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Applying the framework for developing and evaluating complex interventions to increase family communication about hereditary cancer

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ABSTRACT

Objective: Evaluate an intervention to increase family communication (FC) of positive hereditary cancer test results using the Framework for Developing and Evaluating Complex Interventions (FDECI).
Methods: We developed 'programme theory' during the FDECI development phase by aligning intervention components with behavior change techniques (BCTs) and theoretical factors expected to improve FC. During the feasibility phase, we obtained feedback from 12 stakeholder interviews.
Results: Intervention components aligned with a total of 14 unique BCTs for which prior evidence links the BCT to theoretical factors that influence behavior change. Constructive stakeholder feedback included: more information desired, rewording to support autonomy by highlighting options, and improvements to navigation, visuals, and audio. Positive comments included: comprehensiveness of materials, modeling of conversations, and usefulness of the materials for helping a person prepare to share positive test results.
Conclusion: The first FDECI phases were helpful for improving the intervention and planning our ongoing effectiveness and future implementation phases.
Innovation: Our application of the FDECI is novel, including plans to test our 'programme theory' using coincidence analysis (CNA) to determine who accesses which intervention materials, how utilizing certain materials impact the aligned theoretical factors, and whether these in turn make a difference in the behavioral outcome.

1. Introduction

1.1. Family communication (FC) and genetic testing for hereditary cancer

Approximately 5–10% of all cancers are hereditary, meaning they are due to a pathogenic or likely pathogenic (P/LP) variant in one of several genes that substantially increases cancer risks [1,2]. When an individual has a P/LP variant identified, they along with their at-risk family members can benefit from genetic testing and risk-appropriate cancer screening and management options that reduce cancer-related morbidity and mortality [3]. Unfortunately, about 20–40% of family members remain uninformed about P/LP variant ("positive") genetic test results [4-8]. Increasing family communication (FC), with subsequent testing in family members, can maximize the impact of genetic testing to prevent cancers or detect them early among those at highest risk [9-13].

FC in the context of hereditary cancer is often operationalized by researchers as disclosure of a genetic test result to family members [8,14-19]. FC varies based on a number of individual-level factors, including the ability to recognize family members' 'increased risk', perceived responsibility to share information, relationship type, physical or emotional closeness, perceptions of whether relatives want to know, anticipation of relatives' reactions, personal emotions, and perceptions that discussing cancer is not accepted within the family [20-23]. Additional factors that may influence FC include support from family and friends, available resources, healthcare provider support and knowledge, and family structure. [13,16,19,24-28].

Testing rates among family members are substantially lower than rates of results disclosure [4,5,8-10,14,15,25,29-32], with two recent studies reporting that only 30% of relatives had undergone testing [15,30]. Reasons for low testing rates are unclear; however, a study found that some family members who were informed of a pathogenic variant in their family reported believing they are at lower risk for cancer [8], while another study found family members frame their narrative differently [33]. Messages shared may be ineffective at motivating family members to take action or

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family members may need additional information or reminders. Ultimately, we anticipate that successful cascade testing of family members will rely on appropriate, effective messaging in addition to *ongoing* communication between family members.

1.2. Interventions to improve FC and/or genetic testing rates among at-risk family members

Interventions to improve FC and testing have attempted to increase knowledge of familial cancer risks and which relatives to share with, improve self-motivation and self-efficacy to communicate with family members, reduce perceived barriers (i.e., fear and distress of communicating with family), or provide resources to give to relatives [34]. Most FC interventions treat family sharing of test results as a one-time provision of information [22,25,35]; and many showed no statistically significant differences between intervention and control groups with regard to communication with family members or uptake of genetic services by family members [22,25,36-38]. Interventions that demonstrated significant improvements in communication with family, increased knowledge of cancer risks, or increased uptake of genetic services by family members used a variety of delivery methods (e.g., in-person, e-health, mobile app), yet it remains unclear why these interventions were effective while other interventions were not.

1.3. Theory and FC interventions

Theory can help in understanding how or why interventions are effective. However, in a recent systematic review, 5 out of 9 studies seeking to improve FC did not clearly articulate how the theory guided intervention development [34]. Furthermore, not all theoretical frameworks outline explanatory mechanisms by which an intervention is expected to impact FC. For example, an intervention based on the Reciprocal Engagement Model of Genetic Counseling (which focuses primarily on the process of genetic counseling rather than behavior change) was ineffective at increasing uptake of cascade testing for hereditary cancer [38]. Researchers involved in that study later reviewed the communication strategies used by the genetic counselors during the intervention to try and better understand why it may have been ineffective. The researchers did this by applying the Theoretical Domains Framework to align each strategy with behavior change techniques (BCTs) in order to determine how many had established mechanistic links (defined as being both theoretically aligned based on expert consensus and demonstrating statistically significant associations according to a prior literature synthesis) [39-42]. Taylor et al. (2020) found value in their process of aligning theory to the communication strategies, but also acknowledged that ideally this process would be done when initially designing a behavioral intervention.

1.4. Content and other factors may impact FC intervention effectiveness

Although interventions that are developed based on models or theories are more likely to be effective than those not guided by theory [43-46], both effective and ineffective FC interventions have previously used the same theories [22,47-49]. These findings suggest that an intervention's content or the contexts in which interventions are implemented may also impact effectiveness [50,51]. Furthermore, intervention effectiveness can be dependent on differences in individual-level factors (e.g., attitudes, barriers, preferences, etc.) or complex relationships between multiple factors [50]. For example, knowledge is often necessary, but alone is usually insufficient for behavior change [52,53]; thus, other factors, such as self-efficacy or access to resources, may also be needed.

