Published in final edited form as:

Soc Sci Med. 2016 October ; 166: 17–24. doi:10.1016/j.socscimed.2016.07.036.

Parental Intentions to Enroll Children in a Voluntary Expanded Newborn Screening Program

Ryan S. Paquin,

Center for Communication Science, RTI International, Research Triangle Park, North Carolina

Holly L. Peay,

Center for P-12 Education, RTI International, Research Triangle Park, North Carolina

Lisa M. Gehtland,

Center for P-12 Education, RTI International, Research Triangle Park, North Carolina

Megan A. Lewis, and

Center for Communication Science, RTI International, Research Triangle Park, North Carolina

Donald B. Bailey Jr.

Education and Workforce Development, RTI International, Research Triangle Park, North Carolina

Abstract

Background and Objectives—Nearly all babies in the United States are tested at birth for rare, serious, and treatable disorders through mandatory state newborn screening (NBS). Recently, there have been calls for an expanded, voluntary model to facilitate early diagnosis and treatment of a wider range of disorders. We applied the reasoned action framework to examine parental intentions to participate in voluntary expanded screening.

Methods—We recruited a national cohort of recent and expectant parents living in the U.S. who completed a self-administered online survey ($N = 1,001$). Using a mixed-level fractional factorial experiment, we studied parental participation intentions and preferences for timing of consent, cost, consent format, and testing options.

Results—We conducted a hierarchical regression analysis assessing parental intentions to participate in voluntary expanded NBS. Attitudes, perceived normative influence, and perceived behavioral control explained substantial variance in intention, with perceived normative influence emerging as the strongest predictor. We found no evidence that the manipulated program features altered mean levels of intention, but timing of parental permission, cost, and permission format moderated the relative importance of reasoned action constructs on intention.

Correspondence concerning this article should be addressed to Ryan S. Paquin, Center for Communication Science, RTI International, 701 13th ST NW, Suite 750, Washington, DC 20005-3967. Phone: 202-728-2041. Fax: 202-728-2095. rpaquin@rti.org.

Publisher's Disclaimer: This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final citable form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.

Conclusion—Program design features may impact the psychological mechanisms underlying parental decision making for voluntary expanded screening. These results have important implications for parent education, outreach, and informed parental permission procedures.

Keywords

United States; Expanded newborn screening; Informed decision; Reasoned action; Intentions; Theory of planned behavior

Nationwide endorsement of a standardized uniform newborn screening (NBS) panel has been called one of the great public health achievements of the first decade of the 21st Century, has saved the lives of thousands of children, and improved the quality of life of thousands of others (Centers for Disease Control and Prevention, 2011). NBS in the U.S. began more than 50 years ago, when screening was first offered using a heel stick blood test for phenylketonuria (Ross, 2010). In the ensuing years, NBS in the U.S. has grown into a mature public health program. The early decades of screening were characterized by considerable variability among states in the number and types of conditions included on NBS panels (Watson, Mann, Lloyd-Puryear, Rinaldo, & Howell, 2006). To promote greater harmonization, the Department of Health and Human Services Advisory Committee on Heritable Disorders in Newborns and Children (ACHDNC) provides national guidance on candidate conditions for newborn screening. The ACHDNC conducts a rigorous evidence-based review of nominated conditions, applying four primary considerations: (1) the condition represents a significant public health problem, (2) a low-cost and accurate screening test is available to detect it, (3) treatments with proven efficacy exist, and (4) states are capable of implementing screening and follow-up. The ACHDNC then makes a recommendation to the Secretary of Health and Human Services who determines whether the condition should be added to the Recommended Uniform Screening Panel (RUSP; Kemper et al., 2014). There are now 32 conditions on the RUSP, and all states offer screening for at least 29 disorders.

Historically, conditions placed on the RUSP have required urgent medical intervention, the benefits of which include dramatic improvements in quality of life and life expectancy. With this in mind, state NBS programs are generally mandatory; that is, many are conducted without formal requirements for informed parental permission and with limited options for parents to request an exemption. The moral justification for mandatory NBS largely rests in the promise of these public-health programs to protect child welfare and avert preventable harm (Goldenberg & Sharp, 2012).

Despite the success of NBS as a public health program, an alternative model has been suggested to enhance the potential for early diagnosis and treatment of a wider range of disorders, whereby screening for some conditions would remain mandatory while for others it would be voluntary. That is to say, parents would be free to choose whether or not to have screening for the expanded set of disorders, and this screening would only be done with informed parental permission. Underscoring the need for an expanded, voluntary screening option to augment regular NBS are four factors that have the potential to reintroduce state-by-state screening variability, burden state public health infrastructures, and increase

inequities: (1) the slow, condition-by-condition ACHDNC review process in an era of rapid discovery and treatment potential; (2) frustration by patient advocates, who view the bar for evidence as too high; (3) the rapid emergence of commercial options for screening; and (4) new technologies, such as whole genome sequencing that, if adopted for regular NBS, would open the possibility of detecting many conditions that do not currently meet RUSP criteria (Bailey & Gehtland, 2015).

