Supplement

A Multisite Study to Examine the Efficacy of the Otoacoustic Emission/Automated Auditory Brainstem Response Newborn Hearing Screening Protocol: Research Design and Results of the Study

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Purpose: Most newborns are screened for hearing loss, and many hospitals use a 2-stage protocol in which all infants are screened first with otoacoustic emissions (OAEs). In this protocol, no additional testing is done for those passing the OAE screening, but infants failing the OAE are also screened with automated auditory brainstem response (A-ABR). This study evaluated how many infants who failed the OAE and passed the A-ABR had permanent hearing loss (PHL) at 8–12 months of age.
Method: A total of 86,634 infants were screened at 7 birthing centers using a 2-stage OAE/A-ABR hearing screening protocol. Of infants who failed the OAE but passed the A-ABR, 1,524 were enrolled in the study. Diagnostic audiologic evaluations were performed on 64% of the enrolled infants (1,432 ears from 973 infants) when they were 8–12 months old.
Results: Twenty-one infants (30 ears) who passed the newborn A-ABR hearing screening were identified with PHL when they were 8–12 months old. Most (71%) had mild hearing loss.
Conclusions: If all infants were screened for hearing loss using a typical 2-stage OAE/A-ABR protocol, approximately 23% of those with PHL at 8–12 months of age would have passed the A-ABR.

Key Words: newborn hearing screening, otoacoustic emissions, automated auditory brainstem response, false-negatives, congenital hearing loss

Thirty-three children are born every day in the United States with congenital hearing loss—more than any other birth defect with such serious developmental consequences (Leonard, Shen, Howe, & Egler, 1999; Stierman, 1994; White, 1997). In March of 1993, when the National Institutes of Health (NIH) recommended that all newborns be screened for hearing loss, less than 3% of newborns in the United States were being screened. Since that time, the number of infants screened for hearing loss has grown exponentially, and as of January 2005, 93% of newborns in the United States were being screened for hearing loss based on data collected by the National Center for Hearing Assessment and Management (NCHAM, 2005). In fact, with the help of federal funding, every state has now established an early hearing detection and intervention program with a goal of ensuring that all newborns are screened for hearing loss before 1 month of age and that those who fail the screening receive audiologic diagnosis before 3 months of age and are enrolled in appropriate early intervention programs before 6 months of age (White, 2003).

Even though almost all newborns in the United States are now screened for hearing loss, significant problems remain in ensuring that infants who do not pass the hearing screening test are quickly diagnosed and that those with permanent hearing loss (PHL) receive timely and appropriate early intervention services. Indeed, the Centers for Disease Control and Prevention (CDC, 2005) estimated that 44% of the infants referred from newborn hearing screening programs are "lost to the system" before completing a diagnostic evaluation. Given how difficult it has been to complete diagnostic evaluations for all referred infants, it is not surprising that newborn hearing screening programs have tried various strategies to reduce the number of infants who fail the hospital-based screening test and consequently need some type of follow-up (Clemens & Davis, 2001; Finitzo, 2000; Isaacson, 2000).

One frequently used strategy is a two-stage screening protocol prior to hospital discharge. In this protocol, newborns are screened first with either distortion product or transient evoked otoacoustic emissions (OAEs), and those who fail are screened with automated auditory brainstem response (A-ABR). Those who pass the first-stage OAE test or the second-stage A-ABR test are considered to have minimal risk for hearing loss and are not followed further, as recommended by the Joint Committee on Infant Hearing (JCIH, 2000; see also Prieve, 2000). According to White (2003), 17.3% of all newborns were being screened with this two-stage OAE/A-ABR protocol in January 2003, and the percentage seems to be increasing. One of the primary reasons this protocol is being used more frequently is the belief that it will lead to substantially lower referral rates at the time of hospital discharge, often as low as 1% of all newborns (Clemens & Davis, 2001).

Despite the fact that such a protocol can substantially reduce the referral rate (making follow-up easier), there is some concern that it might be missing babies with PHL (see Johnson, White, Widen, Gravel, Vohr, et al., 2005). The purpose of this study was to determine how many infants who fail the OAE but pass the A-ABR in a typically implemented two-stage newborn hearing screening protocol have a PHL when they are 8–12 months old.