Additional complexity occurs when more than one factor can independently lead to the same outcome. For example, one FC intervention may prove effective for individuals who already believe sharing information will benefit family members, but it may be ineffective for others. At the same time, a different intervention could improve FC among individuals who fail to see benefits to FC and are worried about family members' reactions. Therefore, these types of complexities should be considered when developing interventions and evaluating their effectiveness.

1.5. Purpose of the current study

FC, for the purpose of this study, was more broadly defined as an ongoing, transactional process of sharing information about hereditary cancer with at-risk family members, which co-constructs family relationships [54,55]. More specifically, FC requires planning who will communicate with each family member, how and when the test result and basic information about hereditary cancer will be initially shared, how and when to follow-up with family members after initial disclosure as well as being cognizant of how FC may impact family members and relationships [54]. To this end, we used several FC and behavioral change theories to guide the development and refinement of multiple resources that comprise components of a complex FC intervention that is now being tested for effectiveness using a pragmatic randomized controlled trial (RCT) as detailed in our published protocol [56]. Additional study aims involve revising existing FC components and creating additional components or delivery methods to fit unmet or evolving needs we uncover from stakeholder and participant interviews and surveys obtained before, during, and after the RCT. After publishing our protocol, we became aware of the Framework for Developing and Evaluating Complex Interventions (FDECI) [57], which we believe to be valuable for cohesively explaining the methods and results from our other study aims. The FDECI consists of four phases including: (1) development, (2) feasibility, (3) evaluation, and (4) implementation. The authors of this framework recommend engaging multiple stakeholders at each phase and encourage the use of 'programme theory' to develop and modify complex interventions. 'Programme theory' describes how an intervention is expected to lead to its effects through first articulating key components of the intervention and their hypothesized mechanisms of action to impact the target behavior [57].

In this manuscript, we apply the FDECI to: 1) describe the development phase in which we created our 'programme theory' and refined our intervention designed to improve FC of pathogenic variants in hereditary cancer genes; 2) present results of the feasibility phase where we interviewed twelve individuals from our stakeholder population; 3) demonstrate how 'programme theory' and stakeholder feedback helped modify our complex intervention; and 4) describe how we plan to conduct coincidence analysis (CNA) as an innovative method to test our 'programme theory' using longitudinal data gathered during the evaluation phase of our study.

2. Methods

2.1. Intervention materials development phase

During the development phase, we revised educational materials on hereditary cancer and FC that were previously developed by several study team members. We also created additional intervention materials in order to better address FC facilitators, concerns, or barriers identified in prior studies [8,58]. Audio and visuals were incorporated as most participants appreciated use of multiple mediums in our prior evaluation of hereditary cancer education materials [59]. We kept the information basic to ensure key messages are not lost and participants are not overwhelmed. However, given findings in our prior study that some individuals wanted more details, we accommodated through adding "Learn More" buttons to incorporate additional information [59]. Visuals reinforcing key messages were also incorporated to improve understanding among individuals with lower literacy [60-62]. We also focused on visual appeal as that can garner attention [60] and increase the likelihood that the material will hold the attention of individuals and they will find materials acceptable [63]. We made certain several of the resources were easy to print or email to family members

because prior research found that between 30% and 38% of participants shared written materials [16]. Additionally, written materials provide credibility, remove some burden from the patient, and may increase the likelihood that the medical information shared is accurate and less likely to be forgotten [64].

The development and initial refinement of the materials was a highly iterative and creative process that did not involve any formalized data analysis. Rather, intervention development was based on prior literature, theory, expert opinions, and majority consensus. Each component/message underwent review by a minimum of six core study team members including: DC (PhD trained genetic counselor and behavioral/implementation researcher), TP (clinical geneticist and epidemiologic/translational researcher), MD (PhD trained health communication researcher), AW (Master's trained public health researcher), and PH and AT (both Master's trained anthropologists). Several other topic experts reviewed content as well. Components underwent a minimum of three rounds of revisions, although most underwent more. Readability statistics were used to reduce the reading level of various components. Finally, materials were organized into a website to make it easier for individuals to find those that fit their needs and preferences.

Throughout the development phase, we used theories and prior research to create a rationale for each intervention component and worked to ensure materials align with our main behavioral framework for the study (i.e., the capability, opportunity, motivation, behavior (COM-B) model). We then applied methods described by Taylor et al. (2020) to modify our rationale and create our 'programme theory'. Specifically, the first author (DC) aligned intervention components with BCTs from the behavior change taxonomy and mechanisms of action, which are theoretical factors hypothesized to serve as precursors to the behavior, which in this case is family communication [39]. These factors, several of which are shown in parentheses, fall within the following three main areas of the COM-B model: 1) Capability (knowledge, skills, beliefs about capability); 2) Opportunity (social influences, environmental context, and resources); and 3) Motivation (beliefs about consequences, social role/identity, goals, emotions, intentions) [65]. The final 'programme theory' was reviewed by PH and MK and uncertainties or disagreements were discussed and clarified or resolved.

2.2. Intervention feasibility phase

After the University of South Florida Institutional Review Board agreed our feasibility phase constituted an evaluation of the intervention materials and was not human subjects research, PH conducted interviews with six advisory board members and six additional individuals recruited from the Inherited Cancer Registry (ICARE). Participants viewed the materials (i.e., the website, animated audio/visual tools that are referred to as videos, and handouts) while sharing their screen with PH. Evaluation participants were prompted to "think-aloud" by articulating anything that came to mind while reviewing materials and to provide any and all suggestions for improvement [66]. A semi-structured interview guide was also used to elicit perceptions of acceptability, understandability, utility, and visual appeal [59]. Although the majority of interview time was spent discussing areas of improvement, individuals were also asked what they liked. Interviews were recorded and PH took detailed notes documenting all constructive and positive comments while reviewing the recordings. All authors met to discuss and come to a consensus on how to address the constructive comments for each intervention component before revising intervention materials. Due to our iterative refinement process, some participants reviewed modified versions to ensure changes were acceptable. PH then compiled constructive comments for all components within the following categories: content, navigation, visuals/audio, and usefulness. Each category was further divided into agreed upon subcategories by DC, MD, and PH to summarize constructive feedback from the feasibility phase. Authors completed the same process for positive comments in addition to selecting demonstrative quotes.

