyger et al., 2011). Individuals may not share genetic health information when they believe it to be a burden on family members (Dancyger et al., 2011). Family members with strained relationships or estranged family members are less likely to be informed of test results, as to keep with established family dynamics. In general, communication about genetic test results adheres to pre-established boundaries and networks of the family (Dancyger et al., 2011).

Ideally, sharing genetic test results with relatives will empower them to make changes to cancer screening and medical management to allow for early detection and reduce cancer risk. It is also critical to share this information with healthcare providers in order for this goal to be achieved. A study aimed to assess communication patterns of 312 BRCA positive women with health care providers showed that the majority (72%) of women shared their genetic test results with an outside health care professional (i.e. a non-testing provider outside of the tertiary care center) (Ready et al., 2011). Women who did not have a personal cancer history were more likely to share than those with a personal
cancer diagnosis, possibly because these women were less likely to seek care outside of their oncology practice. In general, women shared most frequently with a primary care physician (70%), followed by an OB/GYN (61%), oncologist (42%), surgeon (23%), or other (7%). Most participants did not describe the dissemination of genetic test results to their outside health care providers as difficult, and about 80% of participants felt that their health care provider could appropriately answer questions and address their concerns. Mutation status was not a significant predictor of women sharing genetic health information with outside health care providers. This data suggests that women are willing to share genetic test results with their care providers and that they are satisfied with the discussions that take place with outside health care providers (Ready et al., 2011).

In one of the few male-centered studies, Hallowell et al. explored the communication around and dissemination of BRCA1/2 test results within families of at-risk men who underwent genetic testing (Hallowell et al., 2005). The authors interviewed 17 men, 8 of the men’s partners, and 4 adult daughters about their experiences with cancer and genetic testing, decision-making based on testing results, and communication of test results within the immediate family. The authors found that most fathers viewed the dissemination of their genetic test results as a shared responsibility between them and their partners. Though the initial discussion of genetic testing often took place between both parents, ongoing discussions were more likely to take place between offspring and their mothers. The majority of parents communicated with limited disclosure to their
children, often downplaying the father’s involvement while casually mentioning family cancer history and genetic testing. Parent’s justified their communication styles by considering how they perceived their children would cope and react to the discussion with their right to know the information for their own health. Adult daughters were more likely to be informed of their fathers’ intention to undergo genetic testing before their father’s initial testing than sons, and children deemed to be emotionally fragile at the time were more likely to be informed after testing. Reasonably, all of the parents interviewed expressed more anxiety about daughters’ risks than sons’ risks. None of the men in this study indicated that they were unwilling to communicate genetic test results and none of the parents perceived the disclosure of genetic test as problematic. However, many participants regretted the manner in which they disclosed this information, suggesting that more support may be needed in the communication of genetic information to family members (Hallowell et al., 2005).

Though many more women undergo genetic testing for HBOC than men, both sexes understand the implications of their result for family members, and available data indicates that men share their genetic information with at least one family member (d'Agincourt-Canning, 2001). d'Agincourt-Canning found that men often restrict communication of test results to spouses, children, and siblings (d'Agincourt-Canning, 2001). Less commonly do men share with emotionally close extended family members and/or their physicians. Men are specifically more concerned about their obligation to share information with daughters, granddaughters, and occasionally sons (d'Agincourt-
Canning, 2001). While women may offer up genetic information to family members, men are more likely to share information as a result of direct inquiry from family members, and report significant more difficulty with discussing positive genetic test results than women, particularly when faced with disseminating these results to other men, including fathers, brothers, and sons (Finlay et al., 2008; Stromsvik, Raheim, & Gjengedal, 2011). Furthermore, a study by Stromsvik et al. found that a common desire among 15 Norwegian male BRCA carriers was to keep genetic information completely private, often due to fear of social stigmatization and insurance discrimination (Stromsvik et al., 2011). Men are also less likely than women to share genetic information in order to obtain emotional support (Finlay et al., 2008).

Shiloh et al. compared 51 male mutation carriers with 30 men who tested negative for BRCA1 and BRCA2 mutations via telephone interviews, at a median of 4 years after genetic counseling result disclosure (Shiloh, Dagan, Friedman, Blank, & Friedman, 2013). Among other aims, the study measured participants’ testing-related distress. Results indicate that BRCA carriers were significantly more distressed and reported less positive experiences related to testing compared with non-carriers. This may be due to increased stress related to guilt and grief of passing a BRCA1 mutation on to children. Specific reasons for increased testing-related distress were not explored in this study, though indirect findings suggest that participant’s concerns about daughters’ health outweighed concerns about personal health (Shiloh et al., 2013). Moreover, Stromsvik et al. identified strong emotions in males post results disclosure, including feelings of
anxiety, sadness, loneliness, and unfairness. Due to strong emotions, men often used avoidance as a coping strategy in consideration to personal cancer risk, family cancer history, and psychological implications on everyday life (Stromsvik, Raheim, Oyen, & Gjengedal, 2009).

As HBOC is a condition that primarily affects females, communication patterns among family members, and the gendering of information sharing and receiving, may be a product of the disease natural history. Communication patterns among males and females differ in families affected by genetic disorders that affect both sexes equally (Shiloh et al., 2013; Stromsvik et al., 2011). Results from a retrospective cross-sectional qualitative study evaluating how information surrounding hereditary non polyposis colon cancer (HNPCC) was communicated and disseminated indicate that in the majority of families, probands share information with at least one first degree relative within two weeks of receiving the genetic test results, which is in parallel with HBOC (Stromsvik et al., 2011). However, both male and female probands took responsibility for conveying information to family members, with males assuming an active role in communication and encouraging family members to be tested (Peterson et al., 2003). Furthermore, in a study by Ormond et al. surveying 48 cystic fibrosis carriers with and without a family history of cystic fibrosis, gender was not a significant variable in the dissemination of information to relatives (Ormond, Mills, Lester, & Ross, 2003). Studies regarding the communication of genetic health information in disorders that affect males and females equally indicate that the burden of dissemination is shared more equally by males and
females than in families affected by HBOC (Shiloh et al., 2013; Stromsvik et al., 2011). Moreover, there appears to be less discrepancy over which gender is preferentially informed of genetic results (Shiloh et al., 2013; Stromsvik et al., 2011).

