


# Challenges and Opportunities in Engaging Primary Care Providers in *BRCA* Testing: Results from the BFOR Study



Lydia E. Pace<sup>1,2</sup> , Nadine Tung<sup>3</sup>, Yeonsoo S. Lee<sup>4</sup>, Jada G. Hamilton<sup>5</sup>, Camila Gabriel<sup>6</sup>, Anna Revette<sup>6</sup>, Sahitya Raja<sup>7</sup>, Colby Jenkins<sup>5</sup>, Anthony Braswell<sup>8</sup>, Kelly Morgan<sup>5</sup>, Jeffrey Levin<sup>5</sup>, Jeremy Block<sup>5</sup>, Susan M. Domchek<sup>9</sup>, Katherine Nathanson<sup>9</sup>, Heather Symecko<sup>9</sup>, Kelsey Spielman<sup>9</sup>, Beth Karlan<sup>8</sup>, Daniella Kamard<sup>8</sup>, Jenny Lester<sup>8</sup>, Kenneth Offit<sup>5</sup>, Judy E. Garber<sup>6</sup>, and Nancy L. Keating<sup>1,2</sup>

<sup>1</sup>Brigham and Women's Hospital, Boston, MA, USA; <sup>2</sup>Harvard Medical School, Boston, MA, USA; <sup>3</sup>Beth Israel Deaconess Medical Center, Boston, MA, USA; <sup>4</sup>Mayo Clinic Alix School of Medicine, Scottsdale, AZ, USA; <sup>5</sup>Memorial Sloan Kettering Cancer Center, New York, NY, USA; <sup>6</sup>Dana-Farber Cancer Institute, Boston, MA, USA; <sup>7</sup>Rush Medical College at Rush University, Chicago, IL, USA; <sup>8</sup>David Geffen School of Medicine at UCLA, Los Angeles, CA, USA; <sup>9</sup>Basser Center for BRCA, Perelman School of Medicine, University of Pennsylvania, Philadelphia, PA, USA.

**PURPOSE:** Engaging primary care providers (PCPs) in *BRCA1/2* testing and results disclosure would increase testing access. The *BRCA* Founder OutReach (BFOR) study is a prospective study of *BRCA1/2* founder mutation screening among individuals of Ashkenazi Jewish descent that sought to involve participants' PCPs in results disclosure. We used quantitative and qualitative methods to evaluate PCPs' perspectives, knowledge, and experience disclosing results in BFOR.

**METHODS:** Among PCPs nominated by BFOR participants to disclose *BRCA1/2* results, we assessed the proportion agreeing to disclose. To examine PCP's perspectives, knowledge, and willingness to disclose results, we surveyed 501 nominated PCPs. To examine PCPs' experiences disclosing results in BFOR, we surveyed 101 PCPs and conducted 10 semi-structured interviews.

**RESULTS:** In the BFOR study overall, PCPs agreed to disclose their patient's results 40.5% of the time. Two hundred thirty-four PCPs (46.7%) responded to the initial survey. Responding PCPs were more likely to agree to disclose patients' results than non-responders (57.3% vs. 28.6%,  $p < 0.001$ ). Among all respondents, most felt very (19.7%) or somewhat (39.1%) qualified to share results. Among PCPs declining to disclose, insufficient knowledge was the most common reason. In multivariable logistic regression, feeling qualified was the only variable significantly associated with agreeing to disclose results (OR 6.53, 95% CI 3.31, 12.88). In post-disclosure surveys (response rate=55%), PCPs reported largely positive experiences. Interview findings suggested that although PCPs valued the study-provided educational materials, they desired better integration of results and decision support into workflows.

**CONCLUSION:** Barriers exist to incorporating *BRCA1/2* testing into primary care. Most PCPs declined to disclose their patients' BFOR results, although survey respondents were motivated and had positive disclosure experiences. PCP training and integrated decision support could be beneficial.

**TRIAL REGISTRATION:** [ClinicalTrials.gov](https://clinicaltrials.gov/ct2/show/study/NCT03351803) (NCT03351803), November 24, 2017

J Gen Intern Med 37(8):1862–9  
DOI: 10.1007/s11606-021-06970-8  
© Society of General Internal Medicine 2021

## INTRODUCTION

*BRCA1* and *BRCA2* mutations are the most common cause of hereditary breast and ovarian cancers. Detecting these mutations before a cancer diagnosis provides opportunities for prevention or enhanced screening.<sup>1</sup> Thus, screening appropriate populations for *BRCA1/2* mutations could have substantial clinical and public health impact. To realize the benefit of testing, some experts have advocated for broader screening of high-risk groups (such as individuals with Ashkenazi Jewish (AJ) heritage, who have a ten-fold increased risk of carrying *BRCA1/2* mutations compared with the general population) or screening the general population.<sup>2</sup> Furthermore, in 2019, the United States Preventive Services Task Force (USPSTF) recommended that in addition to family cancer history, clinicians consider ancestry (and specifically, the presence of AJ heritage) as part of their assessment of an individual's risk of carrying *BRCA1/2* mutations.<sup>3</sup> However, how best to operationalize expanded genetic risk assessment and testing in routine practice is uncertain.<sup>4,5</sup> Although testing for *BRCA1/2* mutations has increased,<sup>6–9</sup> there remains undertesting of high-risk individuals in the USA. In addition, racial/ethnic and socioeconomic disparities in testing persist.<sup>5,10</sup> With limited access to genetic counselors and specialists in many areas, reducing existing disparities and further expanding testing will require engaging primary care providers (PCPs) to identify and counsel appropriate patients, interpret and disclose results, and determine initial management.

