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ORIGINAL ARTICLE

Perspectives on communication and engagement with regard to collecting biospecimens and family health histories for cancer research in a rural Alaska Native community

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Abstract

Precision medicine initiatives, such as *Cancer Breakthrough 2020,* promise to improve cancer outcomes by tailoring treatment to an individual's genes, environment, and lifestyle. This promise will fall short unless researchers successfully engage diverse communities, including those with histories of medical and research abuse. We examined a rural Alaska Native community's viewpoints about biospecimen collection and storage; interest and recall in reporting family health history; and interest and engagement in biospecimen collection for conducting a genetic test for cancer. In 2014, four focus groups were held with 28 adult Alaska Native rural community members. Thematic analysis was performed after establishing a coding scheme by team consensus. Study participants shared interest in engaging in genetic cancer research and suggested ways to improve community engagement in research. These included transparency and continuous communication with researchers at all stages of the research, clear communication about the intent of the research, and that research and results take into consideration the community's needs. These suggestions may be beneficial for future efforts to expand precision medicine research in Alaska Native communities and similar, diverse populations.

Keywords Ethics, research · Community engagement · Alaska Native people · Genetic research · Precision medicine

Introduction

Since Congress passed the National Cancer Act in 1971, the landscape of cancer research and treatment has changed substantially in the USA (National Cancer Institute 2017; Powel and Seibert 2017; Tiwari and Roy 2012). In the subsequent five decades, increased funding for research has improved knowledge about cancer-related diseases and led to the development of enhanced strategies for prevention, early detection, and treatment (Tiwari and Roy 2012). These developments are associated with many improved health outcomes such as earlier detection of some cancers and better overall survival rates of some cancers (American Cancer Society 2017; Powel and Seibert 2017; Siegel et al. 2016).

Cancer research continues to advance with the January 2016 announcement of *Cancer Breakthrough 2020* (formerly Cancer Moonshot 2020), an initiative focused on finding vaccine-based immunotherapies for cancer (Jenks 2016). Additionally, the 21st Century Cures Act, passed by Congress in December 2016, authorized US$1.8 billion over 7 years, including US$300 million in fiscal year 2017 (Federal Drug Administation 2017). Furthermore, the *All of Us* Research Program (formerly the Precision Medicine Initiative Cohort Program) is a "historic effort to gather data over many years from one million or more people living in the United States, with the ultimate goal of accelerating research and improving health" through precision medicine (Sankar and Parker 2017). *All of Us* included US$70 million to the National Cancer Institute in 2016 to lead efforts in cancer genomics (National Institutes of Health 2017). These well-funded, national initiatives involving multiple health and technology sectors, effectively doubles the federal funding for cancer research over the next 5 years (McCarthy 2016).

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Despite these impressive advances in research and funding, cancer health disparities persist in the form of disproportionate cancer incidence and mortality rates among certain populations and cancers. Among Alaska Native and American Indian (ANAI) men and women, cancer death rates increased significantly from 1990 to 2009, yet decreased for non-Hispanic White men and women (White et al. 2014). Analysis of the SEER Alaska Native Tumor Registry found the age-adjusted incidence rate among Alaska Native (AN) women for all cancer sites combined was 16% higher than the rate among US White women, while the rate among AN men was not significantly different from the rate among US White men (Kelly et al. 2014). Further, incidence rates for lung and colorectal cancer among AN men and women exceeded rates among US White men and women (Kelly et al. 2014).

AN communities stand to benefit from research funded through the *Cancer Breakthrough 2020* and *All of Us* initiatives; however, past unethical conduct of research and lack of community engagement have contributed to a general mistrust of researchers and reticence to participate in research among ANAI people (Boyer et al. 2011; Foulks 1989; Thomas et al. 2011). Experiences with inadequate protection of individual and community level confidentiality, failure to involve participating communities in research development and implementation, poor practices of informed consent, and failure to share research findings with participating communities are central factors contributing to the distrust of research (Glover et al. 2015). In addition, research practices previously used involving the collection of biological specimens without consent among ANAI community members is concerning (Couzin-Frankel 2010; Tsosie 2007). Thus, there are substantial, historically founded, and understandable barriers to doing much-needed research, especially involving the collection of personal data such as family health history and biospecimens, in AN communities.