In this study, we questioned 21 men via semi-structured interviews to determine what factors influence sharing of genetic information among men with BRCA gene mutations with their family members and health care providers. We aimed specifically to: describe participants’ feelings and opinions about HBOC; ascertain participants’ extent of information sharing with family and primary care medical personnel; and to describe the needs of participants with regard to information and resources provided by genetic counselors and other healthcare providers.
Chapter 2: Methods

Upon approval by The Ohio State University’s behavioral institutional review board, an invitation to participate in a telephone interview was sent to the email listserv for Facing Our Risk of Cancer Empowered (FORCE), a hereditary breast and ovarian cancer patient advocacy group which aims to educate, involve, and aid individuals affected by HBOC, as well as support research dedicated to HBOC (www.facingourrisk.org). The invitation outlined the primary purpose of the study, the intent to examine the impact of HBOC from a male’s perspective, with particular focus on communication of family history and genetic test results. FORCE also posted a study advertisement on their social media pages. Interested individuals were directed to contact study coordinators to set up a date and time to perform the interview. Eligible participants were adult (18 years or older), English-speaking males with a personal or a family history of a BRCA gene mutation. Participants were provided with a $15 Target gift card upon interview completion.

A total of 21 participants (Table 1) were enrolled in the study. All study respondents (21) met eligibility requirements and consented to complete a semi-structured phone interview. This interview focused on family cancer history, experiences with cancer and genetic testing, motivations to pursue genetic testing and subsequently disclose genetic test results, and participants’ emotional support systems. Interviews were recorded and
transcribed in their entirety. Thematic analysis utilizing grounded theory was applied to 18 transcripts in which the men had undergone BRCA genetic testing and were known mutation carriers or were obligate mutation carriers. Individuals with unknown mutation status (n=3) were excluded from analysis. A codebook was developed in order to identify recurrent themes among the 18 transcripts. Codes were applied and modified until coverage of major themes was reached across all 18 transcripts. A total of three coders helped to apply and modify the codebook: one genetic counseling student (AS), one practicing genetic counselor (LS), and one undergraduate student with a background in communication and qualitative data analysis (JB). Both AS and LS underwent training for thematic analysis utilizing grounded theory by an experienced qualitative researcher in the department of Communications, and JS was employed as an undergraduate researcher under this same individual at the time of data analysis. Two coders (AS and LS) independently coded 5 transcripts and discussed differences in interpretation until 100% agreement was met and the codebook modified. Subsequently, JB and AS applied the modified codebook to two different transcripts in order to ensure suitability of codes to the transcripts. One hundred percent consensus was met and the codebook modified accordingly. The final codebook was then independently applied to 18/21 interviews by AS and LS until 80% inter-coder reliability was achieved. Thematic analysis was undertaken utilizing Nvivo, a qualitative software analysis tool that enables the user to identify recurrent themes among a data set and run queries comparing common codes among sub-groups and demographic categories.
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<tr>
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<tr>
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</tr>
<tr>
<td>51-60</td>
<td>3</td>
<td>16.6%</td>
</tr>
<tr>
<td>61-70</td>
<td>7</td>
<td>38.8%</td>
</tr>
<tr>
<td>71-80</td>
<td>6</td>
<td>33.3%</td>
</tr>
<tr>
<td>80+</td>
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<tr>
<td>Graduate/Professional/PhD</td>
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<td>66.6%</td>
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<td>8</td>
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</tr>
<tr>
<td>Retired</td>
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<tr>
<td>Married</td>
<td>17</td>
<td>94.4%</td>
</tr>
<tr>
<td>Widowed</td>
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<th>Time Since Genetic Testing (years)</th>
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<tr>
<td>&lt;1</td>
<td>1</td>
<td>5.55%</td>
</tr>
<tr>
<td>1-3 years</td>
<td>3</td>
<td>16.6%</td>
</tr>
<tr>
<td>4-7 years</td>
<td>7</td>
<td>38.8%</td>
</tr>
<tr>
<td>8+ years</td>
<td>5</td>
<td>27.7%</td>
</tr>
<tr>
<td>Obligate Carrier</td>
<td>2</td>
<td>11.1%</td>
</tr>
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</table>

<table>
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<tr>
<th>Genetic Testing Provider</th>
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<tbody>
<tr>
<td>Genetic counselor</td>
<td>10</td>
<td>20.8%</td>
</tr>
<tr>
<td>Primary care provider</td>
<td>2</td>
<td>11.1%</td>
</tr>
<tr>
<td>Oncologist</td>
<td>2</td>
<td>11.1%</td>
</tr>
<tr>
<td>OB/GYN*</td>
<td>1</td>
<td>5.55%</td>
</tr>
<tr>
<td>Obligate Carrier</td>
<td>2</td>
<td>11.1%</td>
</tr>
<tr>
<td>Unknown</td>
<td>1</td>
<td>5.55%</td>
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<table>
<thead>
<tr>
<th>Offspring</th>
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<th></th>
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</thead>
<tbody>
<tr>
<td>Daughters Only</td>
<td>4</td>
<td>22.2%</td>
</tr>
<tr>
<td>Daughters + Sons</td>
<td>13</td>
<td>72.2%</td>
</tr>
<tr>
<td>Sons only</td>
<td>1</td>
<td>5.55%</td>
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Continued
Table 1 continued

