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ORIGINAL ARTICLE

Engaging rural communities in genetic research: challenges and opportunities

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Abstract Statistical analyses of health and disease in rural communities is frequently limited by low sample counts. Still, some studies indicate increased risk for some diseases even after adjustment for known risk factors. It has been hypothesized that the context of community formation in rural areas facilitates the propagation of genetic founder effects—potentially impacting disease susceptibility. However, outright examination of genetic diversity in such communities has not been performed. Our objective was to engage otherwise research-inexperienced rural communities of largely European descent in genomic research in the context of cancer susceptibility. From September 2015 to February 2016, we implemented a systematic process of progressive community engagement. This iterative method sought project buy-in from first the town mayor, then village council. If approved by both, a focus group of community members examined how residents might view the research, informed consent and specimen collection, and issues of privacy. We were successful in engaging three of the four communities approached for the research project. There was universal enthusiasm for the project by all mayors and village councils. The focus groups' main point of discussion involved wording in the informed consent, with little concern regarding the research question or privacy. Perhaps contrary to popular thought, we found each community we approached to be both welcoming and enthusiastic about collaborating in research on genomic diversity. The systematic method of engagement did much to preserve community respect and autonomy and facilitated buy-in.

Keywords Rural · Genetics · Cancer · Focus groups

Introduction

Studies of disease in rural areas are difficult to conduct and interpret due to diminished population/sample size and poorly captured heterogeneity of exposure/risk. Still, some studies show that rural residents may experience decreased life expectancy and increased all-cause and disease-specific mortality, particularly with respect to some cancers, compared to urban

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residents (Singh and Siahpush 2014a, b; LeVault et al. 2014; Colli et al. 2009; Cole et al. 2012; Colleran et al. 2007; Singh 2012; Fogleman et al. 2015). Some incidence and mortality differences may be attributed to the increased prevalence of smoking, obesity, and alcohol use in many rural areas (Befort et al. 2012; Doescher et al. 2006; Jackson et al. 2005, 2006). Alternatively, the history and geography of many rural American communities are suitable for an underlying genetic heterogeneity in disease susceptibility to arise due to possible founder effects and relative geographic isolation (Jenkins et al. 2016). Might it be possible for some rural communities to have underlying differences in their genetic susceptibility to some diseases?

Population-level cancer disparities have been recognized for well over 100 years and have been characterized by subpopulations such as age, race, and gender (Smuckler 1983; Gehlert and Colditz 2011). There remain specific identified populations about which less is known but who are observed to experience increased cancer risk. Examples include residents of rural Maine of European descent who experience some cancer incidence and mortality exceeding that of African Americans (a recognized high-risk population; Hock et al. 2012; Zeng et al. 2015; Coughlin et al. 2014). Furthermore, a population-level risk may significantly vary across region and tribe as experienced by American Indians/Alaskan Natives (Campbell et al. 2014; Wiggins et al. 2008). The intricacies of cancer incidence rates between European-American rural communities warrant further study. Many experience increased prevalence of behaviors such as smoking, alcohol use, and inactivity which may exacerbate cancer risk due to genetic predispositions (Hock et al. 2012; Weaver et al. 2013). While some researchers report that rural populations experience greater incidence of some cancers, these data are inconsistent (McLafferty and Wang 2009; Singh 2012). Accounting for known risk factors does not always explain differences in rural/urban disease risk, but as the risk is inconsistent across studies and diseases might their be unrecognized local contributing factors?

Previous community-based genetic studies in the USA have been broadly population-based or primarily focused on racial and ethnic minorities, including African-American communities and Native American tribes, but there is little research that has been done looking at isolated rural populations of European descent (Foster et al. 1999; Terry et al. 2012; Rotimi et al. 2007; Skinner et al. 2015). Our group has hypothesized that the nature of the founding of many rural communities in the USA may have facilitated the development of genetic founder effects which may contribute to underlying disease risk variability (Jenkins et al. 2016). To determine if this hypothesis was viable to test, we devised a feasibility study that set out to accomplish three aims: (1) determine if rural communities can be successfully engaged in basic genetic research, (2) provide preliminary data regarding whole-genome variability between rural and urban communities, and (3) acquire baseline data by survey regarding rural residents' knowledge and perceptions regarding genetic research in general. These aims are novel as not only are rural residents underrepresented in research broadly speaking (e.g., UyBico et al. 2007; Bennett 2013), but there is no literature examining genetic diversity in rural US populations or engaging rural communities in such work.

