The False-Positive in Universal Newborn Hearing Screening

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ABSTRACT. *Objectives.* **Concern has been raised about the frequency and subsequent emotional effect of a false-positive result during universal newborn hearing screening (UNHS). This study describes: 1) the results of 1 UNHS program and a potential method to significantly reduce the false-positive rate, and 2) the effect a false-positive result has on lasting maternal anxiety toward their children as well as their views toward UNHS in general.**

Methods. **A retrospective analysis was conducted using data from 5010 infants screened with an automated auditory brainstem response (ABR) at the Women's Hospital of Greensboro (WHOG) from July 6, 1998 to June 30, 1999. In addition, a structured telephone survey was given to mothers of infants who had failed the initial hearing screen (stage 1) and who had completed an outpatient rescreen (stage 2).**

Results. **Confirmed hearing loss occurred in nonneonatal intensive care unit infants at a rate of 1.8/1000. A false-positive rate of 1.9% occurred during stage 1 of UNHS (screening before newborn discharge). We attribute this relatively low rate to rescreening of 51% of those newborns who failed the initial screen before hospital discharge. Eighty percent of these rescreened infants passed, thus needing no additional follow-up. If we had rescreened all infants before discharge, the false-positive rate would have approached .5%.**

Results of the survey were reassuring with regard to lasting emotional effects of false-positive tests. Only 9% of mothers said they "treated their child differently" before outpatient rescreening, and only 14% reported any lasting anxiety after their child passed the outpatient repeat screen. Although none reached statistical significance, potential risk factors for lasting anxiety include more educated mothers, lack of understanding of UNHS, and a false-positive result in both stage 1 and stage 2. Over 90% of all mothers believed that UNHS was a good idea.

Conclusions. **By rescreening all infants before hospital discharge, the false-positive rate of UNHS performed using automated ABR can be reduced to <1%. However, for the false-positive results that do occur, any long-lasting and detrimental emotional impact between mother and infant seems to be small and could be reduced even more with improved understanding about UNHS.** *Pediatrics* **2000;106(1). URL: http://www.pediatrics.org/cgi/content/full/106/1/e7;** ***universal newborn hearing screening, screening, false-positive, maternal anxiety, hearing, audiology.***

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ABBREVIATIONS. UNHS, universal newborn hearing screening; WHOG, Women's Hospital of Greensboro, North Carolina; ABR, auditory brainstem response; NICU, neonatal intensive care unit.

Universal newborn hearing screening (UNHS) is aimed at the early detection of and intervention for children with congenital hearing loss.[1] In the absence of hearing screening, moderate to severe hearing loss is often not identified until 1 to 2 years of age and mild hearing loss not identified until school age.[1–4] In addition, targeted screening of only those children who meet high-risk registry criteria may miss up to 50% of infants with hearing loss.[5] Because of these concerns, in 1993 a National Institute of Health Consensus Statement recommended UNHS.[1] Since then, numerous other organizations including the American Academy of Pediatrics have supported this recommendation.[6] In response to these recommendations, a number of states have legislated UNHS.[5]

However, support for UNHS is not universal. One of the most concerning issues raised is the high rate of false-positive results. The literature reports false-positive rates between 3% and 8%.[5,7–11] This has caused a number of critics to decline to recommend UNHS until the false-positive rate can be decreased and/or there is further knowledge of the emotional effect this false-positive labeling has on families.[12–14] A number of studies from other newborn-screening tests have shown that false-positive results can engender lasting anxiety and adversely affect the parent–child relationship.[15–18] In addition, deUzcategiu and Yoshinga-Itano[19] surveyed mothers immediately after their children had failed the newborn hearing screen and found that 20% to 50% of mothers reported feelings, such as anger, confusion, depression, frustration, shock, and sadness. However, it is still unknown how persistent or detrimental these feelings are.

By using the results of the first year of UNHS at the Women's Hospital of Greensboro, North Carolina (WHOG), these 2 important issues were studied: 1) how to minimize the overall rate of false-positive tests, and 2) the lasting emotional effect a false-positive result has on a newborn's family.

