



Increasing health equity through biospecimen research: Identification of factors that influence willingness of Native Americans to donate biospecimens

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ABSTRACT

American Indian and Alaska Native (AI/AN) people are underrepresented in biomedical research, particularly in biospecimen research, yet little research has been conducted to assess AI/AN attitudes about biospecimen donation. Survey data were collected from 278 AI/AN people in Seattle, Washington in 2016 to assess general willingness to donate, background characteristics related to willingness, and circumstances that would increase or decrease willingness to donate biospecimens. Less than half (43%) of participants were willing to donate. General willingness was related to past donation, and trust in how researchers use and store biospecimens. However, willingness to donate for cancer research was 76%. Fear of research exploitation and spiritual beliefs decreased willingness. Among those who were generally unwilling to donate, willingness increased if the biobank was run by a Native American organization, if the participant or family member had cancer and this was the disease being studied, if the community was involved in developing, reviewing, and approving the research, and if the expertise of researchers was known. Among those who were willing, willingness decreased if they had never heard of the research organization, and if the biobank was run by the federal government. Participation of AI/AN people in biobanking initiatives is critical to address health inequities and improve the health of AI/AN people, realize personalized medicine goals, and address the limited generalizability of current clinical and biospecimen research. These results highlight areas in which interventions could be developed to increase AI/AN donation of biospecimens for research with the ultimate goal of reducing health disparities.

1. Introduction

American Indian and Alaska Native (AI/AN) people have a long history of lower life expectancy and higher disease burden compared with the general US population (Disparities Fact Sheet, 2020) and are more likely to suffer from chronic diseases (CDC and Indian Country working together, 2017). These disparities have roots in historical US federal policies of genocide, removal from ancestral lands, and one-sided treaties (History Through a Native Lens, 2020); and contribute to contemporary conditions of AI/ANs which include poverty (Centers for Disease Control and Prevention, 2013); residence in resource-poor environments, low educational attainment (Bachelor's degrees conferred by postsecondary institutions, by race/ethnicity and sex of student, 2018); ongoing discrimination, and inadequate health care

(Broken Promises, 2018). Similarly harmful has been unethical biomedical research that stigmatized and harmed AI/AN communities and contribute to persistent mistrust of research and researchers (Klausner and Foulks, 1980; Mello and Wolf, 2010; Spruhan, 2006).

Addressing AI/AN health inequities is a multi-faceted, complex issue. Long-term solutions focused on disease prevention that address social determinants of health, or focus on health-risk behaviors, show promise for promoting health equity (Whitesell et al., 2018). However, it is equally important to include AI/ANs in research and biobanks. Biobanking is an especially important topic for AI/ANs because addressing their health disparities relies, in part, on identifying biological risk factors and their interaction with environmental risks. For example, no study has ever examined the prevalence of BRCA mutations in AI/AN women. This is a critical omission since breast cancer is on the rise in AI/

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ANs (Howlander et al., 2013) and up to 65% of women who inherit a harmful BRCA1 mutation will develop breast cancer by age 70 (Chen and Parmigiani, 2007). AI/ANs are more likely to die from cancer and heart disease than are members of the general population, even though these are the top two causes of death in both groups (Summary Health Statistics, 2018). However, the next most frequent causes of death in AI/ANs are quite different from the general population, with intentional or unintentional injury, diabetes, and chronic liver disease among the next most common (Espey et al., 2014). Another example highlighting the importance of AI/AN participation in research was a study of high-risk Human Papillomavirus Virus (hrHPV) in AI women where Lee et al. found that the single most prevalent hrHPV genotype (HPV-51) was quite different from the prevalence in the general population, and this genotype was not covered by the extant HPV vaccines (Lee et al., 2019). This information, which directly impacts health care and cancer prevention, was uncovered only because of participation by a large number of AI women and their willingness to donate biospecimens.

Gaps like this underscore the need to consider the possible benefits of biobanking and genetic research for AI/ANs, as long as this research can be conducted on their own terms (A Spectrum of Perspectives, 2014). Well-publicized research conflicts have raised tribal governments' awareness of the need to regulate research and related activities, such as biobanking. The most visible recent conflict involved the Havasupai Tribe where tissue samples taken from tribal members by a university researcher during an approved diabetes study were later used for other research without the tribe's awareness or consent (Dalton, 2002; Notes, 2010). Tribal research review boards and data ownership are current methods to regulate research and prevent further harm (Morton et al., 2013).