3. Results

3.1. Results of intervention materials development phase

The development phase resulted in a complete intervention as well as our 'programme theory' which aligned intervention components with BCTs and theoretical factors that serve as mechanisms of action (or links) by which intervention components are anticipated to impact FC (see Table 1). All components except the introductory video employed two or more BCTs with confirmed links based on both expert consensus and empirical evidence [41,42,67]. Some components applied the same BCTs; however, we used a total of 14 unique BCTs with confirmed theoretical factors linking the technique with behavior change across all intervention components.

In what follows we describe each of the key intervention components and rationale to support their inclusion. We then provide examples illustrating how intervention components address 'programme theory' including components of the COM-B model – capability, opportunity, and motivation.

3.1.1. Component overview and rationale

Brief Audio-Visual Introduction: To begin, when participants navigate to the FC intervention website, they see a recommendation to begin by viewing a 3-minute, audio-visual tool. This animated tool describes implications of a positive genetic test result, how sharing information may change medical care and improve outcomes for family members, which family members may benefit from the information, and possible next steps participants can take when navigating additional sections of the website, depending on their readiness or needs.

Decide Whether to Share Results with Family: This section of the website provides content that may help patients consider whether to share information with family members. This section also includes a one-page, downloadable PDF that can be printed or completed electronically by checking off commonly reported reasons why people choose to engage in FC [16,19] or writing/typing in their own additional reasons.

Plan Who, When, and How to Share Genetic Test Results (web content and planning guide): This section includes items patients may want to consider about who, when, and how to: 1) share information with family and 2) follow-up afterwards. People may think that sharing a test result once is sufficient, but prior studies have determined that active persuasion and reminders increase genetic testing uptake by family members [68]. Thus, we hypothesized that the lack of follow-up may help explain the low testing rates among family members [14,15,30,31]. The goal of this section is to prompt patients to recognize that family members may not fully understand or be ready to take action after discussing information the first time. Our messaging frames the process of following up as an important way to support family members and ensure they have and remember key information.

The *planning guide*, which is a two-page, downloadable PDF worksheet, allows patients to fill in details about who will share information with whom, when, and how (including a separate column for when follow-up with each family member will occur). Given that people share more often with first-degree relatives and less often with second- and third-degree relatives [69-71], there is a prompt at the top of the worksheet encouraging the family to plan for who will share with more distant relatives [72,73].

Animated Scenarios and Printable PDF– Sharing Your Genetic Test Results with Family Members & Ways You Can Respond to Family Member Reactions: Clickable, animated slides with optional audio provide participants with scenarios demonstrating ways to have a conversation about a positive test result with family members. The impetus for developing samples of what to say came from studies where individuals reported feeling unprepared to share information [16,19,74], and from a participant in our own prior study who suggested having a script might help [19]. Furthermore, over half of tested individuals in one study were uncertain about the exact message to convey to their relatives [36]. Consequently, we also included an accompanying printable handout reinforcing key messages of what to say.

Table 1
Participant website intervention components and 'Programme Theory' (I.e., alignment with behavior change techniques and theory).

