Minority Recruitment in Hereditary Breast Cancer Research

Chanita Hughes,1 Susan K. Peterson,2 Amelie Ramirez,3 Kipling J. Gallion,3 Paige Green McDonald,4 Celette Sugg Skinner,5 and Deborah Bowen6

1Abramson Cancer Center and Department of Psychiatry, University of Pennsylvania, Philadelphia, Pennsylvania; 2Department of Behavioral Science, University of Texas M.D. Anderson Cancer Center, Houston, Texas; 3Chronic Disease Prevention and Control Research Center, Baylor College of Medicine, Houston, Texas; 4Division of Cancer Control and Population Sciences, National Cancer Institute, Bethesda, Maryland; 5Department of Surgery, Duke University Medical Center, Durham, North Carolina; and 6Fred Hutchinson Cancer Research Center, Seattle, Washington

Abstract

Although recruitment of ethnic and racial minorities in medical research has been evaluated in several studies, much less is known about the methods used to recruit these populations to participate in cancer genetics research. This report reviews the resources that have been used to identify and recruit ethnic and racial minorities to participate in hereditary breast cancer research. Overall, hospital-based resources were used most often to identify potential subjects, and active recruitment methods were used most frequently to enroll eligible subjects. This review suggests that there appears to be a finite number of resources and strategies to identify and recruit potential subjects to participate in cancer genetics research; however, options for improving awareness about cancer genetics research among ethnic and racial minorities have not been extensively evaluated. To study ethnic and racial minority participation in cancer genetics research, stronger evaluation components will need to be integrated into research methods. Both observational and experimental studies are needed to determine resources that are most effective for identifying potential subjects who are ethnic and racial minorities and to evaluate the effects of different recruitment strategies on enrollment decisions among these populations. (Cancer Epidemiol Biomarkers Prev 2004;13(7):1146–55)

Introduction

Each year, thousands of individuals are diagnosed with breast cancer (1), and the discovery of BRCA1 and BRCA2 (BRCA1/2) susceptibility genes has catalyzed research on the clinical, ethical, and psychologic implications of genetic testing for inherited breast cancer risk. Although only ~5% to 10% of all breast cancer cases are due to BRCA1/2 mutations, women who carry a risk-conferring gene alteration have an estimated 55% to 85% increased lifetime risk of developing breast cancer and a 15% to 60% increased lifetime risk of developing ovarian cancer (2-5). It is now possible for individuals from all ethnic and racial backgrounds who have a personal and family history of breast cancer that is suggestive of hereditary disease to learn if they carry a cancer predisposing BRCA1/2 mutation. To date, a substantial amount of research has been conducted to understand rates and predictors of genetic test acceptance and to evaluate the clinical, psychologic, and behavioral impact of genetic risk information (6-12). These studies have shown that many but not all high-risk individuals obtain genetic testing for BRCA1/2 mutations, and genetic test acceptance is higher among individuals who have greater perceived risks of developing cancer and those with higher levels of familial cohesion (7, 9). While there is limited evidence that receiving test results generates adverse psychologic effects (6, 10, 13), recent studies have shown that BRCA1/2 test results may generate specific emotional concerns among mutation carriers (14, 15). Further, while early studies indicated that genetic testing for inherited breast cancer risk had a limited impact on cancer screening and prevention behaviors (11), more recent work has shown that genetic test results may motivate use of cancer screening among mutation carriers (12). Despite the significance of these studies, the underrepresentation of ethnic and racial minorities has been a consistent limitation noted in the majority of this work. Less than 5% of participants in recent studies on BRCA1/2 genetic test acceptance were African American (7, 8), and racial and ethnic minorities were not included in early research on the utilization of genetic testing for inherited breast cancer risk (6). Similar trends have been reported in more recent research. Less than 10% of participants in a national cancer genetics consortium, which was designed to be representative of the U.S. population, are ethnic or racial minorities, although these groups (i.e., individuals who are of Black African, Asian and Pacific
Islander, Latino or Hispanic, and Native American/American Indian descent) comprise 25% of the U.S. population (16).

The underrepresentation of racial and ethnic minorities in research studies has been addressed in a substantial number of studies; this work has described several social, psychologic, and structural barriers to ethnic and racial minority participation in medical research. These barriers include mistrust of the medical community and the research process, lack of knowledge and awareness about research studies, poor economic status, and cultural and linguistic factors (17-22).

Although much less is known about barriers to ethnic and racial minority participation in cancer genetics research, previous studies suggest that similar barriers may reduce ethnic and minority participation in these studies. For example, Hughes et al. found that, among African American women who had a college education, those who reported concerns about being exploited were significantly less likely to participate in an education session about hereditary breast cancer and genetic testing compared with women who reported no concerns about exploitation.8 This study also found that younger African American women and those who had lower incomes and greater levels of cancer-specific distress were most likely to decline participation in cancer genetics education. Moreover, although similar strategies were used to recruit African American and Caucasian women to participate in a randomized trial that compared different methods of providing education about hereditary breast cancer, African American women were significantly less likely than Caucasian women to self-refer for study participation (23).

