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Family Communication About Genetic Risk of Hereditary Cardiomyopathies and Arrhythmias: An Integrative Review

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Abstract

Screening for hereditary cardiomyopathies and arrhythmias (HCA) may enable early detection, treatment, targeted surveillance, and result in effective prevention of debilitating complications and sudden cardiac death. Screening at-risk family members for HCA is conducted through cascade screening. Only half of at-risk family members are screened for HCA. To participate in screening, at-risk family members must be aware of their risk. This often relies on communication from diagnosed individuals to their relatives. However, family communication is not well understood and is ripe for developing interventions to improve screening rates. Until very recently, family communication of genetic risk has been mostly studied in non-cardiac disease. Using this non-cardiac literature, we developed the family communication of genetic risk (FCGR) conceptual framework. The FCGR has four main elements of the communication process: influential factors, communication strategies, communication occurrence, and reaction to communication. Using the FCGR, we conducted an integrated review of the available literature on genetic risk communication in HCA families. Descriptive analysis of 12 articles resulted in the development of categories describing details of the FCGR elements in the context of HCA. This review synthesizes what is known about influential factors, communication strategies, communication occurrence, and outcomes of communication in the context of HCA.

Keywords

hypertrophic cardiomyopathy; long QT syndrome; cascade screening; family communication; genetic risk; disclosure; inherited arrhythmia; inherited cardiomyopathies; sudden cardiac death

Introduction

Hereditary cardiomyopathies and arrhythmias (HCA) account for most sudden cardiac death (SCD) in young people (Vetter et al., 2008). The risk for SCD and other complications of

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Conflict of Interest

Lisa L. Shah and Sandra Daack-Hirsch declare that they have no conflict of interest.

Informed Consent

No protected health information or human subjects were used for this article. No animal studies were carried out by the authors for this article.

HCAs (e.g. heart failure, stroke, syncope) are reduced through interventions including medication, implanted defibrillators, and lifestyle modifications. HCAs including hypertrophic cardiomyopathy (HCM), arrhythmogenic right ventricular dysplasia/cardiomyopathy (ARVD/C), familial dilated cardiomyopathy (DCM), long QT syndrome (LQTS), and Brugada Syndrome (BS) are generally inherited in an autosomal dominant (AD) pattern. This means that all first-degree relatives (FDR; parents, siblings, and children) of a diagnosed individual have a 50% chance of having the same disease-causing genetic variant. When an individual is diagnosed with an HCA it is critical that relatives are notified of their risk, tested, and treated prior to development of life-threatening complications. Screening of at-risk family members is conducted through cascade screening, a process of sequentially testing at-risk family members for HCA through genetic testing and clinical exam, and is recommended by the American College of Cardiology Foundation/American Heart Association Task Force (Gersh et al., 2011) and the European Society of Cardiology (Ponikowski et al., 2016). However, to optimize cascade screening, the risk of disease must be communicated timely and accurately to all at-risk relatives.

In several nations worldwide, family communication is the principal mechanism through which family members are notified of increased risk for HCA. Recently, the American Heart Association (AHA)/American College of Cardiology (ACC) described a new quality measure recommending documentation that all FDRs of adult survivors of sudden cardiac arrest from an HCA have been notified of their need to be screened (Al-Khatib et al., 2017). However, the process and extent to which family communication about genetic risk for HCA occurs is poorly understood. Historically, research on family communication of genetic risk focused on non-cardiac diseases; more recently, we find research articles specific to HCAs.

The purpose of this review is to develop a conceptual framework for family communication about genetic risk for HCA in the pre-test phase. Genetic information disclosed in the pre-test phase leads to decisions about testing in at-risk relatives. This is distinct from the post-test state where genetic information is shared between individuals who have been informed about a positive or negative test result and discussions focus on adapting to the HCA diagnosis. This integrative review synthesizes the current literature on family communication of genetic risk for HCA. The outcome of this review is a conceptual framework that can guide clinical practice, research, and policy. First, we reviewed family communication about genetic risk in non-cardiac disease and identified major elements of family communication of genetic risk. This review culminated in the development of the Family Communication of Genetic Risk (FCGR) conceptual framework. We then applied the FCGR conceptual framework to guide an integrative review about family communication of genetic risk for HCA.

Family Communication of Genetic Risk (FCGR) Conceptual Framework

Multiple reviews of the literature on family communication of genetic risk include Wilson et al. (2004), Gaff et al. (2007), Wiseman, Dancyger, & Michie (2010), and Weins et al. (2013). None of these reviews included articles pertaining to families with HCAs. Major findings from each review are summarized in Table I and synthesized into four major elements of family communication of genetic risk: influential factors, communication strategies,

communication occurrence, and outcomes of communication. Influential factors motivate communication decisions and include disease, individual, family, and sociocultural factors that influence the extent of communication (e.g. what will be communicated, when, and to whom).

Communication strategies are the methods used to carry out communication in families. Communication occurrence is defined as how many family members were told about the genetic risk. Outcomes of communication are the emotions, behaviors, and actions resulting from communication experienced by probands, relatives, or families. Figure 1 organizes these four major elements into the family communication of genetic risk conceptual framework (FCGR).

Limitations of reviewed studies included small samples sizes, measurement of attitudes and not actual behavior, data collection soon after undergoing genetic testing which may not have given people enough time to communicate the risk to their relatives, over reporting of communication extremes, absence of studies describing the content of communication, lack of cultural diversity, no reports on the impact of communication on individuals or families, underdevelopment of the unique and sensitive issues related to communication genetic risk to young children, and study focus on general disease communication rather than risk communication (Gaff et al., 2007; Weins et al., 2013; Wilson et al., 2004; Wiseman et al., 2010).

Methods

The FCGR conceptual framework (Figure 1) was used to guide the analysis of this subsequent integrative review on family communication about genetic risk for HCAs. We applied methodology described by Whittemore & Knafl (2005) to conduct the integrative review in five stages: 1. Problem Identification, 2. Literature Search, 3. Data Evaluation, 4. Data Analysis, and 5. Presentation of Results.

Stage 1: Problem Identification

About half of at-risk family members are not screened for HCAs, placing large numbers of individuals at high risk for life-threatening complications of a disease amenable to treatment (Christiaans et al., 2008; Hanninen et al., 2015; Miller et al., 2013). One critical step in cascade screening for HCAs is ensuring that at-risk family members are aware of their increased risk and the need for screening. This generally requires accurate and timely communication from diagnosed individuals to their at-risk family members. However, family communication about genetic risk for HCA is not well understood. This integrative review addresses the problem that family communication about genetic risk, a critical step in cascade screening, is poorly understood in the context of HCA. Improved understanding of how families communicate about genetic risk for HCA may then help address the problem of low screening rates for HCA.

Stage 2: Literature Search

Identification of all relevant literature on family communication about genetic risk for HCA was conducted through electronic database searching and ancestry searching. Gray literature

was not included. Six electronic databases were searched (PubMed, CINAHL, PsychINFO, Scopus, EMBASE, and Family Studies Abstracts) on March 14, 2016 without limits or filters. Search terms were based on terms used in the non-cardiac literature reviews (Wilson et al., 2004; Gaff, et al., 2007; Wiseman, et al., 2010) with additional terms used to focus the search on genetic cardiac diseases, specifically HCM and LQTS. Search terms included long QT syndrome [MeSH], hypertrophic cardiomyopathy [MeSH], inherited heart conditions, inherited heart disease, sudden cardiac death, genetic risk communication, disclosure, family communication, and cascade screening. Endnote X7 was used to manage citations from the multiple searches.

Articles were excluded if they were not in English; reported family screening behaviors only without mention of communication; focused on provider to patient communication, clinical management of individuals, disease pathophysiology or pharmacology, population screening or community programs, registries, or multidisciplinary care programs; published conference abstracts; did not include an inherited cardiomyopathy or channelopathy that can result in SCD; or focused on bereavement or end of life care.