Previous research investigating PCPs' knowledge and perceptions about genetic medicine has identified gaps in

Prior presentation: Poster presentation at the Society of General Internal Medicine Annual Meeting; May 8–11, 2019 Received March 8, 2021

Accepted June 4, 2021

Published online June 25, 2021

knowledge and mixed attitudes about engagement in genetic medicine.<sup>11</sup> However, few prospective investigations have examined the feasibility of engaging PCPs in genetic testing and results disclosure. One study of whole genome sequencing found that patients infrequently discussed results with their PCP, and had mixed experiences doing so.<sup>12</sup> However, other studies demonstrated that PCPs could effectively counsel patients about whole genome sequencing and use this information in clinical decisions.<sup>13</sup> While disclosure of *BRCA1/2* results is more straightforward than whole genome sequencing, PCPs disclosing positive *BRCA1/2* results need to refer patients to appropriate specialists to discuss options for prevention, surveillance, and testing of family members. Disclosing negative results requires identifying need for further testing, for example, when family or personal history suggests the possible presence of a variant not assessed by the test.

The *BRCA* Founder OutReach (BFOR) study is a pilot prospective study of population *BRCA1/2* testing that offered *BRCA1/2* screening to individuals of AJ heritage in four US cities.<sup>14–16</sup> Interested participants enrolled online and received no-cost testing for the three founder mutations that account for 90% of *BRCA1/2* mutations in individuals of AJ heritage.<sup>17–19</sup> In contrast to commercial direct-to-consumer testing models, BFOR participants received online video education before testing and then selected whether they would like to have their results disclosed either by their PCP or a BFOR-affiliated genetics specialist. The BFOR study sought to pilot a model of internet-enabled genetic testing and measure the psychosocial impact and clinical outcomes of this platform for *BRCA1/2* population screening of AJ individuals. An additional study goal was to examine the feasibility of engaging PCPs in results disclosure and follow-up to inform policies and programs for scaling *BRCA1/2* testing. We surveyed PCPs nominated by patients enrolled in the study to disclose their results to understand PCPs' willingness to disclose *BRCA1/2* founder mutation results and their knowledge and attitudes about *BRCA1/2* testing and results disclosure in the primary care setting. After results disclosure, we again surveyed PCPs and conducted semi-structured interviews with a subset of PCPs to explore their experiences and gather recommendations for future interventions.

## METHODS

### BFOR Study Population and Procedures

The BFOR study enrolled participants aged  $\geq 25$  years contacted through community advertising and outreach, including social media. Participant eligibility criteria are described elsewhere.<sup>14</sup> If participants chose to receive results from their PCP, those PCPs were informed about the study by mail and fax and asked (before results were available) to agree or decline to disclose their patient's results. If PCPs

agreed to disclose, study staff sent the results to PCPs via the online study portal or mail. An algorithm created by study investigators classified all results as negative with no further genetic testing needed; negative but with further evaluation needed because of a participant's family or personal medical history ("high-risk negative"); or positive.<sup>14</sup>

**PCP Educational Materials and Resources.** PCPs who agreed to disclose results received their patient's results with written interpretation, including the family history provided by a patient during enrollment and an assessment of whether further testing was needed. PCPs also received online and/or paper-based informational materials about *BRCA1/2* mutations, strategies for results communication, situations when additional testing is recommended, recommendations for screening and prevention strategies for mutation carriers, and implications for relatives.

**Instrument Development.** PCP surveys were developed using items about *BRCA1/2* knowledge, attitudes, and practices from other surveys.<sup>20–22</sup> We additionally developed items to assess PCPs' intent to disclose results to their study patients. Surveys underwent cognitive testing and revisions. The initial PCP survey was four pages ( $\leq 10$  min); the post-disclosure survey was two pages ( $\leq 5$  min). Semi-structured interview guides were developed based on post-disclosure survey findings ( $\leq 30$  min).

### Data Collection

**PCP Engagement.** We used the BFOR study's centralized online database to determine the frequency of PCPs agreeing to disclose their patients' results, from December 2017 to March 2020.