Because of the importance of voluntariness in decisions to participate in research overall, especially in studies that involve genetic testing (24, 25), self-referral is a common method for recruiting subjects into cancer genetics studies (7, 8). However, not all studies related to hereditary breast cancer involve genetic testing, and other methods have been suggested to increase participation among ethnic and racial minorities in medical research studies. These methods include identifying subjects through physician referrals, tumor registries, community outreach, and targeted recruitment materials (20, 26). Each of these approaches has several advantages and disadvantages. For example, physician referrals may be effective because health care providers have access to both personal and family medical histories and can readily identify individuals who are eligible for study participation (27, 28). Similarly, tumor registries and hospital records may be effective for identifying racial and ethnic minorities because they contain demographic and clinical information that can facilitate identification of potential subjects. On the other hand, disadvantages of physician referral and tumor registry approaches may include physician mistrust of the sponsoring institution and limited representation of ethnic and racial minorities in the patient populations at medical facilities. Other approaches that have been suggested include community outreach and mass media (29-32). However, limited information is available on the resources that are used to identify ethnic and racial minorities for participation in cancer genetics research.

Study recruitment is a complex process that involves determination of ascertainment sites, development of enrollment procedures, identification of study participants, and implementation of procedures for subject retention.9 These activities can be conceptualized within a larger framework of study recruitment that includes mechanisms to enhance awareness about the research, acceptance of study procedures, and access to study sites among potential subjects (33). According to this model, awareness includes understanding the procedures involved in the study, while acceptability and access encompass community support for the research and one’s ability to overcome practical barriers (e.g., transportation) to study participation, respectively (33). While awareness has been conceptualized as understanding the procedures involved in participating in a study (33), awareness may also include knowledge about the availability of research studies. Outreach strategies and personalized recruitment approaches have been suggested as possible methods to increase awareness about cancer research in ethnic and racial minority groups (33); however, information on the methods that have been used to recruit these populations to participate in cancer genetics research is not available.

To guide future efforts to recruit ethnic and racial minority groups for participation in cancer genetics research, we conducted a review of the literature to describe the resources that have been used to identify ethnic and racial minority groups for participation in hereditary breast cancer research and the strategies that have been used to recruit these populations into these studies. Specifically, to address the question of what types of resources and strategies are used to recruit ethnic and minorities for hereditary breast cancer studies, we first describe the resources that have been used to identify subjects. We then report on the methods that were used for study recruitment. Because investigators with NIH-funded research projects are required to report enrollment rates for racial and ethnic minority groups, we also summarize enrollment rates for these populations to provide information on the yield of subjects enrolled using various strategies. Developing a better understanding of the methods that have been used to recruit ethnic and racial minorities into hereditary breast cancer studies will help to identify potential barriers and guide future recruitment efforts to increase the generalizability of research on genetic risk factors for cancer.

7 D. Bowen. Minority recruitment in the cancer genetics research: identification of problems and opportunities for new research. Presented at the Cancer Genetics Network Steering Committee Meeting, 2002, Reston, VA.


Methods

We conducted a search of the PubMed database to identify empirical reports published between 1995 and 2003 via a cascade method that used the broadest terms (e.g., hereditary breast cancer and genetic testing) first, with subsequent searches using more specific terms (e.g., BRCA1/2). Specifically, to identify studies related to inherited breast cancer susceptibility, we used the terms “hereditary breast cancer” in our first search and then used the terms “genetic testing, breast cancer” in our second search. In our last search, we used the terms “BRCA1” or “BRCA2.” All searches were conducted using EndNote software to facilitate development of the reference library.

Once the reference library was created, we then conducted separate searches to identify studies with ethically and racially diverse samples. We first used the terms “race” or “ethnic” to identify the initial set of studies and then conducted additional searches using specific racial and ethnic group categories (e.g., African American, Hispanic, and Asian American). Duplicate citations were deleted.

Published reports from epidemiologic studies, clinical trials, and observational studies related to breast cancer susceptibility genes and genetic testing that recruited human subjects and reported data on the race and ethnicity of subjects were eligible for inclusion in this review. Specifically, studies that evaluated the prevalence of hereditary breast cancer susceptibility genes and those that explored psychosocial and behavioral issues in genetic testing (e.g., interest in genetic testing and genetic test acceptance) were eligible for inclusion in this review. We included studies that evaluated intentions to have genetic testing for inherited breast cancer risk to capture studies conducted before testing for BRCA1/2 mutations became available. We excluded studies that did not involve recruitment of human subjects (e.g., review articles, comments and letters, and animal studies) and those that were related to genetic aspects of cancer but were broader in scope (e.g., HER-2). Abstracts of scientific meetings were not reviewed. Because we were interested in identifying the resources and strategies that have been used to recruit racial and ethnic minority groups represented in the U.S. population, we excluded studies in which subjects had been recruited entirely in foreign countries. We also excluded studies that only included individuals of Ashkenazi Jewish ancestry because these individuals are not categorized as racial or ethnic minorities. For this review, recruitment sources were defined as the resources used to identify potential subjects, and recruitment strategies were defined as the procedures used to enroll subjects into the study. Consistent with prior research, we categorized recruitment strategies as either active or passive (34). Active recruitment strategies included methods in which investigators targeted subjects from a known source (e.g., tumor registry and clinic database), and passive recruitment strategies included methods in which investigators provided information about the study and individuals volunteered for study participation by contacting research staff (34). Recruitment through an ongoing research or clinical protocol was categorized as a passive recruitment strategy as was physician and family referral.