The electronic database search generated 506 articles. Duplicate articles were removed resulting in 317 articles. LS reviewed the title and abstract of these 317 articles using the inclusion/exclusion criteria and excluded 235 articles, leaving 82 articles requiring a full text review. Articles with no abstract were automatically included for a full text review. LS then reviewed the full text of all 82 articles using the inclusion/exclusion criteria and excluded 72 articles. Ancestry search methods identified eight additional articles for review from the full-text review of 82 articles and their bibliographies. The full texts of these eight articles were reviewed, applying the inclusion/exclusion criteria and excluded seven articles.

Eleven articles were used for initial data analysis. However, to ensure that all relevant literature was included in analysis, a final search of the electronic databases was repeated prior to publication using limits to identify literature published between March 15, 2016 and June 6, 2017. This search yielded 93 articles which were reviewed using the same process described previously. This final search resulted in the addition of one article and a final sample of 12 articles included in the analysis. Figure 2 illustrates the process of article identification, screening, selection, and inclusion in an adapted PRISMA flow diagram.

Stage 3: Data Evaluation of HCA Literature

The final sample of 12 articles included qualitative, quantitative, mixed methods, ethical case study, and personal narrative designs. Kmet, Lee, & Cook (2004) quality assessment criteria were used to evaluate qualitative, quantitative, and mixed methods studies. Fourteen criteria were used to evaluate quantitative studies and 10 criteria were used for qualitative studies. Criteria were evaluated and scored (2 = Yes, 1 = Partial, 0 = No, N/A = Not Applicable) based on specific definitions and instructions for each criterion, summed, and adjusted to provide a summary score (Kmet et al., 2004). The mixed method study (Haukkala et al., 2013) was evaluated using both quantitative and qualitative criteria. Ethical case studies and personal narratives were not evaluated for quality because validated instruments for these types of studies have not been developed. However, both were judged to be acceptable quality based on being in peer-reviewed journals. Tables II and III show the

quality assessment criteria and evaluation of the articles included in the review. A minimum quality score was set at .75 out of 1.00 (Kmet et al., 2004). All articles exceeded the minimum standard for quality.

Stage 4: Data Analysis of HCA Literature

Data analysis began by extracting the year published, authors, country where the research took place, study design, sample characteristics, sample recruitment, and genetic testing status of participants from the 12 articles. The full text of each article was uploaded into NVivo 10 to facilitate analysis (NVivo Qualitative Data Analysis Software). LS read each article in its entirety and coded study findings into the four major elements of the FCGR (influential factors, communication strategies, communication occurrence, outcomes of communication). Data included in analysis were generally found in the 'results' sections of the articles, however data were also found in the 'discussion' section because some studies present original findings in both sections of an article (Sandelowski & Leeman, 2012). A second reviewer (SDH) then examined coded text for each element and discrepancies were discussed until agreement was reached. The next round of coding consisted of LS reviewing the coded text for each of the four elements and developing categories to capture the details within each element. SDH reviewed the categories and again discrepancies were discussed until agreement was reached.

Results

Stage 5: Presentation of Results of HCA Literature Integrative Review

Table IV summarizes the articles included in this review. Studies were conducted in the United Kingdom (UK; n = 3), United States of America (USA; n = 3), Netherlands (n = 2), Australia (n = 1), Finland (n = 1), Norway (n = 1), and Ireland (n = 1). Studies were generally small with the largest sample size being 383 (Batte et al., 2015). Studies varied in design, including qualitative (n = 7), quantitative (n = 2), and mixed-methods (n = 1). Two non-research articles, an ethical case study (n = 1) and a personal narrative (n = 1), were also included in the review. While all studies included individuals with HCM or LQTS, only three studies included individuals with both HCM and LQTS. Nine studies included only participants that had undergone genetic testing, two studies included participants both with and without genetic testing, and one study did not specify genetic testing status. In studies with participants that had genetic testing, two articles included only those with positive genetic test results and seven included participants with positive, negative, or inconclusive results. Recruitment of participants for the research studies was through clinical settings, disease support groups, national disease registries, and research biobanks.

Results of the qualitative synthesis are presented in Table V by the four major elements of the FCGR (influential factors, communication strategies, communication occurrence, and outcomes of communication). The table contains conceptual definitions for each developed category along with supporting data from the reviewed articles.

Synthesis of Evidence

The integrated review of the HCA literature resulted in a modification of the original FCGR framework (which was constructed based on non-cardiac inherited conditions) in the context of HCA. Figure 3 summarizes modifications of the FCGR based on the HCA integrated review with the original major elements of the FCGR in bold text and modifications to the FCGR in *italics*. All 12 articles discussed influential factors in communication (12 discussed individual factors, 11 discussed disease factors, and nine discussed family factors) while only six articles discussed communication occurrence. The number of articles describing each element, category, and sub-category of the updated FCGR are indicated in parentheses in Figure 3. This integrative review provides a detailed framework for understanding family communication of genetic risk for HCAs.

Discussion

This review analyzed 12 studies related to family communication of genetic risk for HCA. Several findings in this review are congruent with the literature on non-cardiac genetic risk communication in families. Specifically, family, disease, and individual factors were influential in the extent to which families communicated about genetic risk.

Individual factors were the most extensively developed influential factors and were identified in all 12 studies. One category, conflicting interests for children, is of particular relevance for HCA. The onset of HCA often occurs in minor children, whereas in many non-cardiac diseases (e.g., Huntington Disease, Hereditary Breast and Ovarian Cancer), onset in minor children is very rare. Rowland & Metcalfe (2013) reviewed genetic risk communication between parents and their minor children and found that for diseases with young onset (Cystic Fibrosis, Familial Adenomatous Polyposis, Sickle Cell Disease, and Neurofibromatosis), parents communicated more with children who were affected and less with children who were unaffected. Similar to Rowland & Metcalfe's (2013) findings, communication of genetic risk for HCA to minor age children is tightly intertwined with testing and diagnosis because parents take on the role of communicating disease risk and managing their child's health and wellbeing. Unlike communication to adult children or adult relatives, communication about HCA to minor age children seemed to focus on the risks related to the disease itself (such as SCD) rather than the risk for disease and inherited nature of the disease. This creates the potential that children who are told about their disease risk at a young age may not realize the heritability element of their HCA as they transition to adulthood. This presents an opportunity to create developmentally appropriate systems that address HCA risk and inheritance with children throughout their development (Rowland & Metcalfe, 2013).

Influential family factors were not specific to families with HCA as family dynamics, closeness, and contact have been similarly described in the non-cardiac literature (Wilson et al., 2004; Gaff, et al., 2007; Wiseman, et al., 2010). However, unlike in families with non-cardiac disease, some of the milestones such as childbirth, and pregnancy seemed to be meaningful in families with HCA due to the risk for children. Family contact and closeness had varied and somewhat contradictory findings in regards to its influences on communication about HCA in families, however the majority of evidence supports that lack

of contact and closeness with family members is an important barrier to communication of HCA risk.

The description of influential disease factors highlights the contributions of both the understanding and the personal experience with HCA to communication. HCAs, although inherited in a predictable AD pattern, have variable and largely unpredictable expressivity and severity within families. This variable expressivity likely contributes to the difficulties that participants in some studies had in understanding disease inheritance and identifying who was at risk.

Communication strategies includes multiple methods of delivery of communication and variation in the content of these communications, similar to the non-cardiac literature (Gaff, et al., 2007; Wiseman, et al., 2010). Participants in the included HCA studies identified multiple modes of communicating genetic risk to relatives. However, letters were the only clinician-provided mode of communication described. van der Roest et al. (2009) reported that providing family letters to probands led to higher family member screening rates, vs. families not receiving letters (57% vs. 35%, respectively, $p < .01$). Clinicians have the opportunity to support other modes of communication in addition to letters, such as scripts for in-person or telephone conversations, which seemed to be more common modes of communication in families than letters, or identifying a family informant to carry out communication, as is suggested for communication of familial cancer risk (Bowen et al., 2017). As with non-cardiac disease (Gaff, et al., 2007; Wiseman, et al., 2010), the content of communication to relatives was varied among those with HCA, and still very little is known about what information relatives actually received or understand.

