

Public Attitudes Regarding the Use of Residual Newborn Screening Specimens for Research

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KEY WORDS

newborn screening, biobank, informed consent, survey

ABBREVIATIONS

DBS—dried bloodspots

KN—Knowledge Networks

NBS—newborn screening

OR—odds ratio

PRAMS—Pregnancy Risk Assessment Monitoring System

QA—quality assurance

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WHAT'S KNOWN ON THIS SUBJECT: The retention and use of residual bloodspots is a practice of many state newborn screening programs. This practice has become controversial, and little is known about public attitudes on the retention and research use of newborn residual bloodspots.



WHAT THIS STUDY ADDS: This study offers a detailed analysis of public attitudes regarding bloodspot retention and use for biomedical research. The results also offer insights on how education regarding this practice influences support for newborn screening and residual bloodspot use.

abstract



BACKGROUND AND OBJECTIVES: Many state newborn screening (NBS) programs retain residual NBS bloodspots after the completion of screening. Potential uses for residual specimens include laboratory quality assurance, biomedical research, and, rarely, forensic applications. Our objective was to evaluate public opinion about the policies and practices relevant to the retention and use of residual bloodspots for biomedical research.

METHODS: A total of 3855 respondents were recruited using 3 methods: focus groups ($n = 157$), paper or telephone surveys ($n = 1418$), and a Knowledge Networks panel ($n = 2280$). Some participants ($n = 1769$) viewed a 22-minute movie about the retention and use of residual specimens while other participants were provided only written information about this practice. All participants were surveyed using a 38-item questionnaire.

RESULTS: A diverse set of participants was recruited. Respondents were very supportive of NBS in general and accepting of the use of residual bloodspots for important research activities. Respondents were evenly divided on the acceptability of NBS without parental permission, but the majority of respondents supported the use of an “opt-in” process for parental permission for residual bloodspot retention and use. Viewing the educational movie was associated with greater support for bloodspot retention and use.

CONCLUSIONS: Our results show that the general public surveyed here was supportive of NBS and residual sample retention and research use. However, there was a clear preference for an informed permission process for parents regarding these activities. Education about NBS was associated with a higher level of support and may be important to maintain public trust in these important programs. *Pediatrics* 2012;129:231–238

Newborn screening (NBS) is performed by state public health departments on almost all of the 4 million infants born in the United States each year to enable the early identification of certain genetic, metabolic, infectious, and endocrine disorders. Testing is conducted with dried bloodspots (DBS) on filter paper with specimens obtained within the first few days of life. The bloodspots and identifying information are mailed to state NBS laboratories or a commercial partner where testing is performed. Sufficient blood is obtained to enable screening tests to be conducted and to allow for validation of the initial analysis and second-tier testing when indicated.

This approach to screening results in residual bloodspots remaining in the possession of state health departments, at least temporarily, for almost every newborn in the country. These specimens can be used for several purposes, including quality assurance (QA), forensics, and biomedical research.¹ More than 40% of state programs store residual specimens for >1 year, and several states retain them indefinitely.² A number of states have made these residual bloodspots available for biomedical and environmental research for a broad range of projects.³⁻⁶

The storage and use of residual specimens for research have become controversial, primarily because of public concerns over the lack of parental awareness and consent.⁷ Research demonstrates that parents are poorly informed about NBS programs in general, even when conducted with permission, and are largely unfamiliar with the practice of many health departments to retain and use residual bloodspots.^{8,9} Within the last 5 years, 2 states, Texas and Minnesota, have been sued by parents because of the retention of residual bloodspots without parental permission. Texas, in a negotiated settlement with plaintiffs,

agreed to destroy ~5 million retained specimens in 2010.¹⁰ DBS retention has been the subject of a recent Institute of Medicine Roundtable¹¹ and a report by the Secretary's Advisory Committee on Heritable Diseases in Newborns and Children.¹²

In light of the controversial nature of this retention practice, our study was designed to ascertain public opinion on the storage and research use of residual DBS. Given the complexity of this issue and the lack of general public awareness about this practice, we were interested in assessing opinions after the provision of different amounts of information about the topic. Our study used several methods by which public health departments typically garner public opinion, such as focus groups and telephone/paper surveys.

