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A Qualitative Secondary Evaluation of Statewide Follow-Up Interviews for Abnormal Newborn Screening Results for Cystic Fibrosis and Sickle Cell Hemoglobinopathy

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Abstract

Purpose—The purpose of this qualitative analysis was to assess parental acceptability of large-scale, telephone follow-up regarding their infants' newborn screening (NBS) results indicating carrier status for sickle cell hemoglobinopathy (SCH) and cystic fibrosis (CF).

Methods—Analysis of 195 interview transcripts focused on parents' responses to two open-ended questions “*What was your reaction to being called by me?*” and “*What do you think of the state newborn screening program having follow-up people calling parents like you?*” Responses were coded using conventional content analysis procedures and non-parametric tests were performed to analyze quantitative data.

Results—Most parents reported favorable opinions about the follow-up. Favorable opinions were associated with several emotional reactions to receiving follow-up ($p < 0.001$), and three reasons why parents found the interview beneficial ($p < 0.05$): it provided information, clarified NBS results, and answered questions. Seventeen parents of SCH carriers reportedly had not been told their infant's NBS results and received them for the first time during the follow-up interview.

Conclusion—Parents of CF and SCH carrier infants had favorable opinions and identified specific benefits to receiving follow-up contact. This analysis demonstrates an information deficit among carrier parents and illustrates the importance of NBS follow-up and need for comprehensive communication and counseling.

Keywords

newborn screening; follow-up counseling; cystic fibrosis carrier; sickle cell trait; provider-patient communication

Introduction

Nearly every infant born in the U.S. undergoes newborn screening (NBS) through blood samples obtained shortly after birth. This population-scale public health program screens for several conditions in order to save lives and provide early intervention and treatment to affected infants. Wisconsin's NBS program screens for 47 conditions, including two of the world's most common genetic diseases, cystic fibrosis (CF) and sickle cell

hemoglobinopathy (SCH). Early identification of these conditions has proven beneficial in the prevention of infections for infants with SCH ¹ and nutritional deficiencies or lung problems for infants with CF ²⁻⁴.

The U.S. is at the forefront of incorporating innovation into practice as NBS has undergone a recent, rapid expansion with the advent of multiplex and DNA technologies. The high sensitivity of new screening technologies allows for detection of those infants affected with SCH and CF, but also identifies heterozygous genetic “carriers” in the process. A carrier for a recessively inherited condition (sometimes called “trait”) refers to persons with a single deleterious mutation for that genetic condition. The U.S. carrier frequency is estimated to be 1/12 for SCH in African Americans ⁵ and 1/28 for CF ⁶ in Caucasians.

The concept of “carrier status” can be confusing for parents to understand and primary care providers' (PCPs') communication of these results has been criticized ⁷⁻¹⁴. These challenges can lead to the development of parental psychosocial complications such as depression, anxiety, fear, and confusion ¹⁴⁻²¹. We contend that these consequences are potentially avoidable or alleviated by providing access to follow-up and informational resources. We therefore implemented the “Wisconsin Project on Improvement of Communication Processes and Outcomes after Newborn Screening” to serve the nearly 1000 Wisconsin families with infants found to be carriers for CF and SCH via NBS annually ²². This project was designed as a quality improvement effort to assess parents' experiences with their infants' NBS results disclosure for SCH and CF carriers and evaluate their opinions of NBS telephone follow-up.

Many NBS programs are interested in routine telephone follow-up for families of carrier infants, but critics charge that there is no published evidence that parents would accept such contacts from centralized public health systems. Our statewide implementation of an elaborate communication and follow-up study for families of CF and SCH carrier infants provided an excellent opportunity to evaluate parental acceptability of routine, telephone contact for a centralized program that used public health follow-up techniques. In addition to using traditional evaluation surveys ²³, we decided to incorporate two open-ended acceptability questions into our contacts with parents because of the limitations of fixed-choice and close-ended questions. This alleviated potential bias effects often seen in traditional evaluation surveys and allowed parents to provide feedback on any aspect of the follow-up, rather than confining their opinions to specific topics. The purpose of this qualitative, secondary analysis of open-ended interview questions was to assess parental acceptability of large-scale, statewide telephone follow-up regarding their infants' NBS results.