Only six studies described communication occurrence. Overall, reports of communication occurrence were not specific to which family members were told about risk, nor which relatives had not been told about their risk. Most studies focused on immediate family members, who clearly are at highest risk and should be tested first, but did not examine communication to more distant relatives, who are also at high risk. If communication was optimal, relatives who were told of their risk would communicate risk to the next tier of relatives, following the same path as cascade screening. However, communication of risk breaks down in each successive wave and it is unknown if the cascade screening model can rely on communication in each wave or if the communication may be best coming from the original proband or informed family member. Research was also lacking in identifying which relatives are not told, and why, and did not examine communication occurrence from the perspective of the relative.

The most clinically relevant outcome of communication is uptake of clinical or genetic testing of family members because it identifies individuals who would benefit from treatments, and clarifies risk status for additional family members. Findings related to clinical or genetic testing in relatives were scarce, considering the large number of family members at risk. More research is needed to understand the complexity of why at-risk relatives do not get screened for HCA. It is known that making relatives aware of their risk is essential, but not sufficient to ensure that relative will get tested. The familial risk perception model identifies that individuals must develop salience or awareness of risk as an essential

step in their development of personal feelings of vulnerability to risk that lead them take action to control their risk (Walter & Emery, 2005; Daack-Hirsch, Shah, & Cady, 2017). However, although SCD of a family member may be the salient event that brings awareness of the HCA to a family, those experiencing SCD in their family are no more likely to participate in screening than those who have not experienced SCD (Christiaans et al., 2008; Miller et al., 2013) potentially due to the emotions experienced by the sudden loss of a loved one (Christian et al., 2017).

Study Limitations

The 12 articles included in this integrative review are exclusively from western countries where healthcare systems are protective of individual privacy. Healthcare systems and cultures vary significantly in how genetic information is viewed and how families communicate, which may limit the application of these findings in other cultures or healthcare systems which have less restrictive privacy practices. Many of the studies included in this review that had larger sample sizes drew from disease support groups. Samples from support groups likely differ from clinic based samples (Wiseman et al., 2010) in a number of ways including disease experience, coping, and level of interest and knowledge about their HCA. In addition, the retrospective survey designs of the quantitative studies included in this review may be limited by recall bias and confounding, and are unable to establish temporality, a key component in detecting causal relationships between various factors and communication about HCA risk in families.

Practice Implications

The FCGR framework is clinically practical and describes several elements pertinent to the process of family communication about genetic risk for HCAs. Healthcare practitioners providing genetic counseling can directly apply this model by discussing influential factors important to probands and family specific needs and developing personalized communication strategies such as family letters (van der Roest et al. (2009) and utilization of other family members to facilitate communication (Bowen et al., 2017). Ethical-based conceptual frameworks (McConkie-Rosell & Spiridigliozzi, 2004) and multi-disciplinary team approaches (Caleshu et al., 2016) may help clinicians address the variety of issues that impede communication about HCA risk in families particularly with children. A team approach is important because while genetic counseling lays the foundation, communication of risk for HCA is an ongoing process and individuals with HCA will likely have needs related to family communication throughout their life. Probands may interact with physicians or nurses more frequently than genetic counselors for their healthcare and a team approach provides flexibility for various healthcare roles to provide continued support for individuals communicating HCA risk with family members.

Counseling sessions with individuals and families with HCA should specifically address the conflicting interests regarding communication to children and address doubts, ambivalence, and reluctance that may prevent communication to relatives. Practitioners should monitor the success of communication by assessing occurrence and outcomes of communication throughout the counseling process. Probands may benefit from counseling about the range of outcomes of their communication beyond clinical screening and genetic testing. This review

provides insight into range of influential factors that may be important to the proband's communication with family members about HCA risk.

Policy & Research Recommendations

Findings from this study support the recognition of family communication as a key step in cascade screening in the next update of the American Heart Association (AHA)/American College of Cardiology (ACC) Guidelines. Al-Khatib et al.'s (2017) recent quality measure to document communication of genetic risk to first-degree family members is a good step toward the recognition of the key role of family communication in cascade screening. This new quality measure also provides an opportunity for additional research on family communication and family screening which can be used to promote this quality measure into a performance measure, which would be subject to public reporting or pay for performance programs (Al-Khatib et al., 2017).

The updated FCGR in the context of HCA identifies several areas for further research. Future research should seek to understand the specific content of genetic risk communications to family members, as this was a particularly sparse area of research. Understanding the content of communications is key to ensure that family members can make an informed decision about screening. Future research on communication occurrence should strive to determine specifically which family members are not told about their risk in order to more accurately design interventions aimed at improving communication with those relatives who are otherwise unlikely to be told about their risk. Deliberate efforts to include participants with diverse disease experiences and backgrounds in this research is crucial because they may have very different needs and this is a current limitation to the current body of literature. Research aimed at investigating communication about genetic risk for HCA from the point of view of family members would fill an important gap in our understanding about how family members view the communication and their reaction to the communication which would lead to better understanding of how messages are received and translated into screening action. Future research should also aim to develop novel interventions to improve family communication as this may improve the rates of family members participating in screening and genetic testing (Cirino et al., 2017). In addition, utilization of prospective study designs or using control groups can advance our understanding of the relationships among various factors and communication, which may be particularly useful in intervention development.

Conclusion

This article presents an analysis of family communication of genetic risk for HCA and helps to better understand a critical step in the cascade screening process. Family communication about genetic risk for HCA fits into the major elements of the FCGR developed from the literature on family communication about genetic risk for non-cardiac disease. This review expands understanding about family communication of genetic risk for HCA by synthesizing what is known about influential factors, communication strategies, communication occurrence, and the outcomes of communication in the context of HCA.

Findings can be used to inform practice and the development of future studies, interventions, and policy aimed at improving communication and cascade screening in families with HCA.

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Figure 1.
Family Communication of Genetic Risk Conceptual Framework (FCGR) Based on Non-Cardiac Conditions

Figure 2.
Adapted PRISMA Flow Diagram Illustrating the Steps of the Literature Search Process

Figure 3. Modification of the Family Communication of Genetic Risk Conceptual Framework (FCGR) Based on Integrative Review of Family Communication About Genetic Risk for HCA Number in parentheses indicates the number of articles describing each element, category, and sub-category of the updated FCGR.

Table 1**Elements of Communicating Genetic Risk Identified by Reviews**

Element	Description of Element
Wilson et al. (2004)	
Disease factors	Characteristics of the disease that impact the likelihood of communication about genetic risk (e.g., inheritance pattern, certainty and comprehensibility of genetic testing information, and availability of risk management strategies or interventions).
Individual factors	Characteristics of probands that may influence the likelihood that individuals share genetic risk information and with whom they share it (e.g., individual's coping mechanisms, emotional barriers to communication, psychological defenses, and risk perception).
Family factors	Characteristics of the relationships in families and the family system (e.g., communication patterns, family representatives, control of information, and family myths).
Sociocultural factors	Characteristics of the culture or society in which individuals and families live (e.g., gender roles, taboos, concern about discrimination, and notions of kinship and inheritance).
Gaff et al. (2007)	
Deliberation before communication	Probands' considerations of what information to share, when to share it, and the possible effects of communicating inherited disease risk information to family members.
Communication strategies	Individual approaches to communicating risk information (e.g., complete openness with family members, total secrecy, passing on responsibility to others in the family to communicate).
Outcomes of communication	Effects of the disease risk communication on probands, relatives, and families measured as uptake of genetic testing; family members' knowledge of disease risk; or the impact of the communication on probands, relatives, and their relationships.
Wiseman et al. (2010)	
Functions & influences of communication	Reasons for communicating about disease to relatives as well as the factors that either motivate or inhibit disease risk communication.
Processes of communication	Who is told, how they are told, and what they are told.
Outcomes of communication	The impact of the disease risk communication on probands, relatives, and family relationships.
Weins et al. (2013)	
Attitude	The desire to protect; perceptions of relevance, responsibility, and the right to know; and the usefulness of communicating which determine intention to communicate.
Subjective Norm	Pressure from family members, health professionals, and society which determine intention to communicate.
Perceived Behavioral Control	Family dynamics and relationships, communication skills, ability to understand, and coping skills which determine intention to communicate.
Intention	The determination of an individual to engage in communication of genetic risk to another family member as a function of attitude, subjective norms, and perceived behavioral control.
Behavior	Communication of genetic risk to another family member as a function of attitude, subjective norms, and perceived behavioral control.