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<thead>
<tr>
<th>Personal Cancer Dx</th>
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<tbody>
<tr>
<td>No Cancer</td>
<td>13</td>
<td>72.2%</td>
</tr>
<tr>
<td>Breast</td>
<td>2</td>
<td>11.1%</td>
</tr>
<tr>
<td>Prostate</td>
<td>3</td>
<td>16.6%</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Family HBOC Cancer History**</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>One affected FDR</td>
<td>3</td>
<td>16.6%</td>
</tr>
<tr>
<td>&gt; Than 1 affected FDR</td>
<td>1</td>
<td>5.55%</td>
</tr>
<tr>
<td>Only SDR’s or more distant affected</td>
<td>2</td>
<td>11.1%</td>
</tr>
<tr>
<td>1st and 2nd degree/distant affected</td>
<td>12</td>
<td>66.6%</td>
</tr>
</tbody>
</table>

*Sought consultation for testing after having heard OB/GYN speak at a conference
**HBOC cancers included breast, ovarian, prostate, pancreatic, and melanoma
***Demographics not including individuals excluded from data analysis (n=3)
Chapter 3: Results

Primary themes identified reflect our stated aims to describe participants’ feelings and opinions about HBOC; ascertain participants’ extent of information sharing with family and primary care medical personnel; and to describe the needs of participants for information and resources provided by genetic counselors and other healthcare providers. Additional secondary themes emerged upon analysis as well and include: Factors that influenced the decision to undergo genetic testing, and perceived implications of genetic testing.

Emotional Impact of HBOC

Regarding the first major aim of the study, when discussing their family histories of cancer and the personal emotional impact of that family history, 8/18 individuals mentioned that their family history caused them to be most concerned for female family members. In discussing concern for female family members, participants mentioned daughters and granddaughters in particular; sisters or mothers were not mentioned. One participant with both a daughter and a son recounted a conversation he had with his son about their family cancer history and his risk for a \textit{BRCA} mutation; “I was really trying to reinforce with my son because I want him to be in a position to give this information to his daughter, who just turned 18.”
Upon results disclosure, 7/18 men mentioned that they felt fear for their other family members who may also have an increased risk for cancer. Fear was not related to personal cancer history. When fear for other family members was coded, most of the participants explained that they were specifically fearful for their children.

Another common emotion upon results disclosure was sadness, with 5/18 men mentioning being sad. Four of the individuals that mentioned sadness also mentioning fear for family members. When men described why they were sad, none of them mentioned that it was because of new knowledge of their own increased cancer risk, with one man commenting:

Yeah, no I was saddened by it because I have children, otherwise it wasn’t necessarily, “oh my god what’s going to happen to me,” because I was pretty quickly informed about what it means from a male perspective, and once again I knew that I was not the typical male in that I never go to the doctor, no, I go to the doctor.

Many (8/18) participants mentioned that they felt guilty for passing a BRCA mutation on to their children. This was not driven by the gender of the participants’ children—men mentioned feeling guilty about passing the mutation on to both their sons and their daughters, with one man explicitly explaining, “I feel very guilty that I passed this on to my daughter and son.” However, daughters were mentioned more frequently, even
though most participants had both daughters and sons. Of the eight individuals that mentioned guilt, six of them had both daughters and sons, and two of them had only daughters. One participant with both a daughter and a son elaborated on this topic:

I felt uh terrible of course. Now that uh, because of the risk that it would involve for not only my son but especially my daughter, and of course when she found out she was also BRACA2 positive, that’s one of the things that a father doesn’t necessarily want to pass down to his children.

A minority (4/18) of men felt neutral upon results disclosure. When “neutral” was coded, most men explained that the result was not a huge deal for them and was not a surprise. Personal cancer history and family history of cancer did not influence this.

Five participants mentioned that they didn’t need emotional support from a health care professional when they received their results. Three of these five individuals also did not feel that they would personally benefit from a support group. When this was coded, men discussed that they didn’t feel like they needed to talk about it, didn’t really have any questions, and that they could deal with this on their own, as long as they had the factual risk information necessary. Twelve men mentioned that their primary support person was their spouse. Other support systems included participants’ daughters specifically (n=3) or family in general (n=4), with one participant stating the following:
I don’t know if our family is unique or not but, I could tell you that with the exception of one person in our family, we have all wanted to know and haven’t responded adversely to the news, and we’ve been very supportive of one another.

Eleven participants mentioned that they would be interested in a male-focused support group. Many mentioned that having support of other men, at least for those newly diagnosed with a \textit{BRCA} mutation, would be helpful. They felt that gaining information, secondary to talking to other men about HBOC and seeing how they coped with it, would be the primary purpose of a support group. One individual explained:

Oh I’d be curious about it. It probably would be helpful, the reason why because what response, if any, would they have? How concerned would they be? And of course the natural part of any group is not only support but education.

Four participants mentioned that a support group would be more helpful for others than it would for themselves. When this was coded, men mentioned that they wouldn’t need the group personally, but that they would be willing to help others who did. One participant elaborated, “I’m not interested, if I could provide support to someone else I would do it, but it doesn’t keep me from doing anything and it doesn’t limit me in any way so I don’t feel like I need support.” One individual mentioned that a support group would be helpful for himself, but would also be more helpful for others, and is included in both categories. Moreover, four individuals did not feel like a support group would be helpful to them at all, citing that they didn’t really need emotional support, and that they weren’t sure what kind of information they would exchange that would be particularly helpful.
When asked about how HBOC affected their life overall, increased awareness was a major theme, with 11/18 mentioning this. Seven of the men that mentioned having increased cancer awareness due to a positive result also ended up changing their cancer screening practices. One participant said the following:

[having a BRCA mutation] makes me be a little bit more sensitized, and being more careful about certain things. It has affected me in that way, by that I mean I’ve always been, I’ve always taken preventative proactive measures in my health care, so it really hasn’t changed that much at all. The only significant change is not only the level of my awareness and sensitivity to these cancers and to my possible, being a little bit higher probability.

Many of the men explained that they weren’t seriously concerned about getting cancer and that cancer wasn’t something they thought about often, but were just simply more aware of their increased risk to get cancer. A different participant explained that “It’s just a question of awareness, I don’t have any real serious concern about getting cancer.” Of the four men that mentioned increased awareness but did not change their screening based on their genetic testing result, only one of them was already practicing increased screening due to a prior diagnosis of breast cancer.

