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ORIGINAL RESEARCH

Family Communication in a Population at Risk for Hypertrophic Cardiomyopathy

Brittany Batte · Jane P. Sheldon · Patricia Arscott · Darcy J. Huismann · Lisa Salberg · Sharlene M. Day · Beverly M. Yashar

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Abstract Encouraging family communication is an integral component of genetic counseling; therefore, we sought to identify factors impacting communication to family members at risk for Hypertrophic Cardiomyopathy (HCM). Participants (N=383) completed an online survey assessing: 1) demographics (gender, genetic test results, HCM family history, and disease severity); 2) illness representations; 3) family functioning and cohesiveness; 4) coping styles; 5) comprehension of HCM autosomal dominant inheritance; and 6) communication of HCM risk information to at-risk relatives. Participants were a national sample of individuals with HCM, recruited through the Hypertrophic Cardiomyopathy Association. Data from 183 participants were analyzed using a logistic regression analysis, with family communication as a dichotomous dependent variable. We found that female gender and higher comprehension of autosomal dominant inheritance were significant predictors of participants' communication of HCM risk information to all their siblings and children. Our results suggest that utilizing interventions that promote patient comprehension (e.g., a teaching-focused model of genetic counseling) are important and may positively impact family communication within families with HCM.

Keywords Family Communication · Hypertrophic Cardiomyopathy · Risk Information · Gender · Genetic Comprehension · Genetic Counseling

B. Batte · B. M. Yashar
Department of Human Genetics, University of Michigan, Ann Arbor, MI, USA

J. P. Sheldon
Department of Behavioral Sciences, University of Michigan-Dearborn, Dearborn, MI, USA

P. Arscott · S. M. Day
Department of Internal Medicine, University of Michigan, Ann Arbor, MI, USA

D. J. Huismann
Adult Genetics Clinic, University of Colorado Hospital, Aurora, CO, USA

L. Salberg
Hypertrophic Cardiomyopathy Association, Hibernia, NJ, USA

B. Batte (✉)
Department of Gynecologic Oncology, Unit 1362, PO Box 301439, Houston, TX 77230, USA
e-mail: battebr@gmail.com

Introduction

Hypertrophic cardiomyopathy (HCM), a condition characterized by left ventricular hypertrophy, is the most common Mendelian cardiovascular disease. With a prevalence of approximately 1/500, this genetic condition has a profound public health impact (Maron, Maron, & Semsarian, 2012). It is typically inherited in an autosomal dominant pattern, which means that first-degree relatives of a proband are at 50 % risk of inheriting the causal genetic mutation in the family. HCM also shows reduced and age-related penetrance, such that not every person who inherits a mutation in one of the associated genes will be affected and the age of onset or severity cannot be predicted (Jensen et al., 2013). Symptoms of HCM include dyspnea, chest pain, palpitations, and syncope, and disease progression can lead to heart failure. However, there is significant clinical variability, with many patients being asymptomatic with normal longevity (Miller, Wang, & Ware; 2013). A major cause of mortality, especially in young individuals, is sudden cardiac death (SCD; Naghi & Siegel, 2010). Risk stratification is an essential component of care delivery in this patient population, with a substantial proportion of patients eligible for implantable cardioverter-defibrillator (ICD) by

current guidelines (Gersh et al., 2011). The efficacy of these interventions relies on early detection, which can be problematic as knowledge of disease in a family may be limited due to reduced penetrance and the fact that many individuals with HCM are asymptomatic until experiencing an arrhythmia causing cardiac arrest (Miller et al., 2013).

Due to the importance of early detection, recommendations for management of HCM include not only genetic counseling for probands, but also genetic counseling and testing of first- and second-degree family members (Charron et al., 2010; Gersh et al., 2011; Maron et al., 2012). In fact, there has been a recent push in the field of public health to identify individuals at risk for SCD (Michigan Department of Community Health, 2012). Genetic testing allows for the identification of at-risk family members so that individuals with the familial mutation receive proper cardiac care and individuals without the mutation do not undergo costly, unnecessary cardiac screening (Miller et al., 2013). Because of the variability in expressivity and the possibility of mixed phenotypes, family screening also produces additional information necessary for appropriately diagnosing patients (Charron et al., 2010). The identification of at-risk relatives relies heavily upon affected family members communicating information to relatives about the genetic risk for HCM (Hodgson & Gaff, 2013) and the proband's communication to family members is an integral part of cascade testing (Christiaans, Birnie, Bonsel, Wilde, & van Langen, 2008). However, despite the demonstrated importance of family communication in HCM, there is limited psychological research on this population (Aartre & Day, 2011) and thus the specific factors impacting communication are poorly understood. Therefore, the goal of the current study is to assess what factors best predict family communication of risk information in the HCM population.

Studies of information sharing among family members from the field of oncology (Aktan-Collan et al., 2011; LaFrenière, Bouchard, Godard, Simard, & Dorval, 2013; Lindenmeyer et al. 2010; Patenaude et al., 2006; Stoffel et al., 2008) have found that patients' communication of risk information within the family is complex and varied. Consistent with family systems theory, research has demonstrated that family network variables, such as emotional closeness, geographical proximity, social dynamics, and general functioning, are related to family communication of testing results and risk information (LaFrenière et al., 2013; Seymour, Addington-Hall, Lucassen, & Foster, 2010; Weins, Wilson, Honeywell, & Etchegary, 2013; Wiseman, Dancyger, & Michie, 2010). Families create their own culture of "health talk" that may be enhanced by mutual caring and emotional closeness (Lindenmeyer et al., 2010) or impeded by a variety of communication barriers (Forrest et al., 2003). Individuals have multiple motivators for disclosing risk status to their family members, including fulfilling a duty to inform, suggesting testing, and getting advice or emotional support (McGivern et al., 2004; van den Nieuwenhoff, Mesters, Gielen, & de Vries, 2007). The major barriers to communication include social and/or geographical distance from relatives, concerns that family members will not understand, and attempts to protect relatives from worry and distress (Ersig, Hadley, & Koehly, 2011; McGivern et al., 2004; Stoffel et al., 2008).

