Received: 23 June 2021 | Revised: 14 September 2021 | Accepted: 20 September 2021

DOI: 10.1002/pon.5831

WILEY

ORIGINAL ARTICLE

Coping response and family communication of cancer risk in men harboring a *BRCA* mutation: A mixed methods study

Andrew A. Dwyer[1,2] | Sharlene Hesse-Biber[3] | Hannah Shea[3] | Ziwei Zeng[4] | Shiya Yi[5]

[1]William F. Connell School of Nursing, Boston College, Chestnut Hill, Massachusetts, USA

[2]Munn Center for Nursing Research, Massachusetts General Hospital, Boston, Massachusetts, USA

[3]Department of Sociology, Boston College, Chestnut Hill, Massachusetts, USA

[4]Lynch School of Education and Human Development, Boston College, Chestnut Hill, Massachusetts, USA

[5]Department of Measurement, Evaluation, Statistics and Assessment, Boston College, Chestnut Hill, Massachusetts, USA

Correspondence
Sharlene Hesse-Biber, Department of Sociology, Boston College, McGuinn Hall 419, 140 Commonwealth Ave, Chestnut Hill, MA 02467, USA.
Email: sharlene.hesse-biber@bc.edu

Funding information
Boston College Schiller Institute Grant for Exploratory Collaborative Scholarship

Abstract

Objective: Providing genetic counseling and genetic testing to at-risk blood relatives (cascade screening) is important for improving *BRCA* cancer outcomes. Intra-familial communication of risk is critical for cascade screening efforts yet relatively little is known about men's role in communicating *BRCA* risk. We sought to examine men's coping response to their *BRCA* status and intra-familial communication of risk to inform the development of tailored interventions that could promote cascade screening.

Methods: We employed a sequential mixed-methods design. First, we measured coping response (quantitative) using the Multidimensional Impact of Cancer Risk Assessment (MICRA). MICRA scores were compared between *BRCA*+ men, *BRCA*− men and *BRCA*+ women. Subsequently, we used template analysis to analyze qualitative interviews exploring coping and intra-familial communication of risk. The Theory of Planned Behavior (TPB) served as a guiding framework for identifying intervention targets.

Results: *BRCA*+ men ($n = 36$) had significantly higher levels of distress ($p < 0.001$), uncertainty ($p < 0.001$) and negative experiences ($p < 0.05$) compared to *BRCA*− male counterparts ($n = 23$). *BRCA*+ men had significantly lower distress ($p < 0.001$) and uncertainty ($p < 0.001$) than *BRCA*+ women ($n = 406$). Qualitative analysis of in-depth interviews with *BRCA*+ men ($n = 35$) identified promoters and barriers to active coping response and intra-familial communication of risk. Mapping results onto the TPB identified targets for tailoring person-centered approaches for men addressing beliefs/attitude, subjective norms, and perceived behavioral control.

Conclusions: Men and women appear to have different coping responses to learning their *BRCA* status. Developing tailored (sex-based), theory informed interventions may help promote intra-familial communication of *BRCA* risk and support cascade screening.

KEYWORDS
BRCA mutation, cascade screening, communication, coping, genetic risk, genetic testing, hereditary breast ovarian cancer, oncology, Theory of Planned Behavior

Andrew A. Dwyer and Sharlene Hesse-Biber are co-first authors.

1 | INTRODUCTION

Breast cancer is well recognized as a leading cause of cancer in women and is the second leading cause of cancer death in the United States.[1] Pathogenic germline variants in breast cancer 1 (*BRCA1*) and breast cancer 2 (*BRCA2*), hereafter collectively termed *BRCA*, are inherited in an autosomal dominant manner and underlie hereditary breast and ovarian cancer (HBOC) syndrome. Harboring a *BRCA* variant increases breast cancer risk by 45%–65% by age 70.[1] Men with *BRCA* variants are at increased risk for developing breast cancer as well as pancreatic cancer, melanoma and prostate cancer.[1] Importantly, men are just as likely as women to harbor *BRCA* variants and pass *BRCA* variants onto offspring. Thus, men are equally implicated in efforts to improve cancer outcomes by uncovering *BRCA*-related risk (e.g., family history, genetic testing). Additionally, men can play an important role in discussing *BRCA* risk with blood relatives to facilitate cascade screening—systematically providing genetic counseling and genetic testing to at-risk blood relatives.[2]

There are several gaps and barriers that limit men from contributing to improved *BRCA* outcomes. While family history is critical for identifying risk, data suggest that paternal family history is often minimized or neglected[3,4] representing lost opportunities to identify cancer risk. At the level of the individual, hereditary cancer knowledge is often low and misperceptions regarding risk are common.[5] At the interpersonal/family level, men are less likely to be informed of *BRCA* test results[3] and frequently feel left out of discussions of *BRCA* risk.[6] At the environmental (health system) level, healthcare professionals play a key role in risk appraisal. A qualitative study of 25 *BRCA*+ men identified patterns of ineffective communication with healthcare providers that contributed to men underestimating their cancer risk.[7] A study of *BRCA*+ men 4 years following testing showed less than half of men had an increased perception of cancer risk.[8]

Once risk is evident, coping response is a key precursor to intra-familial communication. Limited disclosure within families is a significant barrier to cascade screening contributing to low uptake of genetic testing in blood relatives.[9] Indeed, studies suggest that less than half of families fully communicate *BRCA* risk to at-risk relatives[10] and only about one-half of relatives have cascade screening.[11] A qualitative study of 15 *BRCA*+ men revealed avoidant responses following learning their genetic status.[12] We recently reported a "parent of origin" effect wherein women with a paternally inherited *BRCA* variant were unaware of their cancer risk compared to maternally inherited counterparts.[13] A subsequent qualitative study of 97 women harboring a *BRCA* variant suggests that ineffective, blocked communication within paternally inherited families contributes to divergent risk awareness and outcomes.[14] Many high-risk men lack knowledge about *BRCA* and benefit from educational interventions supporting decision-making.[15] In addition, group interventions can also help support high quality decisions and promote family communication of *BRCA* risk.[16]

There is limited data on men's appraisal of risk, coping response and communication of *BRCA* risk to at-risk blood relatives—information that is critical to informing tailored interventions for men to increase cascade screening for HBOC and improving *BRCA* cancer outcomes. To fill this gap, used a mixed-methods approach (i.e., quantitative and qualitative) to inform development of theory-informed approaches for counseling and coaching men to enhance cascade screening. First, we aimed to quantify coping response in men who underwent *BRCA* testing and compare psychological responses in men who harbor a pathogenic *BRCA* variant with those who are *BRCA* negative. We also sought to examine sex as a biologic variable to determine if men respond similarly to women in their psychosocial response to their *BRCA*+ status. Second, we used qualitative inquiry in the same *BRCA*+ men to explore coping response and human factors underlying intra-familial communication of risk. Last, we integrated the quantitative and qualitative data and mapped findings onto the Theory of Planned Behavior (TPB)[17] to identify theory-informed targets for tailored interventions for men to increase the uptake of cascade screening.

