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Using Mixed-Methods to Evaluate “Transformation of Motivation” as a Construct Influencing Risk Communication among Families at Inherited Risk for Ovarian Cancer

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Abstract

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By Jingsong Zhao

Inherited health conditions represent shared risk for families. Many have called for applying relational frameworks in considering how genetic risk communication could be motivated. Generally, these frameworks suggest concepts such as “transformation of motivation” (*TM*) holding that shared desires to preserve family relationships can prompt positive health steps that would not be taken on the proband’s own behalf. However, the *TM* construct that to date has been poorly operationalized. My overarching aim is to advance our understanding of how to measure the construct of *TM*.

In Aim 1, I undertook a systematic review of 13 FGRC interventions identified between January 2010 to August 2023. This review aimed to address a research gap by examining how current intervention strategies align with theories at the individual, relational, and family-system levels. Additionally, evaluated the effectiveness of these strategies compared to standard-of-care groups in fostering communication of inherited risk among families at higher risk of hereditary conditions.

Aim 2 and Aim 3 studies represent a new methodological effort to bring conceptual clarity to the latent TM construct. Rather than relying on unvalidated measurements, I systematically analyzed whether survivors of ovarian cancer survivors’ natural patterns of use of a study website might serve as indicators of “we-ness” thought to underpin the *TM* construct. In aim 2, I found two factors that characterized survivors’ website engagement, one behavioral and one cognitive factor. In Aim 3, I found that “We”-talk were not associated with family closeness and family size. Yet, qualitative interpretations of interviews revealed relational talks from survivors, suggesting that “we”-talk may not be a reliable marker. Furthermore, high and low we-talkers did not differ in their discussion of *TM*-mapped content, except in their perception of family emotional closeness.

In order to evaluate whether TM can be leveraged in behavior change interventions, we must develop rigorous assessment tools to characterize it. Future research needs to move beyond pronoun counts to consider survivor appraisal processes multiple family members’ perspectives on its occurrence, develop and test measures of relational thinking and their association with a variety of shared coping strategies (including health behaviors as the gold standard).

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CHAPTER 1. LITERATURE REVIEW AND SIGNIFICANCE

1.1 DEFINING THE PROBLEM

1.1.1 Background on Hereditary Breast and Ovarian Cancer Syndrome

Breakthroughs in genomic research have advanced scientists' ability to characterize a growing number of pathogenic genetic variants associated with health conditions (e.g., *breast cancer susceptibility genes 1 [BRCA1]* and *2* [*BRCA2*]).[1-4] Moreover, expectations are high that these advances have the potential to be translated for promoting population health.[5,6] For example, in the United States (U.S.), the Centers for Disease Control and Prevention have called on Public Health Departments to prioritize the identification of individuals and families who are at increased risk of medically actionable hereditary conditions.[7] Evidence based guidelines can be followed to identify individuals deemed to be at higher risk for hereditary conditions.[3,8-10] This process begins with family history-based risk screening to indicate whether individuals should proceed to genetic counseling and as appropriate, genetic testing.

In the context of *BRCA1- and BRCA2-associated Hereditary Breast and Ovarian Cancer Syndrome (HBOC),* compared to the general population, women and men with a *BRCA1*/2 pathogenic variant have a markedly higher risk of developing several types of cancer, most notably breast, ovarian, and pancreatic cancers.[3,11] In the U.S., about 13% of women in the general population will develop breast cancer at some point during their lifetime,[12] while their lifetime risk for ovarian cancer is 1-2%.[13] By contrast, women who inherit a *BRCA1*/2 pathogenic variant have a much higher risk for developing breast or ovarian cancer. Evidence suggests that, by 70–80 years of age, 45-72% of women with increased risk will develop breast cancer, while 11-44% of them will develop ovarian cancer.[14-16] Men who carry a *BRCA1*/2 mutation, have 1-10% risk for developing breast cancer and 2-5% risk for developing pancreatic cancer

during their lifetime. By comparison, men in the general population have a risk of 0.1% for developing breast cancer, and 1% for developing pancreatic cancer.[17] In addition, early age of onset of breast and ovarian cancer (i.e., before the age of 50) is a hallmark feature of carrying a *BRCA1* or *BRCA2* mutation.[3,16]

Early detection of harmful *BRCA1*/2 variants becomes an important part of individual cancer risk management.[9,18-21] For example, the U.S. Preventive Services Task Force recommends that all women receive family history-based risk assessment, and if their personal and/or family history is indicative of hereditary cancers, they are referred to genetic counseling and possibly genetic testing for *BRCA1*/2 and other pathogenic variants.[9] Individuals who carry these harmful variants can be referred for enhanced surveillance procedures and other preventive measures to manage the increased cancer risks.[7,22] For example, evidence suggest that additional magnetic resonance imaging (MRI) screening among *BRCA1*/2 carriers is correlated with an increased rate of early stage (stages 0 or 1) breast cancer detection, and improves metastasis free survival.[21,23,24]

1.1.2 Cascade Screening of Hereditary Breast and Ovarian Cancer Syndrome

Identification of individuals who carry a *BRCA1/2* pathogenic variant also has significant health implications for familial hereditary cancer risk management.[3,25] These individuals, usually the first person who has developed HBOC-associated cancer within a family, are referred to as *probands.* Because *BRCA1*/*2* variants are inherited in an autosomal dominant pattern, that is, a single copy inherited from one parent, either mother or father, increases disease risk.[3,26]

All biological relatives of a proband are at increased risk of carrying a deleterious *BRCA1/2* variant. Each of a proband's first-degree relatives (FDRs), including parents, children, and siblings, have a 50% probability of carrying the same variant; second-degree relatives (SDRs), including aunts, uncles, grandparents, grandchildren, nieces, nephews, or half-siblings, have a 25% chance of carrying the same deleterious *BRCA1/2* variant.[26]

Over the past two decades, guidelines have emphasized the value of cascade screening, that is, a systematic approach for the identification of probands' at-risk FDRs and SDRs and referral to genetic testing.[21,27-30] The process involves a specific order of genetic testing starting with the proband and if the proband is a carrier, continuing to FDRs. FDRs of probands who test positive for *BRCA1/2* variants are then advised to inform their FDRs to further the cascade of testing. SDRs of the proband are included in this process. For example, if genetic testing shows that only a proband's mother carries a *BRCA1* or *BRCA2* variant, biological relatives on the mother's side, such as probands' aunts and grandparents, would be advised to be tested. This cascade screening process is then repeated until all relatives with the pathogenic variant are identified in a family.

Once identified at higher risk for HBOC, these at-risk relatives are advised to pursue enhanced screening procedures and preventive options.[3,21] Female relatives are advised to begin annual or bi-annual clinical breast exams at age 25, and pursue ovarian cancer screening at age 30. Male relatives are advised to undergo an annual clinical breast exam beginning at age 35. At-risk relatives may be prescribed with chemopreventive tamoxifen for long term risk reduction and prevention of disease recurrence.[31,32] Cascade screening provides useful information to relatives who screen negative, as they are not at higher risk of HBOC and can continue routine cancer surveillance schedules.

1.1.3 Challenges of the Identification of Relatives at Higher Risk

A critical downside of cascade screening for ensuring that at-risk relatives are informed of their possible risk for HBOC is that probands are required to act as sole intermediaries in for engaging their relatives to seek genetic services.[28,29] It is important to note that health privacy policies and laws have significant influences on the current approach, not only in the U.S. but worldwide.[33] For example, in the U.S., federal regulations, such as the Health Insurance Portability and Accountability Act, govern the use and disclosure of individual confidential medical information.[34,35] In the context of hereditary conditions, healthcare professionals are not

allowed to directly notify any at-risk relatives about their inherited risk, nor to recommend genetic risk assessment to them. Similar laws and regulations exist in other countries such as Australia,[36] Belgium,[37] and United Kingdom,[38] where healthcare professionals are restricted in the disclosure of genetic information to relatives. To date, the standard of care for healthcare professionals is to encourage probands to send a generic notification letter with informational resources to any at-risk relatives, advising them to pursue genetic services.[33,39]

Thus, the "duty" to inform relatives about *BRCA1/2* risk information falls to the proband. As a result, a sizeable number of FDRs and SDRs are unaware of their risk. Based on the prevalence of *BRCA1/2* pathogenic variants, mathematic modeling suggests that there are approximately 941,155 *BRCA1/2* variant carriers in the U.S.[40] Among this population, only a small fraction of approximately 48,754 individuals has been identified through genetic testing.

In the following section, I will highlight two barriers to family communication about inherited risk of particular importance: selective communication within the family, and suboptimal uptake of genetic testing among at-risk relatives.

1.1.3.a Selective communication within the family

Research suggests that probands are highly selective in with whom they communicate genetic risk information. This selectiveness can be observed as early as their deliberation on whether to tell. For example, a cross-sectional survey study of communication intentions among 329 women who underwent genetic testing for *BRCA1/2* reported that women had significantly higher intention to inform female relatives than male relatives, and to children and siblings than parents.[41] But even with high intentions, probands did not always follow through with actual communication.[42-47] For example, Lieberman and colleagues surveyed individuals of Ashkenazi Jewish descent, who are at higher risk of carrying a *BRCA1/2* mutation.[46] While probands intended to communicate with 223 daughters before receiving the genetic testing results, only 141 of the daughters (64%) were actually informed after the results were received.

1.1.3.b Suboptimal uptake of genetic testing among at-risk relatives

Uptake of genetic testing among at-risk relatives is also relatively low. A systematic review of 22 studies from 2001 to 2017 found in 11 studies that HBOC probands reported only 15% to 57% of their relatives had undergone genetic testing.[48] In the remaining 10 studies, where at-risk relatives' records were obtained from genetics centers, uptake among at-risk relatives ranged from 21% to 44%. It's worth noting that half of these studies only focused on FDRs. Thus, the assumption is that SDR use of genetic services was even lower. Thus, even when informed, many relatives choose not to seek genetic services.

1.1.4 Defining Optimal Family Genetic Risk Communication

FGRC, or discussing genetic information amongst families, is best conceived of as a process rather than as a one-time disclosure event.[49-52] In FGRC studies, communication is often operationalized as "talking", "notifying", "sharing", "discussing", "informing", "disclosing", and "disseminating" genetic risk information with/to relatives.[52-54] Despite the consensus that probands should be the lead communicators,[33,55] there is less agreement concerning what the optimal communication should comprise. Currently, there is no standard set of information deemed essential for probands to convey to at-risk relatives. As described above, the standard of care is for probands to send a generic notification letter from a clinician describing, in clinical language, the proband's circumstances and encouraging relatives to seek genetic services.[33,39] These letters are idiosyncratic to the healthcare provider and do not offer any skills for communication regarding what can be a technically challenging and highly sensitive topic.[56,57]

In the absence of standardized guidance for proband-to-relative communication, empirical studies provide insights on what constitutes optimal FGRC.[49,52,58] In general, the process of FGRC starts for the proband before any actual conversation with their relatives. Probands engage in a decision-making process where they deliberate whether to withhold or disseminate genetic risk information with their family. Probands may have the desire to share genetic risk information for numerous reasons, such as protecting relatives from potential harm or fulfilling familial responsibilities. Studies suggest that probands may consider their

relationships with each of their relatives, concerns about their own medical privacy, and potential negative emotional responses from at-risk relatives in deciding whether or not to proceed with risk discussions. Other considerations include whether to disclose to minor children and young adults,[59,60] how to deal with relatives who indicate they do not want to know.[47,61]

If probands opt to share genetic risk information, they must decide whom to inform, and when to have these conversations. As previously described, current studies have mainly focused on assessing to whom probands have communicated. These studies are based on probands' reports of whom they shared risk information and who sought genetic services.

Probands also must decide how and what to communicate to at-risk relatives including how to approach the topic, the extent of information to disclose, and how to explain follow-up options. Less research attention has been given to this step and the quality of the communication process.[57,62] The complexities of personal, relational, and families predisposed factors involve a good deal of nuance, further complicating this communication step.

For this dissertation, I define optimal FGRC as a communication process that begins with probands' deliberations regarding whether and when to tell, who to tell, and what to tell at-risk relatives and extends through their first and subsequent conversation(s) regarding inherited risk with at-risk relatives.

In the upcoming sections, I will first provide an overview on the current state of FGRC, specifically focusing on communication within the HBOC family context (1.2). Then I will review communication and ethical frameworks as well as behavioral theories that offer insights into achieving optimal FGRC (1.3). In the last section of the Chapter, I will focus on the conceptualization and current research efforts related to interdependence theory and communal coping, which are key relational theories guiding my dissertation.

1.2 CURRENT STATE OF FAMILY GENETIC RISK COMMUNICATION RESEARCH

In this section, I will describe the evidence base related to each step in the FGRC process I described above. This comprises considerations for: (1) whether and when to tell; (2) who to tell; and (3) what to tell. I also will discuss gaps to and opportunities for attaining optimal FGRC.

1.2.1 Current Evidence Base for FGRC

I found 15 peer-reviewed FGRC reviews spanning numerous hereditary conditions.[28,48,52-54,60,63-72] Of these, 3 focused on process and outcomes of FGRC,[52,53,72] 5 assessed facilitators and barriers of FGRC or cascade screening,[28,54,66,68,72] 1 described how FGRC was addressed as part of genetic counseling,[64] 4 focused on genetic risk communication to children and young adults.[60,63,65,67] The uptake of genetic testing among at-risk relatives was assessed in 1 review.[48] And lastly, 3 reviews focused on FGRC interventions to improve the sharing of genetic test results to at-risk relatives, including one review that I conducted as part of Aim 1 of my dissertation.[69-71]

The majority focused on communication in the context of HBOC and Lynch syndrome. For example, Gaff and colleagues identified 26 FGRC studies between 1985 and 2006; 21 studies on HBOC, 3 on Huntington disease, 3 on cystic fibrosis and 1 on chromosome anomalies.[52] Only one review exclusively focused on family communication of hereditary cardiomyopathies and arrhythmias genetic risk. Moreover, consideration of differences between hereditary conditions, such as their penetrance, clinical severity, and timing of clinical onset, were not considered with respect to FGRC.[62,73] Therefore findings regarding FGRC in other hereditary conditions may not be generalizable to the HBOC context.

Evidence for optimal FGRC is based largely on retrospective descriptive study designs. For example, Srinivasan and colleagues found that 18 out 30 studies used qualitative descriptive study designs such as cross-sectional surveys and case series, 9 used qualitative, 2 used mixed-method and only 1 was a randomized controlled trial (RCT).[66] Many of the studies included in these reviews often had small sample sizes. In the review on the uptake of genetic

testing among at-risk relatives in HBOC and Lynch syndrome, the number of probands participated in the 13 HBOC studies ranged from 13 to 200.[48] With such small sizes, current studies may not accurately represent FGRC at the population level, limiting the generalizability of findings about the process, facilitators, and barriers of FGRC. Use of rigorous methods such as determining sample size needed to detect differences that are clinically and practically relevant for improving FGRC largely were not considered.

Lastly, FGRC reviews were published in the 2000s and 2010s. Thus, evidence generated might be somewhat dated to reflect current progress of FRGC needs. While most studies were conducted in the U.S., empirical evidence across countries may not be generalizable to diverse populations in the U.S. With these caveats, I will present considerations relevant to communication of genetic risk within HBOC families based on the FGRC definition.

1.2.2 Consideration Relevant to Family Communication of *BRCA1/2* Risk

1.2.2.a Considerations for whether and when to tell

Probands who: were female, younger, had higher incomes, were White, and those with higher numeracy levels were more likely to share their genetic testing results with their relatives.[43,45,46,74,75] Those who communicated with their relatives often did so to discharge their responsibility of informing the family, gain emotional support and advice, obtain specific family history information from relatives, and protect relatives from potential harms of illness.[52-54,76]

Little is known about the association between probands' HBOC cancer stage and whether to tell. MacDonald et al hypothesized that women with late-stage breast or ovarian cancer were more likely to communicate genetic risk within families, however, their data was underpowered to detect the differences because their sample size was small (women with late-stage cancer = 9).[77] The authors expected a difference because women with late-stage cancer would have a sense of urgency to increase risk awareness among their relatives.

As described above, probands generally have been found to report strong intentions to share their genetic testing result with their family. However, this intention was often assessed in

a binary manner (i.e., yes/no) using cross-sectional survey designs. Questions were often considered at the family-system level, meaning if study participants planned to tell even just one relative, the response would be “yes”. Evidence generated on these study designs may have inflated the extent to which probands intend to communicate.

In accordance with my working definition of optimal FGRC, discussing inherited risk is not a one-time decision. Indeed, studies have shown that most probands communicated to at-least one relative immediately after receiving genetic testing results.[61] For example, Fehniger and colleagues interviewed 73 *BRCA1/2* carriers, and found that among relatives who had been informed (439/606), around half were informed within a week after participants received their test result, while less than 5% found out after a year.[78] Others waited for longer periods and reported delaying because they wanted to wait until they had received all relevant information about their genetic testing result before talking with family. Others indicated no sense of urgency around sharing the information with at-risk relatives. Difficulties in finding the right occasion for FGRC was another reason for delayed communication. For instance, some probands delayed discussions with extended relatives until family gatherings.

In Lieberman et al’s longitudinal mixed-methods study described in *1.1.3*, 13 of 32 probands reported that they had informed at least one relative regarding their test results within the past 6 months at T2 (i.e., 2 years after receiving genetic testing results), suggesting that communication can be very delayed.[46] The authors also interviewed 26 probands in the same study. They found that probands reported delaying communication to find a right or convenient time, or to wait until they felt emotionally ready. Likewise, a qualitative study in Canada found that some hereditary cancer syndrome probands engaged relatives throughout the testing process, even before receiving results. The authors found that the timing of communication was often informed by participants’ comfort level with raising the topic of genetic testing as well as cancer status.[79]

1.2.2.b Considerations for who to tell

Throughout the literature, reports were consistent that probands are more likely to communicate with some at-risk relatives than others.[52,63,78,80] These patterns differ by relationship types, gender, and age of at-risk relatives. Probands tend to communicate *BRCA1/2* genetic risk information with FDRs, female relatives, and adult relatives, but less so with SDRs, male relatives, and young children. Probands are most likely to share genetic risk information with specific relatives when they perceive that the information has relevance to them, when they feel an emotional closeness with the relative, and based on their perception of family rules and patterns.[52-54,61]

Like "whether to tell", empirical evidence on "who to tell" was typically derived from probands' survey responses in cross-sectional descriptive studies. The results may be subject to self-selection bias, self-report bias, and recall bias. Typically, such studies often focus on the specific relatives who were actually informed. They rarely assess the completeness of communication in the family by considering which relatives were not informed.

Koehly and colleagues are among the few researchers to take a family system's approach to characterize whether to tell and who to tell. In a series of studies, the authors used a visual tool for assessing social interactions between study participants and their relatives and friends, namely the Colored Eco-Genetic Relationship Map (CEGRM), among 124 families with known mutations in the *BRCA1/2*.[75,81-85] The studies enrolled 183 female participants and identified a total of 5466 personal network members in their studies. Koehly regards family systems to comprise roles in which some members play the role of genetic risk information "gatherers", "disseminators", and "blockers".

When it comes to whether to tell, findings suggest that individuals who were female, and providers of social support (e.g., tangible assistance, and emotional support) were more likely to disseminate genetic risk information within the family; in contrast, spouses and males were more likely to impede the information flow. In addition, there was a difference in

intergenerational FGRC. Family members in the older or same generation as the proband were more likely to facilitate communication of genetic risk.

A follow-up study investigated reasons for why not to tell at all and who not to tell among 65 information blockers of 42 HBOC families using repeated annual individual CEGRMs of 4 years in the same population.[84] Findings suggest that blocking behavior was transient and based upon upsetting emotional and social situations (e.g., lack of insurance). However, in some families, information "blockers" held consistent ideological opposition to genetic testing over time. In this group, the reasons for blocking were heterogeneous. These "blockers" did so to avoid potentially upsetting social situations with specific relatives, to protect relatives or themselves from emotional fears of cancer information, and to protect their privacy. The notion of gender roles and identities in family traditions was highlighted as a barrier, negatively associated with the flow of genetic risk information among "non-communicative" men.[84]

Findings from studies that moved beyond probands' perspective suggest that a deeper understanding of the multi-level influences on "whether to tell" and "who to tell" is valuable in developing and tailoring family-based intervention strategies. These strategies, target underlying factors that are amendable to intervention and could advance the field from treating FGRC is a simple binary behavior of yes or no communication.

1.2.2.c Considerations for what to tell

The content of communication that probands used when discussing risk with relatives was generally examined as part of a larger research study. In these studies, results were based on proband preferences for timing and mode of communication rather than actual content of the communications. These studies included heterogeneous samples of probands with HBOC or other hereditary conditions.[64,86-88]

These studies find that probands prefer not to convey all genetic risk information at once. For example, an Australia qualitative study of *BRCA1/2* genetic risk found that probands preferred a staged approach in discussing genetic risk with their relatives, where they would

notify at-risk relatives of their increased risk either face-to-face or via a letter, then provide additional educational sources (e.g., website) for those wished to access more in-depth information.[88] Likewise, a Dutch study surveyed 316 probands with a known hereditary condition to assess their preferences about how to inform relatives of genetic risk information. The study found that a majority of probands (285/316) preferred to at first provide a limited amount of information with a possibility to provide more information later.[76]

A few studies have explored what information would be best to support FGRC.[58] Cragun and colleagues found that, among 235 female *BRCA1/2* carriers, printed materials (e.g., brochures, 66%), family letters (62%), and web-based information (e.g., HBOC websites, 48%) were ranked as the most helpful and the most commonly used resources.[45] Participants reported using more than one resource when they communicated genetic test results with relatives. Dheensa et al interviewed 115 healthcare professionals (n=80), and individuals with a person and/or family history of cancer (n=35) in focus groups.[39] The authors found that while family letters better explained genetic risk to relatives, there were difficulties in the process of using the letters. For example, participants felt that letters were too long and complicated while healthcare professionals worried about how directive the letter should be, in a way that information was clear but would not frighten families.[39] At the end, the authors suggested alternatives to family letters to facilitate FGRC, such as contacting at-risk relatives via web-based platforms.

In one of the few studies focused on FGRC content, Himes et al explored breast cancer risk perception among 85 unaffected FDRs of breast cancer probands in a mixed-methods study.[89,90] Very few participants (7%) reported that probands shared a great deal of information about their genetic counseling session, whereas 18.8% reported that probands shared nothing about what they learned in their genetic counseling session. An example of the brevity of what an FDR was told is exemplified in one participant reporting that she was told by the proband- "*I'm negative, but you should still get your mammograms.*" Himes et al also found a positive

association between the perceived amount of information shared and *BRCA1/2* knowledge among unaffected FDRs, that participants who reported higher levels of information shared also showed increased knowledge about breast cancer genetics.

In another survey study, Finlay et al assessed topics commonly discussed during family genetic risk conversations among 115 relatives of 47 probands with known *BRCA1/2* pathogenic variants. The study sample was relatives who had undergone genetic testing and received a positive result.[47] Topics such as "*cancer risk for people with a mutation*" (102/115) and "*chance of having a mutation*" (98/115) were commonly discussed with relatives, while topics such as "*information about genetic discrimination*" (50/115) were less frequently discussed. Because relatives who participated in this study have proactively sought genetic testing, these results may not be generalizable to less selected samples. Taken together, the available evidence suggests research concerning communication content remains sparse.

To date, most of the evidence about optimal risk information to convey has not taken into account the bi-directional nature of communication. For example, virtually no research has explored the level of understanding when at-risk relatives were being informed about inherited risk. Vos and colleagues surveyed 25 probands and 70 relatives in HBOC families about the recall and re-interpretation of genetic risk information received from genetic counselors.[91] The authors found that correlations between probands' and at-risk relatives' recall of the test result (e.g., positive, inconclusive) and re-interpretation of heredity-likelihood (i.e. not-complete at risk to heritable) were weak (r=0.07) to null (r=0).

Another study based in the United Kingdom conducted semi-structured interviews with *BRCA1/2* probands (n=10) and their at-risk relatives (n=22) to assess recall of general genetics information (e.g., inheritance, the gene involved) and hereditary cancer information (e.g., risk management options).[92] The authors found that relatives recalled significantly less accurate information about general genetic and hereditary cancer compared to probands (30% vs.. 53%). In addition, relatives who gained the information from the proband alone recalled significantly

less accurate information compared to those who received information from multiple sources (e.g., proband, genetic counselor) (15% vs. 52%). Evidence to date suggests that relatives often misinterpret what they are told. Yet, there is no evidence to suggest what information elements are needed to redress this misinterpretation and misunderstanding of genetic risk.

1.2.2.d Intervention efforts to promote family genetic risk communication

I conducted a systematic review of intervention studies published between 2010 and 2020 in Aim 1. The review was published in *Patient Education and Counseling* in 2022.[71] I found 9 intervention studies that evaluated strategies for promoting communication of family inherited risk information, each with a comparison group. Details of my review, along with an updated search to identify new interventions from 2020 to 2023, will be provided in Chapter 3.

In addition to my review, two other reviews focused on FGRC interventions. Baroustou et al (2021) evaluating family communication interventions provided in the contexts of HBOC and Lynch Syndrome found only 14 studies conducted over a 17-year period.[69] Of these included studies, 6 overlapped with my review. Three intervention studies were published before 2010, therefore were not included in my review. Family communication outcomes assessed in Baroustou and colleagues' review included the number of relatives contacted/informed about the pathogenic variant, as well as frequency of contact and openness/ease of family communication. The overall effect size for family communication outcomes was small ($g = 0.085$) and not significant ($p = 0.344$). In addition, cascade genetic testing outcomes included the uptake of genetic testing by relatives and/or contact with genetic services and request for genetic consultation. The overall effect size for cascade genetic testing outcomes was small ($g = 0.169$) and significant ($p = 0.014$). However, this significant finding should be interpreted with caution due to the small number of interventions and the self-report outcomes.

After my review in 2022, Ballard et al (2023) published a narrative synthesis of interventions, identifying 5 interventions to support patients in sharing genetic test results with at-risk relatives. Of them, 4 interventions overlapped with my review, with 2 of them focused

exclusively on HBOC. One intervention, which covered a wide range of medically actionable hereditary conditions in addition to HBOC and Lynch Syndrome, was not included in my review as it was published in 2008. The authors concluded that there was no increase in participants' knowledge, motivation, or self-efficacy regarding whom to inform and what information to share. Among interventions that recruited both female and male probands, no gender differences were found in communication behavior.

Taken together, these reviews found limited benefits of current FGRC interventions. Most showed no significant improvement for promoting family communication and/or cascade genetic services compared to the usual care group. Interventions included in these reviews also varied in the theory utilizations, intervention fidelity (e.g., adherence to the study protocol), and intervention "dose" (e.g., duration of the intervention). The predominant null findings highlight a research gap concerning effective family communication intervention strategies.

1.2.2.e Summary: gaps and opportunities of family genetic risk communication research

Evidence suggests that probands are highly selective in whether and when to tell, who to tell, and what to tell. Probands most often choose to share with female relatives with whom they share a close emotional bond. Additionally, probands and relatives' function in family systems where some relatives can block information sharing. There is little guidance on the key information to share with relatives and it appears that relatives often misunderstand what they are told. Moreover, most of what we know is based on probands, who are disproportionately White. Meanwhile, interventions aimed to increase the likelihood of probands communicating genetic risk to relatives have largely been unsuccessful.

1.3 OVERVIEW OF CONCEPTUAL FRAMEWORKS AND BEHAVIORAL THEORIES

I base my proposed dissertation studies on the premise that a family is a complex social *system* based on patterns of emotional and support exchanges between its members.[93,94] In this view, changes in one family member influence the social functioning of other members – how

they interact, communicate with, and support each other, that in turn, can lead to behavioral changes in the unit.

In this section, I will provide an overview of communication and ethical frameworks and behavior change theories that will guide me in moving from the current focus of interventions to influence individuals (i.e., probands) and consider relational frameworks and ethical principles. The latter frameworks and principles view the family system as a web of relationships that surround the proband and influence the extent to which a proband communicates about inherited risk.

I argue that individual-level behavior change theories are also relevant to consider as ultimately actions taken or not taken to communicate cancer genetic risk are probands individual-related behaviors. Of course, influences on health behaviors operate at multiple levels and as such, align well with relational and family system frameworks.[95] Thus, I organize the behavior change theories at three levels: individual level, interpersonal level, and family systems level. Following this section, I will zoom into relational theories that provide conceptual guidance for developing my dissertation studies.

1.3.1 Communication Frameworks

Guidelines informed by communication frameworks could be useful in promoting FGRC. Those of greatest relevance derive from health provider-patient communication. These frameworks were developed to guide health providers to engage in sensitive conversations with patients such as those in life threatening diagnoses.[96] The most commonly applied framework is Buckman's "Breaking Bad News" framework, [97-99] which considers a serious medical diagnosis as an incident of "bad news". This is any notification of a diagnosis that will adversely and seriously affect one's future.[97,98] Such bad news can be intensely emotional for patients and hinder their ability to process and integrate important directive information. To this end, Buckman's framework proposes the following guidelines for providers when delivering bad news concerning health: (1) do so in person, (2) find out how much the patient knows, (3) find out how

much the patient wants to know, (4) share medical information, (5) respond to the patient's feelings, and (6) plan follow-through medical interventions.

In line with Buckman's framework, other serious illness communication frameworks add additional nuanced goals such as eliciting hopes and values and encourage the family to work together to plan for the future.[100,101] For example, the Serious Illness Conversation Guide suggests applying focused language to the conversation. For example, when sharing a medical diagnosis, words like "*I wish we were not in this situation*" could foster a sense of "togetherness" for the patient.

Indeed, these frameworks have been applied in the context of FGRC. Daly developed a six-step skill-building approach to prepare probands to communicate *BRCA1/2* genetic risk information with relatives.[97,98,102] Daly's framework encourages probands to: (1) plan for discussions with relatives, (2) consider optimal settings for the discussions, (3) anticipate information needs of specific relatives, (4) share genetic testing information, (5) respond to at-risk relatives' feelings and reactions, and (6) plan follow-through after communication. Daly's framework aims to build skills and confidence in probands as they consider whether, who and what to communicate with at-risk relatives.

The same study team conducted an RCT to promote FGRC among HBOC families.[103] The intervention included a skills-building session modeled on Daly's framework. The intervention delivered the stepped strategies to 137 probands in the intervention group, and a wellness session to 112 probands in the control group. There was no difference in the percentage of probands who shared test results with at least one relative (99.3% vs. 99.2%; p=0.59). A follow-up survey of 561 FDRs found that intention to pursue cascade genetic counseling was low among at-risk relatives of both groups.[41] However, this framework underpins on probands' individualistic portrayal of autonomy in FGRC, such as self-determination, self-awareness, self-interest.[104] I argue that the relational aspect, that is

considerations of connectedness and interdependency, should be adopted in the context of FGRC.

1.3.2 Ethical Frameworks

Relational autonomy, based in feminist perspectives, is an ethical framework that builds upon the more individual focus of the bad news communication models. Relational autonomy holds that the proband is socially situated in an interdependent web of relationships.[105,106] These relationships are characterized as coming with history, associated obligations, and responsibilities that will influence the proband's decisions about whether, who and what to communicate about inherited risk.[107,108] Scholars argue that the individual-centered communication theories ignores values such as mutual responsibility, support, love, and trust.[106]

Reconceptualization of autonomy in relational terms is not well integrated in clinical genetic services. In a qualitative study conducted by Shepherd and colleagues in Australia,16 genetic counselors were interviewed to assess whether a relational approach has been employed in genetic counseling.[109] As defined by the authors, "a *relational approach considers that individuals are not isolated social units—akin to atoms floating in a social vacuum, detached from their surrounding social environment*." Findings suggest that a relational approach was covertly employed by assessing familial relationships and dynamics. However, explicit discussions about the obligations and responsibilities to relatives were usually missing. This Australian study provides preliminary evidence on the integration of a relational approach in genetic counseling.