Because AI/ANs are underrepresented in biomedical research, it is critical to investigate the factors that would increase participation in research involving donation of biospecimens. A number of qualitative studies about health research in indigenous communities, recently summarized in a scoping review (Woodbury et al., 2019); stress the importance of community engagement, provision of literacy-level appropriate information (Haring et al., 2018); and disclosure about the motivation and intent of the researchers (Hiratsuka et al., 2012). Although these studies have yielded general principles about best practices for engaging in biomedical research in AI/AN communities and collecting biological specimens, they reveal less about individual decision-making about research participation and biospecimen donation. We know very little about the specific characteristics and background factors related to the decision to participate and donate specimens, nor do we know the conditions under which a potential participant would be likely or unlikely to donate. Thus, the primary purpose of this study was to assess, with a survey, general willingness of individual AI/ANs to donate biospecimens, characteristics associated with willingness, and the circumstances under which willingness would increase or decrease.

2. Methods

2.1. Sample & setting

A total of 298 respondents who attended the Annual University of Washington powwow on April 2, 2016 in Seattle, Washington completed an anonymous survey. The survey was distributed from a vendor table by study staff who asked interested individuals if they identified as AI/AN and were 18 years of age or older. A survey and pen were provided to individuals who responded 'yes' to both questions. Survey participants sat at a nearby table and completed the survey in 10–15 min. Upon completion of the survey, a \$10 store gift card was offered to each participant. The study procedures and survey were reviewed and approved as exempt by the University of Washington Institutional Review Board.

3. Survey

Survey items to assess willingness, experience with biospecimen donation, fear of research exploitation, and trust were used verbatim from a survey of older African Americans with minor contextual adaptations (Hagiwara et al., 2014). For example, the African American survey referred to the Tuskegee Trial as an example of research exploitation. In the powwow survey, the conflict between Havasupai Nation and Arizona State University was referred to as an example of research exploitation (Mello and Wolf, 2010). Another question adapted was related to willingness to donate biospecimens to biobanks operated by Native American organizations or the Indian Health Service. While the survey of African Americans was administered to older individuals, the survey items were not specific to any age group.

The first page of the survey included a description of the absence of information about biospecimen donation by AI/AN, the purpose of the survey to assess willingness to donate biospecimens, and that the survey was anonymous. The second page of the survey was a tutorial with images and text written at a 6th grade reading level that defined biospecimens and how they are collected and stored. Following the 1-page tutorial, general willingness to donate a biospecimen was assessed with the question, "Based on what you know about biospecimen collection, biobanking, and how biospecimens are used in medical research, how willing are you to donate your biospecimens?" Response options on a 5-point Likert scale ranged from not willing to very willing. A dichotomous measure was created for respondents who did (somewhat/very willing) and did not (neither unwilling nor willing/somewhat unwilling/not willing) endorse general willingness to donate a biospecimen. Respondents were presented with a series of vignettes to assess barriers and facilitators for biospecimen donation. Questions asked, "How likely would you be to donate your biospecimen if..." Vignette examples included learning about the researchers who would use the biospecimen, knowing that members of your community were involved in developing the research study, knowing why you were asked to donate biospecimens, and several options for what kind of institution operates the biobank where the biospecimen would be stored. Response options for the vignettes were a 4-point Likert scale ranging from very unlikely to very likely. Dichotomous measures were created for respondents who did (likely/very likely) and did not (unlikely, very unlikely) endorse vignette-specific willingness to donate a biospecimen. Respondents were also asked which types of biospecimens they would be willing to donate (i.e., hair, urine, blood, saliva).

Health-related survey questions included respondents' assessment of their overall general health compared to other AI/AN in their community, and self-report of ever having several health conditions, such as diabetes, hypertension, asthma, etc. Previous biospecimen donation history, fear of research exploitation, trust in researchers and biobanks about how biospecimens are used and stored, knowledge of biospecimens and biobanks, and spiritual beliefs about biospecimen donation were also assessed. The survey included 4 items about biospecimen and biobank knowledge participants had prior to reading the tutorial: 1) I knew what biospecimens were, 2) I knew how biospecimens were collected, 3) I knew what biobanks were, and 4) I knew that biospecimens were used in medical research to find cures for diseases. Response options were a 5-point Likert scale ranging from strongly disagree to strongly agree. The average knowledge score was computed across all items; possible scores ranged from 1 to 5 where higher scores indicate more perceived knowledge. Demographic characteristics collected were age, sex, marital status, highest educational attainment, residential zip code, and household income in the last calendar year. A total of 78 close-ended items were included in the survey.