Results

We identified 159 studies on breast cancer susceptibility and genetic testing that met our inclusion criteria. Of these 159 studies, 38 were excluded because they did not involve recruitment of human subjects, 83 were excluded because they were conducted entirely in foreign countries, and 10 were excluded because they only included individuals of Ashkenazi Jewish ancestry or did not include any ethnic and racial minority groups. Of the remaining 28 articles, 5 were excluded because the study sample overlapped with the sample used in a report that had already been included in the review and 8 did not provide sufficient information on the race or ethnicity of subjects or recruitment procedures.

The 15 articles on breast cancer susceptibility and genetic testing included in this review are summarized in Table 1. These studies were conducted in diverse geographic locations in the United States: 33.3% were conducted in southern states, 26.7% were conducted in western states, and 13.3% were conducted in northeastern states. In addition, 26.7% of studies were conducted in multiple states or internationally.10 Two thirds of the studies (66.7%; n = 10) were designed to evaluate hereditary breast cancer (e.g., prevalence of susceptibility genes) in a specific ethnic or racial group, assess ethnic differences in the prevalence of breast cancer susceptibility genes, or compare racial groups in terms of knowledge and attitudes about genetic testing. The studies that evaluated hereditary breast cancer in a specific racial group only included African Americans. Fifty-three percent of studies evaluated psychosocial and behavioral issues in genetic testing, and 47% were epidemiologic studies.

Overview of Recruitment Sources. As shown in Table 1, several types of resources were used to identify potential subjects. These resources included hospital resources such as oncology clinics and tumor registries, state cancer registries, hereditary cancer registries, and high-risk cancer clinics. Other resources such as public records (e.g., motor vehicle registrations) were also used; however, this resource was used in only one case-control study to identify potential subjects who did not have a personal history of cancer (35).

Hospital-Based Resources. Overall, hospital resources, either alone or in combination with other resources, were used most often to identify potential subjects. Resources such as tumor registries, billing records, and clinic databases were used to identify potential subjects in 53.3% of studies (n = 8). Active recruitment strategies were used in most of the studies that used hospital resources to identify subjects; however, the specific methods that were used to recruit subjects in these studies varied. For example, in the study conducted by Armstrong et al. (36), potential subjects were randomly identified from clinic and billing records and were invited to participate in the study by mail. The sample for this study consisted of women who returned a completed