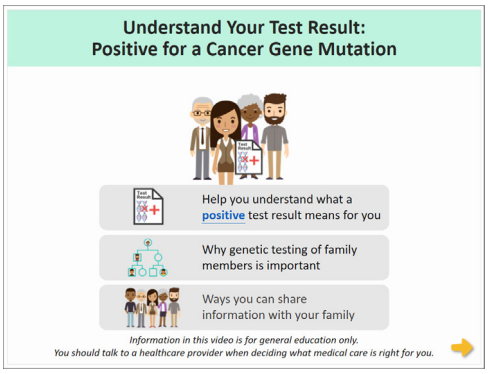
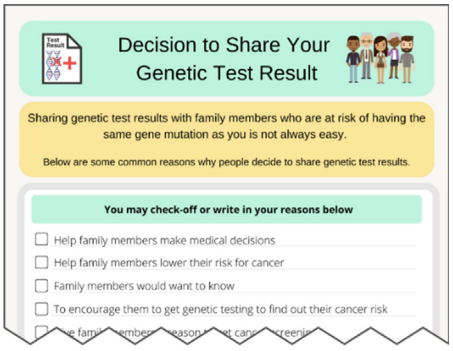
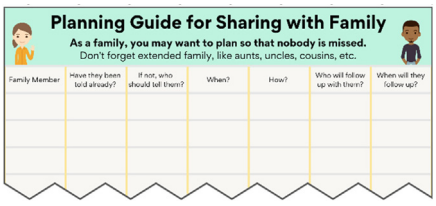
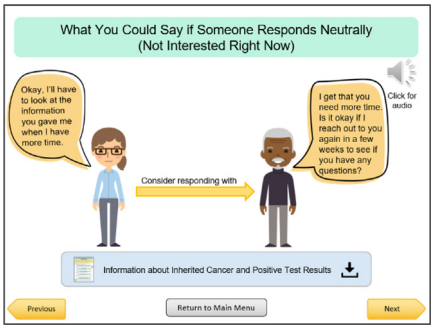
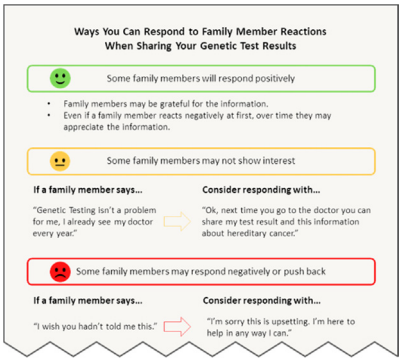
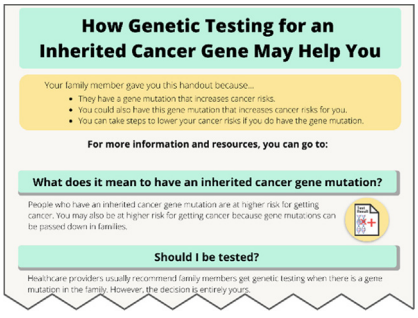
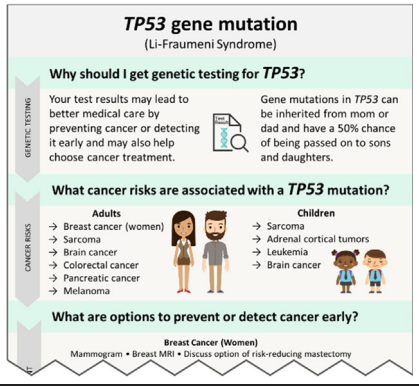
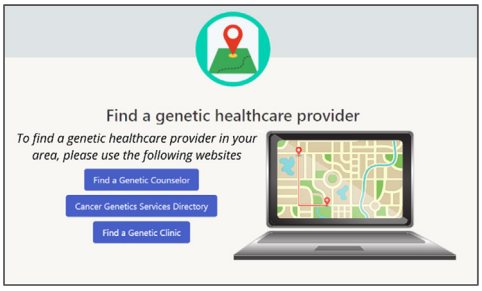
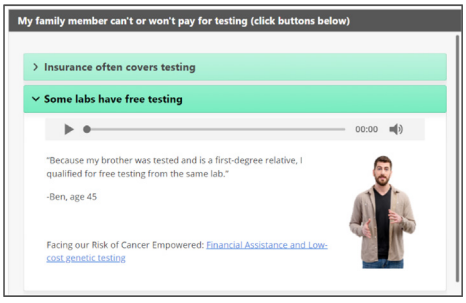
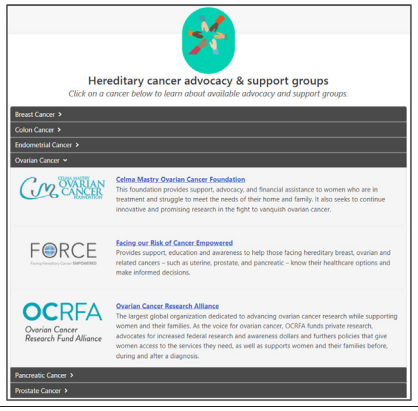
Intervention Component Brief description	Screenshots	COM-B category[a] Behavioral Change Techniques (BCTs)[b] [*Theoretical factors targeted by the intervention component*[c,d,e]]
Homepage 3-minute video (animated slides with audio) • Meaning of positive test result • Utility of sharing positive test result with family members (may change their medical care & even save lives) • Which family members to inform about positive test result • List of possible next steps depending on readiness and needs Links to other website sections listed in rows below.		**Capability and Motivation** • Information about health consequences [*knowledge*[c] ; *intention*[c]; *beliefs about consequences*[c]; *attitudes toward behavior*[c]; *perceived susceptibility*[c]]
Section 1 Webpage – Decide Whether to Share Result with Family • Most family members want the information • Most people decide to share Downloadable PDF Worksheet – Decide Why to Share Your Genetic Test Result (see screenshot) *Reading level: 6.9*		**Opportunity** • Social comparison [*subjective norms*[c]; *normative beliefs*[c]; *social influencing*[c]; *social role/identity*[d]] • Information about others' approval [*subjective norms*[c]; *normative beliefs*[c]; *social influencing*[c] ; *intention*[d]] **Motivation** • Identify benefits of behavior (pros) [*motivation*[c]; *attitudes*[c]; *beliefs about consequences*[c]; *decision making*[e]] • Anticipated regret [*beliefs about consequences*[c]; *emotion* [d]]
Section 2 Webpage — Step-by-Step Guide to Plan Sharing Genetic Test Result: • Which family members • When/how to share • When to follow-up Downloadable PDF worksheet • Planning Guide for Sharing with Family • Includes suggestion to enlist other family members to help with sharing *Reading level: 4.1*	 	**Capability** • Instruction on how to perform the behavior [*knowledge* [c]; *skills* [c]; *beliefs about capabilities*[c]] • Prompts/cues [*Memory, attention, decision processes* [c]] • Goal setting (behavior) *[intentions* [c]; *goals* [c]; *beliefs about capabilities*[d]*]* **Opportunity** • Social support (practical) [*environmental context and resources*[c]; *social influences*[c]]

Table 1 (*continued*)