While our primary aim was to test our hypothesis, we also decided that adopting the principles of community-engaged research would serve to successfully engage lay individuals while navigating the evolving role of ethics and community participation in research (Bromley et al. 2015). Respect for community is a necessary principle of genetic research, as it ensures that communities' and individuals' dignity, interests, and rights are being protected (Jones et al. 2014). Performing genetic research *in partnership with* the community instead of performing genetic research *on* a community allows for this principle to be incorporated. This approach can also help researchers better appreciate the perspectives of the community (Rotimi et al. 2007). Collaborative research with the community often includes the incorporation of a community advisory board, which can help develop equitable partnerships to ensure trust and respect. Community advisory boards often include membership of community leaders and representatives from community organizations. However, Foster et al.'s study has shown that to better identify and address culturally specific risks, it is good to engage and consider the concerns of community members outside of community leaders (1999).

One way to obtain more complete community input and feedback about proposed genetic research is to conduct focus groups (Makowsky Daley et al. 2010). Focus groups allow for participants to provide open-ended feedback and an additional avenue for community engagement and establishing a foundation of trust that can facilitate genetic research, as the community is often engaged in this qualitative research not only as participants but as "recruiters" of participants. Including the community in the recruitment process has been effective for both community-based focus groups and genetic research (Cristancho et al. 2008; Skinner et al. 2015). Focus group findings have provided researchers with better information how to better consider community concerns, recruit research participants, and disseminate research findings in genetic research (Rotimi et al. 2007; Terry et al. 2012).

Our objective here is to describe how we engaged rural communities in the conductance of genetic research, their openness and willingness to participate, and concerns and opportunities revealed in the process.

Methods

The goal of this work is to describe how we engaged communities and individuals for participating in the larger feasibility

study examining genetic diversity among rural populations. To achieve this goal, we sought to engage three rural communities to participate in this study with the aim of collecting 50 saliva samples from each. Following inclusion criteria from a similar study, communities had to have population of approximately 1500 residents and be located at least 20 mi from a town with a population greater than 5000 (Portas et al. 2010).

As a first step in the research process, we sought to engage the community to achieve two key initial goals: (1) to facilitate "buy-in" and develop trust with the community and (2) to ensure that the overall research process, broadly speaking, and the study's informed consent form, specifically, assured protection of participant dignity, autonomy, and integrity of community participants.

Community engagement process Community engagement was a multi-step process that began with the study's principal investigator (PI; Jenkins) initially contacting each community's mayor via email or telephone. After a brief conversation describing the overall nature of the project, the PI had a face-to-face meeting with the mayor to discuss the project in detail and answer questions. If the mayor decided that the project was potentially a good fit and of interest to the community, the PI then presented the project to the village council at a regularly scheduled meeting. After hearing the presentation, asking questions, and expressing any potential concerns, the council would then decide if the project was of potential interest to the community. After buy-in from the mayor and village council, there were two additional steps: selection of local champions and the conductance of a local focus group. This process helped to ensure that the study was acceptable to community leaders and other community members.

Recruitment process Local champions were identified by each town's village council or mayor as individuals who would be enthusiastic champions of the project and assist with generating community support and involvement. As this was an unfunded feasibility study, such individuals were volunteers for their time and efforts and there was no means for compensation. Research coordinators contacted local champions by email and telephone to inform them of the purpose of the focus groups, their role, and the selection criteria for participants. The role of the local champions was to: (a) aid in recruiting community members for the focus groups and coordinate a location and time for a focus group meeting, and (b) assist with project promotion and advertisement within the community and coordinating locations and times for participant recruitment for the larger feasibility sample collection study.