Another Israel study interviewed 28 health care professionals (e.g. genetic counselors, nurses, doctors et al) to understand how the presence of relatives in *BRCA1/2* genetic counseling sessions reflects the application of relational autonomy. The authors argue that having relatives present during the counseling sessions indicates an application of relational autonomy because involving other relatives enables relatives' who are present to express opinions and views.[110]

The CEGRM studies shed lights on how to integrate relational autonomy during genetic counseling.[75,81-84] As previously described, the CEGRM is a psychosocial assessment tool that displays social interaction domains of information, tangible services, and emotional exchanges between probands and relatives, and non-biological social connections (e.g., non-biological family, friends, coworkers). The visual representation of social assessment is designed to foster participants' emotional bonds, promote mutuality of these bonds, and increase experienced empathy in relationships. In this way, the guided processes of family characterization can engage the proband to lift-up emotional connections and bolsters probands' considerations of the interconnectedness of shared genetic risk. While conceptually promising, further studies are needed to examine how relational tools such as CEGRM should achieve a larger impact on FGRC compared to the current standard approach.

1.3.3 Behavior Change Theories: Individual Level

As described earlier, FGRC rests on proband individual behaviors. I will give a brief overview of individual-level theories that are relevant to probands' competencies and behaviors relevant for improving FGRC. Three conceptual models dominate in this discussion and are closely related: Health Belief model,[111] Social Cognitive Theory,[112,113] and the Theory of Planned Behavior.[114,115]

The core elements of Health Belief Model[111] suggest that a proband will communicate risk if they perceive HBOC to be a severe threat and they expect that there is benefit to their relatives to communicate. Thus, FGRC is most likely to occur when a proband perceives HBOC to be a very serious threat to which relatives are susceptible, and that the benefits of informing relatives outweigh the barriers. In turn, Social Cognitive Theory[112,113] suggests that the key driver of FGRC is a proband's level of self-efficacy, that is, the proband has confidence in their ability to effectively communicate genetic risk information. Self-efficacy may be especially salient to genetic risk communication as it involves conveying complex medical information and uses unfamiliar concepts and jargon. Thus, FGRC is most likely to occur when a proband has

high confidence that they can successfully discuss genetic risk information, such as patterns of genetic risk and prevention options, with at-risk relatives.

The Theory of Planned Behavior[114,115] regards probands' intention to engage in FGRC as having critical importance for risk communication, such that the stronger the proband's intention to engage in FGRC, the more likely it will occur. In turn, intentions are driven by attitude (e.g., aligned with the Health Belief Model's concept of outcome expectancies -- such as communication will protect at-risk relatives from developing an inherited disease), social norms (e.g., aligned with Social Cognitive Theory where views of important others can motivate family communication), and perceived behavioral control (e.g., aligned with Social Cognitive theory's concept of self-efficacy).

The appeal of individual-level theories is, again, whether probands communicate genetic risk information with at-risk relatives or not comprises a set of individual behaviors. These theories suggest behavioral determinants that are amenable to interventions to promote enhanced communication. Thus, one would expect that individual-level interventions aimed at promoting FGRC would emphasize probands' outcome expectancies, perceived self-efficacy, and intentions to engage in communication, however, reviews on the current FGRC interventions suggest that they haven't uniformly done so.[69,70]

1.3.4 Behavior Change Theories: Relational Level

Interdependence is a key attribute of family communication at the relational level. It is important to recognize that each proband-relative dyad within a family has a unique relational pattern and level of interdependence. These relational factors, in turn, significantly impact their individual and collective responses to genetic cancer risk. In this section, I will discuss two relational-level theories that serve as theoretical bases of my dissertation: Interdependence theory and communal coping theory.

1.3.4.a Interdependence theory

Interdependence theory, derived from social exchange theory and game theory, was originally proposed to characterize the process by which a dyad can influence each other's behavior motives and actions during a course of social interactions.[116,117] At the heart of interdependence theory is "*transformation of motivation*" (*TM*), a psychological shift in motivation from consideration of immediate self-interest when taking actions to broader consideration of another and/or the collective's interests. Interdependence theory highlights that probands engaging in *TM*, in which considering the best interest of the relationship or the important other instead of oneself, is likely to be key in driving optimal FGRC.[118-121]

The *TM* shift is conceptualized to consist of two domains: a cognitive interpretation of a health threat (e.g., inherited risk of cancer) presenting an existential threat to the "we"; and an emotional interpretation that such a threat has significant meanings to the self, the relative, or the relationship (e.g., fear for the wellbeing of at-risk relatives), which inspires a drive to protect the "we." In some cases, *TM* is thought to occur instantaneously, as part of automatic and habitual responses to social situations, with little cognitive or emotional interpretations.[120] This is likely influenced by other family characteristics. However, the thinking is generally that such transformation, whether spontaneous or effortful, increases the likelihood of jointly behaviors that are mutually beneficial.[121]

In the context of FGRC, *TM* is a process in which probands' motivations become expanded or transformed to include or even focus on the interests of at-risk relatives.[118,122] When a proband learns about their relatives' increased risk for cancer due to their own diagnosis or genetic testing results, their motivation shifts from self-interest (e.g., seeking emotional support) to relationship-focused interests (e.g., sharing information to support each other emotionally). Thus, interdependence manifests in a dyadic sense in which preserving the relationship is the key for engaging in action to reduce a health threat that is shared by genes.

1.3.4.b Communal coping theory

Proposed by Lyons and colleagues, communal coping is prompted when two individuals appraise stressful events together and respond to one's stressful event in a collective and cooperative way, such as reciprocal exchange of support.[123] Communal coping emphasizes the embeddedness of proband-relative dyadic relationships, therefore any change in proband naturally affects their relatives, and affects the relationship as a whole.

Communal coping is conceptualized as comprising two dimensions: appraisal and collaborative coping.[123-125] The appraisal dimension is conceptualized as "*the degree to which problems are construed as shared or individually owned*", in other words, whether inherited cancer risk is appraised as a shared "we"-problem, or an individualistic "my"-problem" or "your"-problem within families. The collaborative coping domain is conceptualized as "*the degree to which coping strategies will be mobilized by involved partners or by the individual*", in other words, whether activation of a collaborative coping process regarding cancer genetic risk is appraised as a collective "we"-responsibility or an individualistic "my"-responsibility or "your"-responsibility.

Communal coping lies in the dimensions of "we-problem" and "we-responsibility", signifying a sense of "we-ness"-thinking that alters both probands and relatives' perspectives from "all about me" or "all about you" to "it's all about us". Other categories of the coping quadrant model, based on the two dimensions, are individualism ("my"-problem and "my"-responsibility), individual help/support provision ("our"-problem and "my"-responsibility), and help/support seeking ("my"-problem and "our"-responsibility).[123]

Similar to *TM,* activation of communal coping could be spontaneous or effortful. In some cases, it could emerge spontaneously as part of an ongoing relationship, or it could be a deliberative choice in absence of such a relationship, where dyads engage in communal coping when one dyad identify the other is facing the same genetic cancer risk.[123]

1.3.4.c Previously proposed integrative model of transformation of motivation and communal coping

Here I briefly note previous theory development efforts aimed to integrate *TM* and communal coping. Lewis and colleagues proposed an integrative model, based on principles and constructs of the two theories, that explicitly considers marital interactions as determinants of romantic couple's behaviors in adoptions of risk-reducing heath habits (e.g. smoking cessation).[118] In their conceptualization, communal coping is activated by *TM*. That is, *TM* occurs when partners consider health threats as meaning for the other partner and their relationship in the context of relational roles and norms. These considerations would prompt thoughts and feelings about attaining mutual beneficial outcomes. The use of communal coping was conceptualized as the utilization of strategies that are communal in natural, such as couples' communication about behavior change, to engage in health-enhancing behaviors. To this end, the couple's motivation becomes more pro-relationship or partner centered, rather than self-centered, and their likelihood of working collaboratively is enhanced.

Lewis and colleagues' model acknowledges the potentially transformative nature of intimate relationships. However, it is less specific in the context of FGRC as there is a lack of emphasis on the shared nature of inherited cancer risk conferred through biological ties. This health threat is intertwined within a complex web of proband-relative dyads, where every dyad may interpret the threat differently based on their relational context and family dynamics. After reviewing two relevant theories at the family-system level, I will further discuss my conceptualization of the integrated *TM* and communal coping model to inform the development of dissertation studies.

1.3.5 Behavior Change Theories: Family-system Level

Family genetic risk communication rests on relational interactions occurring within an interdependent family system. Achieving greater completeness in FGRC requires moving beyond relational theories and incorporating considerations of multiple proband-relative dyadic relationships as a whole. Two theories lend insight into how families may communicate HBOC

as a means to promote health-enhancing behaviors at the family level: Family Systems Perspective (FSP),[126] and Family Communication Patterns theory (FCPT).[127]

Peterson's FSP outlines three family system characteristics related to how probands and the family as a whole cope with a hereditary condition.[126,128] These include: (1) family communication, (2) organization and structure of family relationships, and (3) health-related cognitions and beliefs shared within families. FSP emphasizes that family communication serves a social support function that builds and strength family ties and facilitates coping with external threats.[126] This line of thinking suggests that a proband's HBOC diagnosis will prompt a wider reciprocal exchange of information, as well as emotional, and instrumental support within the family system. Similar to *TM*, family members may respond to the health threat with motivation to collaborate and work together towards managing shared risk.

On the other hand, FCPT theory suggests that the information flow within a family is influenced not just by the immediate meanings and demands of an HBOC diagnosis but by norms within the family that have been established over time.[127,129] These norms include "*the degree to which family members interact and communicate about various topics*" (i.e. conversation orientation) and "*the degree to which a family expects uniformity of beliefs, attitudes, values, and behaviors*" (i.e. conformity orientation).[127,130,131] The more families communicate regularly and openly about a health threat, the more likely they are to be aware of each other's information and emotional needs. In turn, this awareness can promote family-level risk reducing behaviors. This process is augmented when families have a conformity orientation, that is, members support an open family communication environment. Such an orientation enables a freer flow of information. And it is also assumed that freer information flow, in turn, increases the likelihood that family members will appraise genetic risk as a shared health threat.

Upon reviewing relevant communication and ethical frameworks, and theories at the individual, relational, and family-system levels, it becomes evident that *TM* and communal coping may be particularly applicable for understanding how to foster effective FGRC. That is,

while communication must be initiated by probands, the desire to act in the best interest of the relationship or the relative instead of themselves turns FGRC into a relational transaction aligned with actions that preserve the relationship. In the concluding section of the Chapter, I begin by presenting the conceptual model of integrated *TM* and communal coping, which serves as the theoretical premise for my dissertation studies. I also provide an overview of current research on *TM* and communal coping, highlighting research gaps in measurements.

1.4 ADVANCING TRANSFORMATION OF MOTIVATION WITH COMMUNAL COPING

1.4.1 Dissertation Conceptual Model

1.4.1.a Adding a "we"-solution dimension to communal coping

Based on communal coping theory[123] and the integrative model proposed by Lewis et al[118] the process of communal coping involves three components: (1) dyads hold beliefs that joining together to deal with the health threat is beneficial, needed, and/or expected; (2) communication about the details and meaning of the threat is essential if coping efforts are to be shared; and (3) dyads engage in cooperative coping to reduce the negative impacts of the health threat. As outlined, communication is characterized as an intermediate step between "we"-problem and "we"-responsibility.[123]

In the context of FGRC, I propose adding the additional dimension of "we-solution". My thinking is that when probands hold beliefs on "we"-problem and responsibility regarding inherited cancer risk, they would be more inclined to see FGRC as a beneficial solution for addressing the problem and their responsibility to protect at-risk relatives. I conceptualize the "solution" domain as "*engaging in individual or joint actions to reduce the threat*".

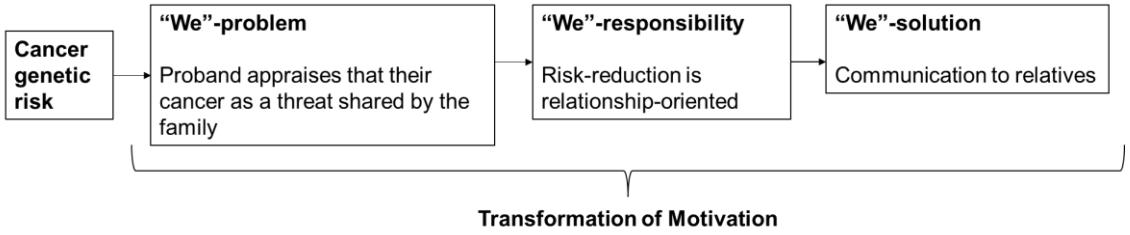
1.4.1.b Final conceptual model

TM of interdependence theory and communal coping share the assumption that the likelihood of optimal FGRC increases when both survivors and relatives experience a sense of "we-ness" in response to threats from a high-risk genetic test result. Interdependence theory

hypothesizes that transformations in motivation – away from the self-centered orientation and towards the pro-relationship orientation – increase the likelihood that dyads will engage in behaviors (or solutions) that reduce the impacts of existential cancer genetic risk shared in "we".

The three indications that probands have experienced a *TM* shift from self-interest to relationship-focused interests is evidence of "we-thinking". Expressions of seeing the health threat as a "we"-problem that becomes a communal responsibility, and requires we actions for the solution. For this dissertation, I define "we"-problem as "*probands appraising that their cancer as a health threat shared by the family*"; "we"- responsibility as "*risk-reduction is relationship-oriented*"; and "we"-solution as "*communication to relatives*". This cognitive and emotional shift is what I and others believe is required to prompt communal coping. I depict these assumptions in *Figure 1A*. I posit that the extent to which survivors regard their cancer as inherited they will view it as a "we-problem". In turn, this "we-ness" thinking about the problem as shared will increase the likelihood that the proband appraises that joint action is needed to reduce risk ("we"-responsibility), and to take a course of action that is required to manage the threat ("we"-solution) in the form of communication to relatives.

Figure 1A. Conceptual model of integrated transformation of motivation and communal coping



1.4.2 Current Research in Transformation of Motivation

To date, efforts to characterize whether the *TM* operates in interpersonal relationships and the mechanisms that underpin it have been sparse. In two studies, Yovetich and Rusbult investigated *TM* in situations where romantic dyads faced conflicts that could affect their relationships.[132] In the first study, they hypothesized that when facing these conflicts, a romantic partner will appraise their broader interaction goals (e.g., eliminating bad feelings in an on-going

relationship) and the range of possible consequences of taking different actions. For these couples, the *TM* process involved the partner filtering out more destructive options (e.g., threatening to break up) and chose behaviors that were less harmful to the relationship (e.g., tried to fix things). To further test this process, 53 college students were asked to retrospectively recall the behaviors that they considered enacting in response to a conflict with a romantic partner, and the behaviors that they actually enacted. The authors found that while the participants did consider destructive options in response to the conflict, they engaged in a cognitive process to broadly consider possible harm to the relationships and chose less destructive options.

In the second study, Yovetich and Rusbult further hypothesized that one's cognitive *TM* tendency would be positively associated with processing time.[132] To test this, the authors designed a 2 x 2 x 3 experiment with 80 college students. Participants were randomly assigned to reaction time conditions (limited vs. plentiful), scenario type (constructive vs. destructive), and relationship type (dating relationship vs. friendship vs. family relationship). An example of a constructive scenario is "*a friend helps you with a very difficult assignment*", while an example of a destructive scenario is "*during an argument, your partner says – sometimes I think I'd be better off without you*." Findings suggest that cognitive *TM* processes require greater message elaboration and more processing time. Participants were more likely to endorse self-oriented actions if they were given a limited time to respond to the threat.

Chen and colleagues replicated these two classic studies using a cross-cultural sample of college student participants from the U.S. and Thailand.[133] Consistent with the original study 1, participants (n=90 in the U.S. and 97 in Thai) were asked to consider increasingly more destructive responses when facing a conflict. The study found that , they actually enacted in a more pro-relationship manner. In a replication of study 2, college students (n=162 in the U.S. and 166 in Thai) chose significantly more relationship destructive responses in the limited time condition compared to the plentiful time condition. In addition, no cross-cultural differences were

found in this process. These studies show that college students in close relationships were willing to take accommodative, relationship-oriented actions, reflecting some aspects of the *TM* latent construct. However, it remains unclear how *TM* operates in health threat contexts when relationships are familial and not romantic.

Evidence to support the existence of *TM* is nascent in the area of health threat response. One qualitative study evaluated how pregnant couples living with HIV would respond to recommended preventive health behaviors. *TM* was operationalized as couples exhibiting more relationship-focus in completing an intervention and adopting positive health behaviors.[134] The authors observed that *TM* was expressed in various ways, such as disclosure of HIV-positive status and fidelity. The authors also observed differences in the timing of *TM*. Some couples chose to engage in the intervention before initiating sexual intimacy, but the majority experienced a more gradual *TM* process, shifting their behavior from engaging in concurrent sexual partnerships to being more faithful. Consistently, these studies found that romantic partners have the tendency to take account of broader considerations in preserving the romantic relationship, as highlighted by *TM*.

1.4.2.a Factors associated with TM

In addition, a small body of research has investigated factors associated with the occurrence of *TM* thinking and action, again predominantly in romantic couples. These studies have consistently found factors such as relationship satisfaction, relationship investment, and feelings of commitment to be associated with increased cognitive interdependence.[135-137] For example, Davis and colleagues found in a cross-sectional survey of 275 individuals in romantic relationships, that cognitive interdependence was positively associated with the inclusion of partners in one's concept of self, and positive thoughts about the relationship.[136]

Self-control, that is one's ability to regulate thoughts, emotions, and behavior in a goal-directed manner, has also been associated with "relationship-preservation motives" (e.g., partners' willingness to sacrifice on the other's behalf).[138] Pronk & Righetti proposed that self-

control allows individuals to overcome "I" destructive behaviors and to act constructively according to "we" long-term goals in an opinion paper.[139] Relatedly, Finkel and Campbell, in a survey study of self-control and accommodative tendencies, found that participants whose cognitive resources were depleted (e.g., overwhelmed by school/work, preoccupied with other things) were less capable of engaging in pro-relationship *TM*, and exhibited weaker accommodative tendencies in response to partner behavior.[140]

Research on *TM* is still in its early stages, with no validated and reliable measures. This lack of measurements limits research understanding of whether, to whom, when and how *TM* occurs in dyadic or broader social networks.

1.4.3 Current Research in Communal Coping

Communal coping also have been studied predominantly among romantic couples and parent-child dyads facing chronic disease management, such as type-2 diabetes, and substance use disorders.[141,142] Emerging evidence suggests that communal coping is associated with better relationship quality, improved psychological well-being, and positive health behavior changes and better physical health.[124] For example, a focus group interview study, conducted by Beverly and Wray, found that when a couple perceived collective benefits of managing diabetes in a team approach (i.e. "*we are in this together*"), the patient dyad was more likely to be adherent to exercise schedules.[143] Similarly, Zajdel and colleagues examined whether communal coping was linked to daily mood and self-care behavior among 123 couples with type-2 diabetes. Using a daily diary questionnaire that assessed communal coping and mood for 14 consecutive days, the study found that those who engaged in communal coping reported better adherence to their medication.[144]

1.4.3.a Communal coping research in cancer

Fewer studies have focused specifically on communal coping in the context of cancer. For example, Wolf conducted 64 interviews with breast cancer survivors, partners, and relatives among 20 families to understand how families might collectively cope with breast cancer.[145] The

author coded linguistic cues that are indicative of ownership discourse (individual vs. communal) when talking about the breast cancer experience (e.g., treatment, hair loss, family rituals). Findings suggest that family members viewed the breast cancer experience as a disruptive experience for individual survivors, but less for the family. The author concluded that communal coping was not highly prevalent among family members as indicated by discourses regarding the cancer experience. Thus, based on this exploratory qualitative study, communal coping may not be an automatic response when family members face the cancer experience.

Other studies investigated communal coping in the context of inherited cancer risk within larger social network systems. For example, Marcum and colleagues took a social network approach and operationalized communal coping as emotional support exchange through the pattern of supportive relationships within a family network (i.e., *which members of your family support you emotionally?*)[146] The study is based on 6 Lynch syndrome families and 12 interviewed informants. Using exponential random graph models, the study found that sharing common genetic testing status (e.g., received or declined genetic testing) was marginally associated with the formation of emotional support exchange. On the other hand, the flow of communal coping was not limited to the boundaries of biological family. Family members who did not share biological risk for inheriting Lynch syndrome also provided reciprocal emotional support exchange within the networks.

Similarly, Koehly and colleagues investigated the association between mental distress and communal coping characteristics among 65 sisters from 31 HBOC families.[85] Communal coping characteristics were derived from the CEGRM (*first described in 1.2.2.b*), and was operationalized as reciprocity of support (i.e., *whether both sisters selected each other as providing a given support type*) and shared supports (i.e. *the number of persons providing support to two or more participating sisters*). Using hierarchical linear modeling, with sisters nested within their families, the study found that shared supports of communal coping were

negatively related to anxiety and somatization, however, reciprocity in support among sisters was not significantly associated with reduced distress.

These studies on communal coping with inherited cancer risk have moved beyond interactions to consider exchanges of support and coping within a broader system. These few studies taken together suggest that cognitive and emotional shifts do occur to preserve relationships in health contexts. However, each of the studies operationalized *TM* differently based on assessments of relationships rather than evidence of "we-thinking" and communal coping. Thus, important research gaps remain regarding the transformative nature of protecting family relationships as a means to prompt communal coping.

1.4.4 Measures of Transformation of Motivation & Communal Coping

1.4.4.a Operationalization of transformation of motivation

To date, there are no validated measures of *TM* conceptualized as a cognitive and/or emotional shift where one's motivation becomes expanded or transformed to include or even focus on the interests of others in a pro-relationship orientation. As a self-report measure, Agnew and colleagues developed a four-item cognitive interdependence scale (α = .82) for assessing romantic relationships. A sample question is that *"In comparison to other parts of your life [e.g., work, family, friends, religion], how central is your relationship with your partner? 1= not at all central, 7 = extremely central).*[135,136] However, what is missing is the identification of dimensions and development of specific items to accurately capture the underpinnings of *TM*.

1.4.4.b Linguistic markers of transformation of motivation

TM is commonly assessed using a linguistic approach, that is, counts of the first-person plural pronouns (e.g., we, us, our) relative to the total words in a study interview (e.g., "we"-talk).[141,147-150] Here, "we"-talk is regarded as an implicit marker of *TM*. When couples talk about a health threat, a higher frequency of plural pronouns could reflect a communal oriented self-construal, while a higher frequency of first-person singular pronouns (e.g., I, me, mine) reflect a more individual oriented self-construal.

A growing body of research has examined “we”-talk in the “we”-problem appraisal dimension of *TM*.[124] For example, in Zajdel et al’s study on 239 caregivers and 124 children with rare diseases, “we”-talk was operationalized as a measure of caregivers’ shared appraisal of whether caregiving is their own stressor to manage or one that is shared among others in their network.[151] Findings suggest that “we”-talk was associated with members of caregivers network to be involved in communal coping that is, direct care of the child and to provide support to the caregivers.

In a couple-focused smoking cessation intervention study, Rohrbaugh and colleagues counted “we”-talk utterances from transcripts (measure of *TM*) of a baseline marital interaction task and intervention sessions. They found that the spouse’s baseline “we”-talk was associated with the smoker partner’s abstinence 12 months after quitting.[147] Similarly, Rentscher and colleagues conducted a couple-focused intervention for problematic alcohol use. They also found an association between the spouse’s “we”-talk (evidence of *TM*) and the alcoholic partner’s maintained abstinence for 30 days prior to treatment termination.[152]

It is important to note that associations of “we”-talk with communal coping have been inconsistent. For example, Davis and colleagues coded participants' relationship thoughts into four categories (i.e. plural pronoun use only, single pronoun use only, mixed plural and singular pronoun use, and no pronoun use), and used it as a quantitative variable to examine its association with participants positive relationship thoughts.[136] They found that plural pronoun use was not a meaningful component of cognitive interdependence in the study.

While being an objective and easily quantifiable measure to assess *TM*, we talk counts have notable limitations. Such counts may not necessarily capture the nuanced meanings behind the use of this pronoun. Especially in different social contexts, like families or broader social networks, "we" can carry different connotations and implications. Therefore, the interpretation of “we” use need to be considered within the full context of the conversation to avoid oversimplifying this measurement.

Measures of the “we”- problem domain have been assessed with questions such as “when you think about problems related to your heart condition, to what extent do you view those as our problem' (shared by you and your spouse equally) or mainly your own problem?” [153] The “we”-responsibility domain has been assessed by questions such as “when a problem related to your diabetes arises, how much do you and your spouse work together to solve it?”[124]

Additionally, there has been efforts to directly observe dyads’ interactions and discussion about a shared threat. Observations are converted to quantitative codes based on a five-point scale ranging from 1 (“*low in communal coping*”) to 5 (“*high in communal coping*”). Questions are posed such as “*to what degree does the patient/spouse view the problem as one individual’s (“my” or “your”) problem or a shared (“our”) problem?*” And “*to what degree does the patient/spouse deal with the problem by working alone or working together as a team?*”

Some studies have used daily diaries as a repeated measure to assess shared appraisal and collaboration.[150,154] Participants answer survey questions such as, “*when you thought about diabetes today, did you view diabetes as ‘our issue’ (shared equally by you and your partner), mainly your own issue, or your partner’s issue*?” daily over a period of time. Responses range from 1 (“*completely my own issue*”) to 5 (“*completely my partner’s issue*”).

Another approach is to assess *TM* is using graphic representations.[124,148] For example, the Inclusion of Other in Self scale (*Appendix 1*), asks a subject to select from a graphic showing different levels of overlapping circles to show their perception of their sense of being interconnected.[135,136,155] Dyads are asked to choose the pair of overlapping circles ranging from two mutually exclusive circles to two nearly overlapping circles that best represents how they cope with a threat. The closer the two circles overlap, the higher the degree of communal coping the dyads perceive.

Several limitations of *TM* research are notable. First, *TM* has only been vaguely conceptualized and there are no validated measures of *TM*. While there has been considerable theoretical conceptualization of *TM*, very little research has occurred specific to inherited cancer

risk. Inherited cancers are an optimal context to further explore *TM* as affected families may be especially likely to experience *TM* and adopt more communal coping. Efforts to understand and conceptualize *TM* in the context of FGRC has importance for development of interventions that could expand at-risk relatives' awareness of their risk and encourage preventive actions.

1.4.4 Summary of Chapter 1

I have provided an overview of the importance of FGRC for families at risk for inherited cancers and the challenges they face regarding whether, when, who and what to tell. I have shown that the current landscape of FGRC interventions is small and lacking in rigor. I have reviewed relevant communication and ethical frameworks and behavior change theories at individual, relational, and family-system level. These theories served as guides in shifting from the predominant focus on influencing individual proband behaviors to considering the relational influences on FGRC.

Central to my dissertation is the assumption that effective communicating about genetic risk rests on relational interactions occurring within an interdependent family system. To this end, I focus on the guiding theories of my dissertation – *TM* from Interdependence theory and communal coping. In the next Chapter, I will provide a detailed overview of my dissertation design and the three specific aims .

CHAPTER 2. OVERVIEW OF THE DISSERTATION

2.1 DISSERTATION OVERARCHING AIM

My *overarching aim* is to advance our understanding of how to measure the construct of transformation of motivation (*TM).* Quantifying *TM* would enable researchers to test interventions that are aimed to encourage communal coping. In the context of my dissertation, this would be to increase communication about inherited risk among families. To address this broad aim, I first conducted a systematic review of current family genetic risk communication (FGRC) intervention studies (Aim 1). My objective was to explore the landscape of strategies used and the extent to which *TM* and communal coping were considered.

My second step (Aim 2) was to test whether and to what extent ovarian cancer survivors' engagement with inherited risk information provided on a website might indicate the experience of TM. I used data from a large ongoing study, Your Family Connects (YFC), a randomized controlled trial (RCT) that evaluated a message-based intervention grounded in relational and family systems level theories.[156-158] The YFC study offered a website for survivors of ovarian cancer to build skills in FGRC and offer free genetic counseling. I propose to code real-time website usage data and conduct factor analyses to identify use patterns that are suggestive of *TM*.

My third step (Aim 3), I propose to expand on the data collected in Aim 2 with qualitative insights into usage patterns identified through structured interviews with survivors of ovarian cancer while they use the YFC website via the think-aloud approach.

2.2 THE YOUR FAMILY CONNECTS INTERVENTION STUDY

In this section, I will first provide an overview of the YFC intervention study design. Then, I will describe the set-up and content of the YFC intervention arm. This section focuses specifically on the intervention arm of the YFC study, as my dissertation research involves mapping use of the intervention arm website as indicators of the TM.

2.2.1 The Your Family Connects Intervention Study Design

The YFC intervention study was designed as a two-arm RCT to compare YFC to the standard cancer registry outreach. The primary outcomes of the trial were the number of ovarian cancer survivors and their close relatives who logged on to the study website by arm. The YFC study was funded by the National Institute of Health (grant number#: 5U01CA240581-02).

Survivors of ovarian cancer were identified through the Georgia Cancer Registry (GCR). Eligible participants of YFC were: 1) diagnosed with ovarian, fallopian tube, or peritoneal cancers between January 2005 and December 2017 in the state of Georgia; 2) residents of Georgia at the time of diagnosis; 3) were alive at the time of invitation based on GCR records; and 4) verified to have a mailing address. A total of 1,938 eligible ovarian cancer survivors were randomly assigned to either a Message based intervention (MBI, n=969) arm or the standard GCR outreach arm (n=969) in a 1:1 ratio.[158]

Survivor participants in the MBI arm were provided with access to an interactive website, *yourfamilyconnect.org*. MBI participants were given access to the YFC website that included content on using different contact options for reaching at-risk relatives, and how to engage in sensitive conversations. An overview of the content with each webpage section is shown in *Table 2A*. In brief, the website features five informational and interactive sections - *Importance of Family Communication*, *Sensitive Conversation Framework*, *Invite Family Members*, *Schedule Genetic Counseling*, and *Other Resources*. These sections are accessible through tabs on the website's dashboard or homepage. Survivors in the MBI arm also were asked to first names of all first- and second-degree relatives (FDRs and SDRs) on the website and select

options for contacting listed relatives to participate in the study that included study team outreach.

Table 2A. The Your Family Connects website content

Head section	Content
Importance of Family Communication	Overview of who could benefit from learning an inherited risk for ovarian cancer Describe motivations to inform close relatives about inherited risk Describe best ways to contact close relatives Describe the menu of three relative contact options
Invite Family Members	Enumerate and invite relatives in 4 steps 1. Enter relatives' first name and specify relationship (e.g., daughter) 2. Select preferred contact option for each enumerated relative 3. Provide relatives' contact information if selects *study team contact* 4. Option to download customizable family letters
Sensitive Conversations Framework	Describe the 5-step sensitive conversation tipsheet 1. Where do I start? 2. What does my relative know about our shared cancer risk? 3. How do I discuss the effects of this cancer on our family? 4. How do I prepare for my relative's reactions? 5. How do I approach the option of genetic counseling with my relatives?
Schedule Genetic Counseling	Background information on genetic counseling and genetic testing Four-question genetic counseling questionnaire Schedule a genetic counseling appointment
Other Resources	Links to four external ovarian cancer resources

The GCR outreach arm received access to a website containing generic information about communication and genetic counseling access. The GCR website offered survivors to access generic contact letters for them to provide to at-risk relatives. Participants in both arms could schedule a family or individual genetic counseling session at no charge.