Intervention Component Brief description	Screenshots	COM-B category[a] Behavioral Change Techniques (BCTs)[b] [*Theoretical factors targeted by the intervention component*[c,d,e]]
Section 3.1 Video (animated slides with optional audio): • Examples of how to start a conversation • Key risk and efficacy messages • Ending the conversation • How to respond to positive, neutral, or negative family member reactions Downloadable PDF Handout – Key Messages to Share When Talking to Family Members about Genetic Test Result & Ways You Can Respond to Family Member Reactions • Printable version of the messages in the video *Reading Level: 6.9*	 	**Capability** • Instruction on how to perform the behavior *[knowledge[c]; skills[c]; beliefs about capabilities[c]]* • Information about social consequences [*knowledge[c]*] • Demonstration of the behavior [*beliefs about capabilities[c]; skills[d]*] **Opportunity** • Social support (practical) *[environmental context and resources[c]; social influences[c]]* • Vicarious consequences [social learning[d]] **Motivation** • Information about health and social consequences [*intention[c]; beliefs about consequences[c]; attitudes toward behavior[c];*] • Reduce negative emotions *[Fear[c], anxiety[c]]* • Anticipated regret [*beliefs about consequences[c]; emotion* [d]]
Section 3.2 Fillable Templates – Family Sharing Email & Family Sharing Letter *Reading Level: 7.7 & 6.6* Downloadable PDF Handout – How Genetic Testing for Inherited Cancer May Help You *Reading Level: 7.6* Downloadable PDF Handout – Gene specific flyers • Information sheets for 23 individual cancer risk genes *Reading Level: 7.2-9.9 (varies by individual gene handout)*	 	**Motivation** (for family members) • Information about health consequences [*knowledge[c]; intention[c]; beliefs about consequences[c]; attitudes toward behavior[c]; perceived susceptibility[c]*] **Opportunity** • Social support (practical) *[environmental context and resources[c]; social influences on family members[c]]*

(continued on next page)

Table 1 (*continued*)

Intervention Component Brief description	Screenshots	COM-B category[a] Behavioral Change Techniques (BCTs)[b] [*Theoretical factors targeted by the intervention component*[c,d,e]]
Section 3.3 Links to External Websites – Find a Genetic Healthcare Provider • NSGC Find a GC • NCI Cancer Genetics Services Directory • ACMG Find a Genetic Service		**Capability** (for family members) • Instruction on how to find a genetic counselor *[knowledge [c]; skills [c]; beliefs about capabilities[c]]* **Opportunity** (for the patient or their family members) • Social support (practical) *[environmental context and resources[c]; social influences on family members[c]]*
Section 4.1 Video – Others' Experiences Sharing Genetic Test Result (FAQ style video)		**Opportunity** • Social comparison [*subjective norms[c]; normative beliefs[c]; social influencing[c]; social role/identity[d]*] • Information about others' approval [*subjective norms[c]; normative beliefs[c]; social influencing[c]; intention[d]*] • Social support (practical) *[social influences[c]]* **Motivation** • Reduce negative emotions *[Fear[c], anxiety[c]]* • Information about health and social consequences [*knowledge[c]; intention[c]; beliefs about consequences[c]; attitudes toward behavior[c]; perceived susceptibility[c]*]
Section 4.2 – links to external websites Support Groups listed by cancer type Resources for support and guidance • Adjusting to cancer/survivorship • Financial resources • Finding and choosing a cancer counselor/therapist • Finding and choosing a support group • Support and resources for caregivers		**Opportunity** • Social support (practical) *[environmental context and resources[c]; social influences[c]]* **Motivation** • Reduce negative emotions *[Fear[c], anxiety[c]]*

[a] Bold headers represent the Capability, Opportunity, Motivation- Behavior (COM-B) model.
[b] Behavior change techniques (BCTs) were aligned with theoretical factors (i.e., causal mechanisms of action) using the Theory and Technique Tool theoryandtechniquetool.humanbehaviourchange.org/tool.
[c] Theoretical factor with confirmed links based on both expert consensus AND empirical evidence.
[d] Factor with inconclusive links - either experts agreed the BCT can cause a change in the factor and subsequent behavior change OR there is sufficient empirical evidence that the factor can serve as a causal link.
[e] Factor is perceived to be necessary but there is no evidence for a causal mechanism of behavior change.

Email and Letter Templates & Handouts for Family Sharing: When patients share with more distant relatives, they are more likely to send them written information [16,31]. Furthermore, written information may increase family members' ability to act on the information [75,76]. Given that traditional family sharing letters are text heavy and written at a 10th grade reading level [77,78], we reduced the reading level of our letter to a 6.6 grade level. We also created a colorful handout written in a question and answer format which breaks up the text and has been shown to be an effective way to convey information [79]. The templates and handouts are simpler than other existing materials because research reveals that many people want less detailed information when they are first told about their risks [80].

Vignettes – Others' Experiences Sharing Genetic Test Results: This section of the website encourages those who are facing barriers or are not ready to engage in FC to view various experiences people have had when sharing genetic test results with family members. These vignettes are represented in 2-3 sentences with a picture and optional audio of the person reading the quotes. Experiences are grouped by topic to help people find relevant information quickly. Vignettes address different barriers reported in the literature and those reported by individuals with pathogenic variants who provided feedback earlier in our process that relate to capability, opportunity, and motivation from the COM-B model. Examples include the following: patient concern about family member reactions or relationships [19], family member is too young or too old to share [19], uncertainty about

how to contact distant family members, family member can't or won't pay for testing, family member doesn't want testing or will not do anything with the information, and family member wonders if testing will affect insurance [20,73,81,82]. Additional examples of how intervention components relate to the COM-B model are described below.

3.1.2. Capability

Basic information sheets for hereditary cancer and for each individual cancer risk gene are expected to increase knowledge for both patients and their family members. Handouts were based on prior research revealing confusion about more recently identified cancer risk genes and findings that some patients were taking actions that were not congruent with the level of cancer risks conferred by their gene [83-87].

The provision of key messages to share with family should increase procedural knowledge. Illustrating ways to respond to different reactions of family members is expected to improve beliefs about capabilities [88]. Content showing how to perform the behavior includes:

1) Examples of what to say when starting a conversation
2) Key risk and efficacy messages to share during the conversation
3) Ending a conversation and opening up the opportunity to follow-up with family
4) Ideas for what to say if the participant doesn't know whether a family member wants the information or if their family member demonstrates a positive, neutral, or negative reaction.