Focus group structure We performed one focus group in each of the three communities. Focus group participant inclusion criteria included age ≥18 years and current residence in the community. The focus groups were held in locations identified by each community's local champions and were easily accessible by community members (e.g., community halls and churches). Each focus group consisted of a written survey on participants' perception of genetic research (see below; to be used as baseline data for further studies) and a semi-structured discussion to explore the participants' thoughts regarding the components of the informed consent form and perceived enthusiasm and concerns of the community regarding their recruitment and participation in genetic research. Details of the focus group procedure can be found in Table 1. The focus group sessions began with participants providing their informed consent to participate in the discussion and to the audiotaping and note-taking of the sessions.

Focus group survey Participants completed a 10-min written survey to obtain their perception of genetic research. The development of survey items was informed by the Health Belief Model (Strecher and Rosenstock 1997), a model utilized in previous studies to assess perception and barriers to participating in genetic research (Cyr et al. 2010; Taylor et al. 2013). The survey contained four sections (Tables 2 and 3), which included participants' demographics, knowledge, attitude and beliefs, and barriers to participating in genetic testing. Face validity of the instrument was determined by having two genetic research experts review the instrument.

- *Demographics*. The instrument captured information for the demographic variables of gender, age (years), education level (categorized), and current employment (open ended).
- *Knowledge*. Two of the 22-item survey assessed participants' knowledge of genetic testing. Both questions allowed for a yes = 1 or no = 0 response.
- *Attitudes and beliefs*. Nine items were used to measure participants' attitudes and beliefs toward genetic testing. Two of the items had a yes = 1 or no = 0 response, and seven items utilized a 5-point Likert-type scale.
- *Barriers*. Eleven items were used to identify barriers to participating in genetic testing. These items were rated with a 3-point Likert-type scale.

Focus group discussion Focus groups were roughly 90 min in length and led by a facilitator who used a semi-structured script for discussion. This semi-structured script included a series of open-ended questions to obtain participants' perspective on the following: (1) consent verbiage and assurance of protection of participant dignity, autonomy, and integrity; (2) strategies for communication of project knowledge and enthusiasm; (3) worth and acceptability of genetic research to the community; (4) perceived barriers to genetic research including stigmatization or mistrust and how such barriers might be

Table 1 Focus group session procedure and content description

Steps	Content description
1	Completed written survey regarding perception of genetic research
2	Discussed genetic research consent form Consent form clearly states: • Objective of the study • Risk of participating • Sharing and privacy of information collected
3	Discussed engaging rural communities Worth and acceptability of genetic research • Openness of community members to genetic research • Community members perception of benefits of genetic research • Personal perception of genetic research Perceived barriers to community participation • Barriers of community members becoming involved in genetic research Communicating project knowledge and community enthusiasm • Effective approaches to recruiting participants • Strategies to creating enthusiasm for a genetic study • Strategies to identifying and recruiting community members to advocate genetic research

addressed. To guide discussion, participants were given an Institutional Review Board (IRB)-approved informed consent form. The consent form detailed the participant requirements: (1) provision of a saliva sample, (2) completion of a genealogy log, and (3) an optional survey. Questions regarding the consent form pertained to the form's ability to clearly illustrate the objective of the study, participants' risk of participating in the study, and the storing and accessing of participant information to ensure privacy. Following the discussion of the IRB consent form, participants were asked questions regarding approaches to increasing awareness, knowledge, and enthusiasm about genetic research projects. The concluding questions provided insight into perceived barriers to participating in genetic research. These groups were audio taped, and one to two note-takers were present.

Table 2 Demographic characteristics of focus group participants, $N = 30$

Variable	*N* (%)
Age, years mean, SD	62 (17.0)
Gender	
Male	8 (26.7)
Female	22 (73.3)
Education level	
Less than high school	1 (3.3)
High school	3 (10.0)
Some college	10 (33.3)
Associate's	4 (13.3)
Bachelor's	8 (26.7)
Master'	2 (6. 7)
Professional	1 (3.3)
Doctoral	1 (3.3)
Employment status	
Employed	17 (56.7)
Retired	13 (43.3)

Statistical analysis plan Descriptive statistics were used to quantify sample characteristics and the responses to survey questions. These analyses were conducted with STATA version 14 (StataCorp 2015). The planned analysis for the focus groups included framework analysis approach where multiple coders would be involved in identifying themes, developing codes, and utilizing multiple coders to code comments until an inter-rater reliability of 90% or higher was achieved. Ultimately, discussion analyses were not performed as planned (see "Results" section).