Nine men mentioned that HBOC had a negligible impact on their life overall, meaning that their BRCA mutation was simply something that had been integrated into their life and that they’ve been made aware of, but that it didn’t really affect their day to day
functioning. One participant mentions “you know it’s not like I’ve got the sword of cancer hanging over my head, good grief it’s been there for a while before I had the genetic testing (laughs) so it hasn’t added any stresses or anxiety to me.” Similarly, another participant explained that his BRCA mutation is “just something [he] carr[ies],” with a third individual explaining that his life is relatively normal, despite having a BRCA mutation “I wouldn’t say it’s been earth shattering, you know I travel, I have a normal life.”

Although having a BRCA mutation did not have an outstanding impact on most of the men’s lives, seven men mentioned that HBOC has impacted them in an emotional sense, due to having close family members affected by cancer or having an increased risk of getting cancer. One individual explains HBOC has affected his life:

> It has changed, it has changed everything I once thought was solid, into chaos. I was dumb enough to think that I was going to live forever, my sister was going to life forever, my parents are going to live forever. And what this has done has explained to me in a very hard lesson that none of that is true. That it is um, yeah, that none of that is true. I think that’s the best way to put it.

Similarly, another participant explains that HBOC has affected him in “just the emotional” ways, due to “seeing [his] sister and [his] brother suffer, two brothers suffer really, [he is] just very very aware of cancer now.” Family history of cancer did not seem to impact participants’ perceived personal impact of HBOC.
**Extent of Information Sharing**

In relation to the second major aim of the study, most men (13/18) shared genetic test results first with their wives, and seven participants shared results second with their children, regardless of gender. One participant shared with his daughter only, even though he also had a son.

Of the 17 men who had daughters, all participants except one shared their *BRCA* result with their daughters, and of the 13 participants who had sons, all except one shared their *BRCA* result with their sons. The individual that did not inform his daughters of his *BRCA* result didn’t because he felt his daughters, who are young adults, knew about his mutation from other family members, specifically their cousin. The individual that did not inform his son of his *BRCA* result withheld this information because his son is under the age of five.

Of the 16 men that shared genetic test results with their children, eight mentioned that discussing HBOC and their genetic test results with their children was not difficult, and eight mentioned that the discussion was not emotional.

Participants mentioned that discussions were pragmatic and primarily focused on information sharing so that their children had the information they needed to make conscious health decisions. One individual explained that “they were not emotional, they
were just matter of fact you know you need to set up and go get tested.” Another participant elaborated:

No I, with both my children I’ve always talked very openly and very much on an equal level with them even when they were smaller. So no I think we…the subject itself is uncomfortable, having the discussion between us is not.

Of the four men that mentioned that the discussion with their children regarding HBOC was emotional, they most often cited concern and fear for their children as the cause of the emotion. One participant explains, “they are very emotional, they’re no longer as difficult as they were initially, I’ve got so many fears that I don’t want to just dump the whole thing on her, there’s stuff I’m afraid of.”

A major theme among study participants was open sharing. When this was coded, men often meant that they were not embarrassed by their BRCA mutation, with one man stating “I mean there is no shame attached, what are you going to do?” and another man stating “Yeah, some people have asked if I’ve had genetic testing, you know people that know what my family has gone through, and I’m pretty open about it, I don’t feel embarrassed by it or anything like that.” Moreover, men felt they would discuss it with anyone if it came up in conversation, and that they didn’t limit information sharing. One individual commented, “I found that I could tell people. Friends, and family and relatives, of course once you tell a relative you know that all of your relatives are going to know pretty soon, but uh, I felt better carrying it,” with another individual mentioning he
tells anybody who is interested, “Anybody who asks. If cancer comes up in conversation, its anybody who, that’s who I’ve told.”

Eight men mentioned leading the discussion about their genetic test results and HBOC primarily with their children, but also with other family members. For men that lead these discussions, most explained that while their wives were aware of the conversation, it was important for the men themselves to initiate these conversations. Men took ownership over their genetic health history and result information and wanted to disseminate that information first hand. When asked whether he or his wife primarily participated in discussions with their children, one participant stated the following:

Um, no my wife knows what’s going on, but I have the discussions mostly with my daughter, there’s nothing that I can discuss with my daughter that would embarrass me, it probably would be a little bit more touchy with her, but I think that she’s becoming more accustomed to these discussions.”

Another participant explained, “You know when my child asks me about my family, I don’t want them to be distracted by my wife.”

Conversely, three individuals mentioned that their wives primarily discussed their BRCA result with their children. When participants mentioned this, they either explained that they have daughters only and felt like their wife could relate more to them as a female, or felt their wife was more equipped to handle the discussion due to a medical or counseling background. One participant explained, “well I think being a woman that there is more of
a connection there,” and another explained that, “because it deals with your personal condition, there is always a possibility of death, and sometimes the concept of discussion between a man and a woman, some of the details can be uncomfortable to talk about as well.” One participant informed his daughter of his results initially, then subsequently made an appointment with his daughter’s gynecologist to ensure that he was aware of the situation:

So my ex and I went to our daughter’s gynecologist, and initially he said “well I can’t share anything, HIPAA, blah blah blah” and I said “no, no no, no, this isn’t about HIPAA, this is me sharing with you, not you sharing with me, I want you to know our family history.

After initially telling his daughter about his BRCA mutation, he mentioned that his ex-wife now has more ongoing discussions with his daughter, because he feels she can relate better given they are the same gender. He was open to discussing this with his daughter, but limited these discussions to more genetic based facts:

So in that sense I think my daughter is old enough to recognize that dad is going to be the most informative genetically, you know as far as decisions as far as surgeries and things like that, you know I’m part of the mix but that’s when other parties get involved which is fine.