10 The international study conducted by Narod et al. was included in this report because almost half of study subjects were recruited from clinics in the United States.
<table>
<thead>
<tr>
<th>Geographic Location (Reference)</th>
<th>Study Objective [Type of Study*]</th>
<th>Study Design</th>
<th>Recruitment Source</th>
<th>Recruitment Strategy</th>
</tr>
</thead>
<tbody>
<tr>
<td>California (49)</td>
<td>To evaluate the frequency of $BRCA1$ mutations in women with breast and ovarian cancer [E/P]</td>
<td>Cohort</td>
<td>State cancer registry</td>
<td>Active: Physicians notified about contacting patients identified from registry; introductory letter mailed to patients; patients were also contacted by telephone to review introductory materials</td>
</tr>
<tr>
<td>Pennsylvania (36)</td>
<td>To evaluate interest in genetic testing for $BRCA1/2$ mutations in a primary care population [P/B]</td>
<td>Cross-sectional</td>
<td>Hospital clinic</td>
<td>Active: Mailed survey to women randomly selected from billing records</td>
</tr>
<tr>
<td>District of Columbia (40)</td>
<td>To describe attitudes about genetic testing for inherited breast cancer risk among African American and Caucasian women with a family history of breast cancer [P/B]</td>
<td>Cross-sectional</td>
<td>Hospital clinic</td>
<td>Active: Breast cancer cases identified from tumor registry and contacted by telephone to identify unaffected FDRs; invitation letter mailed to unaffected FDRs; Passive: FDRs informed about study by physicians or brochures in hospital clinics</td>
</tr>
<tr>
<td>Alabama (38)</td>
<td>To evaluate differences in knowledge about breast cancer and interest in genetic testing among African American and Caucasian women [P/B]</td>
<td>Cross-sectional</td>
<td>Hospital clinic</td>
<td>Active: Subjects approached by project staff while waiting in hospital clinic</td>
</tr>
<tr>
<td>Washington (47)</td>
<td>To evaluate knowledge and interest in genetic testing among demographically diverse women [P/B]</td>
<td>Cross-sectional</td>
<td>Community outreach</td>
<td>Passive: Self-referral from mass media, radio announcements, and newspapers; media approaches to targeted to ethnic-specific outlets</td>
</tr>
<tr>
<td>Illinois and Texas (44)</td>
<td>To evaluate the prevalence of $BRCA1/2$ mutations in African Americans [E]</td>
<td>Cohort</td>
<td>High-risk clinic</td>
<td>Passive: Subjects referred by physicians or self because of family history of cancer</td>
</tr>
<tr>
<td>Utah/Louisiana (46)</td>
<td>To evaluate knowledge about genetic testing and identify predictors of testing intentions in African American $BRCA1$ kindred [P/B]</td>
<td>Cross-sectional</td>
<td>Hereditary breast cancer registry</td>
<td>Active: Mailed study invitation to individuals included a hereditary cancer registry</td>
</tr>
<tr>
<td>North Carolina (39)</td>
<td>To identify predictors of genetic test intentions in African American women with and without a family history of breast cancer [P/B]</td>
<td>Case-control</td>
<td>Hospital clinic</td>
<td>Active: Breast cancer patients identified from tumor registry; contacted by telephone to identify unaffected FDRs; unaffected FDRs invited to participate in the study by telephone; Active: Unaffected women without family history of cancer identified from hospital clinic records; invited to participate in the study by telephone</td>
</tr>
<tr>
<td>International (45)</td>
<td>To evaluate the association between oral contraceptive use and breast cancer risk in $BRCA1/2$ mutation carriers [E]</td>
<td>Case-control</td>
<td>High-risk clinic</td>
<td>Passive: Subjects identified from participants enrolled in genetic counseling and testing protocols</td>
</tr>
<tr>
<td>North Carolina (35)</td>
<td>To evaluate the prevalence of $BRCA1$ mutations in population-based women and to characterize differences in prevalence based on race and clinical factors [E/P]</td>
<td>Case-control</td>
<td>Cases, state cancer registry; Controls, public records</td>
<td>Active: Rapid ascertainment of breast cancer cases from state cancer registry; controls identified from state division of motor vehicles and health care financing records</td>
</tr>
</tbody>
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(Continued on the following page)
survey; of the 400 potential subjects who were identified, 15 were deceased. Of the remaining 385 potential subjects, 71% returned a completed survey and were enrolled (see Table 2). However, in the studies conducted by Thompson et al. (37) and Donovan and Tucker (38), potential subjects were identified during a clinic visit and were invited to participate in the study in-person. Lipkus et al. (39) used a different method to recruit subjects from hospital resources that included an institutional tumor registry and clinic database. The purpose of this study was to identify predictors of intentions to have genetic counseling and testing protocol. Subjects in this study were identified from hospital clinics; recruitment strategies included physician referral, family referral, and self-referral.

Six of the eight (75%) studies that used hospital resources to identify subjects were designed to evaluate differences in knowledge and attitudes about genetic testing between African American and Caucasian women (38, 40), assess genetic testing intentions or test utilization in African American women (37, 39), or to evaluate BRCA1 mutations in African Americans (41, 42). As shown in Table 2, the proportion of African American women enrolled in these studies ranged from 18% to 100%. Consistent with the wide range in the proportions of African American women included in these studies, the number of African American subjects varied widely. For example, 43 African American women were enrolled in the study conducted by Benkendorf et al. (40), whereas 108 African American women were enrolled in the study conducted by Donovan and Tucker (38). Although the studies conducted by Panguluri et al. (42) and Shen et al. (43) were both designed to evaluate BRCA1 mutations in

<table>
<thead>
<tr>
<th>Geographic Location (Reference)</th>
<th>Study Objective [Type of Study]</th>
<th>Study Design</th>
<th>Recruitment Source</th>
<th>Recruitment Strategy</th>
</tr>
</thead>
<tbody>
<tr>
<td>District of Columbia (42)†</td>
<td>To evaluate BRCA1 mutations in African Americans [E]</td>
<td>Case-control</td>
<td>Hospital clinic</td>
<td>Passive: Subjects identified from ongoing epidemiologic study of breast cancer in African American women</td>
</tr>
<tr>
<td>Washington (48)</td>
<td>To evaluate attitudes and decision making about genetic testing [P/B]</td>
<td>Cross-sectional</td>
<td>Community outreach</td>
<td>Passive: Advertisements placed in newspapers and newsletters; flyers displayed at community centers, targeted to ethnic-specific events</td>
</tr>
<tr>
<td>California (43)§</td>
<td>To evaluate BRCA1 mutations in African American breast cancer patients [E]</td>
<td>Cohort</td>
<td>Hospital clinic</td>
<td>Active: Consecutive breast cancer patients identified from individuals treated at oncology clinic</td>
</tr>
<tr>
<td>Michigan and Pennsylvania (41)</td>
<td>To determine the frequency of BRCA1/2 mutations in high-risk individuals [E]</td>
<td>Cohort</td>
<td>Hospital clinic</td>
<td>Passive: Self, physician, and family referrals to genetic counseling and testing protocol</td>
</tr>
<tr>
<td>New York (37)†</td>
<td>To identify predictors of participation in BRCA genetic counseling and testing in African American women [P/B]</td>
<td>Cohort</td>
<td>Hospital clinic</td>
<td>Active: Subjects waiting in clinic approached by project staff; nurse practitioners referred potential subjects for study participation</td>
</tr>
</tbody>
</table>