Although the idea of a voluntary expanded screening program to augment current NBS has been suggested before (Grosse, Boyle, Kenneson, Khoury, & Wilfond, 2006; American Academy of Pediatrics, 2000; President's Council on Bioethics, 2008; Ross, 2010) the social and ethical challenges inherent in implementing state-level public education and establishing a process for informed parental permission remain formidable barriers (Bailey, Skinner, Davis, Whitmarsh, & Powell, 2008). Voluntary expanded screening will require fundamental infrastructure changes to support testing, counseling, education, parental decision making, and follow-up (Dhondt, 2010). Parents tend to support optional population-based screening for conditions not currently part of the RUSP (Bailey, Bishop, Raspa, & Skinner, 2012; Cyrus et al., 2012; Rothwell, Anderson, Swoboda, Stark, & Botkin, 2013; Wood et al., 2014) but theory-driven analyses examining parental intentions to use voluntary NBS are needed to guide program design and to develop educational tools supporting informed decision making.

Here we report an experimental study of recent and expectant parents to determine reactions to a hypothetical voluntary expanded screening program, described as a combined state-level public health and research initiative to test infants for several health conditions not currently part of the RUSP. Such research is vital because the success of expanded NBS depends on the ability of program administrators to build awareness and ensure that parents are given the tools to make informed choices about enrolling their children. To begin addressing the many communication challenges facing a voluntary expanded NBS program, we applied the reasoned action framework to examine the psychosocial mechanisms underlying parental willingness and intentions to have a child participate in expanded NBS.

The Reasoned Action Framework

The *reasoned action framework* is a cumulative model of behavior prediction that subsumes the theory of reasoned action (Fishbein & Ajzen, 1975), the theory of planned behavior (Ajzen, 1985), and the integrative model of behavioral prediction (Fishbein et al., 2001). Like its precursors, the framework postulates that behavioral intention—the subjective likelihood of performing a given behavior—is the most important and immediate predictor of whether people will engage in a behavior (Fishbein & Ajzen, 2010). In turn, behavioral intention arises from the attitudes, perceived normative influence, and perceived behavioral control a person holds with regard to the behavior in question. Generally, the framework suggests that people with more favorable attitudes, more intense perception of approval from others, and a greater sense of control over a behavior will have stronger behavioral intentions, and thus be more likely to perform the behavior (Fishbein, Hennessy, Yzer, & Douglas, 2003). Past meta-analyses have shown that on average the framework accounts for between 39–50% of the variance in intention and 19–36% of the variance in behavior

(Armitage & Conner, 2001; McEachan, Conner, Taylor, & Lawton, 2011; Sutton, 1998). A recent meta-analysis of 33 studies that applied the reasoned action framework to screening behaviors (but not newborn screening, specifically) found that overall attitudes had large associations with intention, norms and perceived behavioral control had medium-sized associations with intention, and intention had medium-sized associations with behavior (Cooke & French, 2008).

Although each of the reasoned action constructs independently influences intentions, the relative strength of their association is partially determined by the specific behavior under consideration, characteristics of the population, or temporary contextual factors. Understanding how these factors moderate the relative importance of the direct antecedents of intention (i.e., attitudes, perceived normative influence, and perceived behavioral control) is useful for anticipating and empirically validating the idiosyncrasies of the population and behavior of interest. One such contextual factor relates directly to the way a behavior is defined in the framework. According to the principle of correspondence, all variables should define the behavior in terms of the same action, target, context, and time (Fishbein, 2008). Even seemingly trivial changes on one or more of these dimensions may fundamentally alter the behavior being examined, and can lead to observable differences within a population in the relative importance of the direct antecedents of intention. From an applied perspective, evidence that program features moderate the psychological pathways underlying parental interest in participating in voluntary expanded screening would have important implications for program design and delivery. For example, education and outreach efforts aimed at promoting informed parental permission for a program with one set of features may require a different communication strategy than would be needed for a program with a different set of features. With this in mind, we set out to answer two research questions:

RQ1: What is the relative importance of attitude, perceived normative influence, and perceived behavioral control as factors associated with parental intentions to participate in an expanded screening program for infants?

RQ2: How do selected variations in screening program features alter the relative importance of attitude, perceived normative influence and perceived behavioral control as factors associated with parental intentions to participate in an expanded screening program for infants?

Method

Participants

We worked with a survey-sampling firm, Research Now, to recruit a nonprobability-based national cohort of participants. All participants were adults living in the U.S. aged 18 years or older. Our target population was currently expectant parents or parents of young children ages three years or less. Women who self-identified as pregnant or who had given birth to at least one child in the past three years were eligible. Men who reported having a pregnant spouse or cohabitating with a pregnant partner were eligible, as were men whose spouse or partner had given birth to a child in the past three years. We over-recruited African Americans to represent 20% of the final sample to enable examination of possible racial

disparities in willingness and ability to participate in the screening program; however, we do not explore these associations in-depth in this report.

Procedure

The study protocol was examined and determined to be exempt from IRB review by the RTI International Committee for the Protection of Human Subjects. Research Now recruited participants for the online survey from their panelist pool using an e-mail invitation. In all, 4,109 panelists responded to the e-mail invitation by clicking an embedded hyperlink to access the online survey. Eligibility was confirmed using an online screener after the participant had agreed to participate in the study. A total of 1,001 eligible respondents completed the survey from April 23rd–28th, 2015, representing 24.4% of the panelists who clicked through from the e-mail invitation.