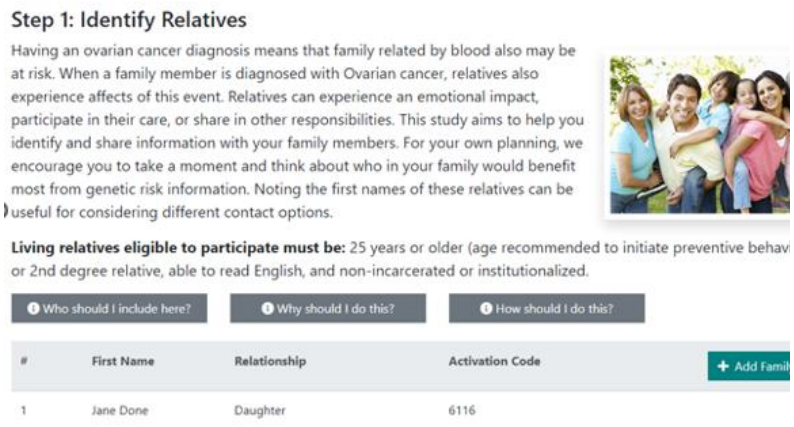
The theoretical foundation for the YFC website development and content was based on the ethical framework and relational level behavior change theories described in Chapter 1: Relational autonomy, interdependence theory, and communal coping.[157]

The YFC infrastructure provided a unique opportunity to explore *TM.* The study sampling frame comprised all surviving individuals diagnosed with ovarian cancer in the state of Georgia between January 2005 and December 2017. Previous communication research related to hereditary conditions has predominantly focused on self-selected samples, consisting of white and highly educated females.[43,45,46,74,75] Thus, exploring *TM* within this study population enables consideration of *TM* within a diverse and representative sample of women who have experienced a life threatening health event that has implications for other family members.

2.2.2 The Your Family Connects Intervention Arm Set-up and Content

During the initial website visit, survivors were required to follow a pre-established sequence when viewing website sections. First, immediately after creating their study account, they were directed to the landing page of the "*Invite Family Members*" section (*Figure 2A*). In this landing page, participants were prompted to list the first name of all FDRs and SDRs and specify the relationship (e.g., mother) in a drop-down list. The section presents instructions including "*who should I include here*," "*why should I do this*," and "*how should I do this*."

Figure 2A. Landing page of the Your Family Connects Website



On the next page, a menu of relative contact options was presented (*Table 2B*). For each listed relative, the menu presents three contact options – "*self-contact*", "*study team contact*", and "*delay contact*". Each contact option was accompanied by a brief description highlighting pros and cons and in which relationships the option might be optimal. For example, for the choice "*self-contacts*," the website narrative suggests that this approach may be best when survivors: have a close personal connection with the relative, want to decide on optimal timing for discussion of inherited risk, and/or have privacy concerns about a third-party's outreach. Survivors were presented with the contrasting viewpoint that this approach may be less ideal when the survivor: does not have frequent contact with the relative, has a personal history with the relative that makes discussions difficult, or is unsure of what to communicate because the specific recommendations are unclear.

Table 2B. Menu of relative contact options

Options	Actions
Self-contact	Survivors download, edit the family letter, and contact their relatives directly
Study team contact	The study team sends the family letter to the selected relative via email or postal mail, as per the survivor's request
Delayed contact	Neither survivors nor the study team reaches out to relatives

This MBI website content highlights that contacting relatives for FGRC is not a one-size-fits-all approach. Instead, participants were encouraged to consider their relationship with each relative, and each relationship's unique needs and preferences. It is important to note that survivors have multiple opportunities to engage with this content as the menu is also presented in the "*Importance of Family Communication*" section.

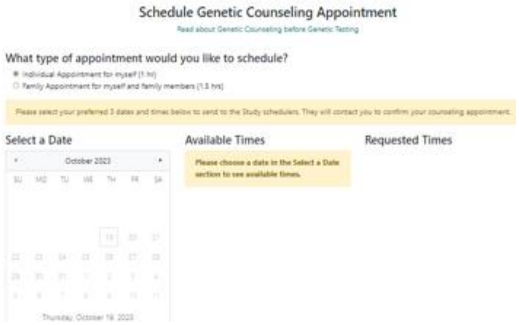
On a subsequent page, survivors were prompted to select their preferred contact approach for each listed relative. If "*self-contact*" was selected, participants themselves were expected to reach out to this relative and could do so with a customizable letter. If "*study team contact*" was chosen, the website would prompt participants to provide the relative's contact information, such as email address and phone number to the study team. In this case, the YFC research team would contact this relative on the survivor's behalf. Lastly, if "*delay contact*" was selected, no further attempts to contact that relative would be made.

On the last page of the "*Invite Family Members*" section, a customizable family letter was available for download. This letter included key messages such as why the relative was being contacted, information about *BRCA1* and *BRCA2* genes and associated cancer risks, and efforts taken to protect confidentiality. Additionally, the letter emphasized the value of visiting the YFC study website to learn more and schedule a genetic counseling session. It also contained a unique login for relatives to access the YFC website.

After navigating through the "*Invite Family Members*" section, survivors were directed to the "*Schedule Genetic Counseling*" section. Here, they were provided with information related to genetic counseling and genetic testing along with frequently asked questions about genetic

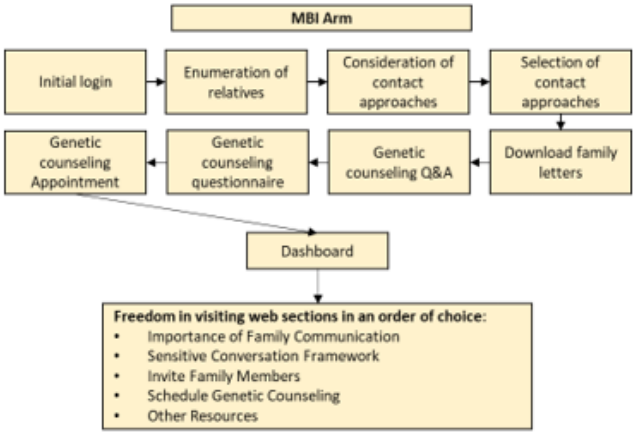
services. Survivors were asked to complete a four-item survey related to their previous experiences with genetic services (e.g., "*Have you or a close relative ever received genetic counseling for cancer? [yes/no/did not answer]*"). Upon the completion of the survey, survivors were prompted to schedule a genetic counseling appointment on the "*My Genetic Counseling*" page. On this page, survivors could choose to schedule a 60-minute individual appointment for themselves, or a 90-minute family appointment for themselves and their relative to attend together (*Figure 2B*).

Figure 2B. Genetic counseling appointment page



Upon completion of visiting the "*Invite Family Members"* and "*Schedule Genetic Counseling"* sections in the specified order, MBI participants could access the website Dashboard to explore other sections. On return visits, the Dashboard was the landing page, and survivors could visit any sections in the order of their choosing. A summary of the website visit set-up is shown in *Figure 2C*.

Figure 2C. Message-based intervention arm website visit set-up



2.3. MAPPING WEBSITE CONTENT WITH TRANSFORMATION OF MOTIVATION

As described in *Chapter 1*, the *TM* shift is conceptualized to consist of two domains: a cognitive interpretation of a health threat (e.g., inherited risk of cancer) presenting an existential threat to the "we"; and an emotional interpretation that such a threat has significant meanings to the self, the relative, or the relationship (e.g., fear for the wellbeing of at-risk relatives), which inspires a drive to protect the "we." The thinking is that *TM* can be prompted by survivors' cognitive and/or emotional interpretation of cancer genetic risk as shared and that mitigating the risk requires pro-relationship responsibility to address and remediate the shared risk. Specifically, I defined "*we"-problem* as "survivor appraising their ovarian cancer as a health threat shared by the family"; "*we"-responsibility* as "risk-reduction is relationship-oriented"; and "*we"-solution* as "engaging relatives in communication about inherited genetic risk".

The MBI website content was based on interdependence and relational autonomy theories. To that end, it is reasonable to expect that survivors who actively engaged with the website content might experience *TM* to the extent that the content prompted appraisals of inherited cancer risk as a "we"-problem, responsibility, -solution. Next, I will present my three assertions on mapping the YFC intervention content to the three *TM* indications (*Table 2C*). To this end, my Aims 2 and 3 are based on the premise that the extent to which ovarian cancer survivors engage with the YFC website *TM*-mapped content may serve as indicators of the *TM* latent construct.

Head section	Website engagement	"We"-problem	"We"-responsibility	"We" -solution
Invite Family Members	FDRs and SDRs enumeration	√		
	Family relationship specification	√		
	Pros and cons of relative contact options		√	
	Selection of contact options		√	
	Family letter			√
Importance of Family Communication	National guidelines for genetic counseling	√		
	Genetic taxonomies (i.e., FDR, SDR)	√		
	Timing of genetic counseling			√
	Motives of FGRC		√	
Sensitive Conversation Framework	5-step sensitive conversation framework tipsheet		√	
	Download of the tip sheet		√	

2.3.1 Assertion 1: Enumeration, Contact Choice, Relative Letter Downloads

I assert that the greater the extent to which survivors' enumerate, choose contact options and download letters for relatives indicates the degree to which survivors interpret inherited cancer risk as a "we"- problem, with collective responsibility, and engaging in family communication as a solution. Evidence for this assertion is reflected in survivors use patterns in the "*Invite Family Members*" section. I hypothesize that survivors listing relatives is suggestive of that the shared health threat is prompting an inclination to communal thinking (i.e., "we"-problem). As described in Chapter 1, this is based in part on relational autonomy wherein a shared health threat prompts survivors to engage in relational thinking as they consider specific family relationships. Survivors' relationships have history, and associated obligations that influence the survivor's appraisal of whether and who to communicate about inherited risk. The enumeration process itself requires survivors to engage in cognitive reflections on the relationship and can prompt emotional responses as survivors anticipate relatives' perspectives and needs. These processes can prompt survivors to recognize the interconnectedness of these relationships within the family. This interconnectedness may prompt survivors to see their relatives as co-owners of the health threat (i.e., "we-problem"). In this way, survivors' enumeration behaviors can be viewed as indicative of *TM* processes.

Survivors' selection of a contact approach for each listed relative could also be indicative of *TM* (*Table 2B*). In choosing a contact approach, survivors were encouraged to weigh factors such as family closeness, privacy concerns, and self-assessed ability to discuss genetic risk information in their selection process. The content also encourages survivors to consider the specific relationship with each listed relative and make judgments of the most suitable approach based on these relationship dynamics. To this end, the process of selecting from the menu of outreach options calls upon the survivors to appraise and reflect on the interconnectedness of their relationship with each relative, shifting selection of contact approach a "relationship-oriented" behavior.

Survivors were able to download customizable family letters for each listed relative. These letters were available for download regardless of the contact approach selection and were designed to facilitate FGRC with relatives. I posit that survivors who downloaded letters were likely more inclined to demonstrate *TM* compared to those who did not download any letters, as such, downloading behaviors should demonstrate a shift toward a "we"-solution appraisal. I assert that survivors who downloaded family letters were more likely to view FGRC as a “we”-solution than those who did not download letters or downloaded fewer letters.

2.3.2 Assertion 2: Family Communication

The notion that for some, ovarian cancer can be attributed to inherited genetic risk is emphasized throughout the “*Importance of Family Communication*” section. Specifically, the section cites guidelines from the U.S. Preventive Services Task Force, recommending that all FDRs and SDRs of women diagnosed with ovarian cancer consider genetic counseling to learn about their potential for inherited risk.[9] The section also outlined the relationship names of all FDRs (i.e., mother, father, sisters, brothers, sons, and daughters) and SDRs (i.e., uncle, aunts, nephews, nieces, grandparents, grandchildren, and half-siblings) and instructed that all of them should be informed of this potential.

In addition, this section outlined the benefits of FGRC for the family, such as “*to prevent more cancer in family*” and “*to see their close relatives thrive and be healthy*.” I would submit that these messages should further encourage survivors towards TM. Plus, survivors have another opportunity to engage with the menu of relative contact options, and its pros and cons as described above. I expect that more use of the *Importance of Family Communication* would prompt survivors to see themselves as part of a dyad/family collective, and as such, could prompt *TM* to protect their relatives by sharing information.

2.3.3 Assertion 3: Sensitive Conversation Framework

The *Sensitive Conversation Framework* section introduces a structured approach to facilitate communication between survivors and their relatives regarding shared genetic risk, aiming to build/enhance survivors communication skills in navigating challenging conversations. The Sensitive Conversation Framework Tipsheet (SCFT) is adapted from frameworks used in delivering difficult news and managing serious illnesses as described in Chapter 1.[97-100] The five steps of SCFT were: (1) developing a communication plan, (2) assessing the relative's understanding of shared cancer risk, (3) emphasizing the "we-ness" behind communication motives, (4) preparing the relative's reactions, (5) encouraging the relative to take advantage of genetic counseling. Survivors can choose to download SCFT to guide their actual genetic risk conversation.

Each step is accompanied by focused language, drawing from the Serious Illness Conversation Guide. For example, in step 3 of emphasizing the "we-ness", survivors are encouraged to foster a relational dialogue by asking about the relative's perception of how the survivor's cancer might impact them (i.e., *"what is your understanding of how my cancer might affect you?"*)

SCFT stands out from other communication frameworks as it acknowledges the need for reciprocal exchange of information and ongoing coping support in the context of a threatening health event. Thus, I posit that survivors who engage more with SCFT will be exhibiting *TM* viewing inherited cancer risk as relationship-oriented that is indicative of perceiving "we-responsibility".

2.3.4 Website Engagement Measures as Transformation of Motivation Indicators

In this section, I will discuss how engagement with the content can serve as indicators of the *TM* latent construct. Research in electronic health disciplines lends support in treating website engagement as indicators of the *TM* latent construct. First, I will provide a brief overview

of commonly used objective and subjective measures of website engagement.[159-161] Then I will justify theoretical premises on the selection of website engagement measures.[160,161]

2.3.4.a Objective measures

Measures of the strength and depth of participants' engagement with website content most commonly rely on automatic tracking of use patterns, including frequency, duration, intensity of use.[159,161] Frequency measures provide information on how often a participant visits a website. These include the number of log-ons and/or website clicks per participant. Indeed, these use indicators can be regarded as behaviors. Additionally, duration is measured as the total time participants spend on the website, as well as mapping patterns of movement through content. In turn, the intensity of usage is commonly reported as the number of pages viewed, quizzes attempted, or discussion posts written. Other types of objective measures include psycho-physiological measures (e.g., eye-tracking and electroencephalography), and mobile sensors. An advantage of these measures is that they can be collected unobtrusively by monitoring online use patterns. Automatic tracking of use patterns can be easily collected with early planning and effective data capture techniques. However, objective measures can be difficult to interpret. Browsing behaviors, such as leaving multiple tabs open, may obscure usage data.[159]

2.3.4.b Subjective measures

Subjective measures focus on users' self-reports of levels of attention, information absorption, and affect and give insight into the user experience.[159-161] These measures are collected via self-administered questionnaires, semi-structured interviews and think-aloud exercises. A number of scales have been developed to quantitatively assess users' subjective website experience.[161,162] Semi-structured interviews and focus groups have been used alone or combined with other methods to gain insights into participants' subjective experiences.

The think-aloud method is commonly used to gain insight into cognitive processes and emotional reactions as users interact with a website in real-time.[160,163] In this method, the

website user is asked to complete a set of specified tasks and spontaneously verbalize what is going through their mind in real time as they perform the tasks. In the classic concurrent think-aloud method, researchers silently observe users' immediate reactions and elaborations without interruption. And researchers may prompt questions to seek explanations and additional details from participants (e.g., "*tell me what you are looking at*?"), or practice active listening to show that participants are being heard.[163,164]

Subjective measures have the benefit of complementing objective measures by enabling an in-depth analysis of user experience, such as awareness of certain topics, and attention to the content. However, data collection can be time-consuming and may be prone to socially desirable responses. Methods such as self-administered questionnaires and interviews are also subject to recall bias. Despite these limitations, subjective measures are critical in understanding website users' experiences and perceptions.

2.3.4.c Theoretical premises of website engagement measurement selections

A working definition of website engagement for this dissertation is the extent to which survivors use and respond cognitively and behaviorally to *TM*-mapped content on the *YFC* website. I will measure website engagement using metrics including duration of time spent on website sections, counts of clicks on pages, and subjective reactions to content assessed using the think-aloud method. Based on the set-up of website sections (2.2.2.), I propose 10 objective measurement variables as the indicators of the *TM* latent construct. These 10 variables are organized under three categories - time spent, clicks, and counts on *TM*-mapped sections (*Table 2D*).

Table 2D. The Your Family Connects website engagement measures

1. Time spent on website content (by minutes)
Enumeration sections
Contact menu sections
Importance of Family Communication section
Sensitive Conversation Framework Tipsheet section
2. Clicks on website content
Enumeration sections
Contact menu sections
Family letter section

3. Counts on the website
Return logins
Relatives enumerated in total
Distinct letters downloaded in total
Sensitive Conversation Framework Tipsheet downloaded in total

Several theories related to information processing provide a theoretical basis for how survivors' website engagement arguably could infer "we-ness".[162,165-168] For example, Elaboration Likelihood Theory suggests that website engagement, measured by time spent, clicks, and counts, is positively associated with depth of information processing.[169,170] When survivors spend more time on the website and engage more frequently through clicks and counts, it indicates that they are processing the information more thoroughly. In contrast, less engagement might indicate peripheral processing of information.

The Model of User Engagement suggests that website engagement could indicate that survivors are experiencing persuasive influence.[167,171] For instance, when *TM*-mapped content is perceived as relevant to survivors communication interests and needs, and the design of the page is inviting and aesthetically appealing, it may create a sense of connection emotionally and encourage them to think deeply about the information presented. In this way, time spent, clicks and counts on the website content could infer *TM*.

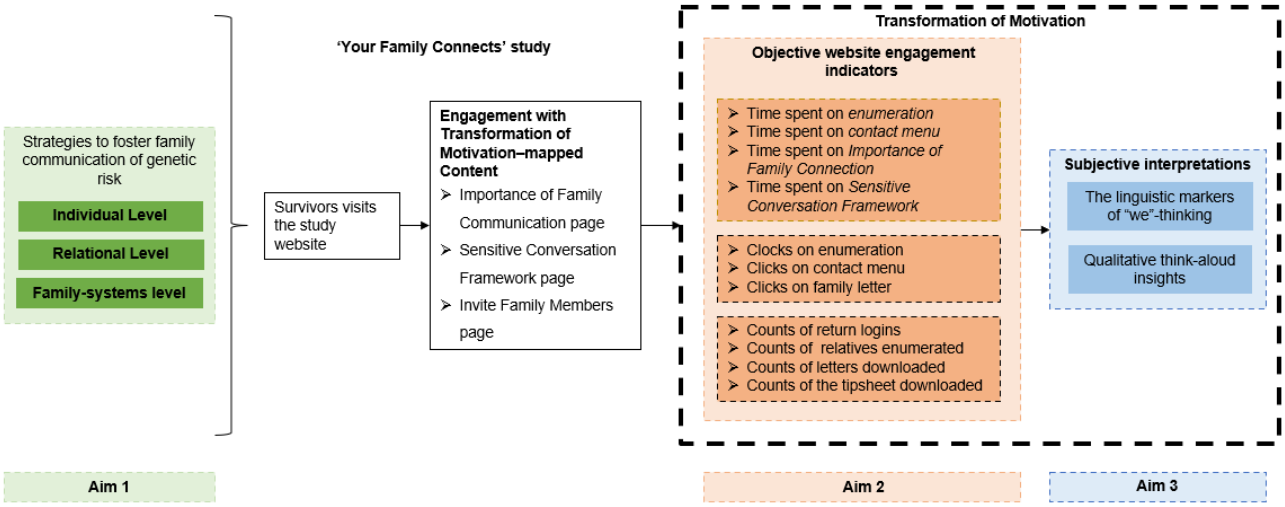
It is important to note that engagement itself is influenced by various external factors, including intervention design such as content and mode of delivery, as well as contextual elements like the physical environment (e.g., internet access) and individual characteristics (e.g., internet self-efficacy). While these factors are not directly measured, they collectively shape the level and nature of engagement on the website.

2.4 PROCESS MODEL AND RESEARCH QUESTIONS

2.4.1. Process Model

I propose the dissertation process model (*Figure 2D*) to illustrate the connections of my three Aims, specifically, how Aim 2 and Aim 3 build iteratively on each other to bring conceptual clarity to the *TM* latent construct. At the end of this section, I also describe the specific research questions posited for each aim.

Figure 2D. Dissertation process model



In Aim 1, I propose to update a review I conducted of interventions aimed to foster communication of genetic risks that are RCTs or quasi-experimental trials, specifically focused on strategies in initiating communication to relatives within family networks. As previously described in Chapter 1, I will draw on several lines of conceptual thinking in reviewing published interventions that have tested FGRC interventions – family-system level, relational level, and individual level. Recognizing the complexity of family communication, I will organize intervention strategies using the proposed multi-level framework. Results from study 1 will help to identify research gaps on whether FGRC interventions have capitalized on a family-based social context. This conceptual guidance would urge a shift in research focuses from proband-centric designs to family-centered designs.

In Aim 2, I propose to assess whether ovarian cancer survivors' YFC website engagement can serve as an indicator of *TM*. Because *TM* is a latent construct that cannot be observed or measured directly, I will conduct Exploratory Factor Analysis guided by theoretical justifications to conceptualize website usage patterns as cognitive and behavioral indicators of the latent *TM* construct. I submit that the greater the website engagement (e.g., time spent viewing the content), the greater the likelihood that the ovarian cancer survivor was experiencing *TM*. In contrast, survivors who more superficially engage with the YFC website content (e.g., less time viewing the content) would be less inclined to "we-ness" thinking.

In Aim 3, I will continue my exploration of *TM* factor(s) identified in Aim 2, using direct observation of survivors' engagement with the YFC website. I will directly observe survivors while they engage with the website and conduct think-aloud interviews in real-time. A sample of 20 survivors will independently visit the YFC website. I will monitor their engagement with *TM*-mapped content. Then, they will complete a brief follow-up survey to assess their family size and perceived family emotional closeness. Lastly, I will conduct one-on-one think-aloud interviews with the same group of survivors, inviting them to revisit the website. The purpose of the last step is twofold. First, I will be able to assess the counts of linguistic indicators of "we-ness"-thinking during their website engagement. And second, I will be able to interpret the insights of *TM* factors through their lived cancer experience. Using the stepped mixed-methods approach, I will be able to gain insights on the identified Aim 2 factor(s) by interpreting the occurrences of "we-ness"-thinking linguistic measures along with their cognitive and emotional reactions in the think-aloud interviews.

To summarize, by paralleling survivors' objective website engagement in Aim 2 with subjective user experience (i.e., think aloud while using the website) in Aim 3 (*Table 2E*), the combined findings will help me achieve the overarching aim, which is to advance our understanding of how to measures *TM*.

Table 2E. Exploration of transformation of motivation in Iterative Aim 2 and Aim 2

Latent Construct	Aim 2: Objective quantitative exploration	Aim 3: Subjective qualitative exploration
Transformation of motivation	Cognitive and behavioral indicators including time spent, clicks, and counts on transformation of motivation - mapped content	Insights on transformation of motivation based on ovarian cancer survivors' lived experience and their use of linguistic markers of "we"-thinking

2.4.2 Aims and Research Questions

Aim 1: Identify Theory-Informed Strategies in Family Genetic Risk Communication Interventions Among Families at Higher Risk of Hereditary Conditions.

Research questions:

1. Which family communication frameworks have been applied?
2. How do the intervention strategies tested align with these theories?
3. To what extent were participants receptive to these strategies and communication increased?

Aim 2: Evaluate Survivors Website Engagement Indicators Comprise a Behavioral Measure of "Transformation of Motivation".

Research question: Can website engagement behaviors including reviewing information, downloading outreach materials, and enumerating relatives serve as indicators of the latent construct of "transformation of motivation"?

Aim 3: To Obtain Preliminary Insights of "Transformation of Motivation" Using the Subjective Think-Aloud Website Engagement.

Research questions:

1. To what extent can linguistic indicators of "we-ness" thinking and communal coping be identified among women who are engaging with the YFC website in real time?

2. To what extent are family size and subjectively rated closeness associated with the frequency of these linguistic indicators?
3. What insights can be gained from individual survivors' lived experiences to inform future research of the latent construct of "transformation of motivation"?

CHAPTER 3. A SYSTEMATIC REVIEW OF THEORY-INFORMED STRATEGIES USED IN INTERVENTIONS FOSTERING FAMILY GENETIC RISK COMMUNICATION

3.1 INTRODUCTION

3.1.1 Genetic Risk is a Family Issue

Increasing uptake of genetic counseling and testing among families at risk for hereditary conditions, such as *BRCA1- and BRCA2-associated Hereditary Breast and Ovarian Cancer Syndrome* (HBOC), is gaining priority. As described in Chapter 1, genetic risk is inherently a family issue. As a reminder, assessing family cancer genetic risk, such as *BRCA1/2*, most commonly begins by identifying a "*proband*", that is, an individual who has been diagnosed with the hereditary condition and/or screens positive for the pathogenic variant. Once identified, follow-up genetic services (e.g., cascade screening) are needed for probands' blood relatives in order to assess who else in the family is also at-risk.[172] National guidelines, in the United States (U.S.) and globally, highly encourage communication to relatives as hereditary cancer prevention and early detection actions are available for asymptomatic relatives who also carry *BRCA1/2.*[22,173-175]

In Chapter 1 (1.1.4), I defined ***optimal*** family genetic risk communication (FGRC) as a communication process that begins with probands' deliberations regarding whether and when to tell, who to tell, and what to tell at-risk relatives and extends through their first and subsequent conversation(s) regarding inherited risk with at-risk relatives.

Currently, confidentiality and privacy protections regulations (e.g., Health Insurance Portability and Accountability Act) compel genetic counselors and other clinicians to advise probands to communicate genetic risk information to their blood relatives.[55,176] As discussed in

Chapter 1, evidence to date suggests that reliance on this approach has resulted in incomplete communication within the family,[41-47] and suboptimal uptake of genetic testing among at-risk relatives.[48]

3.1.2 Barriers to Family Genetic Risk Communication

Prior systematic reviews described facilitators and barriers to family communication about genetic risk that operate at multiple levels of influence. To summarize, at the individual level, probands reported: difficulty understanding genetic risk information (e.g., patterns of inheritance, prevention options), lack of knowledge about who to share and what to share, low of motivation to share, lack of confidence to share the correct information, and difficulty in finding the "right" time for disclosure.[52-54,65,72,177] Relational level barriers reported include: low emotional closeness and infrequent interactions with relatives.[53,72,177] Family level barriers included low family cohesiveness, as well as family norms and interpersonal stigma that limit intergenerational communication of genetic disease experiences.[53,72]

Despite these well-characterized barriers, a recent systematic review of Baroutsou et al (2021) evaluated psychoeducational family communication interventions provided in the contexts of HBOC and Lynch syndrome genetic risk. The reviewed found only 14 studies conducted over a 17-year period. Moreover, the interventions (n=8) resulted in insignificant improvements in the number of relatives contacted/informed about their genetic risks.[69] While the recent review of Baroutsou et al (2021) was comprehensive in scope, information about the extent to which the interventions aimed to address family communication barriers and were grounded in theories of family communication was not the focus. Gaining a better understanding of these factors is needed to inform the next generation of family communication interventions.

3.1.3 Theories Relevant to Family Genetic Risk Communication

As reviewed in Chapter 1, several lines of theories could inform the development of FGRC intervention strategies: family system-level, relational-level, and individual-level.[178] In this section, I will briefly summarize relevant behavioral theories to inform optimal FGRC.

Family system-oriented theories [e.g., Family Systems Perspective (FSP) and Family Communication Patterns Theory (FCPT)], share the view that families operate as systems with unique patterns and processes of communication – often referred to as a family communication climate.[126,127,179] These theories suggest that family relational structures (e.g., size, density of interconnected relationships) facilitate or inhibit the flow of genetic risk information. For example, FCPT suggests that frequent and spontaneous interactions paired with homogeneous values and attitudes amongst relatives can foster freer flow of information. Such information flow, in turn, increases the likelihood that at-risk relatives will share their perceptions of a health threat Additionally, families differ with respect to privacy boundaries, that is, the perceived importance of constraining the flow of genetic risk information that in turn, influences communication patterns and the flow of genetic risk information.[180] Families with a fewer privacy constraints and less hierarchical family relations have been found to be more likely to communicate about genetic risk and to a greater proportion of at-risk relatives.[180-182]

Relational theorists characterize families as a collection of dyads with differing dyadic patterns and levels of relational influence.[183] For example, Interdependence theory and communal coping suggest that family risk communication occurs when inherited risk is perceived to represent a shared threat to specific dyadic relationships (e.g., mother-daughter).[118,184-186] When facing stressful life events (e.g., the potential to carry a pathogenic genetic variant), perceiving the event as a threat to maintaining a specific relationship can activate “communal coping” efforts such as cooperative problem-solving processes (e.g., seeking genetic counseling services).[85,118] Thus, these theories suggest that interventions to capitalize on the relational interdependence of key dyads to motivate interpersonal communication could be most effective.

Whether or not a proband shares inherited risk information with their relatives, ultimately, rests on individual-level actions. Health Belief Model (HBM),[111] Social Cognitive Theory (SCT),[112,113] and the Theory of Planned Behavior (TPB)[114,115] have relevance in this context.

HBM holds that a proband will communicate inherited risk information if s/he perceives at-risk relatives to be susceptible to the risk and that the benefits of informing relatives outweigh the barriers.

SCT adds to this notion of the role of proband's confidence and knowledge, suggesting that probands' low confidence to explain patterns of inherited risk and prevention options could inhibit communication flow. TPB emphasizes the importance of holding specific intentions to communicate as being critical to whether probands will communicate with at-risk relatives. Indeed, this theory holds that the more time- and goal-specific the communication intention is, the more likely it will be attempted.[187] Thus, interventions aimed to improve FGRC would be expected to emphasize awareness of familial risk (e.g., perceptions that relatives are susceptible for the risk), confidence (e.g., communication skills-building), and intentions (e.g., specifying how/when communication will occur) to communicate.

3.1.4 Research Questions

The predominantly null findings of interventions, described in Baroutsou et al.'s (2021) recent systematic review, raise questions about the intervention strategies used. In my systematic review, I characterize and synthesize intervention studies that have included comparison groups to with respect to whether theory was applied to in developing the family genetic risk communication interventions and which strategies were shown to be effective.[188,189] Research gaps identified from the present review could help advance the development of family genetic risk communication interventions.

To this end, I pose the following questions: (1). Which family communication frameworks have been applied?; (2) How do the intervention strategies tested align with these theories?; (3) To what extent were participants receptive to these strategies and communication increased?

3.2 METHODS

3.2.1 Eligibility Criteria

Intervention studies were eligible for the review if they: (1) targeted at least one of 35 hereditary conditions that meet criteria of the American College of Medical Genetics and Genomics (ACMG) as being medically actionable, that is, preventive actions are available that meet established clinical management guidelines;[190] (2) evaluated a strategy or complement of strategies to promote communication of family inherited risk information; (3) included a comparison group; and (4) were published from January 2010 to August 2020 when the most recent guidelines were endorsed. Ineligible studies: (1) were not written in English; (2) focused solely on patient-provider communication, or marital couples' communication and (3) were published as study protocols, conference, and meeting abstracts or in grey literature.

3.2.2 Search Strategy

The search, originally performed in August 2020, spanned three electronic databases (PubMed [National Library of Medicine], PsycINFO [EBSCOhost], and Web of Science), guided by the search terms: "disclosure"; "family communication", "proband", "carrier", "cascade screening", "trial design filter" (*Figure 3A*). All articles were imported into EndNote (X9) to remove the duplicates, and unique studies were later imported into Covidence for study selection.

