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Policy Issues and Stakeholder Concerns Regarding the Storage and Use of Residual Newborn Dried Blood Samples for Research

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Abstract

Newborn screening is an important public health programs in the United States. Over 4 million infants are screened each year for a number of conditions. There is a growing need for more explicit state policies governing the storage and research use of residual newborn samples. This paper provides an overview of newborn screening and issues related to policies of residual newborn samples as well as attitudes and opinions from stakeholders. Three groups (n = 21) were conducted with stakeholders: an African American group, a Pediatrician group and a Mothers of young children group. Despite the differences between these groups, consistent themes emerged from all groups that may be relevant for policy development governing the storage and use of residual newborn samples. The data from this exploratory study suggest that future policy developments with the newborn screening program warrant further public input on these topics.

Keywords

Newborn screening; residual biospecimens; public health; focus groups

Newborn screening is a valuable public health program that has led to significant reductions in the mortality and morbidity of newborns. Because newborn screening is regulated by each state, there is no national legislation governing it. Recently, lawsuits have arisen over the storage and secondary research on residual newborn dried blood samples (DBS). The lack of uniform guidelines among states on policies for the storage and use of DBS raises several ethical, legal, and social dilemmas (Kharaboyan, Avard, & Knoppers, 2004). An American Academy of Pediatrics (AAP) task force recommended that the process of developing policies for newborn screening programs should include concerns and opinions of key stakeholders (health professionals, parents, and the public) (AAP, 2000). One method that has been recommended for obtaining opinions of stakeholders for policy developments within newborn screening are focus groups (Hiller, Landenburger, & Natowicz, 1997). To begin this process, focus groups with stakeholders (mothers, providers, and minority group) were conducted to understand similarities and differences in perspectives on the potential use and storage of DBS for future research purposes.

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Background

Newborn screening (NBS) is one of the most important activities of state public health programs. It affects almost all of the four million children born each year in the United States, and collectively, it is the largest application of genetic and metabolic testing (Botkin, 2005). The NBS program began in the early 1960s to test and decrease the catastrophic effects of phenylketonuria (PKU), a condition that causes neurological damage unless identified and treated early through a modified diet. Because of concern for children with undetected PKU and a high risk for mental retardation, a multidimensional advocacy campaign was initiated to lobby for state legislation to mandate NBS and resulted in state laws for almost every state in the 1960s (AAP, 2000). Since then, the NBS program has grown, and because of recent technological and genetic advancements, the number of tests performed has increased from an average of 6 to more than 40, and there is a national trend to continue to expand NBS programs (Botkin et al., 2006; Waisbren, 2008).

In most jurisdictions (except Maryland, Wyoming, and the District of Columbia), the NBS program is conducted without specific informed consent (AAP, 2000). This means that no communication with parents about NBS is required prior to screening. Mandating screening was justified with the argument that the benefits of NBS for PKU are so compelling that states should require screening under their *parens patriae* authority (Faden, Holtzman, & Chwalow, 1982). The belief remains in most state programs that the benefits to newborn screening are sufficiently great that parental permission[1] need not be sought (AAP, 2000).

NBS is conducted around the birth of the child by obtaining a blood sample from a heel stick dried on filter paper. A battery of screening tests is performed, and more blood is taken than necessary for these tests in case reanalysis is needed. This approach results in a leftover blood sample for every child screened. The leftover blood or the residual DBS are stored for variable lengths of time and storage conditions that differ by state (Therrell, Johnson, & Williams, 2006).