*E, epidemiologic/population based; E/C, epidemiologic/collaborative; P/B, psychosocial/behavioral; and E, epidemiologic.
†Racially or ethnically targeted study.
‡Registry maintained in Utah; most participants lived in LA.
§Includes subjects recruited from high-risk clinics in foreign countries (i.e., Canada and Europe) and the United States.
African Americans and both used hospital clinics to identify potential subjects, 54 subjects were enrolled in the study conducted by Shen et al., whereas 137 subjects were enrolled in the study conducted by Panguluri et al. Despite differences in the sample sizes, the number of affected breast cancer cases was similar in these studies; 45 high-risk African American women were enrolled in the study conducted by Panguluri et al. and 54 African American breast cancer cases were enrolled in the study conducted by Shen et al.

The proportion of ethnic and racial minority subjects in the remaining studies that used hospital clinics to identify potential subjects but were not targeted to a specific ethnic or racial minority group also varied widely. For example, 2% of the subjects enrolled in the study conducted by Shih et al. (41) to determine the

<table>
<thead>
<tr>
<th>Reference</th>
<th>Recruitment Source</th>
<th>Response Rate (%)</th>
<th>Sample Size</th>
<th>Sample Characteristics (No. of Subjects Enrolled)</th>
</tr>
</thead>
<tbody>
<tr>
<td>(49)</td>
<td>Cancer registry</td>
<td>78*</td>
<td>793</td>
<td>87.6% Caucasian (695), 0.5% African American (4), 6.9% Hispanic (54), 5% Asian (40)</td>
</tr>
<tr>
<td>(36)</td>
<td>Hospital clinic</td>
<td>71</td>
<td>272</td>
<td>66.5% Caucasian (181), 25.3% African American (69), 4.0% Asian American (11), 3.1% Hispanic (8), 1.1% Other (3)</td>
</tr>
<tr>
<td>(40)</td>
<td>Hospital clinic</td>
<td>71*</td>
<td>238</td>
<td>76% Caucasian (181), 18% African American (43), 6% Other (14)</td>
</tr>
<tr>
<td>(38)</td>
<td>Hospital clinic</td>
<td>95*</td>
<td>220</td>
<td>51% Caucasian (112), 49% African American (108)</td>
</tr>
<tr>
<td>(47)</td>
<td>Community outreach</td>
<td>Not reported</td>
<td>537</td>
<td>88.3% Caucasian (474), 7.2% African American (39), 1.5% Hispanic (8), 1.1% Asian or Pacific Islander (6), 1.8% Other (10)</td>
</tr>
<tr>
<td>(44)</td>
<td>High-risk clinic</td>
<td>Not reported</td>
<td>28</td>
<td>100% African American (28)</td>
</tr>
<tr>
<td>(46)</td>
<td>Hereditary breast cancer registry</td>
<td>Not reported</td>
<td>95</td>
<td>100% African American (95)</td>
</tr>
<tr>
<td>(39)</td>
<td>Hospital clinic</td>
<td>Cases 86, Controls 70</td>
<td>Cases 130, Controls 136</td>
<td>100% African American (266)</td>
</tr>
<tr>
<td>(45)</td>
<td>High-risk clinic</td>
<td>Not reported</td>
<td>Cases 1,311, Controls 1,311</td>
<td>100% African American (29.8% Jewish (391), 61.1% Other Caucasian (801), 1.1% Black (14), 7.4% French Canadian (97), 0.5% Other non-White (6), 0.1% Missing (1); Controls 31.6% Jewish (414), 57.7% Other Caucasian, 2.1% Black (28), 7.6% French Canadian (100), 1% Other non-White and missing (15)</td>
</tr>
<tr>
<td>(35)</td>
<td>Cases, state cancer registry; Controls, public records</td>
<td>Cases 77, Controls 68</td>
<td>Cases 211, Controls 188</td>
<td>Cases 57% Caucasian (120), 42% Black (89), 1% Native American (2); Controls 56% Caucasian (105), 42% Black (79), 1% Asian (2), 1% Other (2); 100% African American (137)</td>
</tr>
<tr>
<td>(42)</td>
<td>Hospital clinic</td>
<td>Not reported</td>
<td>Cases 45, Controls 92</td>
<td>32.5% Caucasian (80), 20.3% Ashkenazi Jewish (50), 22% African American (54), 25.2% Native American (62)</td>
</tr>
<tr>
<td>(48)</td>
<td>Community outreach</td>
<td>Not reported</td>
<td>246</td>
<td>100% African American (54)</td>
</tr>
<tr>
<td>(43)</td>
<td>Hospital clinic</td>
<td>Not reported</td>
<td>54</td>
<td>95% Caucasian (151), 2% African American (3), 2% Hispanic (3)</td>
</tr>
<tr>
<td>(41)</td>
<td>Hospital clinic</td>
<td>Not reported</td>
<td>159</td>
<td>100% African American (76)</td>
</tr>
<tr>
<td>(37)</td>
<td>Hospital clinic</td>
<td>54</td>
<td>76</td>
<td>100% African American (76)</td>
</tr>
</tbody>
</table>