However, as Nycum, Avard, and Knoppers' (2009) ecological model demonstrates, family communication of risk information is also affected by variables other than those at the familial level. Individual-level factors such as risk perceptions, stress, and personal vulnerability can all impact communication within the family (Nycum et al., 2009; Wilson et al., 2004). Along with perceived risk, other illness representations (e.g., causal attributions, perceived control) have been shown to influence health-related behaviors (Shiloh, 2006). Studies have also found that individuals' coping styles affect family communication (Weins et al., 2013; Wilson et al., 2004), as does their comprehension of complex genetic information (Nycum et al., 2009; van den Nieuwenhoff et al. 2007; Weins et al., 2013). In addition, patients' gender has been found to impact family communication in that women are more likely to be "kinkeepers" in the family (i.e., individuals who keep in touch with other family members) (Wilson et al., 2004) and thus are generally more likely than men to communicate risk information and testing results to other family members (Nycum et al., 2009; Seymour et al., 2010; Weins et al., 2013).

Much of the aforementioned research on factors influencing intrafamilial risk communication has been conducted on individuals with either Lynch syndrome or Hereditary Breast and Ovarian Cancer (HBOC) syndrome; therefore, it is still unknown whether these same factors apply to HCM, a genetic condition for which family communication is also important. Inherited cardiomyopathies, including HCM, are similar to these hereditary cancer syndromes with regard to the age of onset, autosomal dominant inheritance pattern, and mortality risk; however, HCM and HBOC differ in terms of the percentage of males and females affected. As Gaff et al. (2007) asserted, our understanding of intrafamilial communication of genetic information is hampered by the relative lack of research on men's communication experiences. By studying family communication in HCM, we may be able to identify both the unique and the generalizable predictors of family risk communication. Using results from our national sample of 383 women and men with HCM, we hope to gain information that assists genetic counselors and other health-care professionals in ensuring appropriate medical management of HCM for the entire family.

Methods

Participants

Three hundred and eighty-three participants were recruited through the Hypertrophic Cardiomyopathy Association (HCMA), a U.S.-based support group that is dedicated to providing information, support, and advocacy to patients, their families, and medical providers. We excluded participants who were younger than 18 years of age, did not self-report a diagnosis of HCM, and did not report having at-risk adult relatives with which to communicate (i.e., adopted or no living children/siblings over age 18). The disease status of at-risk relatives (defined as blood relations who may have a current diagnosis of HCM or at risk to develop HCM in the future) was based on participant self-report and not verified.

Measures

Demographic Variables Researcher-designed questions were used to assess gender, age, race, ethnicity, and education level, as well as three variables related to clinical vulnerability: a) family history, b) disease severity, and c) genetic test results. Participants reported the number of first- and second-degree relatives (excluding grandparents) with HCM and the number of first-degree relatives who experienced sudden cardiac death. Then, a total family history score was calculated for each participant, with higher scores reflecting a stronger family history. Due to the fact that the study was completed online and participants' medical records were not available, we used ICD status as a proxy for disease severity. Previous work has shown that having an ICD and receiving an ICD shock are associated with psychopathology and lower quality of life (Bostwick & Sola, 2007). We created a categorical scale for assessing ICD status and the three categories were: No ICD, ICD Present But No History of a Shock, and ICD Present with History of a Shock. We coded genetic test results to create a dichotomous variable: Positive Test Result versus No Definitive Test Result (i.e., no mutation detected, variant of uncertain significance, unknown results, or no genetic testing).

Family Communication of Genetic Risk Information Communication of risk information to family members was measured by modifying questions from the McGivern et al. (2004) study on communication of *BRCA1* and *BRCA2* genetic test results. This section consisted of 24 items that assessed reported family structure, methods of communication (e.g., phone, letter, email), and whether the participant had communicated with each relative type about his or her chance to develop HCM. We scored family communication by calculating the proportion of living adult siblings and children in their family that participants told about their risk of developing HCM. We did not include family communication to parents in our measure because many of our respondents' mothers (46.2 %) and fathers (57.7 %) were no longer living at the time of diagnosis. Therefore, we believed we would gain a more accurate representation of family communication by focusing on communication to siblings and adult children only. For the current study, we did not analyze data concerning the method of communication. Because the majority of respondents informed at least one relative and very few communicated with no at-risk relatives, in order to support the statistical analysis, we dichotomized the dependent variable into participants who told all of their at-risk siblings and children versus those who told some or none of their siblings and children.

Family Network Measures We assessed the impact of the family network factors on communication by using validated measures of a) Family Cohesiveness and b) Family Functioning. Specifically, we used a condensed version of the Cohesion subscale of the Family Adaptability and Cohesion Evaluation Scale (FACES-III; Olson, 1986) and we used the 12-item General Functioning Subscale (GFS) of the McMaster Family Assessment Device (Byles, Byrne, Boyle, & Offord, 1988; Epstein, Baldwin, & Bishop, 1983). On the FACES-III subscale participants responded to six statements about their own family's cohesiveness by using a 5-point scale (1=*Almost Never*, 5=*Almost Always*). For the GFS, participants indicated their level of agreement with all 12 statements about their family's functioning, using a scale ranging from 1 (*Strongly Disagree*) to 4 (*Strongly Agree*). Scoring for both measures was performed as previously described (Byles et al., 1988; Epstein et al., 1983; Olson, 1986), with a maximum possible score of 30 for Family Cohesiveness and 48 for Family Functioning. We obtained total scores, with higher scores reflecting greater cohesiveness and increased functioning. Cronbach's alpha coefficients showed strong internal consistency for both measures (.89, .92, respectively).