Currently, 54% (17 programs) of NBS programs store DBS for 18 years or more (National Newborn Screening and Genetics Resource Center [NNSGRC], 2000). The remaining 34 programs (including the District of Columbia) store them for 3 years or less, and of those, 19 store them for 6 months or less (NNSGRC, 2000). At the national level, the U.S. Department of Health and Human Services created a committee to provide advice and recommendations on policies, technologies, tests, and standards within the NBS program. This committee, the Secretary's Advisory Committee on Heritable Disorders in Newborns and Children (ACHDNC), recommended that each state have a formal written policy to address the storage and use of DBS. As of now, the majority of states do not have a formal policy governing storage and secondary research use on DBS. In spite of the lack of policies, research has already been conducted with DBS that has included infectious diseases, environmental exposures, etiological studies of birth defects and developmental disabilities,

[1]Pediatric ethicists generally prefer the term *permission* when discussing *consent* for interventions with children. This is supposed to emphasize the difference between consenting for oneself and providing permission for someone else. The terms are functionally interchangeable.

and population-based studies for genetic disorders (Olney, Moore, Ojodu, Lindegren, & Hannon, 2006).

Current federal regulations governing human subject protections (45 CFR 46) permit use of these samples for research without parental permission when they are anonymized samples. Research on anonymous residual DBS is considered “nonhuman subjects” research and may be exempt from institutional review board (IRB) approval. The U.S. federal regulations (CFR 46.101) explicitly exempt research on stored biological samples when the sources cannot be identified directly or do not have identifiers linked to the samples. Anonymizing samples is assumed to protect sources from risks and eliminate the need for recontacting donors for consent (Wendler & Emanuel, 2002). The current regulation has permitted the vast majority of secondary research on DBS to be conducted without parental permission and, for the most part, without parental awareness (Kharaboyan et al., 2004).

The ethical concern with the retention and use of DBS is significant. Only Maryland, Wyoming, and the District of Columbia require that NBS be conducted with the informed permission of the parents (AAP, 2000). Parents have the ability to refuse NBS in all but five states for religious or philosophic reasons, but parents are not effectively informed of this option (General Accounting Office, 2003). Parents in all states are provided brochures or information sheets about NBS during the peripartum period, but these materials often are intermingled with other educational materials and samples of infant care items so there is little assurance that parents attend to this information. Furthermore, many brochures do not conform with professional recommendations for content and quality such as clear explanations of the benefits and risk of screening and program policies for storage and secondary use of DBS (Fant, Clark, & Kemper, 2005). A 2005 survey found that only 11% of the brochures from all state programs addressed the issues of storage and use of residual samples (Fant et al., 2005).

Given the interest in DBS for research and the ethical complexities involved, a number of professional organizations have begun to address key policy issues. The Council of Regional Networks for Genetic Services advised in 1996 that use of identifiable samples for research should require contact and permission of parents (Therrell et al., 1996). In 1994, the Institute of Medicine Committee on Assessing Genetic Risks recommended information for parents about storage and informed permission for use of identifiable samples, and anonymous use of samples is permissible without informed permission (Andrews, Fullarton, Holtzman, & Motulsky, 1994). The AAP Newborn Screening Task Force in 2000 suggested that use of anonymous samples without permission was acceptable but that use of identifiable samples needs to meet several criteria: IRB approval, parental permission, a determination that NBS samples are the optimal source of tissue, anonymous samples will not suffice, samples from consenting adults will not suffice, and use should be limited to research addressing conditions affecting children (AAP, 2000). Most recently, the ACHDNC recommended any research outside of the NBS program should include indication of the parents’ awareness and willingness in compliance with federal research requirements (45 CFR 46) (Therrell et al., 2009). These recommendations leave ambiguity as to how this should be accomplished, but indicate it will at least entail an education process with parents regardless of whether the

secondary research on DBS uses anonymous samples or identifiable specimens (American College of Medical Genetics, 2009; Therrell et al., 2009).

