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ORIGINAL RESEARCH

All in the Family: Barriers and Motivators to the Use of Cancer Family History Questionnaires and the Impact on Attendance Rates

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Abstract Data has demonstrated that family history questionnaires (FHQs) are an invaluable tool for assessing familial cancer risk and triaging patients for genetic counseling services. Despite their benefits, return rates of mailed FHQs from newly referred patients remain low, suggesting potential barriers to their use. To investigate this, a total of 461 participants, 239 who completed a FHQ (responders) and 222 who did not (non-responders), were surveyed at a subsequent appointment regarding potential barriers and motivators to using the FHQ. With respective rates of 51 and 56 %, there was no significant difference in the proportion of responders and non-responders who reported difficulty in completing the FHQ; however, for both groups factors related to family dynamics (large family size, lack of contact with relatives, and lack of knowledge of family history) were reported as major variables confounding completion of the FHQ. Responders were also significantly more likely to have a personal diagnosis of cancer (p=0.02) and to report that their physician had discussed the reason for the appointment with them (p= 0.01). Overall, 19 % of non-responders returned their FHQ after being mailed an appointment letter and 67 % attended their scheduled genetic counseling appointment. These findings demonstrate that difficulty completing the FHQ is not inherent to its design but due to difficulty accessing one's family history, and that mailed appointment letters are a highly successful way to increase attendance rates in the non-responder population. Furthermore, these results demonstrate the important role that referring physicians play in the utilization of genetic counseling services.

Keywords Genetic counseling · Family history questionnaire · Hereditary breast and ovarian cancer · Service utilization

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Introduction

With increasing knowledge and heightened public awareness of the hereditary component of breast and ovarian cancer, the demand for genetic counseling services has increased. In an effort to meet this growing demand and increase efficiency, many clinics have implemented the use of mailed family history questionnaires (FHQ) to obtain family history information from patients in advance of genetic counseling. From the patient's perspective, the use of a mailed FHQ provides the opportunity to consult relatives and collect accurate family history information prior to counseling; thereby leading to greater confidence in the accuracy of the information received during genetic counseling (Hallowell et al. 1997). From the genetic counselors' perspective, the use of a mailed FHQ may minimize the amount of time spent in direct patient contact as

family history information and relevant pathology records may be reviewed prior to counseling. While clearly desirable to both patients and genetic counselors, a number of studies have also confirmed that a self-administered FHQ is an accurate tool for the systematic collection of family history information (Armel et al. 2009; Qureshi et al. 2009; Vogel et al. 2012). Furthermore, FHQs have been demonstrated to be effective in triaging patients for genetic counseling and assessing genetic risk; however, despite these recognized benefits, response rates to mailed FHQs remain low, ranging from 25 to 53 % (Armel et al. 2009, 2011; Chalmers et al. 2001; Leggatt et al. 1999; Mancuso et al. 2004).

Although FHQs are an invaluable tool in the cancer genetics clinic, low response rates to mailed questionnaires suggests that barriers related to their use may exist. In two small telephone surveys, 89 and 63 % of patients that did not complete a mailed FHQ reported that they intended to, confirming a definitive gap between intent and actual response (Appleby-Tagoe et al. 2012; Poplawski et al. 2009). In an attempt to understand reasons for non-response, two Canadian studies reported that the most commonly cited reasons for non-completion were related to being busy, procrastinating, having limited access to family history information, and bad timing (Appleby-Tagoe et al. 2012; Armel et al. 2011). Of interest, non-completion of the FHQ was not attributed to lack of perceived benefit of genetic counseling or lack of perceived risk of cancer; only 7 % of patients reported that uncertainty about having genetic counseling and testing attributed to their non-response (Armel et al. 2011). As compared to individuals who completed a mailed FHQ, non-responders were significantly more likely to cite fewer overall perceived benefits of cancer genetic counseling (Appleby-Tagoe et al. 2012). In the context of usability, Roth et al. examined the impact of literacy rates and demonstrated no correlation between lower literacy rates and non-completion of a FHQ (Roth et al. 2009). Similarly, when evaluating ethnic background as a predictor of response, Mancuso et al. identified that visible minorities were less likely to respond to a mailed FHQ than non-minorities; however, only 12.3 % cited language as a barrier (Mancuso et al. 2004).

Despite less than ideal response rates, patients have reported that a mailed FHQ was the most desirable method for collecting family history information as compared to alternative forms of delivery such as in person, by telephone, or through email (Appleby-Tagoe et al. 2012). In an effort to improve low response rates to mailed FHQs, a small number of studies have noted increased return rates with the implementation of telephone prompts or the use of a patient navigator. In three studies, the use of telephone prompting resulted in 25–43 % of non-responders returning their completed FHQ (Appleby-Tagoe et al. 2012; Mancuso et al. 2004; Poplawski et al. 2009). In another study employing a patient navigator to contact patients and discuss the referral process, answer questions, and schedule appointments, an increase in genetic counseling attendance rate of 13 % was observed (Rahm et al. 2007).