Illness Representations The Brief Illness Perception Questionnaire (IPQ; Broadbent, Petrie, Main, & Weinman, 2006), a measure of the various components of illness representations, was slightly modified to fit the clinical characteristics of the HCM population. The core components in the scale include identity, causality, timeline, consequences, and cure/control (Leventhal et al., 1997). For each item in the modified, 7-item Brief IPQ, participants responded using a 0 to 10 scale, with descriptors at each anchor. The exact wording of descriptors differed for each item, with a higher rating indicating a more threatening illness perception. Scoring was performed as previously described (Broadbent et al., 2006) for the consequences, identity, coherence, illness concern, emotional representation, personal control, and treatment control items, with higher total scores (out of a maximum of 77) representing a more threatening view of the illness.

Perceived Risk, another component of illness representations, was assessed by two researcher-designed, multiple-choice questions. These questions were: "What is the percentage chance that any one of your children (sons and/or daughters) has a gene mutation that puts them at risk for developing HCM?" and "What is the percentage chance that any one of your siblings (brothers and/or sisters) has a gene mutation that puts them at risk for developing HCM?" Response options for each question were *100%*, *50%*, *25%*, *0%* (*no chance*), and *I don't know.* To code responses, we converted the percentages to decimals and then calculated the mean score for the two items, with higher scores representing greater Perceived Risk. A response of *I don't know* was not included in the scoring.

Comprehension of Autosomal Dominant Inheritance (Comprehension) We measured comprehension of autosomal dominant inheritance by adapting two multiple-choice questions from the Breast Cancer Knowledge Questionnaire (Erblich et al., 2005). The first question asked, "For a person who has been diagnosed with HCM, what is the percentage chance that any one of his or her children (sons and/or daughters) will get a gene mutation that puts them at risk for developing HCM?" The second question asked, "For a person who has been diagnosed with HCM, what is the percentage chance that any one of his or her siblings (brothers and/or sisters) has a gene mutation that puts them at risk for developing HCM?" Response options for each question were *100%*, *50%*, *25%*, *0%* (*no chance*), and *I don't know.* Responses were scored as either correct or incorrect, with one point given for a correct answer (i.e., *50%*) and zero points given for an incorrect answer. A reply of *I don't know* was also given zero points. The mean score of the two questions was then calculated, with a higher mean score representing greater comprehension of autosomal dominant inheritance.

Coping Styles The Brief COPE by Carver (1997) was used to measure coping styles. This measure assesses the main theoretically-derived coping responses, including both problem-focused and emotion-focused styles (Carver, 1997; Carver, Scheier, & Weintraub, 1989). Participants were instructed to respond to each item by remembering how they coped with their illness at the time of diagnosis. For each of the 28 items, respondents use a 4-point scale, with 0 indicating *I Didn't Do This At All* and 3 indicating *I Did This a Lot*.

In order to discover if the factor structure of the measure held for our sample, we performed a factor analysis on responses (N=383), using principal axis factoring with Varimax rotation. Results clustered into aggregates representing each separate coping style and the categories were named according to which items loaded strongly onto each underlying factor (see Table 1). Five factors resulted, each having an eigenvalue greater than 1, and these factors explained 47 % of the variance. The five factors were Planful Problem Solving (λ=5.58), Avoidance and Self-Blame (λ=3.22), Religiosity (λ=1.75), Positive Reframing (λ=1.44), and Substance Use (λ=1.17). Participants' factor scores for each of the five factors were used for analyses, with higher scores representing greater use of the specific coping strategy.

Motivators for and Barriers to Family Communication In order to gain more insight into family communication of HCM risk information, we used measures developed by McGivern et al. (2004) to assess participants' perceived motivators for and barriers to communicating genetic risk information to relatives. Using a scale from 1 (*Not Important at All*) to 5 (*Very Important*), participants rated the importance of five different motivators for communicating with their at-risk relatives (e.g., gain emotional support, inform them of their risk). Participants used the same 5-point scale when rating the importance of seven different potential barriers to family communication (e.g., not wanting to upset relatives, not thinking relatives are at risk). These variables were used for secondary analyses.

Procedures

All study procedures were approved by the Institutional Review Board at the University of Michigan (IRBMED: HUM00014979). HCMA members on the mailing list (N= 5,784) were sent an email notification with a short description of the study and a link to the online survey, which was hosted through the HCMA website (www.4hcm.org). Individuals subscribing to the HCMA mailing list did not necessarily have a diagnosis of HCM. The recruitment message stated: "The purpose of this survey is to better understand the reasons why people with HCM do or do not communicate risk information with their relatives. The focus of this survey will primarily be focused on family communication of risk information and how people perceive and cope with their illness. There are often demographic factors that affect illness perception and family communication; therefore, this survey does contain some questions about your race, ethnicity, and educational background."

The survey was open for a three-week period and three email reminders were sent during that time period. There was also a link to the survey posted on the HCMA website, as well as included in the HCMA newsletter. The 131-item survey was conducted online using Vovici. All participants completed the survey in the same order: demographics, family communication of genetic risk information, motivators of and barriers to communication, family network factors, illness representations, Comprehension, and coping styles.