Materials and Methods

Study Design

This paper describes findings of a qualitative, secondary analysis from statewide follow-up telephone interviews with parents after NBS. Analysis focused on parents' responses to two open-ended acceptability questions:

Question 1: *What was your reaction to being called by me?*

Question 2: *What do you think of the state newborn screening program having follow-up people like me calling parents like you?*

The goal of this analysis was to examine parents' reactions to NBS follow-up in a richer and more descriptive fashion than is possible with more traditional, Likert-scaled evaluation surveys.

Data source: the Project

This analysis used parent interview data from the “Wisconsin Project on Improvement of Communication Processes and Outcomes after Newborn Screening” (henceforth called “the Project”), which represents a collaboration of the Wisconsin State Laboratory of Hygiene (WSLH) and the Department of Health Services (DHS) with the Medical College of Wisconsin as contracted Project agent. This Project was designed as both a research study and a statewide quality improvement initiative, to serve families whose infants were found to be heterozygous genetic carriers for SCH or CF²². The Project aimed to assess parents' psychosocial outcomes after NBS results disclosure and provide brief counseling to parents if needed; however, this report describes only parents' responses to the two acceptability questions evaluating the follow-up interview, itself. Forthcoming manuscripts will discuss results of the larger Project, including parents' experiences and psychosocial outcomes related to newborn screening results disclosure²³⁻²⁵. The Project is consistent with the NIH criteria for waiver of consent for initial contact procedure²⁶. The Project was authorized by DHS and approved by Institutional Review Boards at the Medical College of Wisconsin and University of Wisconsin, Madison.

Participants and Recruitment

Nearly every infant born in Wisconsin undergoes NBS and specimens are sent to WSLH for analysis. Project participants included parents and primary care providers (PCPs) of infants found via NBS to be carriers for SCH or CF. SCH carrier status for the Project was defined as the presence of fetal, adult, and sickle hemoglobin (“F-A-S”) on NBS. Other heterozygous results for hemoglobinopathy (e.g. “F-S-A”) were excluded to minimize confounding effects for the Project's primary analyses. CF carrier status was defined as a screening result that showed elevated immunoreactive trypsinogen (IRT) and a single CFTR mutation, followed by a normal sweat chloride test. The CF Foundation recommends all infants with abnormal CF screening results undergo sweat chloride testing because 2-5% may have a second unidentified mutation, not included in the Wisconsin NBS panel^{27, 28}, resulting in CF the disease. Only confirmed CF carriers, with normal sweat chloride test results, were considered eligible for the Project.

Parents of infants were enrolled in the Project as follows. NBS results were faxed from WSLH to the Project's study team for infants with results indicating F-A-S or elevated IRT with a single CFTR mutation. Infants were immediately excluded based on additional criteria (1) infant had more than one abnormal NBS result, indicative of a co-existing condition; (2) infant's parent had another child previously enrolled and declined to participate; (3) infant considered “medically fragile” defined as less than 35 weeks gestation, spending greater than 5 days in nursery or neonatal intensive care, or being re-hospitalized. Infants were also excluded if their PCP identified any contraindication to the Project's telephone follow-up interview, such as an English-language barrier or psychosocial issues deemed problematic for additional contact and follow-up. Finally, infants were excluded based on sweat chloride testing that had ambiguous results, were performed on infants >5 months old, or resulted in a CF diagnosis.