2 | METHODS

We employed a sequential, mixed-methods research design to investigate *BRCA*+ men's coping response, family communication of cancer risk and cascade screening. We considered coping response as a precursor to intra-familial communication of risk that can support subsequent cascade screening. Briefly, we measured coping response to *BRCA* genetic testing (quantitative survey) then explored *BRCA*+ men's experiences and human factors (qualitative interviews) related to family communication of risk that can facilitate cascade screening. The Boston College IRB approved this study (protocol #16.109.01) and all participants provided opt-in electronic informed consent prior to participation. Findings are reported according to STrengthening the Reporting of OBservational studies in Epidemiology (STROBE).

2.1 | Participants and procedures

We identified a convenience sample of men from the United States who underwent *BRCA* genetic testing by partnering with breast cancer/*BRCA* patient support organizations (Male Breast Cancer Coalition, Facing our Risk of Cancer Empowered [FORCE]) and using social media postings (e.g., Twitter, Facebook) (December 2012 to January 2018). All men (18+ years of age) who had undergone genetic testing for *BRCA* were eligible to participate. Following opt-in electronic consent, participants completed an online survey (Qualtrics™, Provo UT) providing sociodemographic data, clinical information (i.e., personal and family cancer history) and coping response to genetic testing using the Multidimensional Impact of Cancer Risk Assessment (MICRA).[18] Men who had tested positive for a *BRCA* variant were invited to participate in an in-depth phone interview (1–2 h in duration). A single investigator (S.H-B.) conducted all interviews and recorded memos. Each interview commenced by confirming sociodemographic/clinical information then participants were

asked to "share their story." Interviews were audio-recorded then transcribed verbatim for qualitative data analysis. All participants were sent a copy of their interview, given the opportunity to review their transcript and make changes they felt were necessary. No participants chose to edit their transcript.

2.2 | Measures

To assess coping response, we used the validated MICRA.[18] Briefly, the MICRA is a validated 19-item instrument that quantifies psychological response to genetic test results across three subscales: distress (α = 0.86–0.94), uncertainty (α = 0.77–0.92) and positive/negative experiences (α = 0.75–0.85).[18] Participants respond to items using a scale (0 = never, 1 = rarely, 3 = sometimes, 5 = often) and higher scores indicate greater levels of distress, uncertainty and negative experiences respectively.

2.3 | Analyses

For the quantitative survey, we report sociodemographic data using descriptive statistics. We used independent-sample Student's *t*-tests to compare composite MICRA scores (total score and three subscale scores) for men who were *BRCA* positive and negative respectively. Additionally we compared MICRA scores between *BRCA*+ men and a reference group of *BRCA*+ females who were recruited using identical methods—data previously reported.[13] We employed multinomial logistic regression to examine if men's marital status, having children and age predicted MICRA score. A p value < 0.05 was considered statistically significant. For qualitative interviews, we used template analysis as previously described.[19] First, investigators conducted a comprehensive review of existing literature on *BRCA* in men to identify central template themes based on existing literature to identify salient themes (i.e., "top-down" method for identifying central organizing concepts). To complement the deductive approach of template analysis, we used an inductive approach (i.e., "bottom-up" method) by employing aspects of grounded theory.[20] Briefly, we read transcripts line-by-line to identify emergent codes and sub-codes within each template theme. Interview quotes not fitting within the existing organization were used to create a new code or sub-code. The inductive, "bottom-up" approach enabled us to analyze interview data that appeared to deviate from, expand on, or refine the initial deductive template themes. This process helped us better understand and explain a wider range of interview data that did not easily fit into initial templates—thereby broadening and deepening our understanding of the template themes. Iterative qualitative analysis continued until no new relevant codes or sub-codes could be identified from the data (i.e., data saturation). Two investigators (S.H-B., H.S.) independently performed line-by-line readings to organize the template, which was used to analyze interview transcripts. Memos were also written for every transcript to document rationale for how the template was applied. Following coding, three investigators (S.H-B., H.S., A.A.D.) reviewed the emergent elements and organized the codes and sub-codes. This qualitative inquiry process revealed patterns among the participants to expand the quantitative findings and deepen our understanding of *BRCA*+ men's experiences with coping and family communication that can affect subsequent cascade screening.

2.4 | Theoretical framework

To synthesize quantitative and qualitative findings and identify targets for interventions to enhance cascade screening, we utilized the TPB as a guiding framework.[17] Briefly, the TPB posits that intention precedes action/behavior (i.e., family communication of risk and cascade screening). Further, intention is shaped by attitudes, subjective norms and perceived control—all of which are shaped by past experiences. Attitudes are considered to reflect an individual's perceptions of the behavior being good/bad. Subjective norms refer to expectations of family, friends, healthcare providers. Perceived control relates to an individual's self-efficacy and perceived agency. The TPB applies to an individual's intention/behavior. Accordingly, we considered the intention/behavior as the individual carrier sharing information with blood relatives (i.e., family communication of risk). Such communication would be considered a necessary, preceding step for cascade screening (i.e., the at-risk blood relative having genetic counseling/testing). We mapped the most frequently cited codes and sub-codes (i.e., cited by at least half of men) to identify targets for interventions to enhance family communication of risk (which could subsequently support cascade screening).