**Initial PCP Survey.** The initial PCP survey was mailed to the first 125 unique PCPs from each city (126 in one city) nominated by a BFOR study participant to disclose results (total  $N=501$ ), with an option to reply online. PCPs were invited to participate regardless of whether they ultimately agreed to disclose results. Each survey mailing included a personalized cover letter, an upfront incentive of \$50 (check or cash card),<sup>15</sup> and a pre-paid return envelope. The first and second survey reminders were mailed roughly 3 and 6 weeks after the initial mailing.

**Post-results Disclosure PCP Survey.** About 12 weeks following the release of their patient's *BRCA1/2* results, the first 101 PCPs who disclosed patient results were sent a brief survey (by mail or email if available) with a \$25 up-front check assessing their experience sharing results. Mailing and reminder processes were otherwise identical to those used in the initial PCP survey.

**PCP Interviews.** In addition to the post-disclosure survey, we contacted PCPs from a range of geographic regions and specialties who had disclosed results, inviting them to participate in semi-structured phone interviews about their results disclosure experience. We contacted 30 PCPs to achieve our target of 10 PCP interviews. PCPs were asked about challenging or rewarding aspects of results disclosure, perceptions about the educational materials, and attitudes and suggestions about integrating testing and results disclosure into primary care. PCPs received \$100 for participating.

## Analyses

**Survey Data.** We used Chi-square statistics and the Cochran-Armitage test for trend to examine relationships between PCP demographic and practice characteristics, knowledge, attitudes, and *BRCA1/2* experience and whether they agreed to disclose results. We considered 2-sided *p* values <0.05 to be statistically significant. We employed multivariable logistic regression models, including variables associated with agreeing to disclose with a *p* value of <0.2 in unadjusted analyses as well as PCP specialty and city. In descriptive analyses, missing data were infrequent for most variables and were reported separately. In adjusted analyses, one variable had missing data for two PCPs about ordering *BRCA1/2* tests in the past year. These PCPs were considered to have not ordered *BRCA1/2* tests in the past year. A sensitivity analysis excluding these two PCPs had nearly identical results (not shown).

**Qualitative Data.** Each interview was audio-recorded and professionally transcribed. The interdisciplinary team completed a multi-stage thematic content analysis incorporating inductive and deductive principles.<sup>23</sup> The code structure was iteratively developed and reviewed by three team members (LEP, AR, YSL) for logic and breadth. Using Dedoose software, two team members (AR, YSL) independently coded all transcripts, then met to achieve consensus on each transcript. The reduced dataset informed category construction, generating a thematic framework for data interpretation and allowing the research team to explore within and across cases to identify key concepts, patterns, and relationships.

## Ethics

The Advarra central IRB approved the study, with additional oversight by study institutions. Study participants undergoing *BRCA1/2* testing provided electronic informed consent. Consent was implied for surveyed and interviewed PCPs.

## RESULTS

### PCP Engagement and Surveys

By March 1, 2020, 35.1% of BFOR participants (1703 of 4848 enrolled) had requested their PCP disclose results; PCPs

agreed to disclose results for 690 of these participants (40.5% of requests). The remaining participants requested results disclosure from a BFOR genetic specialist. Factors associated with participants' decision to nominate their PCP will be presented elsewhere.

Among 501 PCPs nominated by their patients to disclose results and sent an initial survey, 234 (46.7%) responded. Sociodemographic differences between responding and non-responding PCPs were previously published.<sup>15</sup> Table 1 summarizes characteristics of responding PCPs. Most were

**Table 1 Characteristics of Primary Care Provider Initial Survey Respondents (n=234)**

PCP characteristics	N (%)
City	
Los Angeles	61 (26.1)
Boston	72 (30.8)
Philadelphia	50 (21.4)
New York	51 (21.8)
Sex	
Female	135 (56.7)
Male	99 (42.3)
Specialty	
Internal medicine	152 (65.0)
Family practice	39 (16.7)
Obstetrics/gynecology	37 (15.8)
Other	6 (2.6)
Practice setting	
Office	216 (92.3)
Hospital	18 (7.7)
Academic affiliation	
No	90 (38.5)
Yes	144 (61.5)
Years since health professional school graduation	
0-5	5 (2.1)
6-10	23 (9.8)
11-20	38 (16.2)
Greater than 20	168 (71.4)
Median (IQR)	27 (20, 36)
During the past 12 months have any of your patients asked you if they can or should get tested for a BRCA1/2 mutation?	
No	25 (10.7)
Yes	209 (89.3)
Median number of patients, IQR (n=202 responses)	5 (2, 10)
During the past 12 months, have you referred any of your patients to another health care provider for BRCA1/2 testing, or for an assessment of whether they are candidates for BRCA1/2 testing?	
No	52 (22.2)
Yes	182 (77.8)
Median number of patients, IQR (n=173 responses)	4 (2, 10)
Ordered a BRCA1/2 test in past 12 months**	64 (27.6)
No	168 (71.8)
Yes	64 (27.4)
Missing	2 (0.9)
Number of BRCA1/2 carriers ever cared for	
0	14 (6.0)
1-4	91 (38.8)
6-10	46 (19.7)
11-20	41 (17.5)
Greater than 20	22 (9.4)
Missing	20 (8.5)
Median (IQR) (n=214)	6 (2, 15)
Proportion of patients who are of Ashkenazi Jewish ancestry	
0-5	51 (21.8)
6-10	43 (18.4)
11-20	39 (16.7)
Greater than 20	48 (20.5)
Missing	53 (22.6)
Median, IQR (n=181)	10.0 (5.0, 23.0)

IQR = interquartile range

internists (65.0%) and affiliated with an academic institution (61.5%). The vast majority of PCPs had had patients interested in BRCA1/2 testing, had referred patients for testing, or had patients who were BRCA1/2 mutation carriers. Fewer (26.9%) had ordered a BRCA1/2 test.