Aside from having a personal diagnosis of cancer, little is known as to what motivates completion of a FHQ and ultimately attendance at a genetic counseling appointment. In a previous study, 85 % of breast cancer patients and 76 % of patients without a cancer diagnosis considered their doctor's referral as a strong motivator to attend genetic counseling (Chin et al. 2005). Likewise, among 122 young breast cancer survivors, 41 % cited their doctor's recommendation to attend genetic counseling as a major contributing factor to participating in genetic counseling (Anderson et al. 2012). In contrast, studies in both young breast cancer survivors and relatives of *BRCA1* and *BRCA2* carriers reported their lack of knowledge or lack of recommendation for genetic risk assessment by their physician as frequent barriers for genetic counseling (Anderson et al. 2012; Wakefield et al. 2011). Another study found that only 42 % of patients indicated having received sufficient information from their physician regarding the nature of their referral and genetic counseling (Appleby-Tagoe et al. 2012). Consistent with patient reports, a study using a patient navigator model cited that the most commonly addressed questions during referral follow-up were related to the process of genetic counseling (Rahm et al. 2007). A lack of either patient or provider knowledge was also identified by 36 % of genetic counselors to be a perceived barrier to attending genetic counseling (Rolnick et al. 2011). Therefore, a general lack of knowledge by referring physicians, as well as a lack of both confidence and adherence regarding referral of high-risk families for genetic counseling may further complicate the referral process (Bellcross et al. 2011; Mouchawar et al. 2001; Trivers et al. 2011; Wideroff et al. 2003).

Despite evidence suggesting similar levels of genetic risk in populations of patients that complete a FHQ and those that do not, without completing a FHQ, patients may not be routinely offered an appointment for genetic counseling (Armel et al. 2011). Factors such as increased anxiety, family obligations, insurability, and time commitment have been consistently identified as barriers for prospective patients regarding genetic counseling or testing, and may also play a role in lack of response to a mailed FHQ (Cappelli et al. 1999; Foster et al. 2004). Certainly, evidence suggests that factors related to timing and knowledge of family history can be a significant barrier to this process (Appleby-Tagoe et al. 2012; Armel et al. 2011). Similarly, lack of patient and provider knowledge regarding the value of genetic counseling may also lead to decreased response rates and, in turn, reduced attendance for genetic counseling. In the current study, barriers and motivators related to the design, use, and implementation of a mailed FHQ from a single cancer genetics clinic were examined in both a population of patients who completed the FHQ and those who did not. It is hoped that by understanding how patients perceive the FHQ and by evaluating whether they find inherent barriers in its design and

usability that it will facilitate both an improvement in patient adherence, as well as in the utilization of cancer genetic services on a global scale.

Methods

Referral Process

In the Familial Breast and Ovarian Cancer Clinic (FBOCC) at the Princess Margaret Cancer Center (PM) in Toronto ON, all newly referred patients are asked to complete a family history questionnaire (FHQ) and a personal history questionnaire (PHQ). As described in previous publications, the FHQ consists of a series of questions and tables to obtain a three-generation pedigree for the patient's family while the PHQ consists of a series of questions to obtain the patient's medical history, including personal diagnosis of cancer and previous genetic testing in the family (Armel et al. 2009, 2011). For patients who are referred by physicians within the PM, the FHQ is provided directly to the patient by the referring practitioner's clinic. For referrals received from physicians external to PM, the FHQ is mailed to the patient's home address once the referral is received by the FBOCC. Following review of the completed FHQ by a genetic counselor, patients with a relevant personal history or family history of cancer are offered an appointment. All patients are given a reminder telephone call 1 week prior to their appointment.

Study Design

To determine potential barriers and facilitators to completing the FHQ, a survey-based study was designed. Patients who returned their completed FHQ within 3 months of referral were mailed an appointment time and date. Patients whose completed FHQ was not received within 3 months of referral were also mailed an appointment time and date. Eligible patients, seen between July 3 2012 and August 30 2013, were offered the opportunity to complete a short survey prior to meeting with a genetic counselor. Informed consent was obtained by a study coordinator. Study eligibility was determined during a chart review 1 week prior to appointment dates. Two surveys were designed: one which was given to patients whose completed FHQ was received by the time of chart review and another which was given to patients whose FHQ was not received or was returned after chart review. A data collection sheet was used to obtain demographic information from the patient chart. The study protocol was approved by the research ethics board at the University Health Network.

Study Population

The study population was divided into two groups: responders and non-responders. As described, participants who returned the FHQ more than a week before their appointment were defined as responders; however, responders who returned their FHQ only after receiving an appointment letter were flagged for additional analysis. Based on similarities in baseline characteristics to the responder group, non-responders who returned their FHQ by the time of their appointment were redefined as responders for study analysis. Participants whose FHQ was never returned were defined as non-responders.

Participants were excluded if their appointment was scheduled by telehealth, if they or a family member had been seen by a genetics clinic in the past, or if it had been greater than 1 year since their referral. Non-responders were also excluded if it had been less than 3 months since their referral to allow for sufficient time to complete the FHQ. Participants who did not speak English were eligible for the study provided that an interpreter was available.