METHODS

Survey Instrument

Our 38-item instrument provided brief written educational information on NBS and sample retention and use to all participants. Twenty-four of the questions assessed participant knowledge and attitudes regarding NBS and the retention and use of residual specimens. Validation for comprehension, clarity, and balance was conducted through a Knowledge Networks (KN) panel of 70 general population participants. Public attitudes were ascertained through 3 methods by using different sets of participants: focus groups, paper/telephone surveys, and the KN panel. The project received institutional review board approval at the University of Utah and the Utah Department of Health.

Focus Groups

Focus groups with members of the general population were held in the states of Texas, Colorado, Utah, New Mexico, Arizona, and Oregon. Commercial survey organizations were used to recruit 15 groups; however, data from 1

group were not recorded due to technical problems and are not included in this analysis. Three online focus groups from a random US sample were conducted through KN. Altogether, data were collected from 17 focus groups consisting of members of the general public (7 groups, $n = 68$), African American subjects (3 groups, $n = 20$), Hispanic subjects (3 groups, $n = 31$), and mothers of young children (4 groups, $n = 38$). All focus groups were shown an educational movie regarding NBS and the retention and use of residual specimens. One of the authors (Dr Rothwell) then led a discussion of the issues with expert support on NBS provided by a pediatrician (Dr Botkin). After the discussion, participants completed the survey.

Telephone/Paper Surveys

Participants were surveyed in Texas, Colorado, Utah, New Mexico, Arizona, Montana, and Nevada using telephone-based and paper instruments. A commercial survey company, Dan Jones and Associates, was employed to conduct the telephone surveys in English. This method is similar to methods used by health departments in their Behavioral Risk Factor Surveillance System surveys of the general population.¹³ African American, Hispanic, Native American, and Asian/Pacific Islander subjects were oversampled.

A subset of telephone/paper survey participants ($n = 200$) were recruited through the Pregnancy Risk Assessment Monitoring System (PRAMS) in Utah with the collaboration of the Utah Department of Health. This program, sponsored by the Centers for Disease Control and Prevention, conducts post-natal surveys of new mothers within 1 year of delivery on health issues relevant to pregnancy, childbirth, and infancy.¹⁴ Our project contracted with the Utah State Health Department to separately implement our instrument by using their standard PRAMS methods.

KN Panel

The third method of survey administration was through KN panels. KN is a company that conducts Internet-based surveys and online focus groups using a large, pre-established panel with a nationally representative probability sample.¹⁵ This approach is a high-technology alternative to traditional survey methods and focus groups. The KN participants were both from the Mountain States region and the national sample. The KN panel was oversampled for individuals who describe themselves as African American, Hispanic, Native American, and mothers of young children (<1 year old) to allow for a meaningful analysis of these groups. To assess the impact of the movie, some KN panel participants were randomly selected to view the movie before completion of the survey. The other KN participants were provided only brief written educational information within the survey instrument.

Educational Intervention

A 22-minute movie was developed for this project by the Genetic Science Learning Center at the University of Utah and is available online.¹⁶ The movie explains NBS and the possibility of the retention and use of residual specimens. The benefits of biomedical research are addressed, as are potential concerns including consent issues and risks of breach of privacy, stigma, and discrimination. The movie was validated through use of a separate KN panel for comprehension and balance. Table 1 indicates the educational intervention provided to each of the survey groups.

Statistical Analysis

General linear modeling was conducted with SPSS 19 (SPSS Inc, Chicago, IL) to assess the relationship between participant responses and the basic

TABLE 1 Survey Method and Educational Intervention

Survey Method	n (%)	Educational Intervention	
		Movie + Written	Written Only
Focus group, paper survey	157 (4.0)	157 (100)	0 (0)
Telephone/mailed paper survey	1418 (36.8)	0 (0)	1418 (100)
KN panel/Internet-based survey	2280 (59.1)	1610 (70.6)	670 (29.4)
Total	3855 (100)	1767 (45.8)	2088 (54.2)

demographic data (gender, age, ethnicity, race, income, parenthood, education, status of being a mother with young children, and residing in the Mountain States region), as well as the type of survey method (telephone/paper, focus group, and KN) and level of information provided. Linear regression was chosen over ordinal regression for ease of interpretation of estimated coefficients. Ordinal regression (results not shown) provided equivalent findings. Logistic regression was used to assess the relationship of the aforementioned variables and the dichotomized outcome questions. For all analyses, an α level of 0.05 was considered statistically significant.