A handful of publications to date have reported on AN community members perception of research in an attempt to understand and address concerns (Ayunerak et al. 2014; Hiratsuka et al. 2012; Rasmus 2014; Shaw et al. 2013). There has been limited focus on perceptions of cancer research. Such studies are necessary to develop and implement cancer research that includes the AN population, which is disproportionately affected by these health conditions. As the *Cancer Breakthrough 2020* and *All of Us* develop, efforts are needed at a local and national level to understand and address the multifaceted reasons for cancer disparities. Despite advances in cancer research and clinical practice in recent decades, individuals within minority populations have not reaped the same benefits as non-Hispanic Whites (Ramirez and Thompson 2017).

A current gap in the conduct of initiatives like *Cancer Breakthrough 2020* and *All of Us* is that they do not include direct input about ANAI concerns and perspectives on cancer research. This input is critical to ensure that ANAI populations are involved in and benefit from these efforts. Appropriate communication strategies are needed to achieve the community engagement required to conduct effective health research with ANAI populations. The established principle of community respect in genetic research includes recognition of community values, interests, and rights through collaboration in all aspects of the research process (Chartier et al. 2017; Dean et al. 2017). This applies to the interpretation and dissemination of results; co-development of research priorities and questions; appropriate informed consent; and regular discussion with the community regarding genetic ethics (Bush et al. 2014). Although mistrust for research still exists, ANAI and other Indigenous communities are generally open to participating in research (Buchwald et al. 2006), particularly if it involves studies that are conducted by organizations that have demonstrated trustworthy research practices (Dean et al. 2017). These trustworthy research practices may entail meaningful community involvement, which may include partnerships to identify community needs, concerns, and goals; appropriate community representation in research; participant compensation; and consideration towards acknowledging community knowledge in defining research methodologies (Buchwald et al. 2006; Hiratsuka et al. 2012; Vawer et al. 2013).

Population-specific concerns related to cancer research must be investigated so that effective communication and engagement strategies can be developed to ensure that populations disproportionately affected by cancer are included in novel research and treatment approaches (Ramirez and Thompson 2017). This study engaged members of a rural AN community to explore their attitudes about cancer research pertinent to AN individuals and communities and to investigate their views about research, in general, and, specifically biospecimen collection and family health history.

Methods

Community engagement and approvals

This study was a partnership between AN health and tribal leaders in a rural AN community and researchers from two AN health organizations—Southcentral Foundation and the Alaska Native Tribal Health Consortium. Tribal approval was provided by the tribal government of the participating community and the two AN health organizations. The Alaska Area Institutional Review Board also approved all research procedures. During data collection, community members were asked how they would like to learn about study results and who should receive them. Recommendations included disseminating results to the community through radio interviews and presentations at the local health fair, as well as sending a flyer or brochure to all members of the community regardless of participation in the study.

Setting

This study took place in a coastal community within Alaska's Aleutians-West Census Area. Similar to much of rural Alaska, access to the community is limited to air and water transport. Approximately 400 people live in the community year-round and the average age of residents is 35 years old. Approximately a third of residents are 19 years old or younger (State of Alaska Department of Commerce 2017). A health clinic providing primary and preventative care is staffed by itinerant providers (physician, nurse practitioner, physician assistant) and local community health aides, including a dental health aide therapist and behavioral health aides. The health aide positions are paraprofessional and staffed by trained, local residents. Dentists, optometrists, and other specialty health practitioners from Anchorage provide services to the community one to two times a year. Community members who require advanced care, such as surgery or chemotherapy, must travel by air ambulance or scheduled air service to Anchorage, Alaska's largest city (pop.~ 300,000), approximately 750 miles away (State of Alaska Department of Commerce 2017). Inclement weather often delays travel in and out of the community.

