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Communication about Genetic Cancer Risk to Offspring: a Systematic Review of Children's Perspective

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FAMILY COMMUNICATION ABOUT GENETIC CANCER RISK: A SYSTEMATIC REVIEW OF CHILDREN'S PERSPECTIVE

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Resumo

O presente trabalho consiste numa revisão sistemática da literatura que pretende dar voz às crianças que cresceram em famílias afetadas por uma síndrome de cancro genético. O objetivo é sintetizar a investigação que dá a conhecer as experiências e preferências de crianças e jovens sobre a comunicação familiar acerca do risco do cancro genético.

De acordo com as diretrizes do *Preferred Reporting Items for Systematic Reviews and Meta-Analyses* (PRISMA), depois do registo no PROSPERO (CRD42021229926), foram realizadas pesquisas nas bases de dados MEDLINE e EBSCO, tendo sido identificados e selecionados quinze estudos que cumpriam os critérios de inclusão. Os resultados responderam às questões (1) como, quando e o quê é discutido na família sobre risco de cancro genético, (2) como é que a comunicação familiar sobre o risco de cancro genético impacta as crianças em termos psicossociais e comportamentais, e (3) quais são as preferências das crianças relativamente à comunicação familiar do risco de cancro genético.

Os principais temas investigados nos estudos incluídos focaram-se na perceção de risco e na compreensão do conteúdo e método de divulgação. Mais especificamente, exploraram as perspetivas, a compreensão, expectativas e implicações psicossociais do teste genético e respetivos resultados. Os resultados mostraram que a divulgação é feita maioritariamente pelos pais, ou apenas pela mãe. As crianças indicaram concordar com isso. Contudo, também sugerem que podia ser útil encontrarem-se com um profissional. A maior parte das crianças considera que poderia ter sido útil receber materiais escritos com informação sobre o risco genético de cancro. Sentimentos como estar assustado ou perturbado, surpreso com a informação, preocupado com o risco aumentado de receber um diagnóstico de cancro, sentir medo ou infelicidade à volta do evento, foram descritos na maior parte dos estudos incluídos.

Esta revisão identificou poucos dados quantitativos. Para além disso, os estudos incluídos continham maioritariamente amostras de adultos, revelando uma escassez de estudos que incluam a perspetiva das crianças. As crianças querem envolver-se no aconselhamento genético e fazer parte do processo de tomada de decisão associado à realização do teste genético.

Palavras-chave: Crianças; Família; Cancro Genético; Síndrome de Cancro Genético; Comunicação

Abstract

The present work aims to explore the children's perspectives on family communication about genetic cancer risk. This systematic review of the literature identifies and critically reflects on the research evidence and literature available on this topic. The purpose is to gather research that listens to the voice of children growing up in a family affected by a genetic cancer syndrome.

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement guidelines were used to conduct the review process. After PROSPERO registration (CRD42021229926), searches on MEDLINE and EBSCO were undertaken, and studies were identified using strict criteria. Fifteen studies met the inclusion criteria. The findings informed (1) how, when and what is discussed about genetic cancer risk in the family, (2) how does family communication about genetic cancer risk impact children on psychosocial and behavioral outcomes, and (3) what are the child's preferences regarding genetic cancer risk communication within the family.

The main topics investigated by the included studies focused on risk perception and understanding of the content and method of disclosure. More specifically, the review explored perspectives, understanding, expectations, and psychosocial implications of genetic testing and results. Findings showed that disclosure is done mostly by both parents or mothers only. Children reported agreeing with parents talking to their kids about genetic risk and of them having the role to disclose the genetic risk to them. However, they also suggest that it would be helpful to meet with a genetic counselor. Among offspring who did not receive written materials, the majority felt that it could have been helpful, either for themselves or for others. Feelings, such as being frightened or disturbed, surprised with the information, concern about the increased risk of receiving a cancer diagnosis, feeling scared or unhappy surrounding the event, were described in the majority of the studies.

This review identified little quantitative data. Also, the studies' samples mostly included adults, which indicates the lack of research focused primarily on children's perspective about this topic. Children want to be involved in genetic counseling and be part of the decision-making process.

Keywords: Children; Family; Genetic Cancer; Genetic Cancer Syndromes; Communication

Résumé

Le présent travail vise à explorer le point de vue de l'enfant sur la communication familiale, sur le risque de cancer génétique. L'objectif est de rassembler des études qui prennent en compte la voix des enfants qui ont grandi dans des familles touchées par un syndrome de cancer génétique.

Les lignes directrices PRISMA (*Preferred Reporting Items for Systematic Reviews and Meta-Analyses*) ont été utilisées pour guider les processus. Après l'enregistrement auprès de PROSPERO (CRD42021229926), des recherches ont été menées dans les bases de données MEDLINE et EBSCO, et les études ont été identifiées à l'aide de critères stricts. Quinze études répondaient aux critères d'inclusion. Les résultats ont répondu aux questions (1) comment, quand et ce qui est discuté dans la famille sur le risque de cancer génétique, (2) comment la communication familiale sur le risque de cancer génétique affecte les enfants en termes psychosociaux et comportementaux, et (3) quelles sont les préférences des enfants en ce qui concerne la communication familiale du risque de cancer génétique.

Les principaux thèmes étudiés dans les études sélectionnées portent sur la perception du risque et la compréhension du contenu et de la méthode de diffusion. Plus précisément, ils ont exploré les perspectives, la compréhension, les attentes et les implications psychosociales des tests génétiques et des résultats. Les résultats ont montré que la divulgation est faite principalement par les parents ou seulement par la mère. Les enfants ont indiqué qu'ils étaient d'accord avec cela. Cependant, ils suggèrent également qu'il pourrait être utile de rencontrer un professionnel. Parmi les descendants qui n'avaient pas de documents écrits, la plupart estimaient qu'il aurait également été utile de les recevoir, que ce soit pour eux-mêmes ou pour les autres. Des sentiments tels que la peur ou le fait d'être perturbé, d'être surpris par l'information, l'inquiétude face au risque accru de recevoir un diagnostic de cancer, le sentiment de peur ou de malheur autour de l'événement, ont été décrits dans la plupart des études sélectionnées.

Cet examen n'a permis d'identifier que peu de données quantitatives. En outre, la population étudiée comprenait principalement des enfants et des adultes, ce qui indique peu d'études du point de vue de l'enfant sur ce sujet. Les enfants veulent s'impliquer dans le conseil génétique et faire partie du processus de prise de décision.

Mots-clés: Enfants; Famille; Cancer Génétique; Syndrome du Cancer Génétique; La Communication

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List of Abbreviations

FAP	Familial Adenomatous Polyposis
FSGI	Family Systems Genetic Illness
GC	Genetic Counseling
GRI	Genetic risk information
HBOC	Hereditary Breast and Ovarian Cancer
HDGG	Hereditary Diffuse Gastric Cancer
IGC	Inherited genetic conditions
LFS	Li-Fraumeni syndrome
MFDG	Multi-family discussion groups
MMAT	Mixed Methods Appraisal Tool
PPI	Patient and Public Involvement
VUS	Variance of unknown or uncertain significance

Introduction

1. Genetic Cancer Risk

Genetic information and family history provide means to identify people with inherited cancer predisposition (National Cancer Institute, 2020). The risk assessment is based on medical history and the pattern of disease in the family. Genetic susceptibility tests for hereditary cancer syndromes can identify specific mutations associated with specific syndromes. These tests can also tell if a family member has inherited the same mutation as other members who carry a cancer-associated mutation. However, not everyone who has the mutation will necessarily develop cancer, even if the cancer-predisposing mutation is present in the family (National Cancer Institute, 2017).

Testing can indicate the presence or not of a mutation, although sometimes gives no clear answers. Withal, genetic counselors interpret and explain the test results and what they might mean to the applicant and the family. If a mutation is found, the counselor will talk to the applicant about which family members might also be affected. A positive result means the presence of a mutated gene (or genes) that place the person at increased risk for cancer. A negative result means the person doesn't have the gene mutation that was tested for. However, many genes can have more than one kind of mutation that can result in higher cancer risk, and the test result may be a "false negative", meaning that the test reads negative but the mutation is present (American Cancer Society, 2020). The test result can also be inconclusive when there is not enough information to evaluate whether the genes changes are present. Lastly, the test result can be a variance of unknown or uncertain significance (VUS), when the test shows that a person has an unusual form of a gene that is as of yet not associated with certain or specific knowledge (American Cancer Society, 2020).

According to the American Cancer Society (2020), in the presence of a mutation in either the BRCA1 or BRCA2 gene, the individual is at high risk for breast and ovarian cancer. Men with BRCA1 and BRCA2 variants are also at increased risk of prostate cancer

(National Cancer Institute, 2020). Lynch syndrome, also called hereditary non-polyposis colorectal cancer, can be caused by a mutation in several mismatch repair genes, including MLH1, MSH2, MSH6, PMS1, and PMS2. Inherited mutations in the TP53 gene or the CHEK2 gene, tumor suppressors genes, causes Li-Fraumeni syndrome (LFS). Genetic mutations in the APC gene can cause different syndromes, including Familial Adenomatous Polyposis (FAP; Cancer.Net, 2020). If tests positive for one of these family cancer syndromes, their close relatives (parents, siblings, and children) have a 50% chance of having a mutation, too (American Cancer Society, 2020).

If a mutation is found, the counselor will talk to the applicant about which family members might also be affected. In genetic counseling (GC), the patient is helped to understand how heredity contributes to the disease and the risk of recurrence in specific family members. Following being provided with information about the disease associated risk for, the patient is made aware of the available options to control associated risks. For example, lifestyle changes, early detection, watching for signs and symptoms of cancer, medicines to reduce cancer risk, and preventive surgery are explored (American Cancer Society, 2020) tailored to the person/family. Therefore, GC attempts to make the best possible adjustment to the disorder in an affected family member and/or to the risk of recurrence (Fraser, 1974).

When a person is affected by a particular genetic disorder, it does not only affect one member, but rather the entire family. A genetic disorder can run in more than one person in the family, such as in the case for autosomal dominant genes involved in cancer genetics, and therefore be inherited and passed down from parent to child. Once genetic risks are distributed among family networks, the moment of becoming aware of the increased susceptibility to cancer is considered as a very disturbing event, both at personal and family levels (Evers-Kiebooms et al., 2000).

The moment of taking the genetic test and/or receiving information about its result is very shocking for some. The notion of positive testing can trigger a psychological response, in a similar fashion to that of receiving news about a diagnosis of cancer itself, and it can involve long periods of waiting between the test and the potential diagnosis (Pasacreta, 2003). The individual and the family will be differently affected by the condition, in terms of psychosocial challenges. One model that explains how these genomic conditions affect the family including the family individually responses is the

Family Systems Genetic Illness model (FSGI; Rolland & Williams, 2005). This model uses a schedule of genetic disease, to describe how each phase is associated with specific psychosocial challenges. The FSGI comprises the pre-symptomatic phases of genetic syndromes in the context of GC. The concept of phases over time in this model enables a longitudinal examination over the course of the disease in terms of it involving a process of transitions and changes, requiring the adaptation of the family (Rolland & Williams, 2005).

Growing up in a family with genetic cancer risk can involve additional psychosocial burden and be challenging. For instance, when a child is affected by an inherited genetic condition (IGC), unaffected siblings can subjugate their feelings and needs and may isolate themselves when coping with it (Plumridge et al., 2011). Thus, the impact of genetic cancer risk information on children, affected or not affected, should not be underestimated. However, this is an under-considered clinic area, and there is a call for family communication research that sheds light on what does it mean to live in a family affected by a genetic condition (Metcalfe et al., 2008).

2. Family Communication about Genetic Cancer Risk

The definition of family used here is the same used in the study by Metcalfe et al. (2008, p. 1196) where family is defined as: "any group of individuals united by the legal ties of marriage or partnership, blood or adoption in which the people are committed to one another in an intimate interpersonal relationship where the members see their individual identities as importantly attached to the group they call 'family' which has an identity in its own right through a shared history and shared future, and the adult(s) cooperate emotionally and financially to support dependent individuals (and each other)".

According to a review, the major factors that influence (either motivating or inhibiting) communication of genetic risk within families are the perceived responsibility to tell, relationship type and quality, deciding who to tell, anticipation of relative's reactions, mutation status, and personal feelings (Wiseman et al., 2010). A review of the reviews identified barriers to communication, such as the complexity of risk information and the uncertainty associated with a genetic cancer test, the family culture, history and

myths about the disease, the proximity of the family (not feeling emotionally close posing difficulty to establish contact), and reconstituted families, for instance, where tensions, divorce, separation, or adoption exist (Rodrigues, 2020). The identified facilitators of communication were family cohesion and the perception of being emotionally close to family members, having a family-oriented culture, and having a history of cancer in the family (Rodrigues, 2020).

Research on adjustment to cancer risk mainly focuses on family members who are directly affected with the disease and the communication between this identified person with other family members. When it comes to the mutation carrier, in relation to perceived control within a family genetic health communication context, the sharing of information and coping with genetic information can contribute to an increase sense of personal perceived control (Biesecker, 2001; Pasacreta, 2003; Rolland et al., 2005; Macrae et al., 2013; Madlensky et al., 2017). When it comes to children, cancer genetic health communication was also found to be associated with perceptions of having control about their own health and the surrounding circumstances (Tercyak et al., 2019). Therefore, studies suggest that family communication about genetic cancer risk can positively impact the experience of parents and children. The perceived controllability in managing the disease and its risk can be a crucial dimension in determining the potential adjustment of a family member. Among children, the relationship between cancer genetic health communication and psychological adjustment is moderated by several factors, one of them being the nature of the information being communicated (Tercyak et al., 2019).

Specific content is very important when it comes to communicating health information. For optimal use and to support appropriate decisions and manage information, people need to access it first, process and understand what it means. This capacity is called health literacy (American Public Health Association, 2020), and it is crucial in the consideration of transmitting information concerning genetic syndromes. Genetic literacy, as a subset of health literacy, can be the individual's capacity of using personal genetic information effectively (Erby et al., 2008). When being confronted with personal genetic information, a person attributes meaning to it in terms of a perceived risk for the condition. How a person perceives their risk for a disease is important as it influences behavior, decision-making, and the emotional outcome after testing, even more so than the actual or scientific risk level, in terms of psychosocial aspects of genetic testing (Lerman et al.,

2002). As McDaniel (2005) asserts, it is the meaning that people give to the information that affects the response, not the data itself.

If individuals do the genetic test to aggregate more information, not only for themselves but for other family members too, they are more likely to engage in family communication (Seymour et al., 2010). Studies about hereditary nonpolyposis colorectal cancer (or Lynch syndrome) and hereditary breast and ovarian cancer (HBOC) families found that family members generally are willing to, and do share genetic information with other members, despite concerns and barriers (Pentz et al., 2005; Hallowell et al., 2003; Green et al., 1997). Genetic test results are perceived as being more relevant to some family members than others, based on variables such as gender (for example, for BRCA1/2, information can be perceived as more relevant to female than male relatives), age, relative closeness/relationship (for example, cousins, siblings) and anticipated reactions or perceived receptivity of receiving genetic information of those family members (Seymour et al., 2010).

Families affected by an IGC face the task of communicating genetic risk information (GRI) to their children. Several studies expressed that it should be the parent who primarily is responsible for discussing family genetic information with their children (Metcalfe et al., 2008). Parents showed difficulties with what and when to tell their children, and they recognize that they have different concerns and questions depending on their children' ages. Despite emphasizing the importance of open communication, parents and their children found talking about the condition and sharing risk information difficult, and that openness did not lessen the psychological and emotional pain of living with the condition. However, families can feel empowered when they openly talk about the condition, which can enable individuals to discuss matters and concerns as they arise, and increase their support and care for each other (Metcalfe et al., 2008). Regarding children's experience, mothers report that open communication did not affect children's general behavior or well-being (Metcalfe et al., 2008). In families where the communication was more closed, Metcalfe et al. (2008) found that children felt upset and frustrated with family secrecy, even when the illness was discussed, which resulted in tense relationships between family members. According to a meta-synthesis of the research, if open communication exists, young people may mature into adulthood with a cautious mind regarding reproduction and genetic testing decisions, which can affect their own choices and psychological health. On the contrary, poor communication led to poorer choices and

emotionally-driven decision-making (Metcalfe et al., 2008). Providing information, checking understanding along the way, and explaining and managing the emotional feelings that arise seems to be integral to support children's coping with GRI (Metcalfe et al., 2008).

It has been shown that mothers who test positive for hereditary breast/ovarian cancer risk, as family communicators, want to educate and counsel their children about HBOC as much as possible, even without the benefit of well-developed patient education and counseling resources (Tercyak et al., 2019). However, genetic information is complex, challenging to communicate, and it is expected that the health and genetic literacy of a family communicator will be reflected on children. Tercyak et al. (2019) found that for children, what moderates the relationship between psychological adjustment with genetic cancer health communication is the nature of the information being communicated. For example, in the case when mothers receive uninformative test results, elements of confusion have been expressed in reports by children (Tercyak et al., 2019). Thus, communication affects how information is received and has implications for psychological adjustment for the child (Tercyak et al., 2019).

A meta-synthesis on family communication concluded that many parents need more help and support to assist them in successful communication with their children about genetic conditions (Metcalfe et al., 2008, p.1198).

3. Study Purpose and Aims

The majority of research to date has relied on parental reports and investigated parental factors or the nature of genetic information itself to examine their role in how communication of genetic information is received by, or impacts the psychological well-being of children. Further research is needed with a focus on the child's perspective. Research has shown that regarding children's understanding, they were often more cognizant than their parents anticipated. For example, adolescent girls emphasized more than their parents the potential psychological risks of carrier testing if undertaken at a young age (Metcalfe et al., 2008). Accessing the child's perspective will provide important knowledge about the impact of communication of genetic information and further our

understanding of what specific areas of information should be communicated and inform how best to go about it.

This paper presents a systematic review of the existing research on the impact of communication about genetic cancer risk within the family, from the point of view of the child. A focus on the child's perspective is unique, as most of the research and prior reviews have relied on parental reports or retrospective accounts. Previous systematic reviews and meta-synthesis (Seymour et al., 2010; Metcalfe et al., 2008) on family communication explain what facilitates or impedes family communication (Seymour et al., 2010) and the issues surrounding family communication about inherited genetic risk (Metcalfe et al., 2008). These reviews are innovative because they explore communication to at-risk relatives for hereditary adult-onset cancers and the support required for parents and children through the genetic condition or the risk. However, the compilation of research on the child's experience and perspective is still lacking in the literature. Most of the conclusions are based on parental reports with their perspectives on their children's experience of family communication, with a paucity of conclusive evidence from the voice of the child him/herself.