Eight participants mentioned that the reason they shared their BRCA results with family members was mainly for family members’ own knowledge. Participants mostly discussed that they shared result information so that other family members could take action for
their own health, gain family history awareness, and because family members had a right to know the information. One participant explained, “the hope is that subsequent generations have more information available to them so that they can make more informed decisions,” while another explain that “I did mention it to immediate family members because I thought they had a right to know or need[ed] to know.” No major barriers to sharing information were identified.

Seven individuals mentioned that discussions about HBOC in their families were typically triggered by an event, meaning that the family no longer discussed HBOC frequently, but that discussion would be prompted by new events. Events included newly-published studies or articles, cancer-related deaths in the family, or HBOC-related medical procedures, like a daughter having prophylactic surgery, or a father obtaining a mammogram. Six individuals mentioned discussing HBOC with their daughters most frequently, particularly about involvement in advocacy groups like FORCE, about novel medical information, or regarding their daughters’ cancer screening procedures.

While men were open to sharing *BRCA* result information with family, friends, and health care providers, most men (12) did not know another man outside of their family with a *BRCA* mutation. Four men stated that they did know other men with HBOC, with two of the four attributing this to a support group they participate in.
Expressed Needs to Be Met by HCPs

The third major aim of this study identified needs expressed by participants to be met by health care professionals. The majority of men (12/18) shared their BRCA test result with their primary care physician. Of the individuals that had cancer, 2/4 shared their test result with their oncologist and the other 2/4 had genetic testing through their oncologist, thus their oncologist already knew of their results. Other physicians shared with include psychiatrist (1), Agent Orange physician (1), and urologist (1), and three men mentioned that they share this information with “every single doctor” they see.

The most commonly mentioned theme across the study overall was that the medical field as a whole needs knowledge and awareness about the implications of HBOC for males. This was mentioned 39 times, across the majority (14/18) of transcripts. Men were most interested in promoting awareness surrounding male risks, with one participant explaining, “So to me that would be the greatest thing besides the actual research. Awareness. Awareness, awareness, awareness.” Another participant elaborated:

I just don’t think, no I don’t feel over shadowed, I just don’t think, again its human nature, 90% of people affected are women by the condition, they’re definitely going to get the most attention, that’s fine, I just hope that they don’t overlook the men when they are spending so much time on the ladies, that’s all, I understand what’s going on, now I think awareness needs to be raised, that’s all.
Along with awareness, men mentioned the need for more readily available information and educational outreach specifically for male risk, often in the form of public service announcements and HBOC support groups, because “the more information brought to the public’s attention, the better.” One individual, when discussing an informational poster about HBOC, said the following:

The only thing it said about men was men can be carriers. And you know I felt that um, that they were way underplaying that and that what they should be having is something in there that says men uh, that both genders are at higher risk for you know melanoma, pancreatic, you know etc., and uh women for breast and ovarian at much higher rates, and men for breast and prostate cancer. I mean I think they should in their poster, I thought that they had really relegated us to second-class status.

While many of the men acknowledged that HBOC is a disease that primarily affects women, some individuals expressed frustration for a lack of an established system for treatment and screening, with one individual explaining a conversation he had with his medical team, “there was a lot of shrugging of shoulders and saying ‘we don’t know’. And there are a lot of regimens put in place for women but we don’t know whether any of these work for men.” The men who implemented mammography as a screening practice also expressed frustration over mammography equipment and medical centers being tailored to females.
An additional concern relating to our third primary aim among this cohort was the expressed need to educate health care professionals, specifically primary care physicians and urologists that participate in ongoing follow up with male patients, about male risk. One individual explained that he “talked with various doctors…and they didn’t seem to know a lot about \([BRC\text{A}]\)…particularly for men, and not understanding what the risks are and what needs to be done” in order to minimize those risks. Another participant elaborated with the following statement:

I think men’s doctors don’t pay as much attention to the men as they do the women, um, with the gene. I think they are very sensitive to women’s issues surrounding the gene, I don’t think that they are as sensitive to men’s issues with the gene.

As a whole, when describing the need for awareness and the need for knowledge, men described the need for a collective effort among health care professionals to disseminate more information to the public and sensitize the health care field to male risks and male concerns.

Along with needing knowledge and awareness, 6/18 individuals mentioned the need for better understanding of ethnic background and family history, particularly for Ashkenazi Jewish individuals. Men mentioned that they were prompted to fill out family history questionnaires at their regular health care appointments, but that even when a family history of cancer was noted on the form, no further discussion ensued. Moreover, two
men mentioned that their health care professional was not aware that a BRCA mutation could be passed on to offspring through a male at all. One participant said the following:

They ask about your family history, you know has anybody in your family died of cancer or suffered from cancer? And then that’s it, there’s nothing further. But there is a responsibility to carry that a few more steps if you do know, do you have [a] mutation, have you been tested?

Likewise, seven individuals felt that they needed to personally educate their HCP about male HBOC risks, and that they didn’t feel their provider knew how to follow or manage their cancer risks. Furthermore, men mentioned that they needed to actively pursue care, because “It’s not an automatic thing for men, even when you make it very clear to your physician that you’ve tested positive.” One participant explained “Eh, you know, I understand this is uh kind of Never Never Land for them, they just don’t know how to react” when prompted about his encounters with his physicians. Another participant, when discussing his physician, said the following:

It was very clear, he made it very clear to me that with the knowledge that I had the gene, he did not have the tools or the knowledge to make recommendations or even to follow the progress of having that gene, and the effects it might have on me.

While more individuals mentioned that their health care provider needed to be educated about male risk regarding HBOC, four participants felt that their health care provider was educated about their risks, could answer their questions about HBOC, and could manage
them based off their risk as well as direct them towards new studies. One participant explained the general attitude of his health care providers as, “we know that there aren’t many of you, but however many of you there are, we are paying attention to you because it matters.”