Survey Instrument

The survey was divided into several sections. The section that is the focus of this report assessed parental willingness and interest in having a child participate in a voluntary expanded screening program. The section was premised by a paragraph introducing the concept of a state-level program to screen infants for rare conditions not currently tested for with newborn screening: fragile X syndrome, Duchenne muscular dystrophy, and spinal muscular atrophy. We explained that the purpose of the program would be to identify children with these conditions before symptoms appear and to help researchers develop and/or evaluate treatments. We also explained that the testing would be conducted using the same blood sample taken at birth as part of mandatory newborn screening (i.e., no additional blood draw would be required). We then asked participants a series of questions about their perceptions of the screening program and their intentions to have a child participate. Additional sections focused on health information seeking, public awareness, knowledge, and attitudes toward mandatory newborn screening and testing for fragile X syndrome, Duchenne muscular dystrophy, and spinal muscular atrophy (the background information provided to participants is included in the Supplementary Material).

We embedded a mixed-level ($3^2 \times 2^2$) fractional factorial experiment to study parental preferences for different parental permission mechanisms and timing. In this between-subjects experiment, we manipulated the introductory description of the expanded screening program on four dimensions creating nine distinct program profiles. These profiles represented variations in the timing at which parental permission would be requested to participate in the program (at birth, two weeks after birth, or two months after birth), cost to participate (free, \$10 with optional waiver, or \$10 out-of-pocket), permission format (print or electronic), and testing options (fixed set of conditions or a custom screening panel allowing parent choice of conditions to be tested). See the Supplementary Material for the full text of the screening program description presented to participants, including the experimental manipulations. We generated a nine-level orthogonal main-effects design using the *orthoplan* procedure in SPSS 21.0 (IBM, 2012) and randomly assigned participants to the nine resulting experimental profiles (see Table 1). Although the design restricted our ability to test higher-order interaction effects among the manipulated dimensions manipulated in the program profiles, we were nonetheless able to test interactions with other

observed variables. In the analyses reported here, we tested for moderation effects of the experimental factors on associations between core reasoned-action constructs and parental intentions to have a child participate in a voluntary expanded screening program.

Measures

We used a set of multi-item scales to measure components of the reasoned action framework with regard to having a child participate in an expanded screening program for babies (Fishbein & Ajzen, 2010). All items were measured using six-point Likert-type or semantic differential scales (range 1 through 6). Higher scores represent stronger intentions, more favorable attitudes, stronger perceptions of social support, and a greater sense of control with regard to having a child participate in the program, respectively.

Intention—We used three items to measure intention (e.g., “If you were asked to take part, how likely is it that you would have your child participate in an expanded screening program for babies?” [*not at all likely–extremely likely*]). The inter-item reliability was strong ($\alpha = .93$) and a composite scale was computed by taking the average of the three items ($M = 4.80$, $SD = 1.26$).

Attitude—Four semantic differential items measured global attitudes toward having a child participate in the expanded screening program. The stem for these items was, “If I had my child participate in an expanded screening program for babies, it would be...” The anchors for the semantic differential pairs were *bad–good*, *disturbing–reassuring*, *harmful–beneficial*, and *worthless–valuable*. We averaged these items to create a composite scale ($\alpha = .93$, $M = 4.88$, $SD = 1.23$).

Perceived normative influence—We measured perceived normative influence with three items (e.g., “Would most people who are important to you think that you *should* or *should not* have your child participate in an expanded screening program for babies?” [*definitely should not–definitely should*]). These items were designed to capture how much participants thought important others would approve of them electing to have their child participate in the expanded screening program (i.e., injunctive norms). Given the novelty and hypothetical nature of the target behavior, we did not include items to measure descriptive norms (i.e., whether important social referents would elect to have their own children participate in an expanding screening program). The three items had strong inter-item reliability ($\alpha = .89$) and we averaged them to create a composite scale ($M = 4.73$, $SD = 1.16$).

Perceived Behavioral Control—We used four items to measure perceived behavioral control (e.g., “Having my child participate in an expanded screening program for babies would be completely up to me.” [*strongly disagree–strongly agree*]). These measures had satisfactory inter-item reliability ($\alpha = .74$). We created a composite scale using the average of the four items ($M = 4.78$, $SD = 0.99$).

Statistical Approach

To test our research questions, we conducted a hierarchical multiple regression analysis. In the first block, we entered the three core reasoned action predictors of behavioral intention—attitude, perceived normative influence, and perceived behavioral control. These variables were mean-centered prior to analysis so that the Block 1 model intercept would equal the weighted grand mean intention over the entire sample. In Block 2, we added a set of dummy-coded variables representing the four factors that were manipulated as part of the embedded fractional factorial experiment. The reference levels for these four variables corresponded to the profile in which the cost to participate was free, permission was attained at birth using a print-based process, and parents did not have the option to choose which conditions would be included on the screening panel. In Block 3, we introduced interaction terms, crossing the four manipulated profile factors by each of the three core reasoned action variables. Thus, the model intercepts in Block 2 and Block 3 represent the average intention score among participants exposed to the reference profile when attitude, perceived normative influence, and perceived behavioral control are held constant at their sample means. All analyses were conducted using Stata 13.0 (StataCorp, 2013).

Results

Descriptive statistics

Demographic characteristics of the sample are presented in Table 2. Women make up a greater proportion of adult members in the Research Now panel who are also expectant or recent parents as defined in our eligibility criteria, which is reflected by the distribution of men and women in our study. Entering these demographic variables as controls did not explain significant variance beyond our three-block model, $R^2 < .01$, $F(21, 952) = 1.15$, $p = .285$. For the sake of parsimony, we report estimates that do not control for demographics.