This study intends to contribute to the existent research on GC and cancer genetic health communication, in general, and the children's perspective and needs, in particular, by providing the first systematic review of the literature focusing on children's perspective, impact and needs, inside a family that is affected by genetic cancer risk. The purpose is to gather research that listens to the voice of children growing up in a family affected by a genetic cancer syndrome. Specifically, from the children's perspective:

1. How, when and what is discussed about genetic cancer risk in the family?
2. How does family communication about genetic cancer risk impact children on psychosocial and behavioral outcomes?
3. What are the child's preferences regarding genetic cancer risk communication within the family?

Having access to this type of information, once disseminated, will provide evidence to guide both GC professionals and parents and can inform the development and testing of interventions to enhance and support communication of genetic information to children.

Methods

We used the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement guidelines (Moher et al., 2009) to conduct the review process. We registered the protocol on PROSPERO (CRD42021229926, at https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42021229926).

1. Database search procedure

Two independent researchers used Medline, EBSCO and PsycINFO electronic databases, using search terms chosen in collaboration with the team of authors. Search terms were informed in part by the Metcalfe et al. (2008) and Seymour et al. (2010) systematic reviews, and then adapted for the present investigation. The following search terms were adapted for each database: children, family, genetic cancer, genetic cancer syndromes (breast and ovarian cancer, familial adenomatous polyposis (FAP), Lynch syndrome, Cowden syndrome, Li-Fraumeni syndrome, hereditary diffuse gastric cancer (HDGC), retinoblastoma), communication and outcomes (see, **Appendix A**).

Databases were searched for studies published between 1990, when the BRCA1 gene was identified (Jeff M. Hall et al., 1990; Journal of Clinical Investigation, 2014), and 2020. Forward and backwards citation searches were undertaken on Web of Science and Scopus for included papers. In addition, relevant journals and authors were hand-searched to identify additional relevant articles that could be missed by the search strategy.

Search results were exported to a reference manager, EndNote, and both digitally and manually identified duplicates were discarded. Resulting titles and abstracts were transferred into Excel and reviewed for relevance, and the full-text articles were reviewed for inclusion and data extraction.

2. Article selection: inclusion criteria

Two authors independently screened study titles and abstracts. If included, the article underwent a full-text review. The inclusion and exclusion criteria used for screening the articles were based on the population, concept, and context of the research question (see Table 1).

Studies were included if they focused on family communication from the perspective of the child about genetic cancer risk. Children as minors (< 18 years) or children in retrospective (adults referring to their experience when minors) were the populations included. In studies where children are not the only population target, but rather the whole family was studied, only data pertaining to the children's reports were extracted. Other related studies, such as studies on the development of decision aids, tools, correlational studies, etc., were also taken into consideration. Regarding study design, qualitative and quantitative studies were included.

When referring to genetic cancer risk families, the considered syndromes included breast and ovarian cancer, FAP, Lynch syndrome, Cowden syndrome, LFS, HDGC, and retinoblastoma.

	Include	Exclude
Population	• Children as minors – affected and siblings (< 18 years old). • Genetic Cancer Risk Families (syndromes: breast and ovarian cancer, FAP, Lynch syndrome, Cowden syndrome, LFS, HDGC, retinoblastoma).	• Only parents referring to their children. • Only health professionals' views. • Studies that include offspring > 18 years old.
Concept	• Family communication. • Perspective, experience, and opinions on family communication about genetic cancer risk. • Family communication impact. • Familial discussion/disclosure.	• Only reports of parental perspectives on their children's experience and opinions of family communication about genetic cancer risk.

Table 1 – Inclusion and exclusion criteria used for screening articles.

	Include	Exclude
Context	• Qualitative and quantitative studies with any study design. • Trials. • Correlational studies. • Studies on development of or testing of decision aids. • Expected outcomes from literature about family communication: ○ Emotional reactions (e.g., anxiety, distress, depression). ○ The age at which children were told about the family genetic information and who told that. ○ Communication impact: emotional, behavioral outcomes, impacts on self-esteem, self-concept, self-conscious, identity, body image, body stigma, body, concerns, psychological adjustment, psychological development, vulnerability, arm concerns, well-being, quality of life. ○ Genetic counseling adherence behavior; health behaviors; sharing of information behavior.	• Literature reviews undertaken by other researchers. • Protocol papers.

Table 1 (continued) – Inclusion and exclusion criteria used for screening articles.

3. Paper screening and quality evaluation

All titles and abstracts of the identified studies were independently assessed by more than one author for inclusion or exclusion in the review, until a good level of agreement was reached (97%). Papers were excluded when all reviewers agreed that inclusion criteria were not met, and disagreements were resolved by discussion to reach consensus.

The eligible papers were assessed for their quality using the Mixed Methods Appraisal Tool (MMAT; Hong et al., 2018), which is suitable for five types of study designs: qualitative, randomized controlled trial, nonrandomized, quantitative descriptive, and mixed methods studies. The quality assessment was done by two independent judges. The judges responded "Yes", "No" or "Can't tell" to the screening questions according to the MMAT checklist, depending on the appropriate category of the study appraise.

4. Data extraction

Authors used a detailed spreadsheet to systematically extract data about study, sample, methodology, and outcomes. We identified 4464 abstracts and exported them to EndNote. After de-duplicating, two independent researchers screened the remaining 695 articles for title and abstract relevance. If an abstract fulfilled all inclusion criteria, then we extracted the full-text article. Eleven (1.6% of 695 eligible abstracts) studies met all the inclusion criteria, and four additional studies were identified through other sources. Table 2 summarizes the data extraction procedure. When there was disagreement, the two reviewers consulted a third reviewer to determine article suitability.

5. Data collection procedure

We used tables to extract information from the included articles. In summarizing empirical study data, we recorded the study outcomes, study design and the main findings. We categorized the data according to whether the study pertained information to respond

to the first, the second or the third main question, each of them divided into subtopics (**Appendix B**). We then compiled all the information for each topic and summarized the results according to the research questions they addressed (**Appendix C**).

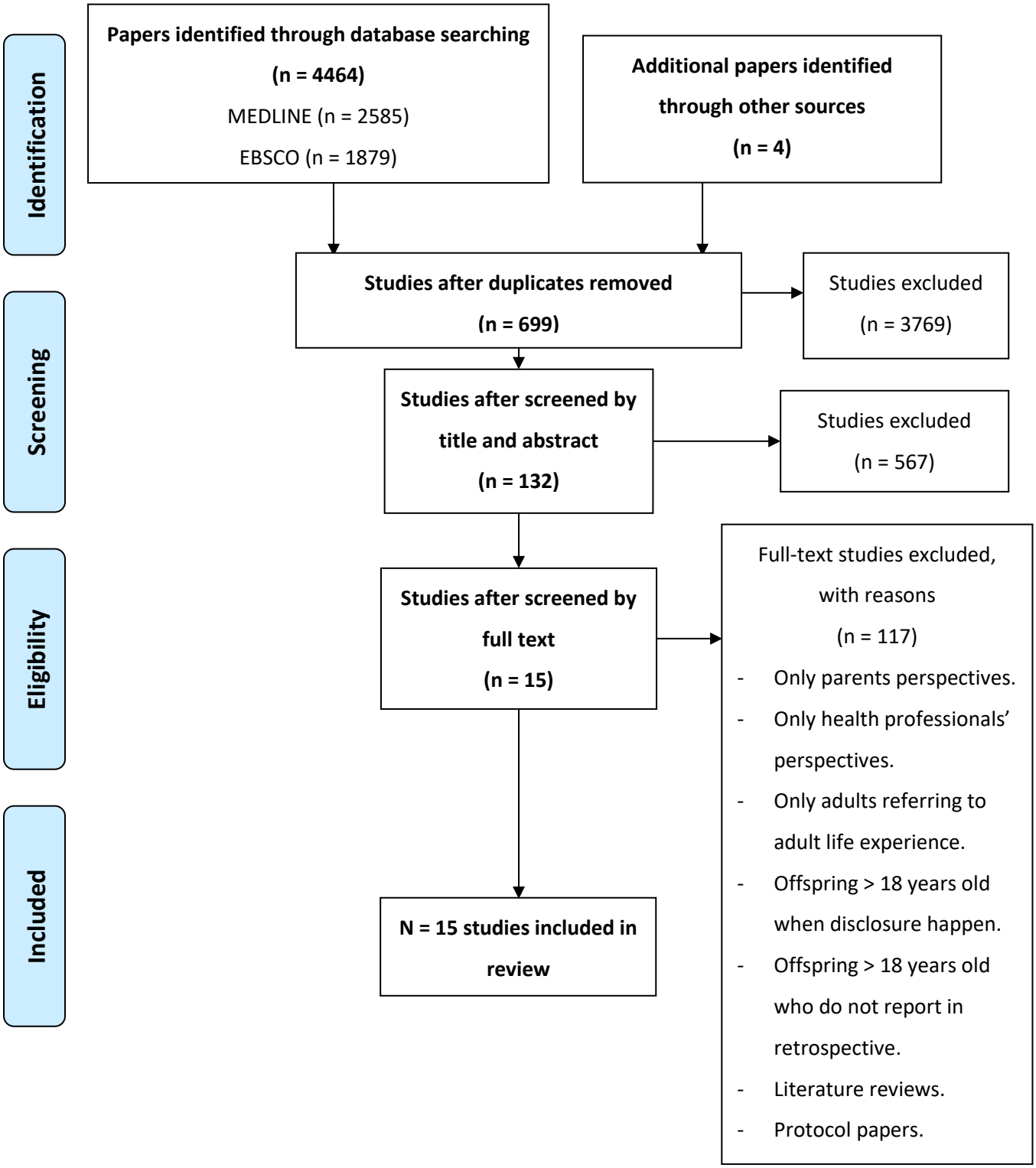


Table 2 – PRISMA flow diagram of study identification and selection process.

Results

1. Available literature

The search returned 4464 articles (2585 results from MEDLINE and 1879 from EBSCO). Endnote removed duplicates (3695 articles) for a total of 769 articles. Four hand-searched articles were added. Then, articles were entered into Excel, where 74 duplicates were hand-removed, for a total of 699 articles. Of these, 567 were excluded, when doing title and abstract screening, for a total of 132 articles. Of these, 117 did not meet inclusion criteria, resulting in 15 articles (Table 1): 10 qualitative (66.67 %), and 5 mixed-method designs (33.33 %). These studies were conducted in the United States (n = 10, 66.67%), Australia (n = 2, 13.33%), United Kingdom (n = 2, 13.33%), and the Netherlands (n = 1, 6.67%). Studies included semi-structured interviews and questionnaires. One article involved an intervention with focus groups. The studies' samples included children and retrospective reporting of children of families at risk for breast and ovarian cancer (n = 10, 66.67%), Li-Fraumeni syndrome (n = 1, 6.67%), multiple syndromes (n = 2, 13.33%), or other syndromes (n = 2, 13.33%). Overall, the population of this study included 20% male children and 76.5% female children, or retrospective children (about 3.25% of the population was not described). Quantitative data were present in five (33.33%) mixed-methods studies. The variables measured in those five papers were: distress (60%), anxiety (60%), depression (60%), anger (40%), need for help (40%), satisfaction and experience with GC (40%), family influence, functioning and communication (40%), quality of life (40%), impact of genetic testing (with variables such as coping, distress, uncertainty, and positive experiences; 40%), cancer worries (40%), regret (20%), decisional conflict (20%), coping style (20%), risk perception (20%), health attitudes and beliefs (20%), and behavior problems (20%). Descriptive data are summarized in Table 3.

Study	Country	Data collection	Purpose	Sample	Family syndrome / at-risk syndrome
[Alderfer et al., 2017]	USA	Interviews	Exploring perspectives of genetic testing among adolescents and emerging adults in families with LFS, examining their understanding and expectations of genetic testing and the psychosocial implications of the testing and results.	Children AND Retrospective children.	Li-Fraumeni syndrome (LFS)
[Bakos et al., 2008]	USA	Interviews	Exploring cancer risk perception and the experience of being a mutation-negative woman within a known BRCA1⁄2 mutation-positive family.	Retrospective children (adults referring to their experiences at < 18 years old)	Breast and ovarian cancer
[Bradbury et al., 2008]	USA	Interviews	Exploring the opinions of BRCA mutation carriers regarding the genetic testing of minors and the cognitive and affective processes underlying these opinions.	Mix sample (children AND adults AND/OR families)	Breast and ovarian cancer
[Bradbury et al., 2009]	USA	Interviews	Exploring offspring perspective of the content and method of disclosure from parents, their understanding and perceptions of hereditary risk and the psychosocial and health-related impact of this communication.	Retrospective children (adults referring to their experiences at < 18 years old)	Breast and ovarian cancer

Table 3 – Summary of Descriptive Data for Studies Included.

Study	Country	Data collection	Purpose	Sample	Family syndrome / at-risk syndrome
[Eisler et al., 2016]	UK	Multi-family discussion groups (MFDG)	Exploring if a MFDG was a suitable approach, and acceptable to both families (parents, children, and young people) affected by IGCs and to genetic counselors; exploratory co-designing of an intervention, involving families and genetic counselors, to meet the needs of families and genetic counselors; and the training of genetic counselors in the intervention's delivery.	Mix sample (children AND adults AND/OR families)	Familial adenomatous polyposis and other inherited genetic conditions (IGC)
[McGill et al., 2019]	Australia	Interviews and questionnaires	Exploring the perspectives and emotional reactions of parents and young adults about cancer-related genetic counseling and testing offered to children in the family.	Mix sample (children AND adults AND/OR families)	Cancer-related genetic mutations
[Norris et al., 2009]	USA	Interviews	Exploring an intergenerational perspective on families' stories to highlight aspects of the family context influencing on communication of risk and decision making about testing and follow-up.	Mix sample (children AND adults AND/OR families)	Breast and ovarian cancer
[Patenaude et al., 2013]	USA	Interviews and questionnaires	Determining what daughters, aged 18–24 years, of BRCA1/2 mutation carriers understand about their 50% chance of carrying a BRCA1/2 mutation and about risk reduction or management options for mutation carriers, the extent and nature of daughters' cancer-related distress, and the effects of knowing mother's mutation status on daughters' future plans.	Retrospective children (adults referring to their experiences at < 18 years old)	Breast and ovarian cancer

Table 3 (continued) – Summary of Descriptive Data for Studies Included.

Study	Country	Data collection	Purpose	Sample	Family syndrome / at-risk syndrome
[Rowland et al., 2016]	UK	Interviews	Exploring what are the experiences of parents, children and young people when discussing genetic risk in families affected by or at risk from a BRCA gene mutation, and how does the information shared impact on children and young people's views about their future risk.	Mix sample (children AND adults AND/OR families)	Breast and ovarian cancer
[Sie et al., 2013]	Netherlands	Questionnaires and open-ended questions.	Exploring whether the testing age should be above 25 years in order to prevent adverse effects such as regret or decisional conflict, by determining the percentage and characteristics of patients reporting such problems in retrospect.	Retrospective children (adults referring to their experiences at < 18 years old)	Breast and ovarian cancer and Lynch syndrome
[Stuttgen et al., 2020]	USA	Interviews	Exploring how minors understand GRI, the disclosure event and what information was given to the minor, and to evaluate how parents' perspectives differ from minors in order to understand factors associated with positive and negative experiences for both parents and minors in the communication of GRI.	Mix sample (children AND adults AND/OR families)	Breast and ovarian cancer, Lynch syndrome, and Huntington's disease
[Tercyak et al., 2001]	USA	Questionnaires and open questions	Exploring conceptions of health, cancer risk, and psychological adjustment among children in families suggestive of carrying BRCA1/2 susceptibility genes.	Only Children (0 - 18 years old)	Breast and ovarian cancer

Table 3 (continued) – Summary of Descriptive Data for Studies Included.

Study	Country	Data collection	Purpose	Sample	Family syndrome / at-risk syndrome
[Tercyak et al., 2019]	USA	Interviews	Exploring early literacy skills and communication about genetics in families, by investigating adolescent and young adult children's psychosocial adjustment following disclosure of their mother's BRCA test results.	Only Children (< 18 years old)	Breast and ovarian cancer
[Werner-Lin et al., 2012]	USA	Interviews	Exploring a patient-centered perspective on the dilemmas faced by 18–24-year-olds considering genetic education, counseling, and testing to identify a BRCA1/2 gene mutation.	Retrospective children (adults referring to their experiences at < 18 years old)	Breast and ovarian cancer
[Young et al., 2019]	Australia	Questionnaires and interviews	Exploring how young adults and their families communicate about a BRCA1/2 pathogenic variant, from a family systems perspective.	Mix sample (children AND adults AND/OR families)	Breast and ovarian cancer

Table 3 (continued) – Summary of Descriptive Data for Studies Included.

Overall, papers typically investigated children's perceptions of cancer risk and their understanding of the content and method of disclosure. Central aims of the papers included to explore perspectives, understanding, expectations, and psychosocial implications of genetic testing and results. Other focus themes were the experience of testing negative within the context of a family that has received positive test results, the experience with genetic services, and "when" to discuss genetic risk in families. For the most part, studies aimed to explore the support needs of the participants, and the effects of the genetic cancer risk/at-risk information on children and young people's views about their future (Table 3).

2. How, when and what is discussed about genetic cancer risk in the family?

In terms of the "how", information regarding who did the disclosure of the genetic condition to the child was only present in six (40%) of the selected studies. Children indicate that they came to know about the genetic test result of their mother/father, through a parent, or by others, for example, a grandmother, aunt, sibling, in-laws, or a genetic counselor. Parents typically shared the test results alone or with their spouses. Daughters of mothers with BRCA1/2 were more likely to learn about the family risk from a mother alone, compared to patterns of disclosure of GRI to sons. In some cases, the offspring's siblings or other family members were present during disclosure. Some offspring reported learning the information by telephone or while participating in an activity with a parent, for example, while driving in the car with their mother (Patenaude et al., 2013):

> *"[My mother] sat me down and told me, 'I am not going to die like [my aunt, my mother's sister].' I was older and understood more, so it was OK because the cancer was so small and early."* – teenage daughter; on HBOC (Norris et al., 2009)

Overall, children reported experiences of open communication in conversation with parents and providers about testing results, risk management, hopes, and fears. However, there were reports about a truncated process of GC, meaning sessions were shorter because children did not actively participate. When there were joint GC sessions, children reported retrospectively a perception of guilt being experienced by their parents for passing the mutation on to them. With this experience, children reported tending to hide their worries, denying concerns, or refrained from asking critical questions about the implications of

their mutation status, their cancer risk, and about various methods of risk reduction, in an attempt to protect their parents from potential additional burden and emotional distress (Werner-Lin et al., 2012). Some young people expressed being aware that their mothers were putting on a "*brave face*". Sometimes, children were afraid that by asking questions they would upset their mothers (Rowland et al., 2016).

During the disclosure phase, five (33.33%) studies reported contact with GC. Only four (26.67%) studies referred to using tools to support the disclosure process. Tools described in the studies included written materials, professional journals, or the internet.

Among HBOC families who carried a BRCA1/2 gene mutation, this review showed that there was very little discussion about male breast cancer (e.g., Young et al., 2019). Mother's communication on HBOC was described as balanced between the sharing of factual information and in aiming to not raise alarm, and by attempting to educate, counsel, and reassure (Tercyak et al., 2019).

