Electronic informed consent information for residual newborn specimen research: findings from focus groups with diverse populations

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Abstract

We developed a video and an app for obtaining consent about allowing newborn blood spots (NBS) to be used as biospecimen resources for biobanking. Newborn screening programs test for treatable diseases and leave residual biospecimens that can be used in future research activities. We conducted focus groups and interviews with three diverse communities to determine (a) how well the consent tools worked and (b) participant familiarity with NBS. Participants preferred the video and noted that they were unaware that NBS could be used for future research. Providing information about how biospecimens could be used was a key issue.

Keywords Electronic informed consent · Diverse populations · Biobanking · Residual newborn specimens

Introduction

Newborn screening (NBS) programs universally test all newborns in the USA for treatable diseases. These programs began in the 1960s, with the advent of the phenylketonuria assay. Children who have a metabolic or genetic condition that would benefit from early intervention are identified through NBS, when this early identification can prevent severe morbidity or mortality (Waisbren et al. 2003). These programs represent one of the only health screens universally available to all families in the USA, and therefore help address population-level disparities, especially within the context of

rare diseases (Brosco et al. 2015). Once NBS is completed, residual dried bloodspots (DBS) may remain for babies who were screened. At the present time, 40 states retain these DBS biospecimens for three or more months; 24 states retain them for more than 12 months. The DBS biospecimens are similar to other clinical biospecimens, and can be used for a variety of clinical and research activities. As noted in the literature, a wide range of valuable research has been conducted using DBS (Rothwell et al. 2019; Maschke 2008).

A significant value of biobanks created through state-run NBS programs is the broad diversity of the participants; biospecimens (i.e., residual dried bloodspots) can represent

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the whole population of interest in a state or geographical area. Currently, the populations most impacted by health disparities, including racial and ethnic minority communities and other underserved groups, are vastly underrepresented in most biological and health data repositories used for research. In response, efforts to promote precision medicine have made the inclusion of minority and underserved populations a primary goal (National Institutes of Health 2020). To meet this challenge, large population databases and cohort studies are working to enroll underrepresented groups; however, there is an increased need to utilize already existing resources that may be more representative than typical biorepositories. One such resource in the USA are the state NBS programs.

While residual DBS from state NBS programs have great potential for inclusive research, recent developments in Congress are prompting changes in state programs to implement a consent process for the storage and use of these residual DBS. Currently, federal human subjects regulations permit the use of biospecimens obtained from clinical care to be used for research without consent in many circumstances but there is a move in Congress to require that, because of the mandatory nature of the NBS public health program, consent should be obtained for the storage and research use of residual DBS (Office of Human Research Protections (OHRP) 2018a; U.S. Congress 2019; Office for Human Research Protections (OHRP) 2018b). Given this debate, it is appropriate for state-run NBS programs to evaluate models of consent and create strategies to educate parents about residual DBS use in research. Unfortunately, the move to using a full written consent form could result in low permission rates for bloodspot storage and research use, potentially undermining the diversity of representation in a biobank. This issue is especially problematic if populations who are historically underrepresented in research opt out at higher rates, limiting the diversity of the data collection and reducing the ability of the biobank to address research questions relevant to health disparities (Winter et al. 2018). Underrepresented populations may have more significant concerns about research done without explicit consent given prior historical use of samples and data without permission (Lee et al. 2019). Yet those very concerns by underrepresented populations may perpetuate disparities if a consent process leads to underrepresentation in public health

research. Therefore, it is imperative to design an efficient consent process that promotes informed decision-making and fosters trust in the integrity of the system.

We recently completed a study for which we created an electronic informed consent information app and video for use with parents whose babies were undergoing newborn screening in Michigan. The results showed significant improvements in the consent process on overall knowledge outcomes, increased satisfaction, and higher rates of decision-making about biobank participation (Rothwell et al. 2020). However, we noticed trends in the results from our knowledge survey suggesting that our multi-media intervention strategy did not work as well with underrepresented groups. Therefore, the purpose of the current focus group study was to explore how underrepresented populations understood the information contained in the app and video and what we could do to improve their understanding. We also assessed their understanding of the knowledge survey questions to determine how it may need to be adjusted for use as an outcome measure in future trials.