Recruitment procedures involved two steps. First, when infants were 3-5 months old, parents were sent recruitment mailings containing an explanation of the Project and a “decline of contact” card. This procedure allowed parents to decline the Project contact without becoming fully informed about the purpose of the research. To avoid parental confusion or distress if, perhaps, parents had not been informed or were confused about their infants' NBS results, recruitment mailings did not mention infants' NBS results. The second step in the recruitment process occurred approximately 10 days after recruitment materials had been mailed to parents. If we had not received “decline to contact” cards, research team members

telephoned parents inviting them to participate in a one-time telephone interview about the disclosure of their infant's NBS results and psychosocial outcomes. Parents who participated in the follow-up interview received a \$20 gift certificate in gratitude for their time.

Description of the Interview

The interview questions and script were developed by a team with methodological and content expertise in pediatric medicine, NBS, genetic counseling, nursing, communication, and bioethics. Parents participated in a one-time, follow-up telephone interview which lasted approximately 20-30 minutes, depending on parent response. Interviews began with informed consent, followed by a carefully scripted, directive, semi-structured discussion balancing fixed-choice and open-ended research questions. The directive nature of the interview script ensured consistent data collection on a set of pre-determined questions, particularly useful with such a large study population; however, the semi-structured approach also allowed for additional unscripted probing questions to clarify parent responses and prompt further elaboration when necessary. The semi-structured approach also allowed for interviewers to provide counseling in the form of parent education and emotional support, if needed. At the end of the interview, after all other research questions were asked and all counseling was provided, if necessary, the two open-ended acceptability questions were asked about parents' opinions of the Project's follow-up.

Interviews were conducted by one member of a multidisciplinary team comprised of nurses, a physician, and a genetic counselor. Prior to the Project's start date, it was anticipated that the parent interviewers would potentially be contacting parents who had not heard their infant's NBS results. Because the follow-up interview with parents of CF carriers happened after the infant's sweat test, where the infant was confirmed a carrier, the study team could be certain that these parents had been informed of their infant's results, likely by more than one provider. Alternatively, we anticipated that any follow-up contact requiring results disclosure would likely happen with parents of SCH carriers, as their PCP would be the primary source for disclosing their infant's NBS results. Because of the definitive nature of the SCH screening, these results are not ambiguous and do not require additional testing for diagnostic purposes (unlike CF carrier infants). Given the potential that some parents may not have been informed of their infant's NBS results, all interviewers had a clinical background and experience delivering clinical test results or diagnoses to parents in a pediatric setting. All interviewers were trained to assess signs of emotional distress or confusion. If either occurred, the interviewee or the interviewer had the option of forgoing the research questions, making the interview a purely clinical intervention. Both research and clinical calls ended with debriefing to ensure misconceptions identified during the interview were clarified and parents knew who to contact with additional questions. Interviews were audio-recorded, transcribed, proofread, and de-identified as they occurred.

Data management and sample

Between March 2008 and August 2010, NBS results for 1,781 infants were faxed to the Project's study team by WSLH. Of these, 617 infants met immediate exclusion criteria (as described above). Recruitment letters were mailed to the remaining 1,164 parents, 11 parents returned a "decline of contact card," and so 1,153 parents were contacted by telephone to participate. Of these, 144 parents verbally declined to participate, 548 had not responded to the recruitment telephone calls by the start of this analysis, and 461 chose to participate in the follow-up interview. When this analysis began, 175 interviews were unavailable for use and therefore excluded because of transcription delays, audio-recording failures, or because they became clinical interventions. Of the remaining 286 transcripts, 195 included parents' responses to at least one of the acceptability questions.

The final set of 195 transcripts consisted of interviews with 190 mothers and 5 fathers. Two-thirds were parents of SCH carriers while 1/3 were parents of CF carriers (Table 1), an identical proportion to that found in the total interviewed sample (n=461) of the Project. While most parents of sickle cell trait infants reported their race/ethnicity as African American (n=90), some identified themselves as Latino (n=7), Caucasian (n=26), or another race (n=7). Similarly, most parents of CF carriers identified themselves as Caucasian (n=53), though some reported their race/ethnicity as African American (n=7) or another race (n=5).