We assessed PCPs' BRCA1/2 knowledge through three questions (Table 2). The median number of questions answered correctly was 2 (interquartile range, 1–2). PCPs' attitudes about genetic testing are shown in Table 2. Overall, most reported feeling very (24.5%) or somewhat qualified (51.5%) to recommend BRCA1/2 testing. Most felt very (19.7%) or somewhat qualified (39.1%) to share results. Overall, 58.2% felt that genetic testing was or could be easily incorporated into their practice. The most frequently identified major obstacles to adopting greater use of BRCA1/2 testing were lack of insurance coverage for genetic testing, testing costs, and PCPs' lack of counseling experience (Fig. 1).

Seventy-five respondents (32.1%) reported they had declined to share results with their patients enrolled in the BFOR study, 128 (54.7%) had agreed, and the remainder had not yet decided or not yet received the invitation to share results at the time of the survey. Among respondents who declined to share results with their patient and who provided their reasons for declining (n=69), the most common reasons given were limited knowledge about BRCA1/2 testing (64.0%) and belief that

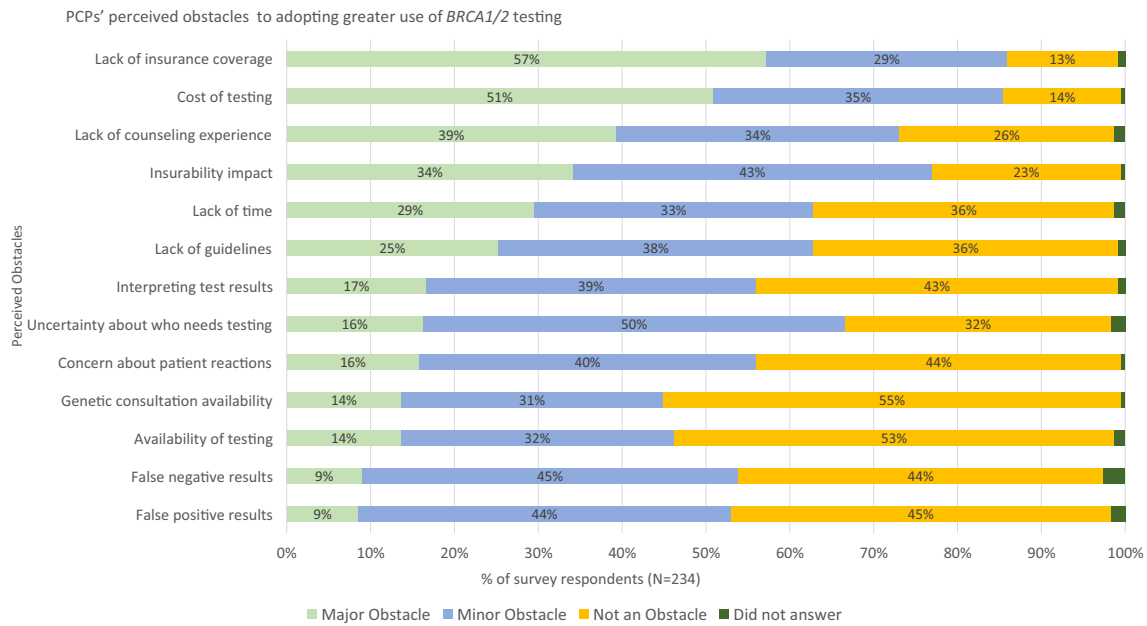
sharing results should be done by a specialist (52.0%) (Table 2). Centralized study data on PCPs' actual agreement to disclose revealed that 100 (42.7%) of the survey respondents ultimately declined to disclose results, and 134 (57.3%) agreed to disclose. Survey respondents were more likely than non-respondents to agree to disclose BRCA1/2 results (57.3% vs. 28.6%, p<0.001).

In unadjusted analyses, feeling somewhat or very qualified to share results and having ordered a BRCA1/2 test in the past year were associated with an increased likelihood of agreeing to disclose results as identified by centralized study data (p<0.001 for both). PCPs' likelihood of agreeing to disclose increased as the number of knowledge questions answered correctly increased (p=0.003). In adjusted analyses, feeling very or somewhat qualified to disclose results was the only PCP characteristic associated with a statistically greater likelihood of agreeing (OR 6.53, 95% CI 3.31, 12.88) (Table 3).