METHODS

The Screening Program

In response to a proposed mandate by the state of North Carolina (which has subsequently been legislated), all newborns at WHOG were screened for hearing loss beginning July 6, 1998. The WHOG is part of the Moses Cone Health System and is the only maternity hospital that serves Guilford County as well as a num-

ber of surrounding counties. A trained technician used an Algo 2 or an Algo 2e auditory brainstem response (ABR) screener (Natus Medical Inc, San Carlos, CA). This automated hearing screener uses a 35-dB nHL alternating polarity click to assess the neural response of the auditory nerve. The equipment has a built-in artifact rejection for myogenic, electrical, and environmental noise interference that stops the screen when testing conditions would preclude adequate testing. The automated screener provides a pass–refer result that requires no interpretation. An immediate retest was performed on obtaining a refer and was considered part of the initial screen. This initial screen (and immediate rescreen if necessary) was designated stage 1a. Screening occurred every day of the week. When time and adequate staffing allowed, another automated ABR was performed before discharge on those infants who had failed the initial screen (ie, within the next 12–24 hours). This was designated as stage 1b. Results of each screening test were reported to the infant's physician, who was responsible to discuss the results and any need for follow-up with the parents.

Newborns failing stage 1 were referred for outpatient screening (stage 2). Stage 2 screening was performed by an audiologist and consisted of an automated ABR and, if necessary, a diagnostic ABR. Failure of stage 2 initiated a referral for additional evaluation (ie, otolaryngologist and hearing aid evaluation). Data were collected on all infants screened at WHOG between July 6, 1998 and June 30, 1999 as well as on any infants who required any follow-up screening or evaluation.

Survey

All non-neonatal intensive care unit -(NICU) infants who completed stage 2 screening were identified, and a parent was contacted by telephone. After obtaining verbal consent, a structured questionnaire was administered by a single person and included questions about the results of the screenings, parental anxiety level, subsequent treatment of and feelings toward the infant, and feelings about UNHS. In addition, demographics, such as age, parity, and educational level, were collected. Respondents whose children had true hearing loss were excluded from the results (ie, only false-positives were included). Whether any lasting anxiety remained after the completion of stage 2 screening was determined by the question, "Has the process of testing and retesting your child's hearing caused you (much anxiety, some anxiety, no anxiety, some reassurance, or much reassurance)?" Using Fisher's exact test, respondents who reported much anxiety or some anxiety after the completion of stage 2 screening (designated lasting anxiety) were compared with those who did not. Statistical analysis was performed using SPSS 8.0 (SPSS, Chicago, IL). The study was approved by the Moses Cone Hospital institutional review board.

RESULTS

The Screening Program

During the study, 5034 healthy term newborns were born at WHOG, of which 5010 (99.5%) had stage 1 hearing screens performed (Table 1). Of the 175 (3.5%) infants who failed the stage 1a screen, 90 (51%) had a stage 1b screen performed, of which 72 (80%) passed. Therefore, 103 infants were referred for stage 2 follow-up screening (2.1%). Eighty-five of the 103 children completed stage 2 rescreening (83%), and of these, 15 (17%) failed and were referred for additional evaluation. Nine subsequently were confirmed to have hearing loss, of which 4 had severe, bilateral loss. One of the 15 children who failed stage 2 screening was lost to follow-up. This is a rate of hearing loss of 1.8/1000 normal newborns and severe bilateral loss in .8/1000. The overall stage 1 false-positive rate was 1.9% (conservatively assuming the 19 children lost to follow-up all had false-positive results) and a positive predictive value of 8.7%. Because we did not collect data on false-negative tests, sensitivity and negative predictive value of the screening process could not be calculated.

TABLE 1. Results of UNHS at WHOG From July 6, 1998 to June 30, 1999 for Normal Newborns

Category	*n*	Percentage of Previous	Percentage of Total Screened
Normal newborns (non-NICU)	5034	—	—
Total screened	5010	99.5	—
Failed stage 1a screen*	175	3.5	3.5
Received stage 1b screen†	90	51	
Failed stage 1b screen	18	20	
Total failed stage 1	103	—	2.1
Received stage 2 screen‡	85	83	1.7
Failed stage 2	15	17	.3
Diagnostic referral	15	100	.3
Diagnostic evaluation	14	93	.28
Confirmed hearing loss	9	64	.18

* Stage 1a screen—initial hearing screen.
† Stage 1b screen—repeat hearing screen before hospital discharge of newborn.
‡ Stage 2 screen—outpatient screen by trained audiologist.

Table 2 lists the 15 children who failed stage 2 and their outcomes. Four had severe bilateral hearing loss and 5 had mild to moderate hearing loss. Three of the 4 children with severe bilateral hearing loss met high-risk registry criteria. Five infants who failed stage 2 screening were later found to have normal hearing (ie, stage 2 false-positives).

All infants admitted to the NICU at WHOG had hearing screening. Of the 454 infants screened, 5 infants failed stage 1 screening. Four of these were subsequently confirmed with hearing loss, while the fifth child was lost to follow-up. This is a rate of hearing loss of 8/1000 of NICU children.