Figure 3A. Search criteria

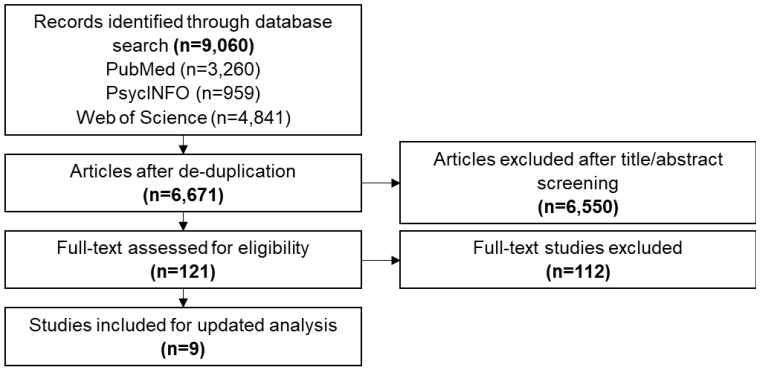
1. **Pubmed**:
   (communication[tw] OR informing[tw] OR disclosure[tw] OR disclose*[tw] OR disclosing[tw]) AND (famil*[tw] OR relative*[tw] OR parent*[tw] OR sibling*[tw]) AND (genetic*[tw] OR proband*[tw]) AND english[lang] AND human[MeSH Terms] AND (""2010""[Date - Publication] : ""3000""[Date - Publication])"
2. **PsycINFO**:
   (Communication OR Any Field: Informing OR Any Field: disclosure OR Any Field: disclose* OR Any Field: disclosing) AND (Any Field: famil* OR Any Field: relative* OR Any Field: parent* OR Any Field: sibling*) AND (Any Field: Genetic* OR Any Field: proband*)
3. **Web of science**:
   (TS=(communication OR informing OR disclosure OR disclose* OR disclosing) AND TS=(famil* OR relative* OR parent* OR sibling*) AND TS= (genetic* OR proband*)) AND LANGUAGE: (English) AND DOCUMENT TYPES: (Article) Indexes=SCI-EXPANDED, SSCI Timespan=2010-2020"

3.2.3 Study Selection

The systematic review followed PRISMA guidelines for reporting the screening and selection processes.[191] The search yielded 9,060 articles (PubMed = 3,260; PsycINFO =959, and Web of Science=4,841). After removing duplicated titles and abstracts, 6,671 unique articles were screened by the first author (J.Z.) for eligibility for inclusion; 121 studies were eligible for full-text screening.

J.Z. reviewed the full texts. A trained master-level research assistant (S.V.) screened 25% of full texts (n=30). All discrepancies were discussed via Zoom meetings to reach a consensus. A Kappa score of 80% was achieved between the two reviewers, indicating good inter-rater agreement [192]. In addition, the reference list of each manuscript was examined to identify additional studies not identified in the search. A total of nine interventions met the inclusion criteria for this review and were included (*Figure 3B*); six of the studies overlapped with the prior review conducted by Baroutsou et al (2021).[103,193-197]

Figure 3B. The original search PRIMSA chart



3.2.4 Data Extraction

Data extracted from each article included: country and year of publication, genetic condition, sample size, sample characteristics, study design, study inclusion/exclusion criteria, intervention and control group descriptions, theories used, and intervention process outcomes assessed.

3.3 RESULTS

3.3.1 Overview of the Studies

As shown in *Table 3A*, five studies were conducted in the U.S.,[103,193,197-199] three in Europe,[194,196,200] and one in Australia.[195] Eight of the nine studies exclusively focused on communication of genetic risk in the context of hereditary cancer syndromes.[103,193,194,196-200]

Seven were conducted in the context of pre-test genetic counseling,[103,199,201] during a post-test genetic counseling session,[103,198] or following post-test genetic counseling.[194,195,200] Five studies were randomized controlled trials (RCTs),[103,193-195,197] three studies were quasi-experimental,[198-200] and one was an observational study conducted as part of an RCT follow-up.[196]

3.3.2 Study Participants

Study samples included healthy adults,[193] patients with the health condition,[196,197,199] probands of a hereditary condition,[103,194,195,198,200] and parents of minor probands.[195] Five interventions were conducted in English,[193,195,197-199] one in Dutch,[194] and one in Swedish.[196,201] Most (n=6) included only women.[103,193,197-200] Because studies included a range of inclusion criteria other than “probands”, I will refer to “participants” in describing findings.

3.3.3 Intervention Outcomes

Only two RCTs reported significant intervention effects (*Table 3B*).[193,197] Bodurtha et al found that participants were more likely to gather and share familial cancer information with relatives than the comparison group at all follow-ups.[193] Similarly, Vogel et al found that participants who received a mobile app health intervention were significantly more likely to discuss genetic counseling with their relatives than participants in the control group.[197]

Table 3A. Study descriptions of original review

Author (Year)	Country	Study design	Genetic condition(s)	Sample Size	Elements of Intervention					Control group	At-risk Relative Enrolled
					Brief descriptions	Content	Theoretical frameworks	Format	Delivered by		
Bodurtha (2014)	U.S.	RCT	Family cancer history	490	Stepwise, skill-based sessions to provide tailored pedigree-based risk information and coach communication skills in two phases.	**Phase 1** (1) Provide tailored risk information and prevention recommendations; (2) Review their pedigree for breast and colon cancer within the CAGene v.5.1 program; (3) Identify any missing information in the family history; **Phase 2** (4) Coach women in communication skills to obtain needed information; (5) Develop a plan for collection and follow-up of this information.	(1) The coordinated management of meaning theory; (2) The expanded Health Belief Model; (3) Buckman's "breaking bad news" framework and it's adapted Daly's model	In-person	Study recruiter	A handout promoting ways to lower breast and colon cancer risks, screening recommendations, and services contact info.	No
Eijzenga (2018)	Netherland	RCT	HBOC or hereditary colorectal cancer	336	Motivational interviewing-based counseling sessions in two phases.	**Phase 1** (1) Agenda setting on the subject of family communication; (2) Exploring probands' pattern of informing relatives; **Phase 2** (3) Providing additional information; (4) Build motivation and strengthen self-efficacy; (5) Discuss possible solutions.	N/A	Via phone	Psychosocial worker	Standard of care	No
Garcia (2020)	U.S.	Pre-test post-test intervention comparison pilot study	HBOC	40	Facing Our Risk of Cancer Empowered (FORCE) resources.	(1) "What you should know about genes and cancer" brochure; (2) "Sharing cancer information with the family" worksheet; (3) "Family letter" template.	N/A	Printed materials	N/A	Standard of care	No
Hodgson (2016)	Australia	Pragmatic RCT	Genetic conditions that have implications for at-risk relatives	95	Individualized genetic counseling follow up phone calls (up to 3 calls) over a 12-month period.	(1) “Getting the picture” about the participant’s experiences of communication to date; (2) Forming an intention to act. This includes maintaining or enhancing the participant’s capacity for communication; facilitating decision-making; and resolving ambivalence; (3) Planning to act.	Reciprocal Engagement Model	Via phone	Genetic counselor	Current practice	No
Kardashian (2012)	U.S.	Pilot quasi-experimental trial	HBOC	19	A ShaRIT educational binder of genetic information and family resources.	(1) The patient’s personalized medical report; (2) Family pedigree; (3) BRCA mutation report from Myriad Genetics; (4) Personalized recommendations for surveillance and prevention; (5) Letter to relatives notifying him/her of BRCA mutation identified in relative; (6) Fact sheet addressing frequently asked questions regarding cancer risk, costs of genetic testing, and insurance issues regarding genetic testing; (7) Contact information for genetic counselors specific to each eligible relative based on their geographic location; (8) Support websites and brochures.	N/A	Printed materials and electronic versions of each resource	N/A	Standard of care	No

Montgomery (2013)	U.S.	RCT	HBOC	422	A six-step communication skills-building intervention delivered in two phases.	**Phase 1** (1) Identifying relatives who could benefit from the information; (2) Choosing the communication format; (3) Assessing how much relatives already knew and how much they might want to know. **Phase 2** (4) Sharing the actual genetic test result with at-risk relatives; (5) Responding to relatives' emotional reaction to the disclosure; (6) Providing genetic counseling resources for relatives.	(1) Buckman's "breaking bad news" framework and it's adapted Daly's model; (2) Theory of Planned Behavior	In-person	Trained health educator	A wellness education intervention	No
Roshanai (2010)	Sweden	A descriptive study as a part of a RCT	Personal or family cancer history	147	An extended counseling session to identify at-risk relatives and overcome perceived obstacles of sharing the information.	(1) Participants intention to inform their at-risk relatives, and barriers of not sharing; (2) Introduction of the breaking bad news model was explained to participants; (3) An information pamphlet was shared to participants.	Buckman's "breaking bad news" framework	In-person	Specialist nurse	Standard of care	Yes; at-risk relatives were contacted after the study completion to understand their experiences of receiving genetic information
Sermjin (2016)	Belgium	Stepwise two-phase quasi-experimental trial	HBOC	172	An informative letter to inform genetic counseling services	An informative letter was directly mailed to at-risk relatives, informing them about the familial cancer risk, the availability of a predictive genetic test and the option of having subsequent counselling.	N/A	Printed materials	N/A	Standard of care	Yes; at-risk relatives were contacted for an informative letter in the intervention phase
Vogel (2019)	U.S.	Pilot RCT	Ovarian cancer	104	A week-long mobile app for Genetic Information on Cancer (mAGIC) intervention	(1) Benefits of speaking with family about genetic counseling/testing and cancer screening; (2) Usefulness of genetic counseling even if adopted or do not have medical history; (3) Gene mutations in men and how they can be affected by cancer.	(1) The Fogg Behavioral Model of Mobile Persuasion; (2) Health Belief Model	Mobile app	N/A	Standard of care	No

Table 3B. Intervention outcome measures and results of original review

Author (Year)	Outcome Measures					Results
	Primary outcome(s)	Other outcome(s)	Collection method	Follow-up time points	Format	
Bodurtha (2014)	(1) Gather family cancer history (yes/no); (2) Share familial cancer information with relatives (yes/no); (3) Frequency of communication.	N/A	Survey/questionnaire	1-, 6-, and 14-months post-baseline	Self-report	Participants in the intervention group were more likely to gather family cancer information ($p<0.001$) and to share familial cancer information with relatives ($p<0.001$) at the follow up. Intervention group reported significantly higher communication frequency at follow-up ($p<0.05$).
Eijzenga (2018)	(1) Cancer genetics knowledge; (2) Motivation to disclose information to at-risk relatives; (3) Self-efficacy.	Intervention evaluation	Survey/questionnaire	1 week after the intervention, and 4 months after the study enrollment	Self-report	No between-group differences were found in all outcomes.
Garcia (2020)	(1) HBOC knowledge; (2) Rates of disclosure to first degree relatives and subsequent genetic testing.	N/A	Survey/questionnaire	6 months following genetic testing	Self-report	The statistical differences between two groups were not performed.
Hodgson (2016)	The number of at-risk relatives who contacted genetic services for information and/or genetic testing.	N/A	Chart audit	18 months after recruitment	Chart audit	No between-group differences were found in all outcomes.
Kardashian (2012)	The feasibility and acceptability of the ShaRIT intervention.	(1) Participants' report of sharing BRCA results with eligible relatives; (2) Participants' report of relatives receiving BRCA testing	Survey/questionnaire	At least 2 months after BRCA positive results disclosure	Self-report	No between-group differences were found in the primary outcome. The proportions of second-degree relatives who tested were statistically different between the control group (67%) and the intervention group (14%).
Montgomery (2013)	(1) The percentage of probands sharing test results; (2) The level of distress associated with sharing.	(1) Attitude about sharing test results; (2) Perceived behavioral control; (3) Behavioral Style Scale; (4) Depression	Survey/questionnaire	3 months	Self-report	The statistical differences between two groups were not performed.
Roshanai (2010)	(1) Disclosure of genetic info to at-risk relatives (yes/no); (2) To who.	At-risk relatives' experiences.	Interview and open-ended questions	8 months	-	The results were quantified and reported. The statistical differences between two groups were not performed.
Sermjin (2016)	(1) Uptake of counselling and predictive testing; (2) The State-Trait Anxiety Inventory score	N/A	Survey/questionnaire	At the study contact	Self-report	The statistical differences between two groups were not performed.
Vogel (2019)	(1) Participants' uptake of genetic services; (2) Self-efficacy in making an appointment for genetic counseling; (3) Discussion of genetic counseling with family (yes/no); (4) Knowledge about hereditary cancer	Usability and acceptability of intervention	Survey/questionnaire; chart audit	1 week and 3 months. Medical record review at 12 months	Self-report and chart audit	Participants in the intervention group were also statistically significantly more likely to self-report talking with their family about genetic counseling ($p= 0.01$).

3.3.3 Intervention Strategies Linked to Communication Theory

All nine studies included an intervention strategy that aligned with a theory; only five specifically named the theory/theoretical framework they used. The theory-based strategies were most commonly to be integrated into genetic counseling sessions. Interventions were primarily delivered in-person,[103,193,196,201] followed by print materials,[198-200] phone, [194,195] and via mobile phone app.[197]

3.3.3.a Family-system level

Family-system level theories hold that over time families develop patterns of functioning that influence their patterns of communication and support. The majority of the interventions involved a clinician co-constructing a family pedigree or family tree with the participant.[103,193-196,198,200-202] I consider constructing a family pedigree to be a family system-level strategy as clinicians (e.g., nurse, genetic counselor) guide the participant to characterize their family's structure of relationships and the relatives who were most likely to be at-risk of carrying an inherited pathogenic genetic variant.[203]

In five studies, the pedigree was provided to participants in both intervention and control groups as part of standard genetic counseling care.[103,195,196,198,200-202] For example, in Sermijn et al, participants reviewed the family pedigree to decide which at-risk relatives should be informed.[200] In Hodgson et al, information on the family pedigree was used to derive the total number of at-risk relatives in a family.[195]

In two studies, a family pedigree was conducted only for intervention group participants. In Bodurtha et al, participants received personalized breast and colon risk based on pedigree information, and if high risk, they were referred to cancer genetic counseling clinic.[193] In Eijzenga et al, a family pedigree was used to systematically assess participants' knowledge about which relatives should be informed about their potential risk, and what information should be provided to these at-risk relatives.[194,204] If participants had informed all at-risk relatives identified in the pedigree, the intervention session would end. If not, participants received a communication skill-building intervention via motivational interviewing.

3.3.3.b Relational level

Relational level strategies recognize that dyads within a family have different relational patterns and levels of interpersonal influence that in turn, can influence how they respond to genetic risk individually and collectively. Four interventions were informed by dyadic-level theories.[103,193,195,196] Three based the intervention on Buckman's "Breaking Bad News" (BBN) six-step framework and its adapted model of Daly's strategy.[103,193,196] Montgomery et al fully aligned intervention communication strategies with the six-step BBN framework.[103] The

intervention was conducted as part of a two-phase genetic counseling session. At the genetic testing pre-disclosure session (phase 1), participants began by identifying at-risk relatives using the pedigree family-system approach described above. Participants then considered optimal communication formats, assessed at-risk relatives' knowledge, and anticipated what relatives might want to know about genetic risk. During the disclosure session (phase 2), participants were encouraged to share the information, anticipate at-risk relatives' reactions, think about possible responses, and refer at-risk relatives to genetic services.[102,205] In Roshanai et al, a nurse interventionist explained each step of BBN to participants and encouraged them to use the framework while informing their at-risk relatives.[196,201] Bodurtha and colleagues incorporated Daly's adapted BBN in a 27-page personalized intervention booklet, and displayed the booklet in an interactive presentation to coach participants in family communication skills.[193]

Two studies mentioned other relational level strategies but provided little detail on how they mapped to theory. For example, Bodurtha et al indicated that the intervention borrowed from the Coordinated Management of Meaning Theory without further explanation.[193] Hodgson et al referred to the Reciprocal Engagement Model, a genetic counseling practice model, but provided no details on how this model informed tele-genetic counseling follow-ups.[202]

3.3.3.c Individual level

Whether or not inherited risk information is shared within families currently relies predominantly on the actions of individual participants. Relevant individual-level theories are those that emphasize *intrapersonal* factors such as cognitive and emotional drivers of communication. While often not specifically attributed, all interventions included strategies aligned with individual-level theories, most frequently HBM, TPB, and SCT. Several interventions began with pedigree discussion complemented with adjunctive individual-level strategies, such as influencing participant's intention [194-196] and building self-efficacy.[193,197] For example, Roshanai et al (2010) complemented the dyadic BBN framework with intervention strategies aligned with the TPB and SCT.[196,201] A nurse interventionist assessed participants' intentions to share information with relatives and explored reasons for not wanting to share information. Nurses then coached participants to overcome identified barriers. Sessions with the nurse coach were videotaped and provided to the participant, along with a pamphlet about basic genetic concepts. Similarly, Vogel et al used a mobile app to promote participants' self-efficacy to communicate with, and effectively manage relatives' emotional reactions.[197] Vogel applied Fogg's Behavioral Model of Mobile Persuasion, a cognitive-behavioral theory, in designing time-cueing prompts to encourage participants to continuously use the mobile app.

A few interventions focused solely on the individual level. For example, in Sermijn et al, the intervention involved a genetic counselor coaching on barriers to communication that aligned with the HBM.[200] After six months of troubleshooting with participants to overcome these barriers, interventionists provided a cueing letter directly to at-risk relatives about genetic counseling. Bodurtha and colleagues included family genetic risk information aimed to raise participants' perceived risk and vulnerability of developing cancer as part of the 27-page booklet.[193] Eijzenga et al used strategies based on HBM and SCT to increase knowledge, motivation, and self-efficacy via motivational interviewing conducted by psychosocial workers or serialized calls from genetic counselors.[194] Garcia and colleagues relied on communication aids developed by Facing Our Risk of Cancer Empowered (FORCE) aimed to increase knowledge of inherited risk.[199] Communication aids included an educational brochure about genes and cancer, a worksheet for sharing cancer information, and a family letter template. Similarly, Kardashian et al provided a ShaRIT information binder, that included a family letter and a cancer genetics fact sheet, for participants to give to relatives to support risk communication.[198]

Individual-level theory was also applied to assess study outcomes. Montgomery et al found that variables of TPB, including participants' attitude about sharing test results, perceived control in sharing genetic test results, and subjective norms of at-risk relatives' perceptions and influences, were positively associated with participants' intention to share.[103] Among a subgroup of participants who shared their genetic test results, the results were similar in that perceived control and subjective norms were positively associated with sharing.

3.3.4 Intervention Strategy Acceptability

Data on intervention adherence could help to interpret might help explain the null findings of the majority of these intervention studies. Six studies collected data on intervention acceptability.[194-199] However, definitions of acceptability varied across the studies. Terms such as usability, usefulness, and satisfaction were also used to describe acceptability. Assessments were relatively brief, and often were conducted with participants and staff. Of the two studies with significant intervention effects, Vogel and colleagues reported high acceptance of and satisfaction with the intervention with respect to the quality of content, and amount of daily messaging.[197]

While five studies found high levels of intervention acceptability, these levels were not associated with significant intervention effects. Eijzenga and colleagues reported that both participants and psychosocial counselors evaluated motivational interviewing sessions to be useful, not too long, and not too confrontational.[194,204] Similarly, Roshanai et al reported that all but three intervention participants claimed the videotape of their counseling session was good,

and 80% of them thought that the pamphlet was good or very good.[196,201] In Garcia et al, 90% of participants regarded FORCE communication aids to be easy to use, and easy to understand.[199] In Kardashian et al, all intervention participants and genetic counselors agreed that the ShaRIT information binder was a useful resource. Compared to 70% of control participants, only 22% of intervention participants wanted additional information regarding sharing results.[198] Additionally, none of the studies linked acceptability outcomes to intervening mechanisms (e.g., self-efficacy, knowledge) suggested by the underlying theories.

3.4 DISCUSSION

My systematic review of theory-based intervention strategies to promote inherited risk communication among families identified just nine studies. Only half applied theory and none explicitly targeted family systems or dyads. Intervention strategies largely focused on hereditary cancer syndromes and were evaluated as a part of, or a supplement to genetic counseling sessions. The theory used most commonly was Buckman's "Breaking Bad News" six-step framework, which is designed to build participants' skills in communicating genetic risk with relatives based on their dyadic relationships. Empirical evidence is lacking for how information on shared genetic risk is received and processed by relatives at the relational and family-system levels.

It is notable that the majority of family genetic risk communication interventions focused on hereditary cancer syndromes. Other hereditary conditions, such as familial hypertrophic cardiomyopathy and familial hypercholesterolemia, received less attention on its lack of family communication and low uptake of cascade screening. For example, a systematic review of familial hypercholesterolemia found that no cascade screening studies were conducted between 2001 to 2018 in the U.S.[206] As suggested by the Theory of Family Systems in Genetic Illness, hereditary conditions differ with respect to characteristics, such as penetrance, clinical severity, and timing of clinical onset.[73] Key differences exist in communication demands of these hereditary conditions, such as likelihood of developing the disease, expected severity of the illness, and testing decisions.[62,207] These differences likely present practical, emotional, and relational challenges for family communication unique to each condition. Thus, interventions for hereditary cancer conditions may not be appropriate to fostering genetic risk communication among families with other hereditary conditions. Results from my present review identify an important research gap in the literature and highlight the need to develop communication strategies on other hereditary conditions.[206]

The majority of reviewed interventions found no benefit for increasing probands' intention or motivations to disclose genetic risk information to at-risk relatives, the number of first-degree relatives that participants who disclosed, or genetic service uptake among at-risk relatives. Of the two intervention studies showing significant effects, one applied multiple theories operating at three levels of influence in the development of their interventions.[193] Both interventions shown to be effective involved asymptomatic individuals, and were conducted outside of genetic counseling contexts. Though the research base is limited, these commonalities support the testing of risk communication interventions outside of genetic counseling settings.

Most studies did not evaluate implementation process factors such as strategy fidelity, feasibility, and sustainability.[208,209] Evidence suggests that these factors are key to enable integration of effective interventions into routine practice.[209-212] The two interventions showing benefit over standard care included intervention strategies (e.g., motivational interviewing) that would require resources not routinely offered in clinical settings, which could limit sustainability. Future family communication interventions should evaluate implementation process factors and outcomes. Additionally, no intermediate outcomes were assessed to align with theory-based intervention mechanisms (e.g., self-efficacy), making it difficult to characterize why the majority of interventions, though highly acceptable to participants, were not effective.

The interconnectedness of inherited health risks in a family system may help to fill research gaps on how sharing of genetic risk information can be promoted and capitalized on a kinship-based social structure. However, empirical evidence is limited regarding how family structure influences communication, and how shared genetic risk is received and processed by at-risk relatives at the relational and family-system levels. For example, FSP holds that whether probands communicate to families about inherited cancer risk depends on family structure, shared health beliefs, and the communication environment.[126] A study that surveyed 175 probands found that individuals coming from families accustomed to open discussion were more willing to communicate difficult topics, such as genetic disease risk, with other relatives.[180] Thus, theory would support that communication interventions do more than note these relational connections, but perhaps, building individual relatives' skills accordingly.[213]

Developing sustainable interventions that broaden reach beyond specialty clinic settings are also needed. Specialty genetic clinics are known to be less accessible to those with low income, families in rural areas, and those from racial-ethnic groups.[214] Thus, current intervention approaches and outcomes may not be generalizable to these populations. New initiatives such as the ongoing National Cancer Institute-funded pilot research program are needed to evaluate

interventions specifically aimed to foster families' communication about inherited risk.[215,216] This initiative and those focused on other highly heritable cardiovascular conditions have the potential to bring attention to family systems and move the field beyond individual-mediated approaches.

This systematic review is not without limitations. Only English-written studies published in scholarly peer-reviewed journals were included, and intervention studies published in other languages that could have been theory-based were not analyzed. Unpublished interventions were not included, thus intervention efforts that were only reported in grey literature are not represented.

Despite these limitations, my and Baroutsou and colleagues' reviews showed relatively few effective interventions promoting family communication about inherited risk; virtually all were focused on hereditary cancers and conducted in specialty clinic settings with women. This suggests an urgent need for the development and evaluation of interventions in a broader set of contexts if the research is to realize the vision of precision public health.[217,218] Additionally, I add to Baroutsou and colleagues' findings to highlight the need for family communication intervention development and evaluation to be informed by multi-level theories of family- and dyadic processes. This review adds to the literature in identifying the absence of interventions aligned with theories of family systems and communication that may explain, in part, why few of the interventions have been effective. Currently, family-system considerations are based almost exclusively on pedigree assessment tools. While these can be clinically useful in interrogating relational connections within families,[84] they have not generally been paired with evidence-based strategies for improving communication.

My systematic review suggests that interventions to foster family genetic risk communication largely have not been effective. Common strategies provided as standard care in genetic counseling practice (e.g., family letters and informational brochures) may be necessary but not sufficient to foster the flow of genetic risk communication within families. Adjunctive strategies such as motivational interviewing, communication tip sheets (e.g., breaking bad news), and theory-based cognitive-behavioral strategies hold promise but their sustainability has yet to be determined. These results highlight the need for accelerating research to develop and test interventions informed by family-system theories, conducted outside of genetic counseling contexts, and for a broader swath of hereditary conditions.

3.5 UPDATES TO SYSTEMATIC LITERATURE REVIEW

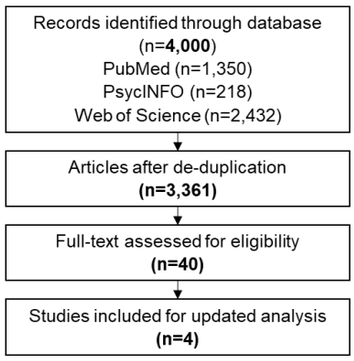
3.5.1 Rationale

The original systematic review was published on *Patient Education and Counseling* in 2021.[71] Cochrane and other organizations recommend that systematic reviews should be updated periodically to integrate new evidence, ensuring that the review remains relevant and meaningful.[219,220] Thus, I initiated an updated search in August 2023 as the conclusions of my published review were based on intervention studies published in the previous decade (January 2010 to August 2020). The aim of the updated search is to identify any new intervention studies that may have been published from 2020 through 2023 that would alter my prior findings or conclusions (e.g., new intervention designs, new study populations, new outcomes, or theory applications).

3.5.2 Methods

I retain the research questions and inclusion criteria from the original review. The only change is that I extended the publication period, searching for intervention studies published between January 2020 and August 2023. In the updated search, I identified 4,000 articles in three databases (PubMed=1,350; PsycINFO=218; and Web of Science = 2,432) (*Figure 3C*). All articles were imported to Covidence for the initial screening process. After removing 639 duplicates, 3,361 studies proceeded to the title and abstract screening stage. Subsequently, 40 studies were identified for the full-text screening. Of these, four met the inclusion criteria and were included in the updated review.[221-224]

Figure 3C. The updated search PRIMSA chart



3.5.3 Results

3.5.3.a Changes in intervention design

As shown in *Table 3C*, one study originated from Australia,[224] one from the Netherlands,[221] and two from the U.S.[221,223] With the exception of the intervention in the Netherlands, all the newly included interventions were delivered in English.

Consistent with my prior findings, the primary study design for interventions was RCT or its variations. The newly included studies were broader in scope and included additional medically actionable genetic conditions other than hereditary cancers. Specifically, three of the four targeted family communication for those affected by inherited cardiac conditions, such as hypertrophic cardiomyopathy and dilated cardiomyopathy. [221,223,224] Additionally, one study focused on communication of multigene testing results associated with prostate cancer.[222] Findings from the prior review were predominantly in inherited cancer syndromes. The broader spectrum suggests an increasingly diversified FGRC research focus in the past five years.

Table 3C. Study descriptions of updated review

Author (Year)	Country	Study design	Genetic condition(s)	Sample Size	Elements of Intervention					Control group	At-risk Relative Enrolled
					Brief descriptions	Content	Theoretical frameworks	Format	Delivered by		
Burns (2022)	Australia	RCT	Hypertrophic cardiomyopathy	50	An additional cardiac genetic counselor-led appointment	The additional appointment was guided by a communication aid. The communication aid was developed to support the delivery of genetic results to the proband and to aid communication with the family. The aid could be written in and taken home	N/A	In-person	Cardiac genetic counselor	Standard clinical cardiology review consultation	No
Kinnamon (2023)	U.S.	A multicenter, open-label, cluster RCT	Dilated cardiomyopathy	1,241	Participants were provided with a Family Heart Talk booklet at the enrollment	The Family Heart Talk booklet included visuals and lay language explanations of the evaluation and care of individuals with dilated cardiomyopathy, and guidance on how to talk with family members about risk.	Leventhal's Self-Regulation Model of Health Behavior	Printed materials	Study staff	Did not receive the Family Heart Talk booklet at the enrollment	Yes; first-degree relatives were contacted with participant's permission
Russo (2021)	U.S.	A patient choice study with a comparison group	Prostate cancer	127	A pretest video-based genetic education	An education video to address cancer inheritance, purpose of testing, risks and benefits of testing, multigene panel options, types of potential results, implications of results for treatment, screening, and cancer management, implications of hereditary cancer risk for blood relatives, genetic discrimination laws, and possible reproductive implications.	N/A	A video link	Study coordinator	Standard of care	No
van den Heuvel (2021)	Netherland	RCT	Inherited cardiac conditions	96	A tailored approach to inform relatives	(1) At the post-test counseling, probands were asked whether they preferred relatives to be informed by themselves or by the genetic counselor; (2) A family letter will be sent to all consented relatives after one month, regardless of whether they have been informed by probands. The family letter is standardized, which also includes a website specifically designed for this study where relatives can find additional information.	N/A	Printed materials	Genetic counselor	Standard of care	Yes; at-risk relatives were consented during participants' pre-testing counseling

Table 3D. Intervention outcome measures and results of updated review

Author (Year)	Outcome Measures					Results
	Primary outcome(s)	Other outcome(s)	Collection method	Follow-up time points	Format	
Burns (2022)	(1) The ability and confidence of the proband to communicate genetic results to their at-risk relatives (2) The number of first-degree relatives informed of their genetic test result	(1) Genetic knowledge (2) Satisfaction with genetic counseling (3) Genetic counseling outcome (e.g. empowerment)	Survey	2 weeks post intervention	Self-report	No between-group differences were found in all outcomes in primary outcomes.
Kinnamon (2023)	Completion of screening among eligible first-degree relatives within 12 months after proband enrollment	N/A	Enrollment of relatives in the study or reports of previous screening within 3 years	12 months	Audit/self-report	A higher percentage of eligible first-degree relatives completed screening in the Family Heart Talk arm (19.5% vs. 16.0%). The adjusted odds of screening completion among these first-degree relatives were higher in the Family Heart Talk arm (OR=1.30; one-sided 95% CI: 1.08 - ∞).
Russo (2021)	Participants' choice of pretest genetic education video (VBGE) vs.. pretest genetic counseling (GC)	(1) Decisional conflict for GT (2) Change in genetics knowledge (3) Satisfaction (4) Intention to share results with family and/or providers	Survey	After the pre-test session, after received genetic counselor results	Self-report	Significantly more participants chose VBGE (71%) versus GC (29%; p<.001). Participants who chose VBGE had higher intention to share GT results (96.4% VBGE v 86.4% GC, P = .02). No differences in other secondary outcomes.

van den Heuvel (2021)	Uptake of genetic counseling in relatives in the first year after test result disclosure	(1) Appreciation of the approach used to inform at-risk relatives; (2) Impact on psychological/family functioning	Survey/interview	3 months and 9 months	Self-report and chart audit	No between-group differences were found in the primary outcome, nor impacts on family/psychological functioning.

I see similarities in delivery settings for the new intervention studies. In this updated search, three of the four new studies were delivered in the context of either pre-test or post-test genetic counseling sessions. [221,222,224] The exception is one study delivered at the time of enrollment, where patients seen in heart failure and cardiac transplant programs were randomized to receive the intervention booklet.[223]

Additionally, participant samples in the updated search were more diverse compared to those identified in the original search. In the original search, the samples comprised women for six of nine studies. Three out of four new interventions enrolled both women and men, except for one study that focused on prostate cancer.[222]

3.5.3.b New evidence in intervention outcomes

Two new studies reported significant intervention effects (*Table 3D*). In Russo's intervention on prostate cancer germline testing, men who chose to participate in the intervention video education arm showed a higher intention to share genetic testing results with their relatives in the post-disclosure survey compared to those assigned to the control arm (96.4% vs. 86.4%, p=0.02).[222]

Kinnamon's Family Heart Talk intervention found that a higher percentage of eligible first-degree relatives completed clinical cardiovascular screening in the intervention arm at 12-month in comparison to relatives in the control arm (19.5% vs. 16.0%), however, the authors did not report whether this difference was statistically significant. Instead, they reported that after adjusting for participants randomization stratum, sex, and age quartile, the odds of screening completion were higher in the intervention group (OR = 1.30; one-sided 95% CI: 1.08 - ∞; one-sided p = 0.01).