Table II

Quality Evaluation of Qualitative Studies

Criteria for Qualitative Studies		Articles							
		Smart et al. (2010)	Geelen et al. (2011a)	Geelen et al. (2011b)	Mangset al. (2014)	Ormondroyd et al. (2014)	Vavolizza et al. (2015)	Haukkala et al. (2013)	Whyte et al. (2016)
1	Question/objective sufficiently described?	2	2	2	2	2	2	2	2
2	Study design evident and appropriate?	2	2	2	2	2	2	2	2
3	Context for the study clear?	2	2	2	2	2	2	2	2
4	Connection to a theoretical framework/wider body of knowledge?	2	2	2	2	2	2	2	2
5	Sampling strategy described, relevant and justified?	2	2	2	2	2	1	2	2
6	Data collection methods clearly described and systematic?	2	2	2	2	2	2	2	2
7	Data analysis clearly described and systematic?	2	2	2	2	2	1	2	2
8	Use of verification procedure(s) to establish credibility?	1	2	2	2	2	2	2	1
9	Conclusions supported by the results?	2	2	2	2	2	2	2	2
10	Reflexivity of the account?	1	1	1	1	1	1	1	1
	Total Sum	18	19	19	19	19	17	19	19
	Total Possible Sum (20)	20	20	20	20	20	20	20	20
	Summary Score (total sum/total possible sum)	.90	.95	.95	.95	.95	.85	.95	.95

2= Yes, 1=Partial, 0=No, N/A=Not Applicable

Table III

Quality Evaluation of Quantitative Studies

Criteria for Quantitative Studies		Articles		
		Batte et al. (2015)	Burns et al. (2016)	Haukkala et al. (2013)
1	Question/objective sufficiently described?	2	2	2
2	Study design evident and appropriate?	2	2	2
3	Method of subject/comparison group selection or source of information/input variables described and appropriate?	2	2	2
4	Subject (and comparison group, if applicable) characteristics sufficiently described?	2	2	2
5	If interventional and random allocation was possible, was it described?	N/A	N/A	N/A
6	If interventional and blinding of investigators was possible, was it reported?	N/A	N/A	N/A
7	If interventional and blinding of subjects was possible, was it reported?	N/A	N/A	N/A
8	Outcome and (if applicable) exposure measure(s) well defined and robust to measurement/misclassification bias? means of assessment reported?	2	2	1
9	Sample size appropriate?	2	2	2
10	Analytic methods described/justified and appropriate?	2	2	2
11	Some estimate of variance is reported for the main results?	2	2	1
12	Controlled for confounding?	1	1	N/A
13	Results reported in sufficient detail?	2	2	2
14	Conclusions supported by the results?	2	2	2
	Total Sum	21	21	18
	Total Possible Sum (28 number of "N/A" *2)	22	22	20
	Summary Score (total sum/total possible sum)	.95	.95	.90

2= Yes, 1=Partial, 0=No, N/A=Not Applicable

Table IV

Summary of Articles Included in Review

Year	Author	Country	Design	Sample	Sample Recruitment	Genetic Testing Status of Participants	Quality Score
2010	Ladd	UK	Personal narrative	N=1	N/A	PGT	Not rated
2010	Smart	UK	Qualitative	N=27 HCM and LQTS	Clinical setting	All participants had genetic testing: 17 PGT, 7 NGT, 3 IGT	.90
2011a	Geelen et al.	Netherlands	Qualitative	N=57 with HCM (from 6 families)	Clinical setting	All families had an identified genetic mutation. Precise results of individuals not given.	.95
2011b	Geelen et al.	Netherlands	Qualitative	N=57 with HCM (from 6 families)	Clinical setting	All families had an identified genetic mutation. Precise results of individuals not given.	.95
2012	Cohen et al.	USA	Ethical case study	3 case studies	N/A	1 PGT, 2 in process of parental decision about genetic testing	Not rated
2013	Haukkala et al.	Finland	Mixed Methods	N=17 LQTS found incidentally	Research biobank	All PGT	.90/.95*
2014	Mangset et al.	Norway	Qualitative	N=13 parents of children with LQTS (9 nuclear families)	Disease support group	All participants had genetic testing. 54% PGT	.95
2014	Ormondroyd et al.	UK	Qualitative	N=22 with HCM or LQTS	Clinical setting	All participants had genetic test. 77% PGT	.95
2015	Batte et al.	USA	Quantitative	N=383 with HCM	Disease support group	64% had genetic testing. 43% PGT	.95
2015	Vavolizza et al.	USA	Qualitative	N=50 with personal or family history of genetic arrhythmia (32 families)	Clinical setting and disease support groups	Gene test status not specified	.85
2016	Burns et al.	Australia	Quantitative	N=75 probands and relatives (38 families) with LQTS	National disease registry	15 probands & 27 relatives PGT. 17 probands IGT. 6 probands and 1 relative did not have genetic testing.	.95
2016	Whyte et al.	Ireland	Qualitative	N=9 family members taking part in cascade screening for HCM/LQTS	Clinical setting	All participants had genetic testing. 4 PGT, 5 NGT	.95

HCM = hypertrophic cardiomyopathy; LQTS = long QT syndrome; BS = Brugada syndrome; ARVC = arrhythmogenic right ventricular cardiomyopathy; DCM = dilated cardiomyopathy; CPVT = catecholaminergic polymorphic ventricular tachycardia; PGT = Positive genetic test; NGG = Negative genetic test; IGT = Indeterminate genetic test.

*Haukkala et al. (2013), a mixed methods study, had a quality score of .90 for the quantitative evaluation and .95 for the qualitative evaluation.

Table V
Qualitative Synthesis of Literature on Family Communication of Genetic Risk for HCA

FCGR Element	Category: <i>Conceptual Definition</i>	
	Citation	Supporting Literature
Influential Factors (Family Factors)		Established Family Dynamics Persist: <i>Patterns and styles already present in a family in terms of communication, conduct towards each other, and relationships.</i>
	Whyte et al. (2016)	<ul style="list-style-type: none"> Parents were expected to communicate with their own children. Older generations were expected to inform the extended family about their risk. When contact with family members was infrequent or limited to holidays, communication about risk did not occur because it was not part of the normal conversations that occurred at these family events.
	Haukkala et al. (2013)	<ul style="list-style-type: none"> One mother who told each of her children about their risk stated that their family always had a very open discussion style.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> One individual felt that having less severe disease than other family members isolated her from her family, however upon further reflection she concluded that the isolation began years before due to a parent's early death and the resulting change in family dynamics.
	Burns et al. (2016)	<ul style="list-style-type: none"> 10% of participants cited previous breakdowns in family relationships as the reason for not communicating risk.
		Family Contact & Closeness: <i>The influence that the family structure, quality of relationships within the family, amount of contact between family members, geographical proximity, and emotional closeness have on the communication of HCA risk.</i>
	Burns et al. (2016)	<ul style="list-style-type: none"> Close relationships with family members made it easier to communicate risk. Half of those who did not communicate to a FDR cited lack of existing contact as the reason for not communicating.
	Smart (2010)	<ul style="list-style-type: none"> Reasons for lack of contact with family members ranged from mundane to complex or sensitive. Some probands simply did not know how to get in touch with relatives.
	Battie et al. (2015), Ormondroyd et al. (2014), Smart (2010)	<ul style="list-style-type: none"> Resentment, estrangement, and poor relationships with family members were barriers to communication of risk.
	Whyte et al. (2016)	<ul style="list-style-type: none"> Large family sizes, particularly in the older generation, resulted in loss of contact with the numerous branches of family over time and a lack of awareness about which family members had to be told about their HCA risk.