3.1.3. Opportunity

Two messages relevant to social opportunity are included in the section of the website on deciding whether to share test results. Prior research has found that when people do not believe a family member wants to know about hereditary cancer, they choose not to share the result [19,72]. Therefore, we highlight how most family members want the information, a finding that is supported by a survey from the United Kingdom showing that 91% of respondents would want to be contacted about a preventable and fatal genetic condition [89]. We also elicit social comparison by stating that most people ultimately decide to share results with family even if they have some concerns.

Narratives, included as part of the patient vignettes, may also influence social norms. Narratives can be effective at promoting health screening behaviors [90]. Furthermore, integrating real-life experiences can enhance trustworthiness and credibility [91].

We also encourage patients to seek social support by recommending within the planning guide that they elicit help from other family members to share information as part of the planning guide. Finally, we include links to hereditary cancer advocacy and support group webpages given research noting that connecting with other people who have been through similar experiences is helpful at not making patients feel alone in their experiences [92].

3.1.4. Motivation

Finally, several intervention components were designed to impact patient motivation. The checklist of common reasons to engage in FC about hereditary cancer was inspired by motivational interviewing (MI). A key way in which MI works is by encouraging 'change talk', whereby people consider and describe reasons they want to or believe they should engage in a behavior [93]. The value of evoking reasons to engage in FC is supported by a study where nearly all participants who communicated with family after completing an MI communication intervention for a different genetic condition consistently demonstrated high levels of change talk during the intervention or they had very high scores on a measure of positive motivations; whereas, neither high change talk nor high positive motivation was found among most participants who failed to complete the behavior [94].

The use of vignettes may also impact motivation given data showing how patient testimonies and stories can often be a powerful, persuasive tool [95]. A couple of patient vignettes were designed to motivate by prompting patients to consider possible outcomes (vicarious consequences), which may evoke anticipated regret from not taking action.

The planning guide section and worksheet include goal setting, which can be effective for behavior change [96]. Using the planning guide may help increase each patient's commitment to follow-up and if they plan for who will share with more distant relatives it may help ensure that cascade testing does not break down, as has commonly been reported [31].

Given that negative emotions about cancer can reduce FC [19,70,97], we developed content to address this barrier. First, demonstrating FC dialogue as part of the scenarios is expected to reduce fears and anxiety over family responses [98]. We also anticipate that negative emotions related to FC can be reduced by informing patients that most family members respond neutrally or positively rather than negatively. Finally, links for hereditary cancer support group webpages and other coping resources may help patients deal with the many challenges of having a pathogenic variant or cancer diagnosis. This is important because patients who are overwhelmed or dealing with their own emotions may be less motivated to share information with family members [92].

Messages included in the templated family letter and handout were designed to increase motivation among the patients' family members. By aligning messages with the extended parallel process model (EPPM), we highlight the threat of hereditary cancer, while making efficacy messages prominent [99]. Given a prior study where perceived risks among family members went down after sharing positive results [8], we wanted to reduce the chance the proband or their family members would minimize the threat message and instead motivate them to mitigate the threat by taking action. Templates include resources to contact a genetics provider and emphasize how knowing about a pathogenic variant provides a way for family members to manage their health (including options that can prevent cancer or find it early when it is easier to treat). These messages align with framing and guidance recommended by Campbell-Salome based on the EPPM and interviews with individuals impacted by a different, medically actionable genetic condition [100].

3.2. Results of the intervention feasibility phase

Characteristics of the twelve stakeholder participants who evaluated intervention components are presented in Table 2. Eleven participants had a pathogenic variant in a hereditary cancer gene, and the twelfth participant had a mother with a pathogenic variant. Most participants identified as White females.

Constructive feedback primarily pertained to content (e.g., more information desired, rewording to support autonomy by highlighting options), followed by improving navigation, visuals, and audio. Constructive comments and how they were addressed (or in a couple of instances our reasoning for not addressing) are shown in Table 3.

After making alterations based on constructive feedback, individuals interviewed later no longer commented on the areas that were altered and it was clear in other comments that the materials prompted some of the participants to consider sharing with more distant family members. Positive comments related mostly to content (e.g., comprehensive materials,

Table 2
Cognitive interviewees demographics table.

ID	Sex	Race	Pathogenic Gene
ICARE 1	Female	Non-Hispanic White	*BRCA 1* +
ICARE 2	Female	Non-Hispanic White	*PALB2* +
ICARE 3	Female	Non-Hispanic White	*BRCA2* +
ICARE 4	Female	Non-Hispanic White	*PALB2* +
ICARE 5	Female	Non-Hispanic White	*BRCA2* +
ICARE 6	Female	Non-Hispanic White	Mother is *BRCA* +
CAB 1	Female	Non-Hispanic White	*PTEN* +
CAB 2	Male	Non-Hispanic White	Lynch Syndrome
CAB 3	Female	Black	Lynch Syndrome
CAB 4	Female	Non-Hispanic White	*RET* +
CAB 5	Female	Non-Hispanic White	*BRCA1* +
CAB 6	Female	Black	*TP53* +

Table 3
Constructive feedback by category and subcategory with supportive illustrative quote and description of changes made.

