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Community Engagement, Trust, and Genetic Testing for Inherited Diseases Among Adopted Persons

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Abstract

We identify concerns pertinent to establishing trust necessary to support adoptees' confidence in and uptake of genetic testing that might allow them to benefit from early screening and medical intervention. Using principles of community-based participatory research, our study sought to build a foundation of trust to document such perspectives. Three focus groups were held with 12 adult adoptees. Transcripts were analyzed using thematic content analysis. Comments highlighted aspects related to genomics and health history in the context of adoption, specifically: (1) trust in the intention of the research study; (2) trust that the adoption community will benefit from the results of research; and (3) trust in the protection from misuse and abuse of genomic data. Results reinforce the call for proper security of data and oversight of the ways it is used and point to the value of including researchers who are a part of the community under study.

Introduction

Historical and contemporary research about how to provide diverse populations with the greatest amount of benefit and fewest risks in health research has demonstrated the important role of trust in that context (Boulware, Cooper, Ratner, LaVeist, & Powe, 2016; Calnan, Rowe, & Gilson, 2006; Cunningham-Burley, 2006; King, 2003; McDonald, Townsend, Cox, Paterson, & Lafrenière, 2008; K. Whetten, Leserman, R. Whetten, Ostermann, Thielman, Swartz, & Stangl, 2006). Those working in public health are very familiar with the important role trust plays in a variety of public health initiatives, including both childhood and seasonal flu vaccination, validity of screening tests such as sexually transmitted infection testing and mammography, food safety compliance, and the use of quarantine to protect healthy populations from those with highly contagious illnesses.

The construct of trust has been particularly relevant in the context of genetic testing, for reasons both warranted and unwarranted. These include past actual misuse of genetic science as illustrated by misguided attempts at eugenics, the highly individual and private nature of the data obtained from this research context, and (often exaggerated or inaccurate) popular portrayals of misuse of genetic science in movies such as "Gattaca" (Baldi, Baronio, De Cristofaro, Gasti, & Tsudik, 2011; Ellis, 2003; Erlich & Narayanan, 2014; Gymrek, McGuire, Golan, Halperin, & Erlich, 2013; Kevles, 1985; Shakespeare, 1998). These challenges to public trust in the context of genetics are exacerbated in some historically underserved communities by closed scientific perspectives that have allowed initially well-meaning researchers to engage in what can only be described as scandalous research. Perhaps the best example of this can be seen in the infamous Tuskegee Syphilis Study, whose initial purpose was one of documenting the natural history of syphilis in order to facilitate research toward treatment, transforming into a project that denied known effective treatment to study subjects without consent or knowledge of the nature of their participation (Centers for Disease Control and Prevention, 2013; Gallagher, 2014; Katz, Conguista, Tien, Chaetcuti, McGowan, Lee, Casagli, Hill, & Dearing, 2016).

The forms of trust that may be relevant in genetic research include trust in the accuracy of genetic testing tools, the intentions of the research and scientific community, safety of private genetic information from abuse or misuse, and the confidentiality of genetic information in a variety of contexts, including insurance (Badzek, Henaghan, Turner, & Monsen, 2013; Choudhury, Fishman, McGowan, & Juengst, 2014; Erlich & Narayanan, 2014; Faulks & Feldman, 2016; Gaskell & Bauer, 2013; Green, Berg, Grody, Kalia, Korf, Martin, McGuire, Nussbaum, O'Daniel, Ormond, Rehm, Watson, Williams, & Bieseker, 2013; Henderson, Wolf, Kuczynski, Joffe, Sharp, Parsons, Knoppers, Yu, & Appelbaum, 2014; Wolf, Annas, & Elias, 2013). Each community has different lived experiences, identities, cultural values, and norms, and therefore it is important to understand each

community's perspectives about what forms of trust are most relevant to address in the context of genetics research. Among the initial communities that should be engaged in this dialogue is the adoptee community. This community has been overrepresented in early uptake of genomic services (Baptista, Christensen, Carerer, Broadley, Roberts, Green, & PGen Study Group, 2016), has been characterized as suffering recognizable health disparities that might be addressed through genomic testing and has a variety of relevant psychosocial issues arising from their separation from biological families that will influence trust (May, Evans, Strong, Zusevics, Derse, Jeruzal, Kirschner, Farrell, & Grotevant, 2016).

As we have described in detail in two recent articles published in the *American Journal of Bioethics*, adoptees experience a systematic and unjust disadvantage from lack of access to family health history that should be recognized as a health disparity on grounds consistent with criteria most commonly used to define this phenomena for purposes of health policy (May et al., 2016). In short, due to the nature of many adoptees' separation from biological relatives, adoptees often lack access to the potentially life-saving information most commonly used to identify increased risk for certain inherited diseases. As May, Strong, Khoury, and Evans, 2015, have put forth strongly, this disparity is potentially avoidable through the application of targeted genetic testing.

In order to address this disparity and allow the adoption community to experience the benefits of early identification of disease available to those who do have access to family health history, we must first address issues of trust that the public health literature identifies as influencing uptake of potentially beneficial screening intervention of the type genetic testing represents. Absent trust, rational argument is unlikely to dramatically influence uptake of preventive health interventions (May, 2017). It is toward this goal that we conducted a qualitative study with adult adoptees to identify both potential benefits and concerns about the use of genetic testing to fill gaps in family health history information for adoptees. In this paper we identify those concerns that are most pertinent in establishing the trust necessary to support adoptees' confidence in and uptake of testing that might allow them to benefit from early screening and intervention to mitigate the threats posed by these inherited diseases. An essential process to build this foundation of trust is through the use of community engagement.