Study Design

As noted by Johnson, White, Widen, Gravel, Vohr, et al. (2005), the study was designed according to specifications contained in a request for applications issued by the CDC. As required by the request for applications, hospitals with existing newborn hearing screening programs using an OAE/A-ABR protocol were recruited to participate. From these hospitals, a sample of infants who failed the OAE but passed the A-ABR were enrolled. These infants, who typically would not have been referred for a diagnostic audiologic evaluation because they passed the A-ABR in the hospital, were assessed with visual reinforcement audiometry (VRA) and other audiologic assessments at between 8 and 12 months of age to determine their hearing status (see Widen et al., 2005, for a detailed explanation of the diagnostic procedures and equipment). The prevalence of PHL in this group of infants who failed the

OAE and passed the A-ABR was also compared with the hearing status of infants in the same birth cohort who had failed both the OAE and the A-ABR and were consequently referred for an audiologic evaluation under that hospital's typical protocol for newborn hearing screening.

Eight hospital systems (11 hospitals)—2 in New York and 1 each in Connecticut, Rhode Island, Florida, Ohio, Kansas, and Hawaiì—were recruited to participate in the study. Each of the hospitals in these systems met the following criteria for participating in the study: (a) operation of a newborn hearing screening program using the two-stage OAE/A-ABR screening protocol for at least 6 months, (b) a historical referral rate of less than 10% for their OAE and less than 4% for their A-ABR screening, and (c) completion of diagnostic evaluations for more than 85% of the referrals from their newborn hearing screening program. As a group, the hospital systems served populations that had ethnic and socioeconomic characteristics similar to the U.S. population.

Because the purpose of this study was to determine how many infants who fail the OAE but pass the A-ABR in a typically implemented two-stage newborn hearing screening protocol have a PHL when they are 8–12 months old, it was important that the hospitals be using screening equipment and protocols that were generally representative of most newborn hearing screening programs. As noted in Johnson, White, Widen, Gravel, Vohr, et al. (2005), first-stage newborn hearing screening in the hospitals was done using either transient evoked otoacoustic emission (TEOAE) equipment from Otodynamics or distortion product otoacoustic emission (DPOAE) screening equipment from Biologic Systems. OAE screening was often repeated a second or third time if the infant did not pass for both ears on the first attempt. The automated TEOAE equipment in the study used nonlinear clicks at 84 ($\pm$3) dB peak equivalent SPL. The stimulus parameters for the DPOAE equipment included F2 frequencies of 2000, 3000, 4000, and 5000, an F2:F1 ratio of 1.22, and L1 intensity of 65 dB SPL and L2 intensity of 55 dB SPL. The Otodynamics EchoCheck and the Biologic AuDX automatically determine the stimulus level, stopping rules, and pass/fail result based on preset criteria. The EchoCheck requires a 6-dB signal-to-noise ratio in the region from 1.8 to 3.6 kHz to pass. A pass for the Biologic AuDX requires a distortion product amplitude of at least 6 dB above the noise floor for three of four frequencies from 2.0, 3.0, 4.0, and 5.0 kHz. For sites using the Otodynamics Echoport or ILO88, technicians judged an infant as having passed the screen if the equipment showed a 6-dB signal-to-noise ratio at 2.0, 3.0, and 4.0 kHz.

All sites in the study used Algo Newborn Hearing Screeners from Natus Medical for A-ABR screening. According to Natus, all of the Algo models use a 35-dB nHL alternating polarity click stimulus, although there have been no independent published studies that have examined the stimulus level of the Natus equipment (see Gravel et al., 2005, for further discussion of this issue). The Natus equipment automatically provides an indication of whether the infant passes or fails based on a proprietary algorithm. Consistent with the goal of the study to examine what happens in typical newborn hearing screening programs using the two-stage OAE/A-ABR protocol, all screening equipment was used as recommended by the manufacturers' user manuals. As is the case with all newborn hearing screening programs to our knowledge, OAE equipment was calibrated by the manufacturer prior to being sold and further calibration was not recommended. A-ABR equipment was calibrated by Natus representatives one time each year as recommended by the manufacturer. Unfortunately, no specific information was available from any of these companies as to how calibration was done. This important issue is discussed in more detail by Gravel et al. (2005). It is clear that calibration issues have not been addressed as thoroughly as they should be by manufacturers, and this point should not be trivialized.