Genetic testing was primarily facilitated through a genetic counselor (10/18), with other health care providers like primary care physicians (2), oncologists (2), and OB/GYN’s (1) ordering the test for the rest of participants. Of the individuals who received genetic testing via a genetic counselor, nine mentioned feeling that the genetic counselor was aware of male HBOC cancer risks. Moreover, 5/10 of these individuals felt that the information their genetic counselor provided was helpful to them. Four out of ten individuals who had genetic testing facilitated by a genetic counselor forgot whether the genetic counselor provided them with specific information pertaining to male impact and risk in the session. A total of seven individuals, regardless of testing provider, mentioned that they were provided with helpful information with their result, and a total of six individuals, regardless of testing provider, mentioned forgetting the information that was provided. Elapsed time since genetic testing was not a predictor of individuals that forgot information provided with their result, with time since genetic testing ranging from six months to twelve years.

In regard to the secondary findings in this study, the primary reason participants underwent genetic testing was due to concern for their children’s potential cancer risk.
This was mentioned by 13/18 participants. Four of these individuals mentioned being concerned for their daughters *specifically*, even though they also had sons. Personal cancer risk was not a major concern among study participants, with only 3/18 individuals mentioning this as a primary reason for being tested.

Although most individuals did not mention personal cancer risk as a primary reason to undergo genetic testing, 7/18 individuals felt that their positive result was important to their health, that it was integral to their medical care, and/or that the result helped sensitize them to possible risks. For individuals that mentioned the result as important to their health, the majority (6/7) of them did not have a personal history of cancer. All individuals with just one affected first degree relative believed that their genetic test result was important to their health, though family history was not a significant predictor of perception of importance among the other family history categories.

The majority of men (10/18) changed their cancer screening when they received a positive genetic testing result, and one additional individual attempted to change screening after receiving a positive test result. This individual described that he had not yet changed his screening because his primary care physician mentioned that it was not necessary, explaining “…my primary care said no, so what I’m doing is I’m making these appointments with these various specialists [to obtain screening] on my own.” On the other hand, 7/18 men did not change their cancer screening routine after learning of their *BRCA* mutation. Two of these individuals did not change their screening routine because
they were already participating in increased screening, as they both had histories of breast cancer. With this considered, 5/18 men had not changed cancer screening based on their positive BRCA mutation status and were not already participating in elevated screening programs prior to genetic testing.
Table 2: Quotes Illustrating a Subset of Primary Themes

<table>
<thead>
<tr>
<th>Theme</th>
<th>Quote</th>
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<tr>
<td>Concern for female family members</td>
<td>“I was really trying to reinforce with my son because I want him to be in a position to give this information to his daughter, who just turned 18”</td>
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<tr>
<td>Interest in male focused support groups</td>
<td>“Oh I’d be curious about it. It probably would be helpful, the reason why because what response, if any, would they have? How concerned would they be? And of course the natural part of any group is not only support but education”</td>
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<td>Increased personal awareness</td>
<td>“It’s just a question of awareness, I don’t have any real serious concern about getting cancer”</td>
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<td>Overall negligible impact</td>
<td>“I wouldn’t say it’s been earth shattering, you know I travel, I have a normal life”</td>
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<tr>
<td>Pragmatic discussions with children</td>
<td>“They were not emotional, they were just matter of fact you know you need to set up and go get tested”</td>
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<td>Open sharing</td>
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<tr>
<td>Initiative with dissemination</td>
<td>“You know when my child asks me about my family, I don’t want them to be distracted by my wife.”</td>
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<td>Sharing to inform</td>
<td>“The hope is that subsequent generations have more information available to them so that they can make more informed decisions”</td>
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<tr>
<td>Need for increased awareness</td>
<td>“So to me that would be the greatest thing besides the actual research. Awareness. Awareness, awareness, awareness.”</td>
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<td>Need for further HCP education</td>
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<tr>
<td>Need to pursue follow up care</td>
<td>“It’s not an automatic thing for men, even when you make it very clear to your physician that you’ve tested positive.”</td>
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Chapter 4: Discussion

HBOC is a condition that confers significant cancer risks that are most dramatic in females. Males are affected by HBOC as well, though, and cancers associated with BRCA gene mutations in males have been well described. Less well-described are the psychological effects and communication patterns among men with HBOC, and perceived preparedness of healthcare providers when managing the care of males with HBOC. This study is one of the few to explore these themes in men and serves as a pilot for future study development.

As a primary aim, we found that there was a general trend toward external focus of concern among this cohort. The majority of men expressed concern for female family members as being a driving factor for undergoing genetic testing. In addition, family cancer history led men to be most concerned about female family members’ risk. This is congruent with previous studies where findings suggest that men may be more concerned by daughters’ health risks than their own (d'Agincourt-Canning, 2001; Shiloh et al., 2013). Due to the dramatically increased cancer risk for females, this concern is appropriate and suggests that participants likely had at least baseline knowledge of the female-specific cancer risks associated with HBOC. This study, however, did not evaluate accuracy of knowledge.
When asked directly, men in the study did not generally express concern for their own personal cancer risk. This seems in conflict, however, with the fact that the same men subsequently integrated their result into their medical care and that it increased their need for more male HBOC knowledge and awareness in the health care system.
awareness about cancer risks. Men who described little concern about their own risk of developing cancer utilized the genetic information to modify cancer screening. Most participants felt that they received adequate counseling about male risks upon result disclosure, possibly indicating that education tailored to men is sufficient to motivate health behavior changes and increase awareness about male risks, even if this was not initially a primary motivator. We did not explore personal cancer worry at multiple time points and it is possible that participants were initially concerned, but that their concerns were alleviated as a result of increased screening. It is also possible that participant’s minimized adverse feelings about personal cancer risk, as has been reported previously (Graves et al., 2011; Stromsvik et al., 2009). Even with these possibilities considered, nearly all participants expressed primary concern for their children throughout multiple lines of questioning.