3.5.3.c New evidence in strategies linked to theory

Similar to the original review, each of four newly identified interventions integrated at least one individual-level strategy. These strategies primarily focused on increasing participants' knowledge of genetic risk and genetic services, and their confidence to effectively communicate risk information with relatives, which can be aligned with SCT.

For example, Kinnamon's Family Heart Talk booklet was designed to provide guidance on how to talk with relatives about dilated cardiomyopathy risk. To build participants' knowledge, the booklet contains section such as "*which relatives should they start with*", "*what topics to cover in the genetic risk conversation*", and "*how to talk to young people about DCM*".[223] While the booklet explicitly mentioned Leventhal's Self-Regulation Model of Health Behavior,[225] few details were provided regarding how the booklet was mapped to the constructs of this individual level theory.

Similarly, Burn's communication aid for hypertrophic cardiomyopathy was intended to help participants effectively communicate genetic risk information to their at-risk relatives. The communication aid contains information such as genetic testing options for family members.[224,226] And in Russo's pre-test genetic education video, information such as consequences of a genetic result for at-risk relatives and implications of hereditary cancer risk for blood relatives were presented.[222]

In addition, van den Heuvel's intervention implicitly applied relational autonomy. As described in Chapter 1, relational autonomy views individuals as situated within a dynamic and interdependent web of relationships. Although the authors did not explicitly mention relational autonomy in developing their intervention, their tailored approach to information provision for relatives aligns with this conceptualization. Specifically, at the post-test counseling session, participants were asked which of these relatives they prefer to inform by themselves, and which relatives they prefer to be directly informed by the genetic counselor. Regardless of participants' preferences, the genetic counselor sent a family letter directly to all relatives for whom the participant has provided consent to contact, one month after the post-test counseling session. This intervention strategy acknowledges participants' knowledge with their specific dyadic relationships, therefore respecting their autonomy in decision-making. Notably, this study is the first in both the original and updated searches for its recognition of dyadic patterns at the relational level.

3.5.3.d Intervention acceptability

Van den Heuvel's study assessed participants' appreciation of the information provision strategy used and preferences regarding alternative contact approaches.[221] In addition, feasibility and acceptability of Burns' communication aid was assessed in a pilot study before the intervention.[224,226] In line with the results of the prior search, these studies reported high levels of acceptability.

3.5.4 Discussion

In summary, newer studies showed expansion in the genetic conditions, involvement of male participants, and adaptation of strategies used in HBOC contexts. However, none of the findings from these new studies would alter my conclusions from the original review. Developing and testing interventions that extend beyond genetic services settings and are tailored to the specific needs of different populations and hereditary conditions are needed. Adjunctive strategies that include relational content hold promise for improving FGRC, but their sustainability and broader applicability need to be tested.

Efforts to capitalize on the inherent interconnectedness of survivor-relative dyads were limited. Most of intervention strategies remain at the individual level, targeting at participants' intention to communication, genetic risk knowledge, and confidence to share. Very few interventions explicitly used behavioral theories to inform the development of strategies. Despite this, certain elements of these strategies were conceptually aligned with theories and constructs at the individual, relational, and family-system levels. But even for interventions that solely targeted individual-level factors, very limited benefits were found in terms of communicating inherited risk to relatives. To this end, I draw my conclusion that developing and rigorously testing interventions that extend beyond standard genetic services and include relational strategies are important for advancing FGRC and improving communication of inherited risks within families, addressing a variety of genetic conditions beyond HBOC.

CHAPTER 4. EXPLORATION OF WEBSITE ENGAGEMENT ACTIVITIES AS INDICATORS OF "TRANSFORMATION OF MOTIVATION"

4.1 INTRODUCTION

4.1.1 Background

As discussed earlier, interventions to promote family genetic risk communication (FGRC) have focused largely on the individual diagnosed with a condition who may carry a pathogenic variant (regardless of whether they have a confirmed diagnosis or not). These intervention strategies only rarely considered family interconnectedness on and its influence on communication to within the family system. Moreover, no prior interventions have explicitly targeted information processing and interpretation among families at a dyadic or family system level. This is surprising in that inherited risk has implications for interconnected blood relatives.

Relational level theories, such as Interdependence theory, argue that a health threat may be a cue to action not only for the proband but for their close relatives. The cue may prompt a transformation of motivation (*TM*) process and subsequent communal coping, leading individuals experiencing the health threat to convey risk information to relatives and in turn, relatives to take preventive actions out of desire to preserve family relationships. For example, an individual who is diagnosed with a heritable condition could be cued to protect relatives from experiencing a similar health outcome and move from a self-focused- ("I-ness") to a relationship-focused orientation ("we-ness").

I have posited that the likelihood of experiencing *TM* increases in the presence of two factors: a cognitive interpretation that the health threat is relevant and meaningful for family relationships, and that is prompts an emotional response such as fears about the well-being of family members. Together, these factors can prompt "we-thinking" in which actions to mitigate threat become a "we"-responsibility that requires "we"-oriented solutions. My aims for this dissertation are to evaluate if the *TM* can be inferred through observable or measurable variables that share a common variance.

This process involves conceptualizing domains of the latent construct of *TM,* developing specific measures, assessing the measures via surveys, or observing behaviors and testing the validity of the measure with quantitative statistics. As I reviewed in Chapter 1, researchers in social and family systems have yet to operationalize the *TM* construct. In Chapter 2, I posed the

question of could survivors' YFC website use patterns serve as an indicator of *TM*. Specifically, I posited that the greater the website engagement (e.g., time spent viewing the content), the greater the likelihood that the survivor was experiencing some *TM*. In contrast, survivors who more superficially engaged with the YFC website content (e.g., less time viewing the content) would be less inclined to "we-ness" thinking.

4.1.2 Changes to the Dissertation Proposal

Based on data availability, I needed to make two adjustments to the dissertation proposal. The YFC website did not record downloads of the Sensitive Conversation Framework. Thus, I was not able to fully test Assertion 3 from my proposal (See below for description). I added an assertion regarding engagement with the genetic counseling section of YFC and its association with *TM* (See below for description). A revised table that maps YFC website content with *TM* indications is provided below (*Table 4A*), as compared to the original *Table 2C* in Chapter 2.

Table 4A. Revised mapping website content with transformation of motivation indications

Head section	Website engagement	We-problem	We-responsibility	We-solution
Invite Family Members	FDRs and SDRs enumeration	√		
	Family relationship specification	√		
	Pros and cons of relative contact options		√	
	Selection of contact options		√	
	Family letter			√
Importance of Family Communication	National guidelines for genetic counseling	√		
	Genetic taxonomies (i.e., FDR, SDR)	√		
	Timing of genetic counseling			√
	Motives of FGRC	√	√	
Sensitive Conversation Framework	5-step sensitive conversation framework tipsheet		√	
	Download of the tip sheet		√	
Schedule Genetic Counseling	Genetic counseling Q&A			√
	Genetic counseling appointment			√

4.1.2.a Revision to assertion 3

In Chapter 2, I presented three assertions for mapping YFC website usage as indication of experiencing a *TM* process. In assertion 1, I expected that survivors' level of engagement with the enumeration section, contact choice selection, and relative letter download would be indicative of interpretating inherited cancer risk as a "we"- problem, responsibility, and solution. In assertion 2, I proposed that survivors' engagement with the "*Importance of Family Communication"* section would suggest that survivors see themselves as part of a family collective and as such, could prompt *TM* to protect their relatives by sharing information. These two assertions remain unchanged.

In assertion 3, I posited that downloading the Sensitive Communication Framework Tipsheet (SCFT) and spending more time reading this section would indicate a *TM* specifically, showing "we-responsibility". The intent of the SCFT information was to emphasize the affective aspect of inherited risk communication due to the shared health threat as a source of fear and

anxiety for families. The greater the time survivors spent time reviewing and then downloading the tip sheet should indicate a TM, specifically "we-responsibility". SCFT downloads were not recorded. However, time spent on the SCFT was retained as an indicator of *TM*.

4.1.2.b New assertion 4

After reviewing the survivor website engagement data and seeing that use of the genetic counseling section was high, I proposed a new assertion (i.e. Assertion 4). I assert that time spent viewing the section including educational information about genetic counseling and testing, and the opportunity to schedule genetic counseling appointments on the website would be indicative of experiencing the *TM*. As described earlier, this also asks survivors to fill out a 4-question survey concerning their past genetic counseling and testing history (e.g., have they or any close relative ever received genetic counseling and/or testing for possible hereditary cancer). And if yes, which relatives have been tested (e.g. self, mother, father, aunts, cousins), and what were the results (e.g. "*have a gene alteration that increases a person's chance of getting cancer*"). The section also describes the steps involved in genetic counseling and the benefits of counseling and testing survivors and family members. Survivors are offered two genetic counseling appointment options: a 1-hour individual appointment or a 1.5-hour appointment for family members to attend together. The inclusion of family appointments could stimulate cognitive and emotional processes and promote "we-solution" thinking.

To this end, my fourth assertion is that survivors' level of engagement with the genetic counseling section could indicate experiencing a *TM* .

4.1.3 Research Question

I now describe Aims 2 and 3, and how they iteratively begin to inform the conceptualization of the latent construct of TM. For Aim 2, I explore whether cancer survivors' engagement of the YFC website in the parent study might serve as behavioral indicators of *TM* (e.g., conceptually verifiable factors). For Aim 3, described in Chapter 5, I conducted qualitative structured interviews with survivors using think-aloud methods to gain insight into the subjective meanings of the factors I identified in Aim 2.

In this chapter, I focus on the methods and findings for Aim 2. The selection of website use measures will be discussed in detail in the 4.2 Methods section below. As a reminder, the research question for Aim 2 is: Can website engagement behaviors including reviewing information, downloading outreach materials, and enumerating relatives serve as indicators of the latent construct of "transformation of motivation"?

4.2 METHODS

4.2.1 Study Sampling

In the YFC parent study,[158] 1,953 ovarian cancer survivors were randomized to either the MBI or the Standard Care arm in a 1:1 ratio. As I described in Chapter 2, only website engagement patterns among ovarian cancer survivors randomized to the MBI arm are included because they were provided with website content that was specifically designed to prompt communal “we-ness” thinking. During the intervention window (July 2021 - September 2023), 152 survivors in the MBI arm accessed the YFC website. A detailed description regarding the website content and set up of the website is available in Chapter 2.

4.2.2 Creating Your Family Connects Engagement Indicators

4.2.2.a Your Family Connects website logs

All website use activities were tracked through a password-protected study portal and recorded along with a time stamp (i.e., hours, minutes, seconds). Each event in which a survivor logged in, logged out, viewed a web page, enumerated or removed a relative, or requested genetic counseling was tracked. For example, when a survivor used their access code for the first time, a log of “*activation code used*” was created. Additionally, if a survivor clicked the “*importance of family communication*” page on the website, an activity log of *“/home/familycommunication*” was generated. When a survivor clicked on the “+*add family members*” and “*save changes*” to add a new relative on the “*Invite Family Members*” page, a log of “*relative created*” was generated. If survivors enumerated multiple relatives on the website, multiple logs of "*relatives created*," with each entry corresponding to a specific relative, would be generated. Likewise, when a survivor removed a previously added relative by clicking the "*delete*" button, a log entry labeled "*relative deleted*" was created. A comprehensive overview of the website activity logs, showing the pre-cleaned raw counts of each type of logs among 152 participants, is present in Appendix 2.

Based on survivors’ enumeration entry, the website generated a relative list that included relative’s first name, family relationship (e.g., daughter, sister), and the contact approach chosen by the survivor for each enumerated relative. Additionally, survivors’ requests for an individual or family genetic counseling session also were tracked on the website. Survivors’ sociodemographic information was obtained from GCR that included current age, race/ethnicity, age at diagnosis, time since diagnosis, cancer stage, marital status, health insurance type, and rurality.

4.2.2.b Final analytic dataset

I took several steps to create the final analytic dataset. First, I removed all survivors’ personal identifiers and confidential information, including their first name, last name, phone

number, and email address in compliance with ethical regulations. I excluded 36 of 152 survivors from the dataset because of the survivor did not do anything on the website after consenting to participate (n=21) or viewed the landing page without any further interaction (n=15).

I identified and removed duplicate activity logs from both the “pages viewed” and “website interaction” logs (n=22). By duplicates, I am referring to activity logs where the same survivor (identified by their unique study ID) had identical activity log records, including log titles, log dates, and timestamps. For instance, if a survivor had two identical “*page viewed*” logs of “*/participant/familyintakesteptwo*” with matching timestamps, I would delete one of the two records to eliminate redundancy. Lastly, I deleted 1,143 automated system generated logs from the dataset. These logs were generated by scheduled notifications triggered within the main YFC trial. For example, if a survivor did not enumerate any relatives on the website, the website would automatically send them an email reminder, then a corresponding system-generated log titled “*Relative entry completion reminder – MBI*” would be logged. The final analytic dataset has 116 participants with a total of 2,456 activity logs.

4.2.3 Measurements

Drawing from current research on website engagement,[159-161] I created a set of 11 objective behavioral variables. These variables included both active and passive behaviors. Passive variables (n=4) comprised indicators of efforts taken to read the website content, that is the duration of time spent on the pages. Active variables (n=7) comprised actions taken by survivors on the website, including clicking on content and enumerating at-risk relatives.

The variables with their respective activity logs and sources, are detailed in *Table 4B*. Each category is mutually exclusive to ensure that the variables are orthogonal, minimizing ambiguity in the interpretation of the website engagement behavioral indicators with respect to the *TM* latent construct.

Table 4B. Website engagement measurement variables

Variable Name	Website logs/sources
Time spent on website content (by minutes)	
1. Enumeration sections	/participant/familyintake, /participant/familyintakestepfour /home/familyletters,
2. Genetic counseling sections	/counseling/aboutcounseling, /counseling/counselingquestions, /counseling/counselingrequest
3. Contact menu sections	/home/familycommunication, /participant/familyintakesteptwo, /participant/familyintakestepthree
4. Sensitive conversation tipsheet section	/home/sensitiveconversations
Clicks on website content	
5. Enumeration sections	relative created, relative deleted, step 1: family members added, step 4: finished family intake, /participant/familyintake, /participant/familyintakestepfour,

	/home/familyletters
6. Genetic counseling sections	read counseling information, counseling requested, counseling questionnaire completed, /counseling/aboutcounseling, /counseling/counselingquestions, /counseling/counselingrequest
Contact menu sections	step 2: viewed types of contact step 3: contact methods set, /home/familycommunication, /participant/familyintakesteptwo, /participant/familyintakestepthree
Family letter section	downloaded letter
Counts on the website	
9. Return logins	Login created Login
10. Relatives enumerated in total	YFC database
11. Distinct letters downloaded in total	YFC database

4.2.3.a Passive behavioral variables

I created 4 passive behavioral variables to assess the duration, presented in minutes, that survivors spent on 4 distinct sections of the YFC website: (1) enumeration sections (*variable 1*), (2) genetic counseling sections (*variable 2*), (3) contact menu sections (*variable 3*), and (4) SCFT section (*variable 4*). In calculating the total time spent on each variable, I followed a two-step approach. First, I calculated the time difference between entering and leaving each individual page using associated time stamps. Then, I summed up the durations of all pages within the *variable* to obtain the total time spent.

For example, in a scenario where a survivor visited the website for the first time, she would be directed to the landing page and presented with enumeration activities (refer to Chapter 2). When she entered the landing page, a log of "*page viewed – /participant/familyintake*" was recorded with a time stamp (e.g. "*06/21/2021 2:04:49 PM*"). Upon clicking "*Next*" to move to the next page presenting the menu of contact options, another activity log (i.e. "*page viewed – /participant/familyintakesteptwo*") was generated and time stamped (e.g. "*06/21/2021 2:14:51 PM*"). In this instance, the time spent viewing this individual enumeration page was calculated as 10.03 minutes, computed by the time difference between the two time stamps.

4.2.3.b Active behavioral variables

Clicks on MBI content is a continuous variable that comprises the counts of mouse clicks on: (1) enumeration sections (*variable 5*), (2) genetic counseling sections (*variable 6*), (3) contact menu sections (*variable 7*), and (4) family letter section (*variable 8*). I computed the total clicks for each variable as the sums of the number of logs in each section. For example, if a survivor initially enumerated three relatives but subsequently deleted one of them, the counts would be 4, counting 3 logs of "*relative created*" and a log of "*relative deleted*".

It is worth noting that activity logs including enumeration variables (*1&5*), genetic counseling variables (*2&6*), and contact menu variables (*3&7*) were coded as two different variables for passive and active behaviors. As described above, the passive behavior variables comprised the time stamp of the "*page viewed – /participant/familyintake*" log for duration calculation (*variable 1*), whereas for the active behavior variable, it is counted as one click when the survivor visited the page.

Return logins is a continuous (*variable 9)* count of the number of times a survivor visits and returns to the website assessed by "*login created*" and "*login*" respectively. Multiple logins on the same date are counted as separate visits, reflecting distinct occurrences of survivor action.

Enumeration of relatives is a continuous (*variable 10)* count of the total number of relatives enumerated by each survivor. Enumeration can occur either during the initial access to the website or through a subsequent visit to the "*invite family members*" page. This data is downloaded from the YFC data portal.

Download of family letters is a continuous count (*variable 11)* of the total number of family letters downloaded by each survivor. If a survivor downloads a letter multiple times for the same relative, it is considered as a single download for the purpose of this behavioral measure. This *variable* is distinct from *variable 8* (the clicks on family letter section) as survivors may click "*download*" multiple times for the same relative without initiating distinct downloads for different relatives. This data is downloaded from the YFC data portal.

4.2.4 Missing and Outliers

I differentiated missing data from a survivor not using specific pages. For example, if a survivor who visited the website did not click on the "*/participant/familyintake*" page, I recorded the count of clicks on that page as 0. Similarly, if there were no relatives enumerated or no family letters downloaded, I coded the counts as 0. Additionally, if there was no time spent on a particular page, the time spent on website content (*variables 1-4*) was coded as 0.

By contrast, data was coded as missing when a survivor failed to click "*logout*" to exit the website, the time spent on the last webpage was truly missing. Patterns of missingness for each time spent variable were inspected to determine whether there is systematic missing data that could bias the analysis.

Regarding outliers, there were three instances in variable *2* where the system recorded that survivors spent more than 60 minutes on the genetic counseling request page, and 1 instance in variable *8* where a survivor had 124 clicks on the family letter section. These

instances were deemed as outliers. Outlier and missing values were set to the mean for each individual page, and then the total new mean was calculated.

4.2.5 Data Analysis

4.2.5.a Exploratory factor analysis overview

I used exploratory factor analysis (EFA) to test whether variable loadings would indicate distinctive the passive and active behavioral factors I posited as potential indicators of *TM* . Factor analysis, a multivariate statistical method, tests whether based on intercorrelations and shared variance observed variables align with overt factors.[227-230] In contrast to confirmatory factor analysis (CFA), EFA assumes that each observed variable has the potential to measure an underlying latent variable. The goal is to identify the strongest linear associations between observed variables and possible underlying factors.

Linear EFA relies on assumptions of sample size and a normal distribution. The YFC dataset meets the rule of thumb of including 5 participants per variable with a minimum of 100 participants.[229] As EFA is grounded in the covariation among measured variables, departures from normality can potentially mislead the Pearson correlation coefficients (r). To mitigate the impact of skewness on EFA results, I ensured that all observed variables were valenced in the same direction.[228] EFA is sensitive to outliers (e.g., out-of-range values), as they can influence the size of the covariances/correlations based on which factor models are estimated. To address this assumption, I converted outliers to the mean as described above.

4.2.5.b Univariate and bivariate analyses

I performed univariate analyses to describe distribution of variables, identify outliers, and events of missingness for each variable. A bivariate correlation matrix was assessed to examine the associations between variables. Then, I assessed the communality estimates of 11 variables. This assessment was performed using squared multiple correlation (SMC), which indicates the extent to which a variable correlates with all other variables. Communality values range from 0 to 1, indicating whether the variable correlates with other variablesin the matrix. In other words, this assessment helps identify whether there are issues of singularity or multicollinearity for each variable. High or low communality values are suggestive of removing a variable from further EFA.[231]

4.2.5.c EFA diagnostic tests

I used two diagnostic measures to assess the appropriateness of the MBI website usage data for EFA. The first two were Bartlett's test of sphericity and the Kaiser-Meyer-Olkin (KMO) measure of sampling adequacy.[228,232] Bartlett's test of sphericity examines whether the correlation matrix is random or not. The null hypothesis of the test is that there is low probability

of observing a statistic this large or larger by chance alone. The KMO measure assesses the proportion of variance in included variables that might be attributed to underlying factors. Values close to 1.0 indicate that factors may be suitable. The generally recommended threshold for the KMO measure is above .80 for overall acceptability, and below .50 is deemed unacceptable, a value above .60 is considered tolerable.

4.2.5.d EFA steps

I conducted EFA using SAS 9.4 PROC FACTOR. To simplify factor solutions and to ensure unidimensional representation of latent variables, I based variables on the *a priori* theoretical mapping I outlined in Chapter 2. I used maximum likelihood extraction to produce more generalizable and reproducible results, as it does not inflate variance estimates. I examined eigenvalues and scree plots of the initial EFA model to determine the number of factors to extract. Eigenvalues greater than 1 were retained. In addition, I conducted a visual inspection of the scree plot to help decide on the number of factors to be retained. The scree plot shows the magnitude of the component eigenvalues in descending order, and factors above a distinct break in the slope (i.e., the "elbow") were retained. Lastly, I submitted factors for promax rotation, a type of oblique rotation, that assumes the factors are correlated.[233] This is contrasted to orthogonal rotation that assumes no correlation. I used promax rotation because it is relatively efficient at achieving a simple oblique structure. Factor scores were generated using factor score coefficient matrix.

4.3 RESULTS

4.3.1 Demographics

116 ovarian cancer survivors with a total of 2,456 activity logs are included in the final analysis (*Table 4C*). The mean age of the group is 62.5 years old (SD=12.2). Survivors' average age at diagnosis was 52.4 years (SD=11.9) and women received their diagnosis an average of 9.6 years (SD=3.8) prior. Most survivors were White (n=90, 77.6%) and non-Hispanic (n=112, 96.6%). Most were married or had a domestic partner (n=73, 64.4%). The majority had health insurance coverage (n=103, 96.3%) and lived in an urban commuting area (n=92, 83.6%). Over two-thirds had ovarian cancers diagnosed at the regional or distant stages (n=81, 69.8%).

Table 4C. Demographics (n=116)

Covariate	Mean/n	SD/Col %
Current age	62.51	12.22
Age at Diagnosis	52.43	11.92
Time since diagnosis	9.66	3.83
Race		
White	90	77.59%
Black	25	21.55%

Asian	1	0.86%
Hispanic		
Non-Hispanic	112	96.55%
Hispanic	4	3.45%
Marital Status		
Single	21	18.58%
Married/domestic partner	73	64.60%
Separated/Divorced/Widowed	19	16.81%
Payer/Insurance		
Not insured	4	3.74%
Private insurance	84	78.50%
Medicaid/Medicare	17	15.89%
Military/VA/Public health services	2	1.87%
Rural/Urban commuting area (RUCA 2010)		
Urban commuting area	92	83.64%
Not an urban commuting area	18	16.36%
Urban/Rural indicator (URIC 2010)		
All urban	61	55.45%
Mostly urban	28	25.45%
Mostly rural	13	11.82%
All rural	8	7.27%
Cancer stage		
In situ	2	1.72%
Localized	35	30.17%
Regional	44	37.93%
Distant	33	28.45%
Unstaged	2	1.72%

4.3.2 Univariate and Bivariate Results

4.3.2.a Univariate analysis results

The univariate analysis of 11 variables is presented in *Table 4D*. On average, survivors logged in to the website 1.57 times (SD=1.25; variable *9*), enumerated one relative (mean = 1.4; SD=1.59; variable *10*), and downloaded one letter (mean = .87; SD=1.39; variable *11*). Survivors spent the most time on relative enumeration sections (mean = 4.8 minutes; SD=5.7; variable *1*) and the least time on the sensitive conversation tipsheet section (mean = 0.44 minutes; SD=1.78; variable *4*). In terms of clicks on the website sections (variables *5-8*), survivors clicked most frequently on the enumeration section (mean=6.09, SD=5.83; variable *5*) and the fewest clicks on the family letter section (mean=1.02, SD=1.69; variable *8*).

Table 4D. Univariate analysis results (n=116)

Variable Name	Mean	Std Dev	Mode	Min	Q1	Median	Q3	Max
Time spent on website content (by minutes)								
1. Enumeration sections	4.85	5.73	0.85	0.2	1.77	2.77	5.54	29.19
2. Genetic counseling sections	4.03	7.27	0	0	0	2.28	3.99	43.92
3. Contact menu sections	3.24	4.22	0.79	0	1.02	1.63	3.32	21.98
4. Sensitive conversation tipsheet section	0.44	1.78	0	0	0	0	0	14.52
Clicks on website content								
5. Enumeration sections	6.09	5.83	2	1	2	5	7	39
6. Genetic counseling sections	4.65	4.29	0	0	0	6	7	22
7. Contact menu sections	4.86	2.74	4	0	3.5	4	6	17
8. Family letter section	1.02	1.69	0	0	0	0	1.5	9
Counts on the website								

9. Return logins	1.57	1.25	1	1	1	1	2	9
10. Relatives enumerated in total	1.41	1.59	0	0	0	1	2	7
11. Distinct letters downloaded in total	0.87	1.39	0	0	0	0	1	7

4.3.2.b Bivariate results

To further inform factor development, I generated a bivariate correlation matrix to look for inter-variable correlations (*Table 4E*). Most of the variables were significantly correlated with each other, ranging from a very high correlation (r=0.94) for clicks in the family letter section (*variable 8)* with distinct letters downloaded (*variable 11),* to lower correlations (r=0.20) such as time spent on the contact option section (*variable 3)* with distinct letters downloaded (*variable 11)*.

Correlations below 0.3 or above 0.8 can signal variables with multicollinearity issues.[229-231] Multicollinearity occurs when two or more measures are highly correlated with each other. Extreme multicollinearity (i.e. singularity) can indicate redundancy among variables, where one variable becomes a linear combination of the others. Multicollinearity could compromise the reliability of the factor structure and result in inconsistent loadings. In particular, two variables related to content concerning family letters (*variables 8 and 11*) were strongly correlated (r=0.94). The counts of clicks on the family letter section aligned closely with the number of distinct letters that were eventually downloaded even though they were theoretically mapped as two distinct actions.

Table 4E. Correlation table of 11 variables

	1[a]	**2[a]**	**3[a]**	**4[a]**	**5[b]**	**6[b]**	**7[b]**	**8[b]**	**9[c]**	**10[c]**	**11[c]**
1[a]	1										
2[a]	0.35***	1									
3[a]	0.24**	0.39***	1								
4[a]	0.20*	0.46***	0.34***	1							
5[b]	0.53***	0.24*	0.44***	0.24**	1						
6[b]	0.30**	0.61***	0.17	0.29**	0.49***	1					
7[b]	0.25**	0.27**	0.52***	0.36***	0.57***	0.33***	1				
8[b]	0.63***	0.21*	0.20*	0.11	0.71***	0.40***	0.29**	1			
9[c]	0.42***	0.53***	0.36***	0.44***	0.38***	0.51***	0.48***	0.33***	1		
10[c]	0.53***	0.27**	0.20*	0.18	0.66***	0.53***	0.24**	0.72***	0.22*	1	
11[c]	0.52***	0.22*	0.21*	0.09	0.66***	0.45***	0.28**	0.94***	0.28**	0.76***	1

a. *Time spent on website content (by minutes): Variable 1. Enumeration sections; Variable 2. Genetic counseling sections; Variable 3. Contact menu sections; Variable 4. Sensitive conversation tipsheet section*
b. *Clicks on website content: Variable 5. Enumeration sections; Variable 6.Genetic counseling sections; Variable 7.Contact menu sections; Variable 8. Family letter section.*
c. *Counts on the website: Variable 9.Return logins; Variable 10. Relatives enumerated in total; Variable 11.Distinct letters downloaded in total*
**p<0.05; ** P<0.01; ***p<0.001*

In addition, using the SMC test, I assessed the communality estimates to determine how each individual variable correlated with all other variables (*Table 4F*). Again, I found that variables 8 and 11 had high communality estimates of 0.93 and 0.92 respectively, indicating an

issue of singularity. Therefore, I made the decision to retain variable *11* (i.e., distinct letters downloaded) but drop variable *8* (i.e., clicks on the family letter section) from EFA analysis. My rationale for this decision is that downloading distinct letters activates survivors' consideration of their relational ties and interconnectedness that I posit to indicate viewing genetic risk communication as a “we”-problem. As a result, EFA was reported on a 10-variable model, excluding variable 11.

Table 4F. Prior communalities for 11 variables

variable		Communalities
Time spent on website content (by minutes)		
1	Enumeration sections	0.56
2	Genetic counseling sections	0.59
3	Contact menu sections	0.46
4	Sensitive conversation tipsheet section	0.32
Clicks on website content		
5	Enumeration sections	0.75
6	Genetic counseling sections	0.65
7	Contact menu sections	0.51
8	Family letter section	0.93
Counts on the website		
9	Return logins	0.53
10	Relatives enumerated in total	0.70
11	Distinct letters downloaded in total	0.92

Extraction Method: Maximum likelihood

4.3.3 Exploratory Factor Analysis Diagnostic Results

4.3.3.a EFA diagnostic assessments

I conducted two EFA diagnostic assessments to evaluate the appropriateness of the 10-variable model. The value of Bartlett’s test for correlation was $\chi2(45) = 563.3726$, $p < .0001$. Therefore, I rejected the null hypothesis and concluded that it is reasonable to consider applying a dimension-reduction technique, such as EFA, to the YFC website use data. The overall KMO value is 0.7649, which falls within the tolerable range, suggesting that the data may still be suitable for factor analysis, albeit with some caution.

4.3.3.b EFA initial model

As shown in the initial 10-variable model (*Table 4G*), two factors have eigenvalues greater than 1, indicating that there is a maximum of two meaningful factors to be retained in the final model. Because this is an unrotated model, the first factor always accounts for the most variance (72.8%), followed by the factor with the second highest proportion of variance (20.7%). The scree plot supported this finding, with two factors positioned to the right of the “elbow” instead of three. From the third factor on, there was a downward trend, indicating that each subsequent factor is accounting for progressively smaller portions of the total variance. Thus, I

conducted additional EFAs to explore models with a combination of rotations and 2 factors extracted.

Table 4G. Pattern factor loadings for EFA of initial 10-variable model

		Factor1	Factor2	Factor3
Time spent on website content (by minutes)				
1	Enumeration sections	0.62	-0.17	-0.02
2	Genetic counseling sections	0.60	0.41	-0.34
3	Contact menu sections	0.51	0.29	0.34
4	Sensitive conversation tipsheet section	0.44	0.39	0.00
Clicks on website content				
5	Enumeration sections	0.79	-0.25	0.27
6	Genetic counseling sections	0.69	0.05	-0.39
7	Contact menu sections	0.59	0.24	0.38
Counts on the website				
9	Return logins	0.64	0.36	-0.06
10	Relatives enumerated in total	0.71	-0.48	-0.10
11	Distinct letters downloaded in total	0.69	-0.48	-0.01
	Eigenvalues	4.03	1.14	0.62

Extraction Method: Maximum likelihood

4.3.3.c EFA final model

The final EFA model comprising 10 variables is presented in *Table 4H.* Following rotation, each variable had a high loading on one factor and a low loading on the other, making the interpretation of factors easier. Based on this model, I consider two factors may align with the latent construct of *TM*. The first factor consists of variables: *1*, *5*, *10*, *and 11*. The second factor consists of variables: *2*, *3*, *4*, *7*, *and 9*. Variable 6 did not load on either factor. The inter-factor correlation was r=0.4816. Eigenvalues, that is variance explained by factor 1 and factor 2, were 2.71 and 2.47.