3.1. Statistical analysis

Descriptive statistics were calculated for the overall sample as number and percent for all study variables. Binary logistic regression

was used to estimate the association of each independent variable with the dichotomous measure for general willingness to donate biospecimens. Results are presented for unadjusted models and adjusting for sex and age (models for sex and age only adjusted for the other factor). Marginal standardization was used to report estimates as prevalence difference with 95% confidence intervals. Prevalence- and risk-based contrasts are more easily interpreted than odds ratios, and absolute differences are most relevant to our overarching goal of increasing the number of AI/ANs who consent to banking biospecimens for research.

Change in willingness to donate biospecimens based on additional information about donation circumstances provided in each vignette was assessed by cross-tabulating the dichotomous measures for general willingness and vignette-specific willingness. McNemar’s test was performed for each cross-tabulation to evaluate if the additional information provided in the vignette influenced willingness to donate biospecimens. Because the vignette response options did not include a neutral category, a sensitivity analysis was performed that excluded respondents who endorsed the neutral category (neither unwilling nor willing) for the general willingness item. Percent and exact 95% confidence intervals are reported. All analyses were conducted using Stata 15.1 (StataCorp, 2017).

4. Results

Of the 298 surveys collected, 11 respondents who did not self-identify as AI/AN on the survey, despite the initial screening question, were excluded. Also excluded were 8 surveys that had missing data on general willingness to donate question, and 1 that had missing data for age, resulting in a final sample size of 278. Complete case data for all other study variables was not required due to the exploratory nature of these analyses with many independent variables and no single model including all variables simultaneously.

Participants showed a wide range of general willingness to donate biospecimens: 12% were very willing, 31% were somewhat willing, 16% were neither unwilling nor willing, 17% were somewhat not willing, and 24% were not willing. Fig. 1 shows willingness to donate specific types of biospecimens. Nearly half of respondents were willing to donate blood (48%) while fewer respondents were willing to donate skin (24%) or tumors (18%).

Descriptive statistics for participants’ demographic characteristics are shown in Table 1. Results of binary logistic regression analyses that estimate the association of demographic characteristics with the dichotomous measure for general willingness to donate biospecimens

Table 1

The association of participant demographic characteristics with general willingness to donate biospecimens for research.

	Descriptive statistics ¹	General willingness to donate biospecimens	
		Unadjusted	Adjusted for sex and age ²
	n (%)	PD % (95% CI)	PD % (95% CI)
Sex			
Male	106 (38)	Ref	Ref
Female	172 (62)	3 (-9, 15)	3 (-9, 15)
Age, years			
18–29	74 (27)	Ref	Ref
30–39	68 (24)	8 (-8, 24)	8 (-9, 24)
40–49	57 (21)	12 (-16, 18)	11 (-16, 18)
50–59	44 (16)	9 (-10, 27)	9 (-10, 27)
60+	35 (13)	7 (-13, 26)	7 (-13, 27)
Marital status			
Never married	130 (48)	Ref	Ref
Married or domestic partnership	67 (25)	5 (-10, 19)	7 (-8, 22)
Separated, Divorced, Widowed	73 (27)	18 (4, 32)	21 (5, 37)
Education			
Less than high school	47 (17)	Ref	Ref
High school and some college	135 (49)	5 (-12, 21)	4 (-12, 21)
College degree or higher	96 (35)	2 (-15, 20)	2 (-15, 20)
Household income in past year			
<\$20,000	131 (48)	Ref	Ref
\$20,000–\$39,999	61 (22)	-7 (-22, 8)	-8 (-23, 7)
\$40,000–\$59,999	49 (18)	-12 (-28, 4)	-14 (-30, 2)
≥ \$60,000	31 (11)	5 (-15, 25)	4 (-16, 23)

¹Cells may not sum to N = 278 due to missing data.