While seven individuals felt that the information they were provided with during results disclosure was helpful and included risks pertaining to men specifically, six of these individuals forgot any information pertaining the impact of BRCA on men and/or increased cancer risk that was provided. Time since genetic testing did not influence forgetfulness, which ranged from six months to twelve years. As healthcare providers, we should be invested in strategizing ways for our patients to retain this important information.
Support groups could serve as a critical venue for this message reinforcement over time, particularly since men did not desire increased support from health care providers. In fact, the majority of men in this study (11) stated that the availability of a male-centered support group would be beneficial, primarily for the purpose of obtaining information about updated cancer risks, screening practices, and/or new studies pertaining to BRCA carriers. Participants also expressed that emotional support would be beneficial, albeit to a lesser extent than informational support. This is similar to findings by Finlay et al. that indicated that men are less likely than women to share information in order to obtain emotional support (Finlay et al., 2008).

Common themes related to the primary aim of dissemination patterns emerging from this data indicate that men may take more of an active role in sharing information with family members than previously thought. Particularly in regard to sharing information with children, men felt it was their duty to disseminate information, and they actively took initiative to do so. Similar to findings reported in the literature about why men communicated genetic information, men felt that they were responsible for communicating their genetic health information to family members because family members had a right to know the information, and in order to facilitate proactive health care measures for female family members (Dancyger et al., 2011; Wiseman et al., 2010). Communication of genetic test results did not appear as a “gendered activity” in our study, with other females in the family or spouses assuming initiative in discussing men’s genetic test results, as described extensively in past literature (d'Agincourt-Canning,
Moreover, men in this study did not communicate with daughters or female relatives more than male relatives, which is a pattern that has been described at length in past literature pertaining to female communication patterns (Cheung et al., 2010; Finlay et al., 2008; Gadzicki et al., 2006; Patenaude et al., 2006; Vadaparampil et al., 2012).

Men in this cohort communicated first with their spouses, and second with both male and female children. This indicates that men view their spouses as a primary support system, which has been described previously in the literature (Stromsvik et al., 2009; Stromsvik et al., 2011). In addition, men were more likely to disseminate information across both genders, although concern was preferentially expressed for daughters and ongoing discussions often occurred with daughters more often than sons.

Men in this cohort reported open and active sharing and communication styles, which conflicts with findings by Finlay et al. that indicate men are more likely to share genetic health information due to inquiry from other family members (Finlay et al., 2008) As previous research indicates, individuals and families with open communication culture are more likely to share genetic result information with other family members (Dancyger et al., 2011), and if applied to the present study, may indicate that men were more likely to share equally across genders and to actively participate in the dissemination process due to previously constructed open communication patterns within their families.
Along with employing open and active communication styles, men in this cohort commonly reported that discussions about HBOC with children were not difficult or emotional to have, even though eight individuals mentioned parental guilt. Family discussions adopted a more fact-based style and preventive focus, whereas Finlay et al. found that men reported much more difficulty discussing positive genetic test results than women did (Finlay et al., 2008). Inconsistencies regarding communication patterns among men in previous studies may be explained by improved inclusion and awareness pertaining to men and HBOC in recent few years, or may be a product of cohort bias in the present study, which consisted of men who are actively engaged in FORCE. Further research aimed at clarifying these discrepancies would be useful.

While men generally communicated openly with family members, the majority of men in this study did not know any other men with a BRCA mutation, despite being involved in the FORCE community. As described previously, Cheung et al. found that women of higher socioeconomic status communicated genetic result information to others more often than individuals of lower socioeconomic status (Cheung et al., 2010), and d'Agincourt-Canning found that females are more inclined to share their test results with friends and acquaintances outside of their family unit (d'Agincourt-Canning, 2001). Based on these data, it could be expected that our relatively homogenous cohort of highly educated men would describe frequent extra-familial communication of genetic testing results, but this did not emerge as a major interview theme. There are many possible
reasons for this, which were not directly investigated but could include lack of comfort in sharing \textit{BRCA} result information with people outside of their family.

Similarly to females described in the literature, with regard to communication with healthcare providers, men frequently shared genetic testing results with primary care physicians. However, in one study Vadaparampil et al. described that 80\% of female participants felt that their primary care physician could adequately answer their questions surrounding HBOC and facilitate appropriate medical management (Vadaparampil et al., 2012), whereas a common theme in the present study was that men needed to educate their HCP’s about male \textit{BRCA}-related cancer risks and screening recommendations. Furthermore, men commonly mentioned that their HCP’s didn’t understand implications of a paternal family history of HBOC related cancers. Interestingly, for one of the three individuals who had not undergone genetic testing, and therefore was not included in the overall thematic analysis, lack of HCP knowledge about male family cancer history risks and the genetic testing process acted as a barrier for him receiving genetic testing, even though he actively pursued this through his HCP. Coordination of educational outreach aimed at HCP’s who routinely follow men may be beneficial.

Our final primary aim regarded needs to be met by HCP’s in reference to male \textit{BRCA} carriers. The most prevalent theme that emerged from this research was the nearly unanimous agreement that female risks should be the primary focus of HBOC related education, but that there needs to be better inclusion of male risk in relation to general
public and health practitioner awareness. Given that this patient population was identified through a cancer advocacy group, it is interesting that increased awareness remains a prevalent concern, as this group of men is likely skewed toward being more involved and educated with regard to HBOC and available resources for the HBOC community. It is possible that this issue is even more prevalent than depicted in this study among men that do not have familiarity with any sort of advocacy group. As one individual described, men may feel that they have been “relegated to second class status.” Moreover, these men expressed discomfort in obtaining some of the recommended cancer screening like mammograms, due to the inherent feminization of these procedures. It is possible that such feminization could deter men from further pursuing preventative care for HBOC.