Parental Intention to Participate in Voluntary Expanded Screening

Results from our hierarchical regression analysis are summarized in Table 3. Although we present the coefficient of determination for each block in Table 3, the regression coefficients shown there are from the Block 3 model after all predictors had been entered.

The three reasoned action constructs entered in Block 1 explained 80% of the variance in intention, $F(3, 997) = 1320.03$, $p < .001$. Before adding Block 2 and 3 variables to the model, perceived normative influence emerged as the strongest predictor of intentions, $\beta = .54$, $B = 0.58$, $SE = 0.03$, $t(997) = 22.67$, $p < .001$. In other words, the Block 1 model estimated that every one-point increase in perceived normative influence would lead, on average, to more than a half-point increase in parental intention to participate in an expanded screening program. By comparison, the relative importance of parental attitudes toward participating ($\beta = .22$, $B = 0.23$, $SE = 0.02$, $t(997) = 10.21$, $p < .001$) and perceived behavioral control ($\beta = .21$, $B = 0.27$, $SE = 0.03$, $t(997) = 9.14$, $p < .001$) was less than that of perceived normative influence in the Block 1 model, which essentially ignores potential group differences derived from the program profiles to which participants were exposed.

Adding dummy variables corresponding to the four manipulated program features in Block 2 did not lead to a statistically significant increase in the amount of variance explained, $F(6, 991) = 0.68, p = .662$. Thus, we found no evidence that mean levels of intention among experimental groups differed based on features manipulated in the nine program profiles.

In Block 3, we added a set of interaction terms to test for differences in the effects of the three primary reasoned action constructs by experimental groups. Variation among program profiles based on timing of parental permission, cost to participate, and permission format significantly contributed to differences in the relative importance of reasoned action constructs on intention, $R^2 = .01, F(18, 973) = 2.35, p = .001$. In all, adding the interaction terms accounted for an additional 1% of the variance in parental intention to participate in an expanded screening program.

The significant positive coefficient for the interaction between perceived normative influence and timing of parental permission indicates that a one-unit increase in perceived normative influence had an impact on intention that was about 0.14-units greater for participants presented with the option to enroll in an expanded screening program that seeks parental permission 2 months after birth rather than at birth, $t(973) = 2.15, p = .032$. Statistically significant negative coefficients for the interactions of attitude with cost of participation and permission format suggest that establishing a \$10 fee to participate in the screening program would decrease the effect of a one-unit increase in attitudes on intention by 0.17 units on average, $t(973) = -3.04, p = .002$, and an electronic permission form would reduce the average effect of attitude on parental intention by 0.16 units, $t(973) = -3.38, p = .001$. Conversely, there were statistically significant positive coefficients for the interaction of perceived behavioral control with cost of participation and permission format, such that a \$10 fee and electronic permission process would each increase the estimated effect of a one-unit change in perceived behavioral control by about 0.15 units on intention, respectively $t(973) = 2.30, p = .022$ and $t(973) = 2.32, p = .021$. Figure 1 summarizes how these interaction effects affect the regression coefficients for reasoned action constructs in models predicting intention to participate based on eight alternative program-design scenarios. The process of generating the estimates for these path models was tantamount to resetting the reference category of the factor variables and their corresponding interaction terms in the Block 3 regression model to match features listed in the figure labels. Notably, whereas the relative association between perceived normative influence and intention is consistently strong across program profiles, the associations of attitude and perceived behavioral control with intention vary widely.

Discussion

Overall, parents interviewed in this study reported strong intentions to participate in a voluntary expanded screening program, as reflected by a mean intention score toward the upper end of the six-point measurement scale. Similarly, parents tended to have favorable attitudes toward participating in the program, sensed that others would support their doing so, and considered the behavior under their control.

These findings are aligned with results from region-specific studies examining public acceptance in the U.S. for expanded newborn screening of fragile X, Duchenne muscular dystrophy, Becker muscular dystrophy, and spinal muscular atrophy. For example, in a recent survey of expectant parents recruited while touring the labor and delivery facilities of a hospital in Pittsburgh, P.A., 93% expressed support for expanded newborn screening of Duchenne muscular dystrophy, Becker muscular dystrophy, and spinal muscular atrophy (Wood et al., 2014). Similarly, 78% of parents offered voluntary Duchenne muscular dystrophy screening for their infant males during routine well-child visits at four pediatric clinics in Atlanta, G.A., were in favor of screening (Cyrus et al., 2012). Caregivers of children with fragile X syndrome or fragile X carriers have also been shown to endorse free voluntary screening (Bailey et al., 2012). Majority support for expanded NBS of serious, untreatable disorders has also been found in studies conducted in Australia (Christie et al., 2013), Canada (Hayeems et al., 2015), Hong Kong (Mak et al., 2012), and the Netherlands (Plass, van El, Pieters, & Cornel, 2010). Nonetheless, public perceptions of expanded newborn screening are just one factor that must be considered when evaluating the merit of extending population screening on a voluntary basis for rare health conditions.

Although the majority of parents in this study indicated a willingness to participate in voluntary expanded screening, some reported they were unlikely to do so. Drawing on this variance, this is the first study applying the reasoned action framework to reveal potential psychological pathways that parents may follow when making decisions about expanded screening. The framework is particularly well-suited for guiding research to better understand factors leading people to perform specific behaviors. In turn, this information lends itself to the design of strategic interventions that focus on affecting change at the most appropriate point in the causal chain.