After the study had been under way for more than a year, one of the hospitals was dropped from the study because it was no longer meeting the prespecified criteria for inclusion. Specifically, the OAE referral rate for infants screened at this hospital was more than double the criteria of 10%. Thus, seven hospital systems (consisting of 10 hospitals) completed the study and provided data for the analyses reported in this article. All participants in the study signed an informed consent, and study protocols and procedures were approved by an institutional review board in each of these hospital systems (hereafter referred to as sites) as well as the University of Hawaiì and the CDC.

In each of the sites, infants who failed the OAE and passed the A-ABR were eligible for participation in the study if the parents spoke English or Spanish (resource constraints made it impossible for the study to translate the study materials and arrange for language interpreters for the small number of families who did not speak English or Spanish). Eligible parents from this group were contacted, the research study was explained, and parents were invited to participate in the study. For parents who agreed and provided signed informed consent, data were collected about demographic characteristics (e.g., number of children, income, race/ethnicity), the health of the infant (e.g., gestational age, birth weight, days in neonatal intensive care unit [NICU]), and the presence of risk indicators specified by the JCIH (2000) for late onset or progressive hearing loss (e.g., family history of congenital hearing loss, in utero infections, syndromes associated with late onset hearing loss). At the time of enrollment, parents agreed to bring their infant back for a diagnostic audiologic assessment at 8–12 months of age and to allow the research team to access the infant's medical record.

As families were enrolled in the study, the previously described data collected at each site were deidentified by replacing personal identifying information with a coded identification number known only to the coinvestigator at that site, and the data were then sent to the data coordination office at NCHAM at Utah State University. Staff at NCHAM created a data file using SPSS and checked for missing values and incorrectly entered data by checking for values outside the range of possible values for each variable. The diagnostic data described below were also

entered into SPSS. Monthly summaries of all data were posted to a secure Web site maintained by NCHAM. The site could be accessed by all coinvestigators at any time with a user name and password. The coinvestigators reviewed these data summaries during monthly telephone conferences in which project design and management issues were discussed. These discussions enabled the team to find and correct data errors, track enrollment and progress with diagnostic evaluations, and make midcourse corrections based on previously collected data.

Of the 3,462 families whose newborn failed the OAE but passed the A-ABR during the enrollment period, 2,678 were invited to enroll. As shown in Table 1, 1,524 of those were enrolled, and 1,154 were invited but declined to participate. Parents of an additional 784 infants were not approached because of staff shortages at the hospital, because of other scheduling and administrative issues, or because the parents did not speak English or Spanish. As will be explained in more detail below, the fact that only 44% of the infants who had failed the OAE but passed the A-ABR were actually enrolled in the study is important in interpreting the findings.

To maintain contact with the family until the infant was old enough to complete the specified diagnostic protocol, postcards were sent to the parents when the infant was 2, 4, and 6 months old. Each postcard reminded the parents about the study and invited them to return a tear-off card with several short questions about the infant's developmental status. The postcards were sent with a request for address correction so that the U.S. Postal Service provided address corrections to the data coordination office at NCHAM when a family had moved and left a forwarding address. When the infant was 7 months old, staff at each site began contacting the family to make an appointment to conduct an audiologic diagnostic assessment of the infant.

Staff members at each site were encouraged to use a variety of procedures to locate difficult-to-find families. For example, at the time of enrollment, families were invited to provide contact information for "baby's doctor/clinic" and for a "relative or friend to contact if we cannot find you." This contact information was used if the family could not be reached using the phone number or address on the enrollment form. Site staff also used Internet address locators (e.g., www.whitepages.com), and in some cases current contact information was obtained from the hospital's billing office.

Infants were enrolled in the study from May 1, 2001, through January 31, 2003. A total of 86,634 infants were born at these hospitals during the enrollment period. Most sites enrolled infants from both the well baby nursery and the NICU, although Sites 3 and 4 enrolled only infants from the well baby nursery (see Johnson, White, Widen, Gravel, Vohr, et al., 2005). During the time that infants were enrolled in the study, the participating hospitals averaged a 4.8% referral rate for OAEs and 1.0% referral rate for A-ABRs. Of the 1,524 infants who were enrolled in the study, 973 (64%) returned for a diagnostic evaluation. For a substantial number of those infants who were enrolled in the study, only 1 ear met the study criteria. Thus, 1,432 ears were evaluated for the study.