Analysis

Parents' responses to the two open-ended acceptability questions were extracted from each transcript to form the dataset for this analysis. Extracted responses were coded following an adapted version of conventional content analysis procedures²⁹. The team conducted line-by-line, open coding on a subset of data to identify categories and develop a coding schema²⁹⁻³². Coders compared their impressions of key concepts found in the text until no new themes emerged from the dataset. Data describing similar ideas were clustered into themes and organized into subthemes, labeled with descriptive codes, and arranged into a codebook^{30, 33, 34}. Disagreements about coding were resolved through discussion, consensus ratings, and revisions of the codebook. A second subset of data was coded to establish the utility of the coding system and the codebook was revised and refined as needed. The final version of the codebook contained 3 major themes, 4 subthemes, and 62 codes describing parents' responses to the acceptability questions.

Coding of the entire data set using NVivo 8 software was performed in rounds. In each round, the 4 members of the coding team individually coded 10 transcripts apiece, followed by consensus coding of an additional 10 transcripts. Consensus coding began with 2 team members coding a single transcript, and then group consensus meetings were held to compare codes with the entire team to promote discussion, avoid coding drift, and intermittently evaluate inter-coder reliability. As a result of these rounds, a randomly selected 80% of transcripts were individually coded, while the remaining 20% were coded by group consensus. Each individual sentence was considered a "unit" for coding and analysis, as subjectively identified by the transcriptionist based on grammar and breaths. Codes represented specific words verbalized by parents during their answer to the interview questions and were incorporated into the development of the coding schema. Sentences were coded as neutral when parents' reactions or opinions did not align with any of the positive/favorable, negative/unfavorable, surprised or confused codes and could not be classified objectively. Quantitative analysis was conducted to better understand the associations between the emergent themes identified in the dataset. NVivo output in the form of a "coding summary report" was transformed into a spreadsheet indicating the presence or absence of codes within transcripts. Non-parametric tests were performed using SPSS.

Results

This section describes parents' responses to two open-ended acceptability questions, with descriptive quotations. Results revealed three main themes: parents' opinions about the Project's follow-up interview, reasons why parents found the Project's follow-up interview beneficial, and parents' emotional reactions to the Project's follow-up interview. Due to the qualitative nature of this analysis, not all parents commented on all themes.

Theme 1: Parents' opinions about the Project's follow-up interview

Of the 195 parents' responses included in this analysis, most contained at least one comment about their general opinions about the Project's follow-up interview (172/195, 88%; parents

of CF carriers 59/65, 91% and SCH carriers 113/130, 87%). Further details about the reasons for these opinions are presented under Theme 2 below.

Most parents reported only favorable opinions of the Project's follow-up interview (n=141/195, 72%; parents of CF carriers 48/65, 74% and SCH carriers 93/130, 72%). Sentences were coded as "favorable" when parents' responses were considered encouraging, complimentary, or implied overall merit, support, or approval of receiving follow-up from the Project.

"Um, I actually was glad that I got the letter because I do think there is a lot more education that needs to be done about newborn screening so I was glad that, you know, that there's a whole project looking into, you know, just bettering the program...I think that's a good idea because I think there are probably a lot of people who don't fully understand it..." (mother of a CF carrier infant)

A code for unfavorable opinion of the follow-up interview was developed, but remained unused. One parent of a CF carrier reported general confusion or uncertainty as their only opinion.

Some parents expressed only neutral opinions about the Project's follow-up interview (n=11/195, 6%; parents of CF carriers 5/65, 8% and SCH carriers 6/130, 5%). Sentences such as "*I think it's okay*" or "*I don't see nothing bad about it*" were coded as neutral. A minority of parents reported combinations of favorable, neutral, or confused opinions about the project (n=19/195, 10%; parents of CF carriers 5/65, 8% and SCH carriers 14/130, 11%).