Overall, we found strong associations between perceived normative influence and intention regardless of program scenario, suggesting that parents who are currently unlikely to participate in expanded screening may feel a lack of social support. Viewed in the broader context of reasoned action research, this finding was unexpected. Generally, perceived normative influence has been shown to play less of a role than attitudes or perceived behavioral control in predicting intentions over a wide range of health behaviors, and this difference is especially pronounced when the reasoned action framework is applied to adults versus adolescents (Conner & Armitage, 1998; McEachan et al., 2011). A notable exception is the large average association between perceived norms and intention observed by Cooke and French (2008) in their meta-analysis for a subset of studies examining prenatal screening tests; however, the average attitude–intention association in that context was also large. Conceivably, the unusually strong association between perceived normative influence and intention may reflect uncertainty within couples about the attitudes of a spouse or partner toward voluntary expanded screening. From this analysis, we do not know who parents think about as important social referents for this behavior, but it is plausible that these participants have spouses or partners in mind as people who would need to be directly involved in decision making. Further analysis is needed to better understand which features of the social context surrounding expanded screening are especially salient to parents.

Additionally, we manipulated program features to explore how alternative configurations of timing, cost, permission format and testing options might influence parental decision making processes related to participating in voluntary expanded screening. From a reasoned action perspective, the resulting differences in program-design scenarios constitute conceptually different behaviors, and the moderation effects we observed extend from those differences. Specifically, variations in timing of parental permission, cost, and permission format altered the predicted relative importance of attitude, normative influence and perceived behavioral control. Changes to the psychosocial mechanism underlying intention to participate based on different configurations of program features provides valuable insights that can be applied to outreach, education, and informed parental permission processes. The reasoned action framework postulates that downstream influences on intention and behavior are generally caused by upstream changes to behavior-relevant salient beliefs. Our results point to the broad kinds of beliefs that may be critical to parental decision making in light of alternative program-design scenarios. For example, between two program scenarios both involving permission at birth, a cost of \$10, and screening for a fixed set of conditions, our results indicate that parental attitudes will impact intentions when a print permission format is used but not with an electronic permission procedure (see Figure 1). The strategic implication of this finding is that normative and control beliefs would be more relevant than attitudinal beliefs in education and outreach materials under than the electronic scenario. Conducting a more detailed belief analysis is the natural next-step to identify specific misconceptions that may have undue influence on screening decisions. For example, if non-intenders construe expanded screening to be socially unacceptable, but further research finds that important social referents tend to be supportive, these inaccurate normative beliefs would be relevant to address in informational campaigns.

Limitations

This survey was not designed to descriptively represent the distribution of opinions and preferences toward expanded newborn screening in the full U.S. population of recent and expectant parents. In addition to not being a probability-based sample, all participants were English speaking. Instead, the experimental design allowed us to draw causal inferences about how altering specific features of a hypothetical screening program would typically affect parental intentions toward participating. Other limitations include our focus on modeling intentions to participate, rather than actual behavior, and the potential impact of factors we did not assess on parental decision making. Along these lines, it is worth noting that the hypothetical screening program focused on rare health conditions, and it is unclear how parental decisions would differ if the program were to offer screening for common conditions. Also, participants were provided with only a brief description of the voluntary expanded screening program, and so their opinions and stated intentions were likely based on less information than would be available to parents deciding to participate in such a program if it were implemented. In that respect, the thoughts and intentions expressed by our participants may be interpreted as reflecting their first impressions of the program rather than the product of an informed and deliberate decision making process. Our findings nonetheless corroborate and extend prior research in this area with data from a national sample of parents.

Conclusion

Many conditions that fall short of the RUSP criteria are of great interest to families, health care providers, and researchers. Early identification can result in substantive benefits for both children and families—reducing the “diagnostic odyssey,” providing access to support services and treatments, informing families of reproductive risk, and providing the opportunity to enroll in experimental treatment studies. Consideration of these potential benefits are counterbalanced by uncertainty over the possible negative implications of testing for children and families. The prospect of voluntary expanded screening will broaden the difficulties faced when attempting to provide public-health scale education to parents about NBS. Information about screening is often just one part of an overwhelming amount of information provided to expectant parents and often escapes notice (Kemper, Fant, & Clark, 2005). As a result, it is of little surprise that studies have found many parents are not aware or do not remember receiving information about NBS (Davis et al., 2006; DeLuca, Kearney, Norton, & Arnold, 2011; Hasegawa, Fergus, Ojeda, & Au, 2011). Because these programs are to a certain degree mandatory, limited public awareness has not translated into low participation rates. Parent education efforts for a voluntary expanded screening program, on the other hand, will need to meet higher disclosure standards, ensure that the opportunity to accept or decline participation is equitably shared, and impartially help parents consider whether or not the program aligns with their goals, values, and preferences. With an opt-in voluntary parental-permission model, ineffective public outreach and weak audience engagement would fundamentally undermine efforts to facilitate informed parental choice. The insights gained from this study represent a first step toward combating this threat and developing a population-level communication strategy for parent education and outreach.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgments

The research reported here was generously supported by the National Center for Advancing Translational Sciences (1UL1TR001111). The John Merck Fund provided additional funding and RTI International provided support for preparing the manuscript.