Of 101 PCPs who disclosed results by December 2019 and were sent a post-disclosure survey, 56 (55.4%) responded. Most felt very (73.2%) or somewhat (23.2%) comfortable sharing the results with their patients (Table 4). Among the 5 PCPs who disclosed positive results, 3 felt somewhat comfortable and 2 felt very comfortable. Nearly all PCPs reported being probably or definitely willing to disclose results again (98.2%).

**Table 2 Knowledge and Attitudes of Baseline Survey Respondents (n=234) About BRCA1/2 Testing and Involving PCPs in Testing**

PCP Attitude	N (%)
How qualified do you consider yourself to recommend BRCA testing?	
Very well qualified	57 (24.5)
Somewhat qualified	120 (51.5)
Not very well qualified	45 (19.3)
Not qualified at all	7 (3.0)
Not sure	4 (1.7)
How qualified do you consider yourself to share BRCA results?	
Very well qualified	46 (19.7)
Somewhat qualified	91 (39.1)
Not very well qualified	65 (27.9)
Not qualified at all	29 (12.5)
Not sure	2 (0.9)
Knowledge questions answered correctly	
1) Suppose you have a female patient whose aunt or grandmother on her father's side carries the BRCA1 gene mutation for breast/ovarian cancer syndrome. In your opinion, could your patient also be a carrier of this mutation? Answer: Yes	190 (81.2)
2) In your opinion, what is the likelihood that a female BRCA1 gene mutation carrier will go on to develop breast or ovarian cancer by age 70? Answer: 40 to 70%	158 (67.5)
3) In your opinion, what percentage of young (<age 40) female breast cancer patients of Ashkenazi (Jewish) ancestry have a BRCA1 or BRCA2 gene mutation? Answer: 20 to 49%	52 (22.2)
All three questions correct	29 (12.4)
Median number of questions answered correctly (Median, interquartile range)	2 (1, 2)
Genetic testing could easily be incorporated into my practice	
Agree	85 (36.3%)
Already incorporated	49 (20.9)
Disagree	97 (41.5)
Missing	1 (0.4%)
Agreed to disclose results based on central study data	134 (57.23)
Reasons for declining to disclose based on survey results (out of n = 69)	
Limited knowledge	48 (69.6)
Should be specialist	40 (58.0)
Lack of time during visits	27 (39.1)
Lack of time for reviewing educational materials	20 (29.0)
Lack of compensation	10 (14.5)
Other	8 (11.6)



**Figure 1** Perceived barriers to adopting greater use of BRCA1/2 testing, according to initial primary care provider survey respondents (N=234).

## Qualitative Findings

In semi-structured interviews, PCP participants ( $n=10$ ), primarily internists, provided feedback on the results disclosure process and potential roles of PCPs in BRCA1/2 testing. Nearly all reported receiving some previous education in genetic medicine but typically referred patients to specialists for testing. Overall, PCPs' perceived favorably their involvement in results disclosure through BFOR. Negative result discussions were often over the phone and described as quick, easy, streamlined, straightforward, and "not a big deal." One PCP stated the experience "opened me up to having this discussion more readily with patients." The PCPs who disclosed positive results felt the process had gone smoothly. However, several PCPs who disclosed negative results said they would want more guidance for positive results, including implications for patients' insurability. PCPs also identified potential challenges, such as ensuring adequate time for results review and communication between visits.

The educational materials mailed to PCPs that interpreted results and suggested next steps were described as "comprehensive" and "thorough," seemingly "designed for quick interpretation and communication at the point of care." "I feel like I learned something," noted one PCP. However, receiving paper materials in the mail was perceived as an unhelpful format. For results, decision support, and educational materials, PCPs prioritized simplicity, convenience, and ready availability at the point of care.

Several interviewees had positive perceptions of PCP involvement in genetic screening overall, feeling that PCPs could leverage a "foundation and a trust" from "preexisting relationship[s]" with patients. One PCP commented, "I think it's very helpful to get [results] from somebody they know and have a working therapeutic relationship with." One PCP noted

that PCPs often care for several generations of the same family, which can facilitate cascade testing of at-risk relatives. However, PCPs also had reservations: for example, not having "the knowledge base," or adding "a relatively large burden to add to [PCPs'] millions of other burdens." Several PCPs were very concerned that conversations with a patient who received positive results would trigger a more in-depth conversation for which they did not feel equipped. Several PCPs emphasized the need for guidance about where and how PCPs should refer BRCA1/2-positive patients and emphasized that subsequent monitoring and management should be led by specialists.

PCPs appreciated that the BFOR study provided patient education and provider decision support. Some PCPs felt that, in general, identifying who needed to be tested was more challenging than disclosing results and wanted more education and support in this process, including through the electronic medical record. Conversely, one PCP noted, "I don't really have the time or necessarily, the training to have a full discussion with patients over the risks and benefit of testing versus not testing."