Table 1 Summary of Items and Factor Loadings for Varimax Rotated, Principal Axis Five-Factor Solution for the Brief COPE scale (N=383)

	Factor Loadings					
Item	1	2	3	4	5	Communality
1 I concentrated on doing Something	**.73**	.02	.02	.07	.04	.54
2 I got support from others	**.58**	.19	.12	-.06	.03	.39
3 I took action to improve things	**.72**	-.18	.06	.13	.00	.57
4 I got help from other people	**.73**	-.08	.10	.03	-.03	.55
5 I came up with a strategy	**.76**	-.16	.04	.11	.06	.63
6 I got comfort from someone	**.63**	.13	.11	.03	-.03	.43
7 I accepted that it happened	**.42**	-.32	.02	.20	.04	.32
8 I expressed my negative feelings	**.39**	.36	.05	.14	.11	.31
9 I got advice from other people	**.69**	.03	.24	.06	-.07	.55
10 I learned to live with it	**.40**	-.17	.05	.23	.02	.24
11 I thought about what steps to Take	**.69**	-.09	.10	.08	.05	.50
12 I turned to work or other Activities	.21	**.30**	.09	.20	.10	.19
13 I said to myself "this isn't real"	-.03	**.63**	-.03	-.01	.00	.40
14 I gave up trying to deal with it	-.30	**.50**	-.01	.12	-.02	.36
15 I refused to believe it happened	-.05	**.61**	-.09	-.05	-.03	.39
16 I let my unpleasant feelings out	.23	**.40**	-.04	.16	.23	.30
17 I criticized myself	.00	**.54**	.07	.08	.18	.34
18 I gave up the attempt to cope	-.25	**.44**	.00	.05	.05	.26
19 I did something to think about it Less	.14	**.40**	.16	.37	.03	.34
20 I blamed myself	-.03	**.53**	.07	-.02	.19	.33
21 I found comfort in my religion	.18	.04	**.89**	.04	-.04	.82
22 I prayed or meditated	.19	.06	**.86**	.00	-.05	.79
23 I tried to see it in a different light	.36	-.02	.29	**.37**	.00	.35
24 I looked for something good in what happened	.29	-.10	.34	**.37**	-.06	.35
25 I made jokes about it	.08	.10	-.01	**.85**	.06	.74
26 I made fun of the situation	.03	.15	-.05	**.69**	.00	.51
27 I used alcohol or other drugs to make myself feel better	.02	.23	-.07	.05	**.87**	.82
28 I used alcohol or other drugs to help me get through it	.01	.16	-.04	.00	**.91**	.86

Note: Boldface indicates highest factor loadings. Factor 1=Planful Problem Solving, Factor 2=Avoidance and Self-Blame, Factor 3=Religiosity, Factor 4=Positive Reframing, Factor 5=Substance Use

Data Analyses

Data were analyzed using SPSS Version 18. To discover what factors predict family communication of HCM risk information, we conducted a multivariate logistic regression analysis, with the dichotomous variable of family risk communication (i.e., told all siblings/children vs. told some or no siblings/children) as the dependent variable. As per standard logistic regression procedures, participants with incomplete data were not included in the regression analysis (referred to as "subsample" hereafter). The independent variables included the four demographic variables of participant gender, family history of HCM, genetic test results, and ICD status (i.e., disease severity). The ten additional independent variables were: a) the two measures of Illness Representations (i.e., score on the Brief IPQ and Perceived Risk score); b) level of Family Functioning, c) degree of Family Cohesiveness, d) factor scores for each of the five Coping Styles; and e) level of comprehension of HCM autosomal dominant inheritance.

To investigate whether the logistic regression's significant predictors were related to family communication when the entire sample was included (i.e., including participants who did not complete the entire survey), we used either Pearson chi-square analyses (for categorical variables) or independent-samples t-tests (for continuous variables).

Results

Demographics

Of the 5,784 HCMA members on the mailing list who received the email invitation, 758 clicked on the email link. Of those, 562 participated in the survey. In addition, 33 individuals clicked on the link via the website. A large number of individuals accessing the survey were ineligible due to not having a diagnosis of HCM, being under 18 years of age, or not having any at-risk adult relatives; therefore, the total number of participants included in analyses was 383. Because HCMA serves not only individuals with HCM, but also unaffected family members and health care providers, the specific number of individuals with HCM who received the email invitation could not be gauged and thus a meaningful response rate could not be determined.

Our study population was composed of 189 women, 192 men, and 2 individuals with unreported gender, all of whom self-reported a diagnosis of hypertrophic cardiomyopathy. Data were incomplete for 200 participants; thus, the final number included in the logistic regression analysis was 183. Demographic characteristics of the entire sample, as well as for the subsample of participants who completed the entire survey, are presented in Table 2.

Family Communication of Risk Information

When we investigated findings for the entire sample (N=383), we found that the majority of participants (72.1 %) communicated with all of their at-risk siblings and children and 23 % communicated with at least one but not all siblings and children. Thus, 95.1 % of the HCMA participants communicated with at least one of their at-risk siblings and/or children. For the subsample (N=183), 74.3 % communicated with all their at-risk siblings and children, 20.4 % communicated with at least one but not all, and 5.3 % communicated with no one.

Predicting Family Communication Results of the logistic regression analysis are summarized in Table 3. The two significant predictors of familial risk communication were participant gender and Comprehension. Gender was predictive of family communication in that women were 2.5 times more likely than men to communicate with all at-risk family members (OR 2.46, 95 % CI 1.11 - 5.43, p=.03). In addition, the greater the participants' Comprehension, the more likely they were to communicate with all at-risk siblings and children (OR 3.57, CI 1.19–10.71, p=.03).