Category	Subcategories	Illustrative Quotes	Changes made
Content [a]	More info needed on Genetic Information Nondiscrimination Act	"Is there a section [in the homepage video] where they talk about how testing could affect insurance?… I ask because that was a big thing for some of my family members, how this would affect their insurance."	Added a slide explaining what GINA covers and what it does not cover to the homepage video
	Support autonomy by highlighting options	"In the column that says, 'what can be done to prevent or detect cancers early?'… the reality is that people choose very different paths with this. Maybe there can be… something along the lines of 'there are different ways to approach a positive diagnosis from surveillance to surgical intervention depending on family history and individual choice'."	Changed section title from "What can be done to prevent or detect cancers early" to "What are options to prevent or detect cancer early"
Navigation [b]	Main menus/navigation slides of videos	"When I first saw it [the menu for the Sharing Your Genetic Test Results video], I guess I was a little confused on what to click exactly."	Revamped the main menu: Added instructions to click on buttons/outline, made topics look more like buttons, made language consistent, etc.
	Outline on videos	"It's [the outline] very helpful to see where you are and how much more you have to go if you're judging your time."	Added an outline to other videos which did not have one.
	Mobile Friendly	"Remember your audience is huge, your age range, so just make sure that every person that might be encountering these materials can manipulate them on whatever device they are on."	Added steps on how to navigate video on mobile devices
Visuals/Audio [c]	Format	"It's [how genetic testing may help you handout] not necessarily an attention grabber in the title… there might be a better way to title it so it makes people feel 'I should get tested'"	Did not change, did not want to be overly persuasive
		"When it talks about kids [in the homepage video], it talks about the testing depends on the type of cancer risk and ages of cancer in family members… those are two separate things and to me it reads like you are combining those two factors."	Edited text to match the audio
		"The sound levels… got more muted on this one screen [of the homepage video]. It's definitely quieter than it was on the first two screens."	Normalized sound levels
	Cartoon characters	"I don't like videos with people in them, I also don't like cartoon people… That's just me, I'm not sure if live people would be appropriate either [in the videos]."	Did not change, most respondents did not comment on this
Usefulness [d]	Encourage asking for help	"This [other's experiences sharing genetic test results] brings up a good point about 'not ready to talk about it', can we direct them anywhere to consider having someone close to them help them share the information or be their point person to give this information to family members?"	Include encouragement to ask family members for assistance in sharing test results in more places.
	Acknowledge that this is difficult	"Acknowledgement that this result is hard and life changing and not going to be easy would be very beneficial, followed by some encouragement that there are resources available… that can help you through the process."	Added this message to two other places in the materials

[a] Content is feedback about the amount, clarity, and ease of understanding of educational information given verbally (in videos) or through written text of materials.
[b] Navigation is how the participant moves throughout the video including navigation instructions and video features.
[c] Visuals/Audio is feedback about the color, visuals, graphics, and sound throughout the video.
[d] Usefulness is feedback on how helpful the materials would be when preparing to share information with family, Feedback on family sharing materials.

modeling of conversations), followed by the usefulness of the materials for helping a person prepare for and share their positive test result (see illustrative quotes in Table 4).

4. Discussion

Using the FDECI, we present methods and results of the first two phases focused on intervention development and refinement for our trial. Through our trial, we will test if our intervention increases FC among patients with a pathogenic variant in one of several hereditary cancer genes that have associated guidelines for cancer screening, surveillance, or prevention options. The development phase resulted in our refined 'programme theory', which is a description of how we anticipate each intervention component will impact theoretical factors and, in turn, improve the outcome (i.e., FC). This is important because intervention research aimed at increasing FC often lacks details related to intervention development and/or 'programme theory' [34,40,57]. Results from our planning phase helped to refine the intervention components and are anticipated to be useful as we explore how individual characteristics and context may impact the effectiveness of our FC intervention. Results from our feasibility phase have also proven useful because detailed stakeholder feedback helped us iteratively improve the FC intervention, thereby making the intervention more appealing and possibly increasing its effectiveness. Feasibility testing of FC interventions have rarely been reported [25], and we hope to encourage other researchers to use the FDECI to complete and report on development and feasibility phases, as we have done here.

Our intervention utilizes and extends previous research on FC about genetic testing and hereditary cancer in three ways. First, we included known facilitators correlated to family sharing throughout our intervention's messages including the following: having an open and close relationship with family members, feeling a personal responsibility to share with others, desiring to inform other family members of their cancer risks to promote positive health behaviors, and providing written resources to give family members [13,15,19,20,76,101]. Additionally, we created a fill-in response section where users can reflect on other reasons for wanting to share results with family and we encourage sharing with more distant relatives. Second, we support autonomy in sharing by including several resources and options for sharing that all highlight the cancer threat while also emphasizing efficacy in acting on the threats. Third, following our definition of FC, we hypothesize another factor that may strongly influence family uptake of genetic services is ongoing communication about genetic risk with family members [68]. Thus, one of the novel components of our intervention is continual messaging that FC about genetic testing and hereditary cancer should be ongoing and include follow-up conversations with family members to prevent cascade testing breakdown.

Despite our attempt to take a rigorous and transparent approach, the opinions of those involved in the development and feasibility phases may have influenced the intervention components in ways that are biased or not readily apparent. However, we believe that by employing feedback and ideas from multiple experts and community members throughout the development and feasibility phases [102], we have improved the intervention. Nevertheless, we cannot conclude whether the intervention components fit the needs of those who were more recently diagnosed with hereditary cancer. Additionally, we did not probe specifically about people's conceptions of FC during our feasibility phase and feedback related to how we operationalized FC within the intervention components was largely lacking.

Table 4
Positive feedback by category and subcategory with supportive illustrative quote.

