Published in final edited form as:

J Empir Res Hum Res Ethics. 2016 July ; 11(3): 231–236. doi:10.1177/1556264616656976.

The Influence of Education on Public Trust and Consent Preferences with Residual Newborn Screening Dried Bloodspots

Erin Rothwell, PhD^{1,3}, Bob Wong, PhD¹, Rebecca A. Anderson, PhD², and Jeffrey R. Botkin, MD, MPH^{2,3}

¹University of Utah College of Nursing, Salt Lake City, Utah

²University of Utah, Department of Pediatrics, Salt Lake City, Utah

³University of Utah Division of Medical Ethics and Humanities, Salt Lake City, Utah

Abstract

The objectives of this study were to evaluate the impact of educational interventions during prenatal care on public trust for newborn screening and consent preferences for the retention and use of leftover newborn screening dried bloodspots. Women who were 30–36 weeks pregnant were recruited and outcomes were measured by telephone survey 2–4 weeks postpartum (n=901). Approximately 40% of the sample chose the opt-out approach but those who watched educational interventions delivered during prenatal care were significantly associated with higher levels of trust and support for an opt-out consent approach. Providing education during prenatal care about newborn screening and the storage and use of leftover dried bloodspots along with brochure-based education provided in the hospital when the baby is born, is associated with improved trust for the program and support for research with the leftover bloodspots.

Keywords

Newborn screening; residual newborn screening dried bloodspots; public health; trust; consent; opt-out; prenatal

Introduction

Newborn screening (NBS) is one of the most successful public health programs in the United States in the past 50 years (Centers for Disease Control and Prevention [CDC], 2011). The program has improved the lives of thousands of infants and their families through early treatment and medical management of infants who have been identified with a variety of serious genetic, congenital, infectious, and endocrine conditions. For the most part, NBS is a mandatory program and only parents who have philosophical or religious objections can opt-out of NBS. The mandatory nature of NBS is based on *parens patriae* doctrine and dependent on public trust that the programs will be conducted in an effective and ethical manner.

When NBS is conducted after the birth of the child, enough blood is collected in case re-analysis needs to be conducted. The leftover dried bloodspots are then stored by state health departments for various lengths of time for almost all infants born in the United States. The leftover dried bloodspots are important for quality assurance of NBS tests and for biomedical research. For the most part, this research has been conducted with de-identified dried bloodspots and without parental consent under past federal human subjects regulations (45CFR46.102(f)). However, criticisms have increased because of the retention and use of leftover dried bloodspots for research outside of NBS. Three lawsuits by parents over the lack of adequate information and parental permission of this practice have occurred (Lewis, 2015). More importantly these lawsuits have raised concerns about the loss of the public's trust for NBS programs.

Recent changes to the laws by the Newborn Screening Saves Lives Reauthorization Act, now require parental consent for the storage and use of leftover dried bloodspots ("Newborn Screening Saves Lives Reauthorization Act," 2015). Additionally, the Notice of Proposed Rulemaking is proposing similar changes to the Common Rule. Both require or recommend broad consent for future, unlimited research of biospecimens whether or not they are de-identified. These changes reflect the growing importance of communicating with the public about research practices that extend beyond newborn screening and other clinical settings that collect biospecimens.

The use of informed consent for the retention and use of leftover dried bloodspots has raised concern among some newborn screening stakeholders. One concern in particular is the lack of available samples for the development of new screening tests. California state newborn screening program implemented a pilot supplemental screening program to evaluate the effectiveness of using tandem mass spectrometry (MS/MS) (Feuchbaum, Cunningham, & Sciortino, 2007). This was considered pilot research by the California legislation and as such parental consent was required. Participation by hospitals throughout the state of California varied. Some hospitals refused to participate (20%, n=63) and only 23% of hospitals offered the supplemental screening to greater than 75% of newborns. This resulted in only 52% of the babies born were invited to participate, and only 47% agreed to participate (CDC, 2012).