3 | RESULTS

3.1 | Quantitative survey

In total, 67 men completed the survey. Fifty-nine men had undergone *BRCA* genetic testing and were included in the quantitative analysis. The sociodemographic and medical information of respondents is shown in Table 1. The sample was quite homogenous in terms of self-reported race with 50/59 (85%) identifying as white/Caucasian (12 did not identify race, Black/African-American n = 1, Asian n = 1). The majority of men, over age 50, were well educated (at least a Bachelor degree), were middle/upper class by self-report, married/partnered and had at least one child. Approximately one-third (19/59, 32%) had a personal history of cancer. In terms of *BRCA* status, 36/59 (61%) harbored a *BRCA* variant and 23/59 (39%) tested negative. Men with/without a *BRCA* variant did not differ in any of the sociodemographic variables. In terms of psychological response, MICRA scores did not differ according to age (<50 years-old vs. 50+ years-old, p = 0.84). Similarly, MICRA scores did not differ in terms of having a personal history of cancer or not (p = 0.16). We considered that time from testing could affect MICRA score. To examine this, we compared men who had completed the MICRA within 12-months of genetic testing

TABLE 1 Participant sociodemographic characteristics

	Male: *BRCA*− (*n* = 23)	Male: *BRCA*+ (*n* = 36)	Total male (*n* = 59)	Female: *BRCA*+ (*n* = 406)
Age (years)				
<30	0	1 (3%)	1 (2%)	72 (18%)
31–40	0	1 (3%)	1 (2%)	129 (32%)
41–50	1 (4%)	8 (22%)	9 (15%)	127 (31%)
51–60	6 (26%)	12 (33%)	18 (30%)	63 (16%)
>60	16 (70%)	14 (39%)	30 (51%)	15 (3%)
Socioeconomic status				
Upper/middle class	20 (87%)	34 (94%)	54 (92%)	344 (85%)
Working/lower middle	3 (13%)	2 (6%)	5 (8%)	62 (15%)
Education				
Bachelor or greater	22 (96%)	33 (92%)	55 (93%)	254 (63%)
Less than bachelor	1 (4%)	2 (6%)	3 (5%)	152 (37%)
Marital status				
Partnered/married	19 (84%)	27 (75%)	46 (78%)	336 (83%)
No partner/single	4 (16%)	9 (25%)	13 (22%)	70 (17%)
Child(ren)				
One child or more	21 (92%)	31 (86%)	52 (88%)	286 (70%)
No child	2 (8%)	5 (14%)	7 (12%)	120 (30%)
Personal cancer history				
Yes, current/past diagnosis	3 (13%)	16 (44%)	19 (32%)	353 (87%)
No personal cancer history	20 (87%)	20 (56%)	40 (68%)	53 (13%)

(*n* = 24) and those who completed the MICRA a year or more after testing (*n* = 35). In *BRCA*+ men, total MICRA scores did not differ according to time from testing ($p = 0.95$). Further, no significant differences were noted in any of the sub-domains. Similarly, *BRCA*− men did not exhibit any differences according to time from genetic testing ($p = 0.42$). Men harboring a *BRCA* variant reported significantly higher levels of distress ($p < 0.001$), uncertainty ($p < 0.001$) and negative experiences ($p < 0.05$) compared to *BRCA*− counterparts (Table S1). Comparing MICRA scores between *BRCA*+ men (*n* = 36) and *BRCA*+ women (*n* = 406) revealed similar scores for negative experiences. However, distress and uncertainty differed according to sex. *BRCA*+ women exhibited significantly higher distress and uncertainty compared to males (both $p < 0.001$). Using multinomial logistic regression, neither men's marital status ($p = 0.17$), having children ($p = 0.12$) nor age ($p = 0.15$) predicted MICRA score.

3.2 | Qualitative interviews

In total, 35/36 *BRCA*+ men consented to an in-depth interview. The three template themes identified from the literature related to: (i) interactions with the healthcare system, (ii) family dynamics and (iii) psychosocial factors.[14] In relation to the "healthcare system" template theme, analyzing interviews revealed codes that promoted (positive) and inhibited (negative) active coping response (i.e., not avoidant) (Figure 1). Three codes, "cost/lack of insurance coverage," "stigmatizing, gendered interactions" and "ineffective communication" with healthcare providers, impeded effective coping responses. Of note, the sub-code "perceived lack of provider *BRCA* knowledge" (under "ineffective communication" code) was cited 49 times by 18/35 (51%) of men. In contrast, "trust and confidence" in healthcare providers was identified as a facilitator of effective coping. Within the template theme of "family dynamics," both promoters and barriers to intra-familial communication of risk were identified (Figure 2). Promoting factors included "awareness of family history" and "family emotional support." Additionally, two codes relating to family norms, "testing norms" (female only vs. testing for all) and "communication norms" (open vs. closed), were found to either promote or hinder family communication of risk. Four codes were deemed to inhibit family communication of risk: "socio-economic status differences," "gendered communication" as well as "geographic distance" and "emotional" distance (i.e., not feeling close or connected to family members) between family members (which frequently co-occurred together) were cited as barriers. The "male stoicism" sub-code frequently appeared in interviews (112 times by 27/35, 77% of

