Predictors of Self-Referral into a Cancer Genetics Registry

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Abstract

Recruitment is a challenging part of developing and maintaining any population-based disease registry and different methods are used to increase enrollment. However, recruitment methods may attract different subgroups of individuals, so examining characteristics of samples recruited using different methods can help detect threats to the external validity of research results.

Objectives: **There were two main objectives of this study: to examine differences between participants who were self-referred and those who were recruited into a cancer genetics registry and to identify predictors of self-referral status.**

Methods: **A cross-sectional survey was done with two groups of cancer genetics registry members (n = 268): (a) members recruited through self-referral and (b) members recruited through population-based sampling.**

Results: **There were no significant differences in demographic variables between the two samples except for education (higher in the self-referral group; $P < 0.01$). The self-referral group showed significantly higher levels of anxiety, depression, and cancer history and was more likely to report the strongest response to statements about cancer risk, screening intentions, and views on genetic testing. Logistic regression modeling indicated these predictors of self-referral status: previous cancer diagnosis, viewing self as a candidate for genetic testing, education higher than high school, and wanting assistance with personal future risk (R^2 = 0.41).**

Conclusions: **Our results suggest that whereas groups recruited via different strategies may seem to be the same based on demographic variables, underlying psychosocial variables differ between those self-referring and those recruited via population-based screening. To accurately estimate the generalizability of population-based studies or studies conducted using a cancer genetics registry, method of recruitment should be examined when interpreting and analyzing results.** (Cancer Epidemiol Biomarkers Prev 2007;16(7):1387–92)

Introduction

Sampling method is important in assessing the external validity of study results for populations beyond the one studied (1, 2). Recruiting for studies by inviting people to participate in research through media outreach such as a study website or print ads often takes relatively fewer resources and presumably attracts people interested in participating in research. As such, these convenience sampling methods can be less costly in recruiting research participants and so are understandably attractive. In contrast, population-based recruiting methods (e.g., random digit dialing or household area sampling) attempt to recruit a population that has fewer biases that come from the effects of volunteering. As such, these probability methods theoretically provide a sample that most closely resembles and ''represents'' the general population, making the results more easily generalizable, but at potentially high administrative costs and time investment (3).

Several investigators have found important differences in the data from samples from different recruitment methods, often stemming from whether the sample was recruited through invited self-referrals or population-based recruitment. For example, differences in demographic variables (2, 3) as well as differences in cancer history and cancer worry (4-6) have been documented in the literature. Results from these studies suggest that self-referred patients may have higher cancer worry, more family history of cancer, more interest in genetic testing, and higher perceived risk than population-based samples.

Differences in sample characteristics have also been associated with timing of the response (7), amount of effort taken to recruit (8, 9), and the method of population-based referral (1, 10). Several studies report evidence for a ''healthy volunteer effect'' that suggests that people with no underlying disease are more likely to agree to participate in a study when approached (4, 5). Others suggest evidence for the converse, a ''sick person effect'' wherein people with decreased health status are more likely to participate in research studies (6). Taken together, these studies show that it is reasonable to consider that recruitment method can result in groups of participants that are meaningfully different. In the context of our study, a research registry on cancer genetics with the goal of conducting research into cancer risk, participants at increased risk might not be easily classified as either healthy or sick according to the terminology above. However, we reasoned that people with more interest in cancer genetics and cancer risk would be more likely to join a study proactively (i.e., via self-recruit) than passively (approached via population-based recruitment). If so, then understanding the nature of the differences between the groups would enhance our understanding of the registry participants and inform study design for studies seeking to enroll registry members.