Community engagement and trust

"Helicopter" models of research in health and social science fields have been criticized for the disregard of participants' lived experiences, cultural attributes, ability to participate in all aspects of research, and insights in science (Brandon, Isaac, & LaVeist, 2005; Cunningham-Burley, 2006; Gamble, 1993; B.L. Green, Maisiak, Wang, Britt, & Ebeling, 1997; Horowitz, Robinson, & Seifer, 2009; Israel, Schulz, Parker, & Becker, 1998; Israel, Schulz, Parker, Becker, & Community-Campus Partnerships for Health, 2001; Minkler & Wallerstein, 2011; Scharff, Mathews, Jackson, Hoffsuemmer, Martin, & Edwards, 2010). As a result, public health scientists and, more recently, medical care researchers have begun to recognize the value of engaging research participants throughout the research process in order to make it more relevant, meaningful, and sustainable in diverse environments. Although the literature is rife with various terms to describe this approach to research, it is often termed broadly as "community engagement."

Community engagement is not a method used in research, but rather a philosophy of how to conduct science in the context of a diverse global populace. This perspective recognizes that the participants of research have knowledge, experience, ideas, and expertise from their lived experiences in their communities that must be incorporated and valued before, during, and after conducting research studies. There are various forms of community engagement employed by researchers, each with a different set of guiding principles and core tenets. For example, community-based participatory research, which has recently been applied to a range of public health research projects globally, has 11 tenets that outline what is necessary to reach the ultimate goals of this approach to research—that of mutual sharing of responsibilities and benefits, empowerment of communities, and sustainability of outcomes. Most relevant for our purpose, this form of community engagement emphasizes embedding members of the community in the research group itself (Israel et al., 2001; Minkler & Wallerstein, 2011; Wallerstein & Duran, 2008).

Preliminary studies indicate that both researchers and research participants report higher engagement, trust, empowerment, and value gained from research studies that have been conducted using a community engagement approach (Christopher, Watts, McCormick, & Young, 2008; Jagosh, Bush, Salsberg, Macaulay, Greenhalgh, Wong, Cargo, Green, Herbert, & Pluye, 2015; Lantz, Viruell-Fuentes, Israel, Softley, &

Guzman, 2001; Minkler & Wallerstein, 2011; Minkler, Vasquez, Warner, Steussey, & Facente, 2006; Salimi, Shahandeh, Malekafzali, Loori, Kheiltash, Jamshidi, Frouzan, & Majdzadeh, 2012; Viswanathan, Ammerman, Eng, Garlehner, Lohr, Griffith, Rhoades, Samuel-Hodge, Maty, Lux, Webb, Sutton, Swinson, Jackman, & Whitener, 2004). Moreover, studies have reported positive relationship, health, and other social outcomes that have been sustained in communities when the research is done using this approach (Bogart & Uyeda, 2009; Minkler et al., 2006; Rhodes et al., 2014; Viswanathan et al., 2004). One of the more resounding benefits of this approach to research is its ability to establish trust among communities and research partners, which, as aforementioned, is a particularly important element in the inherently contextual project of genetics research (Allen, 1996; Busby, 2006; Frewer, 1999; Siegrist, 2000).

Genetics offers an especially unique context in which to apply community engagement to research, as the findings of genetic tests have implications beyond the individual being tested and bare potential risks for individuals and families that other types of research may not pose for participants (May, 2012). Most recently, community engagement has been implemented in genetic research, particularly when the questions under study involve communities that have experienced breaches of trust, medical or research abuse or mistreatment, and/or discrimination in the form of institutional, interpersonal, or intrapersonal racism (Bonhan, Citrin, Modell, Franklin, Bleicher, & Fleck, 2009; Boyer, Mohatt, Pasker, Drew, & McGlone, 2007; Rangi & Terry, 2014; Terry, Christensen, Metosky, Rudofsky, Deignan, Martinez, Johnson-Moore, & Citrin, 2012; Tobias, Richmond, & Luginaah, 2013; Zusevics, 2013). Adopting this philosophy enables researchers and community members to establish trust and work together to answer inherently personal questions that are only answerable through the use of genetic testing.

When community members are not truly engaged in the research process, the results of research can be devastating to both community groups and the scientific community, which loses the trust, respect, and confidence of many communities (Brandon, et al., 2005; Gamble, 1993; Green, Maisiak, Wang, Britt, & Ebeling, 1997; Horowitz et al., 2009; Israel et al., 2001; Minkler & Wallerstein, 2011; Scharff et al., 2010). A recent controversy involving the Havasupai Tribe in Arizona provides an illustrative example. Initial participation in research was for the genetic components of diabetes, a problem of concern to tribal members. Subsequent research, however, was at odds with broader tribal values. Researchers did not intend to misuse research material, but by not engaging that community throughout the research and dissemination process, a severe loss of trust occurred (Cochran, Marshall, Garci-Downing, Kendall, Cook, McCubbin, & Gover, 2008; Drabiak-Syed, 2010; Mello & Wolf, 2010). Therefore, it is important that those exploring sensitive questions at the cutting edge of science, such as genetics, adopt principles of community engagement in order to understand what that community is most concerned about, what their hopes, desires, values, and identity are, and what forms of trust are most pivotal to establish and ensure that the appropriate safeguards are in place. This assures that science produces benefits for both the science and the broader community within a framework of trust.

The perspectives of the adoptee community about genetics and the risks and benefits they may experience from these medical tools to answer family health-history-related questions may be unique to others and have not been well-documented or explored. In order to obtain data about how to reach this unique population and establish trust, there is a need to engage the adoptee community in this research. Therefore, we conducted a study using principles of community engagement to understand the adoptee community's perspectives on genetic testing research and the role of trust in that context. While the broad results of this study are published elsewhere (Strong, May, McCauley, Kirschner, Jeruzal, Wilson, Zusevics, & Knight, 2017), one important dimension of our findings concerns the significance of the types of community engagement we describe previously. A striking theme that emerged from the data was that of trust. This theme took many forms that were further analyzed for the purposes of this manuscript. Here, we will explore the important trust-related themes that emerged from our focus group analyses, themes that we believe highlight the necessity for sustained community engagement to establish trust in research focused on genetics and adoptees.