In terms of the "when", the time interval between the parent receiving the test result and disclosure to offspring can vary in hours to a few days after the family member received the result (Patenaude et al., 2013), or years (Norris et al., 2009; Bradbury et al., 2009). Some retrospective reporting by children indicated that they did not recall how long after the parent learned the test result, they were informed (Patenaude et al., 2013). The youngest age of an offspring learning of a genetic test result was reported as being 8 years old (Stuttgen et al., 2020).

In terms of the "what", in some cases, the specific genetic mutation itself was not described at the disclosure time, although the parents did discuss the hereditary risk of cancer in the family. The most frequently reported information when disclosing to children is content concerning the implications of the test result for the risk of developing cancer, and the ability for the offspring to have genetic testing. Risk reduction strategies (cancer screening, prophylactic surgeries, and avoiding tobacco) were less frequently described at the time of disclosure (Bradbury et al., 2009):

> *"My mom explained that she wasn't sick, but she was getting preventative surgeries so she doesn't get breast or ovarian cancer in the future. She was doing it for us because she wants to be around, unlike her mother who died of ovarian cancer."* - daughter, 18 years old, 11 years old at disclosure; on HBOC (Bradbury et al., 2009)

> *"He [dad] said that testing had been done on his side of the family and a gene was found that makes women prone to breast cancer. He had testing and then brought it up over dinner one evening. He basically said that he is a carrier and at increased risk for cancer in family. Also, that we could be carriers."* – son, 26 years old, 17 years old at disclosure; on HBOC (Bradbury et al., 2009)

One study about BRCA1/2 demonstrated how some parents' messages appeared to be open and empowering (e.g., "*It's your decision if you want testing", "Wants me to know the family history", "If you do get cancer, you can get through it", "You deserve to know"*). From the daughters' perceptions, some parents attempted to provide reassurance for their daughters (e.g., *Mother as role model, you can live with risk, dealing with the "beauty of the unknown"*, "*Don't be scared. It doesn't automatically mean you will get cancer", "(Dad:) Mom, everyone will be OK"*). Other messages, from the daughters' perspectives, suggested more directive communication efforts (e.g., "*She wants me to get testing", "She doesn't want me to get testing now", "Have a good doctor", "Live a healthy life. Be proactive"*). In some cases, parental fears were conveyed (e.g., "*Make sure you know what you need to do", "Have mammograms. Take care of ourselves", "Mother apologetic for passing on the mutation", "I want you to avoid having cancer as I have had"*; Patenaude et al., 2013).

Daughters of a parent with BRCA1/2 indicated that they understood that the information was of the highest importance. It was less clear whether the daughters understood how or when to act on the information, or whether they would just take parental advice around specific responses (Patenaude et al., 2013):

> *"But she [mother] had always pushed just how serious it was and that it was kind of a big deal... it kind of scared me. But she also — she wouldn't say it in a scary way- she would just sort of stress it and I sort of preferred not to think about it."* – daughter; on HBOC (Patenaude et al., 2013)

Studies show that the content of disclosure can vary depending on the age group of the children or the preventive/current health activities of the parent. For example, disclosure of the mother's BRCA1/2 mutation status to daughters was sometimes conflated with the news that the mother would shortly be undergoing preventive surgery (Patenaude et al., 2013). For children < 11 years old, information focused on events that the child

would witness (hospitalization). For younger age groups parents tended to use simplistic language to describe breast cancer: "*poorly boobs*", "*poorly tummy*", "*magic medicine*", "*new boobies*", "*boob job*" (Rowland et al., 2016). For the 12-14 years old age group, disclosure centered more on the hereditary nature of the gene mutation and parents' risk. For the 15-17 years old age group, young people were given more information about risk management strategies. And, when disclosing to offspring at 18 years old, discussions focused more on genetic testing and the implications to the young person's future offspring (Rowland et al., 2016).

3. How does family communication about genetic cancer risk impact children on psychosocial and behavioral outcomes?

3.1. Psychosocial data

As offspring reported on their experiences around the genetic cancer risk disclosure, they described emotional volatility reflected in either increasing or decreasing concern from the time of disclosure until the present (Patenaude et al., 2013). Feelings, such as being frightened or disturbed, surprised with the information, unhappy surrounding the event, or concerns about the increased risk of receiving a cancer diagnosis, were described in most of the studies.

> *"It didn't sink in at first, but once I started having to get all of the testing, it sunk in that I am going to have to deal with this for the rest of my life."* – 16 years old; on LFS (Alderfer et al., 2017)

Daughters reported fear and worry as peaking when they were told about their mother's positive mutation status, but described these feelings as diminishing over time as they acclimatized to the information and learned further about risk-reducing options. In contrast, young women said at the time they were told (in their teens) about a cancer genetic risk, the implications for themselves seemed distant. However, as daughters entered adulthood, fears about developing breast and ovarian cancer tended to increase (Patenaude et al., 2013):

> *"I was nervous for her [mother] more than myself right away and I was nervous for like a year after when my cousin had it and that just really made me think, like, wow, I could have it too."* – daughter; on HBOC (Patenaude et al., 2013)

This review found descriptions about feeling vulnerable as a child when knowing about the cancer risk because the clinical utility of screening and prevention options are not well defined for this younger age group. For example, one participant reported being denied screening for breast cancer because her breasts were considered to be dense at her age and was informed "that it would be a waste of time to even do it" (Werner-Lin et al., 2012). Retrospective reports by children that received low absolute HBOC risk estimates for their age during genetic testing, described that they felt pressured (some by their parents) to act before reaching a certain age they perceived as being unsafe (Werner-Lin et al., 2012). Retrospective reporting by some children indicated that they felt an urgency in wanting some form of risk management. Age often was experienced as a barrier to addressing cancer risk because children were told that they were too young for conventional screening tools (Werner-Lin et al., 2012). When expressing interest in screening or prophylaxis, children reported that professionals would dismiss their concerns as being premature and informed that they were ineligible for clinical trials (Werner-Lin et al., 2012).

Regarding family communication, children reported a multiplicity of positive, neutral, and negative responses to disclosure of genetic risk in the family. The nature of those responses differed depending on the nature of the results and was not necessarily associated with the child's developmental level. When mothers received negative test results, children reported feeling "relief" and being "glad" about the information, both in terms of their own health and in relation to the health of their mothers. In the opposite case of a positive test result, children also had favorable reactions because of the knowledge (health, treatment options, future plans) the genetic test result provides (Tercyak et al., 2019). Some negative responses to disclosure of genetic risk in the family included feeling apprehensive about their own future health and that of their mothers'. Some cases of dispassionate and neutral responses to the disclosure process were described as children simply "not surprised by the news" (Tercyak et al., 2019). When having siblings, children reported feelings that considered those relationships. For example, guilt may be

experienced when a close sibling tested positive; closeness of a sibling may affect how a child felt:

> *"A feeling of guilt that I don't have it. My brother and sister need to deal with this for the rest of their lives."* – retrospective child; on LFS (Alderfer et al., 2007)

> *"It probably brought me and my sister closer together because we now have something (testing positive) that we can closely relate to."* – 16 years old; on LFS (Alderfer et al., 2017)

Although the vast majority of children articulated thoughts and feelings about how they wanted parents to communicate over time with them about a genetic risk, children didn't always engage in family communication in follow-up with their own parents because they had difficulty articulating about their feelings. Some children reported that they did not address their own stress/anxiety, even when recognizing concerned others would support them. Younger children said that they wished to avoid negative feelings (Tercyak et al., 2019):

> *"And then when she [mother] got the testing, I was still, like I didn't like talking about it [BRCA] because I keep things to myself. I don't know whether it's, like, a sense of I need to not make people think that I'm scared of things. I don't know. I just don't, like, openly express my [fears]."* – daughter; on HBOC (Tercyak et al., 2019)

This review showed that children's psychological adjustment may be related to their beliefs about health and thoughts of illness. For example, children with more psychological symptoms experienced more frequent thoughts of becoming sick, thoughts about developing cancer, and worries about cancer developing in a family member (Tercyak et al., 2001).

Some studies indicated that testing and the results have had no impact on family relationships (Alderfer et al., 2017), within or outside their family (Bradbury et al., 2009). However, one study found sadness and social isolation could be triggered by watching older family members die from the same disease, creating a void related to familial mentoring relationships (Bakos et al., 2008):

"...I didn't have a lot of women in my lifetime, family members in my lifetime that I was talking to about [cancer]. I wished I had a woman there I could talk with but that just wasn't the case. I mean I had girlfriends and stuff, but we weren't talking about that kind of thing." – retrospective child; on HBOC (Bakos et al., 2008)

Through their mothers' experiences of breast cancer and their risk management decisions, young people in HBOC families would begin to realize the potential implications of breast cancer risk for their own psychological wellbeing, particularly in relation to their self-esteem and body confidence (Rowland et al., 2016):

"... going on the beach and wearing a bikini, I think that was one of the factors with my mum, like you know [in choosing] to have a reconstruction, and I know that that would affect me as well … because that's when it will knock my confidence and stuff … if that is going to happen, I hope it happens later on when I've got a family and I'm settled down and stuff." – daughter; on HBOC (Rowland et al., 2016)

When a family discussion is facilitated by GC professionals, many children reported gaining confidence and engaging in open communication with family members. The study describing a multi-family discussion group (MFDG) intervention reported positive outcomes, with communication being more effective between family members. In this approach, children who were aged 5 years to 12 years were asked to draw their families before and after attending the program. Before the program some children exhibited patterns where they wrote "why can't you tell me?" and following the intervention, drew children in the family (or themselves) as being happier because they understand what is happening in the family (Eisler et al., 2016).

3.2. Behavioral data

In relation to behaviors following disclosure, some participants reported that learning about their mother's result on HBOC had immediately made them feel that they want to get tested. Others had a more distant time in mind concerning the pursuit of testing for themselves. And others thought that they might not want testing until after they begin having mammograms or when they are ready to have children, with several thinking that

they might decide against having children to prevent the passing of the gene mutation (Patenaude et al., 2013):

> *"I think I want to get tested as soon as possible, even though my mom thinks I don't really need to, just because, why not? Just to, kind of, know. Because I am already assuming I have it. So nothing will change if I do have it, but a lot could change if I don't."* – daughter; on HBOC (Patenaude et al., 2013)

In one study on HBOC, children reported on important reasons for testing when reaching adulthood, which included (in order of importance): to pay closer attention to one's own health; to make more informed decisions about family; to feel better; to make better decisions about smoking or using tobacco; to plan for school or work (Tercyak et al., 2001). In another paper, that utilized retrospective reporting, children highlighted multiple reasons for pursing testing, which included: emotional reasons, the need for increased certainty, family history, preventive measures and (future) children (Sie et al., 2013).

Selective GRI in conjunction with the normalization of cosmetic surgery appeared to impact children and young people's attitudes towards breast cancer and risk management procedures. There are reports on young females wanting genetic testing to receive free breast augmentation (Rowland et al., 2016). When attempting to protect children from the emotional and psychological impacts, parents compromised their child's understanding of GRI. Gendered and partial disclosure influenced male children's perceived risk of the BRCA gene mutation, as being minimal, despite the increased risk of other types of cancer (Rowland et al., 2016):

> *"I understand [BRCA mutation] doesn't affect males that much, it like only gives them like a marginal chance more, whereas females… it gives them a massive chance of getting [breast cancer]. So I couldn't see the interest… it's not something I'd get worried about, me myself… Obviously mum's going to, auntie's going to, [sisters] going to but it hasn't affected me as much as it would them probably…"* - over 13 years old son; on BRCA gene mutation (Rowland et al., 2016)

Still, from the children's perspective, genetic testing in general was reported as having the advantage of allowing them to do efforts in disease prevention, by living a healthier lifestyle and engaging in screening to allow for early disease detection and

treatment (Alderfer et al., 2017). Other health behaviors described as preventive included exercising regularly, eating a healthy diet and avoiding tobacco (Bradbury et al., 2009):

> *"It's worth it to know so that you can have the screenings done… making it so that you can take better care of yourself. It's just something else to know about your body, you know, about your history, so you can take care of yourself in the best way possible"* – retrospective child; on LFS (Alderfer et al., 2017)

Young adults who received genetic information often expressed having the responsibility to inform siblings and cousins, so that they obtain accurate information (Young et al., 2019).

Although one study, suggested that offspring felt that in general children could experience potential negative aspects from learning of a familial mutation (e.g., like changing life plans or feelings of confusion and concern), when they reported on personal experiences, they did not report negative aspects (Bradbury et al., 2009). This study suggested that they themselves did not identify with the reported potential negative responses to genetic testing (e.g., changing life plans or feelings of confusion and concern).

4. What are the child's preferences regarding genetic cancer risk communication within the family?

4.1. Family communication

Concerning the family communication process, children reported agreeing that the parents have the primary role in relation to who should talk to a child about genetic risk (Stuttgen et al., 2020). However, in general, children reported that they wished their parents had given them more information.

Whether or not to undergo genetic testing, adolescents and emerging adults reported that they had enough input on the decision-making process within the family. Those children not involved in the testing decision on LFS said that this was appropriate given their age at the time. Also, children stated that they trusted their parents' judgment by having their best welfare in mind (Alderfer et al., 2017):

"My mom just pretty much told me, 'you're getting a blood test to see if you have this or not.' (Do you feel like you should have had more say in the decision?) No. I was younger and if my mom said, 'hey, do you want to go check to see if you have this or not?' I would have probably said yes anyway" – 16 years old; on LFS (Alderfer et al., 2017)

In one study, children aged between 15 and 17 years old, were asked to report on something that "went well" in their experience with learning about GRI and described a pattern where they were told at a young age about the cancer risk and over time learned more details about it as they got older (Stuttgen et al., 2020). They also indicated the importance of having conversations about the syndromes and/or testing both initially and the importance of it being a topic that is revisited over time (Stuttgen et al., 2020; Tercyak et al., 2019).

Children prefer that parents communicate the information honestly in a way that does not frighten the child. While honesty is important, children also identified tone as being relevant (Stuttgen et al., 2020). The desire and preference for open communication (Norris et al., 2009) and the ability to bring up the topic of genetic risk comfortably with a parent was deemed of value (Stuttgen et al., 2020).

"You can't sugar coat it. You just have to tell them exactly what it is. If you tell them everything's 100-percent perfect and just about won't affect them, it's not true. I'd rather know the full truth" – teenager; on hereditary cancer (Stuttgen et al., 2020)

"My mom, when she first told us she spoke very calmly and explained it in depth and she didn't get too emotional about it, which helped us not immediately freak out and assume that the worst was going to happen" – teenager; on hereditary cancer (Stuttgen et al., 2020)

4.2. Professional care

A key theme was that children expressed interest in knowing, as future adults, about their risk for developing cancer (Tercyak et al., 2001). In all studies, offspring show interest in learning more about genetics and hereditary disease in general. Quantitative data

showed an average of 9 on a scale from 1 to 10 for need of information (Sie et al., 2013). Offspring who reported retrospectively suggested that it would be helpful to meet with a genetic counselor or other experts in general, when they learned of their parent's mutation (Bradbury et al., 2009). Despite twelve studies (80%) reporting that the family attended GC, in only one study the children were included in the service (McGill et al., 2019).

There is limited feedback from children about GC because in general they do not attend GC sessions. Qualitative and quantitative findings showed that young adults commonly expressed satisfaction with the care that they received from genetic services, and when participants indicated that they were not satisfied with the quality of care received, this was commonly related to the experiences associated with a lack of continuity of care. Continuity of care was especially valued:

> *"The (genetic professional) that we go to see has been with us throughout our entire life. So we're very privileged to have someone throughout all those years with us. It's almost like they're a family member."* – son; on cancer-related genetic syndromes (McGill et al., 2019)

Some young adults indicated that GC had prompted debate among family members in relation to the implications of knowing about a hereditary condition in the family (McGill et al., 2019). MFDG can help facilitate family communication, since children and young people often reported difficulties in initiating conversations about their feelings. Parents expressed felt that they did not have enough input from genetics/health professionals to support them in the disclosure process (Eisler et al., 2016).

All offspring who received written materials indicated that they found the information helpful:

> *"Knowing what's going on medically with my body is better than not. I am more mature now and have come to respect certain things. I want to learn more"* – son, 24 years old, 17 years old at disclosure; on HBOC (Bradbury et al., 2009)

Among offspring who did not receive written materials, the majority felt that written materials would have been helpful, either for themselves or for others. Offspring also suggested value in having opportunity to meet with a genetic counselor or other professionals (Bradbury et al., 2009; McGill et al., 2019; Young et al., 2019).

> *"Well, I feel like they didn't explain it enough. I mean, they didn't make me feel scared, but they didn't explain it enough for me to fully grasp it. I didn't really understand the full concepts of this mutation and how it might affect me"* – teenager; on hereditary cancer (Stuttgen et al., 2020)

Regarding preferences, sons and daughters supported the genetic testing of minors, and for those who cautioned against testing minors, the primary reason identified was the lack of medical indication (Bradbury et al., 2008):

> *"It should be their choice (to test). We should leave it open to people."* – son; on HBOC (Bradbury et al., 2008)

Offspring who reported retrospectively suggested various recommendations concerning the appropriate age of testing. For example, for HBOC and Lynch syndrome: 2% of the participants suggested from birth, 4% suggested starting at 16 years, 24% starting 18 years, 20% from 20 years, 18% from age between 21 and 24 years, 18% from 25 years, and 2% from 30 to 35 years. Twelve percent of the participants did not report a recommended age for testing stating that it depends on the person (Sie et al., 2013). Overall, this review found that there was no strong agreement in terms of the age preferences for testing.

Discussion

This is the first systematic review on evidence from the children's only perspective concerning family communication of genetic cancer risk information.

Our findings confirm that disclosure of genetic information is typically performed by parents, and this disclosure pattern aligns with the preferences of children as reported in this review and is a finding consistent with prior studies (Wilson et al., 2004). Results also reveal that it is typically the mother that discloses GRI to children, which leads to a gender factor linked to disclosure. According to the literature, women seem to take on the primary role in disclosure and of acting as family “representative”, “kin keeper” (Wilson et al., 2004), or “gatekeeper” (Nycum et al., 2009; Rowland and Metcalfe, 2013). Studies focused on adults demonstrates that women disclose genetic information to sisters and daughters more often than to brothers and sons (Wilson et al., 2004). Our findings confirm this gendered disclosure process. Also, our results showed few reports of male children expressing about genetic cancer risk. This can have important implications on the perception of risk by male mutation carriers.

Our findings on gendered disclosure suggest that young males may perceive their risk of the BRCA gene mutation as being minimal, despite their increased risk for other types of cancer including prostate, pancreatic and male breast cancer. However, most samples in this review included samples that were predominantly female children. For instance, when talking about breast cancer, our results tended to the disclosure content focus on women’s breast cancer, with men or male cancers rarely mentioned. This finding is consistent with research conducted on adult populations that has found stigmatization of BRCA as being a “woman’s disease”. Because male breast cancer is rare, female breast cancer receives more attention. Influencers such as breast cancer receiving more publicity concerning women through media attention may play a role in perpetuating this context. To reduce this, more publicity of male breast cancer is needed, as well as gender-neutral communication and information material (Midding et al., 2018).