Results

Community engagement Three towns were identified for initial engagement (Towns 1–3). The PI sent introductory emails to the mayors in September 2015. Following the initial email, the mayors were telephoned to describe the nature of the project and to schedule face-to-face meetings. All three mayors expressed enthusiasm with the project and scheduled village council meetings. Again, there was universal enthusiasm for the project and all councils voted to pursue the project. The first challenge came when each town had to identify 1+ local champions to locally coordinate activities. Towns 1 and 2 were able to identify such individuals, but Town 3 was unable to do so. Local champions identified by the community mayors included individuals deeply embedded with the community and who had a personal interest in improving the health of their communities, including a local nurse, emergency medical technician, a cancer survivor, and a county clerk. After 4 months, Town 3 was discontinued from the project and Town 4 approached. We had the same level of enthusiasm here as the previous three, and the council was able to identify local champions. Thus, Towns 1, 2, and 4 participated through the project's duration, which we have respectively coded as Towns A, B, and C while discussing the results.

Focus group recruitment sources/process Initial contacts with local champions began October 2015 and continued through February 2016. The local champions developed a focus group recruitment approach they thought was most suitable for their communities. Methods utilized included social media accounts such as Facebook, published news articles in local newspapers, and individual invitations. Some local champions thought that community members who had a medical background (e.g., nurses) would be ideal focus group participants. The research team emphasized that, while any

Table 3 Participants' perception of genetic research, $N = 30$

Survey items	Total *n* (%)
Knowledge	
Have you heard about genetic screening for certain cancers such as breast (BRCA testing)?	
Yes	22 (73.3)
No	8 (26.7)
Have you heard about genetic screening for other adult onset potentially deadly diseases such as Huntington's disease?	
Yes	12 (40.0)
No	18 (60.0)
Attitude and beliefs	
If you were genetically tested for cancer or other diseases, would you want to know the results?	
Yes	25 (86.2)
No	4 (13.8)
If you were to get genetic testing results that said you were at low risk for a disease, how likely would you be to share the results with your family?	
Very likely	19 (63.3)
Somewhat likely	7 (23.3)
Unsure	4 (13.3)
Somewhat unlikely	0
Very unlikely	0
If you were to get genetic testing results that said you were at high risk for a disease, how likely would you be to share the results with your family?	
Very likely	17 (56.7)
Somewhat likely	3 (10.0)
Unsure	10 (33.3)
Somewhat unlikely	0
Very unlikely	0
Do you think having a positive result to a genetic test guarantees you will get cancer or that disease?	
Yes	4 (13.3)
No	26 (86.7)
Do you think knowing you are at high risk for cancer or other potentially deadly disease would influence your decision to have children?	
Very likely	2 (7.14)
Somewhat likely	7 (25.0)
Unsure	11 (39.3)
Somewhat unlikely	2 (7.14)
Very unlikely	6 (21.4)
Do you think getting a positive result would get you to change your lifestyle, for example quit smoking or eat better?	
Very likely	17 (56.7)
Somewhat likely	13 (43.3)
Unsure	0
Somewhat unlikely	0
Very unlikely	0
Do you think recent advances in genetics such as screening and personalized medicine are generally good or bad?	
Very good	16 (53.3)
Somewhat good	8 (26.7)
Unsure	6 (20.0)
Somewhat bad	0
Very bad	0
Are you afraid scientists will go "too far" with genetic advances (e.g., designer babies or cloning people)?	
Very afraid	2 (6.67)
Somewhat afraid	12 (40.0)
Unsure	9 (30.0)
Not afraid	4 (13.3)
Not afraid at all	3 (10.0)
Do you think celebrities and the media influence public perception of genetic testing?	
A lot	7 (23.3)
Somewhat	19 (63.3)
Unsure	4 (13.3)
Very little	0
Not at all	0
Barriers	
How would you rate each of the following sentences as reasons why you would not get genetic testing	
Afraid of what results would say:	
Major reason	4 (13.3)
Minor reason	12 (40.0)
Not a reason	14 (46.7)
Afraid your insurance company would know the results:	
Major reason	5 (16.7)
Minor reason	12 (40.0)
Not a reason	13 (43.3)
Afraid of telling your family:	
Major reason	0
Minor reason	10 (33.3)
Not a reason	20 (66.7)
Afraid of being fired from your job:	
Major reason	1 (3.33)
Minor reason	3 (10.0)
Not a reason	26 (86.7)
Afraid you couldn't do anything about it (doomed feeling):	
Major reason	6 (20.00)
Minor reason	10 (33.3)
Not a reason	14 (46.7)
Afraid you doctor will not keep the results private:	
Major reason	1 (3.33)
Minor reason	7 (23.3)
Not a reason	22 (73.3)
Did not know it was an option:	
Major reason	3 (10.0)
Minor reason	6 (20.0)