Methods

Description of Community-Engaged Approach to Study

This study's origins and execution utilized several tenets of the community engagement approach to research. One of the unique and central elements of the research framework was the

composition of the research team. The idea for the study came from the principal investigator out of his personal experience and identification with the adoptee community (May, 2015), and the inclusion of adoptees in the design and implementation of the focus group discussion guides and analysis. Specifically, two members of the research team are adopted, one being a student at a local academic institution, another having an adopted sister, and another co-investigator is a clinical psychologist at an international adoption clinic who works closely with members of the adoption community. Therefore, stakeholders in the adoption community were engaged in the design, execution, and analysis of the research from several perspectives. Although not a condition for all forms of community engagement, some, such as CBPR, emphasize the importance of having research team members be members of the communities with which they are conducting research (Minkler & Wallerstein, 2011; Wallerstein & Duran, 2008).

We conducted focus groups with adult adoptees to answer the questions of interest. We held four focus group sessions in the Milwaukee metro area between the months of April and December 2013. The size of each focus group ranged from three to five individuals. Focus groups lasted about 90 minutes and were facilitated by at least two study team members. Detailed information about the focus group guide contents are described elsewhere (Strong et al., 2017). The focus groups were recorded, transcribed, and de-identified for analysis. The Institutional Review Board of the Medical College of Wisconsin approved this research study.

Data Analysis

Focus group transcripts were reviewed by the study team researchers multiple times in order to fully review and understand the data prior to analysis. Data analysis began with the categorization of all data as a collective group of research team members. Subsequently, individual team members reduced the data for further in-depth analysis. Standard memoing and coding methods were used for thematic and content analyses of the data (Boyatzis, 1998; Neuendorf, 2016; Onwuegbuzie & Teddlie, 2003). The qualitative methods we used identify participants' perspectives about the use of genomic testing in general and specifically to fill gaps in family health history. All transcripts were analyzed using QSR NVivo 10 qualitative data analysis software.

Participant Demographics

A total of 12 adults who were either internationally or domestically adopted participated in one of the three focus groups representing five countries of origin (U.S., China, Korea, Chile, and Peru). There were two male and 10 female participants in the focus groups. No additional demographic data was collected from participants.

Results

The analysis for this sub-study explored the main component of trust as identified by the larger focus group analysis (Strong et al., 2017). Trust took on different forms among this community of adoptees: (1) trust in the research study, with particular attention to whether the research was guided by individuals with personal connections to adoption; (2) trust that the adoption community will benefit from the results of the research; and (3) trust in the protection from misuse and abuse of genomic data. Supporting quotes from the transcripts illustrate each form of trust are presented in tables followed by brief analyses.

Quotes in the first form of trust theme reflect participants' trust in genetic research projects that are founded on the principles of genuine community engagement (Table 1). Our participants discussed that trust in the research process is essential, particularly in areas of personalized research such as genetics. Several participants shared the risks of potential engagement in genetic research, focusing on breaches of confidentiality, misuse of data, and loss of trusting relationships between researcher and researched. Knowing that individuals with direct connection to the adopted community have participated in this type of research and are part of the research process itself was discussed as a strategy that would help establish and safeguard trust in genetic research. In particular, respondents stated that knowing that some of the members of the research team (whether researchers or participants) are from the community of adoptees supports their trust in the overall research project on genetics.

Moreover, research participants were interested in being able to engage with other adoptees who have been part of genomic research in order to talk with them (other members of their community) about their experiences with this form of testing. According to our participants, being able to discuss with other adoptees who have gone through the process about their experience would help establish trust and answer questions from those who have similar lived experiences and

Table 1. Supporting Quotes for Form of Trust 1

FORM OF TRUST 1: Trust in the intention of the research study, with particular attention to whether the research was guided by individuals with personal connections to adoption
"…the fact that there's people involved in this research that are adopted, or that have adopted…that investment helps me to have trust in the project. I think when you have that—somebody involved with it that has a personal investment—it, generally speaking, has a tendency, for me, to want to participate more than, for example, outsiders. An example of that is when some of us are invited to speak with groups, or adoptive families or schools…to be 'used' versus to be 'engaged with,' I feel, like, are two different things. So that…I think that may make a difference in the research, or how it's presented back to you. Saying, 'We're a part of this too. This isn't just a job that we're doing…' but there's a…this is a personal investment of some sort. I think…for me at least, that feels safer."
"I'd rather have somebody who's more interested in me, kinda thing—not just running a business. So if adoptees and the community is in this program, I definitely would feel more comfortable with it. Because there's already that kind of trust bond, being an adoptee community kinda thing."
"So then, I would hope for some sort of a legal agreement between, like, participants and the researchers to…whether it's scripted or something we agree on one-on-one…is what I would want out of this. So as a participant… regardless of what you find, regardless of how difficult it is to bring to us or whatever, I want to have verification that it would be given to me. 'Cause otherwise, if there's not that 100 percent transparency for me, I wouldn't participate."

Table 2. Supporting Quotes for Form of Trust 2**.**

FORM OF TRUST 2: Trust that the adoption community will benefit from the results of research
"…for me comes back to one of the adoption issues of just trust. So if somebody else is gonna have access to all of this…and how much of it is gonna be fully given to me? How much is gonna be withheld? I go into that mindset of knowing, again, someone else is controlling parts of what's mine. And so for me, the easy answer—and without getting too, like, into this easy answer—is I'm just not gonna' to do it. Because it just feels safer that way. And so when you engage in something like this, for me it feels very vulnerable. And so that would be the risk of having that out there; trusting or not trusting how much of it's going to be given to me honestly or withheld for research, or those sort of pieces. And then once I have the information, like, the list of medical conditions or this…then what do I do with it?"
"And I'm sure it can stir up emotions within people. And I think that someone that has a less formed self-identity, and a less positive self-identity, especially relating to adoption… it might not be the best idea at that point. Because who knows what it can do to their self-confidence, and how they're feeling about their identity, and where they came from. And, you know, this is in my history… and that's bad…. so that makes me bad kind of situation. Whereas if you have someone that is more confident and okay with having that information, I think they would do a lot better with hearing those results than the latter…."

contexts for identity formation and decision-making. Again, this reflects the principles and benefits of community engagement in which research participants are able to engage not only with the research team, but with each other in order to inform their decision-making in the context of research (Israel et al., 2001; Minkler & Wallerstein, 2011; Wallerstein & Duran, 2008).