Diagnostic Evaluations and Determination of Hearing Status

Infants who failed the OAE but passed the A-ABR screening tests were invited to return for an audiologic diagnostic evaluation when they were 8–12 months old. As described in more detail by Widen et al. (2005), the evaluation consisted of at least VRA, tympanometry, and OAEs using a protocol similar to one used by a large multicenter study funded by NIH in the early 1990s (Widen et al., 2000). Frequency-specific ABR and bone conduction testing were used where appropriate. The goal of the diagnostic evaluation for infants in this study was to collect minimum response levels of 15 dB HL at 0.5, 1.0, 2.0, and 4.0 kHz for each ear. The initial diagnostic evaluation was completed when infants were an average of 9.7 months old. Approximately 32% of the infants for whom diagnostic data were obtained required more than one visit to complete this protocol. A small remuneration, generally $20, was provided to reimburse families for the costs of travel, parking, and child care, whenever they returned for an evaluation.

Table 2 shows the criteria developed by the research team for classifying the hearing status of each child based on the results of the diagnostic audiologic evaluations. Using these criteria, it was possible for an infant to be determined to have a PHL at only one or two frequencies. In some cases, it was impossible to make a definitive determination because a child did not return for subsequent visits. Thus, the categories of "probable not permanent hearing loss," "increased suspicion of permanent hearing loss," and "not sufficient data to rule out permanent hearing loss"

Table 1. Enrollment statistics for participating sites.

Site	Births during enrollment	Eligible for enrollment		Total enrolled		Not recruited		Refusals	
		n	%	*n*	%	*n*	%	*n*	%
1	16,608	1,044	6.3	191	18.3	418	40	435	41.7
2	9,393	421	4.5	370	87.9	18	4.3	33	7.8
3	4,509	285	6.3	84	29.5	186	65.3	15	5.3
4	9,252	209	2.3	147	70.3	30	14.4	32	15.3
5	24,032	456	1.9	170	37.3	11	2.4	275	60.3
6	6,217	433	7.0	266	61.4	50	11.5	117	27.0
7	16,623	614	3.7	296	48.2	71	11.6	247	40.2
Total	86,634	3,462	4.0	1,524	44.0	784	22.6	1,154	33.3

Table 2. Definitions used to determine hearing status.

Hearing status category	Description
Not permanent hearing loss	Using the "best" results from all assessments, MRL thresholds of ≤20 dB HL at 1.0, 2.0, and 4.0 kHz.
Probable not permanent hearing loss	MRL data not available at 1.0, 2.0, and 4.0 kHz, but all frequencies had MRLs ≤20 dB *or* OAEs within normal limits[a] *or* Toneburst ABR data ≤25 dB nHL.
Permanent sensorineural hearing loss	MRLs ≥25 dB at 1.0, 2.0, or 4.0 kHz (tested with good confidence) *or* ABR threshold ≥30 dB nHL *and,* if tested, OAEs below normal limits at the frequencies with elevated MRLs *and* normal middle ear functioning based on tympanometry or bone conduction.
Permanent conductive hearing loss	MRLs ≥25 dB at 1.0, 2.0, or 4.0 kHz (tested with good confidence) *and,* if tested, OAEs below normal limits *and* bone conduction thresholds ≤20 dB with an air/bone gap ≥15 dB at frequencies with MRLs ≥25 dB. Otitis media ruled out based on clinical examination and tympanometry.
High increased suspicion of permanent hearing loss	MRLs ≥25 dB at 1.0, 2.0, or 4.0 kHz, *but* OAEs within normal limits for those frequencies *or* only fair confidence in VRA testing.
Some increased suspicion of permanent hearing loss	MRLs ≥30 dB at one frequency or ≥25 dB at more than one frequency, *but* abnormal tympanometry *and* no bone conduction. *or* Sound field thresholds ≥25 dB (with fair confidence) *and* normal tympanometry *and* OAEs below normal limits.
Not sufficient data to rule out permanent hearing loss	Even though child returned for diagnostic evaluation, no MRLs or OAEs within normal limits for 1.0, 2.0, or 4.0 kHz, and none of the above criteria for permanent hearing loss were met.

Note. MRL = minimum response level; OAEs = otoacoustic emissions; ABR = auditory brainstem response; VRA = visual reinforcement audiometry.