Further Investigation into Predictors of Family Communication Contrary to the logistic regression findings, when the entire sample (N=383) was included in analyses, we found no statistically significant gender difference in familial risk communication, X^2 (1, n=381)=3.01, p=.08. However, in accordance with the regression results, we found in the entire sample that individuals who communicated with all their at-risk children and siblings had higher Comprehension (M=.66, SD=.43) than did individuals who communicated with none or only some of their at-risk family members (M=.55, SD=.46), t (377)=2.14, p=.03.

Secondary Analyses: Motivators for and Barriers to Family Communication

In order to refine our understanding of the process of intrafamilial communication of HCM risk information, we also explored respondents' motivators for and barriers to family risk communication. Using both the entire sample and the subsample (i.e., those who completed the entire survey), for each of the five motivators for communication and seven barriers to communication we computed means and standard deviations for participants' importance ratings. Then, for both samples we investigated how the logistic regression's significant predictors of family communication related to the importance participants ascribed to each potential motivator for and barrier to communication. We assessed the relationships between motivators (or barriers) and categorical variables using independent-samples t-tests. We analyzed relationships between motivators (or barriers) and continuous variables by using Pearson Product Moment correlations.

Table 2 Characteristics of the Study Population and Subsample

Demographic Variables	Study Population (N=383)	Study Subsample (N=183)
Age (Mean)	52.35	49.69
Gender (% Male)	50.1 %	51.9 %
Race (% White)	93.2 %	95.0 %
Ethnicity (% Non-Hispanic)	97.5 %	98.3 %
Education Level (% College Degree or higher)	64.8 %	55.2 %
Family History (% Positive)	63.2 %	66.3 %
Genetic Testing (% who had genetic testing)	46.2 %	64.2 %
Genetic Test Results (% Positive)	38.8 %	43.1 %

Note: The study subsample consists of respondents who completed the entire survey and thus were included in the regression analysis

Table 3 Results of Logistic Regression Analysis Investigating Predictors of Family Communication of HCM Risk Information

Variables	β	S.E.	Wald Test	Odds Ratio	95 % Confidence Interval	*p*-value
Genetic Test Results	.14	.43	.11	1.15	.50 – 2.65	.74
Family History	-.23	.14	2.71	.80	.61 – 1.04	.10
ICD Status	-.12	.26	.20	.89	.53 – 1.49	.65
Participant Gender	.90	.41	4.93*	2.46	1.11 – 5.43	.03
Illness Representations	-.01	.02	.36	.99	.95 – 1.03	.55
Perceived Risk	-.05	1.2	.00	.96	.10 – 9.41	.97
Family Functioning	.07	.04	2.88	1.07	.99 – 1.16	.09
Family Cohesiveness	-.10	.06	2.99	.91	.82 – 1.10	.08
Coping Style: Planful Problem Solving	-.04	.35	.01	.96	.48 – 1.91	.91
Coping Style: Avoidance and Self-Blame	-.32	.44	.52	.73	.31 – 1.62	.47
Coping Style: Religiosity	.00	.18	.00	1.00	.70 – 1.44	.10
Coping Style: Positive Reframing	-.04	.27	.03	.96	.57 – 1.62	.87
Coping Style: Substance Use	-.42	.29	2.15	.66	.37 – 1.15	.14
Comprehension of HCM Autosomal Dominant Inheritance	1.27	.56	5.17*	3.57	1.19 –10.71	.03

Note: The dichotomous dependent variable (family communication) was Told All Siblings/Children vs. Told Some or No Siblings/Children. *$p<.05$. The Hosmer-Lemeshow Goodness-of-Fit Test was not significant, X^2 =8.58, df=8, p=.381, and the Nagelkerke R Square was .171

Motivators for Family Communication As can be seen in Table 4, overall, participants indicated that informing relatives of their risk of developing HCM was the most important motivator for communicating with family members and that suggesting to relatives that they get tested was the second most important motivator.

Analyses using the subsample showed no significant gender differences in motivators for family communication (Table 5). However, as Table 5 demonstrates, analyses of the entire sample demonstrated that women were more likely than men to state that it was important to communicate in order to suggest that family members

Table 4 Motivators for and Barriers to Family Communication: Means and Standard Deviations for Importance Ratings

	Entire Sample (*N*=383)			Subsample (*N*=183)		
Motivators for Communicating with Relatives	*M*	*SD*	*n*	*M*	*SD*	*n*
To inform them of their risk	4.84	.49	381	4.86	.43	183
To suggest that they are also tested	4.66	.75	377	4.72	.67	181
To fulfill your duty to inform them	4.46	1.00	379	4.42	1.00	182
To get advice on medical treatment	3.89	1.52	373	3.91	1.49	181
To get emotional support	3.26	1.50	370	3.31	1.47	179
Barriers to Communicating with Relatives	*M*	*SD*	*n*	*M*	*SD*	*n*
You were not in contact with them	2.19	1.58	259	2.00	1.49	135
You were not close with these relatives	2.12	1.51	252	1.94	1.42	132
You did not think that they would care	1.78	1.26	251	1.64	1.19	133
You did not want to upset them	1.68	1.24	250	1.64	1.21	132
You did not think that they were at risk	1.65	1.20	251	1.69	1.18	130
You were having difficulty coping	1.51	1.09	250	1.52	1.13	133
You did not know what to say	1.44	.99	250	1.32	.78	133