Table 4H. Pattern factor loadings for EFA of final 10-variable model

		Factor1	Factor2
Time spent on website content (by minutes)			
1	Enumeration sections	0.57	0.29
2	Genetic counseling sections	0.16	0.71
3	Contact menu sections	0.18	0.56
4	Sensitive conversation tipsheet section	0.06	0.59
Clicks on website content			
5	Enumeration sections	0.75	0.36
6	Genetic counseling sections	0.47	0.51
7	Contact menu sections	0.27	0.58
Counts on the website			
9	Return logins	0.23	0.70
10	Relatives enumerated in total	0.85	0.13
11	Distinct letters downloaded in total	0.83	0.11
	Eigenvalues	2.71	2.47

Extraction Method: Maximum likelihood
Rotation Method: Promax rotation

4.4 DISCUSSION

Website engagement data from the YFC parent study offered the opportunity to explore possible passive and active behavioral indicators of *TM*. Informed by relational and family systems theories (Chapter 1), I took a preliminary step to explore the extent to which cancer survivors' website use might indicate "we-ness" thinking, consideration of the health event as a shared "we"-problem and responsibility with the communal coping solution of FGRC. My EFA analyses indicated that the data could be summarized in two factors.

4.4.1 Interpretations of Two Factors

Three variables relating to enumeration (e.g., time spent on this section) and one variable related to downloading a family letter loaded on the same factor, conceptually indicating action-oriented behavior. I submit that this factor may reflect survivors' response to becoming informed or reminded that ovarian cancer represents a shared health threat and they are taking the opportunity offered by the website to take an action. The enumeration page offered guidance to survivors to think through their family relationships such that more time spent and more clicks on the enumeration page would be expected to encourage a shift from an individualistic mindset ("I") to a more relational one ("we"). Based on Relational Autonomy, the process of enumerating relatives requires the survivor to consider relatives' perspectives and needs related to the shared health threat. This inclination to communal thinking in turn, could motivate survivors to see the importance of preserving relationships with each relative.

The second factor comprised behaviors used to review website content related to genetic counseling, relative contact options, and initiating sensitive conversations. The website content related to this factor encouraged reciprocal exchange of health information and he need for ongoing FGRC regarding the shared health threat. The contact menu and options for genetic counseling appointments encouraged survivors to consider the specific dynamics of their relationships with each enumerated relative. The number of returns to the website also loaded on this factor.

In Chapter 2, I posited that more time spent in these activities would indicate that survivors were engaging in systematic processing about the importance of communicating genetic risk as beneficial to preserve family health and in turn, sustain the relationship. Taken together, I conclude that this factor is a indicator of systematic information processing regarding the relationality of inherited risk that could be indicative of TM.

As one of the very first attempts to operationalize the latent *TM* construct, my Aim 2 study holds a strong theoretical premise. It was built upon a population-based communication outreach intervention, drawing from the latest theorizing in Relational Autonomy as a guiding

ethical principle.[157] However, these results are preliminary and the two factors do not constitute a measure of *TM*.

It is important to acknowledge the study's limitations. First, the sample size for my EFA is small. As EFA works better with larger sample sizes of greater than 300 participants, the small sample size can lead to unreliable factor structures and unstable estimates. Second, missing data in the time spent variables were replaced with the means. This approach could either overestimate or underestimate the actual time users spent on the page, introducing potential bias into the results. As a result, this method of handling missing data might skew the findings, affecting my ability to draw conclusions from the analysis. Future research should collect data from additional participants to validate and potentially replicate the results.

4.4.2. Conclusions

The purpose of Aim 2 is to fill the research gap where there is no existing validated and reliable measure to assess the latent *TM* construct. I took the efforts to derive ovarian cancer survivors' website engagement behaviors into 2 factors. While I do not suggest that the two factors are the ultimate set for measurement item developments, the results are promising to build on, especially for developing a *TM* scale in the context of FGRC and identifying of amendable intervention strategies to promote the relational thinking. In the next Aim, I will continue my exploration of the extent to which these two factors may indicate "we-thinking",

CHAPTER 5. ASSESSMENT OF "TRANSFORMATION OF MOTIVATION" FACTORS INSIGHTS USING THE THINK-ALOUD APPROACH

5.1 INTRODUCTION

5.1.1 Background for Aim 3

In Aim 3, I continue to explore whether the two website use factors I found in Aim 2 analyses can be further conceptualized as indicators of transformation of motivation (*TM*). To that end, I have the following research questions: (1) To what extent can linguistic indicators of "we-ness" thinking and communal coping be identified among women who are engaging with the YFC website in real time?; (2) To what extent are family size and subjectively rated closeness associated with the frequency of these linguistic indicators?; and (3) What insights can be gained from individual survivors' lived experiences to inform future research of the latent construct of "transformation of motivation"? I will discuss how the qualitative data collected lends deepened insights into the website use factors identified to signify *TM* process.

5.1.2 Rationale for the Think-Aloud Approach

As described in Chapter 1, the classic think-aloud method engages participants to verbalize their thoughts while using the YFC website content. I opted for a retrospective think-aloud method because it is less disruptive than concurrent think aloud as survivors are not expected to navigate the website while answering interview questions. The retrospective approach also allowed ovarian cancer survivors to explore the website independently before the interview, mimicking a natural, uninterrupted environment akin to Aim 2.

There were two caveats to this replication, survivors in the think-aloud Aim 3: (1) did not have the option to return to the website after completing the interview as survivors had in the parent study and (2) the genetic counseling schedule page on the website was disabled at the end of the main trial. Therefore, think-aloud participants were not able to schedule a genetic counseling session.

5.1.3 Replicated Factor Analyses for Survivors in The Structured Interview Study

A comparison of whether variable loadings found in Aim 2 were observed in this small sample of survivors recruited for Aim 3. Due to the small sample size (N=19) the study was underpowered for confirmatory factor analysis. As anticipated, the confirmatory factor analysis model yielded two factors with poor model fit (x^2=0.04, GFI=0.74; SRMR=0.15; RMSEA=0.17). I

found statistically significant differences between the two samples on 4 variables (*Table 5A*). Due to this insufficient sample size, the posit could not be properly assessed.

Table 5A. Aim 2 and Aim 3 website engagement data comparisons

Variable name	Aim 2 (n=116)		Aim 3 (n=19)		T-test
	Mean	Std Dev	Mean	Std Dev	
Time spent on website content (by minutes)					
1. Enumeration sections	4.85	5.73	8.79	9.09	-1.83
2. Genetic counseling sections	4.03	7.27	9.4	9.56	-2.85**
3. Contact menu sections	3.24	4.22	4.24	2.51	-1.44
4. Sensitive conversation tipsheet section	0.44	1.78	1.39	2.59	-1.53
Clicks on website content					
5. Enumeration sections	6.09	5.83	8.79	4.96	-1.90
6. Genetic counseling sections	4.65	4.29	7.47	3.86	-2.70**
7. Contact menu sections	4.86	2.74	7.26	4.13	-2.45*
8. Family letter section	1.02	1.69	1.16	2.06	-0.33
Counts on the website					
9. Return logins	1.57	1.25	2.42	1.43	-2.69**
10. Relatives enumerated in total	1.41	1.59	1.95	2.17	-1.28
11. Distinct letters downloaded in total	0.87	1.39	0.84	1.17	0.08

*p≤ 0.5; **p≤ 0.01; ***p≤0.001

5.1.4 Proxy Measures of Transformation of Motivation Thinking

As introduced in Chapter 1, the two most commonly used measures are the linguistic utterances of first-person plural pronouns[148,150,153,234] and the Inclusion of Other in the Self (IOS) scale.[141,150,155] As the *TM* latent construct remains conceptual, utilizing these existing measures could provide a link between the website use factors and survivors' subjective responses to the website content that was not assessed in the parent study.

5.1.4.a Plural utterances as a linguistic marker of TM

Current research related to interdependence theory has taken first-person plural pronoun use, as opposite to first-person singular pronoun use, as an implicit marker of communal thinking.[141,147-150] When survivors' are confronted with the shared threat of inherited cancer risk their motives can become relationship-oriented, and move from two "I"s to one collective "we". That is, they may be prompted to see self as part of a greater whole. This may be signaled by the frequency in which they use of first-person plural pronouns (e.g., we, us, our; "we"-talk).

After reviewing the interview transcripts, I also noted that survivors used third person plural pronouns (e.g., they, them, their; "they"-talk) with some frequency when talking about family relationships. While third-person pronoun use has not been considered previously, I argue that their use also may suggest that survivors visualize their relatives as connected entities separate from the self. Thus, use of these pronouns could indicate that survivors may be acknowledging concerns for relatives over self as they consider how to address a shared threat.

I posit that ovarian cancer survivors who have higher use of plural utterances in "we"-talk and "they"-talk, compared to "I"-talk, would be more likely to have experienced *TM*.

5.1.4.b Graphic assessment of interdependence

I assessed participants' emotional communal coping using a visual scale. The Inclusion of Other in the Self scale assesses individuals' perception of relational closeness in a pictorial format.[155] As the name implies, this scale assesses participants' perceived relational closeness based on the extent to which they perceive themselves in relation to the other using a scale of overlapping circles.

I adapted the scale instructions to focus on families as a collective unit, rather than as dyadic relationships (*Appendix 3*). A total of seven closeness circles were provided. Survivors were asked to select the pair of circles to best represent their emotional closeness with their family. The highest level of closeness was represented by one fully overlapping circle, while the lowest closeness contains two independent circles. I hypothesize that ovarian cancer survivors who report higher visual emotional closeness would be more likely to have experienced *TM*.

5.2 METHODS

5.2.1 Participant Recruitment

Ovarian cancer survivors were recruited to participate if they: (1) were eligible for the parent YFC study regardless of randomization arm, (2) did not use the website access code during the main trial period, (3) returned a follow-up survey to a parent study survey of survivors who did not participate, (3) spoke English, and (4) had internet access.

Following the closure of the parent YFC trial, the research team conducted a survey of a sample of survivors who chose not to visit the study website. Collaborating with Georgia Cancer Registry (GCR), I randomly sampled 200 of these survivors with the goal to recruit 20 for the think-aloud activity. The GCR mailed out recruitment packages to these 200 survivors. The packet included a flyer, a cover letter, and a consent for contact information release form to women's most recent home address. The consent for contact information release form provided assurance to eligible survivors that their personal identifiers would not be shared with me without first obtaining their consent. The consent form also clarified that agreeing to release their contact information would not automatically commit them to participate but would allow me to contact them directly to provide further information about the think-aloud study.

The recruitment period was between July 2023 to October 2023. Recruitment packages were sent out in two batches, with 100 participants included in each batch. Those who did not respond within four weeks received a follow-up reminder packet that contained the same

materials as in the initial recruitment package. For those who returned the consent for contact information release form, master-student-level research assistants (I.S., and Q.C.) made up to 5 call attempts for consent and screened a total of 40 eligible survivors. Of the 40, 6 were ineligible due to having no internet access, 12 were lost to follow-up, 4 consented but did not visit the website nor respond to call to reschedule.

The final sample size was 20 survivors. Of these, one survivor did not visit the website before the interview and therefore did a concurrent think-aloud interview. Her website engagement was excluded in Aim 2 and Aim 3 comparison *Table 5A*, but other measures of hers, such as linguistic markers and family emotional closeness are included for the rest of analyses.

Verbal consent was obtained during the consent call and an electronic copy of full consent form was emailed to them after. A $150 Amazon electronic gift card was offered to participants who completed the interview in thanks for their time and participation. All study procedures were approved by the Institutional Review Board at Emory University.

5.2.2 Study Design

5.2.2.a Development of the interview guide

I developed the semi-structured interview guide in collaboration with C.M.M.. Questions were aimed to elicit women to talk about their thoughts as they engaged in enumeration-related behaviors (Factor 1) and their thoughts as they reviewed website content (Factor 2). The interview guide drafts were pilot tested with C.M.M. and I.S. before finalization. The final interview guide contained 16 questions, each followed by clarification questions and standard probes for eliciting more detailed elaboration on responses.

The interview guide begins with a brief introduction where the interviewer reviews the survivors' website engagement activities. This includes the time spent on each *TM* theory-mapped section, the number of relatives identified, and the number of family letters downloaded. Following this, survivors were provided with a brief explanation about the process of think-aloud interviews and were asked to view the website content and share their reactions and thoughts about the content.

As described in Chapter 2, the website visit followed a specific sequence, beginning with sections on *Inviting Family Members* and *Genetic Counseling*, and followed by the *Importance of Family Communication* and *Sensitive Conversation Framework* sections. The structure of the interview guide follows in the same sequence. Questions are section-specific and aligned with one of the two identified factors (Aim 2). For example, the first section of interview guide is about *Inviting Family Members and* contains 9 questions. These questions focused on the

survivor's thinking behind their choices of which relatives to identify, and reasons to list these relatives but not others. An example question is "*how did the fact that ovarian cancer can run in families influence your choice of which relatives to identify?*" A copy of the interview guide is included in *Appendix 4.*

5.2.2.b Interviewer Training

I conducted 5 hours of training over 3 days with each of two interviewers (I.S. and Q.C.). The training began with a one-on-one session to introduce the Aim 3 study process and the think-aloud approach. Then each interviewer had two observation sessions: one where they watched me conduct a think-aloud interview with a survivor, and another where I observed them conducting an interview. Following these observations, we held a discussion session to address any issues noticed during the actual interviews. Once this training was completed, I.S. and Q.C. began to independently conduct interviews. Of the final 20 interviews, I conducted the majority of the interviews (70%), followed by I.S. of 20%, and Q.C. of 10%.

5.2.2.c Study procedures

The study consisted of three sequential procedures; all were completed remotely. In step 1, survivors were asked to navigate through the YFC website on their own at least 1-day before the scheduled interview time. If unable to do so, the research team would reach out again to reschedule the interview. The research team did not observe the survivor during this step. Use data was collected for this session as was done for survivors in Aim 2.

In step 2, after the completion of website visit, but before the interview, survivors completed a brief online survey (3 items) that assessed the first names of their blood relatives and their relationship to each that the survivor had chosen ***not*** to enumerate on the website during step 1. Note, the list of enumerated relatives was collected automatically on the website. The online survey also assessed survivors' subjective assessment of family emotional closeness using the revised Inclusion of Other in Self scale.

In step 3, a one-on-one retrospective think-aloud interview was conducted via Zoom and audio-recorded. The interviews were conducted between August to October 2023. The survivor and interviewer viewed the YFC website together. Concurrently, the interviewer asked the survivor to talk-aloud to give their thought process for each of the website pages including the information they found useful, and the actions they took while on the website (e.g., did or did not download family letters). To ensure consistency, a new study account was created to replicate their website visit experience in step 1. All questions in the interview guide were asked regardless of whether survivors had previously visited the website section in its entirety during the initial step.

5.2.3 Quantitative Data Measures

5.2.3.a Linguistic markers of TM

Drawing from previous “we-language” studies, and consistent with the Linguistic Inquiry and Word Count (LIWC) Dictionary,[235] I defined "I”-pronouns as "I", "I’m", "I’ll", "I’d", "I’ve", "me", "my", "myself", "mine"; “we”-pronouns as "we", "we’re", "we’ll", "we’d", "we’ve", "us", "let’s", "our", "ours", "ourselves"; “they”-pronouns as “they", "them", "their", "they're", "they've", "theirs", "themselves".

Counts of survivor utterances were performed using the quanteda package (Version 4.0) in R (version 4.3.2).[236] This package is purposefully designed to efficiently manage and analyze textual data. Before I loaded the interview transcripts to R, Q.C. helped to remove all interviewer lines from the transcripts to avoid the inclusion of “I”-, “we”-, “they”-pronoun counts said by interviewers. Then, the quanteda package quantified and aggregated pronoun counts within each transcript. This analysis yields absolute counts of "I”-pronouns, "we”-pronouns, and “they”-pronouns categories, as well as total interview words. To create linguistic markers of communal coping, I divided the absolute count of each pronoun over total interview words, namely “I”-talk, “we”-talk, and “they”-talk.

5.2.3.b Survey and website measures

Family size of each survivor was assessed in two stages. First, during their independent website visit in Step 1, survivors could enumerate relatives of their choosing on the website. Second, in the subsequent survey (Step 2), survivors were asked two questions, one about first-degree relatives (FDRs) and one about second-degree relatives (SDRs), prompting them to list the names and relationships of relatives ***not*** previously listed on the website. At this time survivors were asked why they did not enumerate the relative (e.g., privacy concerns, large family size). Family size was computed by adding up the numbers of enumerated relatives on the website, and the numbers of relatives not enumerated but reported in the survey.

Survivors tended to omit some of their FDRs and SDRs. This omission occurred especially among participants from larger families, who found reporting all relatives burdensome. This tendency led to inaccuracies in the family size variable. To address this, interviewers also asked participants to share their family size during the interview. I also audited the interviews to identify any relatives that were missed on the survey. To account for these data collection errors, I chose to dichotomize family size with a cut-off at the median (n=8), for a rough designation of larger and smaller families.

Family closeness was measured using an adapted Inclusion of Other in Self scale.[155] A graphic of seven pairs of circles where one circle in each pair is labeled “self,” and the second

circle is labeled “family” was provided. Survivors were asked to choose one of the seven pairs to answer the question, “*Think of your family as a whole, which pair best describes how close you feel with your family in emotional closeness?*” The 7 pair of circles were accompanied with scores that indicate different degrees of overlap: 1 = no overlap, 2 = little overlap, 3 = some overlap, 4 = equal overlap, 5 = strong overlap, 6 = very strong overlap, and 7 = most overlap.

The distribution of data suggested that survivors perceptions of family emotional closeness was highly skewed (mean=5.1; std=2.05; mode=7; min=1; q1=4.5; median=5; q3=7; max=7). Given this distribution, I dichotomized the family closeness variable with a cut-off score of 6. Families with scores of 6 or higher were categorized as having high emotional closeness, while those with scores of 5 or lower categorized as not having high emotional closeness.

History of GC testing was assessed during survivors’ step 1 website visit. Survivors were asked to complete a four-item genetic counseling questionnaire as described in Chapter 2. An example question includes "*Have you or a close relative ever received genetic counseling for cancer? [yes/no/did not answer*]”. If the survivor did not visit the GC survey page during step 1, trained interviewers asked survivors to fill out the survey concurrently during the step 3 think-aloud interview.

5.2.4 Quantitative Data Analysis

SAS® 9.4 (Cary, NC) was used for data cleaning and analysis. I first ran univariate analyses of all variables of interest, including linguistic measures, family closeness, family size, absolute number of relatives listed on the website, and history of genetic testing. I assessed bivariate associations for “I”-, “we”-, and “they”-talk using Pearson’s Correlation, and the difference means for use of these language indicators of interdependence in the high and low family size groups, and high and non-high family closeness groups using interdependence sample t-test.

Because of the qualitative nature of these analysis, and accounting for the very small sample size (n=20), the level of statistical significance must be interpreted with caution.[237-240] The analyses are underpowered to interpret at the level of $p \leq 0.05$. Instead, I rely on confidence intervals to indicate if there were trends in the bivariate associations.

5.2.5 Qualitative Data Analysis

I used MaxQDA 2022 to analyze the verbatim transcriptions of the structured interviews. I used the deductive thematic analysis approach. I started by familiarizing myself with the data. C.M.M and I read five transcripts independently and discussed our general impressions and potential codes to create. Based on these five transcripts, I created a set of initial codes to represent the patterns we observed in the data, along with example excerpts of each initial

code. To test the applicability of the code, we selected three additional interviews, and applied the predefined codes to these interviews and made further modifications as needed. The coding of the remaining 12 transcripts was conducted in two rounds. In each round, C.M.M. and I coded two of the same transcripts to check inter-rater reliability; in the final round, our inter-rater reliability was over 80%.

5.2.5.a Approach to analyzing qualitative interviews to understand the two factors

I reviewed interview responses given while viewing the enumeration sections of the website to identify themes in survivors' discussions about their rationale for enumerating relatives (Factor 1). I identified four themes that influenced survivors' choices of who to enumerate: perceived emotional closeness to the relative, relative's age and gender, a sense of one-and-done, and relational appraisals of the benefits of enumeration.

Similarly, I reviewed interview responses related to engagement with the information content on the website (i.e., menu of relative contact options, the sensitive conversation tipsheet, and genetic counseling sections; Factor 2). I identified three themes: layered contact was preferred, exploring "we"-responsibility, and genetic counseling as part of "we"-solution.

In my efforts to explicate each factor and consistent with my prior supposition, I would posit that enumeration actions and content engagement on the YFC website would be greater among survivors who used relatively more plural "we"-talk (i.e., high we-talkers) compared to those who used very little "we"-talk (i.e., low we-talkers). To explore this supposition, I compared survivors with the highest we-talk profiles to those with the lowest we talk profiles (See Results Section).

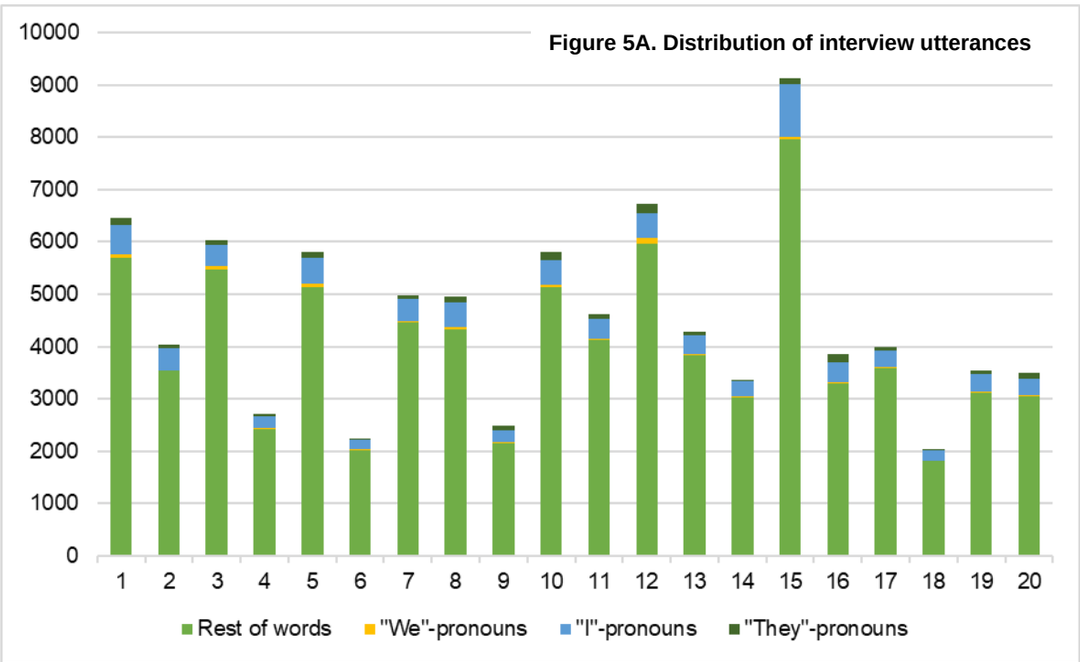


Figure 5A. Distribution of interview utterances

5.3 RESULTS

Table 5B. Univariate results of Aim 3 variables (n=20)

Variable	Mean/n	SD/ Col%
Interview lengths (by minutes)		
Absolute counts		
"I" pronouns	397.15	179.60
"We" pronouns	37.05	28.71
"They" pronouns	90.85	45.98
Total interview words	4528.45	1762.86
Linguistic markers of communal coping		
"I"-talk [a]	8.77%	10.36%
"We"-talk [b]	0.79%	0.39%
"They"-talk [c]	1.99%	0.84%
Number of relatives listed	1.85	2.16
Number of relatives not listed	8.15	4.75
Family size		
Small family (<8 total relatives reported)	8	40%
Large family (>= 8 total relatives reported)	12	60%
Family emotional closeness		
Not high (=<5)	9	45%
High (>=6)	11	55%
GCQ1. Have you or a close relative ever received genetic counseling for cancer?		
No	7	35%
Yes	13	65%
GCQ2. Have your or a close relative ever undergone genetic testing for possible hereditary cancer?		
No	8	40%
Yes	12	60%
GCQ3. If yes, please select who had the genetic testing		
Self	12	92.3%
Daughter	1	7.7%
GCQ4. If yes to GCQ2. and you are aware of the results, please select the appropriate one below		
Have a gene alteration for which the cancer risks are not currently well understood	3	23.1%
No gene alteration related to cancer was found	9	69.2%
Don't remember	1	7.7%

a. Absolute counts of "I" pronouns over total interview words
b. Absolute counts of "We" pronouns over total interview words
c. Absolute counts of "They" pronouns over total interview words
GCQ=genetic counseling questionnaire

5.3.1 Univariate Analysis Results

The distribution of language utterances for each participant is shown in *Figure 5A*. The univariate results are shown in *Table 5B*. On average, interviews with survivors lasted 66.60 minutes (SD=17.60) with an average word count of 4828.45 (SD=1762.86).

"I-talk" pronouns were used most frequently (mean=397.15 times, SD=179.60), followed by "they"-talk pronouns (mean=90.85 times; SD=46.98) and "we"-talk pronouns (mean=37.05 times; SD=28.71). Accordingly, the number of pronoun utterances over the total word count for the interview was the lowest (0.79%; SD=0.39%) for "we"-talk-, 1.99% (SD=0.84%) for "they"-talk, and 8.77% (SD=10.36%) for "I"-talk. Thus, for every 100 words a survivor said in the interview, the survivor used "I-talk", such as "I", "I'm", "I'll", "I'd", "I've", "me", "my", "myself", "mine" about 9 times.

On average, participants listed 1.85 (SD=2.16) relatives on the website and excluded 8.15 (SD=4.75) other eligible blood relatives. As described previously, the family size variable (i.e., the total of the number of relatives reported on the website plus those listed in the survey) was dichotomized with a cut-off at the median (8 relatives). As *table 5B* shows, eight participants reported a small family size, and twelve participants reported a relatively large sample size. Eleven survivors perceived that their families were high on closeness, and nine perceived that their families were not high on closeness.

Thirteen of the 20 survivors reported having had genetic counseling or testing; 9 had a negative result, that is, no gene alteration related to cancer and 3 found that they had a gene alteration for which the cancer risks were not currently well understood.

5.3.2 Bivariate Analysis Results

The cross correlations for the three linguistic markers are shown in *Table 5C*. "I"-talk was significantly and negatively associated with "We"-talk (r=–0.65;95 CI%: -0.84 to -0.27). "They"-talk was not significantly associated with either "I"-talk or "We"-talk.

Table 5C. Pearson correlations of linguistic indicators of communal coping (n=20)

Variable	With Variable	r	95% confidence intervals	
"I"-talk	**"We"- talk**	-0.65	-0.84	-0.27
"I"- talk	**"They"- talk**	0.09	-0.37	0.51
"We"- talk	**"They"- talk**	0.03	-0.41	0.47

My suppositions that linguistic markers would be associated positively with family size and closeness were not confirmed. As *Tables 5D* and *5E* shown below, mean proportions of "I", "we", and "they"-talks did not differ by family size (*Table 5D*) or reported family emotional closeness (*Table 5E*).

Table 5D. Occurrences of pronouns utterances between small and large families (n=20)

	Small family (n=8)		Large family (n=12)		t-test			
	%	SD	%	SD	DF	t-value	95% confidence interval	
"I"- talk	9.02%	1.11%	8.60%	0.99%	18	0.87	-0.006	0.014
"We"- talk	0.76%	0.39%	0.81%	0.41%	18	-0.25	-0.004	0.003
"They"- talk	1.64%	0.52%	2.22%	0.96%	18	-1.55	-0.014	0.002

Table 5E. Occurrences of pronouns utterances between high and not high family emotional closeness (n=20)

	Not high closeness (n=9)		High closeness (n=11)		t-test			
	%	SD	%	SD	DF	t-value	95% confidence interval	
"I"- talk	8.96%	0.99%	8.62%	1.09%	18	0.71	-0.007	0.013
"We"- talk	0.79%	0.34%	0.79%	0.44%	18	-0.05	-0.004	0.004
"They"- talk	2.28%	1.03%	1.76%	0.61%	18	1.4	-0.003	0.013

5.3.3 Selection of High and Low "We"-talk Prototypes

I further explored the think-aloud interviews for whether there use of we pronouns were associated with other relational and "we" thinking that was not pronouns but other utterances. To this end, I identified the highest quartile of "we"-talkers (n=5; "we"-talk > 1.11%), and the lowest quartile of we-talkers (n=5; "we"-talk< 0.52%). The 10 profiles are shown in *Table 5F*. For Factors 1(enumeration behaviors) and 2 (content engagement), I considered whether the thematic content of the survivors' reflections differed for those with relatively more or less "we"-talk.

Table 5F. High and low we-talk profiles

ID	"We"-talk	Who they enumerated	Which contact option they selected	How many letters they downloaded	Whether they've had genetic testing
High we-talkers					
1	1.19%	No enumeration	-	-	Yes, negative
3	1.44%	2 daughters	All self-contact	2	Yes, negative
5	1.45%	1 daughter	Refused contact	0	Yes, negative
6	1.11%	No enumeration	-	-	Yes, negative
12	1.68%	2 aunts, 1 sister	All study-team contact	0	No
Low we-talkers					
2	0.45%	Mother	Self-contact	1	Yes, negative
7	0.28%	no enumeration	-	-	Yes, negative
10	0.52%	1 niece	Refused contact	0	No
15	0.39%	Mother, 1 sister, 1 aunt	Study-team contact	0	No
17	0.35%	1 daughter	Self-contact	1	Yes, VUS*

*VUS (variant of uncertain/unknown significance) – participant has a gene alteration for which the cancer risks are not currently well understood

5.3.4 Enumeration behaviors (Factor 1): High vs. Low We-Talk

5.3.4.a Perceived emotional closeness to the relative

A common theme that survivors discussed when explaining why they did or did not enumerate a specific relative was how emotionally close they regarded the relative (*Table 5G*). Three of the five high we-talkers cited emotional closeness as a key reason for enumeration. For survivor #12, she acknowledged her emotional closeness differed between her listed relatives (i.e., two aunts and a sister) and the rest of the family. She noted that "*we're not a big emotional family. We try to resolve problems and find a solution*," but emphasized that with her aunts and sister, that "*those are three that I thought about that I'm really close to*," and that "*we are all in the same generation. So yes, we grew up together. We're close. My aunts are like my sisters.*" Another high we-talker (#3) listed her two daughters on the website because "*they're two of my closest relatives*."

Closeness was considered even when a survivor did not list a relative. One high we-talker (#6), who did not enumerate any relatives, specifically clarified that emotional closeness was not the reason and stated that "*I'm very close to all of them. I'm extremely close to all of them...the emotional closeness is not the reason for not doing communicate ovarian cancer risk.*"

Low we-talkers also mentioned closeness when discussing why they enumerated certain relatives, but less frequently. Only one survivor (#2) explicitly mentioned emotional closeness as a reason for enumerating her mother because "*we have very close relationships, and we don't hide things. My mom walked through cancer with me*." During her interview, she mentioned that she "*could have listed would have been my aunt, my mom's sister"* because "*They're* [mother and aunt] *just the people I'm closest to as far as what family I have left, other than my children*."

Table 5G. Quotes of high and low talkers regarding perceived emotional closeness to the relative

ID	Quotes
High we-talk	
3	So I feel very close. My oldest daughter … so I'm very comfortable with our relationship. And my youngest daughter … we've got a good ongoing relationship and both of their husbands are wonderful. We have a very good relationship with them. They're [*two daughters*] two of my closest relatives.
5	They're my life
6	I'm very close to all of them. I'm extremely close to all of them...the emotional closeness is not the reason for not doing communicate ovarian cancer risk.
12	Those are three that I thought about that I'm really close to We are all in the same generation. So yes, we grew up together. We're close. My aunts are like my sisters. We commune, fellowship together. And those are the three that I would normally hang out with
Low we-talk	
2	We have very close relationships, and we don't hide things. My mom walked through cancer with me. I could have listed would have been my aunt, my mom's sister. That probably would be, as far as my family would probably be the extend They're just the people I'm closest to as far as what family I have left, other than my children.

High and low we-talkers both noted a lack of emotional closeness as a reason for not enumerating certain relatives (*Table 5H*). For example, on survivor with high we-talk (#1) explained that she did not list any of her nieces and nephews because she did not "*have that close of a relationship with them.*" Similarly, one low we-talker (#10) noted that "*I'm not close enough to the rest*." I found one exception with a low we-talker (#15), despite lacking emotional closeness with her mother, sister and aunt (e.g., "*I have no relationship with my biological mother. I haven't spoken to her in over 15 years*."), listed them because she recognized their biological ties.