References

- American Academy of Pediatrics, Newborn Screening Task Force. Serving the family from birth to the medical home: newborn screening: a blueprint for the future—a call for a national agenda on state newborn screening programs. *Pediatrics*. 2000; 106:389–422. Retrieved from http://pediatrics.aappublications.org/content/pediatrics/106/Supplement_2/389.full.pdf. [PubMed: 10947682]
- Ajzen, I. From intentions to actions: A theory of planned behavior. In: Kuhl, J.; Beckman, J., editors. *Action control*. New York, NY: Springer-Verlag; 1985. p. 11-39.
- Bailey DB Jr, Bishop E, Raspa M, Skinner D. Caregiver opinions about fragile X population screening. *Genetics Medicine*. 2012; 14:115–121. DOI: 10.1038/gim.0b013e31822ebaa6
- Bailey DB Jr, Gehtland L. Newborn screening: evolving challenges in an era of rapid discovery. *JAMA: Journal of the American Medical Association*. 2015; 313:1511–1512. DOI: 10.1001/jama.2014.17488 [PubMed: 25898043]

- Bailey DB Jr, Skinner D, Davis AM, Whitmarsh I, Powell C. Ethical, legal, and social concerns about expanded newborn screening: fragile X syndrome as a prototype for emerging issues. *Pediatrics*. 2008; 121:e693–e704. DOI: 10.1542/peds.2007-0820 [PubMed: 18310190]
- Armitage CJ, Conner M. Efficacy of the Theory of Planned Behaviour: a meta-analytic review. *British Journal of Social Psychology*. 2001; 40:471–499. DOI: 10.1348/014466601164939 [PubMed: 11795063]
- Centers for Disease Control and Prevention. Ten great public health achievements—United States 2001–2010. *MMWR Morbidity and Mortality Weekly Report*. 2011 May 20.60:619–623. Retrieved from <http://www.cdc.gov/mmwr/preview/mmwrhtml/mm6019a5.htm>. [PubMed: 21597455]
- Christie L, Wotton T, Bennetts B, Wiley V, Wicken B, Rogers C, Field M. Maternal attitudes to newborn screening for fragile X syndrome. *American Journal of Medical Genetics Part A*. 2013; 161:301–311. DOI: 10.1002/ajmg.a.35752 [PubMed: 23303663]
- Conner M, Armitage CJ. Extending the theory of planned behavior: A review and avenues for further research. *Journal of Applied Social Psychology*. 1998; 28:1429–1464. DOI: 10.1111/j.1559-1816.1998.tb01685.x
- Cooke R, French DP. How well do the theory of reasoned action and theory of planned behaviour predict intentions and attendance at screening programs? A meta-analysis. *Psychology & Health*. 2008; 23:745–765. [PubMed: 25160879]
- Cyrus A, Street N, Quarry S, Kable J, Kenneson A, Fernhoff P. Clinic-based infant screening for Duchenne muscular dystrophy: A feasibility study. *PLoS Currents*. 2012; 4:e4f99c5654147a.doi: 10.1371/4f99c5654147a
- Davis TC, Humiston SG, Arnold CL, Bocchini JA Jr, Bass PR III, Kennen EM, Lloyd-Puryear M. Recommendations for effective newborn screening communication: results of focus groups with parents, providers, and experts. *Pediatrics*. 2006; 117(Suppl 3):S326–S340. DOI: 10.1542/peds.2005-2633M [PubMed: 16735260]
- DeLuca JM, Kearney MH, Norton SA, Arnold GL. Parents' experiences of expanded newborn screening evaluations. *Pediatrics*. 2011; 128:53–61. DOI: 10.1542/peds.2010-3413 [PubMed: 21708804]
- Dhondt JL. Expanded newborn screening: social and ethical issues. *Journal of Inherited Metabolic Disease*. 2010; 33(Suppl 2):S211–S217. DOI: 10.1007/s10545-010-9138-y [PubMed: 20544288]
- Fishbein M. A reasoned action approach to health promotion. *Medical Decision Making*. 2008; 28:834–844. DOI: 10.1177/0272989X08326092 [PubMed: 19015289]
- Fishbein, M.; Ajzen, I. Belief, attitude, intention, and behavior: An introduction to theory and research [PDF version]. Reading, MA: Addison-Wesley; 1975. Retrieved from <http://people.umass.edu/aizen/f&a1975.html>
- Fishbein, M.; Ajzen, I. Predicting and changing behavior: the reasoned action approach. New York: Psychology Press; 2010.
- Fishbein M, Hennessy M, Yzer M, Douglas J. Can we explain why some people do and some people do not act on their intentions? *Psychology, Health & Medicine*. 2003; 8:3–18. DOI: 10.1080/1354850021000059223
- Fishbein, M.; Triandis, HC.; Kanfer, FH.; Becker, M.; Middlestadt, SE.; Eichler, A. Factors influencing behavior and behavior change. In: Baum, A.; Revenson, TA.; Singer, JE., editors. *Handbook of health psychology*. Mahwah, NJ: Erlbaum; 2001. p. 3-16.
- Goldenberg AJ, Sharp RR. The ethical hazards and programmatic *challenges of genomic newborn screening*. *JAMA: Journal of the American Medical Association*. 2012; 307:461–462. DOI: 10.1001/jama.2012.68 [PubMed: 22298675]
- Grosse SD, Boyle CA, Kenneson A, Khoury MJ, Wilfond BS. From public health emergency to public health service: The implications of evolving criteria for newborn screening panels. *Pediatrics*. 2006; 117:923–929. DOI: 10.1542/peds.2005-0553 [PubMed: 16510675]
- Hasegawa LE, Fergus KA, Ojeda N, Au SM. Parental attitudes toward ethical and social issues surrounding the expansion of newborn screening using new technologies. *Public Health Genomics*. 2011; 14:298–306. DOI: 10.1159/000314644 [PubMed: 20689248]