Statistical Analysis

Continuous variables were reported as medians and ranges and were analyzed using student's *T*-test. Categorical variables were reported as percentages and frequencies and were analyzed using Chi-Square or Fisher's exact, where appropriate. Univariable analysis was conducted to compare baseline characteristics of non-responders with responders as well as their difficulty in answering or completing the FHQ. Multivariable logistic regression analysis was conducted to report statistically significant variables which determined the likelihood of the FHQ being difficult to complete or not completed. The statistical significance level was chosen as a p value of 0.05 or less. SAS v9.2 was used for all statistical analysis.

Results

Study Participants

A total of 1043 new patients were assessed for eligibility; 43.0 % (448) had completed their FHQ at least 1 week prior to their appointment and 57.0 % (595) had not. Of note, 3.6 % (16/448) provided their FHQ only after they were mailed an appointment letter. Of the 448 patients who completed their FHQ, 65.2 % (293) were eligible to participate in the study. Reasons for exclusion included reporting that they (25/155= 16.1 %) or a family member (112/155=72.3 %) had previously been seen by genetics, inappropriate time between referral and appointment date (9/155=5.8 %), and an appointment scheduled by telehealth (9/155=5.8 %). An additional five patients (1.7 %) were excluded as they did not attend their scheduled genetic counseling appointment. Of the 288 patients who completed the FHQ and attended an appointment at the FBOCC, 83.0 % (239) consented to participate and

completed the study questionnaire. All 239 were classified as responders.

Of the 595 patients who had not completed their FHQ at least 1 week prior to their appointment, 63.2 % (376) were eligible to participate in this study. Reasons for exclusion included administration error (9/219=4.1 %), language barrier (2/219=0.9 %), reporting that they (7/219=3.2 %) or a family member (52/219=23.7 %) had previously been seen by genetics, and inappropriate time between referral and appointment date (147/219=67.1 %). Two patients (2/219=0.9 %) were excluded as they were deceased at the time of the study. An additional 34.0 % of patients (128) were excluded as they did not attend their scheduled genetic counseling appointment. Of the 248 patients who were seen at the FBOCC, 89.5 % (222) consented to participate and successfully completed the study questionnaire. Sixty of these participants returned their FHQ during the week of their appointment and were reclassified as responders, giving a total of 299 responders. The remaining 162 participants were classified as non-responders.

Participant demographics comparing non-responders with responders are summarized in Table 1. The majority of participants were highly educated females of Caucasian descent. The average age of responders and non-responders was similar, at 48 (18–82) and 47 (21–74), respectively. The majority of participants reported a positive family history of cancer; however, responders were more likely to report a personal history of cancer ($p<0.001$). This was particularly evident for a personal diagnosis of ovarian cancer. Responders were also more likely to have children ($p=0.003$).

In the province of Ontario, established Ministry of Health eligibility criteria outline which individuals may be offered genetic testing for mutations in the *BRCA1* and *BRCA2* genes. On the date of their appointment, 59.2 % (177) of responders and 32.1 % (54) of non-responders were eligible for genetic testing ($p<0.001$). Only those individuals eligible for genetic testing were offered it. Of note, two responders were unable to proceed with genetic testing due to recent blood transfusions and one responder and two non-responders that were eligible for testing declined it. As shown in Table 1, statistical differences were seen between responders and non-responders who proceeded with genetic testing. Of those tested, 11 % (19) of responders were found to have a *BRCA1* or *BRCA2* mutation and 89.0 % (153) received negative or variant results; 1.9 % (1) of non-responders were found to have a *BRCA1* or *BRCA2* mutation and 98.1 % (51) received negative or variant results ($p=0.05$).

Table 1 Participant demographics

Variable	Non-responders median (range) or % (N) *N*=162	Responders median (range) or % (N) *N*=299	p-value
Age (in years)	47 (21–74)	48 (18–82)	0.82
Sex			0.053
Female	97.5 (158)	99.7 (298)	
Male	2.5 (4)	0.3 (1)	
Ethnicity			0.07
African	0.6 (1)	0.67 (2)	
Asian	12.4 (20)	19.7 (59)	
Caucasian	69.6 (112)	62.2 (186)	
Hispanic	0.6 (1)	3.7 (11)	
Other	16.8 (27)	13.7 (41)	
Personal history of cancer	36.5 (59)	59.5 (178)	<0.001
Type of cancer			<0.001
Breast	31.5 (51)	38.8 (116)	
Ovarian	1.9 (3)	16.7 (50)	
Breast and ovarian	1.2 (2)	1.7 (5)	
Other	1.9 (3)	2.3 (7)	
None	63.5 (103)	40.5 (121)	
Family history of cancer			0.35
Breast and/or ovarian	87.7 (142)	82.5 (246)	
Other	3.7 (6)	4.7 (14)	
None	8.6 (14)	12.8 (38)	
Children	54.3 (88)	68.2 (204)	0.003
Education			0.94
College	22.8 (37)	23.7 (71)	
Grade school	17.3 (28)	15.4 (46)	
University	54.9 (89)	56.5 (169)	
Unknown	4.9 (8)	4.4 (13)	
BRCA testing			
Eligible	33.3 (54)	59.2 (177)	<0.001
Results			0.05
Declined[a]	3.7 (2)	1.7 (3)	
BRCA1/2 Positive	1.9 (1)	11.0 (19)	
Negative/Variant	98.1 (51)	89.0 (153)	