Table 3 (continued)

Survey items	Total *n* (%)
Not a reason	21 (70.0)
The cost:	
Major reason	11 (36.7)
Minor reason	14 (46.7)
Not a reason	5 (16.7)
Not offered at a clinic near me:	
Major reason	9 (30.0)
Minor reason	12 (43.3)
Not a reason	9 (26.7)
No genetic counselor nearby to discuss results:	
Major reason	9 (30.0)
Minor reason	13 (43.3)
Not a reason	8 (26.7)
No time:	
Major reason	2 (6.90)
Minor reason	7 (24.1)
Not a reason	20 (69.0)

community members with a health background and interest would be welcomed to participate, focus group participants who were representative of the demographic and educational composition of the community were encouraged. Local champions expressed challenges in recruiting community members for the focus groups and identifying times that would be convenient for optimal participation. Barriers stated by volunteers included scheduling conflicts with the town's sporting events and holiday activities and identifying community members that reside within the town's borders. This challenge was associated with volunteers' definition of place of residence in which some identified their place of residence as their county, whereas others identified their town as their place of residence.

Focus group participants Focus groups were performed from January 2016 through March 2016. A total of 30 individuals participated in the three focus groups, with group sizes ranging from 5 to 14 members (Table 1). The average age of participants was 62 years. Approximately 73% of participants were female. For education level, the largest percentage of participants had some college (33%), followed by having a bachelor's degree (27%). More than half of the participants are currently employed (57%). Employed participants occupations included teacher, florist, farmer, registered nurse, paramedic, mayor, and village clerk.

Survey results Across the three towns, 73% participants had heard of *BRCA* testing, whereas only 40% had heard of genetic screening for other adult-onset deadly diseases such as Huntington's disease (Table 3). In regards to disclosing diagnosis of high risk for a disease to family, 56.7% of respondents responded very likely, 10.0% responded somewhat likely, and 33.3% responded unsure. Responses varied for how being diagnosed with a high-risk disease would impact their decision to have children, with 7.14% of respondents reporting very likely, 25.0% somewhat likely, 39.3% were unsure, 7.14% somewhat unlikely, and 21.4% very unlikely. There was better agreement on the recent advances in genetics such as screening and personalized medicine with 53.3% reporting that the recent advances in genetics were seen as very good, followed by 26.7% reporting somewhat good, and 20.0% reporting unsure. Across the three towns, 13.3% of participants reported fear of results as a major reason for not participating in genetic testing, followed by 40.0% participants stating it as a minor reason, and 46.7% participants stated it was not a reason. Cost was identified as the largest barrier with 36.7% of respondents reporting it as major reason, 46.7% stated it was a minor reason, and 16.7% reported that it was not a reason. Access to a clinic offering genetic testing or a genetic counselor were the second largest barriers identified. Thirty percent of participants reported not having a clinic that offers genetic testing as a major reason, followed by 43.3% reporting it as a minor reason, and 26.7% reporting it as not a reason. Similarly, for no genetic counselor nearby, 30.0% reported it as a major reason, 43.3% reported it as a minor reason, and 26.7% reported it as not a reason.