Furthermore, if hospitals do offer consent, it is not guaranteed that the information presented during the consent process is given in a meaningful manner that promotes an informed decision. The literature provides substantial evidence that the current process to informed consent process is seriously flawed (Falagas, Korbila, Giannopoulou, Kondilis, & Peeppas, 2009). This means that many individuals sign a consent form without an adequate understanding of the study purpose, procedures, risks, and benefits. Within the context of newborn screening, these challenges to comprehension are magnified because of the timing of newborn screening from the demands of labor and needs of a newborn. If an informed consent process is implemented within hospitals, but the process does not promote meaningful opportunities for comprehension then that in itself may actually cause a decline in public trust. (O'Neill, 2004)

Besides the logistics of offering consent in the hospital, there are other factors that influence participation such as trust (Eyal, 2014; Gikonyo, Bejon, Marsh, & Molyneux, 2008). The

literature has demonstrated that distrust among sub-populations have resulted in lack of participation in research (Braunstein, Sherber, Schulman, Ding, & Powe, 2008; Kennedy, Mathis, & Woods, 2007). With the recent lawsuits by parents, maintaining public trust with newborn screening programs is paramount. Thus it is equally important to identify effective education efforts for parents about the retention and research use of leftover dried bloodspots that promote adequate understanding to make a choice about consent for ensuring public trust with these important public health programs.

This study assessed if improved education during prenatal care about NBS and leftover dried blood spots (as opposed to hospital education around the birth of the child through brochures) influenced perceptions of trust and preferences for opt-in/opt-out consent for the storage and use of leftover dried bloodspots. This study was part of larger study that conducted a randomized controlled trial of prenatal newborn screening education interventions in obstetric clinics (n= 901) (1R01HG006266-02).

Methods

The larger study was designed to assess prenatal education about newborn screening and dried bloodspot retention by state health departments following newborn screening. Institutional review board approval was obtained prior to any research activity. In the parent study, participating women in Utah, New York, and California who were 32 to 38 weeks pregnant were randomly assigned to one of three study groups with each group receiving different education materials. There were three groups and of them all received the usual care that included information about NBS and the storage and use of leftover dried bloodspots provided by their prenatal care providers and from the hospital after birth. The two intervention groups additionally viewed educational videos and were given a companion brochure. The NBS group watched one video intervention that was approximately 5 minutes in length about NBS. The other intervention group watched the video about NBS and an additional video about the storage and use of leftover dried bloodspots that was approximately 7 minutes in length. The video education tools were developed by the Genetic Science Learning Center (GSLC) at the University of Utah.

901 participants were randomized to the three groups. The participants were surveyed over the telephone 2 to 4 weeks after the due date of their babies about their knowledge and attitudes about newborn screening and bloodspot retention (approximately 7 weeks after the intervention). Both the videos and the surveys were pilot tested with a separate group of respondents for validity and reliability prior to implementation in this randomized controlled trial. For this study there was an overall follow-up rate of 70% (N=633) (Author, Author, Author, Author et al., 2016).

Measures

Trust was assessed with a five-question survey. Participants were asked about their level of support and trust for the retention and use of leftover dried bloodspots for research purposes (Table 1). Questions were scored and summed to create a measure of trust ranging from 5 – 20 with higher scores indicating more trust. Reliability of the trust survey was good with a

Cronbach's Alpha = 0.83 for the sample. Opt in / opt out was assessed with a single item. Participants choose between: 1) "Use the leftover blood spots only if parents sign a form saying it is OK", or 2) "Use the leftover blood spots unless the parents contact the health department to have their child's blood spot removed".