Recruitment

Participants were recruited through purposive sampling (Kuzel 1992) during several visits researchers made to the community. Recruitment flyers for family health history and biospecimen focus groups were posted in public locations in the community. Participants signed up to participate at recruitment tables located in the health center and grocery store. The research team presented information on cancer among AN people and answered questions about the study at a community wellness gathering. A local radio station invited community members to discover more about the study by attending the wellness gathering, calling the health center, or visiting researchers at recruitment tables. Recruitment for both family health history and biospecimen focus groups occurred simultaneously and participants were asked to select one group according to scheduling preferences.

Data collection

In 2014, four focus groups were held at the community health center and co-facilitated by researchers trained in qualitative methods following informed consent procedures. Two of the focus groups sought participant views on family health history collection for cancer research and two sought participant views on biospecimen collection for cancer research. All focus groups elicited participant views and experiences regarding research in general. Table 1 displays examples of focus group questions. Focus groups were audio recorded with permission from participants.

Participants

Participants were AN individuals at least 18 years old, able to provide informed consent, and willing to share their views about research in a group discussion. Participants were asked to complete a demographic form before focus group discussions. Participants did not need to have experience with cancer or biospecimen collection or have lived in the community for any specified length of time to participate. Each participant received dinner and a US$50 gift card. All focus groups were audio recorded.

Analysis

Audio recordings were transcribed verbatim with identifiers redacted. Thematic analysis was used to examine data for common themes (Braun and Clarke 2006). Two researchers reviewed and discussed the transcripts to develop a general analysis framework, then independently compiled a list of proposed codes. The proposed codes were reviewed by the two researchers and a finalized coding scheme was developed. Researchers independently coded one transcript using this scheme and established coding consensus prior to proceeding with further analysis. Following consensus, the remaining three transcripts were independently coded and analyzed for key themes by one researcher. Subsequently, the thematic analysis was reviewed by the entire research team, including researchers who coded transcripts and the principal investigators.

Results

Participant characteristics

Nine community members participated in family health history focus groups and 19 individuals in biospecimen focus groups. All focus groups were held in the evening to allow individuals working during the day to participate. Size differences between family history and biospecimen focus groups may be due to participants self-selecting into each group. All participants were of AN heritage. Most were female (68%), over the age of 40 (68%), and had obtained at least a high school education (75%) Table 2. Participants lived in the community from 2 to 71 years.

Themes and sub-themes

Primary themes were established deductively, using focus group questions, to structure and guide the analysis. These include (1) general experiences with research, (2) views specific to the use of biospecimen samples for cancer research, and (3) views on collecting family health history for cancer research. General findings for each theme are described

Table 1 Example focus group questions

Section	Example questions
View of research	What do you think about research?
Views of genetic research	What do you think about researchers collecting biospecimens to study certain genes? If you volunteered to participate in a genetic study in which your blood or spit were collected, how would you feel about giving the researchers permission to look at your medical record?
Views of cancer-related genetic research	What do you think about researchers collecting biospecimens to study how genes may influence cancer in Alaska Native people? If you were asked to participate in a research study in which the researchers wanted to collect your blood or spit to study how genes influence cancer in AN people, what would you think about before making a decision (about whether or not to participate)?
Views of collection of family health history	What should researchers and health care providers think about when asking AN people to share information about family health history? What concerns would you have about giving this information to a researcher How do you feel about researchers reading information about your family health history in your health record?

below. Sub-themes were developed inductively through analysis of the coded data and are described below each corresponding theme. Tables 3, 4, and 5 include exemplary quotes from each theme and sub-theme.

General experiences with research

Many participants had experienced or participated in different types of research, including studies on the environment (e.g., nuclear and heavy metal contaminants in sea mammals and birds); health (e.g., cancer, lead poisoning in children); genetics (e.g., DNA for ancestral migration research); and socioeconomics (e.g., boarding school experience, subsistence "living off the land"). Participation in these prior studies may have shaped their perception and trust in any type of research. They described having had both positive and negative research experiences. Discussion of past negative experience was often paired with recommendations about how to appropriately conduct future research.