## DISCUSSION

Prior studies have examined PCPs' knowledge and attitudes about genetic testing; however, to our knowledge, ours is the first prospective study to examine PCPs' actual willingness to disclose BRCA1/2 mutation results to patients undergoing testing, and to examine the implementation of an intervention to engage PCPs in this process. Importantly, most PCPs invited to disclose their patient's results to them through the BFOR study declined to do so, underscoring the need for focused efforts to engage PCPs. However, the PCPs who responded to our survey were interested in greater engagement in BRCA1/2

**Table 3 Unadjusted and Adjusted Analyses Examining Factors Associated with Primary Care Providers' Agreement to Disclose Their Patient's BRCA1/2 Results**

Characteristics	N (%) agreed to disclose results	p value, unadjusted*	OR (95% CI)**
Sex			
Male	63 (63.6)	0.09	1.00 (reference)
Female	71 (52.6)		0.68 (0.34, 1.36)
Specialty			
Internal medicine	82 (54.0)	0.24	1.00 (reference)
Obstetrics/gynecology	28 (71.8)		0.94 (0.37, 2.39)
Family medicine	21 (56.8)		1.53 (0.64, 3.68)
Other	3 (50.0)		0.38 (0.06, 2.33)
City			
Los Angeles	37 (60.7)	0.39	1.00 (reference)
Boston	36 (50.0)		0.81 (0.35, 1.88)
Philadelphia	28 (56.0)		1.00 (0.40, 2.52)
New York	33 (64.7)		0.82 (0.33, 2.03)
Practice setting			
Office-based practice	124 (57.4)	0.88	n/a
Hospital-based practice	10 (55.6)		
Academic affiliation			
No academic affiliation	53 (58.9)	0.69	n/a
Academic affiliation	81 (56.3)		
Length of time in practice			
>10 years since medical school graduation	117 (56.8)	0.69	n/a
≤10 years since medical school graduation	17 (60.7)		
Feeling qualified to share results			
Not qualified at all to share results, not very well-qualified, or unsure	29 (29.9)	<0.001	1.00 (reference)
Very or somewhat qualified to share results	105 (76.6)		<b>6.53 (3.31, 12.88)</b>
Patient asked about testing in past 12 months			
No	15 (60.0)	0.77	n/a
Yes	119 (56.9)		
Referred patient in past 12 months			
No	34 (65.4)	0.18	1.00 (reference)
Yes	100 (55.0)		0.77 (0.34, 1.71)
Ordered a BRCA1/2 test in the past 12 months			
No or missing (n=2 missing)	83 (48.8)	<0.001	1.00 (reference)
Yes	51 (79.7)		1.70 (0.73, 3.94)
Number of BRCA1/2 carriers ever cared for (n=214)			
0	5 (35.7)	0.46	n/a
1-4	54 (59.3)		
6-10	29 (63.0)		
11-20	25 (61.0)		
Greater than 20	12 (54.6)		
Percent of Ashkenazi Jewish patients (n=181)			
0-5%	31 (60.8)	0.30	n/a
6-10%	25 (58.1)		
11-20%	20 (51.3)		
Greater than 20%	34 (70.8)		
Performance on knowledge questions			
0 correct	6 (30.0)	<b>0.003***</b>	1.36 (0.91, 2.04) <sup>†</sup>
1 correct	31 (54.4)		
2 correct	75 (58.6)		
3 correct	22 (75.9)		

*Bold indicates a statistically significant value*

\*Chi-square tests

<sup>†</sup>Multivariable logistic regression including all variables with  $p < .20$  in unadjusted analyses

<sup>‡</sup>Cochran-Armitage test for trend

<sup>§</sup>Assessed as a continuous variable; odds ratio is per 1-point increase in score

testing and results disclosure, and those who disclosed results had positive experiences, suggesting the potential of targeted interventions to overcome barriers, at least among certain groups of PCPs.

Among our survey respondents, limited knowledge was the most frequently cited barrier to agreeing to share patients' results, suggesting a role for increased education. Higher

performance on knowledge questions was associated with greater likelihood of agreeing to share results in unadjusted analyses, although this did not reach statistical significance in adjusted analyses.

In considering how to incorporate genetic medicine into primary care, some PCPs believed a specialist should disclose BRCA1/2 results. However, over half of PCPs felt genetic

Table 4 PCPs' Experience with Results Disclosure in the BFOR Study Based on Post-disclosure Survey Results (N=56)

Questions about experience	N (%)
Patient results	
Positive	5 (8.9)
Negative but further evaluation recommended	14 (25.0)
Negative with no further evaluation recommended	37 (66.1)
Level of comfort disclosing results	
Very	41 (73.2)
Somewhat	13 (23.2)
Somewhat uncomfortable	2 (3.6)
Not at all comfortable	0 (0.0)
PCP experience (those who "agreed somewhat or strongly" with each statement)	
Able to help patient identify what s/he needed	52 (92.9)
My patient felt better	48 (85.7)
Discussion was right length	50 (89.3)
I had enough information to discuss results	51 (91.1)
I had enough information to refer	50 (89.3)
Participation improved knowledge	
A great deal	14 (25.0)
Somewhat	25 (44.6)
No	16 (28.6)
Participation improved comfort	
A great deal	12 (21.4)
Somewhat	30 (53.6)
No	14 (25.0)
Would probably or definitely be willing to disclose results again	55 (98.2)