Analysis

General linear mixed modeling was conducted with SPSS 22 (SPSS, 2006) to assess the relationship of the outcome measure of trust with race, education, and intervention. Race and education were included due to previous research (Boulware, Cooper, Ratner, LaVeist, & Powe, 2003; Richardson, Allen, Xiao, & Vallone, 2012). Logistic regression was used to analyze the relationship of the dichotomized outcome variable of opt in/out with race, education, and educational intervention. Statistical significance was set with an Alpha = 0.05, no corrections for multiple comparisons were made.

Results

Results from mixed modeling indicated the fixed effects of Intervention ($F = 19.68$, $p < 0.001$), Race ($F = 11.01$, $p < 0.001$), and Education ($F = 4.60$, $p < 0.001$) were significantly associated with trust (Table 2). Within Intervention, results show that the NBS and leftover dried bloodspot video groups were significantly higher in trust than both the NBS group only ($p < 0.001$), and the Control group ($p < 0.001$) indicating that those who received the most education (NBS and DBS) reported higher trust scores (Means and standard errors are reported in Table 2).

Analysis of estimated marginal means (post-hoc) indicate that within race: Whites reported the highest trust scored compared to Blacks, Asians, Others, and those that declined to answer their race (all $p < 0.01$). Within Education, post-hoc analyses show that participant's with high self-reported educational levels had significantly higher levels of trust; some college education was significantly higher in trust than participant's with less than high school education ($p = 0.009$), while participant's with Professional or graduate level education had higher levels of trust when compared to the some college education participants ($p = 0.041$).

Opt in/out – Overall our sample indicated 40% ($n = 255$) choose the opt-out approach while 60% ($n=377$) choose the opt-in approach. Results from logistic regression showed that the Video Intervention (Wald = 18.17, $p < 0.001$), Race (Wald = 11.70, $p = 0.039$), and Education (Wald = 11.83, $p = 0.037$) were significantly associated with the opt-out choice (Table 2). Participants that watched both the NBS and DBS video were over 2 times more likely to choose opt-out versus the Control group (OR = 2.367, $p < 0.001$). Participants viewing only the NBS video were not statistically different from the Control group (OR = 1.232, $p = 0.321$). Within Race, Asians were significantly more likely to choose opt-out versus Whites (OR = 2.42, $p = 0.010$). Within Education, those with 'high school or GED level of education' were more likely to endorse opt-out versus participant's with 'some college education' (OR = 2.51, $p = 0.009$).

Discussion

The results of this study indicate that the video educational interventions delivered in this study about NBS and leftover dried bloodspots were associated with significantly higher levels of trust and support for an opt-out consent approach for the storage and use of leftover dried bloodspots. These results are consistent with the primary outcomes of the main study (Author et al., 2016) in that improved education efforts (education about NBS and leftover dried bloodspots during the third trimester of pregnancy along with brochure based education in the hospital during the birth of the child) are significantly associated with higher levels of knowledge, and support for NBS and the storage and use of leftover dried bloodspots for biomedical research.

Interestingly, participants who received *both* NBS and leftover dried bloodspots video education interventions, reported significantly higher support for NBS itself, and the retention and use leftover dried bloodspots for research than those who just received NBS video education only. This is an interesting finding in that more education about NBS along with education about what happens with the leftover bloodspots after NBS is complete is associated with more positive outcomes for both practices. There were concerns that educating parents about what happens with the leftover bloodspots after NBS, along with education about NBS alone, may cause a decline in support and participation within this important public health program (Author et al., 2011). Reasons for this skepticism are that in addition to quality control and assurance purposes, the leftover blood is used for other biomedical research that are not directly related to NBS. For using leftover dried bloodspots for research outside of NBS and without parental permission for NBS itself, concerns were raised that parents may object to NBS all together. Yet, the primary outcome of this research is that when educated about NBS and leftover dried bloodspots at the same time, and provided a choice, parents are more supportive of an opt-out consent approach for the storage and use of leftover dried bloodspots for research and higher trust for NBS.