Table 2 Focus group participant demographics ($N = 28$)

Type	Sub-type	# of participants	% of participants
Gender	Male	9	32%
	Female	19	68%
Age	18–29 years old	6	21%
	30–39 years old	3	11%
	40–59 years old	8	29%
	60 years or more	11	39%
Education	Up to grade 12	7	25%
	High school graduate	9	32%
	Some college	7	25%
	College graduate	2	7%
	Trade or vocational school	2	7%
	Other (e.g., trade school and some college)	1	4%

Table 3 Focus group theme – General Experiences with Research

Sub-theme	Exemplary quote
Weighing the experience of being “over-studied” and the importance of community benefit	“... there’s always people coming out and doing that [research], plus there’s a lot of research on the animals [birds and sea mammals] and stuff like that. So, you know, so, ‘Oh no, not another study,’ I have kind of heard, you know, remarks from different people about that.” “If it’s going to benefit you so you can get your doctorates [sic] degree, you know, no. I want it to benefit people.” “I think I would go ahead and go with it. If it’s going to--if it does not come out until a few years later, and then it has some sort of a benefit for the community in the future, then I would still participate even if it’s not going to affect me and probably families that [are] here right now, but it--maybe in another generation that they see what’s going on here. I would participate just for that fact.” “Some years it seems like we have a lot of people getting cancer and stuff like that, then do not really know what causes it. Is it diet or lifestyle or something like that? And I have had couple instances of family members that have passed on from different types of cancer, I think. And I think I was kind of interested in research here to see something; you know why are more people getting cancer and stuff like that?”
Lack of follow-up and the importance of continuous community engagement	“Some of the researches [sic] that were done prior to you guys [this project] getting here, the only thing I know was in the community--and I can say that because I have been here forever--is not getting the results back.” “I think it’s important to also maintain follow-up because a lot of people come in and do things and then leave. And then that’s kind of the end of it; there’s not that maintaining the care or maintaining information.” “I think we should … have like locals or local Natives be put through training and get that degree or whatever they need to become researchers so they would not … come in and come out [of the community] … then to have someone there, constantly there to keep up with the follow up.”

Weighing the experience of being “over-studied” and the importance of community benefit Participants described the experience of being “over-studied,” meaning that there were frequently researchers in the community doing one kind of study or another. This experience was not depicted as dissuading them from engaging in future research, but rather as requiring them to seriously question and consider requests for additional research in the community. Research was seen as having potential benefits at both individual and community levels, as well as benefiting the researchers. Some participants said that research should occur only if it serves the “greater good” of the community rather than solely benefiting researchers or the individual community participant. Community benefit was defined as extending to future generations and does not necessarily require immediate benefit or return of services to the community. Finally, some participants viewed a key potential benefit of cancer-related research as being empowered to understand and improve health over time by identifying locally relevant risk and protective factors, for the purpose of decreasing cancer rates in the community.

Lack of follow-up and the importance of continuous community engagement Many participants expressed the need for researchers to have clear communication with community members about the intent of research and its alignment with the community’s interests and priorities. This communication was described as not a one-time event at the time of informed consent, but rather an ongoing process that should occur throughout the research at all stages. Participants talked about researchers’ lack of follow-up with community members about outcomes of studies conducted in the community. Participants recollected that in the past, researchers did not often provide results (regardless of them being individual or aggregate) to the community; thus, participants and community members never knew what knowledge was gained by the research and other outcomes of the information