²Models for age and sex only adjusted for the other factor; PD = prevalence difference; CI = confidence interval; Ref = reference group.

(willing = very or somewhat willing; unwilling = neutral, somewhat not willing, or not willing) are also presented in Table 1. Prevalence difference estimates from the logistic regression models consistently showed higher willingness to donate among older age categories compared to 18–29-year-old individuals (prevalence differences = 7%–12%), but the 95% confidence intervals did not rule out equality. Compared to being never married, people who were formerly married

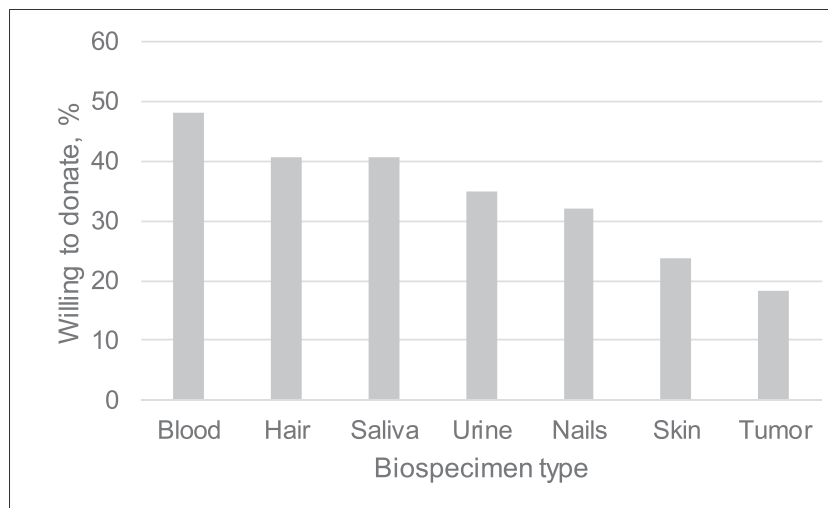


Fig. 1. Willingness to donate specific types of biospecimens (n = 278).

(divorced/widowed/separated) had a higher prevalence of endorsing willingness in the unadjusted model (prevalence difference = 18%, 95% confidence interval 4% to 32%), with a higher magnitude of association after adjusting for sex and age.

Table 2 illustrates the association between participant survey responses and general willingness to donate biospecimens for research. People who considered themselves healthier than other AI/ANs had 19% higher prevalence of willingness than people who considered themselves of similar health to other AI/ANs (neither unhealthy nor healthy, 95% confidence interval 4% to 35%). Self-reported health conditions showed a wide variety in association with willingness to donate. Prevalence differences were markedly higher for people with depression/anxiety (19–20%) and stroke (30–31%). People who had previously donated biospecimens for research had higher prevalence of willingness compared to people who had previously declined to donate and people who had never been asked. However, 92% of participants had never been asked to donate biospecimens. Participants with fear of research exploitation (–22%, 95% confidence interval –37% to –7%) or spiritual beliefs that prohibit biospecimen donation (–43%, 95% confidence interval –57% to –29%) had a lower prevalence of willingness to donate compared to those without those beliefs. Trust in how researchers use biospecimens (30%, 95% confidence interval 14% to 45%) or how biobanks store biospecimens (45%, 95% confidence interval 30% to 60%) was associated with higher prevalence of willingness to donate. Lastly, people with a high level of knowledge about biospecimens and biobanks had moderately higher prevalence of willingness to donate than those with a low level of knowledge (10%, 95% confidence interval –4% to –25%), but confidence intervals included the null.

In the next set of analyses, we cross-tabulated general willingness with vignette-specific willingness to donate biospecimens (Table 3). The additional information provided in each vignette had a significant effect on willingness to donate (p-value range from <0.001 to 0.03). The most notable facilitators to donation were (with percent change from unwilling to willing shown in parentheses): The biobank run by a Native American organization (36%); cancer being studied and the participant or the family member having the disease (36%); knowing about the education, training, and research experience of the researchers (30%); members of the community being involved in developing the research (29%); and the research being reviewed and approved by community leaders (29%). Barriers to donation included if the biospecimen was sent to a research institute the respondent had not heard of (24% changed from willing to unwilling) or if the biobank was run by the federal government, but not the Indian Health Service (27% changed from willing to unwilling). Sensitivity analyses showed a similar pattern of results (results not reported).