Of note, most of these guidelines were not informed by public input on the issues. Failure to invite informed public input into this process of policy developments regarding storage and use of DBS may become problematic. Two lawsuits have arisen in Minnesota and Texas over the retention of DBS samples illustrating the potential tensions and development of mistrust of the NBS program (Citizens' Council on Health Care, 2007; Maschke, 2009). The primary argument for the lawsuits was based on the premise that secondary research use of DBS without parents' consent violated the states' genetic privacy acts. In the case, *Bearder v. State of Minnesota* (2009), the lawsuit was dismissed in favor of the Minnesota Department of Health indicating it did not violate the state's genetic privacy act. In Texas, *Beleno v. Texas Dept. of State Health Services* (2009), the court ruled to destroy stored DBS, and the program will need to include an opt-out option for parents for future storage and research use of DBS. This variability among states gives impetus to develop a line of research to ascertain public opinions and attitudes toward policy development for the storage and secondary research on DBS. To begin this process, focus groups were conducted within the western United States. This project represents initial work for a larger project funded by the National Human Genome Research Institute (R01HG004970-01).

Method

Focus groups were used to address the objectives of this study because they provide a rich research environment to generate data that cannot be attained from one-on-one interviews or questionnaires (Kitzinger, 1995; Morgan, 1988; Stewart, Shamdasani, & Rook, 2007). Specifically, they are useful vehicles for exploring cultural and social dynamic concerns, which is important for complex issues such as NBS (Kitzinger, 1994). These informal group sessions comprise individuals with similar characteristics brought together to discuss their thoughts and beliefs about a specific topic of interest (Krueger, 1994). Focus groups have been used successfully with a number of other studies assessing NBS and genetic issues (Bates, 2005; Davis et al., 2006; Detmar et al., 2007; J. Gamble & Kassardijian, 2008; Wilkinson, 1998).

Participants

A convenience sample of focus group participants were recruited through the aid of local community organizations and contacts within the community. Three focus groups were conducted for this study. The stakeholder groups were chosen based on the researchers' perceptions and experiences of potential groups that may have a more direct impact from policy development around NBS. The first group included 10 individuals who identified themselves as African American. African Americans have a historical negative experience from unethical research that can influence their willingness to engage in future research, especially research that includes demographic identifiers such as race (Corbie-Smith, Thomas, Williams, & Moody-Ayers, 1999; Freimuth et al., 2001; V. N. Gamble, 1997). The second focus group consisted of six pediatricians and one nurse practitioner who worked in a single group practice serving a diverse community in the intermountain west. Pediatric care

providers are directly involved with NBS and are expected to communicate and coordinate NBS results with the parents. The third focus group was conducted with five mothers of young children and, therefore, has the most recent experience with the NBS program itself. These participants all incidentally identified themselves as Caucasian and non-Hispanic. Demographic information such as socioeconomic status, marital status, and age were not recorded, as we did not feel these were relevant because the sample was not intended to be representative. All the groups began with a 10- to 15-minute oral presentation by one of the researchers to provide background information about NBS program policies and practices with regard to storage and research of DBS, and then another researcher experienced in conducting focus groups led the subsequent discussion. Institutional review approval at the corresponding university was obtained prior to the beginning the study.

Data Collection

Focus groups lasted between 1.5 and 2.0 hours. Each group was video-recorded and transcribed by a professional transcriptionist. A member of the research team verified all transcription work. The focus group format followed recommendations by Krueger and Casey (2000) and used a semistructured focus group guide to elicit responses. The semistructured interview guide was developed based on expert opinions and published literature. Questions addressed in the focus groups are listed in Table 1.

Results

Data Analysis

A qualitative descriptive framework was used to address the aims of this study. This framework allows the researcher to sample a broad range of phenomenally or demographically varied cases and to determine the characteristics of a specific experience (Sandelowski, 2000). Congruent with this framework, a qualitative content analysis was used to analyze the data. A distinguishing feature of content analytic approaches is the use of a consistent set of codes to designate data segments that contain similar material (Morgan, 1993). Consistent with previous work (Rothwell & Lamarque, 2010; Rothwell, Sihirath, Badger, Negley, & Piatt, 2008), the codes are generated from the data themselves as directed by the questions from the semistructured interview guide, and instead of using search algorithms, careful readings of the data are performed to code. After coding is complete, the codes are summarized along with recontextualizing of the data to identify patterns (Tesch, 1990). This approach allows for a comparative analysis between different research categories and groups (Morgan & Zhao, 1993) and offers an interpretative approach for emergent meanings participants have toward these issues (Miller & Crabtree, 1992).