Our findings show that the disclosure of genetic cancer risk information is mostly done in a formal way where there may be somewhat of a plan, with a specific time and place when parents sit with their children. Planning ahead at least to some extent is one of

Werner-Lin's recommendations on parents' approach to children about adult-onset hereditary cancer risk. More specifically, the first step is the parent's work towards emotional and informational stability prior to disclosure (Werner-Lin et al., 2018). However, our results found that some parents choose to tell their children in an informal context, for example, while engaged in an activity or while driving the car. Research on how mothers tell their children about breast cancer and treatment shows that a main goal of the parent is to create a process that involves protecting the child from potential distress. To achieve this, a parent may attempt to maintain normality and minimize their own emotional needs and reactions (Asbury et al., 2014).

According to the literature, the disclosure between parent carriers and their offspring is a process that occurs in several distinct phases. These included a pre-disclosure phase, where parents contemplate the sharing of the genetic information; a disclosure phase where parents shared it; and the impact of disclosure phase, where parents reflect over the disclosure moment (Clarke et al., 2008). There is not a specific or "one" way that is recommended for all families as they make their way through the process of disclosing information to offspring. Parents must consider that when initiating discussions with their children about their cancer genetic test result, it is only the beginning of a long-term conversation between family members (Patenaude and Schneider, 2017). Our results confirm this, meaning that children report preferring to have information provided over time gradually, with openness, honesty and that is reassuring at the same time.

Our findings suggest that children may be particularly sensitive to their parent's emotional state at the time of disclosure, which may also continue through into follow-up discussions. Prior reviews on the experiences of parents around disclosure of genetic information have found that they experience challenges in the following areas: how to talk without alarming children, knowing what to say, and in managing their own emotions (Metcalfe et al., 2008). Parents can feel unsure of how much and what specific content children can understand or cope with (Finch and Gibson, 2009). Findings from this review and prior work on adult experiences, taken together, suggest a certain need of parents for emotional preparation and literacy that does not appear to currently exist.

Parents have expressed feeling uncertain concerning knowledge about children's reactions and on how to respond to potential reactions appropriately in a way that minimizes harm (Helseth and Ulfsaet, 2005). Parents may project their own emotional

experiences or perspectives onto their children and/or anticipate children's reactions, based on the relationship to or their perception of their child (e.g., need to protect, feeling of closeness). The nature of the relationship within the family is known to lead to different types and probabilities of communication (Wilson et al., 2004). Our findings, however, suggest not only that children can handle information, but in fact, they desire access to relevant genetic or risk information. GC considers the family unit given its important role in the dissemination of genetic information to family members, and professionals can reassure and support the parent on disclose the information[1].

Findings in this review suggest that the meaning of the genetic information is affected by what is (or has occurred) in the family. For example, a current or past diagnosis in other family members may impact how genetic information is received and responded to. This finding is consistent with prior research that focused on adults receiving genetic information and the role of family context (Smith et al., 1999) and suggests calls for a systemic view of the family and a family-centered approach to GC. The FSGI model describes how genetic conditions affect the family differently through a schedule of genetic disease, where each corresponding phase is associated with specific psychosocial challenges. The concept of phases over time in this model enables a longitudinal examination of responses by various family members, that recognizes that the course of the disease is a process that involves transitions and changes, that will impact and require the adaptation of the family (Rolland & Williams, 2005). This model allows for consideration of "events" of the family being documented or assessed in preparing for disclosure or in follow-up meetings (e.g., recent cancer diagnosis, death of family member).

Children and parents require support through the genetic condition transitions and readjustments (Metcalfe et al., 2008). Our findings and prior works suggest that the impact of genetic knowledge has effects over time that are not necessarily apparent at the time of disclosure (Bonadona et al., 2002). For example, daughters reported fear and worry when their mothers tested positive for the mutation, and when mothers received negative results, children reported feeling "relief". But when watching older family members die from the illness, sadness and social isolation could be triggered. Through the system dynamic of adaptation, children come to realize the potential implications of the genetic information for their own and other family members' wellbeing.

[1] See Forrest et al. (2007), for some guidelines.

When the cancer syndrome causes observable consequences, such as body changes, children become aware of it. In our results, the areas of self-esteem and body confidence were some of the implication's daughters reported in relation to their own psychological wellbeing. This finding is consistent with research on body image (Boquiren et al., 2013; Esplen et al., 2018) and the mother-daughter relationship after breast cancer (Lichtman et al., 1985). It's crucial to monitor how a child is doing and experiencing changes occurring both in relation to their parent and in relation to their own development. The literature recommends and encourages follow-up visits or mental health appointments to discuss potential impacts of cancer/risk knowledge and the progress of family communication (Patenaude and Schneider, 2017).

Health professionals' support and advice are very important in helping parents cope and manage their own feelings (Resta et al., 2006). Optimal communication of GRI by parents to children can have positive long-term effects (Metcalfe et al., 2008). When there are communication challenges or gaps, relationships between family members such as a parent and offspring can be compromised by way of a "protective buffer". This type of relationship dynamic has been described among cancer patients and their partners as a coping strategy - "protective buffering" (Kuijer et al., 2000; Manne et al., 1999), where efforts are made to protect one's partner from upset and burden by cover-up worries, concerns, and yielding to the partner to avoid disagreements (Manne et al., 2007). A similar pattern can occur in the genetic context and contribute to communication challenges or inhibition of disclosure of cancer risk information that has important relevance for the health outcomes of family members who may be at increased risk. Our results suggest that this happened when offspring hide worries or refrained from asking questions about genetic cancer risk, to protect their parents from the additional burden and emotional distress.

1. Limitations

This study has some limitations that are important to note. In this systematic review of the literature, we included some studies with adults retrospectively referring to their experiences as children (when they were < 18 years old). This means that the shared

experience may have already a different meaning as people entered adulthood and assimilate information and experiences differently. Moreover, when the participants' age was not indicated, we had to assume which term to refer to (children or retrospective reporting of children) based on the given information. Moreover, children or retrospective reporting of the children's age was not specified so we had to assume which term to refer to based on the given information. Several studies included entire families as their sample of focus, however, we included those studies because their findings contributed to the topic along with the other included studies.

The possibility of Patient and Public Involvement (PPI) in this systematic review is being discussed because of the potential benefits of it, such as knowledge about the topic of the review and first-hand experience, adding value to the review, improving quality and relevance to public and patients (INVOLVE, 2012). We are considering the involvement of young people, by means of a youth advisory group, that can contribute to the review in different phases of its design, conduct and dissemination. This parent-child theme emerged from the citizens of the TOGETHER project as a priority. However, this component will be worked on later, due to the implications of the process over time. Therefore, this dissertation will not include PPI.

2. Clinical implications

The findings from this systematic review have implications for clinical practice, particularly for the GC professional. With the role to help people understand and adapt to the medical, psychological, and familial implications of genetic contributions to disease (Resta et al., 2006), genetic counselors can support communication within the family system to further support communication efforts to that of the primary recipient of genetic testing/counseling.

This review provided evidence to support openness and interest among offspring in learning more about genetics and hereditary disease in general. While GC models indicate the family as the unit for counseling, these findings suggest greater emphasis of GC in supporting offspring, including children. Perhaps greater focus could include providing

services that specifically target children in families who have elevated cancer risk. Findings may also support a team approach where GC work closely with other health team members, such a social worker or psychologist to support family adaptation and support communication. Children in this review demonstrated strong interest in having follow-up and over time care.

Regarding testing, this review did not find agreement in terms of the age preferences for genetic testing. Although there are some common aspects between current concerns about testing children and concerns about testing adults, GC professionals must monitor each applicant and the family. For instance, addressing any pre-existing risks of psychological distress on parents contributes to better communication and collaboration with the GC professional. Therefore, it ensures the best outcomes when considering genetic testing in a child (Wakefield et al., 2016).

The multi-family interventions study provided positive experiences for children and supported parent-child communication (Eisler et al., 2016). Findings also suggest that children have a need to meet with a genetic counselor and desire more written materials to support their understanding and adjustment. These resources were suggested as having value even prior to being able to do the genetic test. Current GC services may consider further expansion of focus on offspring to support parents and family members in the GC process.

3. Implications for research

This systematic review of the children's perspective on communication about genetic cancer risk to offspring identified only fifteen articles from four countries addressing a few of the proposed genetic syndromes, indicating a paucity of research in this area. Research is scarce regarding quantitative data and syndromes, other than breast and ovarian cancer and LFS. Male children are also a minority in relation to samples being investigated on this subject, including in relation to important topics such as prostate cancer and male breast male cancer. There is also little known about genetic testing for cancer from the offspring perspective in different cultures. Most studies were conducted in

the United States, United Kingdom, and Australia. The inclusion criteria resulted in the omission of studies published in different languages and outside peer reviewed journals. Cultures are known to vary in relation to family belief systems and values (Wilson et al., 2004) so further studies are needed.

Further research driven by family communication is required to examine communication within the family, primarily from the children's perspective. Also, a focus on what kind of information children wants to know or have access to would inform both GC professionals and parents, and give a voice to children growing up in a family affected by a genetic cancer syndrome.

Conclusions

In conclusion, this review aimed to investigate and support the children's voices to be heard in relation to the genetic testing process and how genetic information may be best disseminated to offspring. We believe we have achieved that goal and findings have implications for important potential changes in how children can and may want to be involved in the GC process. Findings suggest that offspring may be open to, and have a need for greater choice in how information concerning their potential risk may be provided prior to the age of their involvement as pointed out by the current guidelines.

This review has shown that family communication about genetic cancer risk is a process where the information can be both empowering and threatening. Parents' literacy influences family communication about genetic cancer risk information. This plays a role in the psychological adjustment of children, as well as future health behaviors adherence.

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Appendix A. Search terms for PubMed

Concept	Keyword	MeSH
Children	Child*[tiab] OR adolescen*[tiab] OR offspring[tiab] OR teen*[tiab] OR minor*[tiab]	"Child"[Mesh]
Family	Famil*[tiab] OR relative*[tiab] OR parent*[tiab] OR sibling*[tiab] OR kin[tiab] OR “family system” [tiab] OR “family network” [tiab]	"Family"[Mesh]
Genetic Cancer	“genetic test*”[tiab] OR “genomic test*”[tiab] OR “genetic counseling” [tiab] OR “genetic counselling”[tiab] OR “genetic screening”[tiab]	"Genetic Predisposition to Disease"[Mesh] OR "Genetic Counseling"[Mesh]
	Cancer[tiab]	
Genetic Cancer Syndromes	“genetic cancer risk”[tiab] OR BRCA1[tiab] OR BRCA2[tiab] OR “familial adenomatous polyposis”[tiab] OR FAP[tiab] OR “Lynch syndrome”[tiab] OR “Cowden syndrome”[tiab] OR “Li Fraumeni syndrome”[tiab] OR “hereditary diffuse gastric cancer”[tiab] OR HDGC[tiab] OR retinoblastoma[tiab]	"Genes, BRCA1"[Mesh] OR "Genes, BRCA2"[Mesh] OR "Adenomatous Polyposis Coli"[Mesh] OR "Colorectal Neoplasms, Hereditary Nonpolyposis"[Mesh] OR "Hamartoma Syndrome, Multiple"[Mesh] OR "Li-Fraumeni Syndrome"[Mesh] OR "Stomach Neoplasms"[Mesh] OR "Retinoblastoma"[Mesh]
Communication	communication[tiab] OR talk*[tiab] OR shar*[tiab] OR discuss*[tiab] OR inform*[tiab] OR “health communication” [tiab]	"Communication"[Mesh] OR "Parent-Child Relations"[Mesh]
Outcomes	“psychological adjustment”[tiab] OR “psychological adaptation”[tiab] OR “self concept”[tiab] OR "self	"Emotional Adjustment"[Mesh] OR "Adaptation,

	esteem"[tiab] OR “body image”[tiab] OR “body shame”[tiab] OR “body stigma”[tiab] OR “body concerns”[tiab] OR stress[tiab] OR distress[tiab] OR vulnerability[tiab] OR anxiety[tiab] OR depressi*[tiab] OR "well being"[tiab] OR "quality of life"[tiab] OR concern*[tiab] OR disclosure[tiab] OR believe*[tiab] OR perception*[tiab] OR "risk perception"[tiab] OR "health literacy"[tiab] OR understanding[tiab]	Psychological"[Mesh] OR "Self Concept"[Mesh] OR "Body Image"[Mesh] OR "Stress, Psychological"[Mesh] OR "Psychological Distress"[Mesh] OR "Anxiety"[Mesh] OR "Depression"[Mesh] OR "Health"[Mesh] OR "Quality of Life"[Mesh] OR "Truth Disclosure"[Mesh] OR "Perception"[Mesh] OR "Health Knowledge, Attitudes, Practice"[Mesh] OR "Comprehension"[Mesh] OR "Health Literacy"[Mesh]

Appendix B. Data extraction table

Article #	Author	Year	Title	Abstract	**Citation** (APA citation of the paper)	**Country** (where the study took place)
17	Melissa A. Alderfer & Robert B. Lindell & Claire I. Viadro & Kristin	2017	Should genetic testing be offered for children? The perspectives of adolescents and emerging adults in families with Li-Fraumeni syndrome	Whether children should be offered genetic testing for cancer risk is much debated but young voices are rarely heard in these conversations. The current study explored perspectives of genetic testing held by adolescents and emerging adults	Alderfer, M. A., Lindell, R. B., Viadro, C. I., Zelley, K., Valdez, J., Mandrell, B., ... & Nichols, K. E. (2017). Should genetic testing be offered for children? The perspectives of	USA
39	A. D. H. Bakos, Sadie P.; Loud, Jennifer T.; Peters, June A.; Giusti, Ruthann M.;	2008	BRCA mutation-negative women from hereditary breast and ovarian cancer families: A qualitative study of the BRCA-negative experience	Background: When women from families with a known BRCA1 or BRCA2 mutation test negative for the family mutation, it is assumed that they will transition their personal cancer risk perception from high to average risk. However, there	Bakos, A. D., Hutson, S. P., Loud, J. T., Peters, J. A., Giusti, R. M., & Greene, M. H. (2008). BRCA mutation-negative women from hereditary breast and ovarian cancer families: a	USA
70	A. R. P.-M. Bradbury, L.; Pawlowski, K.; Ibe, C. N.; Cummings,	2008	Should genetic testing for BRCA1/2 be permitted for minors? Opinions of BRCA mutation carriers and their adult offspring	Although professional guidelines recommend against testing minors for adult-onset genetic conditions, the genetic testing of minors for BRCA1/2 alterations has been debated in the literature. To better	Bradbury AR, Patrick-Miller L, Pawlowski K, Ibe CN, Cummings SA, Olopade OI, Daugherty CK. 2008. Should genetic testing for BRCA1/2 be permitted for minors?	USA
71	Angela R. Bradbury, Linda Patrick-Miller, Kimberly Pawlowski,	2009	Learning of your parent's BRCA mutation during adolescence or early adulthood: a study of offspring experiences	Objective: To examine the experience, comprehension and perceptions of learning of a parent's BRCA mutation during adolescence and early adulthood, and explore the impact on offspring's physical	Bradbury, A. R., Patrick-Miller, L., Pawlowski, K., Ibe, C. N., Cummings, S. A., Hlubocky, F., ... & Daugherty, C. K. (2009). Learning of your	USA
419	B. C. W. McGill, C. E.; Vetsch, J.; Lim, Q.; Warby, M.; Metcalfe, A.; Byrne, J.	2019	I remember how I felt, but I don't remember the gene": Families' experiences of cancer-related genetic testing in childhood"	BACKGROUND: Genetic testing in children for hereditary cancer predisposition syndromes (CPSs) involves unique psychosocial and family-systems considerations. This retrospective study explored the perspectives and emotional	McGill BC, Wakefield CE, Vetsch J, et al. "I remember how I felt, but I don't remember the gene": Families' experiences of cancer-related genetic testing in	Australia
475	Andrea F. Patenaude, Nadine Tung, Paula D. Ryan, Leif W. Ellisen, Larissa	2013	Young adult daughters of BRCA1/2 positive mothers: What do they know about hereditary cancer and how much do they worry?	Objective: The objectives of this study are to determine (i) what daughters, ages 18–24 years, of BRCA1/2 mutation carriers understand about their 50% chance of carrying a BRCA1/2 mutation and about risk reduction or management	Patenaude, A. F., Tung, N., Ryan, P. D., Ellisen, L. W., Hewitt, L., Schneider, K. A., ... & Garber, J. E. (2013). Young adult daughters of BRCA1/2 positive mothers: What do they know about	USA
517	E. P. Rowland, Gill; Considine, Anna-Marie; Metcalfe, Alison	2016	Preparing young people for future decision-making about cancer risk in families affected or at risk from hereditary breast cancer: A qualitative interview study	Purpose: Women carrying the mutated BRCA gene, have approximately an 80% life-time risk of developing breast cancer with 50% risk of their children inheriting the gene mutation. Many parents find it difficult to know when and how to disclose this information to	Rowland, E., Plumridge, G., Considine, A. M., & Metcalfe, A. (2016). Preparing young people for future decision-making about cancer risk in families affected or at risk from hereditary breast	UK
551	A. S. P. Sie, J. B.; Spruijt, L.; Kets, C. M.; Hoogerbrugge, N.	2013	Can we test for hereditary cancer at 18 years when we start surveillance at 25? Patient reported outcomes	DNA-testing for BRCA1/2 or Lynch syndrome is possible from the age of 18 years, although surveillance usually starts at 25. Some patients regret their decision of testing before age 25. This retrospective study evaluates whether the testing age should be above 25 years to	Sie, A. S., Prins, J. B., Spruijt, L., Kets, C. M., & Hoogerbrugge, N. (2013). Can we test for hereditary cancer at 18 years when we start surveillance at 25? Patient reported outcomes. Familial cancer,	Netherlands
594	Kenneth P. Tercyak, Beth N. Peshkin, Randi Streisand and Caryn	2001	Psychological issues among children of hereditary breast cancer gene (BRCA1/2) testing participants	Children growing up in hereditary breast cancer families may experience diminished psychological well-being. In addition to coping with having a cancer-affected parent or close relatives, these children may focus on their	Tercyak, K. P., Peshkin, B. N., Streisand, R., & Lerman, C. (2001). Psychological issues among children of hereditary breast cancer gene (BRCA1/2) testing participants.	USA
664	A. H. Werner-Lin, Lindsey M.; Doyle, Maya H.; Greene, Mark H.	2012	Cancer doesn't have an age': Genetic testing and cancer risk management in BRCA1/2 mutation-positive women aged 18–24	Increasingly, 18–24-year-old women from hereditary breast/ovarian cancer (HBOC) families are pursuing genetic testing, despite their low absolute risks of breast and ovarian cancer and the fact that evidence-based management options used with older high-risk women are not	Werner-Lin, A., Hoskins, L. M., Doyle, M. H., & Greene, M. H. (2012). 'Cancer doesn't have an age': Genetic testing and cancer risk management in BRCA1/2 mutation-positive women aged 18–24.	USA
685	A. L. B. Young, Phyllis N.; Rhodes, Paul; Tucker, Katherine M.;	2019	Talking across generations: Family communication about brca1 and brca2 genetic cancer risk	While family communication about a BRCA1 or BRCA2 (BRCA1/2) pathogenic variant can be a catalyst for the uptake of risk?reducing measures in young adults, disseminating information within families and across generations is complex. This study aimed to	Young AL, Butow PN, Rhodes P, et al. Talking across generations: Family communication about BRCA1 and BRCA2 genetic cancer risk. J Genet Couns. 2019;00:1–17. https://doi.org/10.1002/jgc	Australia
696	Stuttgen, K.; McCague, A.; Bollinger, J.; Dvoskin, R.; Mathews, D.	2020	Whether, when, and how to communicate genetic risk to minors: 'I wanted more information but I think they were scared I couldn't handle it'	Genetic test results are often relevant not only to persons tested, but also to their children. Questions of whether, when, and how to disclose parental test results to children, particularly minors, can be difficult for parents to navigate. Currently, lim ited data are	Stuttgen K, McCague A, Bollinger J, Dvoskin R, Mathews D. Whether, when, and how to communicate genetic risk to minors: 'I wanted more information but I think they were scared I couldn't	USA
697	Norris, J.; Spelic, S. S.; Snyder, C.; Tinley, S.	2009	Five Families Living With Hereditary Breast and Ovarian Cancer Risk	This qualitative study explores the communication and decision-making strategies of five families with hereditary breast and ovarian cancer (HBOC) risk. Investigators asked female carriers of BRCA1 and BRCA2 genetic mutations to recall early	Alderfer, M. A., Lindell, R. B., Viadro, C. I., Zelley, K., Valdez, J., Mandrell, B., ... & Nichols, K. E. (2017). Should genetic testing be offered for children? The perspectives of	USA
698	Ivan Eisler, Matthew Ellison, Frances Flinter, Jo Grey, Suzanne Hutchison	2016	Developing an intervention to facilitate family communication about inherited genetic conditions, and training genetic counsellors in its delivery	Many families experience difficulty in talking about an inherited genetic condition that affects one or more of them. There have now been a number of studies identifying the issues in detail, however few have developed interventions to assist families. The SPRinG collaborative	Eisler, I., Ellison, M., Flinter, F., Grey, J., Hutchison, S., Jackson, C., ... & Ulph, F. (2016). Developing an intervention to facilitate family communication about inherited genetic	UK
699	Kenneth P. Tercyak, Suzanne M. Bronheim, Nicole Kahn, Hillary A. Robertson, Bruno J.	2019	Cancer genetic health communication in families tested for hereditary breast/ovarian cancer risk: a qualitative investigation of impact on children's genetic health literacy and psychosocial adjustment	Children's literacy about the genetics of late-onset hereditary breast/ovarian cancer (HBOC) often develops through conversations with parents about BRCA gene testing and adults' cancer diagnoses. These conversations may promote early understanding of HBOC, but	Tercyak, K. P., Bronheim, S. M., Kahn, N., Robertson, H. A., Anthony, B. J., Mays, D., ... & DeMarco, T. A. (2019). Cancer genetic health communication in families tested for hereditary breast/ovarian cancer risk:	USA