Methods

We conducted four focus groups (FG) and several in-person interviews with three diverse communities in August and September 2019: Hispanic ($\times 2$), Pacific Islander ($\times 1$), and African American ($\times 1$) communities (see Table 1). Participants were parents who had a child within the past year and/or were pregnant. We used phenomenology, as described by Creswell and Poth (2018), as a qualitative framework for planning these discussions so that we could obtain a clear picture of people's current lived experience in understanding the purposes and uses of NBS in clinical and research contexts (Creswell and Poth 2018). The study team worked with the Community Collaboration and Engagement Team (CCET) of the University's Center for Clinical & Translation Science's (CCTS). Based on our documents and research interests, the CCET developed study materials including the informed consent form, the FG/interview discussion questions, a brief explanation of the study that was shared with participants before the FG/interview along with the questions, and a brief survey

Table 1 Participant demographics and tool preference

Community	No. of participants	Age range	Focus group (FG) or individual interview	Tool preference
African American	6 (5 women; 1 man)	24–39 years	3 FG participants; 3 interview participants	Video
Hispanic	16 (14 women; 2 men)	22–41 years	16 FG participants	Video
Pacific Islander	9 (5 women; 4 men)	24–33 years	9 FG participants	Video

evaluating the information from the video and app; all documents were translated into Spanish as well. Participants were recruited by leaders of their communities, who regularly collaborate with the CCET.

Focus group and interview participants (1) reviewed the informed consent, (2) watched a video and used an app about the use of residual DBS for research, (3) responded to FG questions, and (4) completed and discussed a brief knowledge survey about the content in the video and app they had reviewed. Four broad questions were asked at each FG/interview:

- Based on your experience with the app or video, how would you describe newborn screening? What are two topics mentioned that stood out to you?
- Where and how would you like to use these tools? Which do you prefer (the video or the app) and why?
- Would you consent to the use of your child's leftover blood spots in research? Why or why not? What additional information would you need to help you make a decision to consent?
- What are your thoughts on the survey you took after using the tools?

Members of the CCET conducted the FGs and two other study team members observed the discussions and interactions; a trained Spanish-speaking community leader co-lead the Hispanic FGs. Due to low turnout for the African American FG, three individual interviews were also conducted. Ethics approval was obtained through the University of Utah's Institutional Review Board.

Results

Information obtained during these qualitative conversations is divided into three components: demographics, themes from the larger focus group and individual interview discussions, and survey discussion results about the video and app. These components are presented below.

Demographics

Overall, 31 individuals participated in either a focus group or an individual interview. Table 1 presents basic demographic information. A majority of participants were women whose ages ranged between 22 and 41.

Participants were asked when they would prefer to use the video and/or app, if they had a preference between the two, and why. Across the groups, participants noted a preference for providing the information at prenatal visits, clinics, and doctors' offices. They indicated that the information would need to be provided multiple

times. Participants noted that using social media as well as motherhood magazines would be additional options for disseminating information about DBS and research using DBS. Participants felt that the video presentation was clearer and stated that visuals, such as the video, were informative for them. Several participants also commented that they do not like to read and, therefore, preferred the video, which was appealing since they could share through social media venues. Participants noted that more people from diverse backgrounds should be seen in the videos.

Overarching themes from the focus group discussions and individual interviews

For the focus group discussions, participants were asked two overarching questions: "Based on your experience with the app or video, how would you describe newborn screening? What are two topics mentioned that stood out to you?"

In response to the first question, the communities differed somewhat. The African American participants found the information to be unclear and wanted more information about what was being tested. They did not know that NBS were being retained at all. They indicated that they would not be supportive of having the blood spots kept until they were clear about what was being tested for, how the results of the research helped people in the past, what the consequences would be of saying no to the retention of blood spots, and the purpose of any research using the bloodspots. These participants indicated that they would allow the blood spots to be used if the research being completed assisted their child in the future and/or benefited African American communities.

Participants from the Hispanic community noted that they understood the purpose of the blood spots for newborn screening, but were unaware that specimens were leftover once clinical tests were complete. They also mentioned not knowing that the blood spots could be used in research and to help other individuals. Participants thought this information should be presented to people prior to the birth of a baby and that the blood spots could be used, but only if the DBS were "extra" blood. There was some concern about the "DNA" from the blood spots and how that might be obtained and used by researchers. These participants wanted more detail about the research purpose as well as how the information would be used to help others.

Pacific Islander participants found the purpose of NBS very clear, in that babies received a blood stick in the heel to check for diseases in children. They were not aware that the blood spots were saved and could be used for other purposes. There was agreement overall that NBS could be used for research.