5. Discussion

Participation of AI/AN people in biobanking initiatives is critical to address health inequities and improve health outcomes, realize personalized medicine goals, and address the limited generalizability of current clinical and biospecimen research (Dang et al., 2014). Yet, little is known about AI/AN attitudes towards donating biospecimens, the conditions under which such donations would be acceptable, and the characteristics of people willing to donate. In the present study, we sought to address these gaps by gathering quantitative survey data from 278 AI/AN people.

In terms of general willingness to donate biospecimens, we found that less than half of the participants (43%) were somewhat or very willing to donate, with about the same number being somewhat or very unwilling (41%) and 16% undecided. However, when asked about willingness to donate biospecimens if cancer was being studied and the respondent or a family member had cancer, 76% of participants reported they would be willing. These results are similar to a smaller qualitative study with 20 tribal members in the northeastern region of the US and

Table 2

The association between participant survey responses and general willingness to donate biospecimens for research.

	Survey Responses ¹	General willingness to donate biospecimens	
		Unadjusted	Adjusted for sex and age ²
	n (%)	PD % (95% CI)	PD % (95% CI)
Health characteristics			
Overall health compared to other Native Americans in your community			
Very unhealthy/Somewhat unhealthy	60 (22)	12 (–6, 30)	12 (–6, 31)
Neither unhealthy nor healthy	44 (16)	Ref	Ref
Somewhat healthy/Very healthy	169 (62)	18 (3, 34)	19 (4, 35)
Self-reported health conditions³			
High blood pressure	72 (26)	9 (–5, 22)	8 (–6, 22)
Depression/anxiety	69 (25)	19 (6, 33)	20 (7, 33)
Asthma	61 (22)	5 (–9, 19)	6 (–8, 20)
Diabetes	40 (14)	5 (–12, 21)	4 (–13, 21)
High cholesterol	38 (14)	8 (–10, 25)	7 (–11, 24)
Cancer	22 (8)	12 (–10, 34)	12 (–9, 34)
Thyroid disease	21 (8)	–1 (–23, 21)	–1 (–24, 21)
Heart disease (heart attack)	15 (5)	3 (–23, 29)	4 (–22, 30)
Stroke	11 (4)	30 (3, 57)	31 (4, 57)
Experience with biospecimen donation			
Previous donation history			
Never asked to donate	248 (92)	Ref	Ref
Asked but did not donate	8 (3)	–4 (–39, 30)	–4 (–38, 31)
Asked and donated	14 (5)	15 (3, 29)	15 (3, 27)
Fear of research exploitation			
I am concerned that I will be treated as a guinea pig in medical research			
Strongly disagree/Disagree	63 (23)	Ref	Ref
Neither disagree nor agree	101 (37)	–3 (–19, 12)	–3 (–19, 13)
Strongly agree/Agree	112 (41)	–23 (–38, –8)	–22 (–37, –7)
Trust			
I completely trust medical researchers' decision about how my biospecimens are best used			
Strongly disagree/Disagree	93 (34)	Ref	Ref
Neither disagree nor agree	123 (45)	8 (–5, 21)	8 (–5, 21)
Strongly agree/Agree	60 (22)	30 (14, 46)	30 (14, 45)
I completely trust biobanks' procedures on how biospecimens are stored			
Strongly disagree/Disagree	63 (23)	Ref	Ref
Neither disagree nor agree	141 (51)	11 (–3, 24)	11 (–3, 24)
Strongly agree/Agree	71 (26)	45 (30, 60)	45 (30, 60)
Knowledge of biospecimens and biobanks⁴			
Low (1.00–2.75)	101 (36)	Ref	Ref
Moderate (2.76–3.75)	93 (33)	–1 (–15, 13)	–2 (–16, 12)
High (3.76–5.00)	84 (30)	11 (–4, 25)	10 (–4, 25)
Spiritual beliefs			
My specific spiritual beliefs prohibit me from donating biospecimens			
Strongly disagree/Disagree	91 (33)	Ref	Ref
Neither disagree nor agree	113 (41)	–35 (–48, –22)	–35 (–47, –22)
Strongly agree/Agree	71 (26)	–42 (–56, –28)	–43 (–57, –29)

¹Cells may not sum to N = 278 due to missing data.