Survey

Forty-nine (64%) mothers of the 76 eligible infants completed the survey. Of the 27 (36%) who did not complete the survey, 22 were unable to be contacted, and 5 did not speak English. An audiologist administered all the surveys within a 1-month period. Demographic characteristics between the respondents and the nonrespondents are similar as shown in Table 3. For respondents, the survey was administered an average of 4.9 months after UNHS (range: 2–13 months).

Despite a protocol instructing only the physician to tell parents of the stage 1 screening test results, the mothers who were surveyed remember this occurring only 35% of the time. More often, they recalled a nurse or technician reporting the results. Respondents reported not having an opportunity to ask questions about the test or the results 31% of the time, and 47% could not identify which ear(s) failed. Fifty-five percent believed the purpose of the test and the meaning of the results were not adequately explained.

Slightly >80% of mothers reported that they worried about their child's hearing before the follow-up (stage 2) examination. However, 91% claimed that neither they nor any other family member treated their child differently during this time. The other 9% reported speaking more loudly or clapping hands to test their children's hearing.

TABLE 2. Characteristics of the 15 Children With Suspected Hearing Loss

Type of Suspected Hearing Loss	Confirmed Loss	Risk Factor	Diagnostic Referral
1. Unilateral moderate	Yes	None	ENT
2. Unilateral severe	Yes	None	ENT
3. Unilateral mild	Yes	None	Audiology
4. Bilateral profound	Yes	Deaf parents	ENT
5. Unilateral moderate	Normal	None	Audiology
6. Bilateral profound	Yes	Charge syndrome	ENT
7. Unilateral	Normal	None	ENT
8. Unilateral	Did not return	None	None
9. Unilateral	Normal	None	ENT
10. Bilateral moderate	Yes	None	ENT
11. Bilateral profound (auditory neuropathy)	Yes	None	ENT
12. Bilateral profound	Yes	Deaf parents	Audiology
13. Bilateral asymmetric	Yes	Abnormal pinna	ENT
14. Bilateral	Normal	None	ENT
15. Unilateral mild	Normal	None	Audiology

ENT indicates ear, nose, and throat.

Eighty-six percent of stage 2 screens were accomplished within 2 weeks of the infant's birth. After completing stage 2 screening, 86% reported no lasting anxiety concerning the process. Only 1 mother (2%) reported "much anxiety." This was a mother whose child had both false-positive stage 1 and stage 2 results. Six others (12%) reported only "mild anxiety." Table 4 compares differences between the 7 mothers with any lasting anxiety and the 42 without. None of the variables studied reached statistical significance, but a few trends could be seen with regard to risk factors for lasting anxiety. These included: maternal college education, not fully understanding the meaning of the test results, and the infant having a false-positive result at stage 2 screening.

Regardless of anxiety, 94% of all respondents were glad that their children had a hearing test and thought that UNHS was a good idea.

DISCUSSION

Our UNHS program at WHOG identified hearing loss in 9 of the 5010 normal newborns screened for a rate of 1.8/1000. Four infants had severe, bilateral loss for a rate of .8/1000. Both the rates for severe loss and overall loss are similar to those previously reported in the literature.[6,9,20,21] We report a stage 1 false-positive rate of 1.9%, lower than previous reports of 3% to 8%.[5,7–11] We attribute this relatively low rate to rescreening 51% of newborns before hospital discharge (stage 1b). Had we rescreened no infants, our false-positive rate would have been 3.3% (42% higher) and consistent with previous reports. In contrast, if time and resources had permitted, complete stage 1b rescreening (assuming an 80% pass rate) would have resulted in only 35 of the 5010 newborns being referred for outpatient rescreening (stage 2). This 80% decrease in stage 2 referrals would have given us an overall stage 1 false-positive rate of .5%. The potential monetary, manpower, and emotional savings of rescreening all infants before discharge are significant.

In addition to rescreening infants before discharge, the use of the automated ABR as the screening tool was also an important factor in minimizing false-positive results. It has been shown to consistently

TABLE 3. Survey Results of Mothers Whose Infants Failed Stage One of UNHS: Demographic Characteristics

Characteristics	Respondents (n = 49) Mean (Range)	Nonrespondents (n = 27) Mean (Range)
Age (y)	26.5 (16–40)	26.2 (19–37)
Number of children	1.4 (1–4)	1.8 (1–4)
Education (y)	13.2 (9–16)	12.6 (5–17)
Age (mo) of infant at time of survey	4.9 (2–13)	6.2 (2–13)