RESULTS

A total of 3855 surveys were collected from March through November 2010. Table 2 shows frequency counts and percentages for all variables of interest. The majority of responses were collected using the KN method (59.1%). Nearly half of the participants viewed the NBS educational movie (45.8%). The participants were primarily non-Hispanic (77.3%), white (61.4%), a parent (77.6%), and female (63.6%). Minority group participation included African American (23.7%), Asian/Native Hawaiian/other (11%), and Native American subjects (3.9%). Mothers of young children comprised 12% of the participants. Recruitment rates varied according to method: 63% for the KN general population sample, 67% for the PRAMS sample, and 5% for the Dan Jones and Associates general population survey.

TABLE 2 Demographic Characteristics of Sample (N = 3855)

Characteristic	n or Mean	% or SD
Age, y	48.73	17.21
Gender		
Male	1404	36.4
Female	2451	63.6
Ethnicity		
Non-Hispanic	2980	77.3
Hispanic	866	22.5
Race		
Native American	143	3.9
Asian/Native Hawaiian/Other	406	11.0
Black or African American	875	23.7
White	2268	61.4
Mountain Region		
No	1542	40.0
Yes	2313	60.0
Mother with young children (<1 y)		
No	3391	88.0
Yes	464	12.0
Parent		
No	840	22.4
Yes	2911	77.6
Income, \$		
<20 000	630	18.5
20 000–<30 000	428	12.5
30 000–<40 000	382	11.2
40 000–<50 000	322	9.4
50 000–<75 000	678	19.9
75 000–<100 000	468	13.7
100 000–<150 000	329	9.6
≥150 000	177	5.2
Education		
Some high school or less	453	12.0
High school diploma or GED	953	25.2
Some college	884	23.4
Associate's degree	345	9.1
Bachelor's degree	726	19.2
Graduate degree	421	11.1

GED, general equivalency diploma.

Survey Responses

The majority of participants (55%) were aware that NBS is done, although focus group discussions revealed that knowledge about NBS is limited. When asked about their level of support for NBS (Table 3), a large majority was either

TABLE 3 General Linear Modeling Results and Effect Sizes

	How Supportive Are You of Health Departments Doing These Blood Tests on All New Infants?		Do You Think It Is Alright That These Tests Are Done Without Permission From the Parents?		How Concerned Would You Be If Health Departments Saved the Leftover Blood Samples From Infants After the Testing Is Done?	
Mean ± SD response	1.45 ± 0.71		2.58 ± 1.18		2.60 ± 1.15	
1–4 Likert scale anchors (% responses)	1: Very (64.5) 2: Somewhat (29) 3: Somewhat not (3.5) 4: Not (3)		1: Definitely (25.5) 2: Probably (24) 3: Probably not (18) 4: Definitely not (32)		1: Not at all (25) 2: Only a little (20) 3: Somewhat (25) 4: Very (30)	
	F	β	F	β	F	β
NBS education	11.05***	−0.13	5.30*	−0.15	61.52***	−0.51
Survey method (baseline = KN)	2.03	–	1.88	–	4.48*	–
Focus groups	–	–	–	–	1.89	−0.16
Telephone/paper	–	–	–	–	7.72**	−0.24
Age	0.26	0.00	1.77	0.00	1.17	0.00
Female	44.16***	−0.18	4.82*	−0.10	0.42	−0.03
Hispanic	0.24	−0.02	3.77	0.14	4.30*	0.14
Race (baseline = white)	1.89	–	2.37	–	11.41***	–
Black/African American	–	–	–	–	29.78***	0.30
Native American	–	–	–	–	4.14*	0.21
Asian/other	–	–	–	–	1.07	−0.09
Mountain Region	1.31	−0.04	0.04	−0.01	0.20	0.03
Mother with young child	1.12	−0.05	0.26	0.04	6.71**	−0.20
Parent	14.26***	−0.12	0.27	−0.03	0.11	0.02
Income	0.00	0.00	2.21	−0.02	1.09	−0.01
Education	12.17***	−0.03	4.41*	−0.03	8.61**	−0.04

F, F test statistic; β, unstandardized regression coefficient; –, not significant.

* $P < .05$;

** $P < .01$;

*** $P < .001$.

“very supportive” (64.5%) or “somewhat supportive” (29%). Only 3% of participants stated that they were “not supportive” of NBS. As Table 3 shows, variables that had a statistically significant effect in the direction of more support for NBS include: viewing the movie ($P < .001$), being female ($P < .001$), being a parent ($P < .001$), and having a higher education level ($P < .001$).