[a]Normal OAEs were defined as a signal-to-noise ratio of ≥3 dB for 1.0 and ≥6 dB for 2.0–4.0 kHz.

were created (see Widen et al., 2005, for further discussion and examples).

Table 3 shows examples of how these criteria were applied to make determinations about an infant's hearing status. For example, the first row of Table 3 shows the results for an infant's ear classified as "not permanent hearing loss." During the first diagnostic visit, VRA responses for the ear were not obtained as indicated in the spaces for 1.0, 2.0, and 4.0 kHz. However, OAEs were obtained, and those were ≥6 dB signal-to-noise ratio at 2.0 and 4.0 kHz. As explained in the footnote to Table 3, tympanometry results were questionable during this first visit.[1] During a second assessment for this infant, minimum response levels of 15 dB were obtained at all four frequencies (with good confidence in the results), tympanometry results were normal (as defined by Widen et al., 2005), and OAEs of ≥6 dB were obtained at 1.0, 2.0, and 4.0 kHz. Thus, this ear was classified as not having PHL.

As another example, consider the ear in the third row of Table 3 that was classified as having a permanent sensorineural hearing loss. At the first assessment, minimum response levels of 30, 30, and 45 dB HL at 1.0, 2.0, and 4.0 kHz, respectively, were obtained for VRA testing, which was done with good confidence. Tympanometry was normal, and the OAE results were less than 3 dB at 1.0, 2.0, and 4.0 kHz. Thus, the first assessment met the criteria for a PHL. However, this infant, as was the case with almost all infants classified in the study as having PHL, was tested a second time to confirm the results. At the second assessment, the minimum response levels at 1.0, 2.0, and 4.0 kHz were still elevated (45, 30 and 50 dB HL at 1.0, 2.0, and 4.0 kHz, respectively, as indicated in the center of that row under Assessment 2). The quality of this assessment was good at only 2.0 and 4.0 kHz, tympanometry results were normal again, but the OAE signal-to-noise ratio was ≥6 dB at 1.0 and 2.0 kHz. Thus, this ear was classified as having a PHL only at 4.0 kHz since the OAE results at the other frequencies for this assessment were inconsistent with the minimum response levels obtained with VRA.

One additional example emphasizes the conservative approach used in this study for classifying an ear as having a PHL. In the fourth row of Table 3 is an ear classified as having "high suspicion" of PHL. This ear had elevated minimum response levels at 1.0, 2.0, and 4.0 kHz during the first assessment period, but tympanometry results were abnormal, meaning those elevated thresholds could be due to otitis media. During a second VRA session (shown in the middle of the row), minimum response levels were still elevated at 1.0, 2.0, and 4.0 kHz, but the quality of the assessment was good only at 2.0 and 4.0 kHz. Tympanometry was normal, and OAEs were ≤3 dB at 1.0, 2.0, and 4.0 kHz.

Based on these data, it could be argued that the child should have been classified as having a PHL. However,

[1]As explained in Widen et al. (2005), tympanograms were coded as normal (physical volume between 0.3 and 1.0 cc, static admittance 0.2 to 1.0 mmhos, and tympanometric width of <235 daPa), abnormal (normal volume, with static admittance <0.2 mmhos, tympanometric width of >235 daPa, essentially "no peak" per tympanometric screening), questionable (one value was missing, usually tympanometric width), could not test, or did not test.

Table 3. Examples of how hearing status was categorized.

Category assigned	Assessment 1									Assessment 2									Assessment 3								
	MRLs					Tymp result	OAEs			MRLs					Tymp result	OAEs			MRLs					Tymp result	OAEs		
	0.5	1.0	2.0	4.0	Qual		1.0	2.0	4.0	0.5	1.0	2.0	4.0	Qual		1.0	2.0	4.0	0.5	1.0	2.0	4.0	Qual		1.0	2.0	4.0
Not PHL	D	D	D	D		3	D	6	6	15	15	15	15	1	1	6	6	6									
Probable not PHL	15	C	15	20	1	1	6	6	6																		
PHL sensorineural	30	30	30	45	1	1	2	2	2	30	45	30	50	1.5	1	6	6	2									
High suspicion of PHL	D	35	25	25	1	2	2	6	2	D	30	35	35	1.5	1	2	2	2	25	D	D	30	2	3	6	6	6
Some suspicion of PHL	55	50	45	65	1	3	2	2	2																		
Insufficient data	C	C	20	C	1	1	D	6	6	D	D	D	D		4	D	6	6									