²Models for age and sex only adjusted for the other factor.

³Reference for each condition is people who did not endorse that same condition.

⁴Average score for 4 knowledge items scored 1 = Strongly disagree, 2 =

Disagree, 3 = Neither disagree nor agree, 4 = Agree, 5 = Strongly agree where higher scores indicate more perceived knowledge. PD = prevalence difference; CI = confidence interval; Ref = reference group.

who had at least one family member affected by cancer, in which 71% said they would be very likely or somewhat likely to donate a biospecimen for cancer research (Haring et al., 2018). Therefore, willingness to donate biospecimens may be disease-specific. The reasons for willingness to donate biospecimens in our study and in earlier studies is unknown, but these comparisons highlight the importance of identifying the conditions under which biospecimen donation would be more acceptable to AI/AN people. To that end, we found that willingness to donate was related to specific background characteristics and did increase under certain conditions.

When we investigated participant demographic and health characteristics in our AI/AN sample that may be related to general willingness to donate, we found a few significant effects: Older age, people who were formerly married, compared with never married, were more willing to donate; participants who perceived themselves as healthier than other AI/ANs compared with participants who perceived themselves to be of similar health as other AI/ANs were more willing to donate; and those with a history of depression/anxiety or stroke were also more willing.

Not surprisingly, participants in our study were more willing to donate if they had previously done so for research purposes (compared with those who had declined or never been asked), or if they trusted the way in which researchers would use and store biospecimens. However, only 8% of participants had ever been asked to donate biospecimens. These findings are similar to survey results from African Americans in which only 18% had ever been asked to donate biospecimens (Hagiwara et al., 2014). Thus, limited participation in biobanking programs may be due to fewer opportunities to participate rather than an actual unwillingness to donate biospecimens.

AI/AN participants reported they were less willing to donate if they had a fear of research exploitation. Indigenous people have been subject to health and anthropological research without consent. Unethical research contributes to the enduring mistrust that is difficult to overcome. Survey respondents were also less willing to donate if their spiritual or cultural beliefs prohibited it. For Western researchers, the hardest theme to comprehend may be the connection of tissue to one's ancestral lands, ancestors, and culture. For indigenous peoples, asking for biospecimens may be asking for much more than a mere biological sample (Aramoana and Koea, 2019). Survey data revealed that knowledge about biospecimen donation may not be related to willingness to donate, which suggests that simply educating AI/ANs about biobanking is insufficient to encourage willingness.

Of note is the analysis of the circumstances under which participants would change their willingness to donate. Among those who were generally willing to donate, two conditions stood out that would reduce willingness—biospecimens sent to a research institute they had never heard of, and a biobank run by the federal government, but not the Indian Health Service. Of those who were generally unwilling to donate, several specific circumstances increased willingness. The largest increases were associated with: The biobank run by a Native American organization; cancer being studied and the participant or the family member having the disease; the education, training, and research experience of the researchers being known; members of the community being involved in developing the research; and the research being reviewed and approved by community leaders.

Taken together, these results align with findings from other investigations of attitudes and beliefs of indigenous people regarding participation in research involving biospecimens, but also provide new information. Specifically, qualitative studies have highlighted the importance of community involvement in research, disclosure about researchers' motivations and intent, and detailed information about the study and procedures for storage and destruction of specimens

Table 3

Willingness to donate biospecimens and how general willingness changed when presented with additional information about donation circumstances.