*No racial or ethnic differences in response rates.
*Response rate for completion of education session that was required for study participation; African American women were less likely than Caucasian women to participate in education session.
*1African American women were oversampled; an additional 99 African American cases and 108 African American controls were also included in the analysis. However, sample characteristics included in the report are limited to the original sample.
prevalence of \( BRCA1/2 \) mutations in high-risk individuals were African American \( (n = 3) \) and 2% were Hispanic \( (n = 3) \). In contrast, 25% of the subjects enrolled in the study on genetic testing intentions conducted by Armstrong et al. \( (36) \) were African American \( (n = 69) \), 4% were Asian American \( (n = 11) \), and 3.1% were Hispanic \( (n = 8) \).

High-Risk Cancer Clinics. High-risk cancer clinics, including hereditary cancer registries, were used to identify subjects in 20% of studies. Although passive strategies used in these studies included self-referral to a high-risk cancer clinic \( (44) \), passive strategies also included identifying subjects from ongoing research and clinical protocols. For example, subjects who were enrolled in the case-control study conducted by Narod et al. \( (45) \) to evaluate the association between oral contraceptive use and breast cancer risk in \( BRCA1/2 \) mutation carriers were identified through genetic counseling and testing programs throughout the United States and in foreign countries. To be eligible for inclusion in this study, subjects had to have had genetic testing and found to carry a risk-conferring \( BRCA1/2 \) gene alteration. While some of the subjects who were included in the survey study conducted by Kinney et al. \( (46) \) had genetic testing as part of their participation in a hereditary breast cancer registry, testing was for research purposes only, and these individuals had not received \( BRCA1 \) test results prior to their participation in the survey study. Subjects in the study conducted by Narod et al. completed study measures as part of their appointment for genetic counseling or testing or after their clinic appointment, whereas subjects in the study conducted by Kinney et al. were invited to enroll in the study by mail and completed the survey by telephone or in-person. Subjects were enrolled into the study conducted by Gao et al. after provision of counseling about the benefits, limitations, and risks of genetic testing.

Two of the three studies that used high-risk cancer clinics to identify subjects were targeted to a specific ethnic group \( (44, 46) \). Although both of these studies only included African Americans, the study conducted by Gao et al. included 28 subjects and the study conducted by Kinney et al. included 95 subjects. A smaller number of ethnic and racial minorities \( (14 \text{ African American cases and 28 African American controls}) \) were included in the study conducted by Narod et al.

Other Resources. Community outreach was used in only two studies \( (13.3\%) \) to identify subjects; passive strategies were used to recruit subjects in both of these studies. For example, in the study conducted by Durfy et al. \( (47) \) to evaluate knowledge about genetic testing and genetic testing intentions, subjects self-referred for study participation after hearing about the project through mass media such as radio announcements and newspaper advertisements. Because a specific objective of this study was to evaluate knowledge and attitudes about genetic testing in a demographically diverse sample, media advertisements were targeted to ethnic specific outlets such as African American newspapers and community events. Press et al. \( (48) \) used a similar approach to recruit subjects; individuals self-referred to the study after learning about the research through advertisements placed in newspapers and flyers displayed at community centers. Recruitment efforts were also targeted to ethnic specific events in this study.

Although similar recruitment strategies were used in the studies conducted by Press et al. \( (48) \) and Durfy et al. \( (47) \), the representation of ethnic and racial minorities differed. Twenty-two percent of the sample recruited for the study conducted by Press et al. was African American \( (n = 54) \) and 25.2% were Native American \( (n = 62) \), whereas 7.2% of the sample recruited for the study conducted by Durfy et al. was African American \( (n = 39) \), 1.5% was Hispanic \( (n = 8) \), 1.1% was Asian or Pacific Islander \( (n = 6) \), and 1.8% was Other \( (n = 10) \).

State or county cancer registries were used to identify subjects in two studies \( (13.3\%; \text{ refs. 35, 49}) \); however, these studies differed in the strategies that were used to recruit subjects. For example, in the study conducted Anton-Culver et al. \( (49) \), women affected with breast and ovarian cancer were identified from a county-based cancer registry; following physician notification of intent to contact patients, an introductory letter was mailed to potential subjects. After mailing the introductory letter, potential subjects were contacted by telephone for the study invitation and to complete study measures. However, in the study conducted by Newman et al. \( (35) \), nurses completed home visits to collect data after potential subjects were identified from a state-based registry.

As shown in Table 2, similar response rates were reported in the studies that used state or county registries to identify potential subjects \( (35, 49) \); however, the representation of ethnic and racial minorities differed in these studies. For example, 6.9% of the sample in the study conducted by Anton-Culver et al. was Hispanic \( (n = 54) \), 5% was Asian \( (n = 40) \), and 0.5% was African American \( (n = 4) \). Hispanics were not enrolled in the study conducted by Newman et al.; however, 42% of cases were African American \( (n = 89) \) and 1% was Native American \( (n = 2) \). Among controls, 42% was African American \( (n = 79) \), 1% was Asian \( (n = 2) \), and 1% was Other \( (n = 2) \).