Due to proposed changes to the Common Rule and the changes to the Newborn Screening Saves Lives Reauthorization Act, this research raises questions about the best way to maximize limited public health and research resources. The new section in the Newborn Screening Saves Lives Reauthorization Act requires that federally funded research on residual newborn screening dried bloodspots will be considered human subjects research and must include explicit written parental consent. In the state of Michigan, where the state health department created the Michigan BioTrust, there is only a 60% parental consent to allow the samples to be used in research (Michigan BioTrust for Health, 2013). This low uptake is not primarily from parental refusal but more from the logistical challenges of obtaining consent at this time in the hospital. Finally, signatures on consent forms can be easily obtained in many clinical circumstances with limited or no comprehension by individuals signing the form.

The question remains regarding the best approach for obtaining consent for the storage and use of leftover dried bloodspots. If state health departments focus on improved education efforts with the option to opt-out, will that result in more informed decision-making? Or should resources be used to obtain a signature with current education efforts recognizing the

difficulties of obtaining consent in the hospital after recovering from birth and with the demands of a newborn? Increase knowledge is associated with increased trust as supported with this research, therefore shouldn't improving education about NBS be more of a priority than just obtaining a signature for use of leftover dried bloodspots for research (Author, Author, Author, Author & Author, 2012)?

Proposed changes to the Common Rule will require consent for research with prospectively collected biospecimens but it is unknown whether these regulatory changes will be adopted and, if so, how they will be implemented. Within the context of NBS programs, the primary goal is to screen infants for early identification of inherited medical conditions so that a timely treatment and intervention can take place. Efforts should focus more on promoting education and transparency about how leftover dried bloodspots are obtained through a mandatory public health program and not only obtaining a parent signature at the hospital as the Notice of Proposed Rule Making seems to be suggesting. We also have an obligation to enhance comprehension. The results also support other research in that prior education levels and race influence trust and consent preferences (Richardson et al., 2012). For example, in a study that assessed consent preferences for biobank participation between Black and White women, results indicated that Black participants wanted study specific information about future studies prior and after donating their biospecimens. (Brown et al., 2016) Further, Black participants stated they would prefer to be asked for permission before the biospecimens was used in a future study, while White participants preferred only a one time broad consent approach.

Both the states of California and Michigan demonstrate the challenges of hospital-based efforts to obtain consent beyond NBS itself. It is not that all parents are refusing participation but they are not adequately provided an opportunity to provide a choice. Efforts to promote trust and transparency through improved education across diverse groups as opposed to a signature will ensure the continued use of important biospecimen research for improving the healthcare of infant and public health.

Best Practices

This study supports the use of video-based prenatal education materials for parents in conjunction with brochures-based education to promote improved trust with newborn screening. Most importantly, when participants have an adequate understanding about *both* newborn screening and the retention and use of leftover bloodspots from newborn screening, increased trust for research occurred. Interestingly, educating parents prenatally about both topics (NBS and leftover dried bloodspots) at the same time is associated with an opt-out consent preference for research with leftover dried bloodspots.

Research Agenda

More research is needed to further investigate parental attitudes on consent preferences for biomedical research with leftover dried bloodspots. This research demonstrated increased support for an opt-out approach when parents received video education interventions prenatally than just brochure-based education and prenatal video education on NBS alone.

Future research will need to examine when an opt-out approach combined with robust educational interventions may be more appropriate and better use of resources especially within public health programs. This future research needs to include a diverse sample of participants to capture differing opinions toward the use of leftover bloodspots used in research.

Education Implications

Video-based education during prenatal care in this study was more effective than brochure-based only education for promoting trust for newborn screening and support for research with leftover dried bloodspots. Communicating complex topics such as newborn screening and retention and use of leftover dried bloodspots for research require more detailed explanations and use of multimedia interventions improve participant comprehension. Exploring methods to deliver this type of intervention on a population level are needed to ensure continued support for newborn screening and research use of leftover dried bloodspots.