Aim(s) of research	Study design (broad categorization)	Data collection procedure	Intervention? If yes, describe.	Materials (if questionnaires were used, name them)	Sample/ informants	If "mix sample", describe.
To explore perspectives of genetic testing among adolescents and emerging adults in families with LFS, examining their understanding and expectations of genetic testing and the psychosocial implications of the testing and results.	Qualitative	Semi-structured interviews	No	Semi-structured interview guide developed for the current study.	Children AND Retrospective children	adolescents/emerging adults - offspring
Explore cancer risk perception and the experience of being a mutation-negative woman within a known BRCA1/2 mutation-positive family.	Qualitative	Semi-structured telephone interviews	No	A semi-structured interview guide was developed by the nurse researchers	Retrospective children (adults referring to their experiences as < 18 years	N/A
To better understand the opinions of BRCA mutation carriers regarding the genetic testing of minors and the cognitive and affective processes underlying these opinions.	Qualitative	Semi-structured interviews	No	Not reported	Mix sample (children AND adults AND/OR families)	Offspring (retrospective children) and parents.
Interview adult offspring in the developing cohort who learned of their parent's BRCA mutation to better understand the content and method of disclosure, their understanding and perceptions of	Qualitative	Interviews	N/A	N/A	Retrospective children (adults referring to their experiences	N/A
Explore the perspectives and emotional reactions of parents and young adults about cancer-related genetic counseling and testing offered to children in the family. | evaluated families' psychosocial experiences following pediatric	Mix-methods	Semi-structured interview; quantitative measures: The Emotion Thermometers Tool (ETT); The	No	The Emotion Thermometers Tool (ETT), The Genetic Counseling Satisfaction Scale (GCSS),	Mix sample (children AND adults AND/OR families)	Parents and young adults
The objectives of this study are to determine (i) what daughters, ages 18–24 years, of BRCA1/2 mutation carriers understand about their 50% chance of carrying a BRCA1/2 mutation and about risk reduction or management options for	Mix-methods	Phone interviews and questionnaires	N/A	The qualitative interview was developed by the principal investigator. Questionnaire	Retrospective children (adults referring to their experiences as < 18 years	N/A
This study aimed to ask what are the experiences of parents, children and young people when discussing genetic risk in families affected by or at risk from a BRCA gene mutation, and how does the information shared impact on children and young peoples views	Qualitative	Semi-structured interviews	No	N/A	Mix sample (children AND adults AND/OR families)	14 parents and 13 children and young people.
The aim of this retrospective study was to assess whether the testing age should be above 25 years in order to prevent adverse effects such as regret or decisional conflict, by determining the percentage and characteristics of patients reporting such problems in retrospect.	Mix-methods	Questionnaires and open-ended questions. | online self-report survey	No	Regret of the decision to start DNA-testing before 25 years was measured by a 4-point Likert-scale from 1	Retrospective children (adults referring to their experiences as < 18 years old)	N/A
The purpose of this preliminary study is to explore conceptions of health, cancer risk, and psychological adjustment among children in families suggestive of carrying BRCA1/2 susceptibility genes	Mix-methods	Questionaires (quantitative) and open question	N/A	Children's health attitudes and beliefs questionnaire was adapted from those	Only Children (0 - 18 years old)	Eligible subjects were English-speaking children ages 11–17 of mothers enrolled in the Cancer Assessment and Risk Evaluation (CARE) program at the Lombardi
This study sought a patient-centered perspective on the dilemmas faced by 18–24-yearolds considering genetic education, counseling and testing to identify a BRCA1/2 gene mutation. (…) Data for the current study are drawn from three separate qualitative studies,	Qualitative	Interview	No	N/A	Retrospective children (adults referring to their experiences as < 18 years old)	N/A
This study aimed to explore how young adults and their families communicate about a BRCA1/2 pathogenic variant, from a family systems perspective.	Mix-methods	Qualitative (semi-structured family interviews and individual young adult inter-views) and quantitative (questionnaire)	No	Semi-structured interviews; Genogram; Questionnaires/measures: Families at risk of family dysfunction	Mix sample (children AND adults AND/OR families)	Participants were young adults (18–40 years old) and their relatives, with at least one member of the family having a BRCA1/2 pathogenic variant.
To assess how minors understand genetic risk information (GRI), to learn about the disclosure event and what information was given to the minor, and to evaluate how parents' perspectives differ from minors' in order to understand factors associated with positive and	Qualitative	Interview	No	Semi-structured interviews	Mix sample (children AND adults AND/OR families)	Genetic counselors, children and parents.
The present study initiated a family-focused look at living with HBOC risk. The investigators sought to develop an intergenerational perspective on families' stories to highlight aspects of the family context as influences on	Qualitative	Telephone or at-home interviews	No	Open-ended questions	Mix sample (children AND adults AND/OR families)	Five families: parents, sons and daughters
Research questions were 'Are Multi-Family Discussion Groups a suitable intervention for facilitating family communication about an IGC?' and 'Is it possible to train GCs to deliver the intervention?' The three study aims were (i) To ascertain whether a MFDG was a suitable approach, and	Qualitative	Multi-family discussion groups (MFDGs) | Focus groups	Yes. The team work with families and genetic counsellors (GCs) to co-design a psycho-educational	N/A	Mix sample (children AND adults AND/OR families)	Parents, Young People (13 - 18 years old) and children (5 - 12 years old)
To further the study of early literacy skills and communication about genetics in families, we conducted a qualitative investigation of adolescent and young adult children's psychosocial adjustment following disclosure of their mother's BRCA test results. Our	Qualitative	Interviews	No	Semi-structured, qualitative interview for children.	Only Children (0 - 18 years old)	N/A

Participants sociodemographic						
N (total participants)	**N (children + retrospective children)**	**Age (mean, SD, range - total participants)**	**Age (mean, SD, range - children + retrospective children)**	**Sex (N female, N male - total participants)**	**Sex (N female, N male - children + retrospective children)**	**Research setting** (e.g.: hospital, genetic counseling service, e-mail, etc.)
12 participants (7 families)	12 adolescents/ emerging adults (it is not described how many	Participants ranged in age from 12 to 25 years (M = 17.8, SD = 4.82). | Mean age when offered genetic	N/A	Female = 8 (66%); Male = 4 (33%)	N/A	A psychologist (M.A.) and pediatrician (R.L.) administered telephone interviews to all participants using a
N = 13	N = 13	Mean = 49; Range: 43-57	Mean = 49; Range: 43-57	13 female	13 female	Telephone
N = 75	N = 22	Median = 43; Range: 18-66	Mean = 26; Range: 18-33	Women: 59 (79%); Men 16 (21%)	Women: 12 (55%); Men: 10 (45%)	Parent and offspring participants were contacted by telephone. | University of
22 offspring from 13 unrelated parents	9 offspring that were < 18 years old at disclosure	26 (18 - 33)	N/A	Female = 12 (55%); Male = 10 (45%)	Female = 5; Male = 4	Clinic - Univeristy of Chicago Cancer Risk Clinic
N = 35	N = 9	Parents: Mean = 50.4; SD = 7.3; Range = 32–68	Young adults: Mean = 24.8; SD = 2.9; Range = 19–29	Parents: Female = 21; Male = 5	Young adults: Female = 7; Male = 2.	Two Australian hospitals—Prince of Wales Hospital and Sydney Children's Hospital
40 daughters	N/A	Median = 21; Range: 18 - 24	N/A	All female	N/A	Cancer centres - Dana–Farber Cancer Institute, Massachusetts General Hospital and the Beth Israel Deaconess Medical
N = 27	N = 13	Not reported	Range: 10 - 21 years of age	Not reported	Not reported	Regional Genetics Unit in the UK | Semi-structured interviews were undertaken by two researchers (AM and GP) in participants'
N = 111	N/A	Median = 27; Range = 21–40	N/A	Female = 86%; Male = 14%	N/A	Surveys were mailed.
N = 35 (children = 20; mothers = 15) - Only data obtained directly from the children	20 children	Children: mean 14.9, SD 1.8; Mothers: 47.1, SD 3.8	children ages 11–17 (M= 14,9; SD= 1.8)	Mothers all female (N = 15); Children: 6 (30%) boys, 14 (70%) girls.	Children: 6 (30%) boys, 14 (70%) girls	Cancer centre - Cancer Assessment and Risk Evaluation (CARE) program at the Lombardi Cancer Center
N = 32	N/A	Mean = 23.2; Range: 21-27	N/A	All female	N/A	During the 2011 annual meeting of a North American consumer group focused on the information and support needs of BRCA1/2-positive
N = 67	N = 32 (47.76%)	Mean = 44.87, SD = 17.47, range = 18–87	Not reported	Female = 49; Male = 18	Not reported	A trained qualitative researcher (A.L.Y.) conducted all semi-structured interviews either in-person, through video
N = 85	N = 34	Not reported	14 children were 15 years old, 6 children were 16 years old, and 14 children were 17 years old	Genetic counselors (n=19): Female = 16, Male = 3. Children (n=34): Female = 22, Male = 12. Parents (n=32):	Female = 22; Male = 12.	Interviews were conducted via telephone, with the exception of one parent/child pair, who were interviewed in person
N = 17	N = 6	Not reported	Not reported (teenagers)	Female = 11; Male = 6	Female = 4; Male = 2.	Telephone or family home.
They did not collect demographic information from participants.	They did not collect demographic information from participants.	They did not collect demographic information from participants.	They did not collect demographic information from participants.	They did not collect demographic information from participants.	They did not collect demographic information from participants.	Genetics unit in the United Kingdom; charity web-pages; social media; newsletters
N = 34	N/A	M = 17.68; SD = 3.2	N/A	Female = 24 (70.6%); Male = 10 (29.4%)	N/A	The interview was administered over the telefone by a trained interviewer with master's-level expertise in social and behavioral

Family clinical history			
Family syndromes/ at-risk syndromes	**Other**	**Data on the age of onset of genetic cancer in the family**	**Characteristics of family history** (e.g.: the first member being tested, affected family members/ mutation carriers, mutation status, cancer diagnosis, deaths in the family, etc.)
Li-Fraumeni syndrome	N/A	The participants were recruited from families taking part in a larger study of parent decision-making around genetic testing for children. So, the parents already know	Two participants reported that family members had been offered and had completed testing (one reported learning this within days of participating in the project). Not all participants knew they had tested positive and that LFS was diagnosed in their families. In fact, three participants across two families gave no indication that they were aware that LFS had been
Breast and ovarian cancer	N/A	Not reported	All mutation-negative participants in this study had undergone post-genetic test counselling during which they were advised that they were non-carriers of their family's mutation or "BRCA negative". // There are 12 (92%) subjects with the BRCA1 family mutation, and 1 (8%) subject with the BRCA2 family mutation. // The mean of family breast cancer cases is 8, with a range of
Breast and ovarian cancer	N/A	28 parents (53%) have history of cancer, and 0 offspring have history of cancer. 53 parents (100%) have history of genetic testing, and 7 (32%) of	All parents had received genetic counseling and testing. All offspring were eligible for genetic testing and seven had undergone site-specific genetic testing, with three testing positive for the familial mutation. | Parents are BRCA mutation carriers who underwent clinical genetic testing and had children under 25 years
Breast and ovarian cancer	N/A	Offspring were recruited through parent participants in a study examining how and when BRCA mutation carriers disclose their	Not reported
Other syndrome(s) (if yes, describe in column V)	Cancer-related genetic mutations	Not reported	Not reported
Breast and ovarian cancer	N/A	Participants were recruited from among age-eligible daughters of BRCA1/2-positive women tested at the hereditary cancer centers of the Dana–Farber	31 daughters (77.5%) had mothers who had had cancer. | Before disclosure, many DMC had not previously been aware of the high hereditary risk their mothers had for ovarian cancer or breast cancer, whichever had not occurred in their family.
Breast and ovarian cancer	N/A	Not reported	All the families had one person, the mother, who had tested positive for a BRCA mutation and all their children were at risk until they could undergo genetic testing themselves, which is not until a minimum of 18-years old.
Multiple syndromes (if yes, describe in column V)	BRCA1/2 or Lynch syndrome	Responders (n = 111) tested for BRCA1/2 (73%) or Lynch syndrome (27%) at age 18–25 years.// Age of youngest relative with cancer 34 [18–53].	Mother affected with cancer: 48%. Father affected with cancer: 14%.
Breast and ovarian cancer	N/A	Not reported	Mothers: Breast/ovarian cancer history - 12 (80%) affected, 3 (20%) unaffected, BRCA1/2 mutation status - 7 (47%) carriers, 8 (53%) non-carriers.
Breast and ovarian cancer	N/A	Not reported	Thirteen first learned of their genetic risk because a loved one (mother, aunt, sibling) was peri-diagnostic, and were thus motivated to pursue genetic testing. Sixteen grew up familiar with their families' cancer profiles and presented for genetic testing once they were eligible for testing. Three were unaware of their HBOC risk while growing up and were told by a mutation-positive parent or by a healthcare provider
Breast and ovarian cancer	N/A	At least one member of the family having a BRCA1/2 pathogenic variant, identified during index or predictive search genetic testing between 6 months and 20 years prior to	Among women (N = 49), 16 (32.65%) had risk-reducing mastectomy and 12 (24.49%) had risk-reducing hysterectomy/oophorectomy.// 9 participants (13.43%) have cancer diagnosis.// About the result of BRCA1/2 testing, there are 38 (56.72%) positive test results, 6 (8.95%) negative test results, 8 (11.94%) not tested (at-risk), and 15 (22.39%) not tested (spouse, partners).
Multiple syndromes (if yes, describe in column V)	Hereditary cancer (breast/ovarian cancer or colorectal cancer with genetic testing that indicated Lynch syndrome) or Huntington's	Not reported	15 children were at risk for HD; 19 children were at risk for HC (8 for Breast/ovarian, 11 for colorectal). 4 parents were at risk or affected by HD; 11 (parents) spouse/partner of individual were at risk or affected by HD; 17 parents were diagnosed with HC (7 with breast/ovarian, 10 with colorectal).
Breast and ovarian cancer	N/A	Not reported	Family A: Maternal aunt died of breast cancer. Family B: Maternal grandmother died of breast cancer. Family C: Maternal grandmother was a breast cancer survivor. Family D: Maternal grandmother and aunt died of breast cancer. Family E: Maternal aunt was a cancer survivor.
Other syndrome(s) (if yes, describe in column V)	Polycystic kidney disease (PKD), Multiple endocrine neoplasia (MEN), Familial adenomatous polyposis (FAP) - the others	Not reported	Not reported
Breast and ovarian cancer	N/A	Not reported	Maternal. Breast/ovaria cancer history: 22 (64.7%) previously affected (survivor) and 12 (35.3%) unaffected. Proband status: 28 (82.4%) are proband and 6 (17.6%) is the relative. BRCA1/2 genetic test result: 11 (32.4%) are positive (carrier) and 23 (67.6%) are negative (noncarrier) or Uninformative negative.