Discussion

This review described the sources and strategies that have been used to identify and recruit ethnic and racial minorities to participate in hereditary breast cancer research. Overall, hospital-based resources were used most frequently to identify potential subjects. Several different factors are likely to be considered when making decisions about recruitment strategies (e.g., cost, staff time, and recruitment success). Because effective and efficient methods for subject recruitment are critical to achieving study goals, investigators are likely to select resources from which potential subjects can be identified efficiently at minimal cost. Hospital resources contain demographic and clinical information that can facilitate identification of potential subjects without expenditure of significant resources. This may explain why hospital-based resources were used to identify potential subjects in most of the studies reviewed in this report. The goals of the studies may have also influenced the resources that were used to identify potential subjects. More than half of the studies included in this review were designed to evaluate the prevalence of \( BRCA1/2 \) genes among women with a personal history of cancer or to assess knowledge and
attitudes about genetic testing among women with a family history of breast cancer. Identifying women affected with cancer at medical centers where they have been diagnosed and/or treated is likely to be the most efficient approach for identifying potential subjects with a personal history of cancer or a specific risk profile.

While hospital-based resources have several practical advantages for identifying potential subjects to participate in cancer genetics research, an obvious limitation of this resource is that only individuals who are patients at the medical facility will be maintained in the clinic databases and tumor registries at these facilities. Only these individuals have the potential to be made aware about the availability of research studies. Data from national surveys have shown that ethnic and racial minorities are less likely than Caucasians to have private health insurance or have a usual source of care and are more likely to use emergency departments as their usual source of medical care (50-52). Thus, these groups may be underrepresented in the clinic databases and tumor registries at academic medical facilities where research on genetic risk factors for cancer is likely to be conducted. Previous research has shown that, compared with a cancer center tumor registry, a greater number of ethnically diverse women affected with cancer were identified from a tumor registry at a community-based hospital to participate in survivorship research (53). This finding suggests that developing partnerships with community-based hospitals may be one way to increase awareness of and access to cancer genetics research among ethnic and racial minorities. In this report, four of the studies that used hospital clinics to identify potential subjects were conducted at minority medical institutions (n = 2; refs. 41, 42) or in partnership with a community medical facility (n = 2; refs. 37, 40); the number of African American women enrolled in these studies ranged from 43 to 137, and response rates ranged from 54% to 71%.

Thus, even when cancer genetics research is conducted at community hospitals that may have a greater number of ethnic and racial minority patients, the number of individuals who enroll in studies may still vary. Regardless of the settings in which these studies are conducted, it is important for investigators to use recruitment strategies that are effective among ethnic and racial minority groups.

In terms of recruitment approaches, active methods such as in-person recruitment from clinic populations and recruitment by mail or telephone were used most frequently; the representation of ethnic and racial minority subjects was lower in studies that used impersonal recruitment methods. For example, 49% of the sample in the study conducted by Donovan and Tucker (38), which used in-person clinic recruitment, were African American, whereas 18% of the sample in the study conducted by Benkendorf et al. (40), which used a combination of active and passive recruitment strategies, were African American. This suggests that personalized recruitment approaches might be more efficacious for recruiting ethnic and racial minority groups to participate in cancer genetics research. Previous research has shown that, compared with Caucasian breast cancer patients, African American breast cancer patients who were identified from a hospital tumor registry were less likely to enroll in a quality of life study when recruited to participate in the study by mail (30). Thus, active, personalized recruitment methods are possibly effective strategies for recruiting ethnic and racial minorities to participate in cancer genetics research.

Advantages of active recruitment strategies include enrollment of a more generalizable study sample and less chance for self-selection bias (34). Active strategies that use personal approaches may also enhance awareness about cancer genetics research among ethnic and racial minorities by increasing understanding about the importance of the research and by providing potential subjects with an opportunity to ask questions about the procedures involved in study participation (33). Personalized approaches may also minimize mistrust of the research process, which is a barrier to ethnic and racial minority participation in research conducted in other areas (19, 20, 26). However, the effectiveness of active, personalized recruitment methods is likely to depend on whether the recruiter and potential subject are from the same ethnic or racial background. Moorman et al. (18) found that, among women who were eligible to participate in a case-control epidemiologic study of breast cancer, African American women were less likely to complete the interview when they were invited to complete the survey by a non–African American interviewer. In this review, the studies that used personalized recruitment methods did not indicate whether potential subjects and recruiters were concordant in terms of race or ethnic background. Thus, additional research is needed to determine whether the race or ethnicity of the interviewer has an effect on decisions to participate in cancer genetics research among ethnic and racial minority groups.