Theme 2: Reasons why parents found the Project's follow-up interview beneficial

Many parents (n=120/195, 62%; parents of CF carriers 33/65, 51% and SCH carriers 87/130, 67%) reported at least one reason they found the follow-up interview beneficial. Over half of these 120 parents reported two or more reasons (n=64/195, 34%; parents of CF carriers 21/65, 32% and SCH carriers 43/130, 33%). The five most commonly reported reasons are listed in Table 2. The most frequent reason reported was that the follow-up was an opportunity to obtain additional information about such topics as genetics or general NBS information. Parents also reported benefit to receiving an explanation or further clarification about their infants' NBS results, especially when they were confused about the meaning of the results. Consequently, several parents reported an increased understanding about the NBS results. Parents considered the interview an opportunity to obtain answers to their questions not addressed at the time of NBS results disclosure or that arose later. For many parents, the interview reinforced NBS information they had already heard from their infant's provider or reassured them that their infant was healthy. Lastly, parents reasoned that the interview was beneficial because it offered additional resources and emotional support.

Some parents reportedly had not been informed about their infant's NBS results by the PCP and received them for the first time during the follow-up interview (n=17/130, 13% parents of SCH carriers only). For them, the interview represented their only source of information about the NBS results. Most of these parents (n=15) reported this as a beneficial reason for the follow-up interview. Some parents chose not to answer any of our research questions upon hearing their infant's results and requested that the call become a clinical intervention, where the interviewer provided counseling to parents. These interviews were not recorded and therefore do not have transcripts available for analysis. Since no transcripts were available for this set of parents, interview notes were reviewed to find all instances of results disclosure. By the start of our analysis in August of 2010, the study team had completed interviews for 461 parents, to include 306 parents of SCH carriers, and informed an additional 30 parents of their infant's SCH carrier results (47 parents in total). This totals

15% (47 of the 306 interviews) of parents participating as part of this quality improvement Project.

Theme 3: Parents' emotional reactions to the Project's follow-up interview

Most parents (n=174/195, 89%; parents of CF carriers 60/65, 92% and SCH carriers 114/130, 88%) expressed an initial emotional reaction to receiving follow-up from the Project's team. Emotional reactions were organized into four subthemes: positive, negative, surprise, and neutral or no reaction (Table 3). Some parents described more than one emotional reaction (n=29/195, 15%; parents of CF carriers 11/65, 17% and SCH carriers 18/130, 14%), in which case each reaction was coded separately.

Parents mentioned positive emotional reactions to receiving the follow-up that included feeling happy, reassured, comforted, relieved, excited, and appreciative (n=27/195, 14%; parents of CF carriers 8/65, 12% and SCH carriers 19/130, 14%). Although most parents described their positive reactions to our interview with a specific emotion, several parents simply stated they had a “good” or “nice” reaction generally.

Negative emotional reactions were occasionally mentioned (n=2/195, 1%; parents of CF carriers 1/65, 2% and SCH carriers 1/130, <1%). The most common negative emotions were feeling anxious, scared, or worried. Two parents attributed these emotions to not expecting our phone contact. Some parents initially assumed that we were calling to discuss a different, previously undisclosed NBS result. This resulted in initial anxiety or fear, which was alleviated once the Project's study team clarified the reason for the call at the beginning of the interview. In two instances, parents expressed initial annoyance with our follow-up calls because they assumed we were telemarketers.

“Well, at first I thought you were a telemarketer so I was going to read you off. But then you said you know who you were you know what you were calling about and then I was like, “Okay, all right, that's fine, didn't have to tell you off.” (mother of a CF carrier infant)

Some parents reported feeling surprised by our follow-up interview (n=12/195, 6%; parents of CF carriers 4/65, 6% and SCH carriers 8/130, 6%). Of these, several commented that they had not been expecting our phone call and they did not remember receiving our recruitment letter. Other parents reported surprise because of the time lapse (approximately 3-5 months) between the NBS process and the follow-up interview, or they were unaware that research is being done to follow-up with parents after NBS.