- Hayeems RZ, Miller FA, Bombard Y, Avard D, Carroll J, Wilson B, Axler R. Expectations and values about expanded newborn screening: A public engagement study. *Health Expectations*. 2015; 18:419–429. DOI: 10.1111/hex.12047 [PubMed: 23369110]
- IBM Corp. IBM SPSS Statistics for Windows, Version 21.0 [Computer software]. Armonk, NY: IBM Corp; 2012.
- Kemper AR, Fant KE, Clark SJ. Informing parents about newborn screening. *Public Health Nursing*. 2005; 22:332–338. DOI: 10.1111/j.0737-1209.2005.220408.x [PubMed: 16150014]
- Kemper AR, Green NS, Calonge N, Lam WKK, Comeau AM, Goldenberg AJ, Bocchini JA. Decision-making process for conditions nominated to the recommended uniform screening panel: statement of the US Department of Health and Human Services Secretary's Advisory Committee on Heritable Disorders in Newborns and Children. *Genetics in Medicine*. 2014; 16:183–187. DOI: 10.1038/gim.2013.98 [PubMed: 23907646]
- Mak CM, Lam CW, Law CY, Siu WK, Kwong LLT, Chan KL, Chan AYW. Parental attitudes on expanded newborn screening in Hong Kong. *Public Health*. 2012; 126:954–959. DOI: 10.1016/j.puhe.2012.08.002 [PubMed: 23148889]
- McEachan RRC, Conner M, Taylor NJ, Lawton RJ. Prospective prediction of health-related behaviours with the Theory of Planned Behavior: A meta-analysis. *Health Psychology Review*. 2011; 5:97–144. DOI: 10.1080/17437199.2010.521684
- Plass AMC, van El CG, Pieters T, Cornel MC. Neonatal screening for treatable and untreatable disorders: Prospective parents' opinions. *Pediatrics*. 2010; 125:e99–e106. DOI: 10.1542/peds.2009-0269 [PubMed: 20026497]
- President's Council on Bioethics. The changing moral focus of newborn screening: An ethical analysis by the President's Council on Bioethics. 2008. Retrieved from <http://hdl.handle.net/10822/559367>
- Ross LF. Mandatory versus voluntary consent for newborn screening? *Kennedy Institute of Ethics Journal*. 2010; 20:299–328. Retrieved from Project MUSE database. [PubMed: 21338027]
- Rothwell E, Anderson RA, Swoboda KJ, Stark L, Botkin JR. Public attitudes regarding a pilot study of newborn screening for spinal muscular atrophy. *American Journal of Medical Genetics Part A*. 2013; 161:679–686. DOI: 10.1002/ajmg.a.35756 [PubMed: 23443997]
- StataCorp. Stata Release 13.0 [Computer program]. College Station, TX: StataCorp LP; 2013.
- Sutton S. Predicting and explaining intentions and behavior: How well are we doing? *Journal of Applied Social Psychology*. 1998; 28:1317–1338. DOI: 10.1111/j.1559-1816.1998.tb01679.x
- Watson MS, Mann MY, Lloyd-Puryear MA, Rinaldo P, Howell RR. Newborn screening: toward a uniform screening panel and system. *Genetics in Medicine*. 2006; 8(Suppl 1):1S–252S. DOI: 10.1097/01.gim.0000223891.82390.ad [PubMed: 16783161]
- Wood MF, Hughes SC, Hache LP, Naylor EW, Abdel-Hamid HZ, Barmada MM, Clemens PR. Parental attitudes toward newborn screening for Duchenne/Becker muscular dystrophy and spinal muscular atrophy. *Muscle & Nerve*. 2014; 49:822–828. DOI: 10.1002/mus.24100 [PubMed: 24307279]

Research Highlights

- Experiment tests the impact of program features on expanded screening intentions.
- Data from a national online survey of recent and expectant parents.
- Parents tended to support optional population-based newborn screening.
- Program features impact psychological mechanisms underlying decisions to participate.

A. Birth; Free; Print; Fixed

B. Two months; Free; Print; Fixed

C. Birth; \$10; Print; Fixed

D. Birth; Free; Electronic; Fixed

E. Two months; \$10; Print; Fixed

F. Birth; \$10; Electronic; Fixed

G. Two months; Free; Electronic; Fixed

H. Two months; \$10; Electronic; Fixed

Figure 1.

Path estimates among reasoned action variables for eight expanded screening program designs. The designs differ on four dimensions (i.e., timing, cost, format, and test options), which are reflected in the subtitle of each diagram. Standardized regression coefficients are shown. Asterisks indicate coefficients that are statistically different from zero. ATT = Attitude. NORM = Perceived normative influence. PBC = Perceived behavioral control. INT = Behavioral intention.

*p < .05. **p < .01. ***p < .001.