In this study, we explored demographic and background variables between individuals who were recruited into a cancer genetics registry in western Washington using two different recruitment methodologies. This study provides a unique opportunity to study recruitment method. These data stand to affect more than just the used sample in question because participants of this large population-based cancer genetics registry may be invited to participate in numerous studies over time. As we and other investigators draw on this registry for potential research participants, understanding if there are significant differences between the samples based on how they were recruited is essential. If there are no significant

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differences, we can use both self-referred and population-based samples to recruit for several studies. If differences exist, we can suggest the limits of external validity and may also learn characteristics of the self-referred sample that may be targets for intervention. We conducted a cross-sectional analysis of quality of life, demographic, and genetic risk variables in two samples of members of the Northwest Cancer Genetics Network (CGN). The first method sample was self-referred through response to media outreach, and the second through population-based sampling. The aims of the study were to describe differences between these two groups in the CGN and to model predictors of self-referral status to the registry. In accordance with previous studies, we hypothesized that self-referred members of the registry would be more ''engaged'' in participation in the registry than population-based participants, possibly because of more experience with cancer. Thus, we hypothesized that the self-referral sample would exhibit higher anxiety, depression, perceived cancer risk, and interest in genetics than the population-based sample. We also hypothesized that self-referred participants would be more likely to choose the strongest endorsement option (e.g., ''strongly agree'') for questions about genetics, risk, and cancer because of stronger opinions that led them to proactively seek to volunteer in the registry.

Materials and Methods

The Northwest Cancer Genetics Network (CGN) is a registry designed to act as an infrastructure for studies of cancer genetic susceptibility. It is part of a national network funded by the National Cancer Institute, NIH (11). Participants in the CGN are people with cancer identified by state and national cancer registries, first-degree relatives of cancer cases, people who self-referred to the CGN by responding to a media campaign, and controls recruited from a random population sample. This study includes only self-referrals to the CGN and population-based controls.

Self-recruited participants, those that saw an advertisement and responded if interested, were recruited as per instructions in the original request for application. These instructions identified people who were worried about their risk and needed to learn more about that risk as a target group for the larger study. Self-referrals were invited to participate in the CGN registry through media and health professional contacts. Through the associations of CGN with regional genetics professionals, the Cancer Information Service, and local media, members of the public were invited to join the network by calling the CGN telephone number or by submitting an online request through the CGN website. The public recruitment materials asked for individuals interested in participating in a study about familial risk of cancer to call a toll-free number or communicate via a study website. Recruitment materials inviting self-referrals described the benefits of participation in the registry as helping researchers and society learn more about genetics and cancer; being informed by the CGN staff about eligibility for special research studies; and receiving regular updates on research on cancer and genetics. No financial incentives were offered for participation.[4]

Sampling for the Current Study. This study measured psychosocial variables in CGN participants who were self-referrals or population-based controls. We developed a survey that asked participants to answer a battery of measures, including the SF12 version 2 and the Brief Symptom Inventory (BSI), as well as several measures of risk perception, views about genetics, and genetic contribution to disease. The survey also collected detailed information on the surgical history of all participants. The survey was administered by mail, and slow responders were followed-up via telephone and were given the option of completing the survey over the phone. We received 268 responses, 134 each of self-referred participants, and participants from the population-based control sample.

All analyses were conducted using SPSS v.13.0 (7). First, we conducted bivariate analyses on all demographic variables. Next, we examined differences in measures of anxiety and depression from the BSI (8) and the SF12 version 2 physical and mental composite scores (9). We also examined the two groups of participants in terms of several measures that we hypothesized would be different among the two groups: perception of risk of cancer, number of relatives with cancer, and previous genetic testing. The survey asked participants to rate many statements about screening behavior according to a Likert scale (e.g., ''You intend to examine yourself for cancer every month''). We hypothesized that self-referred individuals would be more likely to choose the superlative option to these questions (i.e., the strongly agree option) than the population-based group. To assess this hypothesis, we created a variable to count the number of strongest responses. For all bivariate analyses, we conducted means tests for continuous variables (age and number of relatives with cancer) and χ^2 tests for categorical variables. A forward, stepwise logistic regression model for multivariate analysis was constructed to identify variables that would predict self-referral status. Interviewers were trained to ensure that individuals with cognitive or other difficulty in answering the screening questions were excluded from the study.