Acknowledgments

We would like to thank our colleagues with the parent study of this research and include Louisa Stark, Miriam Kuppermann, Nancy Rose, Siobhan Dolan, and Aaron Goldenberg.

Funding

The parent study was funded by the National Institutes of Health (1R01HG006266-02).

References

- (CDC) Centers for Disease Control Prevention. CDC Grand Rounds: Newborn Screening and Improved Outcomes. Morbidity and Mortality Weekly Report. 2012; 61(21):390–393. [PubMed: 22647744]
- Botkin JR, Goldenberg A, Rothwell E, Anderson R, Lewis MH. Policy guidance for the retention and research use of residual newborn screening bloodspots. Pediatrics. 2012; published online Dec 3. doi: 10.1542/peds.2012-0852
- Botkin JR, Rothwell E, Anderson RA, Rose N, Dolan SM, Kuppermann M, ... Wong B. Prenatal Education of Parents about Newborn Screening and Residual Dried Bloodspots. JAMA Pediatrics. 2016 in press.
- Boulware LE, Cooper LA, Ratner LE, LaVeist TA, Powe NR. Race and trust in the health care system. Public Health Reports. 2003; 118:358–365. [PubMed: 12815085]
- Braunstein JB, Sherber NS, Schulman SP, Ding EL, Powe NR. Race, medical researcher distrust, perceived harm, and willingness to participate in cardiovascular prevention trials. Medicine (Baltimore). 2008; 87(1):1–9. DOI: 10.1097/MD.0b013e3181625d78 [PubMed: 18204365]
- Brown, Katherine M.; Drake, Bettina F.; Gehlert, Sarah; Wolf, Leslie E.; DuBois, James; Seo, Joann; ... Kaphingst, Kimberly A. Differences in preferences for models of consent for biobanks between Black and White women. Journal of Community Genetics. 2016; 7(1):41–49. DOI: 10.1007/s12687-015-0248-y [PubMed: 26304495]
- Eyal N. Using informed consent to save trust. J Med Ethics. 2014; 40(7):437–444. [PubMed: 25180355]
- Falagas ME, Korbila IP, Giannopoulou KP, Kondilis BK, Peeppas G. Informed consent: how much and what do patients understand? Am J Surg. 2009; 198(3):420–435. [PubMed: 19716887]

- Feutchbaum L, Cunningham G, Sciortino S. Questioning the need for informed consent: a case study of California's experience with a pilot newborn screening research project. *Journal of Empirical Research on Human Research Ethics*. 2007; 3:3–14.
- Gikonyo C, Bejon P, Marsh V, Molyneux S. Taking social relationships seriously: lessons learned from the informed consent practices of a vaccine trial on the Kenyan Coast. *Soc Sci Med*. 2008; 67(5): 708–720. DOI: 10.1016/j.socscimed.2008.02.003 [PubMed: 18362046]
- Health, Michigan BioTrust for. Michigan BioTrust for Health: 2013 annual report. 2013. http://michigan.gov/documents/mdch/BioTrust_Annual_Report_2013_FINAL_460368_7.pdf
- Kennedy BR, Mathis CC, Woods AK. African Americans and their distrust of the health care system: healthcare for diverse populations. *J Cult Divers*. 2007; 14(2):56–60. [PubMed: 19175244]
- Lewis MH. Lessons from the residual newborn screening dried blood sample litigation. *Journal of Law, Medicine and Ethics*. 2015; 43(1s):32–35.
- Newborn Screening Saves Lives Reauthorization Act, 113th Congress (2015).
- O'Neill, Onora. Accountability, trust and informed consent in medical practice and research. *Clinical Medicine*. 2004; 4(3):269–276. DOI: 10.7861/clinmedicine.4-3-269 [PubMed: 15244364]
- Prevention, Centers for Disease Control and. Ten Great Public Health Achievements-United States, 2001–2010. *Morbidity and Mortality Weekly Report*. 2011. www.cdc.gov/mmwr/pdf/wk/mm6019.pdf
- Richardson A, Allen JA, Xiao H, Vallone D. Effects of race/ethnicity and socioeconomic status on health information-seeking, confidence, and trust. *J Health Care Poor Underserved*. 2012; 23(4): 1477–1493. DOI: 10.1353/hpu.2012.0181 [PubMed: 23698662]
- Rothwell E, Anderson R, Burbank M, Goldenberg A, Lewis MH, Stark L, ... Botkin JR. Concerns of Newborn Screening Advisory Committee Members Regarding Storage and Use of Residual Newborn Screening Bloodspots. *American Journal of Public Health*. 2011; 101(4):2111–2116. DOI: 10.2105/AJPH.2010.200485 [PubMed: 21330580]
- SPSS. 15.0 Command Syntax Reference. Chicago, Illinois: SPSS Inc; 2006.