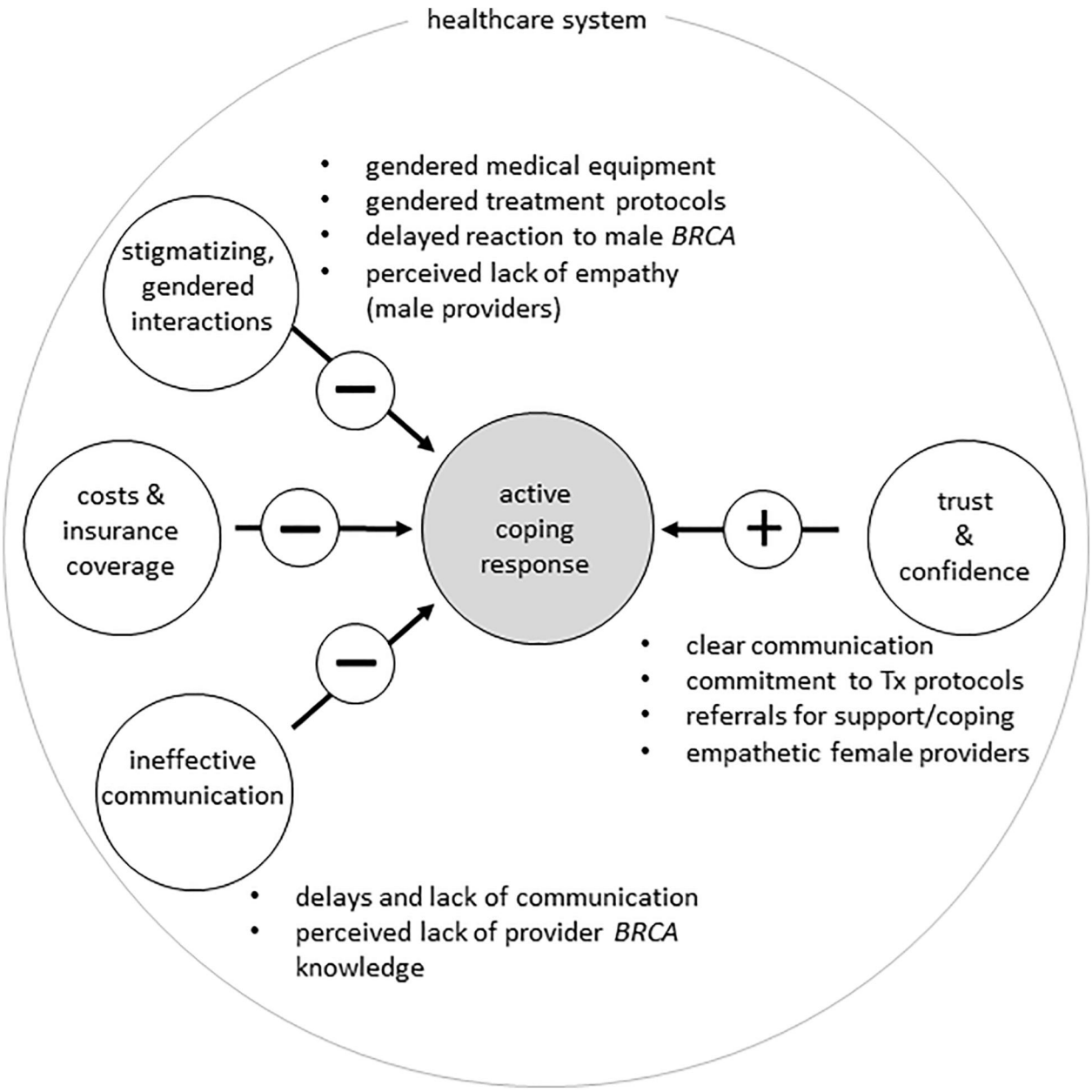

FIGURE 1 Health system: codes relating to coping response. Analyzing interviews with *BRCA*+ men ($n = 35$) identified four codes (circles) and ten sub-codes (bullets) that promote/inhibit active coping response (shaded circle). Three codes impeded effective coping (–) while "trust and confidence" facilitated effective coping (+). Tx, treatment

men) suggesting that perceived social scripts (i.e., men are not supposed to show emotion) may play an important role in coping response and intra-familial communication of *BRCA* risk.

In terms of "psychosocial factors" relating to active coping response and intra-familial communication of risk we identified two barriers and two mixed factors (Figure 3). Similar to "family dynamics," the code of "male stoicism" emerged as a central code within the template theme of "psychosocial factors"—wherein men downplayed their own *BRCA* risk and prioritized concern for family over themselves. "Fear of treatment" was a frequently cited code (49 times by 23/35, 66%) that impeded active coping response and intra-familial communication of *BRCA* risk. Both "psychosocial support" and "changes in identity secondary to *BRCA*/cancer" could either promote or limit coping and communication. Sub-codes such as "changed relational networks" and "community support" (i.e., additional supports from friends and online community) as well as "*BRCA* camaraderie" (i.e., support from female *BRCA* survivors) bolstered coping response. Conversely, the lack of such networks left men feeling unsupported. Men reported changes in their identity that could promote coping and communication including the sub-codes of "body image" (i.e., accepting one's changed body), "somatic identity/role changes" (i.e., embracing *BRCA* survivorship and taking on an activist role as a "sir-vivor") and "relational style" (i.e., embracing a more open attitude towards communication). However, "changes in identity