Note: Importance ratings ranged from 1 (*Not At All Important*) to 5 (*Very Important*). Participants who communicated with all their siblings and children did not respond to items related to communication barriers. The study subsample consists of respondents who completed the entire survey and were included in the regression analysis

Table 5 Men's Versus Women's Motivators for and Barriers to Family Communication: Means and Standard Deviations for Importance Ratings

	Entire Sample					Subsample				
	Men (n=192)		Women (n =187)			Men (n=95)		Women (n=88)		
Motivators for Communicating	*M*	*SD*	*M*	*SD*	*p*	*M*	*SD*	*M*	*SD*	*p*
To inform them of their risk	4.80	.55	4.89	.42	.07	4.82	.46	4.91	.39	.16
To suggest that they are also tested	4.58*	.81	4.75*	.68	.03	4.63	.80	4.82	.47	.06
To fulfill your duty to inform them	4.51	.88	4.41	1.12	.34	4.50	.85	4.34	1.13	.29
To get advice on medical treatment	3.79	1.53	3.99	1.50	.20	3.90	1.47	3.91	1.53	.99
To get emotional support	3.03*	1.53	3.50*	1.44	.002	3.21	1.56	3.42	1.36	.35
	Men (n=129)		Women (n=129)			Men (n=67)		Women (n=68)		
Barriers to Communicating	*M*	*SD*	*M*	*SD*	*p*	*M*	*SD*	*M*	*SD*	*p*
You were not in contact with them	1.94*	1.44	2.43*	1.68	.01	1.70*	1.27	2.29*	1.63	.02
You were not close with these relatives	1.90*	1.36	2.34*	1.61	.02	1.79	1.28	2.09	1.54	.22
You did not think that they would care	1.56	1.13	1.79	1.33	.14	1.55	1.13	1.73	1.25	.40
You did not want to upset them	1.57	1.13	1.73	1.27	.31	1.61	1.21	1.66	1.22	.81
You did not think that they were at risk	1.76	1.22	1.81	1.30	.80	1.65	1.14	1.74	1.23	.66
You were having difficulty coping	1.57	1.16	1.46	1.03	.44	1.63	1.27	1.41	.98	.27
You did not know what to say	1.48	1.02	1.41	.97	.59	1.40	.91	1.24	.63	.24

Note: *p<.05. Analyses were independent-samples t-tests comparing men's and women's ratings on each item. Importance ratings range from 1 (*Not At All Important*) to 5 (*Very Important*). Participants who communicated with all their siblings and children did not respond to items related to communication barriers. The study subsample consists of respondents who completed the entire survey and were included in the regression analysis

get tested [t (373)=2.15, p=.03] and to get emotional support [t (367)=3.05, p=.002].

For the subsample, analyses showed that the greater participants' Comprehension, the more importance they gave to informing family members of their risk (r=.29, p=.0001, n=183) and suggesting that they get tested (r=.22, p=.003, n=181). When using the entire sample, the results demonstrated that the more Comprehension participants had, the more important they thought it was to suggest to family members that they get tested (r=.19, p=.0001, n=374).

Barriers to Family Communication Table 4 shows that, overall, participants reported that the two most important barriers to communication with at-risk relatives were lack of contact and lack of closeness with those relatives.

As can be seen in Table 5, for the subsample we discovered that women were more likely than men to say that an important communication barrier was being out of contact with their relatives, t (133)=2.36, p=.02. Our analyses using the entire sample demonstrated that women were more likely than men to state that important barriers to communication were lack of contact with their relatives [t (256)=2.55, p=.01] and not being close with their family members [t (249)=2.33, p=.02] (Table 5).

For the subsample, results showed that the less comprehension individuals had about autosomal dominant inheritance, the more they felt that an important barrier to their communication was not thinking their relatives were at risk (r=−.19, p=.03, n=130). When using data from the entire sample, the analyses demonstrated that the less Comprehension individuals had, the more they felt that an important barrier to their family communication was not knowing what to say (r=−.15, p=.02, n=249) and not thinking that family members were at risk (r=−.16, p=.01, n=249).

Discussion

In our national study of familial communication in 383 individuals with a diagnosis of hypertrophic cardiomyopathy, we found evidence of a high level of information sharing within the families. Ninety-five percent of study participants reported sharing information about the diagnosis and the risk of developing the condition with at least one of their at-risk, first-degree relatives. Our findings concerning factors that promote and hinder information sharing within the family were consistent with those seen in other studies on risk communication and suggest that comprehension of risk for other family members is a strong driver of communication, whereas impaired family functioning may hamper communication within the family. In addition our findings of gender-related differences in risk communication highlight opportunities for clinicians to improve intrafamilial information sharing.

Our results are consistent with previous research on family communication in hereditary cancer syndromes (McGivern

et al., 2004; Stoffel et al., 2008), particularly with research on family communication in Lynch syndrome that showed that 98 % of respondents communicated with at least one at-risk first-degree relative (Stoffel et al., 2008). Lynch syndrome is similar to HCM in that it affects both genders and has screening recommendations that apply to all family members, regardless of genetic test results. Thus, our findings are highly encouraging in that they show at least some amount of family communication. However, because an important outcome of genetic-based clinical care and genetic counseling is to help patients communicate with *all* at-risk relatives, our regression results, discussed below, may help us better understand specific factors to target in interventions aimed at increasing family communication to *all* at-risk children and siblings.