Many parents expressed a neutral reaction or stated they did not have a reaction to participating in the Project's follow-up interview such as “*I felt fine*,” “*it didn't bother me*,” or “*I didn't have a reaction*” (n=104/195, 53%; parents of CF carriers 36/65, 55% and SCH carriers 68/130, 52%). Many of these parents commented that they were expecting our call because they remembered the recruitment letter or voicemails from the Project's study team.

Quantitative Analysis

Associations between the three themes are marked in Tables 2 and 3 with distinct symbols. We tested for associations between parents' emotional reactions (Theme 3) and their opinions about the Project's follow-up interview (Theme 1). All categories of emotional reactions (positive, negative, neutral, and surprised) were associated with favorable opinions of the follow-up interview (p = 0.001). Only parents with neutral emotional reactions were likely to have neutral opinions about the follow-up interview (p<0.05).

We also found association between overall opinions about the follow-up interview (Theme 1) and parents' reasons for finding the follow-up interview beneficial (Theme 2). The

following three reasons were significantly associated with favorable opinions about the follow-up interview ($p<0.05$): providing information, clarifying NBS results, and answering questions (Table 2). No association was found between neutral opinions about the follow-up interview and reporting a reason for finding the interview beneficial.

Parents' emotional reaction to the follow-up interview (Theme 3) was associated with (Theme 2) reporting a reason for finding the interview beneficial (Table 3). Parents with neutral, negative, and surprised reactions were more likely to report a reason for finding the follow-up interview beneficial ($p<0.05$). Parents with positive reactions were not significantly associated with finding the follow-up interview beneficial in this secondary analysis ($p=0.077$), however the data show a trend toward significance.

Discussion

This qualitative, secondary analysis of interview acceptability questions is the first report of its kind that addresses acceptability of telephone follow-up regarding NBS carrier results. Our data from parent responses to open-ended interview questions show that routine telephone follow-up after NBS is equally acceptable to parents of both SCH and CF carriers. Indeed, the majority of parents interviewed (72%) had favorable opinions of the follow-up interview and many parents (62%) reported reasons why they found the follow-up beneficial.

Parents' emotional reactions to receiving follow-up contact were reported among both groups of parents proportionally, and all reactions were significantly associated with an overall favorable opinion of the follow-up contact. It should be noted that even parents with initial neutral, negative or surprised reactions were significantly likely to identify reasons they found the interview beneficial, while a non-significant trend for finding the follow-up interview beneficial was found among parents who reported an initial positive reaction. We hypothesize these parents with positive reactions already had sufficient information and understanding about their infant's NBS results and therefore did not need further counseling or clarification from the interviewer. This potentially minimized their perceived benefit from the follow-up contact.

The data suggest that parents of carriers for CF and SCH found the follow-up calls beneficial, but perhaps for different reasons. For CF carrier parents, a larger proportion reported that the interview provided them with another resource and an opportunity to have questions answered that either were not addressed by PCPs at initial results disclosure or specialists upon sweat testing. Perhaps, new questions arose in the interim between results disclosure and the follow-up interview.

Alternatively, a larger proportion of SCH carrier parents reported that the interview was beneficial because it provided them with additional information and increased their understanding by clarifying the NBS results. Often, parents wanted more information about inheritance, especially regarding the possibility of carrier status in themselves and family members. Other parents were confused about the differences between SCH trait and SCH disease and the implications of these differences such as questioning whether trait could become disease in the future.