Note. Frequencies are kilohertz. For tympanometry (tymp) results, 1 = normal tympanograms (all variables with normal limits), 2 = abnormal tympanograms (at least one variable in abnormal range), 3 = questionable due to missing tymp width, and 4 = missing data (all four variables were coded as missing). Qual = quality of the VRA result and is an average of the frequencies rated, with 1 = good and 2 = fair; PHL = permanent hearing loss; D = did not test; C = could not test. OAE data are coded as 2 = <3 dB signal-to-noise ratio, 4 = 3–6 dB signal-to-noise ratio, and 6 = >6 dB signal-to-noise ratio.

Table 4. Hearing status of ears that failed OAE and passed A-ABR.

Site	Not PHL ears	Probable not PHL ears	Ears with PHL		Ears with increased suspicion of PHL		Ears with insufficient data	No. of infants with diagnostic data	Percentage of enrolled infants with diagnostic data
			SN	PC	High	Some			
1 (148 ears)	131	6	0	0	0	0	11	81	42.4
2 (478 ears)	432	35	7	0	0	0	4	299	80.8
3 (59 ears)	40	6	0	0	0	2	11	42	50.0
4 (165 ears)	82	24	10	5	17	12	15	109	74.1
5 (111 ears)	58	16	2	0	0	5	30	86	50.6
6 (241 ears)	202	8	4	0	2	8	17	184	69.2
7 (230 ears)	195	5	2	0	0	1	27	172	58.1
Total (1,432 ears)	1,140	100	25	5	19	28	115	973	63.8
%	79.6	7.0	1.7	0.3	1.3	2.0	8.0		

Note. A-ABR = automated auditory brainstem response; SN = sensorineural; PC = permanent conductive.

during a third VRA session, the tympanometry results were questionable, but the infant had OAEs of $\geq$6 dB signal-to-noise ratio at 1.0, 2.0, and 4.0 kHz. Such robust OAEs made it less certain that a PHL was present. Consequently, the infant was classified as high suspicion. Even though several attempts were made to bring the parents back for a fourth assessment, which would have been the most desirable outcome, the parents did not return. Because of conflicting information between the VRA testing and the OAEs, this ear was placed in the high suspicion category instead of classifying it as PHL. That such a conservative approach was used to classify an ear as having PHL is important to remember when the results of the study are interpreted.

Results

Table 4 shows how the ears of the 973 infants who returned for diagnostic audiologic assessment were classified, using the criteria outlined in Table 2. As can be seen, 79.6% of the 1,432 ears were classified as not PHL, with an additional 7% classified as probable not PHL. Thirty ears (2.1% of the ears in the study) from 21 infants were classified as having PHL, and an additional 19 ears from 12 infants (1.2% of the ears in the study group) were classified as having a high suspicion of PHL. For 8% of the ears, there was not enough diagnostic evaluation data to make a determination about hearing status. As discussed more fully by Widen et al. (2005), it is important to note that only 63.8% of the infants in the study group returned for diagnostic evaluation. Thus, there is no information about the hearing status for 36.2% of the infants in the group initially enrolled in the study.

Table 5 summarizes demographic and neonatal characteristics of each infant in the study group who was diagnosed with unilateral (n = 12) or bilateral (n = 9) PHL at 8–12 months of age. As can be seen, 6 of the 21 infants (29%) had spent time in the NICU, and 8 of 21 (38%) had one or more of the JCIH risk indicators for progressive or late onset hearing loss.[2] Although it would be interesting to evaluate how degree and laterality of hearing loss (unilateral vs. bilateral) was related to NICU stay and presence of JCIH risk indicators, the small number of infants in each condition made this impossible.