PCP = primary care provider

testing could be incorporated into their practices. Respondents identified several barriers to incorporating greater use of *BRCA1/2* testing in practice, including concerns about costs, PCPs' lack of counseling experience, and time. Nevertheless, three-quarters of PCPs felt "very comfortable" disclosing results. In interviews, several PCPs voiced reservations about their readiness to disclose positive results. Ongoing follow-up is underway to evaluate management of mutation carriers and high-risk negative results. This will allow us to compare management initiated by PCPs versus BFOR-affiliated genetics specialists, and assess PCPs' readiness to make appropriate initial recommendations, including referral for further genetic testing in the case of high-risk negative tests, or prevention/surveillance strategies and cascade testing for those testing positive.

Interviewed PCPs also suggested opportunities to improve PCP engagement efforts. We relied primarily on paper mail, which limited PCPs' consistent receipt of the educational materials and their convenience. However, their content and scope were well-received. Future work should test electronic decision support based on family and personal history data to help PCPs determine whether initial testing is needed, or whether further genetic testing is needed despite a negative *BRCA1/2* test, ideally through their own electronic medical record system.

Only 1/3 of BFOR study participants nominated their PCP to disclose their *BRCA1/2* results. Participants who did not choose their PCP most frequently cited a preference for getting these results from a clinician with expertise in cancer genetics

as the reason; these reasons and associated factors will be described in a separate manuscript. These low rates of choosing a PCP underscore that barriers to integrating *BRCA1/2* results communication into primary care exist for both providers and patients.

Our study has limitations. Most PCPs were affiliated with academic institutions in one of four major cities, so our results may not generalize to PCPs across the USA. Although our response rates compare favorably with other physician surveys, slightly less than half of surveyed PCPs responded to the initial survey, and PCPs who agreed to disclose results were more likely to respond. PCPs who did not respond may have been more hesitant about incorporating genetic medicine into their practices, and our results do not reflect their perspectives. We did not survey PCPs who were not nominated by their patients to disclose results; those PCPs also may have been recognized by their patients as having limited familiarity with or interest in *BRCA1/2* testing and genetic medicine.

There is a pressing need to test strategies to engage PCPs in genetic medicine. This will be particularly important in non-academic settings and rural and underserved areas with less access to genetics specialists, so future research must engage PCPs and patients in those settings. To mitigate undertesting, reduce disparities, and meet growing demand, PCPs also will need to be more involved in identifying patients at risk for *BRCA1/2* mutations and ordering appropriate tests. Online training, electronic decision support, and virtual consultation services could be important tools to increase PCPs' comfort and skills in genetic medicine.

**Corresponding Author:** Lydia E. Pace, Brigham and Women's Hospital, Boston, MA, USA (e-mail: lpace@bwh.harvard.edu).

**Funding** This study was funded by an American Cancer Society Cancer Control Career Development Award for Primary Care Physicians (LEP, grant number 130741-CCDA-17-072-01-CCDA), The Sharon Levine Corzine Research Fund, Breast Cancer Research Foundation, The Basser Center for BRCA, The Nancy Ann Mellen Fund for Hereditary Cancer Research, Robin and Ken Isaacs, Brooke and Eric Meltzer, Jerold O. and Abbe Beth Young, Michael Stoler, Anonymous Donors, American Cancer Society Mentored Research Scholar Grants in Applied and Clinical Research (JGH, grant number MRS-16-020-01-CPPB), NCI P30 CA008748 (JGH, JDL, PRT, VM, TW, BD, MER, KM, KO), NCI 5-P30-CA-016520-38 (SMD, KLN, HS, KS), NCI P30CA016042 (BYK, DK, JL, AB). Quest Diagnostics provided testing at discounted costs to the BFOR study and no cost to participants; Quest Diagnostics was not involved in study design or presentation of findings.

#### Declarations:

**Conflict of Interest:** Dr. Garber discloses relationships with Helix Genetics, Konica Minolta and the Breast Cancer Research Foundation. Dr. Tung discloses relationships with Astra-Zeneca and Myriad. All other authors reported no conflicts of interest.