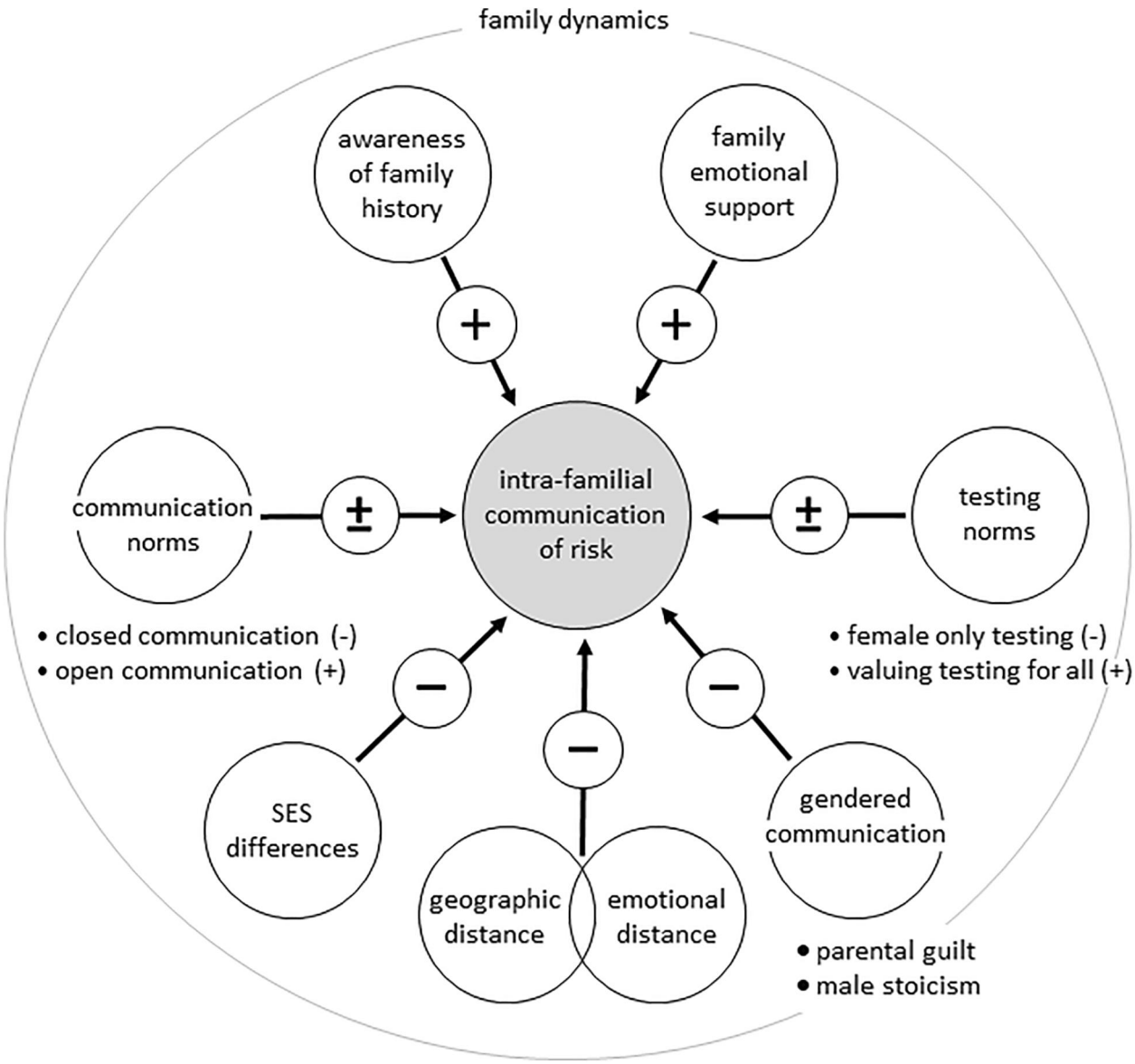

FIGURE 2 Family dynamics: codes relating to intra-familial communication of risk. Analyzing interviews with *BRCA*+ men ($n = 35$) identified 11 codes (circles) and six sub-codes (bullets) that promote/inhibit communication of risk to family members (shaded circle). Two codes (top, +) promoted intra-familial communication of risk. Two codes (left/right, ±) could either promote or hinder intra-familial communication of risk. Five codes (bottom, −) contributed to "blocked" family communication patterns. Codes "geographic distance" and "emotional distance" (overlapping circles) frequently co-occurred in interviews. SES, socioeconomic status

secondary to *BRCA*/cancer" could also work in the opposing direction to undermine effective coping and limit intra-familial communication of *BRCA* risk to potentially at-risk blood relatives (see examples in Supporting Information S1).

3.3 | Mapping findings onto the TPB

Drawing on the significant quantitative findings of increased uncertainty, distress and negative experiences in *BRCA*+/− men, we reviewed the emergent qualitative themes from interviews with *BRCA*+ men (i.e., template themes: "psychosocial" and "family dynamics"). Frequently occurring codes/sub-codes (i.e., cited by more than half of men) that were deemed amenable to intervention were mapped to the elements of the TPB (Figure S1). Representative quotes of each of the most commonly cited codes/sub-codes is provided in Supporting Information S1. Under TPB behavioral beliefs/attitudes, "Identity role changes secondary to *BRCA*/cancer" was cited 179 times by 30/35 (85%) of men. The code "fear of treatment" was cited 49 times (23/35, 66%). This code encapsulated the sub-codes "fear of surgery/death" (29 times, 16/35, 46%) relating to mortality concerns, and "reluctance to seek treatment" (20 times, 12/35, 34%) relating to delayed care seeking behavior. The sub-code "body image" (i.e., gendered self, seeing oneself as macho or not)

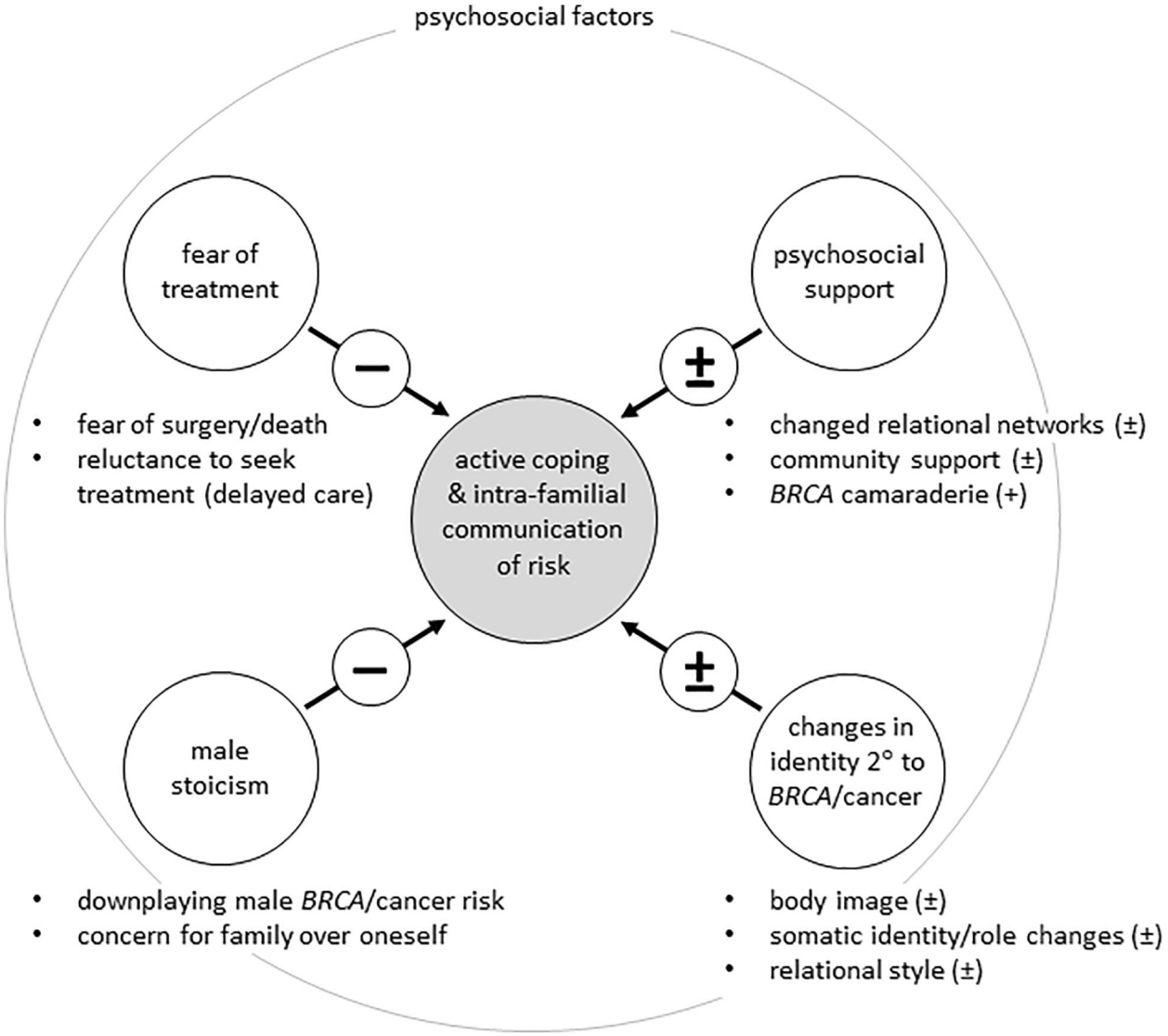

FIGURE 3 Codes relating to psychosocial factors affecting active coping response and intra-familial communication of risk. Analyzing interviews with *BRCA*+ men ($n = 35$) identified four codes (circles) and 10 sub-codes (bullets) that promote/inhibit active coping response and communication of risk to family members (shaded circle). Two codes (left, –) posed barriers to active coping and effective communication. Two codes (right, ±) had mixed-effects

was noted 83 times by 27/35 (77%) of men. While the "body image" code was significant, we opted not to map it onto the TPB. The rationale for this was that it is not clear that "body image" is amenable to a specific intervention and may better considered as a modifying factor.