Results

Demographics. Results of the bivariate analysis on demographic variables are reported in Table 1. On average, the sample was in the early 1950s, male, married, and White. Roughly half of each group reported smoking >100 cigarettes in their lifetime, and 11% of each group reported being a current smoker. Of the sociodemographic variables analyzed, only education was significantly different between the two groups. More members of the self-referred group reported some college (43%) or college or beyond (47%) than the population-based sample (37% and 39%, respectively).

Table 1. Demographics

	Population-based (n = 134), n (%)	Self-referral (n = 134), n (%)
Age (mean ± SD), y	52.8 ± 16.6	51.4 ± 14.4
Median (interquartile range)	54.5 (29)	53.0 (22)
18-29	13 (10)	11 (8)
30-39	16 (12)	21 (16)
40-49	28 (21)	25 (19)
50-59	21 (16)	33 (25)
60-69	26 (19)	27 (21)
70+	30 (22)	16 (12)
Female	49 (37)	54 (40)
Marital status		
Never married	20 (15)	19 (14)
Married	94 (70)	89 (66)
Formerly married	20 (15)	26 (19)
Education*		
High school/GED or less	33 (25)	13 (10)
Some college/technical school	49 (37)	58 (43)
College or beyond	52 (39)	63 (47)
White/Caucasian, non-Hispanic White, n (%)	116 (87)	122 (91)

*$P < 0.01$.

[4] See http://www.fhcrc.org/science/phs/cgn/for more information about the CGN.

Table 2. Current health and family history

	Population-based (n = 134), n (%)	Self-referral (n = 134), n (%)
Smoked at least 100 cigarettes in lifetime, n (%)	69 (51)	64 (48)
Current smoker, n (%)	15 (11)	16 (12)
Have at least one cancer diagnosis, n (%)*	19 (14)	53 (40)
Relatives with cancer, n (%)*		
None	18 (13)	7 (5)
1	34 (25)	11 (8.2)
2+	82 (61)	116 (86.6)
No. relatives with cancer (mean ± SD)*	2.4 ± 1.9	4.0 ± 2.8
Median (interquartile range)	2.0 (2)	3.0 (3.0)
Have had genetic test for any reason, n (%) †	4 (3)	11 (8.3)
Self is ''family health informant'' (n = 215), n (%)	15 (15)	18 (15.9)
Had hysterectomy, n (%) ‡	8 (16)	23 (42.6)
Had oophorectomy, n (%) ‡	2 (4.1)	15 (27.8)
SF12 version 2 PCS	49.6 ± 9.6	48.9 ± 11.1
SF12 version 2 MCS †	51.2 ± 9.6	48.7 ± 11.2
BSI anxiety (mean ± SD) ‡	0.34 ± 0.44	0.52 ± 0.62
BSI depression (mean ± D)§	0.41 ± 0.55	0.56 ± 0.66

Abbreviations: PCS, physical composite score; MCS, mental composite score.
*$P < 0.001$.
†$P < 0.1$.
‡$P < 0.01$.
§$P < 0.05$.

Health Variables. As shown in Table 2, we found significant differences in both personal and family cancer history between the self-referred and population-based groups. People in the self-referral group were more likely to have had cancer (40% versus 14%; $P < 0.001$). Although the majority of both groups reported family members with cancer, a greater majority of the self-referred group (87%) reported at least two family members with cancer compared with the population-based group (61%). The number of relatives with cancer also differed between the two groups, with those in the self-referral group reporting 3.97 on average compared with 2.43 for the population-based group. People in the self-referral group were also more likely to report having had a genetic test for any reason (8% versus 3%). Participants were asked to report the identity of their family's health informant, the person who they ''would feel comfortable communicating with other family members about health issues and who would know about health issues in the family.'' We hypothesized that self-referred participants would be more likely to report being the family health informant than population-based participants. However, there was no significant difference between the two groups and ~15% of participants in each group reported having this role.