Table 5H. Quotes of high and low talkers regarding perceived lack of emotional closeness

ID	Quotes
High we-talk	
1	I just don't have that close of a relationship with them.
3	And so, I try to eliminate stress as much as I can, which sometimes involves keeping some people like my sister at a distance. I still love her. Still love her, don't get me wrong.
Low we-talk	
7	Because there were family members that I did not tell, only because it wasn't – I didn't feel like there was really a need to tell them because I don't have a close relationship with them.
10	I'm not close enough to the rest. I'm not close enough with any of my nephews. I see them. I get a hug from them. But, I don't feel close enough in talking with them. And, they would definitely shut down and wouldn't be interested in it.
15	I have no relationship with my biological mother. I haven't spoken to her in over 15 years. When I think about the people who helped me, who sort of needed to be empowered to help me as the person going through it, none of these three people were the right choices. They were the right choices from a biological perspective to think about the cancer risk
17	It was probably more emotional with the ones that were close. The one further away, it was sort of like a fact.

Most often, this lack of emotional closeness was attributed by survivors to having little regular contact with relatives, often due to greater geographic distance (*Table 5I*). Both high and low we-talkers mentioned that they saw certain relatives infrequently, which led to their decision not to enumerate them.

Table 5I. Quotes of high and low talkers regarding little contact

ID	Quotes
High we-talk	
1	As I mentioned before, I don't live in the same city with any of them, or any of my sisters. I text with a couple of them from time to time, but I pretty much see them in the summer and I'll maybe see some at some of the holidays. The fact that I don't get to see them that often I'm not real close with any of them and it's primarily just because we don't live in the same place and there are so many.
3	I feel since my little brother and sister both live in other states, we're not able to get together as much now as we did when my mom and dad were still living because we're all grandparents now. We all have our families. We all have demands. So, that's the main reason I didn't list my sister and my brother.
6	I wish there were more opportunities we could see each other, but as it stands now, it's maybe a few times a year. It's not an ideal situation for this kind of discussion.
Low we-talk	
10	The ones that I didn't list is maybe some of the older nieces that I see a lot, but they're younger than me. So, I don't feel very comfortable talking with them. I'm just not close enough to them because they live out of town.
17	I think the biggest thing is probably proximity…I'm all over. So, they weren't in my physical orbit.

5.3.4.b Relative's age and gender

Both high and low we-talkers considered their relatives' male gender or their age (whether young or old) when deciding whether to enumerate them or not. First, as shown in the profile *Table 5F*, neither high nor low we-talkers listed any male relatives during their initial website visit. Both groups generally recognized female relatives as the only ones for whom inherited risk for ovarian cancer was relevant (*Table 5J*), because "*it's a woman thing (high we-talker #3).*"

Interestingly, low we-talkers (#7 and 10) raised a point that even female relatives would not be considered for listing if they had undergone surgeries to remove their female reproductive organs. As survivor #7 stated, "*as an example, with my mother, she wouldn't apply because she had a hysterectomy when I was younger*."

Table 5J. Quotes of high and low talkers regarding female relatives

ID	Quotes
High we-talk	
1	That would be just an assumption I would have but I would think that typically when it comes to ovarian or breast cancer you're thinking in terms of your female family members.
3	I was mostly concerned because of my two daughters. When you're talking ovarian cancer, it's a woman thing.
12	Ovarian and the breast cancers usually pertain to women.
Low we-talk	
2	It was more just thinking about women and communicating with them. It's not that it's not relevant to men, I was just thinking about people who would be going through it themselves I guess in my mind, I really thought more about females than males in this situation.
7	Just because they're men. Yeah. That wouldn't even be a thought. Because ovarian, I'm thinking about female relatives only. As an example, with my mother, she wouldn't apply because she had a hysterectomy when I was younger. She wouldn't apply in terms of ovarian cancer because she no longer has her ovaries.
10	They [older sisters] all had their hysterectomy and everything.

Despite the website clearly stating that male relatives were eligible for genetic cancer risk communication and providing visual cues of family pictures with both female and male family members, both high and low we-talkers did not perceive the topic to be relevant for their male relatives (*Table 5K*). This came from two perspectives. First, most of them considered that the inherited risk due to ovarian cancer did not apply to male relatives. As a high-talker #1 noted, regarding her male relatives were not "*even registering on my radar that there would be a need for that.*" A low we-talker #7 expressed a similar opinion that "*just because they're men. Yeah. That wouldn't even be a thought*."

Second, survivors' lived experiences in ovarian cancer treatment made them feel that there is a sense of ovarian cancer being uniquely female, therefore inherited risk, coming from their ovarian cancer diagnosis, did not apply to male relatives. As explained by one low we-talker #15, "*that female experience was a big part of my post-surgical care experience that I focused on the experience women unfortunately have. And it made it very easy to exclude the potential for the men in my family to be at risk*."

Table 5K. Quotes of high and low talkers regarding male relatives

ID	
High we-talk	
1	I wasn't really thinking of males at all. That was something that came to mind for me. I will say this is a surprise to me in that I tended to think about contacting female relatives but not male relatives. I realize now that they can be carriers of the gene, but it was just something that didn't even – wasn't even registering on my radar that there would be a need for that, yeah. I will say this is a surprise to me in that I tended to think about contacting female relatives but not male relatives.
5	And I know men can get breast cancer but it's not as prevalent as it is in women. So, that's why I didn't list my two grandsons.
6	One [*son*] is not married and he has no children, so it wouldn't be applicable to him.
Low we-talk	
2	I was just more thinking Ñ obviously if they have wives or daughters it impacts them. But I just think in my thought process this morning it was more just thinking about women and communicating with them. It's not that it's not relevant to men, I was just thinking about people who would be going through it themselves.
7	I have a son. yeah. And then, immediate – well, I have a younger brother. I have an uncle. But nobody that would pertain to this study. Just because they're men. Yeah. That wouldn't even be a thought.
10	But, I didn't know in terms of genetic counseling for ovarian cancer. I didn't know it's the male's part of the family.
15	No males, immediately I took all males [inaudible] all men out of the conversation My uncles, I don't know how they fit into – I guess breast cancer if there's the tie between ovarian and breast, then breast cancer becomes a risk for them. So, I think some of my experiences that were, frankly, horrifying, really had me focusing on the experiences of women. What they'd go through, why it takes so long for them to get diagnosed, why they're in Stage 3 because they are sort of sent in circles, disregarded, that female experience was a big part of my post-surgical care experience that I focused on the experience women unfortunately have. And it made it very easy to exclude the potential for the men in my family to be at risk. Probably unfairly, I kind of blocked out the men in my life and their risk. I probably unfairly excluded the men from my list. Intellectually, yes, but I think it's taken me some time to intellectually get to, hey, ovarian, BRCA, don't forget that there a connections and connectivities here. But it took me some time.

Age was another factor that influenced survivors' decision on who to enumerate (*Table L*). High and low we-talk survivors universally did not list younger relatives under 25 years old as they did not meet the study inclusion criteria on the website. However, several survivors

believed that their young relatives would benefit from enumeration. Others noted that they did not list younger and eligible relatives because "*right now, health issues are not there for them yet*." Similarly, older age was a common reason for both high and low we-talkers to exclude some relatives from enumeration. Except for one high we-talker (#12), who believed that enumeration would be beneficial to older relatives because "*that's when health crises and stuff starts*."

Table 5L. Quotes of high and low talkers regarding age

ID	Quotes related to older age	Quotes related to younger age
High we-talk		
1	I actually would not contact him. He's 89 years old .	
3		I do have a granddaughter, which I think I mentioned on another page, but she's only nine years old. She's too young at this point.
5		The boys are so young. They're 10 and 7.
6		It says on here living relatives eligible to participate, must be 25 years or older. The only two that would conceivably benefit would be my two granddaughters, but they're far too young. The 16-year-old would be old enough to understand, but I don't know if this is an ideal time in her life for me to do this.
12	Because of their age. That's when health crises and stuff starts.	The younger ones, they're in their 30s, 35 and under. So, right now, health issues are not there for them yet
Low we-talk		
2		I will say what was most important to me is having three daughters, but you have listed here 25 years or older. And so, they're 14, 20, and 21. So, that was a little disappointing that they were not able to be put on here because of their age.
7	For my uncle who's in his 70s. Well, because – I mean, it's kind of like he's already older. So far – I mean he's never had any health issues. He's already 70-something. I don't really have to think about her just because she's also 76, and she's already been through it.	My daughter, based on what I clicked on the "Who should I include here?" So, for example, my daughter, she's under 25 based on the parameters laid out there. As far as where it says, "Living relatives eligible to participate must be 25," if that were not there, then I would have added my daughter.
10	Because they're in the range of 70 and 69, So, I don't know if – Especially one of them. Would she understand the process? She's the 69-year-old that probably wouldn't understand it a whole lot...she's not computer literate.	
17		And part of it is I'm looking at this screen. It says, "Living relatives eligible to participate." My granddaughters are not 25, which is why I didn't put them down.

5.3.4.c A sense of one-and-done

Survivors explicitly considered communication with some as being "done". They had communicated with relatives in the past and perceived that no further discussion was needed (*Table 5M*). This is particularly noted among high we-talkers. They remarked that they did not list certain relatives as they had already discussed relevant topics with them. For example, one high we-talker (#6) did not list her son because "*I've actually spoken to him about it. When I was diagnosed with cancer, I went through genetic testing and counseling for the BRCA gene and, fortunately, I tested negative… and at this point, I don't think there's a significant enough risk to warrant it unless further information comes down the pipeline.*" This sentiment was not expressed by low we-talkers.

In addition, some survivors also expressed a notion of health consciousness being an influence on their decisions about who to enumerate. That is, some survivors felt that their relatives were already well informed due to their education or professional background, making it unnecessary to list them. This line of thinking emerged in one low we-talker (#10), who thought out loud about her sister's medical background (i.e., nurse) in enumeration decisions, in a sense that it might make her sister interested in being listed.

Table 5M. Quotes of high and low talkers regarding a sense of one-and-done

ID	Quotes
High we-talk	
3	I just really feel like the others have been following me these 18 years, and maybe if there was a greater genetic disposition, I might include others. But since that's not the case, I just really thought my main concern is for my two daughters. (3142_3**13, Pos. 41)
5	My sister, as I stated earlier, also had the genetic testing. So, she has done with her daughters what I did with my daughter. And that is to share the results of the testing. We're all very good about going to the doctor and getting regular checkups and things like this. And I have shared just about everything that I can think to share with my daughter and my sister. So, I'm not sure what else I can do to let them know.
6	The other one, I've actually spoken to him about it. When I was diagnosed with cancer, I went through genetic testing and counseling for the BRCA gene and, fortunately, I tested negative, but I spoke with him at that time At this point, I don't think there's a significant enough risk to warrant it unless further information comes down the pipeline. If something changes later on or more information becomes available later on, that could change, but for right now, I don't think so. (1666_6**16, Pos. 43)
12	I have spoke my family is pretty close, and we pretty much have discussed it. Yes. My nieces and whatnot. We've talked about it and told them what to look out for, that kind of stuff.
Low we-talk	
2	I'm a 10-year survivor, so I'm not in the middle of this.
10	She is a nurse and she probably would've been very much interested in it

5.3.4.d. Relational appraisals of the benefits of enumeration

Some high and low we-talkers appraised their position in the family system when making decisions about who to enumerate (*Table 5N*). For example, high we-talkers #1,5,6, believed that enumeration of their nieces and nephews would be overstepping their sisters' role as the mother. These survivors expressed concern that enumerating her nieces and nephews might negatively impact her relationship with her sister. One high we-talker (#1) explained that she did not enumerate any of her nieces or nephews because she "*would probably let my sisters make that decision, if they wanted to then share it with their children. That would be my first thought is my obligation to my own immediate family and not so much beyond that. It would feel to me like overstepping if I were to just go ahead and list them all*." And she later added "*I really would not want to circumvent that relationship with their mothers, my sisters. It's not really a matter of – even if I was close to them*."

Also, some high and low we-talkers shared a generational perspective, believing that their younger relatives were not ready for such communication, thus they did not include them in the enumeration. As one high we-talker #12 explained, "*the next generation down would not be interested. Those are the ladies that are still left in the family that are in the age group with illnesses and health issues and stuff like that. The younger generation have not reached that*

level yet." Similarly, low we-talker #10 noted that "*the other nieces, like I said, are fairly young in their 20s and living their own life. I don't know they're gonna be open to hear anything.*"

In addition, family system thinking also appeared when discussing the privacy of their relatives and the need for permission before listing them. Survivor #12 stated that "*I would have to think about it because I've got a big family. And then I have to think about addresses and phone numbers and like I said, invading their privacy.*" Likewise, low we-talker #3 did not enumeration as she "*would want to ask permission before I did that*."

Appraisals of relationships within the family system tended to occur at the dyadic level. Survivors, regardless of we-talk level, tended to consider each of their relatives' specific situation before deciding whether to list them. This is showcased in one low we-talker (#10). She only listed one of her nieces during the website visit because she "*thought of the family members that I think was actually going through something similar in that she was having some thyroid issues. So, her* [the niece's] *name came up right away as a person that I thought would really benefit from doing that*. " When asked about another niece and her brother, she clearly appraised their specific situations. She did not list another niece because "*she's got a newborn and working*." For her brother, she noted that "*he has some issues going on, and I just don't see him taking on anymore. Anything else that don't pertain to him.*" High we-talkers also considered specific relationships when deciding who to enumerate. For example, one survivor (#5) did not list one of her sisters because "*her husband just passed away...Right now, with my sister, she's grieving the loss of her husband. So, to talk to her about cancer would not be the greatest thing*."

Table 5N. Quotes of high and low talkers regarding relational appraisals on enumeration

ID	
High we-talk	
1	I don't know that I would do it for my nieces or nephews. I would probably let my sisters make that decision, if they wanted to then share it with their children. That would be my first thought is my obligation to my own immediate family and not so much beyond that. It would feel to me like overstepping if I were to just go ahead and list them all. I really would not want to circumvent that relationship with their mothers, my sisters. It's not really a matter of – even if I was close to them But I wouldn't overstep that relationship with their children. I wouldn't jump it because, again, it can be sensitive. I would rather let them discuss it or approach it to their own kids.
5	Again, as I said, I'd have to talk to my sister about it and see what her thoughts are. And I have her – now, her husband just passed away. ..Right now, with my sister, she's grieving the loss of her husband. So, to talk to her about cancer would not be the greatest thing.
6	I would speak with my son before I spoke with my granddaughter, because I think that's the proper thing to do, since he's her parent.
12	The next generation down would not be interested. Those are the ladies that are still left in the family that are in the age group with illnesses and health issues and stuff like that. The younger generation have not reached that level yet. So, there's no need to even inform them because like I said, they will probably do like I did, they would drop it all in the garbage and be like, Oh, I don't have to worry about it. I don't have any answers I would have to think about it because I've got a lot a big family. And then I have to think about addresses and phone numbers and like I said, invading their privacy. They're pretty private with their personal. And we try not to, if it's nothing wrong, we don't. if they can handle it, they handle it. If there's nothing wrong, we don't get involved. If there's something we need to know, they let us know. So, I would not invade their privacy for one, to just say, Hey, you need to change.
Low we-talk	

2	I wanted to ask you the question about if you were if the website was going to reach out to them, I would want to ask permission before I did that.
10	The other nieces, like I said, are fairly young in their 20s and living their own life. I don't know they're gonna be open to hear anything that – I think it's important for them to hear, but I just don't think their mind gonna be set on it. They've got other stuff going on in their lives. Because I thought of the family members that I think was actually going through something similar in that she was having some thyroid issues. So, her name came up right away as a person that I thought would really benefit from doing that. The topic really did come up maybe a couple of weeks ago when she was talking about a new job that she was getting. That she needed to go and have some testing done, and she didn't wanna be taking off a lot of time to do that. So, she was having some issues with thyroid. So, that came up. 'Cause I did remind her that I had a diagnosis of ovarian cancer. That maybe she needed to keep that in mind and not put off appointments. That she needed to get them as soon as she can. So, she was the one person I thought would benefit from it. She's got a newborn and working. He has some issues going on, and I just don't see him taking on any more. Anything else that don't pertain to him.

5.3.5 Content Engagement (Factor 2): High vs. Low we-talk

5.3.5.a Layered contact was preferred

As observed in the main trial and shown in profile *Table 5F*, after reviewing the content on different contact options, survivors most frequently chose to contact relatives themselves. I also found that, as in the main trial, regardless of we-talk levels, survivors who listed more than one relative chose the same contact approach for all the relatives they have listed. However, when they thought out aloud, it appeared that the decision process about how to contact relatives was layered (*Table O*).

As described earlier, survivors were presented with discrete options for contacting relatives. For example, if a survivor chose "*self-contact*", she could not also choose "*study team contact*". She could, however, choose different contact approaches for different relatives.

Regardless of we-talk levels, survivors believed that they should be the first one to initiate contact with the relatives they enumerated. Most felt that personal contact initiated by themselves was a given. One high we-talker (#5) explained, "*the best option is the close, personal connection with a relative*." Another high we-talker (#3) selected "self-contact" because "*I want to make sure that they understand what I am going through.*" Similarly, a low we-talker (#7) noted that "*because the information is more factual. It's more detailed. You're getting it from the source*." Another, low we-talker (#7) emphasized that "*the survivor, of course, should be the one. Or the patient should be the one to pass on that news, share that news*."

However, viewing "*self-contact*" as the first and default choice does not mean that survivors would not have been open to other contact options. High we-talker #3 noted that for distant relatives who she did not enumerate, she would not have chosen "self-contact". As she explained, "*and for a distant relative, maybe not so much, and they just really wouldn't want to have anything to do with it if they were like a distant relative.*" Similarly, low we-talker #7 also showed openness to other contact choices, as she stated "*I'll just use my mother as an*

example. Knowing how she is, emotional and all that, the health professional contact might be – someone might feel more comfortable with it, so they won't have to have that conversation or deal with that overwhelming, stressful situation of sharing the news."

Choosing "*study team contact' was less common than self-contact*. Survivors shared a number of motives for choosing study team contact. One high we-talker (#12) preferred to preserve her relatives' autonomy in deciding how to get risk information. As she stated, she wanted to let her relatives "*make their own decisions about whether they're going to participate. I don't wanna be the influence. I want them to be something they wanna do on their own*."

Other survivors talked about relatives who they were not emotionally close to or those they had not enumerated. For example, low we-talker #15 listed her mother, sister and aunt, who she was not emotionally close to, due to the recognition of inherited cancer genetic risk. In her case, she preferred study team contact because "*choosing the study team to reach out to them, for me, was putting a degree of separation between me and those who would not be part of, who could not be a part of, who I would not allow to be a part of, taking care of me*." A high we-talker (#3) further explained, "*with a person that I'm not as close to, it might would be okay for the study team to contact that person if I wasn't really close or confident that they wanted this information*."

Of the 10 "we"-talk prototypes, only one low we-talker chose to "*delay contact*" for her niece. She considered the timing of the communication, explaining that it might be an inconvenient topic for her niece at that moment. As she explained "*because I think she's going through a couple things...and, I just think until she can get settled and can concentrate a little bit more, I think she would be open to it. I just don't think right now will be good*."

Table 5O. Quotes of high and low talkers regarding layered contacts

ID	Self-contact	Study team contact
High we-talk		
1	I have good relationships with them that I would think they would want to hear it from me before they would want somebody from outside as your first contact about it. I think that they would think it was strange if they just got this random thing from somewhere that I hadn't said anything to them about. For me, personal contact and letting them know before I sent them anything or did it concurrently.	
3	I want to make sure that they understand what I am going through. Because of the relationship we have, a very close relationship. I would want to know if my daughter was having issues, and I am 99.9% sure she would let me know. And I think they feel the same about me. They want to know about what goes on.	And for a distant relative, maybe not so much, and they just really wouldn't want to have anything to do with it if they were like a distant relative And with a person that I'm not as close to, it might would be okay for the study team to contact that person if I wasn't really close or confident that they wanted this information.
5	The best option is the close, personal connection with a relative.	
12		To contact family members without me being a part of it at all. And let them decide and let them make their own discretion about whether they want to do it or not.

		Let them make their own decisions about whether they're going to participate. I don't wanna be the influence. I want them to be something they wanna do on their own.
Low we-talk		
2	I definitely reached out to people who had been in my shoes, or family. I think most people are going to wanna reach out to somebody they know.	
7	I would say I would never choose to have the health professional contact family members. Something like this, I mean, the survivor, of course, should be the one. Or the patient should be the one to pass on that news, share that news. But in the end, I mean, it's best to just contact them. Have the survivor contact, just to get it over with. Yeah. So, for me, again, I'm just a straight shooter. Always have been. So, if I'm in a situation, and I need to communicate and let people know, I'm going to tell them directly. Because the information is more factual. It's more detailed. You're getting it from the source.	I'll just use my mother as an example. Knowing how she is, emotional and all that, the health professional contact might be – someone might feel more comfortable with it, so they won't have to have that conversation or deal with that overwhelming, stressful situation of sharing the news
15		For my biological group, I probably would have preferred, I would have chosen to let the study team contact them. We're contacting you on behalf of your family member. This is what we want to let you know, and kind of choose to do that from there. For my biological, because I knew that there would be no impact, I was not going to allow any impact. And I would've wanted – I did, I selected, I chose a degree of separation. And so, choosing the study team to reach out to them, for me, was putting a degree of separation between me and those who would not be part of, who could not be a part of, who I would not allow to be a part of, taking care of me. Again, culturally, also not something that is easy for me to say. Carving them out eliminated the burden of having the conversation, eliminated the burden of saying you're not helping me. Please step away.
17	I have a very close relationship with my daughter, and my husband was with also. And it got it over with. Let's put it that way.	

5.3.5.b Exploring "we"-responsibility

My suppositions about how *TM* might lead to risk communication was that survivors would acquire a sense of "we"-responsibility if they viewed inherited risk as a we-problem. There was evidence of this thinking among both high and low we-talkers (*Table 5P*). One high we-talker (#3) noted that emphasizing the "*we*" would "*convey hope and family connection.*" Similarly, a low we-talker (#17) agreed that hereditary cancer prevention "*is a shared goal. It affects the family. It's not just the individual*."

However, this aspect of communication as a means of preserving family relationships was more granularly expressed among high we-talkers. For example, one high we-talker (#3) explained her responsibility as member in the family that "*It's like we're in this together. We're family. We share a lot of wonderful stuff, our wonderful stories, our wonderful memories, but there are also responsibilities we have as family members. It's best if you can come to grips with*

this possibility. Yeah. It may occur in me. I really need to know." Similarly, another high we-talker (#12) implied, "*we're gonna talk about it as a family group. This is our conversation*."

Some low we-talkers struggled with the idea that inherited risk communication is relationship-oriented. For example, one low we-talk survivor (#10) expressed uncertainty about the idea that sharing communication about cancer as a shared health threat would impact their relationship with other family members. She noted "*I don't know if my relationship would be hurt one way or the other if they didn't take it."* Similarly, another low we-talker (#11) remarked that "*I don't feel like I'm protecting, I feel like I'm continuing a relationship that is there by communication.*"

Table 5P. Quotes of high and low talkers regarding exploring "we"-responsibility

ID	Quotes
High we-talkers	
3	I would keep the Weness part in this. And you convey hope and family connection. Yeah, it's really good. It's like we're in this together. We're family. We share a lot of wonderful stuff, our wonderful stories, our wonderful memories, but there are also responsibilities we have as family members. It's best if you can come to grips with this possibility. Yeah. It may occur in me. I really need to know.
5	My daughter and I have done a lot of this already...I'm very familiar. And I like to emphasize the we-ness behind your motive to acknowledge that the shared risk has the potential to interfere with their life… And it wouldn't hurt to be proactive
12	But just to Ñ I'm gonna discuss this with you we're gonna talk about it as a family group. This is our conversation. It's nothing nobody wants to talk about. Now if it's if you're thrown into that situation and it's actually happening to you, it's just a hard conversation to have.
Low we-talkers	
2	I'm not really sure how to read that. I don't feel like I'm protecting, I feel like I'm continuing a relationship that is there by communication.
7	Because it's a we thing. It's not just – yeah. So, I do like that. That's good. We-ness.
10	I don't know if my relationship would be hurt one way or the other if they didn't take it. But, I don't know that that one would make a difference one way or the other. We've got a pretty good relationship
17	I like the – it is a shared goal. It affects the family. It's not just the individual.

5.3.5.c Genetic counseling as part of "we"-solution

Website engagement on the genetic counseling section was mapped as part of the "we"-solution. High and low we-talk survivors were similar in expressing hesitancy towards undertaking genetic counseling (*Table 5Q*). One high we-talker (#14) remarked, "*It's like how much can my body stand to hear? Do I want to subject myself to some knowledge that, yeah, there is a good likelihood I may get cancer, and this would just bring it all to the front of the mind. I'd rather keep it back here in the back of my mind where I don't have to think about it too much*." Similarly, one low we-talker (#9) stated that she is taking "*a little bit of a coward's way out of not doing genetic testing. But also kind of just acknowledging my flaws. I am a person who if I knew I would constantly be worrying about it*."

Table 5Q. Quotes of high and low talkers regarding genetic counseling hesitancy

ID	Quotes
High we-talk	
12	I'm gonna say no because I don't know anything about it. I don't know enough about it. It's like how much can my body stand to hear? Do I want to subject myself to some knowledge that, yeah, there is a good likelihood I may get cancer, and this would just bring it all to the front of the mind. I'd rather keep it back here in the back of my mind where I don't have to think about it too much.
Low we-talk	
10	I'm not pursuing just because I'm surrounded by this amazing group of medical professionals who are very aggressive about staying tied to my health. A little bit of a coward's way out. You can say no. You're very kind. But that's what's stopped me, really just knowing who I am.

2	Just because it is time, its people have a lot on their plates and raising kids and family. I do see doctors regularly and I'm Ñ I stay healthy. So I guess I'd want to see, yeah, the benefit of that hour.

However, high and low we-talkers universally acknowledged the significance of genetic counseling for younger generations through a relational lens (*Table 5R*). In their generational thinking, genetic counseling was seen as important for younger generations in terms of cancer prevention. For example, one high we-talker (#14) appraised that "*it seems like it would be geared toward the younger generation in my family, and next generation. So, they can kind of watch out and be prepared not be prepared but do testing and be more knowledgeable about the cancer and the gene pool with the cancer in our family.*" Similarly, a low we-talker (#11) emphasized that "*I had a hysterectomy so, that helped clear some of the issue with myself. So, the gene testing was more really for my daughters*."

Both groups also valued the insights of genetic counseling for informing future family planning decisions. One high we-talker noted that "*my older daughter probably would want to schedule a counseling appointment because one of the things that helps prevent you from getting ovarian is having children and breastfeeding*." Similarly, a low we-talker (#17) explained, "*just for their family history going forward. Thinking ahead in the future. So, if they decide to get married and have a family of their own, they'll be well-equipped with that knowledge of any issues genetically*."

Table 5R. Quotes of high and low talkers regarding generational thinking of genetic counseling

ID	Quotes
High we-talk	
3	My older daughter probably would want to schedule a counseling appointment because one of the things that helps prevent you from getting ovarian is having children and breastfeeding.
5	And the same thing with my sister. But it's been a long time since both she and I have had cancer. And they're older now. They may need to have an update on what's going on in their bodies. To be informed. I know that her doctor is very aware, especially the gynecologist. He's very aware of my history and her history. I just want to make sure that it doesn't hurt to have another opinion.
6	Obviously, to find out, in time, if she – hypothetically, if she were to contract something, some form of cancer, that she could get treatment immediately. Better her chances of dealing with the disease.
12	it seems like it would be geared toward the younger generation in my family, and next generation. So, they can kind of watch out and be prepared Ñ not be prepared but do testing and be more knowledgeable about the cancer and the gene pool with the cancer in our family. I think it would be excellent if they [younger generations] would take part in and want to be want to know and get that knowledge before something happens pray tell that nothing does.
Low we-talk	
2	I had a hysterectomy so, that helped clear some of the issue with myself. So, the gene testing was more really for my daughters. I think it's very important for my children to know if that was a gene, something that we needed to as they got older, be looking for.
10	I would like to know whether I have the gene that could possibly lead to breast cancer. So, yes. I would be interested in it. Yes, I do. I think if there is a history of ovarian cancer or breast cancer, if there's a chance that it's in their gene, I think they would wanna know. So, I think the plus side of it is learning about it. And, it's something they would wanna know, especially since they had one family member that has it. I see the benefits and I think it's always a benefit in knowing if you had a family member who had any type of cancer to know if it's a possibility that's in your family. If you had that opportunity to do that.
17	I'd want to know more about my particular cancer so that I could research or get help researching different treatment plans. Yeah. And whether or not my daughter or granddaughters are at risk. Yeah. I would definitely do that. For my son and my daughter, yes. I would have them schedule genetic counseling. Yes. Yes. Just to rule out any – yeah, just to know. Just for their family history going forward. Thinking ahead in the future. So, if they decide to get married and have a family of their own, they'll be well-equipped with that knowledge of any issues genetically.

5.4 DISCUSSION

5.4.1 Role of "I", "We", "They"-Talk to Indicate Transformation of Motivation

Using think-aloud methods I sought to gain insights into the *TM* process. I posited that utterances of "we"-pronouns could be implicit markers of *TM* and communal coping. My rationale was that using "we"-talk while engaging with the website content could lend insight into the cognitive and emotional process that might underpin survivors experiencing a transformation of motivation that in turn could motive them to communicate with family members about inherited cancer risk.

Overall, survivors used "we"-talk infrequently as compared to "I"- and "they"-talk. Less than 1% of total utterances was "we"-talk; 9% was "I"-talk and 2% was "they" talk. The low frequency of "we"-talk is consistent with previous studies[147,152]. For example, in a couple-focused smoking cessation intervention study, the baseline "we"- and "I"-talk utterances of 40 participants were 1.5% (SD=1.8%) and 6.0% (SD=2.4%) respectively. After the intervention, the corresponding rates were just 1.1% (SD=1.0%) for "we"-talk and 6.0% for "I"-talk (SD=1.7%).[147] Another study of couple-focused interventions for problematic alcohol use, alcohol users baseline "we"-talk was 0.77% and "I"-talk was 9.8%.[152] Benchmarks for what level of "we"-talk would indicate the *TM* required to prompt communal coping, as opposed to other coping mechanisms that I described in Chapter 1 (e.g., "we"-problem, "your"-responsibility) have yet to be established.

Additionally, "I"-talk and "we"-talk were negatively correlated such that the greater the use of "I"-talk the lower the use of "we"-talk. "I", "we", and "they"-talk was not associated with survivors' subjective ratings of their family's emotional closeness measured by the adapted Inclusion of Other in the Self scale, or family size. These findings are also consistent with existing evidence.[149,241] Helgeson and colleagues also found no association between a diabetes-specific Inclusion of Other in the Self scale and patients' or partners' "we"-talk among 70 couples in a type-2 diabetes management study.[241] Additionally, Zajdel et al found that caregivers' network size, defined by the number of individuals identified in their network circles (inner close circle, middle close circle, and outer least close circle), was not associated with "we"-talk or "I"-talk.[149]

It is worth noting that more than half of these 20 ovarian cancer survivors rated 6 or 7 in their emotional closeness with their family, with the meaning rating of 5.1. Thus, the lack of variation in this variable likely reduced the already low power, due to the small sample size, to detect these associations. As revealed in the interviews, participants defined "family" differently,

with some only counting their core family members or those who they felt the closest, whereas others included distant relatives as well.

This is the first study to count “they”-talk. [234] In the context of family genetic risk communication (FGRC), the alternative “they” language becomes conceptually interesting. While my study did not report any significant findings, future research is warranted to build evidence on how probands view the rest of family as a unified identity. Taken together, these descriptive analyses and tests of conceptual associations between “we”-talk and family structure suggest that counts of “we”-talk may not be an optimal measure of *TM*.