FCGR Element	Category: <i>Conceptual Definition</i>	
	Sub-Category: <i>Conceptual Definition</i>	
	Citation	Supporting Literature
	Batte et al. (2015)	<ul style="list-style-type: none"> Lack of closeness and lack of contact with relatives were not very important barriers to communication (although they ranked as the most important barrier among several potential barriers). Family functioning and cohesiveness had little influence on the occurrence of communication. Women were more likely to describe communication barriers related to family contact and closeness than men.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> In one family, questionable paternity lessened the perception of HCA risk for siblings, creating emotional distance.
	Ladd (2010)	<ul style="list-style-type: none"> Some parents from 14 different geographically disparate families, with no prior contact, eventually developed relationships based on shared paternity and shared disease among their children who were related only by a sperm donor.
	Ormondroyd et al. (2014), Smart (2010)	<ul style="list-style-type: none"> Geographically dispersed families and complex family relationships increased uncertainty about telling relatives about HCA risk.
	Cohen et al. (2012)	<ul style="list-style-type: none"> Geographic and emotional distance contributed to the unwillingness of one family member to communicate about HCA risk.
	Milestones: <i>Normal life happenings such as marriage, pregnancy, or childbirth that influence HCA risk communication.</i>	
	Geelen et al. (2011b)	<ul style="list-style-type: none"> Pregnancy, childbirth, increasing severity of physical complaints potentially related to HCA, or death of family members raised the issue of family risk for some families.
Influential Factors (Disease Factors)	Haukkala et al. (2013)	<ul style="list-style-type: none"> Birth of a grandchild motivated communication in one family.
	Understanding of Disease: <i>An individual's measured or perceived understanding or confidence in their knowledge of various aspects of their disease and the disease risk for others (e.g. inheritance, genetic tests, perception of risk).</i>	
	Batte et al. (2015)	<ul style="list-style-type: none"> Lower comprehension of HCA was weakly associated with more importance placed on barriers to communication including not thinking family members were at risk ($r=-0.19$, $p=.03$), not knowing what to say to family members about HCA and their risk ($r=-.15$, $p=.02$), or not knowing how to explain medical genetics ($r=-.16$, $p=.01$).
	Burns et al. (2016)	<ul style="list-style-type: none"> Not knowing which relatives were at risk was a barrier to communication for 12% of the participants and was significantly more common for those experiencing anxiety (50% vs. 13% without anxiety, $p=.025$) and depression (67% vs. 21% of those without depression, $p=.028$).

FCGR Element	Category: Conceptual Definition	
	Sub-Category: Conceptual Definition	
	Citation	Supporting Literature
	Smart (2010), Vavolizza et al. (2015)	<ul style="list-style-type: none"> Participants did not know which family members were at risk and how to explain complexities of the disease including the genetic component.
	Batte et al. (2015)	<ul style="list-style-type: none"> Perceived lack of risk in relatives was not considered an important barrier of communication nor a statistically significant predictor of communication ($\beta=-.05$, $p=.97$).
	Haukkala et al. (2013)	<ul style="list-style-type: none"> One proband felt they didn't know enough about the disease to communicate risk to family members.
	Whyte et al. (2016)	<ul style="list-style-type: none"> Participants (incorrectly) perceived that family members who did not have children would not require testing for HCA and that having a mutation that caused HCA was no different than any other genetically variable characteristic.
	Batte et al. (2015)	<ul style="list-style-type: none"> Increased comprehension of HCA inheritance patterns was observed among individuals who told all of their siblings and children compared to those who only told some siblings and children ($OR=3.57$, $p=.03$). Higher levels of comprehension of HCA inheritance patterns were also associated with assigning more importance to informing family members of their risk ($r=.29$, $p=.0001$) and suggesting they get tested ($r=.22$, $p=.003$).
	Smart (2010)	<ul style="list-style-type: none"> Some individuals believed they had no problem knowing which family members to contact and what to say.
	Burns et al. (2016)	<ul style="list-style-type: none"> 71% of participants believed a good understanding of what their genetic test results meant made it easier to communicate.
	Mangset et al. (2014)	<ul style="list-style-type: none"> Perception of others' risks, particularly children, is not completely rational and often involves emotional components.
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> Non-rational risk perception for families experiencing death from HCA, who use various mechanisms to justify their perceptions of a lower risk of disease for themselves and their children.
	Disease Experience: <i>An individual's experience with the disease, including how they came to be diagnosed or aware of their risk, symptoms, coping with the disease, and test results.</i>	
	Geelen et al. (2011b), Ormondroyd et al. (2014)	<ul style="list-style-type: none"> Some grew up knowing about the HCA in their family and described an evolving understanding of their risk.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> Some knew of heart issues in family members, but not that the heart issues were familial until genetic testing was done.
	Haukkala et al. (2013)	<ul style="list-style-type: none"> For some, learning about their HCA changed their perception of their family history and explained symptoms in family members that had never previously been linked to a genetic heart problem.

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	Sub-Category: <i>Conceptual Definition</i>	
	Citation	Supporting Literature
	Geelen et al. (2011b)	<ul style="list-style-type: none"> The HCA was a surprise for some who were the first in their family to be diagnosed and had no evident family history.
	Ladd (2010)	<ul style="list-style-type: none"> In one case of a child born through a sperm donor, the family learned about the HCA risk through the sperm bank.
	Haukkala et al. (2013)	<ul style="list-style-type: none"> All participants learned about their risk incidentally through participation in a biobank research study.
	Whyte et al. (2016)	<ul style="list-style-type: none"> All participants diagnosed with an HCA due to a family member's diagnosis, 66% of whom had a sibling die from an HCA and 33% who had a relative experience a cardiac event.
	Batte et al. (2015)	<ul style="list-style-type: none"> Disease severity of the proband, measured by ICD status, was not related to communication (OR= .89, $p=.65$).
	Geelen et al. (2011a)	<ul style="list-style-type: none"> Disease severity within the whole family was not indicative of communication to children.
	Haukkala et al. (2013)	<ul style="list-style-type: none"> For three individuals discovering their HCA incidentally through biobank participation, disease severity and risk information took on more personal relevance because their symptoms could now be explained by the diagnosis.
		<ul style="list-style-type: none"> Three individuals who lacked symptoms were less concerned about the disease and thought that the absence of symptoms in their family made it easier to communicate risk.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> For one individual, personal difficulties dealing with the HCA (symptoms and reactions of those around her) were more concerning than risk to family members.
	Geelen et al. (2011a)	<ul style="list-style-type: none"> Parents of children affected with HCA were concerned with the physical health of their child more than the genetic nature of HCA and the possibility of inheritance.
		<ul style="list-style-type: none"> In situations where a couple was faced with communicating risk to children, the parent who had HCA or was at risk for HCA generally took the lead in making decisions about how to manage the child's risk.
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> Some families minimized risk despite death from HCA in family members.
	Smart (2010)	<ul style="list-style-type: none"> For some, risk of sudden death (especially in children) motivated communication but also created guilt and anxiety.