The second form of trust highlighted that among the issues identified by adoptees were concerns that not all results would be available to each individual, and/or that results that were returned would be based on some scientific or agenda other than the desires or needs of the individual tested (Table 2). These representative quotes also highlight some of the participants' skepticism about how much weight to put into the results of genomic testing. Adoptees in this study identified concerns with the potential for genetic results eliciting psychological distress if results contradict already formed self-identities or reveal potentially damaging health risk factors/outcomes. Respondents relayed the importance of self-awareness and readiness when it comes to genetic testing among adoptees whose self-identity may be developing or fluctuate drastically based on what might be revealed through genetic testing. Participants value being able to trust that results are accurate so that negative influences on self-identity or reproductive decision-making are minimized.

In order for adoptee community members to participate in this research, it is important that they be provided with enough education about the current state of the science and the breadth, scope, and reliability of any test results they may receive. This has direct implications for informed consent procedures and documents that must include language/explanations about the limitations of the science, discuss issues related to follow-up about

new discoveries and whether or not re-contact takes place if new tests reveal that initial results were wrong or provided only partial answers, how and with whom results would be revealed, and how the results will be reviewed with participants upon release. These forms of trust are not unique to the community of adoptees, but have resounded from others who have engaged in genetic testing in the past (Bonham et al., 2009; Boyer, Mohatt, Pasker, Drew, & McGlone, 2007; Cochran et al., 2008; Drabiak-Syed, 2010; Mello & Wolf, 2010; Rangi & Terry, 2014; Terry et al., 2012; Tobias, Richmond, & Luginaah, 2013; Zusevics, 2013).

Finally, the quotes supporting the third form of trust that arose in this analysis demonstrate the importance participants placed on trusting that the information from genetic testing would be used ethically, not abused, misused, or shared with others for whom they were not initially intended (Table 3). Adoptees in this study expressed the need to have confidence in how and with whom results would be shared in order to feel safe and protected in the genomic context. They were particularly wary of data going beyond themselves or their doctors to insurance companies that might use it for potentially harmful financial purposes. Respondents expressed the need for adequate informed consent procedures to be in place to support this form of trust in genetic research from the adopted community.

Table 3. Supporting Quotes for Form of Trust 3.

FORM OF TRUST 3: Trust in the protection from misuse and abuse of genomic data
"And so I don't know if something like that is possible with the DNA you take from us. But anything is possible nowadays, especially the way technology advances. So to know that information...that genetic material is secure and is only being used for your purpose, not anybody else's. And I know you have informed consent and things like that, nowadays. But, I mean, it's something that was also just recently in the headlines."
"That information would have to be really, really, really strictly confidential between you and your doctor. And as it is now, insurance companies know everything about you, because that's the only way the doctor can get paid."
"I think that as long as you make the informed consent as detailed as possible, so that people are pre-warned about the results that they're going to give...as long as you cover that basis, then I think that you're fine. Because as long as people have a forewarning about what they're getting into, then they should be able to handle what the results are."

Discussion

The results of this analysis highlight various forms of trust valued in the context of genomic testing for the purposes of filling in potential gaps in family health history by some members of the adoptee community. In some areas, adoptees are no different than other groups when it comes to trust in genetic research. In others, adoptees have different concerns or priorities.

A noteworthy finding that was unique to our study was the value our participants placed on the community-engaged approach we used to learn their perspectives on genetic testing. Specifically, our study points to the importance of having trust in researchers and the research approach in the context of genomics among adoptees. Our participants discussed that trust in the research process is essential, particularly in areas of personalized research such as genomics. This perspective has resounded in other community engagement in other areas of health research (Boulware, et al., 2016; Calnan et al., 2006; Christopher et al., 2008; Cunningham-Burley, 2006; Jagosh et al., 2015; King, 2003; Lantz et al., 2001; McDonald et al., 2008; Minkler & Wallerstein, 2011; Salimi et al., 2012; Viswanathan et al., 2004; Whetten et al., 2006). Adoptees in this study felt that they could trust the technology, safety of their data, and overall research protocol more knowing that members of the research team had direct membership in the adoptee community. Relate this to "hidden agendas" and ulterior motives, which is what we think the quotes are getting at. These concerns, in turn relate to the types of misguided research priorities that can overtake good judgment in cases like that of the Tuskegee Syphilis Study.

Furthermore, the results point to the potential value of including researchers who are a part of the community under study in the research process to establish trust in the research and the researchers. As expressed by our participants, knowing that a part of the research team is part of the adoptee community establishes a genuine connection to the topics and questions being asked and therefore begins to develop a level of trust that is difficult to obtain if research members are completely disconnected from the community of research participants. This demonstrates the value of using community engagement approaches in genomic science.

Similar to research with other populations, the findings from our study suggest a need to establish trust in the release of genomic/genetic test results. In particular, this study highlights the importance of outlining to whom and how test results are

shared. As expressed by previous research and the adoptees in this study, individuals have the understanding that genetic findings can shape and alter an individual's self-identity as well as that of their broader community (Bonham et al., 2009; Boyer et al., 2007; Chestney, 2001; Hoopes, 1990; Rangi & Terry, 2014; Sorosky, Baran, & Pannor, 1975; Terry et al., 2012; Tobias et al., 2013; Winter & Cohen, 2005; Zusevics, 2013). Past research—such as that done with the Havasupai tribe—that overlooks the significant impact genetic test results have on community identity, risks losing trust from research participants (Cochran et al., 2008; Drabiak-Syed, 2010; Mello & Wolf, 2010). Providing education about what genetic information may reveal about an individual, their family, and their community, as well as having safeguards in the informed consent process and the release of test results are approaches that can help build and sustain trust from community members engaging in genomic science.