Missing data is attributed to patient non-response

[a] Two eligible responders who were not tested due to recent blood transfusions were not included in this calculation

Survey Responses

Knowledge and Interest in Genetics Appointment

Participant responses to survey questions are summarized in Table 2. With respect to referral, a significant difference was noted between responders and non-responders ($p<0.001$). While the majority of participants in both groups reported their physician recommended the referral, fewer responders reported that they were unaware that a referral had been made. Responders were also more likely to state that they were aware of the reason for their appointment ($p=0.02$) and that their physician had discussed the reason for the appointment with them ($p<0.001$). A greater proportion of responders than non-responders reported that they were highly interested in the appointment ($p=0.03$). After receiving the FHQ, responders

Table 2 Survey responses

Question	Non-responders % (N) *N*=162	Responders % (N) *N*=299	p-value
Knowledge and interest in genetics appointment			
Reason for referral			<0.001
Asked Doctor	22.2 (36)	21.5 (64)	
Doctor requisition	66.1 (107)	74.2 (221)	
Other	3.7 (6)	4.0 (12)	
Not aware	8.0 (13)	0.3 (1)	
Aware of reason for appointment			0.02
No	23.5 (38)	14.8 (44)	
Yes	76.5 (124)	85.2 (254)	
Doctor discussed the reason for appointment			<0.001
No	39.6 (63)	19.5 (58)	
Yes	60.4 (96)	80.5 (239)	
Initial interest level for appointment			0.03
Very interested	58.0 (94)	71.8 (214)	
Somewhat interested	30.3 (49)	20.8 (62)	
Neutral	11.1 (18)	7.1 (21)	
Uninterested	0.6 (1)	0.3 (1)	
Change in interest level after receiving FHQ[a]	*N*=106[a]		<0.001
Decrease	12.9 (13)	4.7 (14)	
Same	74.2 (75)	64.4 (192)	
Increase	12.9 (13)	30.9 (92)	
Difficulty getting to medical appointments			0.21
No	77.8 (126)	82.6 (246)	
Yes	22.2 (36)	17.4 (52)	
Use of the family history questionnaire			
Reasons FHQ was difficult/incomplete[b]	*N*=89[b]	*N*=152[b]	
Family is very large	25.0 (22)	47.4 (72)	<0.001
It was too long/took too long to complete	26.7 (24)	19.7 (30)	0.21
English was not good enough	3.3 (3)	4.6 (7)	0.64
No family history of cancer	1.1 (1)	3.3 (5)	0.29
Do not know family history	36.0 (32)	47.4 (72)	0.08
Am not in contact with relatives	31.5 (28)	51.3 (78)	0.003
Did not want to ask relatives	17.9 (16)	18.4 (28)	0.93
Adopted	2.3 (2)	0.65 (1)	0.55
Relative/personally in treatment	7.9 (7)	3.9 (6)	0.19
Afraid	6.7 (6)	3.3 (5)	0.21
FHQ was confusing	4.5 (4)	5.9 (9)	0.64
Other	11.2 (10)	2.6 (4)	0.006
Understood the purpose of questions in the FHQ[a]	*N*=106[a]		0.08
No	6.9 (7)	2.8 (8)	
Yes	93.1 (95)	97.2 (275)	
FHQ Instructions were clear[a]	*N*=106[a]		0.003
No	19.8 (20)	8.6 (24)	
Yes	80.2 (81)	91.4 (253)	
FHQ was too long[a]	*N*=106[a]		0.003
No	49.0 (49)	65.9 (182)	
Yes	51.0 (51)	34.1 (94)	
Information was too personal to provide[a]	*N*=106[a]		0.32
No	91.9 (91)	94.7 (266)	
Yes	8.1 (8)	5.3 (15)	
Preferred method to providing family history[a]	N=106[a]		<0.001
By questionnaire	31.3 (30)	77.9 (218)	
By phone	11.5 (11)	3.6 (10)	
In person	57.3 (55)	18.6 (52)	

Missing data is attributed to patient non-response

[a] Reduced sample size as these questions were not asked to non-responders who did not receive the FHQ

[b] Reduced sample size as this question was not asked to participants if they did not receive or did not report difficulty completing the FHQ

were more likely to report an increase in interest whereas non-responders were equally likely to report either an increase or a decrease in interest ($p<0.001$). Most patients did not report having difficulty attending medical appointments ($p=0.21$).