Genetic counseling				
Applicant (who did the genetic test)	**Does/did the family attend genetic counseling?**	**If yes, were the children included?**	**Children's (0 - 18 years old) experience/feedback of genetic counseling.**	**Retrospective children's (adults referring to their experiences as < 18 years old) experience/feedback of genetic counseling.**
According to their parents' reports, 10 of these 12 participants were	Yes	No	Not reported	Not reported
Not reported	Yes	No	N/A	Not reported
Parents	Yes	No	N/A	Not reported
Parents	Yes	No	N/A	"I would get it [genetic testing] if and when I have children, so that I can be aware of the potential effects for them [his children]" (son, 21 years old, 14 years old at disclosure). | All offspring thought it would be helpful to meet with a genetic counselor or other expert in general, and the majority
Retrospective children	Yes	Yes	N/A	"The [genetic professional] that we go to see has been with us throughout our entire life. So we're very privileged to have someone throughout all those years with us. It's almost like they're a family member" (F10, young adult (male), positive, affected). // Both parents and young adults commonly expressed satisfaction with the care
Mothers	Yes	No	N/A	30 daughters had no counseling or testing; 2 had genetic counseling only; 3 had genetic testing only, 5 had both counseling and testing (1 was positive to be BRCA1/2 mutation carrier, 7 were negative). The majority of our sample had had neither genetic counseling nor BRCA1/2 testing. Untested daughters expressed a wide range of intentions
Mother	Yes	No	Not reported	Not reported
All participants; In 43 %, another relative was simultaneously tested	Not reported	No	N/A	Counseling information and services were considered sufficient (median 3.3 [1.4–4.0], scale 1–4).
Mothers	Not reported	No	Not reported	N/A
Participants	Yes	No	N/A	Julia described the experience of having the family history mapped out for her already: "The (genetic counselor) already had an overall background about our family history and had worked with both my aunt and my mother and was just really, really exceptional." For others, however, this relationship compromised the privacy of young adult women. For example, in six instances, providers shared the
Often, one or two women initiated genetic testing and advocated for others	Yes	No	N/A	Many families expressed appreciation or a desire for external support from health professionals to process the experiences they had undergone by providing emotional support, facilitating communication and for one family, providing a follow-up consultation (n = 14).
Parents	Yes	No	Not reported	N/A
Parents	Yes	No	Not reported	Not reported
Not reported	Not reported	No	N/A	N/A
Mother	Yes	No	Not reported	Not reported

1. How, when and what is discussed about genetic cancer risk in the family?

Who disclosure genetic cancer risk to the participants?	**If "other(s)", who?**	**Time interval between the test result and disclosure.**	**Age in years when told.**	**Content of disclosure** (what was said/ explained to the children. E.g.: who had genetic testing, the implications of the test result, etc.)	**Quotes AND/OR existing categories of disclosure content** (if there's not quotes and/or existing categories of disclosure content, you can write the information in the previous column).	**How was the genetic cancer risk disclosure to the participants? Describe** (e.g.: by telephone, 'sit-down' family meeting, private conversation)	**During the disclosure phase, was there any contact with genetic counseling services?**	**If yes, describe how the genetic counseling service was involved in the disclosure.**	**Were tools involved in the disclosure?** (to assist or support disclosure. E.g.: literacy resources, communication tools, brochures, etc.) **OR did the participants have access to information after the genetic cancer risk disclosure?**	**If yes, describe.**	**How do family communicate about genetic cancer/risk** (e.g.: open communication; limited communication; with fear, coping strategies; etc.)
Not reported	N/A	Not reported	Not reported. But 11.4 yrs (3-20) when offered testing. 8 participants	Not reported	N/A	Not reported	Not reported	N/A	Not reported	N/A	Not reported
Other(s)	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	N/A	Not reported	Not reported	Not reported
Other(s)	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	N/A	Not reported	N/A	Not reported
Both parents	All offspring reported that the parent with the mutation shared the result; the majority alone and	(PARENT REPORT) Based on parent reports, the majority of parents disclosed to their offspring	The estimated age range at disclosure is 11 - 24.	In three cases the genetic mutation was not initially described, although the parents (all mothers) did discuss the hereditary risk of cancer in the family. Offspring descriptions of the content shared varied; some mentioned multiple	"He [dad] said that testing had been done on his side of the family and a gene was found that makes women prone to breast cancer. He had testing and then brought it up over dinner one evening. He basically said that he is a carrier and at increased risk for cancer in	All offspring reported that the parent with the mutation shared the result; the majority alone and in eight cases with their spouses. Daughters were more likely to learn of the familial mutation	Not reported	N/A	Yes	In some cases (6/22), parents provided written materials. All offspring who received written materials indicated that they were helpful. Among offspring who did not receive written	Not reported
Other(s)	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Yes	Not reported	Not reported	N/A	Some parents and young adults indicated that genetic counseling had prompted debate among family members about the implications of knowing about a hereditary condition in the family.
Other(s)	Mothers, parents, grandmothers, or genetic counselor. | Almost all the daughters were informed of their mother's result in	More than half said they were informed 'within hours' or a few days after their mother had received her	None was younger than 12 years of age when informed; about half were told	Some messages were open and empowering, some attempted to model reassurance for the daughter, others were quite directive, and some conveyed deep parental fears. What daughters reported strongly suggested that they had received their parent's message that	Overview of what the daughters reported was the information parents stressed in disclosing the maternal result: - She wants me to get testing - She doesn't want me to get testing now - It's your decision if you want testing - Wants me to know the family history	For most, the result disclosure was not in a 'sit-down', formal family meeting but, rather, occurred as a casual encounter. About 10% learned about their mother's result over the telephone; several learned about	Yes	Not reported	Not reported	N/A	Not reported
Mother	N/A	Not reported	Information disclosed fell into four age categories; 8-11, 12-14, 15-17 and > 18 years.	In the majority of families with children <11 years old, information focused on events that the child would witness for example reasons for hospitalisation, which included discussions surrounding a family gene and breast cancer diagnosis, (prophylactic) mastectomy and/or breast reconstruction. For this age group	N/A	Mothers affected or at risk of BRCA gene mutation predominately disclosed genetic risk information to their children, in the home, typically in the children's bedroom. Where siblings were of similar ages, gender and personalities	Not reported	N/A	Yes	By 15-17 years old, young people were considered adequately mature to cope with and understand the potential risk of breast cancer to themselves and were given more information about risk management strategies. At 18 years old.	Some young people were aware that their mothers were putting on a "brave face". This comprehension did not however, encourage communication because children and young people were afraid that asking questions would upset their mothers further.
Other(s)	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	N/A	Not reported	N/A	Not reported
Mother	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported
Other(s)	mother, father, aunt, sibling, healthcare provider	Not reported	Not reported	Not reported	N/A	Five participants and their parents sought genetic counseling and testing together, generally when the parent or another close relative was peri-diagnostic. Many described open and fluid communication in conversation with parents and	Yes	Not reported	Yes	Many sought out information online or in professional journals.	Many described open and fluid communication in conversation with parents and providers about testing results, risk management, hopes and fears.
Other(s)	Family members (father, mother, sister, in-law, etc.)	The mean length of time since they had received their test results was 5.79 years.	Not reported	Not reported	N/A	Not reported	Yes	Many families expressed appreciation or a desire for external support from health professionals to process the experiences they had undergone by providing emotional support, facilitating communication and for one family, providing a follow-up consultation (n = 14). Of the families	No	Some families explicitly expressed a need for written material (F7) or family member discussions facilitated by a health professional (F1, F3, F4, F8, F10) to assist with family communication.	Family functioning assessed with the FES identified families that reported to openly communicate with their family about genetic risk (F1, F9, F12), whereas families that were less communicative scored lower (F4, F5, F6, F11, F14). Interestingly, some families despite scoring high on the
Other(s)	Both parents or other sources. 6 of 34 mature minors reported situations where the minor initially learned of their	(PARENT REPORT) The majority of parents felt it was important to take time to process one's own results before	8 years old.	Not reported	N/A	They (minors) were told at a young age and learned more details as they got older.	Not reported	N/A	No	14 of 34 mature minors reported they did not receive the information necessary to understand their risk.// While most parents reported they consulted the Internet, most mature minors reported they did	When asked whether parents in other families should talk to their kids about genetic risk, all mature minors interviewed responded 'yes', and when mature minors were asked whether they were happy their parent had disclosed GRI to them, all
Both parents	N/A	Years	Not reported	Not reported	N/A	The daughter said, "[My mother] sat me down and told me, 'I am not going to die like [my sister].' I was older and understood more, so it was OK because the cancer was so small and early".	Not reported	N/A	No	N/A	The younger daughter reported always leaving the room when discussions of breast cancer or risk took place.
Other(s)	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Yes	Not reported	Not reported	N/A	For many participants gaining confidence and engaging in open communication with family members was regarded as the main benefits, by giving them the tools and methods to communicate more effectively with each other and build their
Both parents	N/A	Not reported	Not reported	For those with mothers who were previously affected by breast/ovarian cancer and/or BRCA+, the content of communication focused more on supportive messaging and providing psychosocial reassurances so as to not transmit worry to family members.	"Umm, she wanted us to understand, like this [the results] she didn't want it to, like scare us. Like, she didn't want to scare us, like to think, like we have to think about this our whole lives because we could have it, too. And she didn't want us to think about, like how she could maybe get it again. She just wanted us to, like focus on living our lives and not be scared	Children reported a range of reactions to disclosure, with mixed feelings about what they learned. That is, children reported a multiplicity of positive, neutral, and negative responses to family communication. For example,	Not reported	it should be kept in mind that these discussions were unaided by healthcare professionals, occurred without the benefit of patient education materials, or the provision of psychosocial support families beyond mothers' genetic counseling.	Yes	Several children sought-out information on their own on the Internet or from science teachers at school. | Some wanted more facts and information about the test itself, its reliability, and general statistics about HBOC (i.e., medical information	Children reported that their mothers attempted to balance sharing factual information without raising alarm and this was common across ages. | cancer genetic health communication in families about HBOC and BRCA testing was relatively succinct and to the point ; one participant

2. How does family communication about genetic cancer risk impact children on psychosocial and behavioral outcomes?																																		
How did genetic cancer risk disclosure impact participants?														How do family communication impact participants?																				
Psychosocial outcomes								Behavioral outcomes					Free text	Psychosocial outcomes												Behavioral outcomes						Free text (experience on living in a at-risk/ genetic cancer risk family)
Uncertainty	Discomfort	Vulnerability	Guilt	Depression	Fear	Anxiety	Other(s) statements	Decision-making	Life projects/ planning	Involvement in new tasks	Hypervigilance	Other(s) statements		Psychological adjustment	Discomfort	Vulnerability	Shame	Depression	Fear	Anxiety	Self-esteem/ concept/ conscious	Body-image/ stigma	Reports/ Information on family relationships after disclosure (e.g.: the test result brought brothers closer together; the guilt in the relationship; dyadic etc.)	Other(s) statements	Coping strategies	Decision-making	Life projects/ planning	Hypervigilance	Perceived risk	Other(s) statements	
Another participant explained "... the whole 'ignorance is bliss' factor. So if you had a	"I guess if you found out that you do have a disease, it would be upsetting" (13-year-old) - repetido, está	Not reported	A child testing negative, whose 2 siblings have tested	"I guess if you found out that you do have a disease, it would be upsetting" (13-year-old) -	The participants (n = 6)	Half of the participants mentioned that knowing one's risk status,	The participants mentioned the possibility of shock, anger, disappointment, worry, and fear associated with testing. Three participants, all of whom tested positive, indicated	Of the four participants involved in the decision regarding testing, two (50%) reported that they considered the impact the results of the testing might have on their future	Some participants also mentioned possible benefits of genetic testing to the family and specifically future generations (n = 4;	The most frequently mentioned advantages of genetic testing for children (see Table 2) were to learn of risk status or genetic	Not reported	When asked how testing and the results shaped their outlook, all participants indicated no change or a positive change, even when their result was positive: "I don't really feel that it's	All of the participants who underwent genetic testing said that they would do the testing again.	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Most participants indicated that testing and the results have had no impact on family relationships (n = 4; 57%). The remaining three participants represented a single family in which two children tested positive and one negative. One participant from this family stated: "It probably brought me and my sister closer together because we now	Not reported	Not reported	The eight participants who knew they had been offered and/or had gone through testing	Not reported	Not reported	Not reported	N/A	N/A	
Not reported	Not reported	Not reported	Not reported	The sadness and social isolation triggered by watching older family members die created a void related to	Not reported	Not reported	N/A	Not reported	Longevity and life expectancy meant something different for women from multiple-case families: "Honestly, at that time, I was	Not reported	The vigilance that compels these women to seek early diagnostic care is rooted in their heightened awareness of death; proactive	N/A	N/A	The sadness and social isolation triggered by watching older family members die	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Watching the premature deaths of so many young family members contributed to the sense that life was short and needed to be lived to its fullest: "...when my mom passed away, I was relatively young...and I think
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	N/A	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	N/A		
Not reported	Not reported	Other descriptions representing this theme included "feeling doomed' or 'denying' the	Not reported	Other descriptions representing this theme included 'feeling	Some offspring reported feeling frightene d or	Some offspring reported feeling frightened or disturbed	When asked to describe their initial reaction when their parent shared this information, few offspring reported being surprised. Some offspring reported a	Not reported	The majority of offspring reported no impact on their childbearing plans; however, two sons reported that it might	Offspring were asked what they think they could do now to avoid cancer. The most frequent responses were to	Not reported	Offspring uniformly reported no negative impact on relationships within or outside their family. Many offspring reported that the information their parent shared influenced	Most offspring (17/22) reported that they felt they had a good understanding of the information shared, although many stated	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Offspring uniformly reported no negative impact on relationships within or outside their family.	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	Many offspring reported that the information their parent shared influenced their personal health	N/A
Not reported	Young adult participants rated the cancer diagnosis and treatment as more distressing than the decision	Proximal disease threat overshadows the long-term - "...The genetic testing, that wasn't distressing or	Not reported	Although some young adults did not feel that they required psychosocial support for their/their	Not reported	Both parents and young adult participants rated the cancer diagnosis	Young adult participants were hopeful about having better health outcomes (in testing) than older generations in their family.] Young adults in the study who were old enough to	Not reported	Not reported	Not reported	Not reported	N/A	N/A	Some young adults indicated that genetic counseling had prompted	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Emotional and social isolation - "It's an extremely rare condition, you can't talk to your friends about it. It's really alienating..." (F20,	Of families whose children tested positive for a CPS, some parents (n = 6/9, 66.6%) and all young adults (n =	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	N/A
Not reported	Not reported	The daughters said, if found to be carriers, that information could be 'dampening' or make them feel 'doomed' or	Not reported	Not reported	Disclosur e of the mother's mutation status to her daughter	General distress among DMC was not high. However, levels of	The daughters described the emotional volatility reflected in either increasing or decreasing concern from the time of disclosure until the time of the interview. For some,	Some participants said learning about their mother's result had immediately made them want to get tested. "I think I want to get tested as soon as possible, even	They worried about the impact genetic knowledge could have on their life plans, especially plans for childbearing. Many	Not reported	Knowledge of their mother's risk status had not undermined their own sense of being generally healthy, and cancer risk did not permeate	Many daughters said they wished they were a bit more careful about their diet and physical exercise patterns, but most described their health as 'it's great', 'no major medical concerns', "I think I ama really	The daughters reported many foci for worries they associated with knowing their mother's hereditary cancer predisposition. These included worry about	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Many daughters said they wished they were a bit more careful about their diet and physical exercise patterns, but most described their health as 'it's great', 'no major medical concerns', "I think I am a really
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Selective genetic risk information in conjunction with the normalisation of cosmetic surgery impacted on children and young people's attitudes towards breast cancer and risk management procedures.	Greater knowledge of the BRCA mutation also caused young people to consider the impact that breast cancer risk might have on their future relationships	Not reported	Not reported	N/A	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	Some young people were aware that their mothers were	Not reported	At this time young people began to realise the potential implications of breast cancer risk for their own psychological	At this time young people began to realise the potential implications of breast cancer risk for their	Not reported	N/A	Not reported	Selective genetic risk information in conjunction with the normalisation of cosmetic surgery	Not reported	One young female participant demonstrated she understood the importance of monitoring and screening	Gendered disclosure lead several young males to perceive their risk of the BRCA gene mutation was	N/A	Interviews highlighted a discrepancy between parental perceptions of risk and what they disclosed to their children. Selective communication and playing down risks, particularly related to prophylactic surgery, impacted on children and young
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	N/A	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	As multiple reasons were possible per responder, percentages are not cumulative: half (47 %) included an emotional reason for DNA-testing, most	The majority of responders (78 %) had been influenced by the medical history of their parents, versus 13 % by that of sisters and 1 % of brothers. Nearly half of responders had received no advice of relatives or was supported in making
Not reported	Not reported	Though many children were young when their mother's cancer first occurred, themes that emerged included	Not reported	Children's scores on both the STAIC and RADS fell within normal limits. Children who reported	Though many children were young when their	Anxiety and depression. Children's scores on both the STAIC and RADS fell	Stress and worry related to family member's cancer. All of the children in this sample reported knowing, or having known, at least one first- or second-degree relative or close friend with	Children's rating of the relative importance of reasons for participating in cancer genetic testing upon reaching adulthood (n=20), by order of importance:	Not reported	Not reported	In terms of the impact of children's thoughts about their mother's cancer risk, over one-half (57%, n=8) stated that these thoughts affected their feelings	Behavior problems. On the YSR, average internalizing and externalizing scores were normal. Consistent with our findings on depression and anxiety, children who reported fewer internalizing (ω-0.63	Interest in cancer risk information. Among the 20 children enrolled in this study, 18 (90%) expressed at least a fair amount of interest in knowing as adults	several outcome indicators suggest that children's psychological adjustment	Not reported	Not reported	Not reported	Anxiety and depression: Children's scores on both the STAIC and RADS fell	Consisten t with the quantitati ve data are the qualitativ e	Anxiety and depressio n: Children's scores on both the	Not reported	Not reported	Not reported	Stress and worry related to family member's cancer: Among children with a previously affected mother, 57% (n=8) thought	Children whose mothers had had breast or ovarian cancer were asked an openended question about	Among the 20 children enrolled in this study, 18 (90%) expressed at least a fair amount of	Not reported	Not reported	When the focus of attention turns to cancer, nearly one-half of children wa	Behavior problems: On the YSR, average internalizing and externalizing scores were normal. Consistent with our findings on	Children whose mothers had had breast or ovarian cancer were asked an open-ended question about their experience with this event. Review of their responses (n=8) revealed that though many children were
Not reported	Not reported	Despite being given low absolute breast and ovarian cancer risk estimates for their age during genetic testing	Not reported	Not reported	Not reported	Not reported	N/A	Five had completed risk-reducing bilateral mastectomy (RRBM) or had one scheduled in the months following the interview, and none had scheduled or completed risk-reducing salpingo-	Not reported	Nichelle shared: "I'm 21, so I can't do mammograms and MRIs until I turn 25. So, it's four years of just self-examinations and clinical examinations.	Not reported	N/A	N/A	Not reported	Some participants using shared providers experience d a truncated	Participants felt vulnerable to an impending cancer diagnosis and felt pressured to take action	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	For others, parents exerted pressure to pursue risk reduction.	Although parents remained a significant source of emotional and pragmatic support for young mutation-positive women, parental	Not reported	For others, parents exerted pressure to pursue risk reduction. - repetido atrás	Not reported	Not reported	Not reported	N/A	N/A
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	N/A	Young adults often expressed responsibility for siblings and cousins. Daughters felt responsible for informing brothers, particularly when they	Psychosocial adjustment of young adults assessed with the PAHC alone identified	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Young adults often expressed responsibility for siblings and cousins. Daughters felt responsible for informing brothers, particularly when they perceived their mother to be less proactive (F1, F13). Jessica (28-year-old, carrier) was more concerned about her younger cousins obtaining accurate information than she was about their mother receiving information. "I think it's because	Sons of fathers who had died from prostate cancer were more likely to report concern about their cancer risk (F9, F16).	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Parental guilt about passing on the pathogenic variant also made it difficult for children in the parent–child relationship to express their concerns freely (F4, F6, F18, F19). For example, Gemma (21-year-old; carrier) said she was unable to talk to
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	N/A	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	17 of 34 mature minors reported they could sense	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	N/A		
Not reported	Not reported	Not reported	Not reported	Not reported	The two daughters reported that they frequentl y claimed to be sick	The son said he was paranoid about getting cancer himself.	N/A	Family B - "I want to get tested when I'm older so I can make sure to get it out if there is something there, just the same as my mom did. If I were my mother or aunt, I would	Not reported	Not reported	Not reported	N/A	The younger daughter said she was being tested because her sister wanted her to do it but did not plan to obtain her results; she said "[I] just didn't want	Not reported	Family E - The older daughter said that negative consequenc es of	Mrs. A described a sense of inevitability from childhood that she and	Not reported	Not reported	Family A - Not when the older daughter began to develop "breast	Not reported	Not reported	Not reported	Not reported	About thinking the mother's risk, the daughter responded, "I would say I think about it more than they (parents) do. It's in	Not reported	Family B - daughter (about mammograms, breast self-examinations, and Pap	The son said he did not know much about the hereditary cancer risk but did have	Not reported	"When my mom started talking to me about that (begin mammograms, breast self-	Family A - At home, the children played "chemo" and "finger stick" and the youngest girl repeatedly drew suture marks like her	Mrs. A reported that, as teenagers, the sisters taunted each other with gallows humor about who would be the first to "get it.// The children played "chemo" and "finger stick" and the youngest girl repeatedly
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	Not reported	Not reported	Not reported	Not reported	N/A	N/A	Not reported	Not reported	Not reported	Children and young people reported feeling embarass ed in	Not reported	Not reported	Children and young people reported feeling anxious, embarrass ed or	Not reported	MFDGs need to be delivered at weekends as families do not want to talk to their employers or children's	For many participants gaining confidence and engaging in open communication with family members was regarded as the main benefits, by giving them the tools and methods to communicate more effectively with each other and build their confidence in posing and/or answering questions effectively (see Figure 2- A picture showing a family hugging and after attending the	Children and young people reported feeling anxious, embarrassed or nervous in attempting to initiate conversations about	Not reported	Not reported	Not reported	Not reported	Not reported	N/A	After MFDG pilot, youth reported: - Through their interactions with other families and sharing their experiences with peers, who were able to understand and relate to what it is like living with an HBC, they thought the
"And then when she told me, then I guess I was, umm, it was kind of unsettling because that means that I	Not reported	Not reported	Not reported	Not reported	"And, umm, like I knew it ran in the family, so but my mom had always	Not reported	Children were concerned that they could not obtain definitive or quantitative information about their own cancer risks in specific and accurate numbers.// Half of children (50%) reported during interview that	Not reported	"Umm, it's [HBOC and genetic testing] definitely made me reflect more on what could happen to me, umm, because it was sort of—it was out of the blue for me.	All children (100%) interviewed noted that they followed or would seek to follow a more healthy life-style to influence the portion of cancer risk that may be under	Not reported	N/A	Although the vast majority (91%) of children articulated thoughts and feelings about how they wanted parents to communicate with them about HBOC and	Children reported a range of reactions to disclosure, with mixed feelings about what	Not reported	Not reported	Not reported	Not reported	Not reported	Although the vast majority (91%) of children articulate d thoughts	Not reported	Not reported	Others were reluctant to reveal any negative thoughts and feelings to their mothers because they did not want to increase maternal stress. There, children appeared to protectively buffer their parents against undue concern about the child's emotional state and alleviate parental guilt about HBOC or the possibility of having transmitted an altered BRCA gene to their	On the whole, children did not report long-term, persistent, or adverse reactions to the communication of their mothers' BRCA test results. In	50% of the children affirmed that they felt they could cope well with any future health issues that might arise,	Not reported	Not reported	Not reported	All children (many of them male and 12–17 years) whose mothers had received uninformative	Many children had only vague remembrances of the very first discussion and a few conflated learning about BRCA testing with learning about	That is, children of mothers with a BRCA+ result appeared no more or less likely to endorse this than did the others. Children's concerns about cancer (more broadly) seemingly stemmed from the totality of the lived experiences