Table 4 Focus group theme – Views on Biospecimen Collection

Sub-theme	Exemplary quote
Access to biospecimen collection results	"... if somebody did get a sample done and stuff like that, I think if they asked, they should be able to get a copy of what the report looked like."
Getting biospecimen collection results back	"Just getting the results back to them that would make them--that would make a big difference. I mean, 'Oh, you'll get the results in two weeks.' A month, a year, two years, three years go by and we are like, 'Why did we even bother?' That's the stigma between what was done in the past, growing up, versus now. How fast can you get me my results after I give you a spit and hair sample, a swab? Granted, we all know it's going to take a week to get your blood work back from here, sometimes ten days, depending on if the plane makes it. But it's just getting the results back." *Author Note: This comment was in reference not only to biospecimen collected for research but also for clinical diagnostic purposes.
	"I think it's all good, but I'd like to see results in writing, in print or however you want to say it. I think the studies should have been done ten years ago. I have seen--I used to work with elders, and not being a health--I mean, not being a doctor or nurse, I would look at someone, and they would be sent back and forth to Anchorage for all the pain they had, and they would tell me, "They told me it was in my head." And I just looked at them, and I was--I just, in my heart, I knew they have cancer. Just seeing them, the way they looked, how thin and pale, and they were in pain, and then it turns out they die a couple of months later."

they shared with researchers. Participants recommended that researchers establish an ongoing relationship with the community, which includes sustaining relationships after data collection is complete. Participants provided insight into how researchers can more effectively engage with the community in general and specifically with regard to following-up and returning results while including tribal leadership in all stages of the research process (development, design, data collection, and dissemination). One participant suggested hiring community members to join the research team (e.g., to take part in recruitment and data collection activities) as a means of engagement.

Researchers were encouraged to provide participants with a timeline at the beginning of the study outlining the research process and when to anticipate results, and participants stated that researchers should provide timely results to the

Table 5 Focus group theme – Views on Collection of Family Health History

Sub-theme	Exemplary quote
Sharing family health history	"… in the future if I should have any grandchildren, they would say, "Okay, this is from grandma's father's side, grandma's mother's side." What were--what issues did they have in the family back then? And that way we would know--they may be someone that's interested in their health concerns like a lot of us".
Medical records	"… our identity is in the records, our names, numbers, and our information … .who has access to it, who is going to look at them, and what are you getting out of it, what are you looking for."
	"… in the history of my family, I have lost loved ones with cancer. So it kind of rings a little bell … a warning sign for me at this age. I consider myself still young but concerns, yes. I would feel comfortable if they did have to go through the whole history of my life just to find out [if I am at risk of cancer]."
Contacting family members to collect family health history	"I am pretty sure people would be more willing to share family history, as long as the people you are asking from, like from me, it's kept confidential and it really does hit a hard note with cancer patients and people that died of cancer.".

community (e.g., within 2 months of determining final results). Many participants believed they were not being informed of research study outcomes. They may have never received results or evidence that their participation made significant differences in general or with regard to their community. Furthermore, it was suggested that researchers provide written results back to the community, including researcher contact information. It was noted that researchers should be easily accessible to respond to queries from community members; in addition to the timely return of results, this means telling participants when to anticipate that results will be available and keeping an open line of communication. Participants reported that community members expect transparent communication from researchers about both the challenges and successes encountered throughout the study, from initial engagement through publication of findings and beyond, again highlighting the importance of researchers maintaining continuous, long-term engagement with the community.

Views on biospecimen collection for cancer research

Participants were asked to share feedback on the practice of collecting biospecimen (e.g., saliva, blood) for genetic cancer research including who should collect biospecimen, what they should be used for, how they are collected, and when they should be collected. Generally, participants were willing to take part in biospecimen collection for cancer research as well as for clinical screening purposes. However, participants also described the need to have data collection procedures and dissemination processes planned in collaboration with community leadership before community members agreed to participate. Participants stated that biospecimen collection should be voluntary and results must be confidential and provided back to participants. More specific findings on this theme are described below.

Access to results from biospecimen collection analysis Participants shared numerous perspectives about who should have access to results of biospecimen analysis for cancer research. Some did not see a problem with sharing individual de-identified results with anyone, regardless of their relationship to the community while others did not want results to be shared with anyone but the individual participant who provided the biospecimen to researchers. Others endorsed sharing individual results first with the individual who provided the biospecimen, followed by sharing results in the aggregate with the community to both protect privacy of individual participants and inform the community of results. With regard to biospecimen collection for clinical purposes (e.g., cancer screening), some participants wanted clinical screening results involving biospecimen collection to be stored in their medical records, but were hesitant to have genetic information stored in their medical records without restrictions in place for who has access and for what this information would be used for. For example, some saw benefit to having their genetic information stored in their medical record for preventative reasons as well as for the potential to help family in the future and would want genetic family members to have access to their records. Most participants wanted access to results, at the very least, at an individual level.