We then asked if it is “alright” to do NBS without the parents’ permission. Our participants were evenly split with 25.5% responding “definitely alright,” 24% “probably alright,” 18% “probably not alright,” and 32% “definitely not alright.” In the regression model, variables that had a significant effect on responses in the direction of higher approval include movie education ($P < .05$), older age ($P < .05$), female gender ($P < .05$), and higher education level ($P < .05$).

Our participants were evenly distributed across a spectrum of concern with respect to the practice of bloodspot retention. Although 25% indicated that they were “not at all concerned” about this practice, 30% were “very concerned.” In the regression model, several variables had a significant effect in the direction of lower concern, including: movie education ($P < .001$), white race compared with African American ($P < .001$), white race compared with Native American ($P < .05$), non-Hispanic ethnicity ($P < .05$), mother of young children ($P < .01$), paper/telephone survey method ($P < .01$), and higher education level ($P < .01$).

As detailed in Tables 4 and 5, we posed 3 questions that addressed potential uses of residual bloodspots, including maintaining the quality of existing tests, research to address diseases affecting mothers and infants, and research addressing diseases that affect the

general public. For each of these questions, ~80% of the participants indicated that these uses were either “definitely” or “probably alright.” Therefore, participants did not make a clear distinction between residual sample use for QA and health research and/or between pediatric research and general research applications.

The survey included a brief vignette for the next question. Participants were asked to imagine that a health department has been saving DBS for 10 years without the permission of parents. Then they were asked to imagine that the health department wanted to use the samples for important health research. We asked: “It may be very difficult and costly to find many parents after several years. If parents cannot be contacted, what would be the best thing to do with their baby’s leftover samples?” The majority of our participants (71%) would allow the samples

TABLE 4 General Linear Modeling Results and Effect Sizes

	In Your Opinion, Should Health Departments Use These Leftover Blood Samples to Maintain the Quality of Existing Tests for Newborns?		Do You Think It Would Be Alright for These Leftover Blood Samples To Be Used for Important Research on Diseases That Affect Mothers and Infants?	
Mean ± response	1.92 ± 0.95		1.72 ± 0.88	
1–4 Likert scale anchors (% responses)	1: Definitely (39) 2: Probably (39) 3: Probably not (12) 4: Definitely not (10)		1: Definitely (50) 2: Probably (36) 3: Probably not (7) 4: Definitely not (7)	
	F	β	F	β
NBS education	56.23***	−0.40	34.67***	−0.28
Survey method (baseline = KN)	9.07***	−	14.62***	−
Focus groups	3.68	−0.19	6.66**	−0.23
Traditional telephone/paper	15.69***	−0.27	24.80***	−0.31
Age	1.93	0.00	0.31	0.00
Female	11.35***	−0.12	8.19**	−0.10
Hispanic	0.94	−0.05	0.52	−0.04
Race (baseline = white)	1.49	−	2.04	−
Black/African American	−	−	−	−
Native American	−	−	−	−
Asian/other	−	−	−	−
Mountain Region	0.70	0.04	0.24	0.02
Mother with young child	5.45*	−0.15	1.40	−0.07
Parent	0.25	−0.02	1.27	−0.04
Income	1.18	0.01	7.10**	0.02
Education	10.44**	−0.04	0.11	0.00

F, F test statistic; β, unstandardized regression coefficient; −, not significant.

* $P < .05$;

** $P < .01$;

*** $P < .001$.

to be used for important health research with privacy safeguards in place (Table 6). Our survey also addressed the potential tradeoff between removing identifiers from samples and the return of results. A short paragraph informed participants that removing identifiers would increase privacy protections but, if identifying information is kept with the samples, important results about the infant's health could be returned to the parents. When asked which approach is more appropriate, 64% responded that allowing notification of parents of results was more important than providing greater privacy protection. A number of variables in our model had a significant effect on this response, as indicated in Table 6. The survey instrument included information explaining that some people believe that state health departments should keep residual samples only if the parents sign a form giving

permission (an opt-in consent model), whereas others believe it is acceptable to retain the samples unless parents contact the health department to have their child's sample removed (an opt-out model). When asked to choose between these approaches, 62% responded that the samples should be kept only if parents sign a form. Variables associated with selection of the opt-out approach included movie education (odds ratio [OR]: 2.02; $P < .001$), older age (OR: 1.01; $P < .001$), and a higher level of education (OR: 1.09; $P < .01$). The final question in the survey returned to the general question of acceptability of sample retention and research use. Our rationale was that completion of the survey instrument itself could be educational by highlighting aspects of policy and practice. We asked: "After thinking about these questions for the last few minutes, we want your final opinion: Do you think it

is alright to use these leftover blood samples for doing important research?" Responses indicated that 81.5% were either "definitely" or "probably alright" with this practice (Table 5). Only 9.5% stated that use of residual samples for research was "definitely not alright."