Predictors of Family Communication

We found that participants with higher overall comprehension of the inheritance of HCM as an autosomal dominant condition were significantly more likely than those with less comprehension to communicate with all of their at-risk siblings and children. Additionally, lower comprehension was associated with assigning greater importance to the communication barrier of not thinking other family members were at risk. Van den Nieuwenhoff et al. (2007) found that a common reason for non-disclosure to relatives at risk for inherited hypercholesterolemia was a lack of comprehension concerning the risk for relatives to be affected. In addition, Cheung, Olson, Yu, Han, and Beattie (2010) saw a positive association between family communication of *BRCA1* or *BRCA2* test results and participants' comprehension of HBOC screening and of risk reduction recommendations. Although the type of communication that Cheung et al. measured was not identical to that in our study, their findings, along with van den Nieuwenhoff et al.'s (2007) and ours, suggest that improving patient education may be a way to increase family communication of risk information. Particularly important in patient education may be an explanation of the clinical variability of HCM, because previous research has shown that patients may have lay beliefs that inheritance inevitably results in physical and/or behavioral resemblance between family members (Lindenmeyer et al., 2010). Thus, the meaning of a positive genetic test result may be difficult for individuals to conceptualize, both for themselves and for other family members, in the absence of clinical evidence of disease. Direct assessment of patients' level of comprehension and an exploration of how the patient translates this knowledge into disease risk for family members, therefore, are paramount in clinical encounters.

In terms of the effect of gender on familial risk communication, we found conflicting results. For the entire sample we found no gender difference, which is consistent with some of the previous research on communication of hereditary cancer syndrome risk (Aktan-Collan et al., 2011; Ishii et al., 2011; McGivern et al., 2004; Patenaude et al., 2006; Ratnayake et al., 2011; Stoffel et al., 2008; Vos et al., 2011). However, our findings from the regression analysis showed that women were more likely than men to have communicated with all their at-risk relatives. These findings are in accordance with D'Agincourt-Canning's (2001) contention that family communication about health issues tends to be the assumed responsibility of women. In support of this view, when Koehly et al. (2009) looked at characteristics of information gatherers, disseminators, and blockers in families with a *BRCA1* or *BRCA2* mutation, they discovered that gatherers and disseminators were significantly more likely to be women, whereas the blockers were more likely to be men. Others (Koehly et al., 2003; Lindenmeyer et al., 2010; Wiseman et al., 2010) have also found this gender difference in family communication. However, some research has indicated that men are more likely to use intermediaries to communicate health information within the family (Gaff et al., 2007); therefore men's communication may be similar in scope to women's, but may take a different form. Thus, the fact that we did not find a gender difference in intrafamilial risk communication when we used the entire sample may partially reflect Gaff et al.'s (2007) finding regarding men's use of intermediaries. Future research on family risk communication will benefit from assessing not only the amount of direct communication, but also the intrafamilial recruitment of intermediaries.

Our findings provide support for using family systems theory (Gaff, Galvin, & Bylund, 2010; Rolland & Williams, 2005) to help explain motivators for and barriers to familial communication of HCM risk information, in that we discovered, for both the entire sample and the subsample, that the two most important barriers to participants' intrafamilial communication were not being in contact with relatives and not being close to them. Therefore, our results concerning barriers to communication are consistent with research showing significant relationships between family communication and family network constructs in patients with cancer and other hereditary diseases (Fehniger et al., 2013; Harris et al., 2010; Koehly et al., 2003; LaFrenière et al., 2013; Seymour et al., 2010; Weins et al., 2013; Wiseman et al., 2010). Our findings are also consistent with research (Gaff et al., 2007) showing that most of the failure to communicate genetic information is due to passive rather than active nondisclosure, so that individuals are not deliberately choosing to withhold information but instead are influenced by family dynamics that hinder communication. Family closeness is often manifested and/or created by the sharing of vital health information and the development of a collective, family health narrative (Lindenmeyer et al., 2010); therefore, individuals are less likely to share health information when cohesiveness within the family is lessened, such as by lack of contact or lack of closeness.

Given our finding that two factors that pertain to the family network, lack of contact and lack of closeness, are barriers to communication, we would have expected in our regression analysis that the two family network variables—Family Functioning and Family Cohesiveness—would have been significant predictors of intrafamilial risk communication. Importantly, however, whereas the items concerning barriers to communication specifically pertain to communication concerning the health risk of HCM, the Family Functioning and Family Cohesiveness scales are global measures that do not include health-related assessments. Therefore, it may be that such general measures of family functioning and cohesiveness are not adequate assessments of family closeness as it specifically relates to our respondents' collective, family health narratives.

Practice Implications

A goal of our study was to analyze factors that may impact the communication of risk information to family members of HCM patients. There are several important implications that are evident in our results for clinical practice in general and genetic counseling practice specifically. Our finding that comprehension of autosomal dominant inheritance is a significant predictor of family communication demonstrates the critical role of education in creating positive outcomes from both types of clinical encounters. In a review on the content and process of genetic counseling (Meiser, Irle, Lobb, & Barlow-Stewart, 2008), two main professional approaches to genetic counseling were identified: the teaching model and the counseling model. Meiser et al. (2008) found that a large proportion of communication in counseling sessions is biomedical rather than psychosocial, with the teaching model of genetic counseling being most widely implemented. Our results support the value of the teaching that occurs during the genetic counseling process and suggest that some reliance on a teaching-focused model of practice could be an effective way to improve familial risk communication. However, while genetic counselors may be preferentially utilizing a teaching model, relying exclusively on this approach may not support long-term comprehension. Research (DiCastro et al., 2002) indicates that genetic counseling patients do not always retain the risk information provided to them in a readily accessible format, which can further inhibit their ability to communicate this risk information to relatives. In addition, understanding communication within a family from a transactional perspective ("sender-receiver model") highlights the fact that each time information is communicated within a family, the interaction between the family members simultaneously affects each participant (Bylund, Galvin, & Gaff, 2010). Consequently, it is critical to ensure that the teaching model is intertwined with careful exploration of the patients' belief system regarding genetic causation and illness and to explore potential reactions from family members to the information that the patient will be sharing (Eunpu, 2010).