Discussion results—informed consent The focus group sessions were not analyzed as planned. Several participants at the beginning of the sessions reported that their local champions informed them that the focus group session was an information session about genetic research. As a result, during the sessions, participants inquired about genetic research, which diverted the discussion from the planned questions. Because of this and in keeping with the spirit of ensuring community trust and engendering community buy-in, the facilitator allowed for an open discussion of the project in addition to facilitating discussion around the semi-structured script questions. Thus, the content of the focus group discussion was not as centered on the intended questions as anticipated, making it difficult to analyze using proposed qualitative methods. Instead, focus groups notes and audio tapes were reviewed thoroughly by study coordinators to identify key comments and common concepts from the sessions.

In regards to the IRB consent form, a majority of focus group members agreed that community members would experience challenges in comprehending components of the consent form due to their reading level. A specific participant from Town A highlighted that many individuals in their community have a third-grade reading level; therefore, their lack of understanding of the consent form may be a barrier to participating in a genetic research study. Participants also in

reading the objective requested background information about genetics, and how and when genetic mutations occur. Additionally, clarifying the amount of genealogy information (e.g., how much family history needed) is also important to discuss in the objective of the consent form. Additional information regarding research findings was requested. For instance, participants want clarification on obtaining individual results from the study.

In discussing the risk of participating in genetic research, participants wanted clarity on the level of risk. More specifically, participants in two of the three towns wanted to know the effect of the Health Insurance Portability and Accountability Act on genetic research studies and their role in protecting their information. Additionally, participants in two of the three towns inquired about the length of time their information will be stored and requested more detail on the official start date. Although the consent form provided detailed information regarding their risk levels, some participants stated that they were still uncertain about their level of risk and were fearful of unanticipated harm. Additionally, a participant expressed concern that unauthorized individuals may hack the password and access the information. Thus, it is important to delineate the level of security of the environment.

Some focus group participants indicated that they wanted detailed information, names and contact information, of individuals that will be able to access their information. On the other hand, other members were not concerned about sharing and privacy, as the information would coincide with information in their medical records.

Discussion results—genetic research involvement and perceptions Members of Town A were open to participating in genetic research and believe that other community members would share their interest. Several members of Town A reported that they have previously participated in a genetic research study and perceived genetic research to be beneficial to improve health outcomes. However, members of Town B and C expressed concerns in participating in genetic research. The concerns were associated with the use and sharing of their information. Participants expressed concerns about findings creating challenges in accessing health insurance. The sharing of the genetic information also influenced their perception of the benefits of participating in genetic research. Nonetheless, members of three towns stated that communicating the purpose and benefits to the community would increase acceptability.

Discussion results—barriers Focus group participants identified multiple potential barriers to participation in genetic research. Focus group participants indicated that the sharing of information was a perceived barrier and reiterated the importance of communicating who would have access to the information and related privacy laws. Another barrier was lack of awareness of a research study being conducted due to potentially limited advertising efforts. Thus, participants highlighted the importance of utilizing multiple sources (e.g., social media and news articles) to inform the community of the study. Additionally, fear of results was reported as a barrier for participants from two of the towns. These participants indicated that the preconception that genetic studies would provide results of specimen analysis indicative of health risk may make participants uncomfortable, thus precluding involvement. Another potential barrier is completing necessary forms for research participation, as one participant stated that this process could create anxiety.

Participants' responses to approaches to recruiting community members and creating enthusiasm coincided. Participants indicated that information about the study should be publicized in the local newspaper, social media accounts, and the town's website, and posters/flyers should be hung around the community. Having a champion within the community was reported as an effective approach by all three communities. Publicizing the study during community meetings and local events was mentioned as another potential approach to increasing awareness and enthusiasm.