3. What are the child's preferences regarding genetic cancer risk communication within the family?						
Disclosure event			Family communication process			
Do participants report what they liked? If yes, describe (facilitators factors, positive experiences).	**Do participants report what they didn't like? If yes, describe** (barriers factors, negative experiences).	**Do participants report what could have been done differently/ happened differently? Describe** (e.g.: more or less information/ tools, inclusion in the decision-making, etc.).	**Do participants report what they liked? If yes, describe** (facilitators factors, positive experiences).	**Do participants report what they didn't like? If yes, describe** (barriers factors, negative experiences).	**Do participants report what could have been done differently/ happened differently? Describe** (e.g.: more or less information/ tools, inclusion in the decision-making, etc.).	**Free text**
All eight participants indicated that they had enough say in the decision-making process. Those not involved in the testing decision reported that this was appropriate given their age at the time. They also stated that they trusted their parents' judgment, would have gone along with the decision if asked,	Not reported	All eight participants indicated that they had enough say in the decision-making process. Those not involved in the testing decision reported that this was appropriate given their age at the time. They also stated that they trusted their parents' judgment, would have gone along with the decision if asked,	Not reported	Not reported	Not reported	About testing, all of the participants indicated that children should be involved in the decision regarding whether or not to get genetic testing, to an extent, and that parents could unilaterally decide to have their children tested for genetic cancer predispositions without involving their child
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A
Not reported	Not reported	Sons (8 of 10) and daughters (7 of 12) supported the genetic testing of minors. One frequently reported reason for not supporting the testing of minors was the lack of a medical indication: "You can't do anything at that age. I don't really see how	Not reported	Not reported	Not reported	Sons and daughters: 1. Opposed to genetic testing of minors (). Reasons: - There is no medical indication 3 females, 1 male
Not reported	Not reported	In some cases (6/22), parents provided written materials. All offspring who received written materials indicated that they were helpful. Among offspring who did not receive written materials, the majority felt that written materials could have been	Not reported	Not reported	Not reported	N/A
Perceived benefits of testing - "It's just knowing about it and managing it and then I won't get into the situation that my dad got in" (F26, young adult,positive, affected). | Young adults commonly expressed satisfaction with the care they received from genetic services. Continuity of care,	When participants indicated that they were not satisfied with the quality of care received, this was commonly related to experiences of lack of continuity of care.	Information and support needs. Need to "remember what I do and don't know" - "I understood it when [the genetic professional] put it down, she made it sound very simple, but explaining it now I have to think back and remember it exactly…I'm going to be kicking myself for not being able	Some parents and young adults indicated that genetic counseling had prompted debate among family members about the implications of knowing about a hereditary condition in the family.	Not reported	Support in times of transition - "I've been more aware of getting more information [so I can] explain it to my girlfriend because I feel like I need to know more answers for her" (F10, young adult (male), positive, affected). // Participants highlighted that their desire for follow-up from genetic services was	N/A
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	So, although most daughters clearly conveyed that they had heard their mother's BRCA1/2 test result and had received their parent's message about the information being important, gaps and misconceptions in these young women's understanding of the relevant facts were common.
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A
Not reported	Not reported	Need for information (scale 1–10): 9.// Recommended age for DNA-testing: 2 % of responders would recommend DNA-testing from birth, 4 % from 16 years, 24 % from 18 years, 20 % from 20 years, 18 % from an age between 21 and 24 years, 18 % from 25 years and 2 % from 30 to 35 years. 12 % of responders did not report an age as this	Not reported	Not reported	Not reported	N/A
Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	N/A
Julia described the experience of having the family history mapped out for her already: "The (genetic counselor) already had an overall background about our family history and had worked with both my aunt and my mother and was just really, really exceptional." | Many described open and fluid communication in conversation with	Some participants using shared providers experienced a truncated process of genetic counseling; these participants perceived their mutation-positive parent as harboring guilt and self-blame about having passed the mutation to the adult child. As a result, during joint counseling sessions, participants hid worries, denied concerns, or refrained from	Not reported	Not reported	Not reported	Not reported	N/A
Many families expressed appreciation or a desire for external support from health professionals to process the experiences they had undergone by providing emotional support, facilitating communication and for one family, providing a follow-up consultation (n = 14).	Not reported	Of the young adults that were identified to have a psychosocial adjustment difficulty (n = 14), 64% reported needing emotional support during the interview (n = 9).// Some families explicitly expressed a need for written material (F7) or family member discussions facilitated by a health professional (F1, F3, F4, F8, F10) to assist with	Not reported	The daughter stated (Elizabeth, 22), "my family isn't great in saying what they need from one another" (F10).	Robert (26-year-old, non-carrier) wished his father, Fred (56-year-old, carrier) was more forthright and open about his test results. Robert said Fred was not open with him "because he doesn't want to see it as a sign of weakness" (F7). // Interestingly, of the young adults that were identified to have a psychosocial adjustment difficulty (n = 14),	N/A
A majority of mature minors reported something that 'went well' in their experience with learning GRI was that they were told at a young age and learned more details as they got older. Mature minors reported having first learned about GRI as young as age 8. "I knew that my mom had it because it was genetic. But I didn't know	14 of 34 mature minors reported they did not receive the information necessary to understand their risk.	14 of 34 mature minors reported they did not receive the information necessary to understand their risk, and 21 of 34 mature minors reported they wished their parent had given them more information: "Well, I feel like they didn't explain it enough. I mean, they didn't make me feel scared, but they didn't explain it enough for me to fully	When asked whether parents in other families should talk to their kids about genetic risk, all mature minors interviewed responded 'yes', and when mature minors were asked whether they were happy their parent had disclosed GRI to them, all mature minors who responded answered 'yes'.	21 of 34 mature minors reported they wished their parent had given them more information. "I think my father assumed that a lot of it, I wouldn't understand, and he didn't want to tell me. And I would go out and find that information some other ways, and then I would talk to him about it. I'd say, "Well, Dad, I found out about this [other	When mature minors were asked how parents should communicate information, almost all mature minors stated that the parent should communicate the information honestly in a way that does not scare the child. "You can't sugar coat it. You just have to tell them exactly what it is. If you tell them everything's 100-percent perfect and	Zero mature minors reported that their parent gave them too much information. | When asked whether parents in other families should talk to their kids about genetic risk, all mature minors interviewed responded 'yes', and when mature minors were asked whether they were happy their parent had disclosed GRI to them, all mature
Not reported	Not reported	Not reported	Not reported	Not reported	Older daughter's comments that open communication would have been preferable.	N/A
AFTER PILOT: - All participants (parents, children, and young people) reported that an intervention specifically designed for families affected by or at risk from IGCs was very necessary to facilitate family communication.	AFTER PILOT: - All participants (parents, young people, and children) were adamant that hospitals or medical facilities should not be used because they conjure up memories.. -The MFDG should be no longer than half a	Not reported	All participants (parents, children, and young people) reported that an intervention specifically designed for families affected by or at risk from IGCs was very necessary to facilitate family communication. Participants appreciated that engaging in activities would encourage discussions and help family	Children and young people specifically wanted their friends to attend (the discussion group) because they would be more comfortable talking to their friends than to their parents or siblings. Despite this, the majority of children and young people stated that being in an MFDG (multi-family	Family members wanted to decide who attended the MFDG and whether they should invite extended family members or friends. Children and young people specifically wanted their friends to attend because they would be more comfortable talking to their friends than to their parents or siblings.	Initially participants, especially young people and children, found the idea of sharing personal thoughts, feelings, and experiences with other families daunting or intimidating. However, through their interactions with other families and sharing their experiences with peers, who were able to understand
Nearly all children (94%) affirmed the importance of these conversations both initially and with the passage of time. (…) children reported that their mothers attempted to balance sharing factual information without raising alarm and this was common across ages.	Not reported	There were 68% of interviewed children with questions that were unanswered, or more frequently, were unanswerable at the time of the disclosure: some wanted more facts and information about the test itself, its reliability, and general statistics about HBOC (i.e., medical information typically encountered during genetic counseling	Nearly all children (94%) affirmed the importance of these conversations (about HBOC and BRCA testing) both initially and with the passage of time.	Not reported	All children (100%) also articulated preferences for how they would like to learn more about genetics and health. Among 94% of children, this included discussion of children having genetic testing—and a slight majority (59%) reported deferring this topic for serious consideration until adulthood. In those instances, children reported they	N/A

Conclusions		
Study limitations (copy-paste)	**Do the authors propose guidelines based on the results/ future directions? If yes, describe.**	**Notes** (free text)
The findings of this study need to be considered within the context of its methodological limitations. Our sample was small, heterogeneous and may be biased. Those with more negative views or experiences	Parctice recomendation - importance of developmentally-appropriate conversations between children, parents and providers prior to genetic testing of children.	N/A
Not reported	We recommend that providers who care for these women take a family-centred approach incorporating both mutation-positive and mutation-negative women. This study suggests that we recognize that one cannot	N/A
The data are limited	Providing offspring access to testing may be a means for men to fulfill their perceived parental responsibility regarding their genetic risk.	N/A
Several limitations of this study that are inherent in the methodology must be acknowledged. First, the study cohort represents a select group who may not be the	Further research is necessary to evaluate relationships among content and methods of disclosure, adaptive psychological responses and engagement in risk-reducing	N/A
Limitations include the study's retrospective methodology and potential recall bias Our heterogeneous sample, although allowing us to determine important	Future studies should use prospective methodology and focus more closely on comparing cancer and genetics-related distress, and other psychosocial outcomes, between groups, including different CPSs, and	N/A
Like many study cohorts in research on hereditary cancer impact, our sample was not ethnically, geographically, or socioeconomically diverse, so our results may not generalize to more heterogeneous	- Counseling approaches to this population may require modification; daughters may need sequential counseling to consider first the importance of starting breast screening at age 25 years and then later	N/A
The study is limited by quite a small self-selecting sample size but included different types of family structures and socioeconomic backgrounds and data saturation was achieved. The study's strengths lie in its in-depth analysis of	this group is potentially biased because most of the participating mothers had or were awaiting mastectomies and no families came forward where the women at risk or affected were not taking a surgical option; this possibly delayed the need for these	By avoiding risk communication parents thought that they were protecting their children from harmful information however, avoiding or delaying disclosure had three profound effects on children
Our study is limited by a possible selection bias due to low response rate (51 %) and significantly fewer males responding (p<0.03). This is unsurprising: both males and younger patients are known to have lower participation rates	Decisional conflict could then be a target for future interventions. While standardized support by a social worker during the decision-making process is currently provided, this support could be made more effective by focusing on those young adults	N/A
As a prerequisite to conducting this type of research, we recognized it was important to protect the confidentiality of a parent's participation in testing against any unwanted disclosure to a child. Thus	For clinicians who consult on issues regarding children of parents participating in BRCA1/2 testing, our study highlights: - The sensitivity surround not to discuss testing with their children at this particular phase in the	N/A
Secondary data analysis is always limited by the design and shortcomings of the original studies (Thorne et al., 2004). AWL's participants were interviewed before and LMH's participants were interviewed shortly after passage of the	Research is needed to understand provider training and genetic service delivery, with particular attention to the implications of public policy shifts, specific to this young age group. We have implemented a separate protocol to study the	N/A
Family communication research is limited by the nature of the recruitment process since those who participate are more likely to have open lines of communication and are willing to share their experiences. The length of time since a family	In Australia, and most countries worldwide, the clinical procedure to inform relatives of a known BRCA1/2 pathogenic variant is to encourage the proband or presenting patient to disseminate the information to the rest of the family. However,	N/A
Parents and mature minors in this study self-selected to participate and share their experience communicating about GRI, resulting in a cohort biased in favor of sharing GRI with minors. Also, our cohort may be biased in favor of	Proposed guidelines 1. Minors should be informed of genetic risk information (GRI) 2. Parents should take time to process their own genetic risk or pathogenic variant status before informing their minor children of GRI	N/A
Not reported	The investigators believe that a larger, qualitative study is needed. A grounded theory approach could provide a theoretical framework that encompasses the issues, including influential factors,	N/A
- The GCs required more training and development than we had originally anticipated, which we hypothesise is largely due to the limited training in family systems therapy and the lack of experience in working with multiple families in a	- MFDGs need to be delivered at weekends as families do not want to talk to their employers or children's schools because they fear being stigmatised, an issue that was raised repeatedly during our MFDGs.	N/A
This study has several limitations, including the narrow diversity in the sample and underrepresentation of males and certain BRCA genotypes that precluded some subgroup analyses and the ability to draw more definitive	Presently, there remains a need for developmentally informed and evidence-based interventions to help parents (and especially mothers) make decisions about communicating with their children about HBOC and BRCA testing and promote	N/A

Appendix C. Summarized results table

	Year	Country (where the study took place)	Aim(s) of research	Study design (broad categorization)	Data collection procedure	Intervention? If yes, describe.	Materials (if questionnaires were used, name them)	Sample/ informants
RESULTS (15 papers)	Papers from 2001 to 2020	USA: 10; UK: 2; Australia: 2; Netherlands: 1.	Main aims of the papers: - explore perspectives, understandind, expectations and psychosocial implication of genetic testing and results; - risk perception; - experience of being negative in a positive family; - understand the content and method of disclosure; - experience with genetic services; - support needs;	Qualitative: 10 Mix-methods: 5	Qualitative: interview Mix-methods: interview + questionnaires	Yes, one intervention with focus groups.	Interviews were semi-structured and questionnaires were: - The Emotion Thermometers Tool (ETT), The Genetic Counseling Satisfaction Scale (GCSS), Multidimensional Impact of Cancer Risk Assessment (MICRA) questionnaire, Quality of Life Patient/Cancer Survivor (QoL-	Only Children (0 - 18 years old): 2 Retrospective children (adults referring to their experiences as < 18 years old): 5 Children AND Retrospective children: 1 Mix sample (children AND adults AND/OR families): 7
QUOTES								

Participants sociodemographic							Family clinical history			
N (total participants)	**N (children + retrospective children)**	**Age (mean, SD, range - total participants)**	**Age (mean, SD, range - children + retrospective children)**	**Sex (N female, N male - total participants)**	**Sex (N female, N male - children + retrospective children)**	**Research setting** (e.g.: hospital, genetic counseling service, e-mail, etc.)	**Family syndromes/ at-risk syndromes**		**Data on the age of onset of genetic cancer in the family**	**Characteristics of family history** (e.g.: the first member being tested, affected family members/ mutation carriers, mutation status, cancer diagnosis, deaths in the family, etc.)
							Li-Fraumeni syndrome: 1 Breast and ovarian cancer: 10 Other syndrome(s): (1) cancer-related genetic mutation, (2) Polycystic kidney disease (PKD), Multiple endocrine neoplasia (MEN), Familial adenomatous polyposis (FAP) - the others Inherited	Most of the papers are about breast and ovarian cancer (10), Li-Fraumeni syndrome (1), multiple syndromes (2) and other syndromes (2).		