Although the racial or ethnic concordance between potential subjects and study recruiters may influence the effectiveness of active, personalized recruitment approaches, the yield of recruitment sources and methods may also vary depending on the study population, eligibility requirements, and study procedures. We observed that the number of racial and ethnic minorities decreased as the complexity of study participation increased. For example, although the studies conducted by Lipkus et al. (39) and Thompson et al. (37) both used active recruitment strategies and had similar eligibility requirements, enrollment rates were lower in the study conducted by Thompson et al. (54%) relative to the study conducted by Lipkus et al. (86%). It is possible that, differences in enrollment rates for these studies may have been due to the procedures involved in participation. In the Lipkus et al. study, subjects had to complete a telephone interview and agree to participate in an education program about breast cancer risk factors and mammography, whereas subjects in the Thompson et al. study had to complete psychologic measures and consider participating in genetic counseling and testing. A recent study found that African Americans were most willing to participate in studies that did not involve invasive procedures (54). Thus, knowledge of the role of the complexity of the study design and burden would be critical in attempts to enhance ethnic and racial minority participation in cancer genetics research. Less knowledge about hereditary breast cancer and exposure to information about
genetic testing for inherited cancer risk (55) may contribute to greater concerns about genetic testing procedures among African Americans. Therefore, when implementing personalized approaches, it may also be important to consider the target population’s level of exposure to new medical procedures and to identify potential barriers (e.g., cost of medical procedures and access to study sites) so that strategies are designed to address these issues.

Prior studies have shown that community outreach may be effective resources for identifying ethnic and racial minority groups for participation in cancer prevention and control research (29, 31); however, only two studies included in this review used this resource to identify potential subjects. In both of the studies that used community outreach, potential subjects were recruited by self-referrals from mass media. Both of these studies were also conducted in the Pacific Northwest (Seattle, WA); however, the study conducted by Durfy et al. (47) included a smaller number of racial and ethnic minorities compared with the study conducted by Press et al. (48). This may have been due to differences in the sampling strategies used in these studies. The study conducted by Press et al. used a specific sampling frame to identify potential subjects from diverse ethnic backgrounds, while the study conducted by Durfy et al. used a general approach. The different enrollment rates for ethnic and racial minority groups in these studies suggest that, even in geographic areas that are not densely populated with ethnic and racial minorities, diverse study samples can be recruited if sampling plans are designed to achieve this objective.

Moreover, living in a geographic area with a large proportion of ethnic and racial minorities does not guarantee that study samples will be ethnically or racially diverse. Although North Carolina is one of the top 10 states with the highest number of Black residents, African American women were oversampled in the study conducted by Newman et al. to ensure sufficient representation of this population (35). Thus, regardless of the geographic location for study sites, planning, additional time, and extra effort may be needed to recruit ethnic and racial minorities for participation in cancer genetics studies.

Although this is the first report to review recruitment of ethnic and racial minorities for participation in research on hereditary breast cancer, some limitations should be considered. First, we limited our review to ethnic and racial minority participation in research on breast cancer susceptibility and genetic testing. Additional research is needed to describe ethnic and racial minority participation in research on other hereditary cancer conditions. An additional potential limitation is that we may not have identified all studies that included ethnic and racial minorities with our search strategy. However, we used a structured search method to identify studies on breast cancer susceptibility and genetic testing that included ethnic and racial minority subjects. A final limitation is that we conducted a descriptive, not a quantitative, review of recruitment methods and messages would provide important information on the strategies that are most and least effective for recruiting ethnic and racial minorities to participate in research on breast cancer susceptibility and genetic testing and the yield of subjects using these methods.

It is important to emphasize that decisions not to enroll in a study are to be respected in all individuals; however, lack of awareness about available studies decreases decision making about study participation. One issue emerging from this report is that evaluation components should be incorporated into recruitment methods used in cancer genetics research studies. It was not possible to determine which methods were most and least effective for recruiting ethnic and racial minority groups because response rates were not reported consistently. In addition, for the studies that used hospital-based resources to identify potential subjects, it could be argued that the proportion of ethnic and racial minorities who were enrolled in these studies were representative of the number of ethnic and racial minority patients who receive care at these centers. However, the proportion of ethnic and racial minority groups represented in these resources was not reported in the studies included in this review. As increased attention is focused on the inclusion of ethnic and racial minorities in NIH-funded research (56), it will also be important to establish guidelines that can be used to determine adequate representation of ethnic and racial minorities in cancer genetics research. Previous research has based adequate representation of ethnic and racial minorities in cancer treatment clinical trials on the incidence of disease in these populations (57); however, this approach may not be reasonable to apply in cancer genetics research, where information about the prevalence of susceptibility genes among ethnic and racial minorities may be based on a limited number of studies with small samples (42-44). In addition to more specific evaluations of recruitment methods and representativeness of study samples, future research should examine reasons for refusing to participate in cancer genetics research as well as retention of ethnic and racial minorities in these studies. Observational studies would be useful to determine the most effective settings (e.g., academic medical center and community hospitals) for identifying potential subjects, while experimental study designs that compare the effects of different recruitment methods and messages would provide important information on the strategies that are most and least effective for recruiting ethnic and racial minorities to participate in cancer genetics research.

References