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	Sub-Category: <i>Conceptual Definition</i>	
	Citation	Supporting Literature
Influential Factors (Individual Factors)	Batte et al. (2015)	<ul style="list-style-type: none"> Regardless of coping style (measured by the Brief COPE), coping did not have an effect on communication (OR range .66–1.00, all $p > .10$) nor was difficulty coping with one's disease considered a very important barrier to communication.
	Batte et al. (2015), Burns et al. (2016)	<ul style="list-style-type: none"> Different genetic test results (positive, negative, variants of unknown significance) were not found to affect communication occurrence to relatives (OR=1.15, $p=.74$).
	Reasons to Communicate Risk: <i>An individual's conscious reasons to communicate risk and included three distinct subcategories: moral and ethical conviction; desire to inform, encourage, and help; and reciprocal communication.</i>	
	Moral & Ethical Conviction: <i>Feelings of responsibility, duty, or obligation to communicate risk to various relatives.</i>	
	Batte et al. (2015), Burns et al. (2016), Geelen et al. (2011b), Mangset et al. (2014), Ormondroyd et al. (2014), Vavolizza et al. (2015), Whyte et al. (2016)	<ul style="list-style-type: none"> Participants in seven studies were motivated to communicate about genetic risk based on feeling a responsibility, duty, or obligation to communicate about the genetic risk for HCA to family members.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> Responsibility was generally focused toward relatives and children, but responsibility to follow healthcare provider's directions was also described.
	Geelen et al. (2011b), Mangset et al. (2014)	<ul style="list-style-type: none"> Some participants envisioned scenarios where a child or relative died due to not knowing their risk.
	Vavolizza et al. (2015)	<ul style="list-style-type: none"> One participant described communication of HCA as something personal between family members and not an obligation. 27 participants (54%) did not bring up obligation or duty at all as a reason to communicate however explanations of why they did not are unknown.
	Ormondroyd et al. (2014), Vavolizza et al. (2015)	<ul style="list-style-type: none"> Belief that relatives have a right to know about their HCA risk was emphasized as a reason for communication.
	Ladd (2010)	<ul style="list-style-type: none"> In one family connected only by a sperm donor, one mother contemplated if these now connected individuals should have an obligation to share information with each other.
(Mangset et al. (2014))		<ul style="list-style-type: none"> Moral and ethical convictions were often accompanied by other feelings including anger, guilt, and doubts, making communication difficult.
Altruism: <i>Desire to inform, encourage, and help as reasons to communicate.</i>		

FCGR Element	Category: <i>Conceptual Definition</i>	
	Sub-Category: <i>Conceptual Definition</i>	
	Citation	Supporting Literature
	Batte et al. (2015), Burns et al. (2016), Haukka et al. (2013), Mangset et al. (2014), Ormondroyd et al. (2014), Vavolizza et al. (2015)	<ul style="list-style-type: none"> • Probands often communicated about genetic risk because they believed the information would be useful to their relative's health and medical decision making and had the potential to save a life.
	Burns et al. (2016), Haukka et al. (2013), Mangset et al. (2014), Vavolizza et al. (2015)	<ul style="list-style-type: none"> • Altruism was particularly strong when considering children or future generations.
	Batte et al. (2015), Burns et al. (2016)	<ul style="list-style-type: none"> • Some were motivated to communicate to encourage their relatives to get genetic testing.
	Burns et al. (2016)	<ul style="list-style-type: none"> • Some felt that their relatives would want this information and would want to get tested.
	Ormondroyd et al. (2014), Vavolizza et al. (2015)	<ul style="list-style-type: none"> • Some wanted to provide information that might be useful for the future, if HCA symptoms developed.
	Reciprocal Communication: <i>The expectation that communication of risk to relatives would have some benefit to the proband.</i>	
	Batte et al. (2015)	<ul style="list-style-type: none"> • Some participants communicated with relatives to get emotional support and advice on their own medical treatment.
	Ladd (2010)	<ul style="list-style-type: none"> • One respondent communicated her child's test results back to the sperm bank to add to the genetic information of the entire biologic (through sperm donor) family and was hopeful others would do the same. She recognized that reciprocal communication may not be possible and respected others' desire for privacy.
	Psychological Functioning: <i>General or disease related anxiety, depression, or stress that affects or is associated with communication about genetic risk.</i>	
	Haukka et al. (2013)	<ul style="list-style-type: none"> • The need to communicate risk to relatives and children was an additional stress of an HCA diagnosis.
	Burns et al. (2016)	<ul style="list-style-type: none"> • Individuals with clinical anxiety, compared to those without, were more likely to report not knowing which family members were at risk (50% vs. 13% without anxiety, $p=.025$), emotional difficulties sharing genetic risk information (43% vs. 6% without anxiety, $p=.018$), not wanting to upset relatives (43% vs. 6% without anxiety, $p=.018$), and feeling guilty or anxious about communication of risk (36% vs. 0% without anxiety, $p=.009$).
		<ul style="list-style-type: none"> • Depression was associated with increased feelings of guilt or anxiety about communicating risk (67% vs. 4% without depression, $p=.001$), and difficulty knowing who in the family was at risk (67% vs. 21% without depression, $p=.028$).
	Smart (2010)	<ul style="list-style-type: none"> • Anxiety was related to the potential for severe and lethal symptoms of HCA, especially for children.
	Haukka et al. (2013)	<ul style="list-style-type: none"> • One individual described feeling scared to communicate risk and only doing so because of births in the family.

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	Citation	Supporting Literature
		<ul style="list-style-type: none"> Anxiety was also reported by some individuals due to having to repeat risk information several times to several relatives.
	Mangset et al. (2014)	<ul style="list-style-type: none"> One parent described anger, guilt, and vulnerability about communicating risk and felt the entire situation was awful.
	Mangset et al. (2014), Smart (2010)	<ul style="list-style-type: none"> Anxiety was often accompanied by guilt of nondisclosure or just guilt in general.
	Smart (2010), Vavolizza et al. (2015)	<ul style="list-style-type: none"> Participants worried that communication would cause stress and negative emotions for themselves or their relatives due to the bad news they would share.
		Doubts, Ambivalence, & Reluctance: <i>The view that negative consequences of risk communication may outweigh the positive, leading to hesitation or blocking communication about genetic risk to relatives.</i>
	Geelen et al. (2011b), Ormondroyd et al. (2014), Smart (2010)	<ul style="list-style-type: none"> Participants reported being uneasy and having doubts or ambivalence about burdening family with information they would not want to hear and wondered what good it would do given that there was no way to prevent the disease.
	Burns et al. (2016), Smart (2010), Vavolizza et al. (2015)	<ul style="list-style-type: none"> Some participants were reluctant to communicate because of the potential to upset family members with what was perceived as bad news.
	Burns et al. (2016)	<ul style="list-style-type: none"> Reluctance especially when the proband was experiencing generalized anxiety.
	Cohen et al. (2012)	<ul style="list-style-type: none"> Described three clinical scenarios in which ambivalence was evident about disclosing genetic risk to family members and children despite apparently obvious benefits.
	Smart (2010)	<ul style="list-style-type: none"> Doubts and concerns were complicated by conflicting advice or pressure from healthcare providers and different family members about who to tell about the risk.
	Mangset et al. (2014), Ormondroyd et al. (2014)	<ul style="list-style-type: none"> Feelings of doubt or ambivalence about the value of HCA risk communication was accompanied by a sense of obligation to communicate, creating a difficult dilemma.
	Burns et al. (2016)	<ul style="list-style-type: none"> Reluctance to communicate because of a perception that family members may not care about the information.
	Batte et al. (2015)	<ul style="list-style-type: none"> Thinking relatives would not care and not wanting to upset relatives were rated as 'not very important' barriers to communication.