Overview of Categories

Five key categories related to issues and concerns about storage and research on residual NBS samples were identified from the transcripts of the focus groups. The categories included the following: the need for informed consent for biomedical research on anonymous or identifiable residual samples, consent for storage of NBS samples, fears of potential discrimination from research on residual samples, lack of perceived individual benefits of anonymous research, and lack of awareness about the NBS program itself. More

research with additional groups is needed to make further generalizations outside the group dynamics of these discussions, but this information is useful for providing a starting point about the attitudes and opinions of potential stakeholders.

Informed consent for research

If the samples were stored and used for biomedical research, participants stated that informed consent would be needed with some type of IRB governing board for protection. Comments also addressed concerns about what the process would entail if they were currently asked for samples of their DNA in a clinical setting for a general research project. Examples included the following: “But I think the whole storing and using without permission is a really slippery slope” (M)[2]; “But now if they wanted to take a DNA sample from you today, would they ask me for consent?” (AA); and “Like an IRB where they’re deciding what studies to allow access like granting money but granting access to these samples” (P). Informed consent was perceived as needed for research on DBS samples because most participants in the groups felt as parents they owned, or should own, the samples of their children. “This is property of the state, but the state is like a nonentity and the people make up the state so we all own it” (AA) and “The parents should be able to choose what they want to do with that” (M). One participant felt that informed consent was needed because screening was not optional (“It is impacted by the fact that it’s not optional to have this drawn” [P]).

Participants felt that informed consent would also establish more communication and control over the type of studies conducted on the samples. Informed consent would also involve state health departments that would help develop and define a process for involving the parents. Examples included the following: “I want to have some control over my property” (AA); “I want to know for sure, you know, what’s going to happen with this specific specimen” (M); and “I think it needs to be guidelined not only by the state department but by the individual” (P).

Informed consent for storage of samples

Participants in this study were also asked about how long residual samples should be stored if no research was planned with them. Most of the participants felt the samples should be destroyed after the testing was complete even if they were anonymous. Examples included the following: “I am uncomfortable with indefinitely” (M); “I’d probably not let them keep it just because of the privacy thing” (P); and “I think well keeping it for a year, fine” (M). Again, as mentioned above, participants stated that informed consent would be needed if the samples were retained (anonymous or with identifiers). Examples included the following: “Why isn’t there any mandatory laws, I mean for how long they can hold it or store it” (M); “I like to have safeguards [for storage of the samples]” (AA); and “I mean we have to get informed consent for everything else that recently is required” (AA).

Interestingly, participants in the pediatrician group differed slightly from participants in the other two groups voicing support for storage indefinitely if the residual NBS samples were

[2]M = Mothers focus group; AA = African American focus group; and P = Pediatrician focus group.

anonymous. Reasons for this perspective included the following: “As you said it’s the entire population. Where else do you get an opportunity” (P); “I mean you’ve got such a great base here” (P); and “At some point that you can look at the development of some disease process over time and be able to make linkages to toxicology or exposure to medicine or drugs or nonmedicine and drugs and all sorts of stuff” (P).

Potential discrimination

Another theme that emerged from the transcripts was the issue of potential discrimination. Individuals repeatedly stated that they were fearful of possible negative consequences of research if they were found to be predisposed to a disease. For example, the following were some of the statements: “You’re not going to get this job because you’re predisposed to or you’re predisposed to whatever” (M); “Saying you have this genetic disease and you know, with all the insurance companies” (AA); and “How do you protect the rights of the patient” (P).