5.4.2 Actions and Content Engagement Factors Among High and Low We-Talkers

I explored whether the level of we pronouns survivors used was associated with other relational talk. In comparing the highest quartile of "we"-talkers and the lowest quartile of we-talkers, I found that high we-talkers had more positive expressions of emotional closeness when thinking about which relatives to communicate with about inherited cancer risk. The highest we-talkers emphasized emotional closeness as the key for their decisions on whom to enumerate. Even when high we-talkers did not enumerate certain relatives, they clarified that lack of emotional closeness was not the reason. The lowest we-talkers mentioned emotional closeness infrequently. However, both groups viewed a lack of emotional closeness, often due to infrequent contact, as a key factor influencing their decision not to enumerate certain relatives.

Consistent with Relational Autonomy theory, when survivors were engaging in enumeration behaviors, they tended to appraise who to enumerate for inherited risk conversations at dyadic level, not a family system level. Regardless of level of we-talk, participants considered each relative's specific situation before deciding whether to list them. For example, survivors were concerned about communicating with older relatives, believing they might not comprehend the information.

Regardless of we-talk level, survivors frequently did not consider male relatives for enumeration, considering ovarian cancer as relevant only to women. Survivors lacked awareness of HBOC also being associated with breast, pancreatic, and prostate cancers.[3,11]

As proposed by Family Systems Perspective,[126] FGRC has a social support function that builds and strengthens family ties. The highest and lowest we-talkers applied family system thinking when they considered their role in and responsibility for communicating risk. High and low we-talkers considered their ties with close relatives in deciding about enumerating. For example, survivors were reluctant to enumerate nieces and nephews feeling that they might be overstepping and that it would be more appropriate for their siblings (i.e., the parents of their nieces and nephews) to play that role. This relational thinking did not differ by “we”-talk level.

Consistent with previous findings, both high and low we-talkers considered communication about FGRC to be a one-time event.[52,63,78,80] If the survivor had discussed it previously with a family member or members, they were reluctant to bring it up again, even if there was a clear opportunity to reopen the discussion. This suggests that survivors may experience a strong inclination toward *TM* and communal coping that is dispensed once they feel information has been shared. In this respect, the survivor shifts from this perception of shared responsibility ("we"-responsibility) to perceiving relatives are informed and it is now the relative's responsibility ("your"-responsibility) to take necessary action. This seemed to be especially true when family members had professional or educational experiences that the survivor regarded as making them highly capable to take recommended actions.

The majority of survivors, regardless of level of "we"-talk, selected "*self-contact*". Survivors regarded self-contact as a no brainer and frequently aligned it with their emotional closeness. Most regarded the provided standard letters as too impersonal and jargony and rejected the idea that this type of communication should come directly from an outside source. On the face of it, greater endorsement of self-contact would be an indication of strong *TM* and communal coping. However, the survivors seemed to come quickly to this decision without weighing any pros and cons, as the website encouraged them to do. Thus, this choice would be unlikely to discriminate those who experience *TM* from those who do not.

While the construct of *TM* that would motivate FGRC is thought to arise from concerns about preserving relationships survivors, particularly those with the lowest "we" counts, had trouble with the concept that risk communication would preserve their relationships. However, clearly the dyadic thinking and related appraisals the survivors shared largely were about protecting their relatives from information that might come at a bad time. Thus, not sharing information was as much a part of *TM* as the decision to share information.

Lastly, both high and low we-talk survivors prioritized genetic counseling as less important for themselves, believing it would be most beneficial for their children and other younger relatives. This adds another level of relational thinking. Even when survivors choose to enumerate specific relatives on the website (Factor 1), they may or may not regard genetic counseling as important for themselves. It reflects a complex interplay involving survivors' sense of "we"- responsibility and their considerations of generational cancer risks within the context of FGRC. Taken together, relational thinking for the most part was equally present among high and low we-talkers. Thus, the notion that we-talk can be an indication of *TM* is flawed. *TM* appears to be a more complex construct that is not captured by counts of "we" talk.

5.4.3 Strengths and Limitations

There were several limitations of the study. First, the small sample size of 20 ovarian cancer survivors that did not enable me to proceed with a confirmatory factor analysis for the two factors found in Aim 2. Also, the study participants in this study were volunteers who received an incentive ($150). As the comparisons of Aim 2 and Aim 3 engagement data suggested, survivors in the think-aloud study had a higher level of website engagement in this experimental setting. On the other hand, this could also be a strength because participants were more thoughtful in verbalizing their views.

Second, I was not able to report the sample characteristics of the study. Demographic variables, such as date of birth, date of cancer diagnosis, cancer stage, were not included in this present study, therefore I was not able to examine if Aim 3 sample was differ from Aim 2 sample. Because I utilized Aim 3 participants' think-aloud interview data to make meanings of factors identified in Aim 2, this limitation affects the generalizability of my findings. Moreover, the Georgia Cancer Registry does allow the release of participants private data to the study team.

There also could be subconscious social desirability response bias generated when interviewers probed participants for responses. However, at the setup of the study, all interviewers went through a 3-day training process where we did a rotation of interview observations before independently carrying out interviews.

Interpretations of two factors suggested that *TM* is likely a complex and nuanced construct far beyond that of "we"-talk. In Chapter 6, I will draw implications based on results from each Aim to inform suggestions for future directions in research.

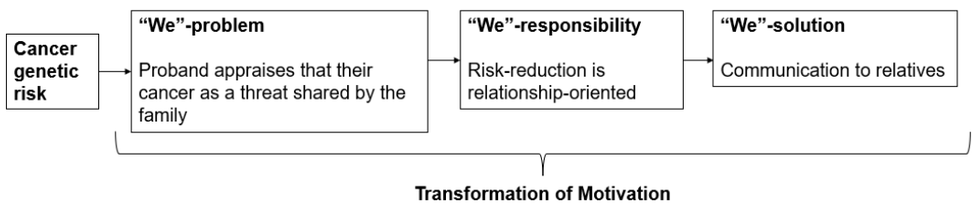
CHAPTER 6. DISCUSSION AND CONCLUSION

6.1 SUMMARY OF THREE INTERCONNECTED STUDIES

In this last Chapter, I will first present the summary of three interconnected studies. Then, I will discuss two major areas for future research on transformation of motivation (*TM*) as an intervenable construct: "we"-talk measurements, and Communication in enumeration and cognitive aspects.

My *overarching aim* is to advance our understanding of how to measure the construct of TM. The research I have conducted is preliminary but has the potential to increase understanding about whether activation of relational thinking could prompt survivors' *TM* to communicate with relatives about their inherited cancer risk. As defined in Chapter 1, *TM* is a psychological shift mechanism in motivation, transitioning from consideration of immediate self-interest when taking actions to broader consideration of another and/or the collective's interests. As a reminder, the conceptual model of *TM* and communal coping, presented in Chapter 1, is shown in *Figure 6A*.

Figure 6A. Conceptual model of integrated transformation of motivation and communal coping



In Aim 1, I undertook a systematic review to explore whether current family risk communication interventions were theoretically informed and at which levels of influence (i.e., individual, relational, and family-system). For Aim 2 and Aim 3 studies I analyzed data from an ongoing website-based study and think-aloud structured interviews to bring conceptual clarity to the latent *TM* construct. I systematically analyzed whether survivors of ovarian cancer survivors' natural patterns of use of a study website might serve as indicators of "we-ness" thought to underpin the *TM*. Other studies have used traditional and largely unvalidated approaches such as survey questions (e.g., *"when you think about problems related to your heart condition, to what extent do you view those as our problem' (shared by you and your spouse equally) or mainly your own problem?"* [153]) to assess we-ness. My mixed method approach aimed to provide a deepened understanding of the latent construct of *TM* in the context of family genetic risk communication (FGRC).

I found two factors of survivors' website use, one action oriented and one content engagement. As reviewed in Chapter 1, *TM* was conceptualized to consist of two domains: a cognitive interpretation of inherited cancer risk presenting an existential threat to the "we"; and an emotional interpretation that such a threat has significant meanings to the self, the relative, or the relationship, which inspires a drive to protect the "we." The behavioral action factor, identified in Aim 2, could be another domain to indicate *TM*. As I proposed, "we"-solution is communication relatives in the conceptualization, therefore, this new identified action factor could imply "we"-solution.

In Aim 3, I found that "we"-talk pronoun counts, often used as an implicit marker of TM, was not associated with family closeness or family size. Yet, qualitative interpretations of interviews revealed a lot of relational talk by survivors that also was not associated with among of we-talk. Taken together, these results suggest that "we"-talk may not be a reliable marker and that there is context-specific relational thinking missing from the pronouns analysis. Emotional closeness was offered as a universal reason for enumerating relatives for FGRC. However, the measure I used for emotional closeness did not perform well, being highly skewed. Other measures of family closeness might be used or developed to give insights into TM. Assertions and hypotheses of Aim 2 and Aim 3 are summarized in *Table 6A*.

Table 6A. Assertions and hypotheses of Aim 2 and Aim 3

Aim 2 assertions	
Assertion 1	The greater the extent to which survivors' enumerate, choose contact options and download letters for relatives indicates the degree to which survivors interpret inherited cancer risk as a "we"- problem, with collective responsibility, and engaging in family communication as a solution.
Assertion 2	More use of the Importance of Family Communication would prompt survivors to see themselves as part of a dyad/family collective, and as such, could prompt TM to protect their relatives by sharing information.
Assertion 3	Survivors who engage more with sensitive conversation framework tipsheet will be exhibiting TM viewing inherited cancer risk as relationship-oriented that is indicative of perceiving "we-responsibility".
Assertion 4	Survivors' level of engagement with the genetic counseling section could indicate experiencing a TM .
Aim 3 hypotheses	
Hypothesis 1	Ovarian cancer survivors who have higher use of plural utterances in "we"-talk and "they"-talk, compared to "I"-talk, would be more likely to have experienced TM.
Hypothesis 2	ovarian cancer survivors who report higher visual emotional closeness would be more likely to have experienced TM.

6.1.1 Strengths and Limitations

As I have discussed specific strengths and limitations related to each Aim in previous chapters, here, I briefly note strengths and limitations of the work on the whole. First, my research questions have focused on the context of ovarian cancer. This sample limits the applicability of findings to other medically actionable hereditary conditions that affect both males and females. However, the ovarian cancer context and related national guidelines enabled me to pose questions that would not have been possible in hereditary colon cancer for example. All first- or second-degree relatives of those diagnosed with ovarian cancer are recommended to seek genetic counseling, regardless of whether the family member diagnosed has undergone

genetic testing and found to carry a pathogenic variant.[18,19,242] Since privacy policies and laws mean that survivors must be tested and then are the conveyers of risk information based on their results.

Additionally, my research involved only the behaviors and perspectives of ovarian cancer survivors. To improve generalizability of a transformation of motivation as inherently relational process additional insights how the collective family navigates arrives at "we"-thinking might be gained from consider the perspectives of relatives. In addition, while *TM* is conceptualized as a psychological shift, the cross-sectional designs of the Aims 2 and 3 studies did not allow for a comparison between baseline *TM* and after website visit *TM*. For example, it is plausible that participants who exhibited high *TM* in the linguistic makers of "we-ness"-thinking may have already engaged in *TM* before website exposures.

6.2 DIRECTIONS FOR FUTURE RESEARCH

I conclude that using "we"-talk counts to indicate relational thinking is likely not an adequate indicator of the *TM* and communal coping. Further qualitative analyses suggest that survivors use a good deal of relational talk about emotional closeness. Other relational aspects in their decisions about sharing risk information, such as familiarity with the life circumstances and personalities of their relatives, were not reflected in "we"-talk. Future qualitative studies are needed to inform the development of relational thinking measures that might be associated with motivation to cope communally by communicating about inherited risk. , I will focus on two major areas to recommend future research directions and enhance the understanding of *TM* as an intervenable construct. This will be essential for developing and validating a *TM* scale.

6.2.1 "We"-talk Measurement and Beyond

Future studies should focus on qualitative explorations on linguistic makers of "I", "we", and "they". First, it is important to note that interdependence of family relationships cannot be fully captured from an proband-centered field of view.[243] There is a need for obtaining a broader perspective to understand at-risk relatives' *TM* shift, and its reciprocal influences on probands' *TM* within specific dyadic relationships, as well as its moderating effects on relationships with other family members. Future research is warranted to include perspectives of at-risk relatives.

For example, there could be studies involving interviews and observations of dyads or families together. The interview process becomes much more of a relational process, which enables in-depth interactions among members within one family, leading to co-constructions of *TM* meaning.[244,245] In addition, shared understanding within a dyad can be more effectively examined in a relational context, as it involves responses that are context-specific and require

background knowledge about family functioning and norms. To this end, the dyadic interviews offer a more integrated understanding of the family dynamics, including insights into relatives who are not directly involved in the interviews

In the design of these studies, “we”-talk and other linguistic marker counts would be counted from probands, relatives, and as a whole. Examining these linguistic markers qualitatively and quantitatively would further illustrate how the concept of "we" is used in conversations and understood within the family context. In addition, future research should also explore other potential markers derived from interviews to enhance understanding of *TM*. What I identified in the interviews is “they”-talk, which was more frequently used compared to “we”-talk. "They-talk" could provide insights into how probands refer to others within their networks, potentially revealing additional layers of relational appraisals in *TM* shifts. However, similar to "we"-talk, the measure of "they"-talk is context-dependent and may require additional measures such as qualitative interpretations or self-reports of *TM* and communal coping to strengthen its validity and reliability.

Secondly, analyses for Aims 2 and 3 were cross-sectional. I posited that *TM* is a process that occurs over time. Thus, I am studying survivors behaviors and thoughts at a moment in time and was only able to understand the process in retrospect. For analyses in Aim 2, these survivors were diagnosed between January 2005 and December 2017. It is plausible that survivors who chose not to enumerate had already transformed from "I" to "we" at the time of diagnosis or after undergoing genetic testing. I conceptualized ovarian cancer survivors’ website (YFC) use to infer “we-ness”. However, there was no baseline comparison and so it cannot be determined if survivors have initially viewed inherited cancer risk as “we”-problem, responsibility, and/or solution before visiting the website.

In addition, as discussed in Chapter 1, there is a spectrum of communication timing for FGRC. Studies have shown that most of probands communicated to at-least one relative immediately after receiving genetic testing results.[61] However, some waited longer until they had all relevant information about their genetic testing result, and others felt no urgency to share the information with at-risk relatives. Thus, to better understand *TM* as an intervenable construct, it is important to examine the *TM* process among participants near the time of diagnosis or at the beginning of their treatment. This would allow us to assess their baseline perspectives and measure any shifts after viewing *TM*-based intervention content, such as YFC.

Another recommendation is to incorporate kin-keeping scales to further understand *TM*.[246,247] Kin-keeping, as introduced by Rosenthal, is a family system level construct referred as a "*designated family role within the family division of labor*."[246] Research has shown that the kin

keeper role typically involves a key family member managing and maintaining family connections and communication in a close family unit.

Understanding the interplay between kin-keeping and *TM* provides a more comprehensive view of how family roles affect FGRC with different relatives. Especially in the context of ovarian cancer, where guidelines recommend that at-risk relatives seek genetic counseling, opening additional avenues in communication with relatives. Examining how kin-keeping roles overlap with survivors' communication role could identify potential alternative mechanisms among kin-keepers, who may not be probands, to initial and promote *TM* beyond proband-centric interventions.

Previous research on FGRC has shown mixed results regarding its impact on family relationships; some studies indicate that sharing genetic testing results with relatives can strengthen family relationships, while others suggest it may lead to isolation.[248,249] In my study, when thinking aloud while visiting YFC, some ovarian cancer survivors demonstrated an orientation to transform their motives towards relationship-oriented. That is, for many of them, their rationales behind action and cognitive engagement were based on their very specific relationships with each relative. Regardless of whether they ultimately chose to list them, survivors can speak of specific reasons behind their decisions, reflecting a sense of relationship maintenance.

These findings highlight the need to understand how *TM* and communal coping processes across different proband-relative dyads and family structures. As described in Chapter 1, theories at family-system level lend insights into what triggers probands to consider differently in their enumeration behaviors and cognitive perceptions of their relatives' risk. For example, Family Communication Patterns theory suggested examining family norms that have been established and shaped over time, and its influence on relational appraisal.[127,129] And Family Systems Perspective suggested assessing organization and structure of family relationships, and health-related cognitions and beliefs shared within families.[126] Future research should still focus on qualitative exploration, and develop interview questions that focus on these family-system level factors to better understand how it influences probands' enumeration decisions and perceptions regarding genetic risk communication.

Furthermore, as identified in cognitive factor insights; many survivors did not view communication as a means of preserving family relationships. For some, it is plausible that the relationship was not established from beginning. For example, gender misconception greatly affected participants enumeration of male relatives in factor 1. While ovarian cancer primarily affects women, it is important to note that the underlying genetic risk of *BRCA1/2* also impacts

men in the families. However, this connection did not seem to be recognized by ovarian cancer survivors. As a result, they may not have perceived the need to maintain a relationship with male relatives regarding genetic risk. Thus, future research could benefit from adopting a gendered perspective. This should include studying male pathogenic variant carriers to gain a comprehensive understanding of how to establish relationships beyond simply having probands view information on a website. Additionally, it would be valuable to explore how the *TM* process operates differently for male and female probands.

In addition, as layered contact approaches were preferred, it is important to assess how a combination of different outreach approaches could improve the quality of FGRC. For some probands, direct communication with relatives was considered the most straightforward “no”-brainer approach. As this outreach approach aligns with the standard of care, accompanied by a generic notification letter, it is important to examine the content of these communications. Current studies often treated communication of inherited risk as a binary action (yes/no).[52,63,78,80] Much needed is qualitative research to explore what actually occurs during these conversations. Specifically, it is important to investigate the extent to which relational aspects, such as a sense of “we”-responsibility and solution are inferred or explicitly discussed, and whether these relational talks could effectively motivate relatives to seek genetic counseling.

For those relatives to be contacted by a third party, such as the study team in YFC, it is important to acknowledge that the probands still play an important role in communication. Survivors’ enumeration actions relied on thoughtful appraisals of individual relationships as they did not simply forget to list certain relatives on the website. Instead, they could articulate well on specific reasons for not listing or reaching out to specific relatives, such as a desire to preserve their relatives’ autonomy in getting risk information and emotional closeness. On the other hand, privacy policies and laws emphasize the need for self-control when it comes to disclosing personal medical information.[34,35] These disclosure regulations put probands in the spotlight, “, as any third-party outreach must inconveniently begin with the proband’s consent. Thus, future research should assess the acceptability of having a stepped outreach approach, where a third party communicates with those relatives only after obtaining the proband’s consent.

6.3 CONCLUSIONS

The *TM* is a conceptually compelling construct that to date has been poorly operationalized. In order to evaluate whether *TM* can be leveraged in behavior change interventions, we must develop rigorous assessment tools to characterize it. Future research needs to move beyond pronoun counts to consider survivor appraisal processes multiple family

members' perspectives on its occurrence, develop and test measures of relational thinking and their association with a variety of shared coping strategies (including health behaviors as the gold standard). Once the abstract latent *TM* can be measures future effort should focus on developing and testing of *TM*-informed strategies to foster behaviors such as family inherited risk communication.

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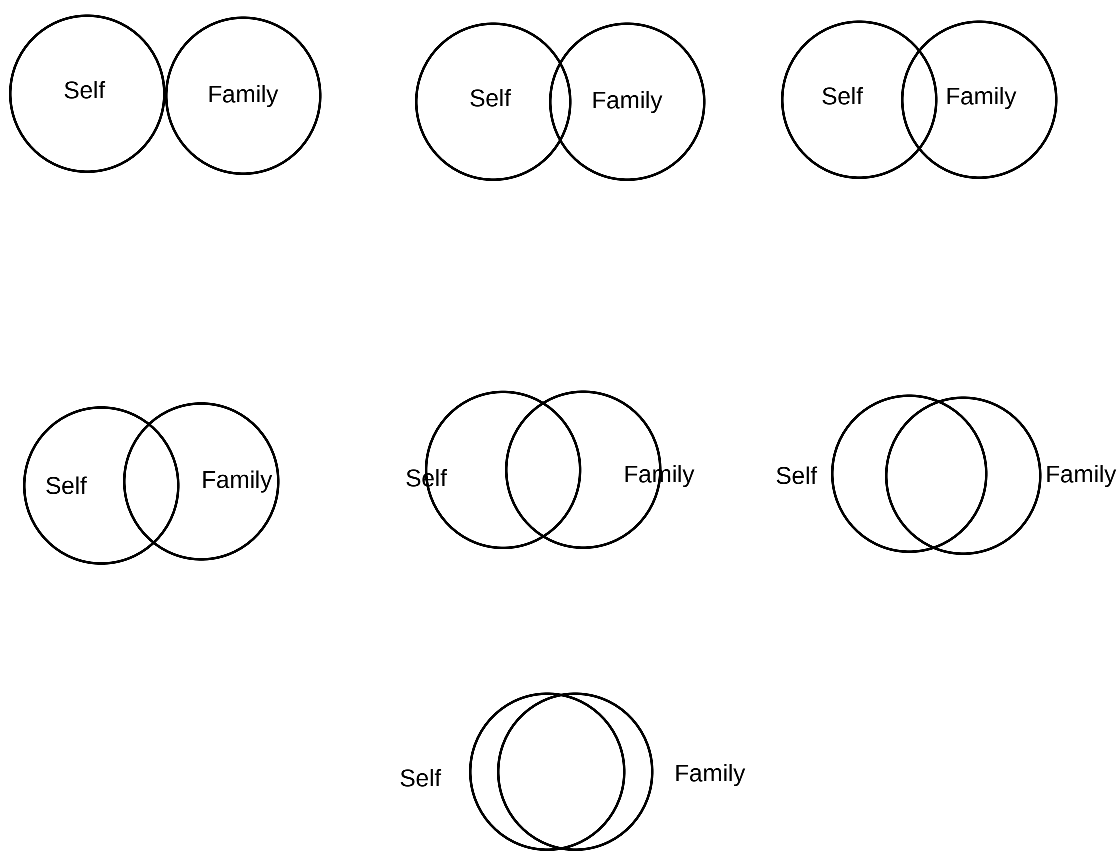
APPENDIX 1. THE ORIGINAL INCLUSION OF OTHER IN THE SELF SCALE

Scoring:

Respondents choose a pair of circles from seven with different degrees of overlap. 1 = no overlap; 2 = little overlap; 3 = some overlap; 4 = equal overlap; 5 = strong overlap; 6 = very strong overlap; 7 = most overlap. The number chosen is the respondent's score.

Instructions:

Please circle the picture below that best describes your relationship.

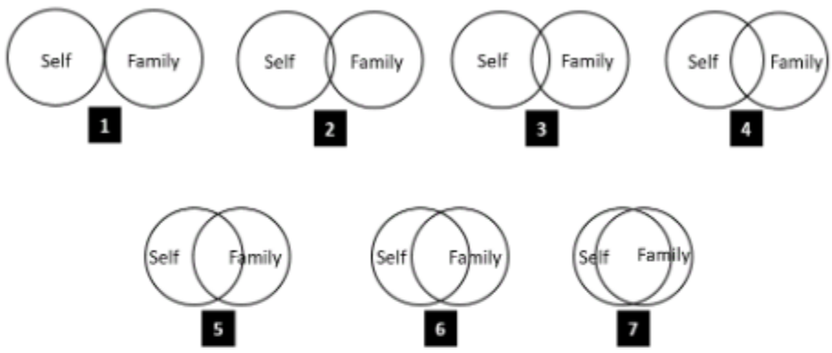


APPENDIX 2. WEBSITE ACTIVITY LOG COUNTS

Automated system generated logs	Counts	Page viewed logs	Counts	Website interaction logs	Counts
Access code used	22	/Counseling/aboutcounseling	94	Counseling questionnaire completed	65
Access code used for the first time	158	/Counseling/counselingquestions	145	Counseling requested	27
Consented to study	145	/Counseling/counselingrequest	139	Downloaded letter	235
Consented to website	141	/Home/contactedit	9	Login	76
Counseling session confirmation	26	/Home/dashboard	62	Login created	132
Counseling session requested	28	/Home/familycommunication	9	Logout	69
Email validated	54	/Home/otherresources	12	Read counseling information	69
Message status	127	/Home/sensitiveconversations	21	Relative created	182
Mobile phone validated	127	/Home/support	16	Relative deleted	49
Participant is designated contact - MBI	47	/Home/updatepassword	1	Step 1: family members added	115
Relative entry completion reminder - MBI	188	/Participant/familyintake	197	Step 2: viewed types of contact	108
Relative info entry completion reminder	45	/Participant/familyintakesteptwo	181	Step 3: contact methods set	65
Schedule counseling reminder - MBI	115	/Participant/familyintakestepthree	201	Step 4: finished family intake	62
Validated mobile phone number	127	/Participant/familyintakestepfour	115		
Validation email sent	132				
Welcome text sent	127				

APPENDIX 3. ADAPTED INCLUSION OF OTHER IN THE SELF SCALE

Below are 7 pairs of circles that range from just touching to almost completely overlapping. One circle in each pair is labeled "self," and the second circle is labeled "family." Please choose one of the seven pairs to answer the following question.



Think of your family as a whole, which pair best describes how close you feel with your family in emotional closeness?

1

2

3

4

5

6

7

APPENDIX 4. INTERVIEW GUIDE

Notes to interviewers:

- Please be fully visible and have good lighting on Zoom
- Please use the "access code for interview" to pre-enter participants relative enumeration activities on the landing page of the website right before the interview
- Please ask all probing questions that are labeled "must cover"
- Please feel free to ask probing questions that are not on the interview guide (especially when you feel the need to follow-up with participants responses)
- Please be sensitive that participants have had ovarian cancer

Give a brief introduction of yourself

Remind participants that they have agreed to audio-record the interview in the consent phone call, and they can turn off the camera if they choose to. We are going to ask to engage with the website as if this is something you believe that would be helpful for you.

TURN ON RECORDING

Intro (5 minutes)

I see that as requested, you were able to visit the Your Family Connects website before this interview. I now have had a chance to look over the activities you reviewed on the website. As we move into the next phase of our discussion, I am seeing that you spent about X minutes on the website, you identified X relatives, and you downloaded X family letters.

In this phase of our discussion, I want to watch you and ask you to share what you were thinking as you went through the Your Family Connects website sections. For each section, we will talk about the actions you took and actions you chose not to take so that I can get a better understanding of your reasoning. The interview will take about 90 minutes. As we explained in the consent form, this interview will be audio-recorded.

In each section, I will give you a couple of minutes to remind yourself of the content and when you are ready I will ask you to tell me your reactions to the content. First stop is the "*Invite Family Members."*

Task 1 – Invite Family Members (30 minutes)

Now we'll start with the first section, *Invite Family Members*. Please glance over the *Invite Family Members* section, and let me know when you are finished.

1. When you first visited the website, you spent just about X minutes on this section. Can you tell me a little about your thoughts about the content?

- PROBE: What do you think the main message was for this section?
- PROBE: Can you say a little more about that?
- PROBE: Anything else that strikes you?

2. How did the fact that ovarian cancer can run in families influence your choice of which relatives to identify?
    - PROBE: Could you tell me more about that?
    - PROBE: Please explain what you mean.
3. Now looking over the three options that were offered for contacting relatives, what were your thoughts about using these options?
    - PROBE: What are your thoughts about using different contact approaches based on each specific relationship?
    - PROBE: Could you tell me more about?
    - PROBE: Please explain what you mean.

[*If participant listed at least one relative, go to question 4; if participant did not list any relatives, go to question 6*]

4. I see that you listed X first-degree relatives and/or X second-degree relatives. Is this all the relatives you could have listed? [USE THIS ANSWER TO GUIDE THROUGH QUESTIONS TO FOLLOW]
5. Can you tell me more about what influenced your decision to list these relatives (AND NOT OTHERS IF APPROPRIATE)?
    - PROBE (must cover): In terms of emotional closeness, could you tell me more about how it differs between relatives you listed and relatives you did not list?
    - PROBE (must cover): How about communication styles in your family -- Do they differ between you and the relatives you listed and the relatives you did not list?
    - PROBE: Why did you list more first-degree relatives than second-degree relatives (or vice versa)?
6. You did not identify any relatives. Can you tell me more about your decision not to list any relatives?
    - PROBE (must cover): Could you tell me a little about your feelings of emotional closeness to these relatives?
    - PROBE (must cover): How about communication styles in your family -- Do they differ between you and the relatives you could have listed but did not?

[If participant selected only one contact approach, go to question 7; if participant selected more than one approach, go to question 8; If participants did not select contact approach because she did not enumerate relatives, go to question 9]

7. You chose the same contact approach for all relatives/Your chose ____ for the one relative that you identified. Tell me a little about your thinking as you considered the options for contact?
    - PROBE: Why not the other approaches?
    - PROBE: How did your emotional closeness and communication style influence your choice?
    - PROBE: How do you view your role in sharing this risk information with your family?
    - PROBE: Could you tell me more about that?
8. You chose different options of contact for the family members you identified. Tell me more about why you chose different contact approaches for your relatives?
    - PROBE: How did your emotional closeness to these relatives and communication styles influence the selection of different approaches?
    - PROBE: How do you view your role with respect to conveying risk information to your family?
    - PROBE: Could you tell me more about that?
9. In a hypothetical situation where you were asked to select contact approaches for your relatives. What contact approaches would you consider?
    - PROBE: How does your emotional closeness to these relatives and communication styles influence the selection of different approaches?
    - PROBE: How do you view your role with respect to conveying risk information to your family?
    - PROBE: Could you tell me more about that?
10. I see that you downloaded N family letters. Can you tell me more about your thinking when you decided to download family letters?
    - PROBE: How do you think your relatives would respond to receiving the family letter from you?
    - PROBE: Did you customize the letter in any way for each family member?
    - PROBE: Tell me more about that?

Task 2 – Genetic Counseling [15 minutes]

Now let's move on to the next section, Genetic counseling. Please take a quick look at the section to remind yourself of the content.

11. We were offering genetic counseling when the main trial is ongoing. If the genetic counseling is offered to you on the website, would you choose to request a genetic counseling session. Tell me more about your decision
    - PROBE: What, if any, benefits do you see to [THE FAMILY MEMBERS YOU IDENTIFIED] meeting with a genetic counselor?
    - PROBE: What, if any, benefits do you see to [THE FAMILY MEMBERS YOU DIDN'T IDENTIFY] meeting with a genetic counselor?
    - PROBE: Can you tell me more about that?

Task 3 – Importance of Family Communication (20 minutes)

Now we are moving on to the *Importance of Family Communication* section. Please take a quick look at the content to remind yourself.

12. How does your family regard the importance of communication with each other, generally?
    - PROBE: Can you tell me a little about how communication flows in your family with respect to news about family events – births, marriages, health events?
    - PROBE: Could you tell me more about that?
13. How did your journey with ovarian cancer influence your views about the importance of family communication?
    - PROBE: How did the awareness that ovarian cancer can run in families influence your thinking?
    - PROBE: Could you tell me more about that?
    - PROBE: Did having cancer make it harder/easier to communicate this news to your family?
14. Why did you decide [NOT] to visit this webpage?
    - PROBE: Could you tell me more about that?
    - PROBE: Please explain what you mean.

Task 4 – Sensitive Conversation Framework (15 minutes)

Now we'll move to the last section, *Sensitive Conversation Framework.* Please take a quick look to refresh your memory.

15. Continuing our discussion of family communication, how did the tips in the sensitive conversation framework land with you in relation to your family's communication style?
    - PROBE: What about it seemed useful to you?
    - PROBE: What about is did not seem useful to you?
    - PROBE: Can you tell me more about that?
16. Can you tell me more about your decision [TO DOWNLOAD] [NOT TO DOWNLOAD] the sensitive conversation framework tip sheet?
    - PROBE: Why/why not?
    - PROBE DOWNLOADED: Which of the tips did you use in your conversations with family members?

When finished with the interview (5 minutes)

That's all the questions I have. Do you have any last thoughts on the website we viewed together? Do you have any questions or concerns?

Thank you for your time.

TURN OFF RECORDING