Our study also reinforces the call for the proper security of genomic data, protections from psychological/emotional consequences, and the oversight of the ways genomic data is being used/shared outside of the research context. Reflecting previous research from different populations about concerns about the misuse of data by employers and insurance companies (Baldi, Baroneio, De Cristofaro, Gasti & Tsudik, 2011; Ellis, 2003; Erlich & Narayanan, 2014; Gymrek, McGuire, Golan, Halperin, & Erlich, 2013; Kevles, 1985; Shakespeare, 1998), the adoptees in this study expressed considerable anxiety about needing to trust that their genomic/genetic data would not be shared with anyone besides them and the research team. As expressed by our participants and those of other studies, many were not aware of the protections from discrimination provided by the Genetic Information Nondiscrimination Act (Allain, Friedman, & Senter, 2012; Feldman, 2012; Parkman, Foland, Anderson, Duquette, Sobotka, Lynn, Nottingham, Dotson, Kolor, & Cox, 2015) and what it does and does not protect. As the data indicate, the participants in this study would want to be able to trust that their genetic tests would not be used against them by employers or insurance companies and that their information would be safely stored. This form of trust calls for informed consent procedures to provide clear identification of the limitations of the Genetic Information Nondiscrimination Act (GINA) of 2008 and a discussion of the possibility of identifiability despite efforts to maintain privacy and confidentiality, which have been identified in research with other communities about genomics (Kevles, 1985; Slaughter, 2008). Examples of breaches of confidentiality in genomic information support some anxiety among various communities (Baldi et al., 2011; Ellis, 2003; Erlich & Narayanan, 2014; Gymrek et al., 2013; Kevles, 1985; Shakespeare, 1998) and therefore should be openly acknowledged and addressed prior to enrolling communities in these studies. In addition, information about GINA should be included in research consent procedures so that participants gain awareness about this legal protection and also understand its shortcomings.

Trusting that appropriate safety measures are in place for the possible negative mental health consequences of genetic testing was also a finding of this research. Adoptees in this study identified concerns with the potential for genetic results eliciting psychological distress if results contradict already formed self-identities or reveal potentially damaging health implications. Although some research has documented increased anxiety/stress from receiving some genetic results for certain populations, these impacts have typically only lasted for the short-term with limited long-term consequences (Broadstock, Michie, & Marteau, 2000; Heshka, Palleschi, Howley, Wilson, & Wells, 2008; Michie, Bobrow, & Marteau, 2001). And yet, the adoptees of our study valued trusting that appropriate services and supports would be available in the case that any negative psychological outcome is experienced after genomic testing. This safety net may be of particular interest to the community of adoptees who may experience identity-related questions due to their unique lived experience separate from their biological family (Chestney, 2001; Hoopes, 1990; Sorosky et al., 1975; Winter & Cohen, 2005).

Conclusion

In our research engaging the adoptee community, the importance of the type of engagement in research described above became striking. Our study has demonstrated the ways in which the importance of both engagement and trust emerged as essential in the context of genetic research with this community. These findings highlight the importance of adopting principles of community engagement in genetic research with diverse communities in order to understand their unique needs of trust in that context and to establish a genuine foundation of trust between researchers and communities.

Previous research has shown how research

team membership in a community in which they are conducting research supports relationship-building, buy-in, participation, sustainability of outcomes, among other positive results (Israel et al., 2001; Minkler & Wallerstein, 2011; Wallerstein & Duran, 2008), many of which were identified by the participants of this study. Although it may take additional time to build relationships and develop research processes that are mutually defined and beneficial, this approach can help build that trust that is so necessary for long-term investment in science from potentially vulnerable communities.

Toward this end, the senior author of this paper, Thomas May, has led a group assembling a collection of stories representing first-hand accounts—written by adoptees—concerning their experiences interacting with the U.S. healthcare system while lacking family health history, to appear as a themed "symposium" in a forthcoming issue of *Narrative Inquiry in Bioethics*. In addition, project personnel have engaged the adoption psychology literature (Strong et al., 2017) as well as attending the University of Massachusetts Amherst Rudd Adoption Research Project's annual New Worlds in Adoption conference as invited speakers. Finally, dissemination of information about our own Genomic Family History for Adoptees project to adoptees in the general population has occurred through articles featuring these issues in newspapers and in radio and media interviews. (Campbell, 2017; Criscione, 2017; Hinds, Shah, & May, 2017, WLRH Hunstville 89.3 FM).

All of these methods reflect attempts to make transparent the goals and progress of the project overall; to continue to refine our understanding of the specific nature of adoptees' lived experience and how genomic technologies might be properly targeted and utilized to optimally benefit this community; to identify concerns that would inhibit some adoptees' willingness to uptake interventions that could, if properly framed, offer substantial benefit; and to engage the adoptee community in a direct and participatory fashion in articulating their own stories and needs. In this, we believe our project reflects the stated values of transparency, engagement, empowerment, and partnership articulated by the White House Precision Medicine Initiative's November, 2015 "Statement of Privacy and Trust Principles"(The Precision Medicine Initiative, 2015).

References

Allain, D.C., Friedman, S., & Senter, L. (2012). Consumer awareness and attitudes about insurance discrimination post enactment of the genetic information nondiscrimination act. *Familial Cancer, 11*(4), 637–644.

Allen, A.L. (1996). Genetic testing, nature, and trust. *Seton Hall Law Review 1997, 27*(*3*), 887–892.

Badzek, L., Henaghan, M., Turner, M., & Monsen, R. (2013). Ethical, legal, and social issues in the translation of genomics into health care. *Journal of Nursing Scholarship, 45*(1), 15–24.