Surgical History. Both hysterectomy and oophorectomy were reported at significantly higher rates in the self-referred group with 43% reporting a hysterectomy and 28% reporting an oophorectomy compared with 16% and 4% in the population-based group. We did not measure if these surgeries were prophylactic or not. Reported history of breast lump, colon polyps, ovarian cysts, polycystic ovaries, colectomy, thyroidectomy, mastectomy, prostatectomy, and transurethral resection of the prostate were also examined. However, there were no significant differences between the two groups with respect to these variables (results not shown).

Quality of Life. We calculated the composite score for anxiety and depression for the BSI and physical and mental composite scores for the SF-12 v.2. These scores are reported in Table 2. For the BSI measures, the self-referred group reported higher levels of both anxiety and depression (0.52 and 0.56, respectively; $P < 0.01$) compared with the population-based group (0.34 and 0.41). For the SF-12 v.2 composite measures, there were no significant differences between the groups in physical composite score (49.9 versus 48.9); the differences in the mental composite score were marginally significant at $P = 0.055$.

Views on Cancer Risk. Participants were asked to choose from a list the factors contributing to risk of cancer (Table 3). In keeping with our hypothesis that self-referred participants would be more likely to choose the strongly agree options, we report the proportion of each group selecting that option. Of 23 items, we found significant differences in four: ''genetics'' (44% of self-referral group chose strongly agree versus 25%; $P < 0.0001$), ''family history of colon cancer'' (43% versus 22%), ''your own body chemistry'' (36% versus 18%), and ''added chemicals in the air'' (24% versus 15%). Two items showed borderline significantly higher results in the self-referral group: ''added chemicals in food'' and ''eating too much'' ($P < 0.10$). Items that did not differ significantly between the two groups included chemicals in the drinking water, ground, and at work; alcohol; cigarettes; being overweight; power lines; radiation from the sun; stress at home and work; high-fat diet; too many fruits and vegetables; personality; contraceptives; hormone replacements; bad sleeping patterns; and plastic products (results not shown).

Cancer Screening. As shown in Table 3, the majority of the self-referral group (69%) reported ''definitely'' planning to have cancer screening, compared with half of the population-based group (51%; $P < 0.01$). People in the self-referral group were more likely to indicate strongly agree to intentions to follow routine screening recommendations (50%), to screen more frequently than routine recommendations advise (16% compared with 5%), and to screen themselves for cancer every month (23% compared with 10%). Self-referrals were also more likely to report that they talk with their health care provider ''a lot.''

Views on Genetic Testing. Results of items asking about interest in genetic testing are reported in Table 3. There were significant differences between the two groups on several of these items. More participants in the self-referral group reported that they are definitely a candidate for genetic testing (40%; $P < 0.05$). More participants in the self-referral group reported having read a lot about genetic testing (6% versus 2%). Approximately half (49%) of the self-referral group reported definitely being interested in genetic testing, and 62% reported being interested if the test were covered by insurance. Far fewer in both groups indicated that they remained interested even if they had to pay for a genetic test themselves. However, there was a significant different between the two groups with more people in the self-reported

Table 3. Interest in genetic testing and cancer screening

	Population-based (n = 134), n (%)	Self-referral (n = 134), n (%)
Increases your risk for cancer very much		
Genetics*	31 (25)	58 (44)
Family history of colorectal cancer*	28 (22)	56 (43)
Your own body chemistry*	23 (18)	47 (36)
Added chemicals in the air †	19 (15)	32 (24)
Added chemicals in food ‡	20 (15)	33 (25)
Eating too much ‡	14 (11)	25 (19)
Are a candidate for genetic testing, definitely yes*	10 (8)	52 (40)
Interested in genetic test, definitely yes*	19 (15)	64 (49)
Interested if insurance covers cost, definitely yes*	32 (25)	80 (61)
Interested if you had to pay for it yourself †	3 (2)	13 (10)
Know a lot about own cancer risk*	15 (11)	39 (30)
Read or heard a lot about genetic testing for cancer risk ‡	2 (2)	8 (6)
Read or heard a lot about your cancer risk*	19 (14)	43 (32)
Definitely planning to have cancer screening§	68 (51)	91 (69)
Intend to screen self for cancer every month, strongly agree§	13 (10)	31 (23)
Intend to follow the routine recommendations for cancer screening for people your age, strongly agree*	31 (24)	57 (43)
Intend to have more frequent screening than is routinely recommended for people your age, strongly agree§	6 (5)	21 (16)

*$P < 0.001$.
†$P < 0.05$.
‡$P < 0.1$.
§$P < 0.01$.

group indicating interest (10% in the self-referral group compared with 2% in the population-based group).