Category	Subcategories	Supportive Illustrative Quotes
Content [a]	Comprehensiveness	"It pretty well covers from the time you get tested to sharing it with, you know, people in your family that need to know that…I think it included everything". "I think everything you guys have got is good. It's not threatening, it's not too much, it's not too fast. It's simplistic, which you want because it's a scary conversation, but it's got meat, it's not light weight it's not trivial."
	Concise, clear, and plain language	"They [family sharing letter and email] were plain English, but they're not dumbed down too much, but they're not too high of thinking either… They're articulate and deliver the message pretty cleanly. I think brevity is important too."
	Optional content	"I like that they [the learn more buttons in the homepage video] are there, because there's a lot that goes into what's been discussed."
	Printable materials	"I love that you are giving options to print information."
	Patient experiences	"These [the others' experiences sharing genetic test results] are really good validations for people." "I could relate to some of them [other's experiences sharing genetic test results], it was like 'yes, I know how you feel'… It would have been good to hear that when I was first sharing."
	Autonomy supportive and positive tone	"It's [planning guide for sharing with family] just saying 'ideas', it's not telling somebody 'here's what you need to do'. I always think that's helpful." "I like that everything is set in a positive tone, even the responses coming back [in the sharing your genetic test results video]. Setting a positive tone neutralizes the situation."
Navigation [b]	Outline/organization	"I do really like [the video outline]… Because if you don't want to listen to 'How to Navigate this video', you can jump… or go back to something." "I think it's [the website] user friendly, I like the breakdown of the information."
	Ease of navigation	"The way it's [the website] laid out, it seems like it should be very intuitive… They click on the bubble, very simple for someone to navigate"
Visuals/Audio [c]	Visuals	"The graphics are nice and friendly" "All the icons are really good. There's diversity, age differences…"
	Color scheme	"I'm glad that you didn't make it [how genetic testing may help you handout] all pink, because we need the guys to get in on this, cause a lot of men do pass down the gene." "I like the fact that it's [gene flyer] not too bright, but it's just enough to keep my attention."
	Cartoon characters	"I appreciate how many different life scenarios you represented in those people [in the sharing your genetic test results video]… That was significant to get that diversity thing going on there."
	Audio	"The person speaking [in the homepage video]… is very clear to understand. Very slow talking."
Usefulness [d]	Planning/preparing	"I did not expect the [family member] reactions I got, so I'm glad you say that it can be hard and something people need to prepare for." "I wish this [ways you can respond to family member reactions print-out] was available when I was diagnosed! I do like how it gives people a way out, options to deal with family members." "Some people are list makers, and the idea of having a chart [planning guide for sharing with family] would be very helpful, or you might think of a relative you haven't thought about in a while."
	Would have made sharing easier	"I think it [how genetic testing may help you handout] would make it easy [to share] especially with people you don't know well to slip it in an envelope and print out a generic letter… I know some people don't want to talk about it for a while, and if that's the case, this would make it easy." "I had one particular relative who was resistant to get tested. So I think if I had had some of those words [others' experiences sharing genetic test results video], maybe I would have been more persuasive sooner."
	Changed understanding	"… I didn't think about, you know, family that I don't have a lot of contact with. And that [planning guide for sharing with family] would be great to write down the information with everything and just kinda disperse it and let them decide if that's something they want to do." "…like I said, until I watched [the homepage video], it never crossed my mind for my brothers, but it would probably be good for them too."

[a] Content is feedback about the amount, clarity, and ease of understanding of educational information given verbally (in videos) or through written text of materials.
[b] Navigation is how the participant moves throughout the video including navigation instructions and video features.
[c] Visuals and Audio is feedback about the color, visuals, graphics, and sound throughout the video.
[d] Usefulness for Preparing is feedback on how helpful the materials would be when preparing to share information with family, Feedback on family sharing materials.

Although the focus of this manuscript is on our development and feasibility phases, we considered issues related to future implementation phases by ensuring the intervention would be easy to scale up at minimal cost. We also recognized that the intervention may not work for some patients and have begun to consider ways to tailor or alter delivery of the intervention components to increase our ability to reach more patients. For example, if people do not login to the website, we may need a more direct way to send them various materials that align with their needs and/or barriers. Alternatively, some individuals may require a more resource intensive intervention. After we conduct the RCT, we will use the data to evaluate how the intervention may interact with individual characteristics or contextual factors to better understand how to adapt or revise intervention components or delivery methods to better meet individual needs.

Congruent with the FDECI, our ongoing evaluation phase is testing intervention effectiveness. We will also use the RCT data to test our 'programme theory' using an innovative application of a relatively new methodology called coincidence analysis (CNA) that we discuss in our innovation section. Our latest iteration of the intervention will not change during our ongoing evaluation phase. However, we will use our 'programme theory' and information about participants for whom the intervention is and is not effective to develop and pilot an adaptive or stepped intervention [57,103,104].

4.1. Innovation

There are several innovative aspects to our multi-faceted intervention and methods. First, unlike most existing FC interventions, we developed our 'programme theory' before evaluating intervention effectiveness; and we plan to use results from the evaluation phase to test our 'programme theory' and intervention. We will then modify and improve these or create additional components to be implemented among those for whom the current intervention is ineffective. Second, study participants in the RCT are given a higher level of flexibility than they have in other FC interventions in terms of what information/resources they access for themselves and what information/resources they share with family members. Thus, we are capturing data on which of the intervention components each participant clicks on and downloads, as well as asking what resources they gave to family members. Third, data will be used to conduct CNA to determine who accesses which intervention materials, how accessing or utilizing certain materials impact various theoretical factors related to capability, opportunity, or

motivation and whether these in turn alter the behavioral outcome. CNA is a method of causal modeling that can uncover complexities inherent to the intervention and individual differences across participants or context; this includes the ability to find underlying causal chains if they are supported by the data. CNA will test hypothesized causal mechanisms by which the complex intervention leads to changes in behavior. CNA has recently been used by the first author to identify factors that differentiated between participants for whom a different, more resource intensive FC intervention was effective and those who did not achieve the behavioral outcome [94]. To our knowledge, CNA has not yet been used within the FDECI as a method to empirically confirm 'programme theory', gain a better understanding of how and why a complex intervention is or is not effective, and ultimately contribute to the modification or future development of FC interventions and 'programme theories' upon which they are based.

4.2. Conclusion

We have described the formulation and refinement of our FC intervention, which serves as an example of methods and results from the development and feasibility phases of the FDECI. We also illustrate how these phases connect with the evaluation and implementation phases. Our application of the FDECI and CNA may aid other researchers who seek to better understand how and under what circumstances an intervention is effective.

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Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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