Baldi, P., Baronio, R., De Cristofaro, E., Gasti, P., & Tsudik, G. (2011). Countering Gattaca: Efficient and secure testing of fully-sequenced human genomes. Proceedings of the 18th ACM Conference on Computer and Communications Security, 691–702.

Baptista, N.M., Christensen, K.D., Carere, D.A., Broadley, S.A., Roberts, J.S., Green, R.C., & PGen Study Group (2016). Adopting genetics: Motivations and outcomes of personal genomic testing in adult adoptees. *Genetics in Medicine, 18*(9), 924–932.

Bogart, L.M., & Uyeda, K. (2009). Community-based participatory research: Partnering with communities for effective and sustainable behavioral health interventions. *Health Psychology 28*(4), 391–393.

Bonham, V.L., Citrin, T., Modell, S.M., Franklin, T.H., Bleicher, E.W., & Fleck, L.M. (2009). Community-based dialogue: Engaging communities of color in the United States' genetics policy conversation. *Journal of Health Politics, Policy and Law, 34*(3), 325–359. doi:10.1215/03616878-2009-009.

Boulware, L.E., Cooper, L.A., Ratner, L.E., LaVeist, T.A., & Powe, N.R. (2016). Race and trust in the health care system. *Public Health Reports, 118*(4), 358–365.

Boyatzis, R.E. (1998). *Transforming qualitative information: Thematic analysis and code development*. Thousand Oaks, CA: Sage Publications Inc.

Boyer, B.B., Mohatt, G.V., Pasker, R.L., Drew, E.M., & McGlone, K.K. (2007). Sharing results from complex disease genetics studies: A community based participatory research approach. *International Journal of Circumpolar Health, 66*(1), 19–30.

Brandon, D.T., Isaac, L.A., & LaVeist, T.A. (2005). The legacy of Tuskegee and trust in medical care: Is Tuskegee responsible for race differences in mistrust of medical care? *Journal of the National Medical Association, 97*(7), 951–956.

Broadstock, M., Michie, S., & Marteau, T. (2000). Psychological consequences of predictive genetic testing: A systematic review. *European Journal of Human Genetics, Oct. 8*(10), *731–738.*

Busby, H. (2006). Consent, trust and ethics: Reflections on the findings of an interview based study with people donating blood for genetic research for research within the NHS. *Clinical Ethics, 1*(4), 211–215.

Calnan, M., Rowe, R., & Gilson, L. (2006). Trust in health care: Theoretical perspectives and research needs. *Journal of Health Organization and Management, 20*(5), 359–375.

Campbell, O. (2017, January 17). How adoptees are filling in the holes in their family medical history. *New York Magazine*. Retrieved from https://www.thecut.com/2017/01/how-adoptees-fill-the-gaps-in-their-family-medical-history.html.

Centers for Disease Control and Prevention. (2013). US public health service syphilis study at Tuskegee. Retrieved from https://www.cdc.gov/tuskegee/index.html.

Chestney, E.S. (2001). The right to know one's genetic origin: Can, should, or must a state that extends this right to adoptees extend an analogous right to children conceived with donor gametes. *Texas Law Review 80*(2), 365–391.

Choudhury, S., Fishman, J.R., McGowan, M.L., & Juengst, E.T. (2014). Big data, open science and the brain: Lessons learned from genomics. *Frontiers in Human Neuroscience*. Retrieved from https://www.frontiersin.org/articles/10.3389/fnhum.2014.00239/full.

Christopher, S., Watts, V., McCormick (Alma Knows His Gun), & Young, S. (2008). Building and maintaining trust in a community-based participatory research partnership. *American Journal of Public Health, 98*(8), 1398–1406.

Cochran, P.A., Marshall, C.A., Garcia-Downing, C., Kendall, E., Cook, D., McCubbin, L., & Gover, R.M.S. (2008). Indigenous ways of knowing: Implications for participatory research and community. *American Journal of Public Health, 98*(1), 22–27.

Criscione, W. (2017, June 6). What's in your genes? INLANDER. Retrieved from https://www.inlander.com/spokane/whats-in-your-genes/Content?oid=4276892.

Cunningham-Burley, S. (2006). Public knowledge and public trust. *Community Genetics, 9*(3), 204–210. doi:92658 [pii].

Drabiak-Syed, K. (2010). Lessons from Havasupai tribe v. Arizona State University Board of Regents: Recognizing group, cultural, and dignity harms as legitimate risks warranting integration into research practice. *Journal of Health & Biomedical Law, 6*(2), 175–226.

Ellis, K. (2003). Reinforcing the stigma-the representation of disability in Gattaca. *Australian Screen Education Online,* 31, 111–114.

Erlich, Y., & Narayanan, A. (2014). Routes for breaching and protecting genetic privacy. *Nature Reviews Genetics, 15*(6), 409–421.

Faulks, S.W., & Feldman, S.R. (2016). Genomics, ethics, and compliance, pp. 75–89, in R.A. Norman (ed.), *Personalized, evolutionary, and ecological dermatology.* New York: Springer.

Feldman, E.A. (2012). The genetic information nondiscrimination act (GINA): Public policy and medical practice in the age of personalized medicine. *Journal of General Internal Medicine, 27*(6), 743–746.

Frewer, L. (1999). Risk perception, social trust, and public participation in strategic decision making: Implications for emerging technologies. *Ambio*, *28*(6), 569–574.

Gallagher, A. (2014). Learning from Tuskegee: From moral outrage to integrative ethics. *Nursing Ethics, 21*(2), 125–126.

Gamble, V.N. (1993). A legacy of distrust: African Americans and medical research. *American Journal of Preventive Medicine, 9*(6 Suppl.), 35–38.

Gaskell, G., & Bauer, M.W. (2013). *Genomics and society: Legal, ethical and social dimensions.* London: Routledge.

Green, B.L., Maisiak, R., Wang, M.Q., Britt, M.F., & Ebeling, N. (1997). Participation in health education, health promotion, and health research by African Americans: Effects of the Tuskegee syphilis experiment. *Journal of Health Education, 28*(4), 196–201.