Superlative Answers to Questions. One hypothesis of our study was that people in the self-referral group would be more likely to choose responses that were the superlative option of a scale question (e.g., strongly agree, definitely yes). To assess this variable, we counted the total number of these responses to 58 questions on risk, screening intentions, and education/assistance needs in each group. The result was highly significant at $P < 0.001$, with a mean of 9.4 superlative responses in the population-based group and 15.3 in the self-referral group.

Education/Assistance Needs. The survey contained several questions about the participants' perceived assistance and educational needs. As shown in Table 4, self-referrals were more likely to indicate wanting ''very much'' assistance on personal risk, screening tests, coping with feelings about cancer, learning about cancer risk factors, learning about cancer treatment, coping with feelings, helping others with cancer, and talking to health care providers.

For educational needs, self-referrals were more likely to indicate wanting very much education about cancer through phone calls with providers, television shows, audiotapes, videotapes, individual sessions, magazines, the internet, and written pamphlets (Table 4). There were no significant differences between the two groups with respect to their interest in receiving education from a ''group of people like you'' or during visits with health care providers.

The survey asked respondents their views about their health care providers. There were not significant differences between

Table 4. Education needs

	Population-based (n = 134), n (%)	Self-referral (n = 134), n (%)
Would like very much assistance with...		
...changing behavior*	34 (26.0)	63 (47.4)
...getting information on general risk factors*	21 (15.9)	45 (34.1)
...learning about cancer treatment*	15 (11.7)	43 (32.6)
...personal risk*	25 (19.2)	67 (50.4)
...screening tests*	22 (17.1)	57 (42.9)
...coping with feelings about cancer †	13 (9.9)	31 (23.7)
...helping others ‡	26 (20.2)	43 (33.3)
...talking to health care provider ‡	21 (16.0)	36 (27.3)
...hearing others' experiences§	13 (10.0)	23 (17.42)
Would like education about cancer very much...		
...in written health pamphlets*	23 (17.6)	51 (38.9)
...during instructional television shows †	13 (10.2)	31 (23.8)
...from videotapes †	10 (7.7)	25 (18.9)
...phone call with a health care professional ‡	10 (7.6)	22 (16.9)
...from audiotapes ‡	6 (4.7)	16 (12.2)
...in an individual session ‡	24 (18.6)	41 (31.3)
...through a telephone help line ‡	5 (3.8)	15 (11.5)
...over the internet ‡	23 (17.8)	37 (28.9)
...in a visit with health care provider§	29 (22.1)	42 (31.8)
How much do you talk to your health care provider about risk for cancer (a lot) †	9 (6.8)	25 (19.1)
Total no. strongest response to 58 questions*	9.4 ± 8.4	15.3 ± 9.2

*$P < 0.001$.
†$P < 0.01$.
‡$P < 0.05$.
§$P < 0.1$.

the two groups in expressed attitudes about satisfaction with their health care providers (not shown). However, the self-referral group was more likely to report talking a lot with their provider about cancer risk ($P < 0.01$; Table 4).

Results of Logistic Regression Model. We conducted a forward stepwise logistic regression model to identify predictors of self-referral status to the CGN, entering all variables that had shown statistical significance at the 0.25 level in the bivariate analyses. Although most of the demographic variables were not statistically significant in the bivariate analysis, we forced them to remain in the regression analysis to not miss any multivariate effect of these variables.