Within TPB normative beliefs/subjective norms, positive "testing norms" was most frequently cited (85 times by 27/35 [77%] of men) and promoted intra-familial communication of risk. Conversely, family testing norms indicating testing was only relevant for women posed a barrier to cascade screening (Supporting Information S1). Similarly, family communication of risk was impeded by "male stoicism" (112 times by 27/35, 77%)—encompassed sub-codes "downplaying male *BRCA*/cancer risk" and "concern for family over oneself." The sub-code "parental guilt" was cited 40 times by 23/35 (64%) of men. Finally, under TPB control beliefs/perceived behavioral control, "awareness of family history" was frequently cited in interviews (77 times, 27/35, 77% of men). In such instances, knowing the family history of cancer was seen as empowering for men to act on this information whereas those who were unaware did not appraise risk and were "blindsided" by the genetic risk (Supporting Information S1). Similarly, "psychosocial support" was noted 35 times by 19/35 (54%) of men with instances of promoting and impeding coping, communication and cascade screening (Supporting Information S1).

4 | DISCUSSION

Herein we report men's quantified psychological response to genetic *BRCA* testing and subsequent qualitative exploration of mean's coping response and intra-familial communication of *BRCA* risk. Men

harboring a *BRCA* variant had significantly greater distress, uncertainty and negative experiences compared to *BRCA*− counterparts. All men had genetic testing due to family cancer history, so the heightened psychological impact on *BRCA*+ men suggests that genetic test results per se are a key factor in coping response. Our findings are consistent with prior studies documenting that increased distress in *BRCA*+ men compared to men who tested negative.[8,21] Graves and colleagues found male *BRCA* carriers reported significantly MICRA distress scores at 6 and 12-months compared to *BRCA* − men.[21] While absolute differences were noted over time, no differences were observed between groups after controlling for baseline levels of distress. A 2013 Israeli study of 69 men found significantly greater distress, more negative experiences and higher overall MICRA scores in men harboring a *BRCA* variant.[8] We observed similar results in the present study as well as greater uncertainty in *BRCA*+ men. It is worthwhile to note that the MICRA is used to report relative differences in psychological response to genetic testing. Thus, neither 'normal' reference ranges nor thresholds are established for the instrument. We also examined sex as a biologic variable by comparing psychological response between *BRCA*+ men and women. Analysis revealed women had significantly greater distress and uncertainty compared to males suggesting sex-specific appraisals of risk conferred by a pathogenic *BRCA* variant.

Appraisal of risk is linked with coping response. Prior qualitative studies have identified divergent coping responses among *BRCA*+ men. Some men respond with an active response (i.e., seeking information, communicating with family/healthcare professionals, engaging in screening/treatment) while others have a passive/avoidant coping response (i.e., deny/ignore *BRCA* status, passively avoidant, know but do not act on information).[6,22,23] Our qualitative interviews with *BRCA* + men helped elucidate men's coping response and subsequent intra-familial communication of risk. Within the template theme of "healthcare system," we identify "trust and confidence" in healthcare professionals as a critical factor supporting active coping response (Figure 1). The validity of this observation is supported by prior studies and systematic reviews underscoring the powerful role that providers (e.g., physicians, nurses, genetic counselors) have in promoting intra-familial communication and cascade screening.[24] Additionally, key findings from our qualitative inquiry ("stigmatizing, gendered interactions" and "ineffective communication") mirror prior work examining psychosocial aspects of men with *BRCA* variants that identified similar themes of stigma/shame[12,22,23,25] and limited disclosure of *BRCA* status.[3,7,25] More broadly, recent systematic and scoping reviews indicate that men are reluctant to discuss health issues and seek help for health concerns—particularly for mental health concerns.[26,27] Collectively, such findings point to the need for more inclusive and person-centered approaches to male obligate *BRCA* carriers.

Previous studies highlight men's need for *BRCA* information as well as negative experiences of providers being dismissive or minimizing men's *BRCA* risk.[7,12,25,28] Thus, evidence from the present study and others point to a need for change in healthcare systems to create more inclusive, less gendered clinical environments as well as re-doubling of efforts to adopt more person-centered approaches to support accurate *BRCA* risk appraisals and shared decision-making.[14] While intra-familial communication of risk can support cascade screening efforts, it merits mentioning that there are legal frameworks in place to protect individuals. In the United States, the Genetic Information and Non-Discrimination Act (GINA) protects individual genetic information privacy and prevents genetic discrimination.[29] Thus, concerns about workplace or health insurance discrimination should not pose barriers to communication of genetic risk. However, GINA protections do not include life and disability insurance protections.

Within the template theme "family dynamics," effective intra-familial communication of risk was bolstered by "awareness of family history" and positive "testing norms" (Figure 2). Indeed, a recent study examining sharing genetic test results identified family ties and sense of duty as important motivators for sharing test results with family members.[30] Notably, in the present study, "parental guilt" was often cited as a barrier to communication. Parental guilt is common to many genetic conditions[31] including *BRCA*.[32,33] Feelings of guilt likely contribute to emotional reactions including shame and secrecy that impede disclosure of genetic test results within families.[12,34] We identify additional barriers to active coping and intra-familial communication of *BRCA* risk in the template theme "psychosocial factors." Notably, "male stoicism" (i.e., men are supposed to be strong and not show emotion) was cited by nearly two-thirds (27/35, 77%) of men. It is plausible that such *BRCA*-related appraisals and responses (male stoicism" and "parental guilt") may help explain why men are less likely to report family history of cancer,[4] pursue genetic testing and undergo genetic testing.[35]

Key promoters of active coping and intra-familial communication included "psychosocial support" and positive "changes in identity secondary to *BRCA*/cancer" (Figure 3). Men shared stories of how receiving support form spouse/partner, friends and *BRCA* peer-to-peer support (face-to-face and virtual) helped them re-conceptualize their identity. In line with our observations, prior qualitative studies have identified spousal support[6,7] or having a "*BRCA* informant" sister[14] as pivotal for supporting men in disclosing *BRCA* risk within families. Strong family ties repeatedly appear in the literature on *BRCA*+ men and often motivate men to focus on offspring[6,22,23] and view communication of risk as a "duty."[12,32,34,36] Interestingly, our prior work indicates that age plays a modifying factor.[37] Younger men are more stigmatized (akin to the "body image" sub-code in the present study) by learning their *BRCA* status (i.e., a "women's disease") and focused more on their own personal health. In contrast, older men (>50 years-old) were more concerned about risk to family.[37] Thus, when considering between-sex responses to *BRCA* status, one should also consider temporal aspects relating to where an individual is in their respective life-course.