Green, R.C., Berg, J.S., Grody, W.W., Kalia, S.S., Korf, B.R., Martin, C.L., McGuire, A.L., Nussbaum, R.L., O'Daniel, J.M., Ormond, K.E., Rehm, H.L., Watson, M.S., Williams, M.S. & Bieseker, L.G. (2013). ACMG recommendations for reporting of incidental findings in clinical exome and genome sequencing. *Genetics in Medicine, 15*(7), 565–574.

Gymrek, M., McGuire, A.L., Golan, D., Halperin, E., & Erlich, Y. (2013). Identifying personal genomes by surname inference. *Science 339*(6117), 321–324, doi:10.1126/science.1229566.

Henderson, G.E., Wolf, S.M., Kuczynski, K.J., Joffe, S., Sharp, R.R., Parsons, D.W., Knoppers, B.M., Yu, J., & Appelbaum, P.S. (2014). The challenge of informed consent and return of results in translational genomics: Empirical analysis and recommendations. *The Journal of Law, Medicine & Ethics, 42*(3), 344–355.

Heshka, J.T., Palleschi, C., Howley, H., Wilson, B., & Wells, P.S. (2008). A systematic review of perceived risks, psychological and behavioral impacts of genetic testing. *Genetics in Medicine, 10*(1), 19–32.

Hinds, A., Shah, P., & May, T. (2017). "The challenge of not having relatives' health histories," aired September 23, 24, and 25 on Your Health Radio, UNC Family Medicine's weekly health radio show (see https://yourhealthradio.org/2017/09/22/the-challenge-of-not-having-relatives-health-histories/).

Hoopes, J.L. (1990). The psychology of adoption. In D.M. Brodzinsky and M.D. Schechter (Eds.), Adoption and identity formation, pp. 144–166. New York: Oxford University Press.

Horowitz, C.R., Robinson, M., & Seifer, S. (2009). Community-based participatory research from the margin to the mainstream: Are researchers prepared? *Circulation, 119*(19), 2633–2642. doi:10.1161/CIRCULATIONAHA.107.729863.

Israel, B.A., Schulz, A.J., Parker, E.A., & Becker, A.B. (1998). Review of community-based research: Assessing partnership approaches to improve public health. *Annual Review of Public Health, 19*(1), 173–202.

Israel, B.A., Schulz, A.J., Parker, E.A., Becker, A.B., & Community-Campus Partnerships for Health (2001). Community-based participatory research: Policy recommendations for promoting a partnership approach in health research. *Education for Health 14*(2), 182–197. doi:10.1080/13576280110051055.

Jagosh, J., Bush, P.L., Salsberg, J., Macaulay, A.C., Greenhalgh, T., Wong, G., Cargo, M., Green, L.W., Herbert, C.P., & Pluye, P. (2015). A realist evaluation of community-based participatory research: Partnership synergy, trust building and related ripple effects. *BMC Public Health, 15*(1), 725.

Katz, R., Conguista, A., Tien, T., Chetcuti, M., McGowan, R., Lee, J., Casagli, D.B., Hill, K.C., & Dearing, B.A. (2016). Was the development of the mistrust-in-research (MIR)field of study yet another 'legacy of the USPHS syphilis study at Tuskegee'? A Case Study in Passing Ethics Research from 'generation to generation,' *Journal of Healthcare, Science and the Humanities*, *6*(1), 21–35.

Kevles, D.J. (1985). *In the name of eugenics: Genetics and the uses of human heredity.* Cambridge: Harvard University Press.

King, W.D. (2003). Examining African Americans' mistrust of the health care system: Expanding the research question. *Public Health Reports 118*, 366–367.

Lantz, P.M., Viruell-Fuentes, E., Israel, B.A., Softley, D., & Guzman, R. (2001). Can communities and academia work together on public health research? Evaluation results from a community-based participatory research partnership in Detroit. *Journal of Urban Health, 78*(3), 495–507.

May, T. (2012). Rethinking clinical risk for DNA sequencing. *The American Journal of Bioethics, 12*(10), 24–26.

May, T. (2017). The importance of trust in the vaccine safety enterprise. *The American Journal of Bioethics, 17*(4), 1–3.

May, T., Evans, J.P., Strong, K.A., Zusevics, K.L., Derse, A.R., Jeruzal, J., Kirschner, A.L., Farrell, M.H., & Grotevant, H.D. (2016). Issues of "cost, capabilities, and scope" in characterizing adoptees' lack of "genetic-relative family health history" as an avoidable health disparity: Response to open peer commentaries on "does lack of 'genetic-relative family health history' represent a potentially avoidable health disparity for adoptees?". *The American Journal of Bioethics, 16*(12), w4–w8.

May, T., Strong, K.A., Khoury, M.J., & Evans, J.P. (2015). Can targeted genetic testing offer useful health information to adoptees? *Genetics in Medicine, 17*(7), 533–535.

May, T., Strong, K.A., Zusevics, K.L., Jeruzal, J., Farrell, M.H., Kirschner, A.L., Derse, A.R., Evans, J.P., & Grotevant, H.D. (2016). Does lack of "genetic-relative family health history" represent a potentially avoidable health disparity for adoptees? *The American Journal of Bioethics, 16*(12), 33–38.

May, T. (2015). An adoptive parental perspective on personal genomic screening. *Pediatrics, 135*(4), e811–e814.

McDonald, M., Townsend, A., Cox, S.M., Paterson, N.D., & Lafrenière, D. (2008). Trust in health research relationships: Accounts of human subjects. *Journal of Empirical Research on Human Research Ethics, 3*(4), 35–47.

Mello, M.M., & Wolf, L.E. (2010). The Havasupai Indian Tribe case—lessons for research involving stored biologic samples. *New England Journal of Medicine, 363*(3), 204–207.

Michie, S., Bobrow, M., & Marteau, T.M. (2001). Predictive genetic testing in children and adults: A study of emotional impact. *Journal of Medical Genetics, 38*(8), 519–526.