DISCUSSION

Residual bloodspots are essential for QA purposes for NBS programs but also can be used for a broad range of biomedical research. However, given the limited information readily available to parents about NBS and bloodspot retention and use, it is not surprising that this practice has become controversial. Lay advocates in both Minnesota and Texas characterize the practice in alarmist terms, including the sale of infant t-shirts exclaiming "Help!!! The Govt Has My DNA."¹⁷ Both states have been involved in litigation over this issue, and many states are concerned about public backlash against NBS programs in general.

A number of professional organizations have commented on the retention of DBS. The American Academy of Pediatrics Task Force issued a detailed set of recommendations in 2001.¹ The task force recommended that research with unlinked specimens (not individually identifiable) was appropriate when consistent with the goals of NBS programs and that parents should be informed that residual specimens might be used for QA or epidemiologic research. Furthermore, they recommended that research with identifiable specimens should be conducted with parental permission and only when the DBS are the optimal resource for the proposed research. The Institute of Medicine organized a roundtable discussion in 2010 that highlighted many of the benefits and complexities of QA and research with retained DBS, and speakers emphasized the need for better communication and collaboration

TABLE 5 General Linear Modeling Results and Effect Sizes

	Do You Think It Would Be Alright for These Leftover Blood Samples To Be Used for Important Research on Diseases That Affect the General Public?		Do You Think It Is Alright To Use These Leftover Blood Samples for Doing Important Research?	
Mean ± response	1.83 ± 0.93		1.85 ± 0.93	
1–4 Likert scale anchors (% responses)	1: Definitely (45) 2: Probably (37) 3: Probably not (9) 4: Definitely not (9)		1: Definitely (42.5) 2: Probably (39) 3: Probably not (9) 4: Definitely not (9.5)	
	F	β	F	β
NBS education	37.65***	−0.32	40.75***	−0.33
Survey method (baseline = KN)	12.25***	−	8.79***	−
Focus groups	6.71**	−0.24	4.84*	−0.21
Telephone/paper	19.79***	−0.30	14.21***	−0.25
Age	1.54	0.00	2.35	0.00
Female	1.26	−0.04	7.91**	−0.10
Hispanic	0.84	−0.05	0.04	0.01
Race (baseline = white)	2.74*	−	1.49	−
Black/African American	0.46	−0.03	−	−
Native American	0.67	0.07	−	−
Asian/other	7.09**	−0.19	−	−
Mountain region	0.31	0.03	0.65	−0.04
Mother with young child	1.73	−0.08	1.59	−0.08
Parent	1.42	−0.05	0.35	−0.02
Income	1.42	0.01	2.74	0.01
Education	9.40**	−0.04	2.13	−0.02

F, F test statistic; β, unstandardized regression coefficient; −, not significant.

* P < .05;

** P < .01;

*** P < .001.

with parents to maintain trust in these public health programs.⁸

The Secretary's Advisory Committee on Heritable Diseases in Newborns and Children issued a briefing paper in 2010 addressing policy issues in the retention and use of DBS.⁹ They recommended the development of well-defined strategies and policies, including the promotion of education for parents and health professionals. Both the American College of Medical Genetics and the Association of Public Health Laboratories have issued statements on this topic that emphasize the value of the specimens and the need for new policies to protect the privacy and confidentiality of families.^{18,19}

Research using DBS is generally conducted with de-identified specimens, and we have not identified any reports of harm or breaches of privacy from projects using DBS. Beyond the NBS context, use of residual clinical bio-specimens is a common practice in biomedical research.²⁰ Although the ethical and regulatory issues in biobanking