Discussion

We were able to successfully engage three rural communities to participate in this study. Through the initial engagement described in this paper, we identified local leaders and champions to facilitate the recruitment and conduct of focus groups in each community. These focus groups allowed for us to further secure community buy-in, get community input on the consent form, and address concerns on the conduct of genetic research in their communities. Focus group participants identified multiple concerns about the informed consent forms, including literacy level and use of their genetic data. Additionally, focus group findings also suggested that although participants had concerns, they still thought that their communities would be agreeable to participation in such research.

The method of approaching each potential partner community through its elected leadership proved effective and demonstrated feasibility of the process for future, funded studies. All four communities approached expressed considerable enthusiasm and support for the project. Rather than expressing concern over possible stigmatization, the most commonly expressed feeling toward the project was a desire to help in the fight against cancer, even in this basic way. Identifying local champions and having volunteers publicize focus group sessions on social media, in the local newspaper, and through posters were effective approaches for recruiting focus group participants. Utilizing these communication channels assisted

in meeting the focus group session size goals and to obtain a diverse group of participants (e.g., age, education level, and employment status). Additionally, similar to previous study, it facilitated community buy-in by engaging more than just community leaders (Foster et al. 1999).

Focus group discussions did not proceed as planned due to variable participant expectations. While we were unable to perform qualitative analyses from the discussion transcripts, some common concepts were identified. These included potential participants wanting more information on the background of genetics and on how their personal information would be kept private. Findings regarding the concerns about privacy were not surprising, as previous studies have identified privacy as a major concern (Kaufman et al. 2009). They also wanted to make sure that the reading level was compatible with that of their community members. Most participants were also concerned about knowing all the information for the genealogy log.

From the genetic research perception survey, some of the major findings were that rural community members appear to understand that genetic testing varies by disease. More than half of participants were knowledgeable of *BRCA* testing, whereas 60% of participants had never heard of the genetic testing for adult-onset potentially deadly diseases such as Huntington's disease. Moreover, it can be speculated that the higher knowledge of *BRCA* testing compared to other adult-onset diseases such as Huntington's disease testing is related to individuals having a family history of breast cancer and/or ovarian cancer as these family members are recommended to receive testing (Mai et al. 2014) An additional reason for higher knowledge of *BRCA* testing is direct-to-consumer (DTC) advertising. Several DTC campaigns for cancer-related genetic tests were launched in the past decade (Mai et al. 2014). In regards to attitudes and beliefs, an interesting finding is one third of respondents reported that they were unsure if they would inform their family of findings of high risk of a disease with their family. This result is consistent with literature that report individuals find disclosing their diagnosis as challenging (Ewing et al. 2016) and express worry and concern about the reaction (Hilton et al. 2009). Another interesting finding was that many (~40%) participants reported they were unsure if knowing they were at high risk of developing any deadly disease would influence their decision to have children. This finding may reflect the mean age of participants, 62 years. Thus, a majority of participants are past the decision-stage of raising a family.

Lessons learned

Opportunities to improve communication with communities Providing mayors and local champions with a written lay summary of the study to disseminate to community members may have aided in recruiting participants for focus groups and eliminated confusion regarding the objective of the focus group sessions. Similarly, some local champions and focus group participants alike indicated that they thought that the study was intended to assess increased cancer risk in rural areas. While identification of genetic homogeneity through this feasibility study may provide the foundation for future study on genetic relationships to cancer risk in rural areas, that itself was not the purpose this study. Thus, a lesson learned from this study is that the research team should take special care to ensure that the local champions and community members understand the nature and purpose of a feasibility study and to ensure that communities understand the broader health implications of such research, rather than the potential specific health concerns. This is important for the sake of appropriate understanding between researchers and community members which enables the building of trust between researchers and community members. Additionally, appropriate communications to rural populations during the continuum of the research process may help facilitate buy-in, as rural populations may be more motivated to be involved if researcher engagement with a community is framed as an opportunity to solve an important community problem, rather than solely an opportunity to participate in research (Martin et al. 2016).