These issues of potential discrimination from biomedical research on these samples focused on the possibility of unknown future genetic and technological advancements. For example, the following statements were made: “future of uncertainty” (AA) and “horror stories about genetic testing and what that could evolve later on down the line” (M). Some of these concerns stemmed from the possibility of technological advances in genetics and that eventually DNA may be identifiable (“twenty years down the road all the genetic stuff is completely mapped” [P]).

Individual benefits of anonymous research

Questions were asked about the potential for research on the residual DBS samples if they were anonymized. Participants questioned the benefit of anonymized research if they could not communicate the results back to the person and that those conducting research on samples would have an obligation to inform the individual about an identified disease or illness. Statements included the following: “how does that benefit me if my kid has a disease and after two months I can’t get contacted” (M); “What is the feedback once a kid has developed something and they recognize something in the testing within two months? What is the feedback? What is the process?” (AA); and “If you find something that’s significant that’s going to affect them you should let them know” (P). Interestingly, participants within the pediatrician group also expressed support for research on anonymous samples for developing a database for epidemiological studies such as identifying environmental toxins and prevalence of diseases or illnesses, but as parents their opinions differed. One statement that expressed this duality included “As a parent I’d want that information back if my child’s blood tested positive for something. As a physician I still think all the numbers [prevalence rates] are valuable regardless of whether or not the parents get any feedback” (P).

Knowledge of NBS

Another theme that emerged from the data was the lack of knowledge of the NBS program. The pediatricians were aware of the NBS program itself because of the fact that they are responsible for conducting follow-ups within NBS such as initial positive results, but only a few participants in the other two groups were aware of the program before participating in

the focus groups. The timing of NBS is a hectic time with the arrival of a baby, and participants recognized this may be a difficult to inform parents about NBS. Examples included the following: “I think they will always have, there will always be some problems [education about testing] with any testing that you do” (P) and “I don’t think I would, you know, truly question anything that’s being done at the time [birth of a child]” (M). Participants felt more should be done to educate people about this program but did not specify one organization as responsible. One participated stated, “I think that is everybody’s job to educate everybody” (M).

Discussion

The focus group findings from this study indicate that participants would prefer informed consent for storage and biomedical research on DBS even if they were anonymous. Participants also indicated that informed consent would be beneficial because it would include involvement and leadership from state health departments. This finding was similar to other studies exploring issues with the retention and research use of biological specimens for research obtained from adults in the clinical setting. For example, most participants were supportive of future unlimited research on anonymous samples when they were asked (Chen et al., 2005; Wendler & Emanuel, 2002). In a study by Chen et al. (2005), more than 85% of the research participants authorized future unlimited research on anonymous samples if they were asked beforehand, and Wendler and Emanuel (2002) found 65.8% of the sample would want consent for research on identified samples derived from clinical settings. These research studies recommended more in-depth research to further understand attitudes and perceptions toward genetic research.

Research on children’s residual DBS samples may have additional concerns and barriers in contrast to considering only one’s personal specimens for genetic research. For example, participants in this study wanted to know if their child tested positive from research on the DBS. This perspective was also evident in the pediatrician’s group. Participants in the pediatrician group acknowledged the uniqueness and value of anonymous population-based research with DBS, but also indicated that as parents they would want to know if their child’s sample was identified as having a disease or harmful exposure.

Future uncertainty of research advancements was indicated as another reason for wanting control and informed consent for research conducted on DBS. Most of the concerns from the participants focused on negative consequences of genetic research such as denying employment or insurance. This has also been supported with research assessing attitudes toward biobanks and unknown implications from future genetic and technological advancements (Clayton, 2003). Last, the majority of participants including health professionals know little about NBS issues. These results tend to underscore the current education practices is low (Davis et al., 2006; Detmar et al., 2007) and the importance for improving and developing new educational tools for parents and professionals (AAP, 2000). The ACHDNC has recommended that education about NBS occur prenatally. This is based on the assumption that the birth of a child is a hectic time and parents are focusing on the new demands of parenthood and therefore are not as attentive to information at this time. The need to improve communication strategies will only increase as NBS programs expand

to screen for additional conditions and as advances in technology and genetics increase opportunities for potential research on DBS.