Genetic counseling				
Applicant (who did the genetic test)	**Does/did the family attend genetic counseling?**	**If yes, were the children included?**	**Children's (0 - 18 years old) experience/feedback of genetic counseling.**	**Retrospective children's (adults referring to their experiences as < 18 years old) experience/feedback of genetic counseling.**
	Yes: 12 Not reported: 3	Yes: 1 No: 14	Because children didn't attend genetic counseling, there's no feedback of the service.	- All offspring thought it would be helpful to meet with a genetic counselor or other expert in general, and the majority indicated that they thought it would have been helpful when they learned of their parent's BRCA mutation. They indicated an interest in learning more about genetics and hereditary disease in general and the opportunity to ask specific questions. - Both parents and young adults commonly expressed satisfaction with the care they received
				"The [genetic professional] that we go to see has been with us throughout our entire life. So we're very privileged to have someone throughout all those years with us. It's almost like they're a family member".

1. How, when and what is discussed about genetic cancer risk in the family?

Who disclosure genetic cancer risk to the participants?	**Time interval between the test result and disclosure.**	**Age in years when told.**	**Content of disclosure** (what was said/ explained to the children. E.g.: who had genetic testing, the implications of the test result, etc.)	**How was the genetic cancer risk disclosure to the participants? Describe** (e.g.: by telephone, 'sit-down' family meeting, private conversation)	**During the disclosure phase, was there any contact with genetic counseling**	**If yes, describe how the genetic counseling service was involved in the disclosure.**	**Were tools involved in the disclosure?** (to assist or support disclosure. E.g.: literacy resources, communication tools,	**If yes, describe.**	**How do family communicate about genetic cancer/risk** (e.g.: open communication; limited communication; with fear, coping strategies; etc.)
Mother: 2 Both parents: 3 Other(s): (4) Mothers, parents, grandmothers, aunt, sibling, in-law, or genetic counselor. Not reported: 6	One paper says that they were informed 'within hours' or a few days after their mother had received her result. Most others did not recall how long, after their mother learned her result, they were informed. Others papers recall years between the test result and disclosure.	The youngest age at disclosure was 8 years old.	- In some cases, the genetic mutation was not initially described, although the parents (all mothers) did discuss the hereditary risk of cancer in the family. - The topics offspring most frequently reported included who had genetic testing, the implications of the test result for the risk of developing cancer (for the parent, other family members or for the offspring) and the ability for the offspring to have genetic testing.	- Parents shared the results alone or with their spouses. - Daughters were more likely to learn of the familial mutation from a mother alone than sons. - In some cases, offspring's siblings or other family members were present during disclosure. - Some offspring reported learning the information by telephone or while driving in	Yes: 5 Not reported: 10	Normalizing emotional responses and exploring possible lifestyle changes or raising topics that are potentially emotionally difficult (e.g., guilt) were other benefits raised by participants to facilitate family discussions. The families that expressed a desire for family discussions had a family functioning score that depicted relatively higher scores in cohesiveness, expressiveness and lower on conflict.	Yes: 4 No: 3 Not reported: 8	- written materials - internet and professional journals - health-care providers, family, school (science teachers) and books - family member discussions facilitated by a health professional to assist with family communication	- Some parents and young adults indicated that genetic counseling had prompted debate among family members about the implications of knowing about a hereditary condition in the family. - Some young people were aware that their mothers were putting on a "brave face". This comprehension did not however, encourage communication because children and
			- *"But she had always pushed just how serious it was and that it was kind of a big deal... it kind of scared me. But she also — she wouldn't say it in a scary way-she would just sort of stress it and I sort of preferred not to think about it."* - *"He [dad] said that testing had been done on his side of the family and a gene was found that makes women prone to breast cancer. He had testing and then brought it up over dinner*	*"[My mother] sat me down and told me, 'I am not going to die like [my sister].' I was older and understood more, so it was OK because the cancer was so small and early."*		*"I didn't know that mum felt guilty…and [aunt said]…'awh it sucks that my sister is going through this', so…maybe it is good for…a health professional to probe and ask questions, and go, oh you felt this but you didn't know that she felt this…and I can go, 'that's a stupid thing to feel, mum don't feel guilty about that'…"*			- *"My family isn't great in saying what they need from one another."* - *"I think it's the responsibility of your parents to share that information with you if it's going to affect you or affect your family. I think that it's irresponsible not to talk to your kids about that in whatever way that you may need to."* - *"I just don't think it would be possible for me to not know something was going on.*
			Overview of what the daughters reported was the information parents stressed in disclosing the maternal result (individual, open-ended responses): - She wants me to get testing - She doesn't want me to get testing now - It's your decision if you want testing - Wants me to know the family history - Have a good doctor						

2. How does family communication about genetic cancer risk impact children on psychosocial and behavioral outcomes?																																		
How did genetic cancer risk disclosure impact participants?																	How do family communication impact participants?																	
Psychosocial outcomes								Behavioral outcomes					Free text	Psychosocial outcomes									Behavioral outcomes						Free text (experience on living in a at-risk/ genetic cancer risk family)	
Uncertainty	Discomfort	Vulnerability	Guilt	Depression	Fear	Anxiety	Other(s) statements	Decision-making	Life projects/ planning	Involvement in new tasks	Hypervigilance	Other(s) statements		Psychological adjustment	Discomfort	Vulnerability	Depression	Fear	Anxiety	Self-esteem/ concept/ conscious	Body-image/ stigma	Reports/ Information on family relationships after disclosure (e.g.: the test result brought brothers closer together; the guilt in the relationship; dyadic; etc.)	Other(s) statements	Coping strategies	Decision-making	Life projects/ planning	Hypervigilance	Perceived risk	Other(s) statements		
- Participants were concerned that they could not obtain definitive or quantitative information about their own cancer risks in specific and accurate numbers. These children wanted to understand if they themselves were at greater or lesser risk for cancer in numerical and more absolute terms, especially in light of practices	Young adult participants rated the cancer diagnosis and treatment as more distressing than the decision to have genetic counseling/testing, and the genetic counseling/testing process itself.	- The daughters said, if found to be carriers, that information could be 'dampening' or make them feel 'doomed' or 'destroy' their life. - The daughters described the emotional volatility reflected in either increasing or decreasing concern from the time of disclosure until the		- The participants mentioned the possibility of disappointment associated with testing. - The sadness and social isolation triggered by watching older	- The participants mentioned the possibility of fear associated with testing. - Some offspring reported feeling frightened or disturbed. - Some offspring reported a concern about the increased risk of receiving a cancer diagnosis, either for	- Participants mentioned that knowing one's risk status, regardless of the result, allows one to prepare, reduces anxiety and increases one's power in the situation. - Participants reported that negative emotions associated with a positive result were a potential disadvantage of testing and/or that	- Participants that tested positive, indicated that the seriousness of having this mutation/deletion and the lifelong implications did not set in until after testing when the details of the surveillance procedure were known. They indicated that they have now accepted this	- Participants involved in the decision regarding testing reported that they considered the impact the results of the testing might have on their future prior to testing. Some considered the need for cancer surveillance and health behavior changes,	- Participants mentioned possible benefits of genetic testing to the family and specifically future generations. - Participants spoke about the importance of knowing their genetic status in	- The most frequently mentioned advantages of genetic testing for children were to learn of risk status or genetic predisposition for disease and to allow for disease prevention efforts	- The vigilance that compels these women to seek early diagnostic care is rooted in their heightened awareness of death. - One participant worried that if she knew she was	- When asked how testing and the results shaped their outlook, all participants indicated no change or a positive change, even when their result was positive. - A range of perceived advantages to testing were reported including: a) learning of risk status; b) allowing for	- Although many offspring reported potential negative aspects to learning of a familial mutation in general, the majority (15/22) reported no personal negative aspects for themselves. - Children expressed at	- The sadness and social isolation triggered by watching older family members die created a void related to familial	Some participants using shared providers experience d a truncated process of genetic counseling,	- Participants felt vulnerable to an impending cancer diagnosis and felt pressured to take action before they crossed a	Anxiety and depression: Children's scores on both the STAIC and RADS fell within normal limits. Children who reported themselves to be in better overall	- Some young people were aware that their mothers were putting on a "brave face". This comprehension did not however, encourage communication	- Anxiety and depression: Children's scores on both the STAIC and RADS fell within normal limits. Children who reported themselves to be in better overall	At this time young people began to realise the potential implications of breast cancer risk for their own psychological wellbeing, particularly to their self-esteem and – going on one	MFDGs need to be delivered at weekends as families do not want to talk to their employers or children's schools	- Most participants indicated that testing and the results have had no impact on family relationships. - No participants indicated that testing had an impact on their friendships. - Offspring uniformly reported no negative impact on relationships within or outside their family. - For others, parents exerted pressure to pursue risk reduction. - Young adults often expressed responsibility for siblings and cousins. Daughters felt responsible for	Initially participants, especially young people and children, found the idea of sharing personal thoughts, feelings, and experiences with other families daunting or intimidating. However, through	- Of families whose children tested positive for a CPS, some parents (n = 6/9, 66.8%) and all young adults (n = 4/4, 100%) indicated that the positive test	- Two participants, in the same family, over age 16 at the time of the decision and undergoing predictive testing,	The son said he did not know much about the hereditary cancer risk but did have concerns for his sister and might consider	One young female participant demonstrated she understood the importance of monitoring and screening as a risk management strategy but	- Sons of fathers who had died from prostate cancer were more likely to report concern about their cancer risk. - Gendered	- At home, the children played "chemo" and "finger stick" and the youngest girl repeatedly drew suture marks like her mother's on her own chest. The two daughters reported	- Watching the premature deaths of so many young family members contributed to the sense that life was short and needed to be lived to its fullest. - Levels of cancer-related distress were considerably higher on both the study-specific self-report questionnaire and the IES
- "...the whole 'ignorance is bliss' factor. So if you had a gene mutation, it doesn't necessarily mean that you're going to develop full-blown cancer, but it's there and (confirmed by testing) it's something you have to live with." - "And then when she told me, then I guess I was, umm, it was kind of unsettling because that means that			A child testing negative, whose 2 siblings have tested positive, reported: *"A feeling of guilt that I don't have it. My brother and sister need to deal with this for the rest of their lives."*	*"I guess if you found out that you do have a disease, it would be upsetting."*	- *"I was shocked, scared. I wondered if I was going to get the gene and realized I could pass it to my [future] kids. I would feel like it was my fault if they got cancer"* - *"I was nervous for her [mother] more than myself right away and I was nervous for like a year after*	- *"I really wanted to know whether or not I had that [the mutation], so that I wouldn't have to worry anymore about if I had it or not."* - "I was a little worried, but then I got over it and I was like, okay, I'm all right now and now we know *that I can be checked up."* - *"Knowing or not doesn't change* anything in terms of my actual risk	- *"It didn't sink in at first, but once I started having to get all of the testing, it sunk in that I am going to have to deal with this for the rest of my life."* - *"It was surprising, but helped explain all the breast cancer in the family. I wasn't worried about myself,*	- *"I was just kinda thinking about, you know, that [positive test result] could be hard with relationships. Like if you were in a relationship with someone and you knew that you had this ticking time bomb within you"* - *"I think I want to get*	- *"I guess if something came up with my kids and they needed to know if I carried the gene, then, you know, I would do it [genetic testing] for their sake"* - *"It made me have a*	- About the advantages of genetic testing for children: *"...it shows what kind of genes you have and the risk of when – like if you might get a disease. It tells the doctors in advance so they can fix it maybe,*		- *"I don't really feel that it's changed my sense of who I am or what I want to do with my life, or what my life options are in any way"*	- *"[It could cause] confusion, concern. Someone may be too young to understand the information. Also, individuals may feel this is a death warrant"*	- *"...I didn't have a lot of women in my lifetime, family members in my lifetime that I was talking to about*	- Alysha went on to discuss receiving her test results with her parents and her brother present: *"It*		and social isolation - *"It's an extremely rare condition, you can't talk to your friends about it. It's*	- *"Like she tried to make it look like it didn't affect her ..."* - *"She made like a not a joke out of it but she made it seem like everything was ok, like she didn't*	*"And then when she [mother] got the testing, I was still, like I didn't like talking about it [BRCA] because I keep things to myself. I don't know whether it's, like, a sense*	– going on one beach and wearing a bikini, I think that was one of the *factors with my mum, like you know [in choosing]* to have a reconstruction, and *I know that that would affect me as*		- *"It probably brought me and my sister closer together because we now have something (testing positive) that we can closely relate to"* - *"A feeling of guilt that I don't have it. My brother and sister need to deal with this for the rest of their lives"* - Jessica was more concerned about her younger cousins obtaining accurate information than she was about their mother receiving information, *"I think it's because they're my little cousins and I feel*	- About thinking the mother's risk, the daughter responded, *"I would say I think about it more than they [parents] do. It's in the past for them, but it's in my future."*	- *"But I guess, if something did come up, I would just roll with it as it came and assess my situation as I got through each step and, like, figure things out*	- *"I was allowed to decide whether I wanted to (testing) or not, but I discussed it with my parents and my sister and my family"*		*"I've got a friend whose just had a boob job ... and I was like I really want to have it done, I can't have it done now ... because there's a barrier*	- *"I understand [BRCA mutation] doesn't affect males that much, it like only gives them like a marginal*	*"I mean, yeah, I definitely worry about it [cancer], but it's—it's seemingly, like out of my control. I mean, both my grandmas had breast cancer. And now my aunt. So, it's, I mean, it's*	- *"...when my mom passed away, I was relatively young...and I think you just grow up a lot faster when you are around that, especially when there is more than one [death] in your family. So, I've always kind of made every day count"* - *"My aunty had to deal with it at a younger age, as opposed to*

3. What are the child's preferences regarding genetic cancer risk communication within the family?						
Disclosure event			**Family communication process**			**Free text**
Do participants report what they liked? If yes, describe (facilitators factors, positive experiences)**.**	**Do participants report what they didn't like? If yes, describe** (barriers factors, negative experiences)**.**	**Do participants report what could have been done differently/ happened differently? Describe** (e.g.: more or less information/ tools, inclusion in the decision-making, etc.)**.**	**Do participants report what they liked? If yes, describe** (facilitators factors, positive experiences)**.**	**Do participants report what they didn't like? If yes, describe** (barriers factors, negative experiences)**.**	**Do participants report what could have been done differently/ happened differently? Describe** (e.g.: more or less information/ tools, inclusion in the decision-making, etc.)**.**	
- Participants indicated that they had enough say in the decision-making process. Those not involved in the testing decision reported that this was appropriate given their age at the time. They also stated that they trusted their parents' judgment, would have gone along with the decision if asked, and knew that their parents had their best welfare in mind. - Young adults commonly expressed	- When participants indicated that they were not satisfied with the quality of care received, this was commonly related to experiences of lack of continuity of care. - Some participants using shared providers experienced a truncated process of genetic counseling; these participants perceived their mutation-positive parent as harboring guilt and self-blame about having passed the mutation to the adult child. As a result, during	- Although parents remained a significant source of emotional and pragmatic support for young mutation-positive women, parental involvement risked compromise of participant privacy and the loss of control over disclosure of their mutation status to loved ones. - Most sons and daughters supported the genetic testing of minors. One frequently reported reason for not supporting the	- When asked whether parents in other families should talk to their kids about genetic risk, all mature minors interviewed responded 'yes', and when mature minors were asked whether they were happy their parent had disclosed GRI to them, all mature minors who responded answered 'yes'. - All participants (parents, children, and young people) reported that an intervention specifically designed for families affected by	- Mature minors reported they wished their parent had given them more information. - Children and young people specifically wanted their friends to attend (the discussion group) because they would be more comfortable talking to their friends than to their parents or siblings. Despite this, the majority of children and young people stated that being in an MFDG (multi-family discussion groups) with their siblings was not	- Some participants indicated that not perceiving LFS as a death sentence influenced their decision. - Participants highlighted that their desire for follow-up from genetic services was heighted at times of family transition, for example, when children in the family reached adulthood and were making family planning decisions. Young adult participants also indicated that they would value support	- About testing, all of the participants indicated that children should be involved in the decision regarding whether or not to get genetic testing, to an extent, and that parents could unilaterally decide to have their children tested for genetic cancer predispositions without involving their child under certain circumstances. - Some participants felt that the age of the child was an important consideration.
- "My mom just pretty much told me, 'you're getting a blood test to see if you have this or not.' (Do you feel like you should have had more say in the decision?) No. I was younger and if my mom said, 'hey, do you want to go check to see if you have this or not?' I would have probably said yes anyway" *- "Knowing what's going on medically with my body is better than not. I am more mature now and have come to respect*	- In response to a question about disadvantages of genetic testing, one participant stated: *"Not really. I mean, learning you have it could be a shock, but in the end, it's better to know"*. Similarly, a second participant said, *"I know that some people do get worried about getting – like uncovering some type of thing that they would be worried about, but for me, no"*.	*- "You can't do anything at that age. I don't really see how the information could be helpful at this age."* *- "It is not really necessary to know that young …because you can't act on the information."* *- "If there is no physical health risk, give them knowledge early on. It is better to know at an early age because it could change early health habits."*		*- "My family isn't great in saying what they need from one another"* *- "I think my father assumed that a lot of it, I wouldn't understand, and he didn't want to tell me. And I would go out and find that information some other ways, and then I would talk to him about it. I'd say, "Well, Dad, I found out about this [other information about HD]. Why didn't you tell me [that]?" And he would just say, "Well, I*	*- "In some Li-Fraumeni families, it's basically a death sentence. Had that been the case in my family, had that been my experience with it, my desire to be tested may have been very, very different"* *- "I've been more aware of getting more information [so I can] explain it to my girlfriend because I feel like I need to know more answers for her"* - A son wished is father was more forthright	*- "…if you told someone who was six, they wouldn't get it at all, … but once they're nine or ten, I think they would understand what's going on"* *- "Parents make medical decisions for their children all the time and I think that if it were something important for the parents to know to be able to take care of their child, then I think they would be justified in doing it without involving the child"*

Conclusions		
Study limitations (copy-paste)	**Do the authors propose guidelines based on the results/ future directions? If yes, describe.**	**Notes** (free text)