TABLE 4. Survey Results of Mothers Whose Infants Failed Stage One of UNHS*

Survey Questions	Lasting Anxiety† n = 7	No Lasting Anxiety† n = 42
Maternal age >24 y	7 (100%)	31 (74%)
<1 child	4 (57%)	22 (52%)
>High school education	6 (86%)	23 (55%)
Infant <2 mo of age at time of survey	2 (29%)	7 (17%)
Results were explained to me by a physician	2 (29%)	15 (36%)
I could ask questions about UNHS and the results	3 (43%)	31 (74%)
I know which ear(s) failed	3 (43%)	23 (55%)
I fully understood the meaning of the results of the hearing screen	2 (29%)	20 (48%)
I worried about my infant's hearing after stage 1 screening	6 (86%)	35 (83%)
The repeat test took <2 wk to occur	6 (86%)	36 (76%)
I or another family member treated our infant differently because of the results of the UNHS	1 (14%)	3 (7%)
My child gets sick more often than other children	0 (0%)	1 (2%)
My child failed stage 2 screening but upon referral had normal hearing	2 (29%)	2 (5%)
I am glad that my infant's hearing was tested at birth	6 (86%)	40 (95%)
I think that UNHS is a good idea	6 (86%)	40 (95%)

* Response to questions stratified by presence of lasting anxiety attributable to the UNHS process (n = 49).
† All P values >.05.

produce lower false-positive rates than the otoacoustic emissions test, the other commonly used screening tool.[9]

Our survey of mothers whose children had a false-positive screening test is reassuring with regard to any lasting anxiety after stage 2 screening. Although our survey was designed only to look at lasting anxiety, it is reassuring to know that despite a high level of anxiety before stage 2 screening, 91% of families treated their infants normally during this time. It seems that false-positive testing is unlikely to create a "vulnerable child syndrome" that can occur in situations such as this.

No factor was overwhelmingly responsible for causing lasting anxiety, but the factors that may play some part include: more educated mothers, a misunderstanding of the meaning of the screening results, and a false-positive test during stage 2 screening. However, even in this group of mothers with the most reason to have negative feelings toward UNHS, support of UNHS was overwhelmingly positive. Compared with the results of the survey by deUzcategiu and Yoshinaga-Itano,[19] it seems that initial feelings of anxiety are common but dissipate quickly after the child passes stage 2 screening.

Despite the reassuring findings of our survey, the potential for negative feelings caused by UNHS remains and must be continually addressed. Based on our study, we believe that a good strategy to minimize undesirable outcomes with UNHS is twofold. First, minimize false-positives by requiring infant rescreening before hospital discharge. Current recommendations in North Carolina state only that "reasonable efforts should be made to repeat the initial screening protocol close to the time of discharge" in those infants who have failed.[22] The potential 80% decrease in false-positive results should make the advantages of this practice apparent.

A second way to minimize negative parental feelings regarding UNHS is to improve communication during the stage 1 screen. According to our survey, the lack of parental understanding with regard to the purpose of the test and the meaning of a "fail" was common. More study needs to go into who discusses the results and what language is used. However, in our anecdotal experience, a trained audiologist or nurse with more time to spend with the family may be more effective than a busy or less knowledgeable physician.

This study has some limitations. First, we do not know the number of false-negatives (ie, those newborns passing the screen and subsequently identified as hearing impaired). Although previous studies have documented a very high sensitivity of the ABR,[23] there are a number of reports where hearing loss has been missed. For example, ABR screening of newborns does not detect hearing loss with onset later in infancy or childhood.[24–26] Guarding against a false sense of security cannot be overemphasized.

Second, our survey sample size is small and generalizing the results to other populations should be done with caution. Our cohort of mothers may be older and more educated than many. In addition, results may be biased in a few important ways. Selection bias may be present attributable to the fact that one third of potential respondents were not surveyed. We cannot be certain that respondents are representative of the entire group. For example, we do not know whether there are cultural differences in feelings toward UNHS, because only English speaking mothers completed the survey. However, as seen in Table 3, characteristics of respondents seem similar to those of nonrespondents. Finally, recall bias may be present. As shown in Table 3, the average time elapsed between UNHS and the survey administration was 4.9 months. It is likely that significant negative feelings are forgotten or minimized as the memory of UNHS becomes more distant. However, as seen in Table 4, it did not seem that those mothers surveyed closer to the time of UNHS (within 2 months) were significantly more likely to have developed any lasting anxiety.

CONCLUSION

In summary, lasting anxiety from false-positive hearing screens seems to be uncommon. Despite this reassuring finding, rescreening all eligible newborns before hospital discharge and clear communication with families are necessary to make this screening tool even better.

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