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	Supporting Literature	
	Citation	
	Whyte et al. (2016)	<ul style="list-style-type: none"> Some felt relatives would not be interested nor proactive if given the information, particularly in regards to relatives with whom they had no contact.
		<p>Difficulty with the Conversation:</p> <p><i>Not knowing what to say or how to start a conversation about risk for HCA.</i></p>
	Smart (2010)	<ul style="list-style-type: none"> Some participants described simply not knowing how to even start a conversation about being at risk for sudden death and the accompanying activity restrictions.
	Batte et al. (2015)	<ul style="list-style-type: none"> Not knowing what to say in a conversation about HCA risk was not an important barrier to communication.
		<p>Gender Influence:</p> <p><i>Differences in communication styles that are thought to be related to gender.</i></p>
	Batte et al. (2015)	<ul style="list-style-type: none"> For the 383 participants who completed at least some of this study, there was no association between gender and communication ($X^2=3.01$, $p=.08$). For the 183 who completed the entire study, females were more than twice as likely than males to communicate with all their siblings and children about HCA risk ($OR=2.46$, $p=.03$). Women gave slightly higher importance to suggesting relatives get tested ($t(373)=2.15$, $p=.03$) and receiving emotional support ($t(367)=3.05$, $p=.002$) as motivators for communication. Women were also more likely than men to state that not being in contact ($t(255)=2.55$, $p=.01$) or not being close to relatives ($t(249)=2.33$, $p=.02$) were more important barriers to communication.
		<p>Conflicting Interests for Children:</p> <p><i>Conflict between the need to protect children from physical, psychological, or social burdens related to HCA and HCA risk, now and in the future.</i></p>
	Smart (2010)	<ul style="list-style-type: none"> The risk of sudden death from HCA in children could be a motivator to communicate risk to family members, but it could also limit communication, particularly when other barriers were present. Concern for other children in the extended family was given as a reason to communicate.
	Cohen et al. (2012), Mangset et al. (2014), Vavolizza et al. (2015)	<ul style="list-style-type: none"> The desire to protect children from physical, psychological, and social burdens was a major concern of parents of children with or at risk for HCA.
	Cohen et al. (2012)	<ul style="list-style-type: none"> Parents acted as guardians of the health and welfare of their children and feared that communication of risk information would be traumatic or disruptive to their child's life.
	Mangset et al. (2014)	<ul style="list-style-type: none"> Parents concerned with protecting their children from harm while promoting their future autonomy. Although parents were aware that despite the risk for psychological harm to children, they also were aware of the benefits of sharing the risk information with their children.
	Vavolizza et al. (2015).	<ul style="list-style-type: none"> Parents had difficulty balancing being open with their children and not scaring them.

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	Citation	Supporting Literature
		<ul style="list-style-type: none"> Parents based the extent of their communication, details shared, and terminology on their child's age.
	Geelen et al. (2011a)	<ul style="list-style-type: none"> Some parents purposely pushed the HCA and its genetic nature into the background to not burden children and facilitate the child's future decision making regarding testing.
	Haukkala et al. (2013), Mangset et al. (2014)	<ul style="list-style-type: none"> Perception of HCA risk in children had a substantial emotional component and brought forth a multitude of conflicting emotions including anger, anxiety, responsibility, vulnerability for criticism, guilt, trust, and distrust.
		<p>Desire to Protect Elderly:</p> <p><i>The need to protect elderly relatives, generally parents, from psychological burdens related to HCA (e.g. guilt of passing on bad gene, worry, stigma).</i></p>
	Ormondroyd et al. (2014), Smart (2010)	<ul style="list-style-type: none"> Participants described concerns about involving elderly parents in communication and screening of HCA largely to protect them from worry.
	Smart (2010)	<ul style="list-style-type: none"> Reasons for not telling elderly parents about HCA risk were not thinking the diagnosis would be beneficial at later stages of life, feeling they were at lower risk due to their advanced age, and a desire to protect from parents from the potential guilt of passing on the disease-causing gene.
Communication	Whyte et al. (2016)	<ul style="list-style-type: none"> Parents were expected to take a major role in communicate HCA risk to the extended family.
		<p>Delivery:</p> <p><i>Various modalities, styles, tones, or approaches used to communicate HCA risk to relatives.</i></p>
	Burns et al. (2016) Strategies	<ul style="list-style-type: none"> 11% of participants learned about their own risk from family members or a family letter. 89% of participants used multiple methods to communicate risk to different family members, most commonly a combination of in-person and phone communication.
	Haukkala et al. (2013), Ormondroyd et al. (2014)	<ul style="list-style-type: none"> Clinicians provided a family letter to help probands inform their relatives. However, this did not seem to be a uniform practice.
	Burns et al. (2016) Geelen et al. (2011b)	<ul style="list-style-type: none"> One individual (in each study) composed their own letter to communicate HCA risk to family members, sometimes along with communicating in person.
	Vavolizza et al. (2015).	<ul style="list-style-type: none"> Email was used to communicate risk to relatives.
	Whyte et al. (2016)	<ul style="list-style-type: none"> Participants unanimously preferred to give and receive HCA risk information in-person rather than using letters.

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		<ul style="list-style-type: none"> Participants made use of Facebook to keep in contact with cousins and also communicate about HCA risk.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> One participant who described his family as distant, informed relatives of their risk by phone then had no further discussion with them about the risk. Another participant held a somewhat formal family meeting, inviting all her siblings and their spouses, providing handouts and employing a straightforward and frank approach to informing all her siblings about their risk at the same time. Other, less formal, approaches included discussing HCA risk during normal family life such as during a walk, regular phone calls, or family dinners. Discussions about the health of affected family members often led to discussion of HCA risk. In some families, relatives visited the doctor together and openly discussed the HCA and its risks. While one proband described urgency to communicate with family members and immediately disseminated family letters. However, she felt that her newly informed relatives did not have the same sense of urgency and that copies of the letters were given out much more casually to other at-risk relatives.
	Ladd (2010)	<ul style="list-style-type: none"> In the case where a sperm donor was used, all communication was written, anonymized, and always between the individual and the sperm bank; however, through a website called the Donor Sibling Registry, some parents of different children from the same sperm donor connected and shared information.
	Smart (2010)	<ul style="list-style-type: none"> Family members used persuasion and pressure to motivate others to share HCA risk information for the interest of the larger family, particularly the younger generation.
	Mangset et al. (2014)	<ul style="list-style-type: none"> Families expected the assistance of healthcare professionals in communicating information to their relatives.
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> Participants felt that follow up for recently informed relatives from a healthcare provider may reinforce the communication started by the proband.
	Cohen et al. (2012)	<ul style="list-style-type: none"> Approaching communication of HCA risk as a process rather than a single event was supported by genetic professionals assisting parents with communication to their children about HCA risk.
	Content: <i>Specific content of communications about HCA risk.</i>	
	Vavolizza et al. (2015)	<ul style="list-style-type: none"> Variation in how much and what information was shared with relatives.
	Smart (2010)	<ul style="list-style-type: none"> Those who communicated risk shared details about the HCA, genetic tests, and the process of cascade screening.

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Communication Occurrence	Geelen et al. (2011b)	<ul style="list-style-type: none"> One family letter included an explanation of the situation, asked family members to spread the news, and offered to help anyone who wanted to know more.
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> An example of a letter provided by clinicians to give to family members contained brief information about the HCA and the risk to relatives.
	Vavolizza et al., 2015, p. 612).	<ul style="list-style-type: none"> One individual included a paragraph in an email communication about her child's diagnosis that said "unfortunately this means that it's in our genetic makeup... you might consider getting tested"
	Ladd (2010)	<ul style="list-style-type: none"> Initial communication from the sperm bank described the situation in which HCM had been discovered in a child related to her children through a mutual sperm donor, what HCM was, and advised seeking specific testing as soon as possible. Subsequent communications urged genetic testing and gave details of genetic test results from other family members.
	Whyte et al. (2016)	<ul style="list-style-type: none"> Almost half of the participants described talking about famous athletes with the HCA to converse more easily about the risk.
	Batte et al. (2015)	<ul style="list-style-type: none"> 72% of participants said they communicated with all of their siblings and children, while 23% reported communicating with at least one but not all siblings and children, and about 5% communicated with no one.
	Burns (2016)	<ul style="list-style-type: none"> 100% of their participants said they had told at least one FDR, 73% said they had told at least one SDR, and 60% said they had told at least one TDR.
	Haukkala et al. (2013)	<ul style="list-style-type: none"> 33 out of 35 children (94%) of affected parents had been told about their LQTS risk; the two who were not told were reported to be quite young.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> When a child was the first diagnosed in the family, the parents reported communicating risk information to all of the mother's siblings and parents, since it was determined their child inherited the risk from the mother.
	Ormondroyd et al. (2014), p. 92	<ul style="list-style-type: none"> "most participants know of relatives who have not been told".
Outcomes of Communication	Burns et al. (2016)	<ul style="list-style-type: none"> 10% of their sample said at least one FDR had not been informed of their risk.
	Ladd (2010)	<ul style="list-style-type: none"> In the case where at-risk biological siblings were related through a sperm donor, the sperm bank communicated the risk to all 24 of the known offspring's families, however there was a good possibility that the sperm bank was not aware of all the children born to this specific sperm donor.
Clinical Screening & Genetic Testing:		