One strategy to improve this communication could be utilizing follow-up clinical encounters to re-emphasize the importance of family communication, identify family members who are at risk, and explore the discussions that have already occurred from the perspective of the patient and his or her family members. Participation of genetic counselors in these follow-up visits could also have a positive impact on the use of genetic services by the at-risk family members. Educational interventions, including using the family pedigree to identify at-risk relatives and specifically addressing the importance of family communication, have been shown to be efficacious (Forrest, Burke, Bacic, & Amor, 2008) and continue to be developed, refined, and tested (Gaff & Hodgson, 2014; Hodgson, Metcalfe, Aitken, Donath, Gaff, & Winship, 2014). Our results, too, suggest that these types of interventions may be an excellent way to improve family communication. For instance, using the family pedigree may not only help in identifying at-risk relatives, but may also increase patients' comprehension of autosomal dominant inheritance—a form of comprehension we found to be predictive of strong family communication.

Additionally, when discussing with patients the importance of family communication, it is beneficial to explore how both genders in the family network can take responsibility for communicating risk information to at-risk relatives. Providing family notification letters to all patients with HCM may be an important tool to aid communication with relatives (van der Roest, Pennings, Bakker, van den Berg, & van Tintelen, 2009), as well as reduce the over-reliance on female family members for the communication of familial risk information. This would also ensure accuracy in the information presented to family members, as research has shown that many errors occur in the transmission of genetic test results to relatives (Vos et al., 2011).

Study Limitations

Despite our study's strengths, including sample size, diverse study population, inclusion of men, and assessment of novel variables impacting family communication, it is important to be cognizant of several limitations. During recruitment we informed the participants that the survey was about communication; therefore, there is the possibility of a response bias so that those who did not communicate with their at-risk relatives may have been less likely to complete the survey. Additionally, although the recruitment of participants from a national HCM support group increased our sample's geographic diversity, our sample may not be representative of clinic populations with HCM. For example, individuals from HCMA—a support group providing numerous educational resources—may have greater comprehension of HCM inheritance. Our

sampling technique (i.e., utilizing the HCMA mailing list) also precluded us from determining whether respondents were from the same family and from calculating a meaningful response rate for our study. Thus, multiple affected relatives from the same family may have participated in this study. We were also unable to determine whether participants completed the questionnaire through the email link, website posting, or both. Disease status of at-risk relatives was not included in our questionnaire, and thus some at-risk relatives may have been affected with HCM themselves.

As with all self-report research, there may also have been a social desirability bias in that participants may have over-reported the number of siblings and children with whom they communicated. However, because all data were de-identified and anonymous, this is less likely to have been problematic in our study. Because we did not survey all family members, we faced the limitations found in other family communication research (e.g., Fehniger et al., 2013; van den Nieuwenhoff et al., 2007) that we were unable to confirm that our participants actually communicated with their relatives and that they provided family members with accurate information. Also, due to our inability to verify the content and providers of any genetic counseling that participants may have obtained, we could not accurately measure this construct; therefore, we do not have data regarding participants' genetic counseling experience.

Although the two samples we used for analyses were comparable in terms of all demographic variables except the percentage that obtained genetic testing, it is possible that this particular group disparity may represent a more general, intrinsic group difference (e.g., in motivation, interest, or proactive behavior) that may have contributed to our different findings for the two samples. In addition, because our small numbers necessitated that we group together participants who communicated with some affected relatives and those who talked to no relatives, we were not able to make comparisons between these two subgroups and participants who communicated with all affected relatives. For our exploratory analyses regarding motivators and barriers, we used an alpha level of .05 for our multiple comparisons; therefore, it is possible that some of our findings are due to chance. Finally, as with many other studies on familial risk communication (e.g., Fehniger et al., 2013; Wilson et al., 2004), we do not have any long-term data on the outcomes of family communication, including rate of genetic testing, clinical screening, genetic counseling, sudden cardiac arrest, and heart failure in at-risk relatives.

Research Recommendations

Because this was an exploratory study, future studies with a larger sample size and in other patient cohorts are needed in order to confirm these results. As mentioned previously, it will also be important for future studies to assess long-term outcomes, such as uptake of genetic testing and counseling, in order to highlight the importance of familial risk communication on improving medical management in at-risk relatives. By also surveying the at-risk relatives, not just individuals with an HCM diagnosis, these additional studies may be able to confirm the accuracy of information provided and determine the psychological effects of being informed of the risk to develop HCM.

Conclusions

Family communication of genetic information is a complex process composed of various motivators and barriers. In this study of individuals with a diagnosis of HCM, we have shown that gender and comprehension of autosomal dominant inheritance are significant predictors of communication with all at-risk siblings and children. These findings have important implications for clinical practice in that genetic counselors can tailor their counseling to ensure patients understand exactly which relatives are at risk, help their patients explore the barriers that could hamper the sharing of information, and identify personal characteristics and aspects of family functioning that would strengthen their ability to talk about this difficult topic.

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Conflict of Interest Brittany Batte, Jane P. Sheldon, Patricia Arscott, Darcy J. Huismann, Lisa Salberg, Sharlene M. Day, and Beverly M. Yashar declare that they have no conflict of interest.

Human Studies and Informed Consent All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000. Informed consent was obtained from all patients for being included in the study.

Animal Studies No animal studies were carried out by the authors for this article.

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