TABLE 6 Logistic Regression Results

	More Important To Allow Notification of Parents If Something Important Is Learned Versus Provide Greater Privacy Protection		Better To Keep Samples Unless Parents Contact the Health Department To Have Them Destroyed Versus Keep Samples Only If Parents Sign a Form		With Privacy Safeguards in Place, Allow the Samples To Be Used for Important Health Research Versus Do Not Allow the Samples To Be Used for Important Health Research	
Frequency and percentages	2441 (64%) notification vs 1386 (36%) privacy		1448 (38%) opt-out vs 2383 (62%) opt-in		2167 (71%) allow vs 904 (29%) do not allow	
	Wald	OR	Wald	OR	Wald	OR
NBS education	25.21***	1.80	34.43***	2.02	8.83**	1.44
Survey method (baseline = KN)	14.55***	−	6.05*	−	8.00*	−
Focus groups	7.70**	2.04	4.36*	1.56	0.10	1.08
Phone/paper	7.99**	1.55	2.22	1.26	7.63**	0.64
Age	4.92*	1.01	15.59***	1.01	3.04	1.00
Female	6.91**	1.24	2.91	1.15	8.65**	1.29
Hispanic	1.07	0.88	2.88	0.81	0.29	0.93
Race (baseline = white)	17.22***	−	3.94	−	10.49*	−
Black/African American	15.60***	1.52	−	−	0.13	0.96
Native American	0.02	1.03	−	−	2.58	0.73
Asian/other	2.61	1.32	−	−	7.26**	1.63
Mountain region	4.48*	1.27	0.01	0.99	0.94	1.13
Mother with young child	0.31	1.08	0.25	1.08	0.57	1.12
Parent	1.82	0.88	3.69	0.83	0.31	1.06
Income	12.46***	0.93	0.19	0.99	0.54	0.54
Education	11.24***	0.91	8.60**	1.08	4.43*	1.06

Wald, Wald test statistic; −, not significant.

* P < .05;

** P < .01;

*** P < .001.

have been a topic of active discussion, there has been little public controversy over these practices. Public sensitivities may be heightened in the NBS context due to the lack of parental consent or adequate education for the clinical service, the acquisition of specimens from a vulnerable population, a direct role of state government, and the perception that genetic research poses special risks.

A significant concern in the pediatric and public health communities is that greater public awareness of the retention and use of residual specimens will impair the efficacy of NBS.²¹ The worry is that parents will refuse NBS because of concerns over DBS retention and use. Our results demonstrate a high level of support for NBS in general and are consistent with other recent results in the field.^{22,23} Furthermore, we found that support for NBS was enhanced through more in-depth education on the issues. Therefore, our results do not suggest that there is a major conflict between education about NBS and support for NBS generally or sample retention and use specifically.

However, our results also demonstrate that there is a substantial minority of individuals who are highly concerned about this practice. The recent legal cases illustrate that vocal citizens with strongly held opinions can have significant impacts on program policy and function. The litigants in these cases are primarily demanding a parental permission process. We found that despite an evenly divided set of opinions about parental permission for NBS clinical services, a majority of our participants (62%) support an opt-in approach to

DBS retention and use. The focus group discussions clearly demonstrated that participants support NBS and DBS retention and use but that they want more information about both and a choice over the latter. Our results are consistent with those of Tarini et al²⁴ in this regard. It is notable that the American Academy of Pediatrics, the Institute of Medicine, and the Secretary's Advisory Committee on Heritable Diseases in Newborns and Children statements advocate more education and public transparency relevant to this practice and so appear consistent with our measure of public expectations.

We found that the single most consistent independent variable associated with support for DBS use was viewing the educational movie. Despite the strong statistical significance of the association between viewing the movie and support for DBS use, the magnitude of this effect (β) was modest in practical terms. Other variables associated with more support on these particular questions include female gender, survey method, higher education, older age, and being the mother of a young child. These variables also had modest effects. Variables such as race, ethnicity, income, parenthood, and geographic location had no meaningful effect on responses to most questions.

A weakness of the study is that we received approval from only 1 state, Utah, to conduct the PRAMS-like survey, raising uncertainty about the generalizability of data from this important group. Another weakness is a dependence on the movie and the written information in the survey for educating the general public about a complex, unfamiliar topic.

CONCLUSIONS

The general public included in this study had only limited awareness of NBS and essentially no awareness of the retention and use of residual bloodspots. When made aware of the issues, the public was strongly supportive of NBS and was accepting of the retention and research use of these specimens. Acceptability by the public of sample retention and use was enhanced by providing substantive information about the pros and cons of this practice through an educational movie. Although accepting of the use of these valuable resources, the public preferred an opt-in approach to decision-making by new or expectant parents in determining whether residual bloodspots are retained and used. The need for more attention to prenatal education and informed choice is clear. Greater transparency and increased education by public health programs about these policies and practices are likely to be important in maintaining public trust and acceptance for these valuable programs and research activities.

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