Use of the Family History Questionnaire

The second half of the survey was designed to gather potential barriers to completing the FHQ. Of the 299 responders, three did not indicate whether or not they had difficulty completing the FHQ and were excluded from this analysis. Of the total 162 non-responders, 56 stated that they did not receive the FHQ and were also excluded from the remaining analysis. Among the responders, 51.4 % (152/296) reported having difficulty completing the FHQ as compared to 56.4 % (22/39) of non-responders who reported having completed it. In contrast, 48.6 % (144/296) of responders reported no difficulty completing the FHQ as compared to 43.6 % (17/39) of non-responders. In addition to the 22 non-responders who completed the FHQ and reported difficulty with it, 28 non-responders who reported that they did not start the FHQ and 39 non-responders who reported that they had started the FHQ but not finish it, were also surveyed regarding barriers to not having completed it.

As shown in Table 2 as well as Fig. 1a and b, the primary reasons for which participants reported difficulty with the FHQ or their reasons for not starting or completing it were related to information about their family history. Specifically,

51.3 % of responders and 31.5 % of non-responders reported limited contact with relatives, 47.4 % of responders and 25.0 % of non-responders reported that their family was too large, and 47.4 % of responders and 36.0 % of non-responders did not know their family history. Many responders and non-responders stated that they did not want to ask their relatives for information (18.4 %; 17.9 %). A proportion of responders (19.7 %) and non-responders (26.7 %) also stated that the reason for having difficulty with or not completing the FHQ was related to its length. Significantly fewer responders (2.6 %) than non-responders (11.2 %) provided additional reasons for difficulty, which included concerns with privacy, procrastination or being busy, losing the FHQ, and concerns with the format of the FHQ ($p=0.006$). The 67 non-responders who did not finish or start the FHQ were also asked about additional barriers (Fig. 1b). The most common barriers were reported as forgetting about the FHQ (18.0 %) or not having time to complete it (11.9 %). Additional reasons included emotional difficulty completing the FHQ (9.0 %), feeling that the appointment was not needed (6.0 %), or not wanting an appointment (3.0 %).

When surveyed about their impressions of the FHQ, the majority of responders (97.2 %) and non-responders (93.1 %) reported that they understood why they were being asked about their family history. While most participants reported that the FHQ instructions were clear, a higher proportion of non-responders reported that these instructions were unclear (19.8 vs 8.6 %; $p=0.003$). Non-responders were also more likely to report that the FHQ was too long ($p=0.003$). Study participants were asked how they would prefer to provide their family history. Participants tended to indicate a preference for the method that they used during the study, with majority of responders preferring a questionnaire and majority of non-responders preferring an in-person appointment ($p<0.001$).

Predictors of Having Difficulty or Not Completing the FHQ

In order to identify variables associated with not starting, not finishing, or having difficulty completing the FHQ, univariable Chi-square tests or T-tests and multivariable logistic regression analyses were conducted. As previously described, 144 responders and 17 non-responders reported no difficulty completing the FHQ while 153 responders reported some difficulty and 89 non-responders reported either some difficulty or did not start/finish the FHQ. As shown in Tables 3 and 4, several differences were noted between these groups. A higher proportion of responders reported no difficulty completing the FHQ compared with non-responders ($p<0.001$). Study participants with a personal diagnosis of cancer were less likely to report difficulty in completing the FHQ ($p=0.02$), which was confirmed by multivariable analysis (OR=1.7, $p=0.02$). Participants whose physician discussed the reason for the genetic counseling appointment with them were almost twice as likely to report no difficulty completing the FHQ (OR 1.8 p=0.02). There was a significant difference among reported changes in interest level following receipt of the FHQ ($p<0.001$). This was particularly evident on multivariable analysis as participants who reported difficulty completing, or who did not complete the FHQ, were 13.4 times more likely to report a decrease in interest level ($p=0.01$) and less likely to report an increase in interest level (OR=0.46, $p=0.001$). Similarly, participants who stated that the FHQ instructions were unclear were almost three times more likely to report difficulty completing the questionnaire ($p=0.004$; OR=2.8, $p=0.01$). Participants who reported having difficulty attending medical appointments or who found the FHQ was too long were also more likely to report difficulty completing the FHQ ($p=0.04$ and $p<0.001$, respectively).