Return of results from biospecimen analysis Some participants specifically commented that they had not received results from testing done on biospecimen collected for past studies or from diagnostic screening. It is unclear whether or not return of results was promised, but it was clear that participants would have preferred to have results returned to them and may have believed that this would occur. With regard to biospecimen collected for clinical purposes, some participants mentioned a need for increased cancer screening and follow-up, citing a lack of clinical cancer screening being done in the community and limited health care provider follow-up on diagnostic cancer screening. This was seen by some as a reason many community members were being diagnosed with late-stage cancer.

Views on collection of family health history

In two focus groups, participants were asked about family health history; all four groups were asked about family history of cancer. Themes concerning family health history are described in more detail below. Participants discussed opinions on collecting family health histories and using them for cancer prevention and general genetic research.

Sharing family health history for cancer research Participants shared divergent perspectives about sharing family health histories for research purposes. Some saw sharing family health history for research as a positive, preventative measure with the potential to encourage screening for diseases known to run in families, like some cancers (e.g., breast, colorectal). Similarly, some supported documenting and sharing family health histories, saying that this would benefit family members in the future. Others; however, wanted the family health histories to be collected only with regard to general health information and not personal lifestyle choices (e.g., diet and exercise). One participant recommended that if collecting family health history was going to be widely implemented, behavioral health counseling services should be made available to people in the community, for instances where remembering a family member's health history (e.g., death from cancer) triggers a challenging emotional experience.

In addition to perspectives on whether family health history *should* be shared, participants discussed whether family history *could* be shared. They described challenges in collecting family history information due to a community history of forced government relocation during World War II and beyond, as well as introduced epidemics that resulted in the loss of many family members. Acquiring reliable multigenerational family health information would thus be difficult in many families due to limited or complete lack of knowledge about previous generations' health history.

Medical records Participants also provided views on collecting and storing family health history information from medical records for research. Most participants shared that access to medical records should be restricted to protect individual and family privacy. Many clarified that before extracting family health history from medical records for research purposes, patients should be given the opportunity to accept or decline consent specifically for those data to be accessed and used for research. Others were open to having their medical record information reviewed for family health history as long as the data were deidentified prior to researchers having access and using it in a study. Participants also shared that placing family health history information collected for research in the medical record should be a personal choice and should not be mandated. Still others voiced a desire for transparency about medical records access; they wanted to know more about when and why their medical records were used as well as the duration of time that information would be accessible as well as who has access to their data.

Contacting family members to collect family health history for research Participants were also asked to provide feedback on the acceptability of researchers contacting family members to gather family history information. Some were supportive without stipulations, while others said they would want to know what questions would be asked and the names of individuals or institutions requesting information before agreeing to have family members contacted. Others said they were amenable to researchers contacting family members as long as confidentiality was maintained for both themselves and the contacted family members.

Discussion

Participants were generally supportive of research involving biospecimen and family health history collection and provided pragmatic feedback for positive community engagement for cancer research highlighting a need for research communication pertinent to biospecimen-related cancer research. Participants discussed ways that research may benefit or harm their community. Results highlight a need for transparent and continuous researcher-community communication about biospecimen-related cancer research.

Improvements for community engagement and communication suggested by participants in this study aligned with other studies concerning AN/AI research. Successful community engagement entails active and meaningful community involvement at all levels of the research process (Boyer et al. 2005; Burhansstipanov et al. 2005; Lopez et al. 2012; Peterson 2010; Williams et al. 2010) which may include providing educational, financial, or technical resources to the community (Burhansstipanov et al. 2005; Noe et al. 2007; Shaw et al. 2013). Successful engagement also requires communication on the part of researchers, who must take into account and respect community beliefs and values (Allen et al. 2014; Legaspi and Orr 2007; Rasmus 2014; Rivkin et al. 2013). Research must take community needs into consideration and research intent and results should be clearly and iteratively communicated. Consistent with community-based participatory research theory (Israel et al. 2010), participants believed that community members should have an active, meaningful role—including being a dynamic part of the research team. Meaningful and principled community engagement can be cultivated through transparent and continuous communication.