Author Manuscript

Author Manuscript

Author Manuscript

Author Manuscript

Table 1

Distinguishing characteristics of nine orthogonal program profiles generated for a mixed-level factorial experiment with four factors

Profile	Factor			
	Timing	Cost	Format	Test options
1	Birth	Free	Print	Fixed
2	2 weeks	Free	Electronic	Custom
3	2 months	Free	Print	Fixed
4	Birth	\$10 + waiver	Electronic	Fixed
5	2 weeks	\$10 + waiver	Print	Fixed
6	2 month	\$10 + waiver	Print	Custom
7	Birth	\$10	Print	Custom
8	2 weeks	\$10	Print	Fixed
9	2 months	\$10	Electronic	Fixed

Table 2

Participant characteristics

Variable	<i>n</i>	%
Age		
18 – 24	122	12.2
25 – 34	488	48.8
35 – 44	303	30.3
45+	88	8.8
Gender		
Female	788	78.7
Male	213	21.3
Race/ethnicity		
White, non-Hispanic	576	57.5
Black, non-Hispanic	166	16.6
Hispanic, all-races	147	14.7
Other, non-Hispanic	112	11.2
Education		
High school or less	164	16.4
Some college or technical school	266	26.6
College graduate	398	39.8
Advanced or postgraduate degree	173	17.3
Annual household income		
Less than \$25,000	116	11.6
\$25,000 – \$49,999	274	27.4
\$50,000 – \$74,999	246	24.6
\$75,000 – \$99,999	179	17.9
\$100,000 or more	186	18.6
Geographical region		
North	193	19.3
South	355	35.5
Midwest	239	23.9
West	214	21.4
Marital status ^a		
Married	717	71.6
Living with partner	128	12.8
Single	156	15.6
Children in household ages 3 years or younger		
One or fewer	746	74.5
Two or more	255	25.5
Currently pregnant ^b		
No/Unsure	851	85.0
Yes	150	15.0

Note. $N = 1,001$. Percentages within variables may not sum to 100 due to rounding error.

^aThe *single* category combines respondents who selected *never been married*, *widowed*, *separated*, or *divorced*.

^bThe currently-pregnant data refer to female participants who self-reported as pregnant, as well as male participants who reported having a pregnant spouse or partner.

Table 3
Hierarchical regression analysis predicting intention to participate in an expanded screening program for infants

Variable	β	B	SE	95% CI	R ²
Block 1					.80***
ATT	.39	0.40***	0.06	0.29, 0.52	
NORM	.39	0.43***	0.06	0.30, 0.56	
PBC	.12	0.15*	0.07	0.02, 0.29	
Block 2					.80***
Timing (ref=Birth)					
Two weeks	-.01	-0.02	0.04	-0.11, 0.07	
Two months	-.01	-0.03	0.04	-0.12, 0.06	
Cost (ref=Free)					
\$10	-.01	-0.03	0.04	-0.12, 0.05	
\$10 + waiver	-.00	-0.01	0.04	-0.10, 0.07	
Format (ref=Print)					
Electronic	.02	0.04	0.04	-0.03, 0.12	
Test options (ref=Fixed)					
Custom	-.01	-0.04	0.04	-0.11, 0.04	
Block 3					.81**
ATT × Timing (ref=Birth)					
Two weeks	-.01	-0.02	0.06	-0.13, 0.09	
Two months	-.06	-0.10	0.06	-0.21, 0.01	
NORM × Timing (ref=Birth)					
Two weeks	.05	0.09	0.06	-0.03, 0.21	
Two months	.07	0.14*	0.07	0.01, 0.27	
PBC × Timing (ref=Birth)					
Two weeks	-.03	-0.06	0.07	-0.20, 0.09	
Two months	.03	-0.06	0.07	-0.09, 0.20	
ATT × Cost (ref=Free)					
\$10	-.09	-0.18**	0.06	-0.29, -0.06	

Variable	β	B	SE	95% CI	R ²
\$10 + waiver	-.05	-0.08	0.05	-0.18, 0.03	
NORM $\times$ Cost (ref=Free)					
\$10	.07	0.15*	0.07	0.02, 0.28	
\$10 + waiver	.04	0.08	0.06	-0.05, 0.20	
PBC $\times$ Cost (ref=Free)					
\$10	.04	0.09	0.08	-0.06, 0.24	
\$10 + waiver	.04	0.08	0.07	-0.06, 0.22	
ATT $\times$ Format (ref=Print)					
Electronic	-.09	-0.16**	0.05	-0.25, -0.07	
NORM $\times$ Format (ref=Print)					
Electronic	.01	0.03	0.06	-0.08, 0.14	
PBC $\times$ Format (ref=Print)					
Electronic	.07	0.15*	0.06	0.02, 0.27	
ATT $\times$ Test options (ref=Fixed)					
Custom	.01	0.02	0.05	-0.07, 0.12	
NORM $\times$ Test options (ref=Fixed)					
Custom	-.02	-0.03	0.05	-0.14, 0.08	
PBC $\times$ Test options (ref=Fixed)					
Custom	.03	0.06	0.07	-0.07, 0.19	

Note: All regression coefficients are estimates from the Block 3 model, intercept $B = 4.83$, $SE = .04$, $ATT = \text{Attitude}$, $NORM = \text{Perceived normative influence}$, $PBC = \text{Perceived behavioral control}$, ref = Reference category.

*
 $p < .05$.

**
 $p < .01$.

 $p < .001$.