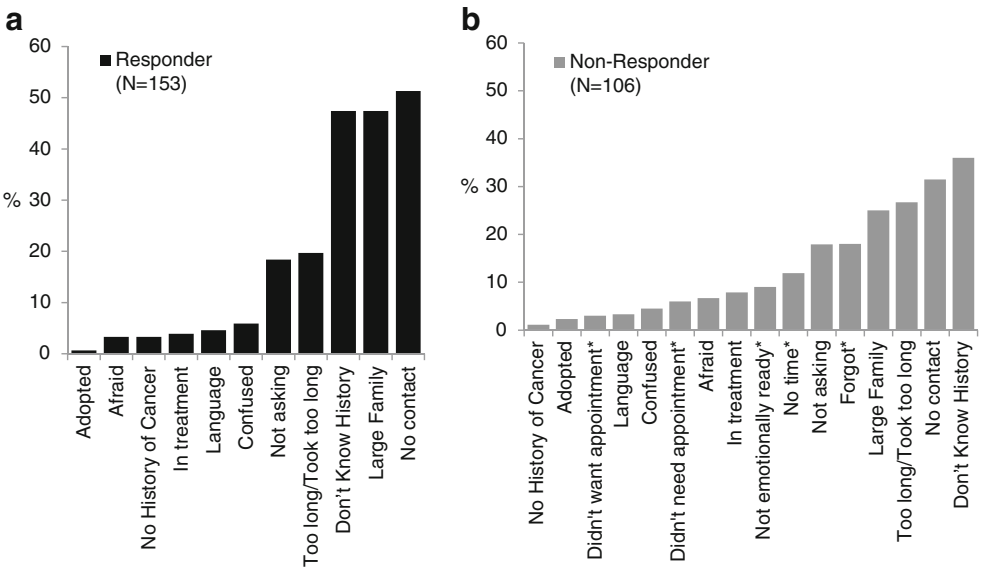

Fig. 1 Reasons the family history questionnaire was incomplete or difficult to complete **a** Percentage of responders who indicated specific barriers to completing the FHQ **b** Percentage of non-responders who indicated specific barriers to completing the FHQ. *Option only available to non-responders who did not finish or did not start the FHQ ($N=67$)

Table 3 Univariable analysis of factors associated with difficulty answering or completing the family history questionnaire

Variable	Difficulty answering or did not complete the FHQ		
	No Median (range) or % (N) *N*=161	Yes Median (range) or % (N) *N*=241	p-value
Group			<0.001
Responders	89.4 (144)	63.1 (152)	
Non-responders	10.6 (17)	36.9 (89)	
Age	47 (18–82)	46 (22–79)	0.17
Personal history of cancer	59.6 (96)	48.1 (116)	0.02
Doctor discussed the reason for appointment			0.01
Yes	82.6 (133)	71.4 (170)	
No	17.4 (28)	28.6 (68)	
Change in interest level after receiving FHQ			<0.001
Decrease	1.2 (2)	10.2 (24)	
Same	63.9 (103)	69.8 (164)	
Increase	34.8 (56)	20.0 (47)	
Difficulty getting to medical appointments			0.04
Yes	14.3 (23)	22.5 (54)	
No	85.7 (138)	77.5 (186)	
FHQ instructions were clear			0.004
Yes	94.2 (145)	84.6 (187)	
No	5.8 (9)	15.4 (34)	
FHQ was too long			<0.001
Yes	28.4 (44)	45.4 (99)	
No	71.6 (111)	54.6 (119)	

Analysis does not include non-responders who had not received the FHQ (*N*=56) nor responders who did not indicate whether they had difficulty completing the FHQ (*N*=3). Missing data is attributed to non-response

Discussion

FHQ Return Rates/ Genetic Counseling Attendance Rates

A number of studies have demonstrated that response rates to mailed cancer family history questionnaires are low, typically around 50 % or less (Appleby-Tagoe et al. 2012; Armel et al. 2011; Kohut et al. 2012; Mancuso et al. 2004; Rahm et al. 2007).

Table 4 Multivariable analysis of factors associated with difficulty answering or completing the family history questionnaire

Variable	Odds ratio	95% CI	p-value
Personal history of Cancer			
No vs. Yes	1.7	1.1–2.6	0.02
Doctor discussed the reason for appointment			
No vs. Yes	1.8	1.1–3.2	0.02
Change in interest level after receiving FHQ			
Decrease vs. same	13.4	1.8–102	0.01
Increase vs. same	0.46	0.28–0.74	0.001
FHQ instructions were clear			
No vs. Yes	2.8	1.2–6.2	0.01

Analysis does not include non-responders who had not received the FHQ (*N*=56) and responders who did not indicate whether they had difficulty completing the FHQ (*N*=3). Missing data is attributed to non-response

In order to improve response rates and ultimately genetic counseling attendance rates, some have suggested the use of telephone prompts or a patient navigator (Appleby-Tagoe et al. 2012; Mancuso et al. 2004; Poplawski et al. 2009; Rahm et al. 2007). In the current study, a total of 612 patients had not returned their FHQ within 3 months of mailing from the clinic. Of these 612 patients, 12.4 % (76) returned their FHQ following prompt by an appointment letter. This response rate rose to 19.4 % (76/392) when patients who were deceased, whose family had already had genetic counseling, or whose referral was greater than 1 year old were excluded. When examining attendance rates, 67.3 % (264/392) of patients who had not returned their FHQ within 3 months of mailing attended a genetic counseling appointment following prompt by mailing of an appointment letter. This data demonstrates that while a mailed appointment letter may only increase response to the FHQ by 9.2 %, overall attendance for genetic counseling can be increased by 32 %. While it may be ideal to obtain the family history in advance of genetic counseling, it would undoubtedly be preferable to have a patient attend genetic counseling without a completed FHQ, than to not be offered an appointment at all. In one small study the use of a patient navigator was successful in increasing overall attendance rates (Rahm et al. 2007); however, the cost of implementing such a service may not be feasible for many, if not most, cancer genetics clinics. In contrast,