Similar to others findings (Hiratsuka et al. 2012), participants in this study expect more frequent and more detailed communication about the research process and outcomes as compared to past research that was conducted in their community. Communication should include pertinent details about why data is collected, who has access to it; what it is used for; how long it will be used; and what happens to biospecimen when they are no longer needed for a specific study, as well as aggregate and individual results of the study, depending on local and individual preferences. Overall, communication plays a fundamental role in community-engaged research including understanding community cultural values, formal and informal networks, and patterns of communication, which makes active community involvement imperative for the conduct of respectful, meaningful research (Strickland 2006; Trinidad et al. 2015). Researchers should not make assumptions about communication preferences based on other research or populations (Burhansstipanov et al. 2005), but should instead consider using communication strategies that use iterative inquiry to learn about and engage communities in which they wish to conduct research. Moreover, in the current precision medicine research environment, community partnership is an expectation (Scherr et al. 2017). Consistent with this expectation, bidirectional interaction between community members and researchers emphasizing mutual respect and trust encourages and sustains community motivation to participate in research (Baquet et al. 2013; Burhansstipanov et al. 2005), making regular contact between key community members and researchers a high priority.

Participants were generally open to biospecimen collection for cancer research; however, they wanted to ensure processes were in place, including returning results back to those who provided biospecimen using appropriate measures such as mail, integration with electronic health records, or in-person conversation. Suggestions varied and emphasized access to results, at the very least to the individual, since results from biospecimen collection in past research and medical care were inconsistently and sometimes never shared with community members. Participants also discussed the expectation of receiving aggregate research summary results. Paying heed to this expectation, the study team disseminated results using all suggested methods (radio, health fair, and mail). This was financially feasible given the small number of community households (~300 household mailboxes); future studies would benefit from including this expense in the research budget. Some suggested that community involvement is needed when developing results summaries (Kwan et al. 2014). Other studies have stressed the importance of community involvement in sharing results, including which results to share and how to do so in a culturally respectful manner; following these principles enables researchers to achieve the dually critical goals of advancing scientific knowledge and ensuring beneficence to the community (Kwan et al. 2014; Trinidad et al. 2015). Remarkably, Buchwald et al. (2006) determined that among urban AI/AN people who completed a survey about research participation, community involvement in interpreting genetic research results did not increase the odds a person would participate in a study and suggest that this may be due to community members' recognition of a need for outside technical expertise. On the other hand, Rivkin et al. (2013) suggest that utilizing Indigenous knowledge and community expertise generate progress towards research results dissemination which facilitates enhancements in relation to cultural sensitivity.

A prior study conducted with ANAI participants in an urban setting found barriers to collecting family health history including an absence of health history information due to adoption or death, lack of familiarity with the concept of family health history, and emotional upset when discussing family members deaths (Southcentral Foundation 2008). Individuals in that study also indicated some AN people may distrust non-Native providers and thus be reluctant to share certain information. As the aforementioned study took place in an urban setting, this current study provides additional knowledge regarding rural AN perspectives on collecting family health history information. In this current study, the idea of collecting family health histories for cancer research showed varied perspectives ranging from beneficial to unrealistic. It was considered unrealistic for some families because of limited generational knowledge participating community members had regarding family health. Similar to other communities that have experienced past injustice and historical traumas (e.g., such as colonization, forced government relocation, and epidemics), participants in the community described challenges locating historical records, particularly those that involve family histories often shared orally. If family health history were to be collected for cancer research via access to medical records, then transparent communication about how family health histories will be retrieved and used must be taken into consideration.