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	<i>Reported uptake of clinical care including screening, and genetic testing because of communication of HCA risk.</i>	
	Burns et al. (2016)	<ul style="list-style-type: none"> • Uptake of genetic testing was 60% for FDR and 50% for SDRs and TDRs. • Average time from proband diagnosis to genetic testing of other at-risk relatives was six months.
	Cohen et al. (2012)	<ul style="list-style-type: none"> • One teen was tested and diagnosed with the same genetic mutation as her deceased sister.
	Ladd (2010)	<ul style="list-style-type: none"> • The mother of two children born from a sperm donor had both children clinically tested and later genetically tested.
	Haukkala et al. (2013)	<ul style="list-style-type: none"> • Of the 33 children who were told about their risk, 19 were tested for disease, four chose not to be tested, four planned to test in the future, and for six it was not known what the children did with the information.
	Geelen et al. (2011a)	<ul style="list-style-type: none"> • In one family, none of the four brothers nor the parents decided to get genetic testing after they were told about their risk by the mother of an affected child.
	Family Functioning: <i>Describes positive and negative changes in family dynamics as a result of communication of HCA risk.</i>	
	Vavolizza et al. (2015)	<ul style="list-style-type: none"> • Some families became closer, with communication and sharing of HCA risk information forming a bond between relatives who had not been close before.
	Whyte et al. (2016)	<ul style="list-style-type: none"> • The HCA strengthened the already close bond shared by families.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> • One individual reported that it was nice to get in touch and catch up with relatives with whom they had been out of touch with by communicating HCA risk.
	Ladd (2010)	<ul style="list-style-type: none"> • Communication about HCA risk connected people who previously had no reason to communicate and these connections were described as positive and cooperative.
	Vavolizza et al. (2015)	<ul style="list-style-type: none"> • Some participants were frustrated and felt isolated from their family when family members chose to not seek testing.
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> • Some participants believed that a healthcare provider might be helpful at building bridges between estranged relatives by being more directly involved in the communication process.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> • In one family, conflicts persisted although they were not attributed directly to the genetic risk communication.

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		<ul style="list-style-type: none"> Despite variations in how individuals handled the HCA risk communication, overall family life stayed relatively normal and on good terms when communication occurred.
		<p>Responsibility Completed:</p> <p><i>Discharging of responsibility for disease risk after communication is done.</i></p> <ul style="list-style-type: none"> Geelen et al. (2011b), p. 1755 One participant's statement, "Now its up to them" reflects the feeling that by communicating to relatives they felt their duty to tell about the risk was complete and that action or inaction by the relatives was no longer their responsibility or concern.
	Mangset et al. (2014)	<ul style="list-style-type: none"> One participant did not even want to know what their relatives chose to do with the risk information.
	Ladd (2010)	<ul style="list-style-type: none"> Not wanting to pressure others was described in one case where a mother urged others to contact a particular cardiologist but left it at that to purposely avoid pressuring others decisions.
		<p>Relative's Lack of Interest or Denial:</p> <p><i>Describes relatives whose response to learning their risk was apparent or real denial or lack of interest.</i></p>
	Whyte et al. (2016)	<ul style="list-style-type: none"> Family member reactions included being uninterested in the HCA risk information.
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> Some participants felt their family members had not taken the risk information seriously enough.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> One participant felt that none of her siblings or parents seemed very interested in the risk information. In a different family, a seemingly healthy relative was not very concerned about the risk, believing there was no way that all the siblings would have the HCA.
	Haukkala et al. (2013) p. 249	<ul style="list-style-type: none"> Upon communicating about the disease risk to her adult children, one mother reported simply that her children thought "it was not a topic for them".
	Vavolizza et al., 2015	<ul style="list-style-type: none"> One participant attributed their relative's decision to not pursue testing to stubbornness. One participant who had sent an email to her entire family did not receive a single response from any family members. Another family had one family member who appeared to ignore the communication.
	Ladd (2010)	<ul style="list-style-type: none"> Felt denial upon learning that her children were at risk for HCM since they appeared healthy and athletic; however, this denial was not sustained because she went on to have her children tested.
		<p>Parental Concern:</p> <p><i>Concerns or worry related to a non-adult child's health, coping, or response to being aware of their disease or disease risk.</i></p>

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	Geelen et al. (2011b)	<ul style="list-style-type: none"> Concern about children preoccupied some parents and diminished their concerns about communicating with other relatives.
	Haukkala et al. (2013)	<ul style="list-style-type: none"> One participant described feeling sad about her daughter's diagnosis because her granddaughter now had to be checked.
	Cohen et al. (2012)	<ul style="list-style-type: none"> Parents felt that the results of positive genetic tests would be difficult for their adolescent children to hear. In one case, parents of a diagnosed teenager wanted more invasive and protective measures for their child since they felt she wasn't compliant with the activity restrictions given after her diagnosis.
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> One mother regretted having her child tested due to the child's extremely poor coping with the disease, despite having felt that it was very logical to get her child tested initially.
	Emotional Reaction: <i>Emotions of the relative that occurred in response to the communication of HCA risk.</i>	
	Haukkala et al. (2013)	<ul style="list-style-type: none"> 24/33 (73%) adult children told about their risk were reportedly 'satisfied' with receiving communication that they were at risk for HCA, however there were no details reported about what being satisfied entailed.
	Vavolizza et al. (2015)	One relative expressed anger and fear about learning about the HCA risk and wished they had never known the information.
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> One child took the information very hard, constantly thinking he was going to die, making his parents question their decision to let him know about his risk, However, two years later the child and his parents had adjusted. Participants were unanimously appreciative of having been told by a relative about their risk.
	Geelen et al. (2011b)	<ul style="list-style-type: none"> One family was shocked to learn about the HCA risk in their family.
	Ladd (2010)	<ul style="list-style-type: none"> The mother of an affected child conceived from a sperm donor imagined what a terrible shock it was for the donor to learn about his own risk, the risk to his children, and the risk he unknowingly passed to many others. Described going from absolute joy to deep devastation upon learning about her child's HCA risk.
	Discontinuation of the Cascade: <i>The breakdown in communication that occurs when relatives who learn about HCA risk fail to pass on risk information to subsequent relatives.</i>	
	Ormondroyd et al. (2014)	<ul style="list-style-type: none"> Some participants believed the HCA risk they communicated to relatives had not been passed on to adult or minor children.
	Haukkala et al. (2013)	<ul style="list-style-type: none"> One participant described a relative who was against passing on the HCA risk information to their children but the reasons for this discontinuation of communication were not known.

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	Geelen et al. (2011b)	<ul style="list-style-type: none"> No outright refusal to continue communication but a perceived lack of immediacy in continuation HCA risk information to children or siblings.
	Haukkala et al. (2013), Vavolizza et al. (2015)	<ul style="list-style-type: none"> Situations where relatives refused to pass on information to their own children were described as extremely difficult and frustrating for probands.
	Difficulty with Relatives' Reactions: <i>Frustration, disappointment, anger, or disagreement with how relatives responded to communication of HCA risk.</i>	
	Geelen et al. (2011a; 2011b)	<ul style="list-style-type: none"> Despite respecting relatives' inactions and remaining on good terms, two participants reported disappointment and difficulty with relatives' lack of response.
	Haukkala et al. (2013), Vavolizza et al. (2015), Whyte et al. (2016)	<ul style="list-style-type: none"> Probands were hurt by relatives' lack of response, frustrated by those who did not want to get testing, or angry or frustrated when relatives declined to pass information on to other family members