The final model included 254 (95%) of the original 268 participants. Besides demographic variables, four additional variables remained in the final model: having had a cancer diagnosis, considering oneself definitely a candidate for genetic testing, wanting assistance with personal cancer risk very much, and having attained education greater than high school. Of these, the biggest contributor was a previous diagnosis of cancer (odds ratio, 7.2; 95% confidence interval, 3.2-15.8) and considering oneself a candidate for genetic testing (odds ratio, 5.9; 95% confidence interval, 2.5-13.9). The results are shown in Table 5; the R^2 for the final model was 0.41.

Hysterectomy and oophorectomy were reported significantly more often in the self-referral group. Because these are gender-specific variables, we did not include them in the main model. To examine whether these were predictors of self-referral, we conducted a subanalysis of women only that included both variables. History of hysterectomy emerged as a predictor of self-referral status (odds ratio, 6.51; 95% confidence interval, 1.80-23.51, not shown), as did lower anxiety, previous cancer diagnosis, and definitely viewing oneself as candidate for genetic test. Overall, this analysis is less robust because it includes a smaller subset ($n = 85$) of the whole sample and should be considered hypothesis-generating.

Discussion

We conducted a cross-sectional analysis of quality of life, demographic, and genetic risk variables in two samples of members of the CGN recruited in different ways: self-referral or population-based recruitment. Our findings are consistent with other studies that have shown differences between samples depending on recruitment method and add a new understanding of potential predictors of volunteerism for research studies that add to our understanding of both generalizability of research results and potential education and assistance needs of study volunteers. Interestingly, the discrepancies between recruitment groups were not obvious by focusing only on traditionally measured demographic variables; in fact, our results indicated that, demographically, the two groups were quite similar. Education was the only demographic variable that was significantly different between the two groups, but both groups possessed a higher education than the general population as indicated by the U.S. Census (10).

Consistent with our hypotheses, the self-referral sample was more likely to report a personal history of cancer and greater numbers of relatives with cancer; in fact, cancer diagnosis was the single biggest predictor of self-referral status. We could not determine from these data if cancer survivors are proactively seeking enrollment into a cancer registry because of a sense of unmet needs from health providers, a heightened sense of altruism, or other factors. Likewise, we did not measure the participation of cancer survivors in cancer support groups, individual counseling, or other support. The questions we asked only offer a sense of perceived needs for education and assistance around cancer risk. Whereas the needs were higher in the self-referral group, our results indicated no difference in the two groups in overall satisfaction with health care providers. This may reflect the trend that most people tend to be satisfied with their providers, but it also does not suggest that health care satisfaction is correlated with recruitment method.

Our results showed statistically significant differences in the two groups in depression, anxiety, and SF-12 mental composite score, suggesting that the self-referral sample was experiencing lower quality of life as measured by these variables. Although different in the two samples, these results were well within the normative range reported in nonpsychiatric patients in validation studies of the BSI instrument and in general population scores of the SF-12 version 2 for both groups (8, 10). We did not explicitly screen for major psychiatric disturbance pre-enrollment so only future study could rule it out, but it is unlikely that the differences we observed were related to such conditions. In multivariate analyses, quality of life variables fell away as predictors of self-referral status, suggesting that difference in quality of life between the two groups is explained by the other predictors of self-referral status: personal cancer history, considering self a candidate for genetic testing, wanting assistance with personal risk, and higher education.

The self-referral group was more highly motivated for cancer screening behaviors, specifically in intentions to follow cancer screening recommendations, previous knowledge, genetic testing, and perception of self as a candidate for genetic testing. Self-referred members were also more likely to choose the strongest response (e.g., strongly agree, definitely yes) to a series of questions about interest in genetic testing, environmental contributions to cancer risk, and needs for assistance and education about cancer risk. Because self-referred participants indicated a consistently higher and stronger interest in education and assistance about cancer risk than the population-based group, this might indicate an unmet need for support around cancer risk in the self-referral sample, perhaps especially in the large proportion of cancer survivors in that group.