mailing an appointment letter requires minimal expense with a significant improvement in attendance rate, particularly in the non-responsive population of patients. Those who take the time to complete the FHQ are likely more invested in the referral process and therefore more likely to attend a genetic counseling appointment. In the current study, only 1.1 % (5/432) of individuals who completed their FHQ following mailing did not attend a genetic counseling appointment as compared to 32.7 % (128/392) of non-responders. Of note, 100 % of all non-responders who returned their FHQ following receipt of an appointment letter subsequently attended their genetic counseling appointment. Although little data exists regarding the use of telephone prompts following mailed FHQs, the use of a mailed appointment letter seems to be a highly effective method for increasing utilization of genetic counseling services. In order to compensate for a 33 % no-show rate for such patients, group counseling or overbooking could be considered. In this clinic's experience, by overbooking specific appointment times for these patients, this problem has been effectively addressed.

Study Participants

In comparing responders and non-responders, it is evident that responders are more likely to have children and a personal diagnosis of cancer. Additionally, responders are more likely to be eligible for genetic testing and to have a *BRCA1* or *BRCA2* mutation. In contrast, no significant difference was seen between these two groups with respect to family history of breast or ovarian cancer. These results are consistent with previously published data demonstrating that while responders are more likely to be eligible for genetic testing, their familial genetic risk estimates are not significantly different from those of non-responders (Armel et al. 2009, 2011). These findings support the idea that non-responders are not from lower risk families, but rather may not be the most appropriate person in the family in whom to initiate genetic testing. As such, while offering genetic counseling to the non-responder population may not increase the numbers of patients offered genetic testing, it will lead to the identification of those family members in whom genetic testing should be initiated, and ultimately a population of patients with *BRCA1* or *BRCA2* mutations that would have otherwise been missed.

Barriers to Using FHQs

While it has been demonstrated that completion of a mailed cancer FHQ does not make patients more anxious or worried about cancer (Leggatt et al. 2000), minimal data exists exploring barriers to FHQs and ultimately why response rates are lower than desired. In the current study, both non-responders and responders were surveyed regarding the reasons that they had difficulty completing the FHQ, reasons for not finishing the FHQ, or not starting it. In total, 51.3 % of responders reported that they had some difficulty completing the FHQ. In comparison, 56.4 % of non-responders who claimed they had completed the FHQ reported difficulty completing it. Overall, in both the responder and non-responder groups the most commonly cited reasons for difficulty completing the FHQ were related to family dynamics; in particular, not having contact with their relatives, having a large family size, and not knowing their family history. In contrast, fear, confusion, and language barriers were less likely to be cited as barriers by both groups. For non-responders, not having time to complete the FHQ and forgetting to complete it were secondary to issues related to obtaining family history information. These results are consistent with previous data demonstrating that difficulty accessing family history information plays a significant role in non-response (Appleby-Tagoe et al. 2012; Armel et al. 2011). For responders, difficulty completing the FHQ was overwhelmingly related to the ability to obtain information regarding their family history, whereas for non-responders a number of other reasons, albeit less common, were also cited for non-response. Nevertheless, for both groups, lack of knowledge of family history information, whether due to family size, not knowing the family history, or not having access to relatives, played the most significant role in having difficulty completing the FHQ; however, responders were clearly more motivated to overcome the challenges related to obtaining their family history than non-responders.

In addition to exploring reasons why the FHQ was not completed or was difficult to complete, participants were also surveyed about usability of the FHQ. In a previous study, a number of factors were identified to help improve ease of use and accuracy of information provided on FHQs (Armel et al. 2009). In particular, a carefully worded and developed FHQ with clear instructions indicating the purpose of obtaining the family history may be the first step towards increasing response rates. In the current study, the majority of both responders and non-responders indicated that they understood the purpose of the questions in the FHQ and that they did not believe the information was too personal to provide. Similarly, the majority of participants in each group indicated that the instructions to complete the FHQ were clear and only a minority cited confusion with the FHQ as one of the reasons that they had difficulty or did not complete it. Regarding the length of the FHQ, 51.0 % of non-responders felt it was too long as compared to 34.1 % of responders. When specifically surveyed about whether the length of the FHQ contributed to the reason they had difficulty with the FHQ or did not complete it, only 26.7 % of non-responders and 19.4 % of responders stated that it was a hindrance. This would suggest that although a significant proportion of non-responders feel that the FHQ is too long, that this in itself is not a barrier to completing it.

When exploring factors that predict non-response or difficulty completing the FHQ, a number of significant variables

were identified. In both univariable and multivariable analysis, those individuals who reported difficulty completing the FHQ or did not complete it were more likely to cite that they were unaffected, that their doctor did not discuss the appointment with them, that their interest level decreased after receiving the FHQ, and that the instructions to complete it were not clear. This would suggest that in patients who are less likely to be motivated to complete the questionnaire, such as those unaffected with breast or ovarian cancer, having their doctor explain the importance of the referral and the nature of genetic counseling is critical. Finally, when asked how they would prefer to provide their family history if given the choice, 78 % of responders selected FHQ as compared to 31 % of non-responders. The majority of non-responders selected a preference to providing their family history in person, suggesting the existence of a confirmation bias. Overall, a total of 65.9 % of participants selected the FHQ as their preferred method to provide their family history. This is consistent with other data demonstrating that patients perceive FHQs as an acceptable method for providing family history information (Appleby-Tagoe et al. 2012; Armel et al. 2009; Chalmers et al. 2001).