This study provides important insights into psychosocial impacts, communication patterns, encounters with health care practitioners, and expressed needs of males with HBOC. However, some study limitations should be noted. This study included a small number of participants that were identified through the FORCE network. This study is vulnerable to self-selection bias among the sample population, particularly because participants were notified via the study invitation about study aims in regard to communication patterns among men with HBOC. This may have inherently limited our cohort to men who disseminate this information in the first place, and excluded men who do not discuss HBOC with their family members and/or health care providers. In addition, the study cohort was largely homogenous, comprising of highly educated
Ashkenazi Jewish men. Ashkenazi Jewish individuals have an increased risk of carrying a BRCA mutation based on ethnic background and have contributed widely to genetic research involving BRCA in the past. These individuals may have differing concerns and behaviors specific to their culture and background. Thus, study findings may not be generalizable across a broader population of men with BRCA gene mutations.

Information such as family history and family communication patterns, BRCA mutation status, screening behaviors, and health care provider interactions were self-reported and were not verified with documentation. It is possible that recall of this information may be inaccurate or misrepresentative of actual events. Due to the thematic nature of this research, statistical analysis was not implemented, leading to the absence of statistical significance regarding correlation between demographics and study responses. This study serves as a pilot study for future research. Further research aimed at expanding on these themes would be valuable.
References


Appendix A: Interview Guide

Thank you for agreeing to participate in this interview. Your responses will help us understand how men affected by breast and ovarian cancers in their family share genetic health information with others. I appreciate any and all information you may be able to provide, however, if you would like to skip a question you may at any time. As a reminder, we are recording this call so that I may refer back to your answers later.

Are you ready to continue? Great!

1. First, I’d like to hear about your experience with breast and ovarian cancer in your family.
   a. Who in your family has had cancer?
   b. Have any men in your family had cancer?
   c. Tell me about what the diagnosis was like for them. Did your relative(s) have a long battle?
   d. Were you a direct care provider for any the people in your family who have had cancer?

2. How do you think the cancer in your family has affected you, personally?
   a. Emotionally?
   b. Do you think the cancer in your family affects your risk of getting cancer someday?
   c. How concerned are you about your family history of cancer? Very concerned, a little concerned, not concerned at all?

   [If not disclosed previously]

3. Have you yourself ever had a diagnosis of cancer? If so, please tell me about it.

   [If not disclosed previously]

4. Have you had cancer genetic testing?
   a. If yes: did you test positive for a mutation?
      i. Were you the first person in your family to have genetic testing?
      ii. How long ago did you have genetic testing?
      iii. What was your primary reason for having genetic testing?
      iv. Were there other reasons that you chose to have genetic testing?
      v. When you had genetic testing, who did you go to in order to have it done?
      vi. Do you remember what they told you when you got your result?
1. Were you on the phone or in person?
2. How did receiving your result make you feel?
3. Who did you tell first about your genetic test result?
4. Who else have you told?
5. What are people’s reactions when you tell them about your genetic testing experience?
6. Did you feel that the person providing you with your result had information for you about the impact of the result on males, specifically?
   a. Tell me more about that.

[if positive for BRCA mutation ONLY]
7. Has your cancer screening changed since you found out about your result? In what way?
8. Do you know any other men outside of your family with HBOC?
9. How has having this condition in your family affected your life?
10. Would you be interested in more male focused support groups?

11. Let’s talk about sharing family history information with your primary care physician.
   a. Have you been asked about your family history of cancer?
   b. Do you share information about the females with breast cancer or ovarian cancer in your family?
   c. Has your primary care physician commented on your family history of cancer with regard to your risk?
   d. Has he/she asked if you had genetic testing?

6. Have you had cancer genetic testing?
   a. If no: has anyone in your family had genetic testing?
      i. If yes: did they have a cancer gene mutation?
         1. Have you thought about genetic testing?
         2. What are your reasons for not having genetic testing?
      ii. If no: have you or your relatives considered it?
         1. What are your reasons for not having genetic testing?

   b. Let’s talk about how sharing family history information with your primary care physician.
   c. Have you been asked about your family history of cancer?
d. Do you share information about the females with breast cancer or ovarian cancer in your family?
e. Has your primary care physician commented on your family history of cancer with regard to your risk?

7. Tell me about your typical cancer screening routine.
   [ask specifically about breast, prostate, colon, skin screening if not automatically disclosed]

8. Let’s switch gears a little bit. What type of discussions do you have with your family about your family history of breast and ovarian cancer?
   a. Do you have children?
      i. Have you discussed your family history with them?
   b. If you discuss your family history of cancer with your family members, do you usually bring it up or does someone else?
   c. What are those discussions like? Are they difficult/emotional? Is it something you mention frequently?
   d. Who have you discussed your family history with? Daughters? Sons? Siblings?

9. How important do you think your family history is to your medical care? Very, a little, not important?

10. Where do you think the medical community could improve in dealing with male patients that either have a BCRA mutation or a family history of breast and ovarian cancer?

11. Is there anything not asked in this survey you would like to add?

If you don’t mind, I would like to begin with a few demographic questions, which will help me determine whether there are certain trends among groups with similar backgrounds. Please remember that you may opt to skip any question.

1. What is your age group?
   a. 18-29
   b. 30-39
   c. 40-49
   d. 50-59
   e. 60-69
   f. 70+
   g. I’d prefer not to answer

2. What is your ethnicity?
   a. American Indian or Alaska Native
b. Hawaiian or Other Pacific Islander
c. Asian or Asian American
d. Black or African American
e. Hispanic or Latino
f. Non-Hispanic White
g. I’d prefer not to answer

3. What is your religious affiliation, if any?
a. I’d prefer not to answer

4. What is the highest degree or level of education you have completed?
a. Less than high school
b. High school graduate (includes equivalency)
c. Some college, no degree
d. Associate's degree
e. Bachelor's degree
f. Ph.D.
g. Graduate or professional degree
h. I’d prefer not to answer

5. What is your employment status?
a. Student
b. Unemployed
c. Retired
d. Employed
e. I’d prefer not to answer

6. What is your marital status?
a. Single
b. Married
c. Divorced
d. Widowed
e. In a committed partnership
f. I’d prefer not to answer

We have now concluded this interview, thank you for your participation. Your responses are invaluable to this research. If there is something I forgot to ask, is it ok if I call you to follow up?

Yes/No

Thank you very much for your time.