Implications

There are several limitations to this study. First, single focus groups were used within three different communities/contexts, and future research should conduct multiple focus groups within a target population to gain a more complete understanding of their perspectives surrounding NBS. Also, the lack of knowledge about the NBS screening program may have influenced the results. The presentation delivered at the beginning of the focus groups was the first time most of the participants heard about the NBS program. If the participants were more informed about their state's program and policies regarding this program, they may have expressed different attitudes and opinions during the focus groups. For example, research on attitudes toward biobanks from clinical specimens was supportive of research without consent on anonymous samples (Clayton, 2003). However, these participants were aware of why the specimen was obtained beforehand. Last, although the participants in this study were open to research on DBS with informed consent, this does not indicate that the public is supportive of this possible use. More research is needed using multiple methods of ascertaining public input toward the storage and secondary use of DBS (Hiller et al., 1997). Despite these limitations, this study draws attention to the lack of knowledge and concerns the participants consistently raised about informed consent for storage and research on residual DBS samples.

More research is needed to engage the public on the expansion and future policy developments of the NBS program, especially over the retention of DBS. Future policy developments should address first the lack of awareness of the NBS program and then begin a dialogue about acceptable policies governing storage and use of residual NBS samples with the public. Lack of communication may hinder the development of any policy development for future research by inadvertently giving the perception that parents are not a valued component of the screening process.

Research involving anonymous human biological materials is considered "nonhuman subjects" research, but the participants in this study felt that DNA was personal and that if anonymous research was conducted, they wanted an informed consent process. This finding differs from several recommendations from national organizations in that research on anonymous samples was acceptable without parental consent (AAP, 2000). However, the lack of knowledge about NBS program in general may have influenced attitudes and opinions within these groups. NBS is a complex process because of the numerous issues surrounding this public health program such as why NBS is conducted without parental consent, the type and number of tests screened, and why additional blood is needed to conduct NBS. Improving educational efforts will be difficult, but necessary.

In spite of these concerns, the preliminary results from this study are consistent with recommendations from several national groups in that parents need to be better informed about NBS. Several recommendations have come forth that education of parents about NBS needs to occur prenatally (AAP, 2000; Therrell et al., 2009), but there has not been a study to date that assessed if prenatal education is more effective than the current approach of

educating parents through brochures. Also, prenatal education will involve prenatal care providers at a greater level than before and research is needed to identify the most acceptable approach.

Finally, the development of policies over the storage and research of DBS may cause unwanted consequences within the newborn program itself as well as the complexities and logistical challenges of informing and asking permission of parents for research on DBS. NBS is a state mandated program, and the success of the program is dependent on the population participating to ensure all babies are screened. Additional information about uses of the DBS not associated with NBS may impede participation in the program itself. At the present time, NBS is conducted with minimum parental education. It is unknown how these changes to NBS will affect participation and willingness to allow research on DBS. However, the scientific value of DBS is immense, and without parents' willingness and awareness for research with DBS, lack of trust with this public health program and researchers may occur.

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Table 1

Questions Addressed in the Focus Groups

Knowledge and awareness about NBS
Could any of you please explain any experiences you have had with newborn screening?
As parents (or pediatricians or community members), what was your level of awareness about this program? (If not, how does that make you feel? If yes, how do feel about this process?)
Do you have any concerns about this program?
Anonymous issues
Would you care if these samples were being used for other research purposes if they were anonymous?
Storage
Knowing the current uses and potential benefits for these newborn blood spot samples, what are some of your opinions, comments, and suggestions for storage of them?
How long do you believe they should be stored?
Research of residual samples
What would be some acceptable and unacceptable research purposes for blood samples?
What do you think the process should be for using the residual samples for research purpose?
Community
How would you communicate this issue to your community? How would you actually deliver the information?

Note: NBS = newborn screening.