Perhaps the most noteworthy finding in this analysis was among parents of SCH carriers. The follow-up interview served as the primary source of information for 12% - 15% of parents regarding their infants' status as SCH carriers. There are several reasons parents may not have heard results. Parents may have changed PCPs or clinics, and so the result would have been sent to the incorrect location, or they may have missed appointments when the results would have been discussed. On several occasions throughout this study, our team

found that a simple newborn screening card omission such as the infant's first name could lead to confusion, or if the infant's name had changed from the time the baby was screened in the hospital (i.e. NBS card was submitted with mom's last name "baby boy Smith," but infant's full name was actually "John Jones"). It is possible that another family member knew the infant's NBS results other than the parent interviewed (i.e. mom took infant to results disclosure appointment, but our study team interviewed dad). Some parents reported thinking that their infant's NBS results were mailed to them in a formal letter, but they never read the letter. Lastly, it may be possible that the infant's parent was actually told the NBS result, but didn't remember hearing them.

The healthcare expectation in pediatric and family medicine is that all parents are told their infant's NBS results, provided adequate information, and offered necessary counseling by PCPs (and later by other health professionals, in the case of CF). We were surprised to find that such a large portion of parents interviewed had either not been told their infant's NBS results or had lingering concerns, questions, and knowledge gap. Even with PCPs doing their best to inform parents of their infant's NBS results, these findings suggest that our health care system may not adequately meet the informational needs of this population. Furthermore, this information deficit illustrates the importance of NBS follow-up, and the need for comprehensive NBS communication and counseling.

There are several limitations to our analysis. First, interview participation was potentially subject to self-selection bias. Only parents wanting to participate in the Project's follow-up interview completed the interview, possibly skewing data toward favorable opinions and feedback. Second, social desirability may have influenced parents' responses to our interview questions. It is possible that participants did not want to report negative opinions to the interviewer, skewing data toward favorable responses. Lastly, PCPs may have introduced bias by advising against inclusion of families meeting the Project's contraindications. Most contraindications identified were language barriers (non-English speaking), and only a few families were identified as having "psychosocial" issues. In fact, PCPs were mostly encouraging of the Project's follow-up interview for families with possible "psychosocial" issues, noting the benefits of additional attention and follow-up.

Despite these limitations, the clinical implications of these data suggest the importance for PCPs to fully disclose NBS carrier results to all parents. Additionally, access to clinical follow-up after initial results disclosure is valuable and relevant to parents regarding their infant's CF or SCH carrier status, perhaps via telephone or at subsequent PCP appointments. Though telephone follow-up from the statewide NBS program may be impractical, clinical follow-up by PCPs or other clinical professionals may be more feasible and can provide parents an opportunity to ask additional questions and access further information.

Conclusion

Routine telephone follow-up for parents of CF and SCH carriers identified via NBS is both feasible and acceptable to parents. Most parents interviewed had favorable opinions and identified specific benefits to receiving the follow-up contact. Furthermore, this analysis demonstrates a potential information deficit among carrier parents and illustrates the importance of NBS follow-up, and the need for comprehensive NBS communication and counseling. Future studies of the larger Project will examine these informational deficits, the need for clinical follow-up, and the gaps in reporting NBS results.

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Participant Demographics**Table 1**

Variable	n	%
Parent Gender		
Female	190	97.4
Male	5	2.6
Parent Race/Ethnicity		
Black	92	47.2
White	79	40.5
Latino	6	3.1
Other	12	6.2
Not asked	6	3.1
Parent Age (years)		
<20	22	11.3
20-29	114	58.5
30-39	51	26.2
40-49	3	1.5
Not asked	5	2.6
Infant's carrier status		
Carrier for sickle cell disease (trait)	130	66.7
Carrier for cystic fibrosis ^a	65	33.3

^aCF carrier infants in this analysis had an elevated immunoreactive trypsinogen (IRT), a single CF-associated mutation, and a normal sweat chloride test.