Minkler, M., & Wallerstein, N. (2011). Community-based participatory research for health: From process to outcomes. New York: John Wiley & Sons.

Minkler, M., Vasquez, V.B., Warner, J.R., Steussey, H., & Facente, S. (2006). Sowing the seeds for sustainable change: A community-based participatory research partnership for health promotion in Indiana, USA and its aftermath. H*ealth Promotion International, 21*(4), 293–300.

Neuendorf, K.A. (2016). T*he content analysis guidebook*. Thousand Oaks, CA.: Sage publications.

Onwuegbuzie, A.J., & Teddlie, C. (2003). A framework for analyzing data in mixed methods research. In A. Tashakkori and C. Teddlie (Eds. *Handbook of Mixed Methods in Social and Behavioral Research,* pp. 397–430.

Parkman, A.A., Foland, J., Anderson, B., Duquette, D., Sobotka, H., Lynn, M., Nottingham, S., Dotson, W.D., Kolor, K. & Cox, S.L. (2015). Public awareness of genetic nondiscrimination laws in four states and perceived importance of life insurance protections. *Journal of Genetic Counseling, 24*(3), 512–521.

Rangi, S.K., & Terry, S.F. (2014). Genetic testing and native peoples: The call for community-based participatory research. *Genetic Testing and Molecular Biomarkers, 18*(8), 531–532.

Rhodes, S.D., Mann, L., Alonzo, J., Downs, M., Abraham, C., Miller, C., Stowers, J., Ramsey, B. Simán, F.M., Song, E., Vissman, A.T., Eng, E. & Reboussin, B.A. (2014). CBPR to prevent HIV within racial/ethnic, sexual, and gender minority communities: Successes with long-term sustainability. Innovations in HIV prevention research and practice through community engagement, pp. 135–160, in S. Rhodes, (Ed.), *Innovations in HIV prevention research and practice through community engagement*. New York, NY: Springer.

Salimi, Y., Shahandeh, K., Malekafzali, H., Loori, N., Kheiltash, A., Jamshidi, E., Frouzan, A.S., & Majdzadeh, R. (2012). Is community-based participatory research (CBPR) useful? A systematic review on papers in a decade. *International Journal of Preventive Medicine, 3*(6), 386–393.

Scharff, D.P., Mathews, K.J., Jackson, P., Hoffsuemmer, J., Martin, E., & Edwards, D. (2010). More than Tuskegee: Understanding mistrust about research participation. *Journal of Health Care for the Poor and Underserved, 21*(3), 879–897.

Shakespeare, T. (1998). Choices and rights: Eugenics, genetics and disability equality. *Disability & Society, 13*(5), 665–681.

Siegrist, M. (2000). The influence of trust and perceptions of risks and benefits on the acceptance of gene technology. *Risk Analysis, 20*(2), 195–204.

Slaughter, L.M. (2008). The genetic information nondiscrimination act: Why your personal genetics are still vulnerable to discrimination. *Surgical Clinics of North America, 88*(4), 723–738.

Sorosky, A.D., Baran, A., & Pannor, R. (1975). Identity conflicts in adoptees. *American Journal of Orthopsychiatry, 45*(1), 18–27.

Strong, K.A., May, T., McCauley, M.P., Kirschner, A.L Jeruzal, J., Wilson, S.L., Zusevics, K.L., & Knight, C. (2017). Adult adoptees' attitudes regarding the potential use of genetic information to fill the gap in their family health history. *Adoption & Fostering*, *41*(2), 159–169.

Terry, S.F., Christensen, K.D., Metosky, S., Rudofsky, G., Deignan, K.P., Martinez, H., Johnson-Moore, P., & Citrin, T. (2012). Community engagement about genetic variation research. *Population Health Management, 15*(2), 78–89.

The Precision Medicine Initiative (2015). Precision medicine initiative: Privacy and trust principles. Washington, DC: National Institutes of Health.

Tobias, J.K., Richmond, C.A., & Luginaah, I. (2013). Community-based participatory research (CBPR) with indigenous communities: Producing respectful and reciprocal research. *Journal of Empirical Research on Human Research Ethics, 8*(2), 129–140.

Viswanthan, M., Ammerman, A., Eng, E., Gartlehner, G., Lohr, K.N., Griffith, D., Rhodes, S., Samuel-Hodge, C., Maty, S., Lux, L., Webb, L., Sutton, S.F., Swinson, T., Jackman, A., & Whitener, L. (2004). Community-based participatory research: Assessing the evidence. *Evidence Reports/ Technology Assessments, No. 99*, 1–8. Rockville, MD: Agency for Healthcare Research and Quality.

Wallerstein, N., & Duran, B. (2008). The theoretical, historical, and practice roots of CBPR. In C*ommunity-based participatory research for health: From process to outcomes*, N. Wallerstein and B. Duran (Eds.), pp, 25–46. San Francisco, CA: Josey-Bass.

Whetten, K., Leserman, J., Whetten, R., Ostermann, J., Thielman, N., Swartz, M., & Stangl, D. (2006). Exploring lack of trust in care providers and the government as a barrier to health service use. *American Journal of Public Health, 96*(4), 716–721.

Winter, K., & Cohen, O. (2005). Identity issues for looked after children with no knowledge of their origins: Implications for research and practice. *Adoption & Fostering, 29*(2), 44–52.

WLRH Hunstville 89.3 FM, & May, T. (2017). Adoptees discover more with genomic sequencing (see http://wlrh.org/News/adoptees-discover-more-genomic-sequencing ed).

Wolf, S.M., Annas, G.J., & Elias, S. (2013). Patient autonomy and incidental findings in clinical genomics (Policy Forum: Point Counterpoint)*Science 340*(6136), 1049–1050.

Zusevics, K. (2013). Ancillary care, genomics, and the need and opportunity for community-based participatory research. *The American Journal of Bioethics, 13*(2), 54–56.

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