Our study suggests that there is reason to examine effect of recruitment method to more accurately estimate the threats to external validity of study results. In this particular registry, an

Table 5. Multivariate model

	Odds ratio (95% confidence interval)
Age (18-29 is reference), y	
30-39	1.62 (0.40-6.54)
40-49	1.07 (0.28-4.17)
50-59	1.43 (0.36-5.72)
60-69	0.84 (0.20-3.58)
70+	0.48 (0.10-2.23)
Female	1.36 (0.69-2.69)
Marital status (never married is reference)	
Formerly married	0.75 (0.22-2.56)
Married	0.72 (0.26-1.97)
White, non-Hispanic White	2.21 (0.80-6.13)
Had at least one cancer diagnosis	7.02 (3.20-15.82)
Thinks self is candidate for genetic test	5.94 (2.54-13.86)
Wants assistance with personal future risk	3.38 (1.75-6.55)
Education more than high school	2.52 (1.08-5.90)
Total used: 254 (94.8%)	
Max-rescaled R^2	0.41
Prediction ability, %	
Population	73.4
Self-referral	75.4
Area under ROC	0.82
Hosmer & Lemeshow goodness of fit test (8 degrees of freedom)	
χ^2	5.56
$P > \chi^2$	0.70

infrastructure for several types of studies, the degree to which external validity is in question may depend on the type of study using the CGN as a basis for recruitment. In studies of genetic susceptibility to cancer that only involve the contribution of genetic material, bias may be limited to the differential personal and family histories of cancer in the two groups; this bias may be controllable. However, studies designed to test psychosocial or educational interventions may face more difficulty in generalizing study results to a larger population if using a self-referred sample. In our study, high interest in genetic risk is to some degree expected in self-referrals because, presumably, people interested in the topic would be most motivated to seek enrollment. However, the consistently higher levels of interest in support and education compared with the population-based group remains interesting, not least because the CGN enrollment process involves a fairly time-intensive enrollment process that involves an exhaustive family history. It would not have been a surprise to see high levels of interest in genetic cancer risk in those consenting to join the population-based group as well.

In intervention research, recruiting self-referrals might help to target people who need it the most, and issues of external validity to the general population may not have the highest priority. In our study, it was an aim of the larger network to identify a target group interested in learning about genetic cancer risk; thus, logically, the next step is to identify interventions that may further serve this group. Our study suggests that our self-referred sample is disproportionately made up of cancer survivors and people with family history of cancer and is interested in genetic testing and education, assistance, or counseling about cancer risk. Self-referred participants may have higher anxiety, depression, and lower general mental functioning, possibly driven by personal cancer history and perceptions of the importance of genetics and genetic testing. Interventions aimed at reducing anxiety around genetic risk for cancer through education may help to support this group.

We acknowledge several limitations in the present study. First, this was a cross-sectional study; we cannot assume causality and can only estimate correlation between our independent variables and recruitment status. Prospective research is needed to test a conceptual model of predictors of self-referral status. Second, some differences are to be expected in the self-referral group because the original CGN promotional materials targeted people who were interested in genetics and cancer screening. However, we did not ask self-referrals their reasons for participation, and therefore we cannot speak to the specific effects of our promotional materials. Third, we did not measure pre-enrollment psychiatric conditions, participation in cancer support groups, individual counseling, or other psychological support; thus, we can only guess at the true psychosocial implications for the large proportion of cancer survivors in the self-referral sample. Fourth, this was a heavily Caucasian sample; the demographic makeup of the sample alone highlights the need for targeted minority recruitment so that we may further know how and if our efforts can best serve multiple cultural groups.

These results contribute to the growing body of research that examines characteristics of samples recruited to research studies by different methods. They suggest reason to look beyond traditionally measured demographic variables to assess similarities or differences in samples according to recruitment method. Although there may be limits to the external validity of studies that involve self-referred participants, inviting self-referrals may identify a group of people who are disproportionately cancer survivors and who are actively seeking information and support about genetic risk of cancer. Future studies that examine further the support available to cancer survivors may suggest potential interventions to provide needed support and education or identify unmet needs regarding continuing care and education about genetic cancer risk.

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