	n (%)	n (%)	n (%)	n (%)
General willingness to donate biospecimens	Willing ¹	Change in willingness		
Based on what you know about biospecimen collection, biobanking, and how biospecimens are used in medical research, how willing are you to donate your biospecimens	121 (43)	Reference	Reference	Reference
How likely would you be to donate your biospecimen if ... ²		Willingness unchanged ³	Willing to unwilling ³	Unwilling to willing ³
You learn about the researchers who will be using your biospecimen, like their education, training, and research experience	183 (67)	175 (64)	18 (7)	82 (30)
You knew that members of your community were involved in developing the research study	181 (66)	178 (64)	18 (7)	80 (29)
You knew that the research study was reviewed and approved by your community leaders	181 (66)	176 (64)	19 (7)	81 (29)
What was being studied was cancer and you or a family member had this disease	209 (76)	167 (61)	10 (4)	99 (36)
The biobank was run by a Native American organization	207 (74)	164 (59)	14 (5)	100 (36)
The biobank was run by a local cancer research center	174 (63)	179 (65)	21 (8)	76 (28)
You know that your biospecimens are sent to research institutes you have never heard of	87 (32)	174 (63)	67 (24)	35 (13)
The biobank was run by a university	157 (57)	189 (69)	23 (8)	62 (23)
The biobank was run by the Indian Health Service	164 (60)	175 (64)	25 (9)	73 (27)
You knew why you are asked to donate biospecimens	174 (64)	174 (64)	22 (8)	77 (28)
The biobank was run by the federal governments but not Indian Health Service	81 (29)	165 (60)	74 (27)	36 (13)
Personal information that could be used to identify you was not collected with your biospecimens	142 (52)	165 (60)	43 (16)	66 (24)

¹Willing = Very willing, Somewhat willing for general willingness; or Very likely, Somewhat likely for vignette-specific willingness.

²McNemar's test showed the additional information provided in each vignette had a significant effect on willingness to donate (all p-values < 0.05).

³Compared to general willingness to donate biospecimen, without additional information.

(Woodbury et al., 2019; Haring et al., 2018; Hiratsuka et al., 2012). In our study, community involvement and approval impacted purported willingness to donate, as did trust in the researchers and institutions involved in the research. Spiritual beliefs were also important and, to some extent, health conditions.

A striking finding from our study is that willingness to donate biospecimens decreased if the federal government, but not the Indian Health Service, operated the biobank and increased if a Native American organization operated it. These results echo an earlier study of the conditions under which AI/AN people would participate in a genetic study, with a decrease in willingness associated with the federal government leading the study (Buchwald et al., 2006). This is not surprising, given the long history of mistrust of the federal government by AI/AN people (Pacheco et al., 2013). Moreover, these attitudes do not bode well for a massive U.S. effort called, “All of Us,” which is designed to study how biology, lifestyle, and environment affect health (The Precision Medicine Initiative, 2020). However, AI/AN communities were slow to be consulted and have been wary of participating (Kaiser, 2019). In contrast, organizations such as the Alaska Area Specimen Bank that is co-owned by the Alaska Native Tribal Health Consortium and federal and state agencies (Hiratsuka et al., 2012) may hold greater promise for engaging AI/AN people in research involving biospecimens.

6. Strengths and limitations

One of the strengths of our study is the large number of urban and reservation dwelling AI/AN participants compared with prior studies which were primarily qualitative investigations involving focus groups and key informant interviews (Aramoana and Koea, 2019). Those studies provided important information about community perceptions and concerns about biobanking and biospecimen donation, albeit with a smaller number of people. In contrast, our survey research was focused on individual attitudes and factors that might influence, or change, willingness to donate, and identified specific ways in which low AI/AN participation in research involving biospecimens might be mitigated.

Other characteristics of the study—the recruitment method and the nature of the sample—could be viewed as either strengths or limitations. We were able to recruit a large number of AI/AN participants in a relatively short period of time through convenience sampling, but we cannot claim that our sample is representative of the AI/AN population. Zip codes varied widely among survey participants and an analysis of associations between willingness and urban and reservation were not meaningful. Additionally, participants presumably had various tribal affiliations rather than being associated with a single tribe. On the one hand, this could increase generalizability but on the other hand, we do not know if individual concerns and attitudes were tribe specific. The cross-sectional survey was also limited to measuring willingness to donate rather than actual donation of biospecimens. However, 92% respondents had never been asked to donate biospecimens.

7. Conclusions

This study, one of the few quantitative studies ever conducted, advances the literature by focusing on individual characteristics and circumstances to consider when attempting to enroll AI/AN people in research involving biospecimen donation, and provides a roadmap for the development of interventions to increase participation with the ultimate goal of reducing health disparities.

CRedit authorship contribution statement

Ka’imi A. Sinclair: Project administration, Writing - original draft. **Clemma Muller:** Methodology, Writing - original draft. **Carolyn Noonan:** Formal analysis, Writing - original draft. **Cathryn Booth-LaForce:** Writing - original draft. **Dedra S. Buchwald:** Conceptualization, Funding acquisition.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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