Survey findings

After viewing the video and app (tools), and prior to the FG/interviews, participants were asked to complete a survey with knowledge questions about the topics presented in the tools. This survey had previously been used in a clinical trial to assess how well the tools improved users' knowledge compared to the standard educational materials provided for the Michigan BioTrust, a residual DBS biobank in Michigan. The purpose of including the survey in these FG/interviews was to assess minority community understanding of the questions on the survey and determine its utility when assessing efficacy of the app and video in underserved populations. The question participants responded to was "What are your thoughts on the survey you took after using the tools?"

The participants all noted that the survey was too long and unclear to them. They stated that some of the questions felt like trick questions because of the language as well as the length of the survey questions themselves. Participants would prefer "yes/no" or "true/false" responses to the questions as opposed to the 5-item Likert scale responses.

Participants noted that simpler words should be used to ask about people's knowledge from the video and app. The Hispanic community participants noted that some of the words were unclear to them, i.e., "biobank." In addition, the translation was not clear to all Spanish-speaking participants, who were from different regions of Central and South America, and used different Spanish dialects.

Discussion

This qualitative study identified areas that need to be addressed in order for minority communities to be adequately informed about the importance of using DBS from NBS. Educational programs will need to be developed to provide information about what NBS is, how the residual DBS are stored, and how they can be used for future research. Several issues were raised about how well the app and video educated participants about NBS and the use of DBS in research. Most participants noted they would prefer to use the video rather than the app because the visuals in the video contributed to their understanding more than the text in the app. Therefore, a video tool in the appropriate language would be very useful for educational purposes. However, the knowledge survey we used in the previous study was clearly too complex, and the questions and answers about the app and video were unclear to the participants. Thus, the survey will need revision prior to use in future research studies in order to ensure we are collecting accurate data from minority community participants. We noted particular items that participants indicated were problematic and will begin revisions using this information.

In terms of residual DBS, the African American community was skeptical about the purpose of using residual DBS. They wanted clearer guidelines about what researchers hope to find by using these biospecimens. In addition, this community was concerned about how the data from these biospecimens would impact the health of their community. For these communities, education about biobanking and the residual DBS from NBS programs will need to be developed. Having individuals from African American communities involved in developing educational materials and programming and in exploring how to promote biobanking opportunities will be key in moving the use of DBS forward in the USA. One issue that will need to be examined is how to ensure African Americans approve the storage and use of residual DBS for clinical and research purposes. Working with the communities to determine how to appropriately discuss these opportunities will be crucial if DBS are to be made available in a variety of research contexts.

For the Hispanic communities, there was clear understanding about NBS; however, the participants noted that they were not fully aware that these biospecimens could be used to assist in promoting the health of other individuals and in other communities. The genetic determinations that could be made from these biospecimens were of concern to these communities. That said, the Hispanic communities were interested in learning more about the use of residual DBS and would be supportive of clinical and research uses of these biospecimens. Educational materials and programming will be needed to ensure that clear information is provided to Hispanic communities about the use of DBS as well as how it links to NBS programs.

The Pacific Islander community seemed clearer about the purpose of NBS and that these blood spots were used to identify diseases in children. However, this group was also unaware that the biospecimens could be used to study disease in the future. Again, more educational options that are culturally appropriate and understandable will need to be provided for the Pacific Islander community.

Conclusion

Overall, parents from these three communities were interested in NBS and how the use of DBS could aid in future research activities. Each community noted needing more information about the overall purpose of the future research and how the findings from that research would be used to improve healthcare in their communities. Educational interventions will need to focus on how DBS can support both the clinical and research realms so that all communities benefit from the knowledge. State-organized efforts to educate people about the purpose of NBS, how DBS would be used and why, and what types of research would be conducted with the

biospecimens would be crucial in forwarding the development of biobanks with residual DBS from diverse populations. These types of concerns could be addressed at physician's offices, in clinic settings, and in motherhood magazines. Because it is difficult to add burden to the prenatal care environment, it would be beneficial to spread the word through sources particular communities find useful. As noted by the Hispanic communities, social media would be a method through which to educate communities; however, this avenue should be considered for all communities for information sharing. The African American group noted that churches would be an effective mechanism through which to share information. Finally, the "Island Wave Podcast" reaches the Pacific Islander communities in Utah. Including stakeholders from diverse communities in the development of these educational opportunities, in identifying the best "advertising" methods, and in the clinical and research settings using the residual DBS will be key to creating an ongoing, community-based discussion that can improve how many people agree to have biospecimens included in a biobanking database. This community-based approach should be considered for all research.

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Compliance with ethical standards

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

Ethical approval This article does not contain any studies with animals performed by any of the authors. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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