Table 1

Survey questions

Trust 1:	In your opinion, do you think it would be alright for health departments to use leftover bloodspots to maintain the quality of newborn screening tests?	(1 = Definitely Alright – 4 = Definitely not alright)
Trust 2:	In your opinion, do you think it would be alright for health departments to use leftover bloodspots to develop new newborn screening tests?	(1 = Definitely Alright – 4 = Definitely not alright)
Trust 3:	Do you think it is alright for these leftover blood samples to be used for important research on diseases that affect mothers and babies?	(1 = Definitely Alright – 4 = Definitely not alright)
Trust 4:	How much do you trust state health departments to use leftover blood spots in an ethical manner?	(1 = I completely trust them – 4 = I don't trust them at all)
Trust 5:	How supportive are you of using these blood spots for research?	(1 = Not supportive at all – 4 = Very supportive)
Opt-In/Opt-Out Question:	Some states store and use leftover blood spots only if the parents sign a form to give permission. Other states allow the leftover blood spots to be used unless the parents contact the health department to remove their child's blood spots from storage. What do you think is the best approach?	_____ Use the leftover blood spots only if parents sign a form saying it is OK _____ Use the leftover blood spots unless the parents contact the health department to have their child's blood spot removed

Table 2

Mixed Model, and Logistic Regression Results

Trust N = 633 Range = 5 – 20 Mean = 16.80 SD = 2.76		N = 632 255 (40%) opt-out VS. 377 (60%) opt-in	
F	Mean (SE)	Wald	OR
Intervention Group Reference = NBS & DBS videos			
	19.68 ***	18.17 ***	
NBS & DBS Video	16.95 (.20)	16.20 ***	2.37
NBS Video	15.90 (.21)	1.05 ***	0.99
Control	15.41 (.21)	1.54 ***	1.23
Race Reference = White			
	11.00 ***	11.70 *	
Asian	15.52 (.38)	1.75 ***	6.63 *
Black/AA	15.71 (.26)	1.56 ***	0.26
Multiracial	16.92 (.42)	0.36	2.80
Others (Alaska/PI)	16.41 (.26)	0.86 **	1.23
White	17.27 (.17)		
Declined	14.71 (.47)	2.56 ***	3.84
Education Reference = Some college			
	4.60 ***	11.83 *	
< high school	15.22 (.40)	1.20 **	0.62
HS grad/GED	15.83 (.28)	0.59	8.53 **
Some college	16.42 (.24)		
AD, CC, VT	15.70 (.34)	0.72	1.63
College grad	16.26 (.22)	0.16	0.29
Prof or graduate	17.10 (.27)	–0.68 *	0.15

* p<.05,

** p<.01,

Author Manuscript

Author Manuscript

Author Manuscript

Author Manuscript

>.001

PI = Pacific Islander, AA = African American, HS = High school, GED = General Equivalence Degree, AD = Associates Degree, CC = Community College degree, VT = Vocational Technology degree