4.1 | Clinical implications

Mapping codes/sub-codes onto the TPB identified several targets for interventions to support men's coping and facilitate intra-familial

communication in order to facilitate subsequent cascade screening. A strengths-based approach could be used to help men perceive their *BRCA* status as an opportunity to re-conceptualize their identify[28] and embrace traditional masculine gender roles[27] of being a protector (i.e., communicating *BRCA* risk = being a "good" father). One possible approach to overcome the stigma of *BRCA*/breast cancer (i.e., a "woman's disease") is to adopt a harm reduction perspective and use "chest" as opposed to "breast" as appropriate during discussions with men.[25] Of note, *BRCA* variants are passed to offspring in an autosomal dominant manner (i.e., 50% chance) yet parental guilt is common in genetic disorders.[31] Healthcare providers play an important role in absolving parents of culpability. Specifically, healthcare professionals can reinforce that passing a gene onto offspring is not intentional. Rather, heritability is like flipping a coin. Additionally, each and every interaction with *BRCA*+ men is an important opportunity to inquire about social supports, provide resources (i.e., peer-to-peer support groups) and underscore the value of having trusted confidants to process reactions to learning *BRCA* status and possible cancer risk. Importantly, factors underlying coping response and intra-familial communication of risk are multifactorial, complex and dynamic. Accordingly, interventions to promote active coping and communication of *BRCA* risk could be characterized as a complex intervention.[38] In line with recommendations from the Medical Research Council,[39] we used a behavioral theory (TPB) as a lens to interpret our findings and identify targets amenable to intervention. Moreover, there are numerous factors mediating intra-familial communication including resilience, vulnerability, communication patterns, emotions/geographic distance, fear (cancer and treatments) as well as family cohesion/dynamics. Accordingly, stakeholder engagement may be very useful for co-creating solutions to respond to the unmet health and informational needs of these men.[40]

4.2 | Study limitations

Relative strengths of this study are that all *BRCA*+ men completed the survey and had an in-depth interview. This mixed methods approach (i.e., quantitative and qualitative) provides deeper insight into coping response and intra-familial communication of risk than either approach in isolation. However, it is important to note that the study has a number of limitations. First, as we recruited through patient support organizations and social media introducing a risk of recruitment bias. Moreover, the enrollment period was relatively long and it is possible that data collected earlier in the study may not accurately reflect current patient attitudes, beliefs, and coping as genetic testing has become increasingly normalized in clinical practice. In addition, sample is comprised of men from the United States, so findings may not applicable to other countries and cultures. The sample is limited in size and relatively homogeneous (85% White/Caucasian) thereby limiting generalizability. Notably, there is a paucity of data regarding the experiences of *BRCA*+ men of color and more work is needed to engage and better understand their needs.

5 | CONCLUSIONS

We identified significant distress, uncertainty and negative experiences in *BRCA*+ men compared to *BRCA*− counterparts. Additional analysis revealed sex differences. Compared to *BRCA*+ men, women had even greater distress and uncertainty after learning their *BRCA* status. Developing tailored sex-based interventions may help shift current perspectives from guideline-based approaches targeting populations to more precision interventions guided by the unique experiences of individuals. Qualitative analysis revealed psychosocial factors as well as family dynamics and the healthcare system act as promoters and barriers to active coping in *BRCA*+ men and intra-familial communication of risk. Mapping findings on to the TPB, we identify several targets for interventions for men to increase the uptake of cascade screening in families. To realize the full potential of our current understanding of *BRCA* and improve cancer outcomes, there is a need to develop complex interventions supporting cascade screening in families harboring pathogenic *BRCA* variants.

ACKNOWLEDGMENTS

This study was funded by a Boston College Schiller Institute Grant for Exploratory Collaborative Scholarship. We are thankful for the generous participation of the patients and recognize the Boston College *BRCA* Team for their support.

CONFLICT OF INTEREST

The authors have no competing interests to declare.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ORCID

Andrew A. Dwyer https://orcid.org/0000-0002-7023-6794

REFERENCES

1. Nelson HD, Pappas M, Cantor A, Haney E, Holmes R. Risk assessment, genetic counseling, and genetic testing for BRCA-related cancer in women: updated evidence report and systematic review for the US Preventive Services Task Force. *J Am Med Assoc*. 2019;322(7):666-685.
2. Bednar EM, Sun CC, McCurdy S, Vernon SW. Assessing relatives' readiness for hereditary cancer cascade genetic testing. *Genet Med*. 2020;22(4):719-726.
3. Daly MB, Montgomery S, Bingler R, Ruth K. Communicating genetic test results within the family: is it lost in translation? A survey of relatives in the randomized six-step study. *Fam Cancer*. 2016;15(4):697-706.
4. Ozanne EM, O'Connell A, Bouzan C, et al. Bias in the reporting of family history: implications for clinical care. *J Genet Counsel*. 2012;21(4):547-556.
5. Young AL, Butow PN, Rhodes P, et al. Talking across generations: family communication about BRCA1 and BRCA2 genetic cancer risk. *J Genet Counsel*. 2019;28(3):516-532.
6. Suttman A, Pilarski R, Agnese DM, Senter L. "Second-class status?" insight into communication patterns and common concerns among