Facilitators to Using FHQs

When surveyed regarding who had initiated their referral, the majority of both responders and non-responders indicated that their doctors had recommended it; however, non-responders were significantly more likely to say that they were unaware they had been referred. Similarly, responders were significantly more likely to state that they knew the reason for their appointment and that their doctor had discussed it with them. This data demonstrates the importance that physician guidance plays in the referral process. By having a discussion with patients about the purpose and relevance of genetic counseling, patients are able to make an informed decision about whether or not to complete the referral process and ultimately attend a genetic counseling appointment. To date, only a limited number of studies have looked at barriers and facilitators for the utilization of genetic counseling services; however, it is evident from the data available that patient and provider knowledge of the value of genetic counseling plays a significant role (Anderson et al. 2012; Rolnick et al. 2011; Sussner et al. 2013). In one study, a primary reason cited for not attending genetic counseling was that no one had recommended it (Anderson et al. 2012). In another, physician referral was a significant predictor of intention to undergo genetic counseling (Sussner et al. 2013). Clearly the answers to the survey questions in this study were based on patient recall; yet, these results suggest that patients whose physicians entered into a discussion regarding the purpose of genetic counseling were significantly more likely to complete their FHQ and ultimately attend their genetic counseling appointment. Not surprisingly, when a patient navigator was used to schedule genetic counseling appointments, the most common questions the navigator received were related to the process of genetic counseling (Rahm et al. 2007). This only further highlights the important role that healthcare providers play in the utilization of cancer genetic services. Unfortunately, lack of physician knowledge about or comfort with referring patients for genetic testing may be a significant barrier to this process (Mouchawar et al. 2001; Wideroff et al. 2003). Some physicians may also not be discussing or offering referrals due to fear of discrimination or negative emotional impact for their patients (Acton et al. 2000).

Results of the current study also demonstrate that interest level in genetic counseling is a significant predictor for completion of the FHQ. Responders were significantly more likely to report an interest in genetic counseling prior to receiving the FHQ and significantly more likely to report an increase in interest following receipt of the FHQ. Because a greater number of responders had children and a personal diagnosis of cancer, it is possible that these factors motivated them to complete their family history questionnaire. Nevertheless, because there was no significant difference in family history of cancer for both responders and non-responders, and that previous studies have demonstrated that risk estimates are no different between these two groups (Armel et al. 2011), one would anticipate that a genetic counseling appointment would be equally as valuable to each population.

Strengths and Limitations

Strengths of this study include its large sample size, the use of an anonymous survey rather than a telephone interview, and the fact that both the responder and non-responder populations were surveyed. Limitations of the study include the fact that patient responses were based on recall and as such it was not possible to know whether or not referring physicians had discussed referrals with their patients. Additionally, the population of patients surveyed was homogeneous and no data was available on the group of patients that did not attend their scheduled appointment. The group of responders who reported that they had no difficulty with the FHQ was also not surveyed about any potential barriers.

Conclusion and Practice Implications

With advances in technology and the availability of web-based FHQs that can feed directly into electronic databases, it is anticipated that the desire by cancer genetics clinics to use FHQs will only increase. As such, it is not only important to explore the ways in which technology can change the delivery of genetic counseling services, but also to better understand the barriers posed by using such tools. Results of this study demonstrate that difficulty completing the FHQ is not necessarily a barrier

inherent to its design but rather due to difficulty accessing one's family history. As a result, genetic counselors need to be aware that lack of knowledge of one's family history, whether due to large family size or lack of contact with relatives, can stall or inhibit the scheduling process and prevent some patients from ultimately being seen for genetic counseling. It is therefore important that this population not be ignored based on an assumption that non-response is an indicator of lack of interest. Additionally, this study has served to demonstrate that mailed appointment letters are a highly successful and cost-effective way to increase attendance rates in a population of patients that is non-responsive to a mailed FHQ. Lastly, and perhaps most importantly, the data demonstrate the valuable role that referring physicians play in the utilization of genetic counseling services. Educating both patients and providers about the value of this service is critical and can likely be achieved by offering educational events targeting physicians and patient friendly brochures addressing the purpose and benefits of genetic counseling. By increasing knowledge about and awareness of genetic counseling and the beneficial role it plays in identifying high-risk individuals, it is anticipated that this will lead to a reduction in size of the non-responder population and an overall increase in the utilization of genetic counseling services.

Conflict of Interest Author Susan Randall Armel, author Jeanna McCuaig, author Nicole Gojska, author Rochelle Demsky, author Manjula Maganti, author Joan Murphy, and author Barry Rosen declare that they have no conflicts 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 (5). 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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