## REFERENCES

- Kuchenbaecker KB, Hopper JL, Barnes DR, et al. Risks of Breast, Ovarian, and Contralateral Breast Cancer for BRCA1 and BRCA2 Mutation Carriers. *JAMA*. 2017;317(23):2402-2416.
- King MC, Levy-Lahad E, Lahad A. Population-based screening for BRCA1 and BRCA2: 2014 Lasker Award. *JAMA*. 2014;312(11):1091-1092.
- U.S Preventive Services Task Force *Final Recommendation Statement: BRCA-Related Cancer: Risk Assessment, Genetic Counseling, and Genetic Testing*. Rockville, MD August 20 2019.
- Yurgelun MB, Hiller E, Garber JE. Population-Wide Screening for Germline BRCA1 and BRCA2 Mutations: Too Much of a Good Thing? *J Clin Oncol*. 2015;33(28):3092-3095.
- Domchek S, Robson M. Broadening Criteria for BRCA1/2 Evaluation: Placing the USPSTF Recommendation in Context. *JAMA*. 2019;322(7):619-621.
- Chen Z, Kolor K, Grosse SD, et al. Trends in utilization and costs of BRCA testing among women aged 18-64 years in the United States 2003-2014. *Genet Med*. 2017.
- Guo F, Hirth JM, Lin YL, et al. Use of BRCA Mutation Test in the U.S., 2004-2014 *Am J Prev Med*. 2017;52(6):702-709.
- Han X, Jemal A. Recent Patterns in Genetic Testing for Breast and Ovarian Cancer Risk in the U.S. *Am J Prev Med*. 2017;53(4):504-507.
- Kolor K, Chen Z, Grosse SD, et al. BRCA Genetic Testing and Receipt of Preventive Interventions Among Women Aged 18-64 Years with Employer-Sponsored Health Insurance in Nonmetropolitan and Metropolitan Areas - United States, 2009-2014. *MMWR Surveill Summ*. 2017;66(15).
- Cragun D, Weidner A, Lewis C, et al. Racial disparities in BRCA testing and cancer risk management across a population-based sample of young breast cancer survivors. *Cancer*. 2017;123(13):2497-2505.
- Hamilton JG, Abdiwahab E, Edwards HM, Fang ML, Jdayani A, Breslau ES. Primary care providers' cancer genetic testing-related knowledge, attitudes, and communication behaviors: A systematic review and research agenda. *J Gen Intern Med*. 2017;32(3):315-324.
- van der Wouden CH, Carere DA, Maitland-van der Zee AH, Ruffin MT, Roberts JS, Green RC. Consumer Perceptions of Interactions With Primary Care Providers After Direct-to-Consumer Personal Genomic Testing. *Ann Intern Med*. 2016;164(8):513-522.
- Vassy JL, Davis JK, Kirby C, et al. How Primary Care Providers Talk to Patients about Genome Sequencing Results: Risk, Rationale, and Recommendation. *J Gen Intern Med*. 2018;33(6):877-885.
- Morgan KM HJ, Symecko H, Kamara D, Jenkins C, Lester J, Spielman K, Pace LE, Gabriel C, Levin JD, Tejada PR, Braswell A, Marcell V, Wildman T, Devolder B, Baum R, Block JN, Fesko Y, Boehler K, Howell V, Heitler J, Robson ME, Nathanson KL, Tung N, Karlan BY, Domchek SM, Garber JE, Ofit K Population Screening for BRCA1/2 Founder Mutations Utilizing an Internet-enabled Medical Model. *Submitted manuscript*.
- Pace LE, Lee YS, Tung N, et al. Comparison of up-front cash cards and checks as incentives for participation in a clinician survey: a study within a trial. *BMC Med Res Methodol*. 2020;20(1):210.
- Morgan K, Symecko H, Kamara D, et al. The BRCA founder outreach study: Initial results of a digital health model. *J Clin Oncol*. 2020;38(15\_suppl):2007-2007.
- Roa BB, Boyd AA, Volcik K, Richards CS. Ashkenazi Jewish population frequencies for common mutations in BRCA1 and BRCA2. *Nat Genet*. 1996;14(2):185-187.
- Struwing JP, Hartge P, Wacholder S, et al. The risk of cancer associated with specific mutations of BRCA1 and BRCA2 among Ashkenazi Jews. *N Engl J Med*. 1997;336(20):1401-1408.
- Prevalence and penetrance of BRCA1 and BRCA2 mutations in a population-based series of breast cancer cases. Anglian Breast Cancer Study Group. *Br J Cancer*. 2000;83(10):1301-1308.
- Wideroff L, Freedman AN, Olson L, et al. Physician Use of Genetic Testing for Cancer Susceptibility: Results of a National Survey. *Cancer Epidemiol Biomarkers*. 2003;12:295-303.
- Freedman AN, Wideroff L, Olson L, et al. US physicians' attitudes toward genetic testing for cancer susceptibility. *Am J Med Genet A*. 2003;120a(1):63-71.
- Wideroff L, Vadaparampil ST, Greene MH, Taplin S, Olson L, Freedman AN. Hereditary breast/ovarian and colorectal cancer genetics knowledge in a national sample of US physicians. *J Med Genet*. 2005;42(10):749-755.
- Green J, Thorogood N. *Qualitative Methods for Health Research, 3rd Edition*. Los Angeles: Sage Publications; 2014.

**Publisher's Note:** Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.