men with hereditary breast and ovarian cancer syndrome. *J Genet Counsel*. 2018;27(4):885-893.
7. Rauscher EA, Dean M. "I've just never gotten around to doing it": men's approaches to managing BRCA-related cancer risks. *Patient Educ Counsel*. 2018;101(2):340-345.
8. Shiloh S, Dagan E, Friedman I, Blank N, Friedman E. A follow-up study on men tested for BRCA1/BRCA2 mutations: impacts and coping processes. *Psycho Oncol*. 2013;22(2):417-425.
9. Alegre N, Perre PV, Bignon YJ, et al. Psychosocial and clinical factors of probands impacting intrafamilial disclosure and uptake of genetic testing among families with BRCA1/2 or MMR gene mutations. *Psycho Oncol*. 2019;28(8):1679-1686.
10. Healey E, Taylor N, Greening S, et al. Quantifying family dissemination and identifying barriers to communication of risk information in Australian BRCA families. *Genet Med*. 2017;19(12):1323-1331.
11. Lieberman S, Lahad A, Tomer A, et al. Familial communication and cascade testing among relatives of BRCA population screening participants. *Genet Med*. 2018;20(11):1446-1454.
12. Moynihan C, Bancroft EK, Mitra A, et al. Ambiguity in a masculine world: being a BRCA1/2 mutation carrier and a man with prostate cancer. *Psycho Oncol*. 2017;26(11):1987-1993.
13. Hesse-Biber S, Dwyer AA, Yi S. Parent of origin differences in psychosocial burden and approach to BRCA risk management. *Breast J* 2020;26(4):734-738.
14. Dwyer AA, Hesse-Biber S, Flynn B, Remick S. Parent of origin effects on family communication of risk in BRCA+ women: a qualitative investigation of human factors in cascade screening. *Cancers*. 2020;12(8):2316.
15. Quinn GP, Vadaparampil ST, Miree CA, et al. High risk men's perceptions of pre-implantation genetic diagnosis for hereditary breast and ovarian cancer. *Hum Reprod*. 2010;25(10):2543-2550.
16. McKinnon W, Naud S, Ashikaga T, Colletti R, Wood M. Results of an intervention for individuals and families with BRCA mutations: a model for providing medical updates and psychosocial support following genetic testing. *J Genet Counsel*. 2007;16(4):433-456.
17. Ajzen I. The theory of planned behavior. *Organ Behav Hum Decis Process*. 1991;50:179-211.
18. Cella D, Hughes C, Peterman A, et al. A brief assessment of concerns associated with genetic testing for cancer: the Multidimensional Impact of Cancer Risk Assessment (MICRA) questionnaire. *Health Psychol* 2002;21(6):564-572.
19. King N. Template analysis. In: Symon G, Cassell C, eds. *Qualitative Methods and Analysis in Organizational Research: A Practical Guide*. Sage Publications Ltd.; 1998:118-134.
20. Bryant ACK. *The SAGE Handbook of Grounded Theory*. Sage; 2007.
21. Graves KD, Gatammah R, Peshkin BN, et al. BRCA1/2 genetic testing uptake and psychosocial outcomes in men. *Fam Cancer*. 2011;10(2):213-223.
22. Stromsvik N, Raheim M, Gjengedal E. Cancer worry among Norwegian male BRCA1/2 mutation carriers. *Fam Cancer*. 2011;10(3):597-603.
23. Stromsvik N, Raheim M, Oyen N, Engebretsen LF, Gjengedal E. Stigmatization and male identity: Norwegian males' experience after identification as BRCA1/2 mutation carriers. *J Genet Counsel*. 2010;19(4):360-370.
24. Schwiter R, Rahm AK, Williams JL, Sturm AC, Sturm AC. How can we reach at-risk relatives: efforts to enhance communication and cascade testing uptake: a mini-review. *Curr Genet Med Rep*. 2018;6:21-27.
25. Skop M, Lorentz J, Jassi M, Vesprini D, Einstein G. "Guys don't have breasts": the lived experience of men who have BRCA gene mutations and are at risk for male breast cancer. *Am J Men's Health*. 2018;12(4):961-972.
26. Seidler ZE, Dawes AJ, Rice SM, Oliffe JL, Dhillon HM. The role of masculinity in men's help-seeking for depression: a systematic review. *Clin Psychol Rev*. 2016;49:106-118.
27. Seidler ZE, Rice SM, Ogrodniczuk JS, Oliffe JL, Dhillon HM. Engaging men in psychological treatment: a scoping review. *Am J Men's Health*. 2018;12(6):1882-1900.
28. Dean M, Rauscher E, Gomez E, Fischer C. Expectations versus reality: the impact of men's expectancy violations in conversations with healthcare providers about BRCA-related cancer risks. *Patient Educ Counsel*. 2019;102(9):1650-1655.
29. Rothstein MA. GINA at ten and the future of genetic nondiscrimination law. *Hastings Cent Rep*. 2018;48(3):5-7.
30. Dean M, Tezak AL, Johnson S, et al. Sharing genetic test results with family members of BRCA, PALB2, CHEK2, and ATM carriers. *Patient Educ Counsel*. 2021;104(4):720-725.
31. James CA, Hadley DW, Holtzman NA, Winkelstein JA. How does the mode of inheritance of a genetic condition influence families? A study of guilt, blame, stigma, and understanding of inheritance and reproductive risks in families with X-linked and autosomal recessive diseases. *Genet Med*. 2006;8(4):234-242.
32. Hallowell N, Arden-Jones A, Eeles R, Foster C, Lucassen A, Moynihan C. Guilt, blame and responsibility: men's understanding of their role in the transmission of BRCA1/2 mutations within their family. *Sociol Health Illness*. 2006;28(7):969-988.
33. Stromsvik N, Raheim M, Oyen N, Gjengedal E. Men in the women's world of hereditary breast and ovarian cancer—a systematic review. *Fam Cancer*. 2009;8(3):221-229.
34. Hallowell N, Ardern-Jones A, Eeles R, et al. Communication about genetic testing in families of male BRCA1/2 carriers and non-carriers: patterns, priorities and problems. *Clin Genet*. 2005;67(6):492-502.
35. Daly MB. The impact of social roles on the experience of men in BRCA1/2 families: implications for counseling. *J Genet Counsel*. 2009;18(1):42-48.
36. Hallowell N, Ardern-Jones A, Eeles R, et al. Men's decision-making about predictive BRCA1/2 testing: the role of family. *J Genet Counsel*. 2005;14(3):207-217.
37. Hesse-Biber S, An C. Within-gender differences in medical decision making among male carriers of the BRCA genetic mutation for hereditary breast cancer. *Am J Men's Health*. 2017;11(5):1444-1459.
38. Baroutsou V, Underhill-Blazey ML, Appenzeller-Herzog C, Katapodi MC. Interventions facilitating family communication of genetic testing results and cascade screening in hereditary breast/ovarian cancer or lynch syndrome: a systematic review and meta-analysis. *Cancers*. 2021;13(4):925.
39. O'Cathain A, Croot L, Duncan E, et al. Guidance on how to develop complex interventions to improve health and healthcare. *BMJ Open*. 2019;9(8):e029954.
40. Dwyer AA, Au MG, Smith N, et al. Evaluating co-created patient-facing materials to increase understanding of genetic test results. *J Genet Counsel*. 2021;30(2):598-605.

SUPPORTING INFORMATION

Additional supporting information may be found in the online version of the article at the publisher's website.

How to cite this article: Dwyer AA, Hesse-Biber S, Shea H, Zeng Z, Yi S. Coping response and family communication of cancer risk in men harboring a *BRCA* mutation: a mixed methods study. *Psychooncology*. 2022;31(3):486-495. https://doi.org/10.1002/pon.5831