Consistency among local champions Another lesson to take forward to larger and funded studies is that engagement of community members in the continuum of the research process may serve to both focus research and provide critical results interpretation. While utilizing local champions helped facilitate the recruitment of participants, it was clear that focus group participants came to the meetings with differing ideas regarding the meeting's purpose and their role (thus limiting the ability for consistent data collection across groups). While the research team surmised that this was likely due to differing understanding and description by the local champions, the limited resources available to this feasibility study did not allow for the champions themselves to be either trained in their expected roles of facilitator and recruiter or be an integral part of the results analysis and interpretation process. We therefore cannot adequately describe any biases in presentation or project "selling" that may have occurred as the individual champions approached community members. Further work should explicitly examine how to ensure consistency of communication and engagement across multiple communities.

Opportunities to improve community participation The very nature of such community-engaged research is subject at times to the motivations and abilities of the partner community members. Thus, success in local champion recruitment, and advertising and organization of the focus groups and project participants was variable and unique for each community. Ultimately Community 3 was unable to identify any local champions and had to drop out. Nonetheless, providing

mayors with tools and resources, such as written overview of the study, may allay challenges in recruiting local champions. Likewise, the numbers of individuals attending the focus groups and project recruiting venues varied considerably and is likely a reflection of both community-level motivations as well as local champion outreach skills. It should be noted that as this was an unfunded feasibility study, we were unable to provide compensation for the local champions, likely limiting their continued motivation and expended effort. The lack of funding also limited our ability to perform further work, such as modifying and testing newer promotional materials or engaging champions and community members in other aspects of the project such as data analysis and interpretation. We expect that future, funded studies would benefit from the direct compensation of local champions for their time and efforts. In this manner, we might expect greater outreach and coordination among larger proportions of the engaged communities.

Limitations and strengths

This study has several limitations. First and foremost was the reliance upon local champions to advertise and recruit for the focus groups. This is inherently biased toward unique capabilities, time, and spheres of influence of each and is unlikely to yield a truly randomized, representative cross-section of each community. Second, the small sample size for the survey of perceptions of genetic research results prohibits the generalizability of these findings. Third, significant differences in how focus group participants understood the nature of the focus groups led to them being unsuitable for rigorous analysis—therefore perhaps limiting understanding of rural communities' perception of genetic research. These weaknesses may be mitigated in part by the tight-knit structure of such communities where there is greater likelihood for similar life experiences and social norms contributing to greater homogeneity in opinions than more urban areas. This study also has several strengths. For example, participants were recruited from three different communities separated by hundreds of miles, which increases the likelihood that collected responses represent the perceptions of multiple rural communities. Additionally, the community was very engaged in the research process, including developing recruitment strategies.

Conclusion

The outcomes of this study can serve as a guide to engaging rural community members in participating in research. Among such small communities, the great majority of individuals have had friends and/or family members experience cancer. Such personal experiences directly contributed to the near-universal enthusiasm for the project itself and its aims. Beginning with each town's elected officials, we successively and respectfully engaged each community, combining our ideas for the research side of the project to their knowledge and opinions on how best to work within their own community. From this collaboration, we were able to obtain data that are perhaps generalizable (e.g., the great majority of residents think genetic research is beneficial and that genetic analyses showing increased cancer risk would influence their lifestyle) and local specific (e.g., changes in the consent form to reflect the communities education levels and specific means to advertise the project to attract participants). We plan to continue our relationship with these communities, for example by returning to the communities with the results of the analysis and discussing how they may best be presented, and hope that others may be encouraged to begin such collaborations. Rural residents have historically been underrepresented in research, and yet they are frequently an accessible and motivated population. Care must be taken, as with any other study, to maintain attitudes of respect and beneficence as there may be perceptions of "city folks" trying to come to town and "research us." Much of this may be alleviated by our methods of successive engagement and true community partnership. Rural community members believe that genetic research is beneficial. All of the participants reported that receiving positive results would influence their decision to alter their lifestyle, and more than 80% of respondents reported that the advances in genetic research are very or somewhat good.

Compliance with ethical standards All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000(5). Informed consent was obtained from all patients for being included in the study.

Conflict of interest C.D., A.F., W.Z., A.L., R.M., K.D., and W.J. declare that they have no conflict of interest.

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