Table 2
Reasons why parents found the Project's follow-up interview beneficial ^a

Reason	Total % (n)	SCH % (n)	CF % (n)	Quote
Provided Information ^b	26% (51/195)	31% (40/130)	17% (11/65)	"...I'm happy because that was something that I actually wish that I could've talked to somebody about because I felt like, you know, I know that there's other parents going through this, too, and it would've been really nice to have more information especially right up front." (mother, CF carrier infant)
Clarified results or increased understanding ^c	23% (44/195)	25% (33/130)	17% (11/65)	"Yes, 'cause as you said its probably a lot of people that don't understand the difference in the trait and the disease and I don't even really understand the...the ...how they [PCP] were breaking down the likelihood of if we had more kids you know the chances of that kid getting the disease and not the trait so it gets even more confusing for me so ...um and a lot of people probably won't reach out and ...and ask so it's I think is good that you're seeking out the parents to make sure that they do understand." (mother, SCH carrier infant)
Answered Questions ^b	18% (35/195)	16% (21/130)	22% (14/65)	"I think that there's a lot of us out here who have questions and sometimes we don't get the answers that we need, or we don't have the time to get the answers to the questions that we have, so if we can just sit down with people and, you know, ask questions and have them answer them for us and understand and, you know, see where we're coming from, that's a great thing." (mother, CF carrier infant)
Provided another resource	9% (18/195)	8% (11/130)	11% (7/65)	"I think it's a really good idea because, you know, especially if there are some parents that are still worried or concerned or have questions...they have another person they could talk to about it." (mother, CF carrier infant)
Received results for first time	8% (15/195)	12% (15/130)	n/a	"I like [the follow-up call] very much because before you called me I didn't even know he had the trait...So...I... I do appreciate [the study team] taking the time out to call new ... new mothers like myself 'cause I didn't know anything about this at all or the results." (mother, SCH carrier infant)

^a"SCH" refers to parents of SCH carrier infants; "CF" refers to parents of CF carrier infants; percents are rounded to the nearest whole percent

^bAssociated with (Theme 1) favorable opinion of the Project (p<0.05)

^cAssociated with (Theme 1) favorable opinion of the Project (p<0.001)

Table 3
Parents' emotional reactions to the Project's follow-up interview ^a

Emotional Reaction	Total % (n)	SCH % (n)	CF % (n)	Quote
Positive ^b	14% (27/195)	14% (19/130)	12% (8/65)	"Um, it was actually a relief [to get the follow-up call]...because... like I was saying I was in the hospital and I really didn't know what was really going on...'cause I wasn't really in my mind. But it's actually a relief...[pause]... I'm very thankful really." (mother, SCH carrier infant)
Negative ^{b d}	1% (2/195)	<1% (1/130)	2% (1/65)	"At first, you know, you get a little bit worried...'cause you might... something might be wrong...but, um, you know, once I started the conversation and found out more about the call, you know, I was much more relaxed, knowing that, you know, nothing was wrong." (mother, SCH carrier infant)
Neutral ^{b c e}	53% (104/195)	52% (68/130)	55% (36/65)	"Um, actually, I was expecting the phone call because they had said you guys were going to call on the letter you guys sent to me so I was kind of expecting it." (mother, SCH carrier infant)
Surprise ^{b e}	6% (12/195)	6% (8/130)	6% (4/65)	"Um, well I was surprised – I didn't realize that they did, um, follow-ups on this sort of thing, but, I'm – I'm glad to see that they do." (mother, CF carrier infant)

^a"SCH" refers to parents of SCH carrier infants; "CF" refers to parents of CF carrier infants; percents are rounded to the nearest whole percent

^b Associated with (Theme 1) favorable opinion about the follow-up interview (p<0.001)

^c Associated with (Theme 1) neutral opinion about the follow-up interview (p<0.05)

^d Associated with (Theme 2) reporting a reason for finding the interview beneficial (p<0.05)

^e Associated with (Theme 2) reporting a reason for finding the interview beneficial (p<0.001)