Expectations shared by community members in this study are apt for how biospecimen-related research should be conducted in their community and in similar diverse rural and frontier communities. However, this is not to say challenges to implementing these expectations are inconsequential. A lack of time and presence in the community can limit the frequency and quality of community engagement. Researchers must, therefore, plan for the critical and valued time needed to develop and maintain trusting partnerships (Baquet et al. 2013; Legaspi and Orr 2007). Moreover, expenses related to regular in-person contact to develop and maintain relationships may be impractical and require adequate, often long-term planning to obtain, especially when traveling to rural Alaska communities where air travel is costly (Hoeft et al. 2014; Johnson et al. 2006; Katz 2004). Rural and frontier communities may lack interest in research or the availability to participate due to busy workloads and other responsibilities (Chipp et al. 2011; Landon et al. 2004). These limitations are justification for research protocols to include provisions for research education and training opportunities for the community. On the side of researchers, more flexibility in funding resources and timelines may help encourage the development of mutual trusting partnerships with community members.

Limitations

This study took a community engagement approach and strived to involve community at all levels, including the hire of a local research coordinator who provided on-the-ground logistic support within the community. In spite of this, we had difficulty finding a community resident to serve as research coordinator who was available for the duration of the study, thus decreasing our ability to fully engage community members. Community tribal leadership reviewed and provided input on all study aspects, from conceptualizing the project to disseminating results, which included guidance on when, where, and how results should be shared. This study only includes participants from one rural AN community and should not be construed as representative of all AN communities. Another limitation was that only a third of participants were men (32%), so results may not necessarily represent male viewpoints in the community. Although the majority of community members were under 35 years old, most participants in this study were older (40+ years), so results may not reflect opinions of younger community members. Focus groups were held in the community health care setting, which may have contributed to confusion regarding biospecimen collected for clinical research from those collected for clinical practice. However, holding the focus groups in the health care setting may have provided more credibility for the research as people generally have trust in services provided in that setting. Future studies could be improved by clearly distinguishing between biospecimen collection for genetic research purposes and for clinical practice. For example, some participants responded to questions about collecting blood samples for genetic research testing as if they were being asked about a health care provider collecting blood samples as part of a complete blood count (CBC) or another diagnostic test. Finally, there was a substantial difference in size of the focus groups on biospecimen collection and focus groups on family history collection for cancer research, probably due to participants self-selecting into one group or another, and this may have biased the results to some extent.

Implications

This study provides perspective about how researchers could improve communication with communities involved in biospecimen-related cancer research. However, much of what was learned can be applied to all forms of research conducted in rural AN communities and potentially other rural frontier communities. Participants in this study shared important perspectives on using family health history for cancer research. It is worth noting that researchers may encounter substantial challenges when communicating about family health histories in communities with a history of trauma (e.g., government relocation, widespread mortality due to epidemics). Generational information may be difficult to obtain since family members and others who knew them may no longer be available to share histories. Although family health history data may be incomplete due to loss of historical records and limitations on availability of oral knowledge, AN individuals and families can, with adequate support, start collecting what they do know about their family's health history to begin documenting potential genetic health risks.

Current efforts are planned to increase diversity in the *Cancer Breakthrough 2020* initiative which includes patient navigation, using mobile technology to collect data, and establishing syndicates dedicated to increasing participant diversity in precision medicine research (Ramirez and Thompson 2017). Participants who contributed to this study may potentially be interested in taking part in future cancer research involving biospecimen collection, as long as that research is voluntary, confidential, conducted in full partnership with the community, and conducted with respect to all community members through practices of openness, transparency, and reciprocity. Opening the door to precision medicine studies such as those occurring through the *Cancer Breakthrough 2020* initiative and *All of Us* could potentially provide a means of increasing participant diversity in large-scale studies with promise to change the landscape of cancer prevention and treatment in the next 50 years. Keeping that door open will require the active, iterative, and ongoing engagement of researchers with AN communities in a manner that is valued and meaningful to all involved.

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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).

Conflict of interest The authors declare that they have no conflict of interest.

Statement of informed consent Informed consent was obtained from all participants for being included in the study.

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