To interpret the clinical significance of finding PHL at about 9 months of age for 21 infants who passed an A-ABR newborn hearing screening test, the following reference points were used:

- First, how many infants were identified with PHL in addition to those who would have been identified otherwise based on failing the OAE and failing the A-ABR? In other words, how many infants with PHL were identified in the comparison group?
- Second, a number of infants qualified to be in the study group even though one of their ears passed the initial OAE; this was because the other ear failed the OAE and subsequently passed an A-ABR. Thus, a substantial number of infants who had passed the initial screening test in one ear returned for diagnostic evaluations. The question of whether any, and if so how many, initially passed ears were classified as having a PHL during the diagnostic assessment provides an important reference point for interpreting the significance of the number of ears found with PHL among those that failed the initial OAE but passed the A-ABR.
- Third, it is important to consider how many of these 21 infants were likely to have had congenital versus late onset hearing loss.

[2]Although the JCIH position statement (JCIH, 2000) combines risk indicators for "late onset" and "progressive" hearing loss, the terms have very different meanings. A child with late onset hearing loss has normal hearing at birth and acquires a hearing loss at a later age. Thus, congenital and late onset hearing losses are mutually exclusive. A progressive hearing loss is one that becomes worse over time. Thus, either a congenital or a late onset hearing loss can be progressive. For this study, differentiating between congenital and late onset losses was most important. Although there are many important reasons to know whether a hearing loss is progressive or stable, determining whether losses were progressive was not a goal of this study.

Table 5. Demographic and neonatal characteristics of infants with PHL in study group.

Infant	Site	ID	Ear	Type of hearing loss	Hearing status of "other" ear	No. of days in NICU	Gestational age (weeks)	Birthweight (g)	JCIH risk indicators for late onset or progressive PHL	Gender	Annual household income	Mother's ethnicity	Health insurance
1	2	053	R	Mild SN		0	38	3150		M	$35,000	White	Private insurance
			L	Mild SN									
2	2	091	R	Mild SN		64	27	645	PPH; mechanical ventilation	M	$35,000	Black	Private HMO
			L	Mild SN									
3	2	130	L	Mild SN	PHL	5	38	3160	Stickler syndrome	F	$45,000	White	Private insurance
4	2	131	L	Mild SN	PHL	42	27	1450		M	$45,000	White	Private insurance
5	2	148	R	Mild SN	nh	37	28	1205	PPH; mechanical ventilation	M	$25,000	White	Medicaid
6	4	005	R	Moderate PC		0	39	2942		M	$15,000	Asian	Private insurance
			L	Mild PC									
7	4	020	L	Mild SN	nh	0	40	2940		F	$50,000+	White	Private insurance
8	4	027	R	Mild PC		0	40	3456		M	$25,000	Asian	Private insurance
			L	Mild PC									
9	4	029	R	Mild SN	nh	0	40	3136	In utero infection (herpes)	F	$50,000+	Mixed	Private HMO
10	4	055	R	Mild SN	nh	0	37	4060		M	$50,000+	Mixed	Private HMO
11	4	066	R	Mild PC	nh	0	39	2892	Stigmata associated with syndrome	M	$45,000	Pacific Islander	Private HMO
12	4	089	R	Mild SN	nh	0	37	3535	Family history	M	$15,000	Mixed	Medicaid
13	4	122	R	Moderate SN		0	38	2368		F	$7,500	Asian	Private insurance
			L	Moderate SN									
14	4	126	R	Mild SN		0	42	3379		F	$25,000	Asian	Medicaid
			L	Mild SN									
15	4	138	R	Mild SN		0	38	2832		M	$50,000+	Mixed	Private HMO
			L	Mild SN									
16	5	046	R	Severe SN	nh	0	40	3288		M	$45,000	White	Private HMO
17	5	065	R	Profound SN	PHL	5	36	2126	In utero infection (CMV)	F	$50,000+	Black	Private HMO
18	6	002	R	Mild SN		0	37	2830		M	<$5,000	White	Medicaid
			R	Mild SN									
19	6	003	L	Moderate SN	nh	0	37	3040		M	<$5,000	White	Medicaid
20	6	072	L	Mild SN	nh	40	40	2942	Down syndrome	M	$50,000+	White	Private insurance
21	7	258	R	Moderate SN		0	36	2892		M	$35,000	White	Private insurance
			L	Mild SN									

Note. NICU = neonatal intensive care unit; JCIH = Joint Committee on Infant Hearing; R = right; L = left; nh = normal hearing; PPH = persistent pulmonary hypertension; CMV = cytomegalovirus.

Table 6. PHL in comparison group of infants who failed OAE and failed A-ABR.

Site/births	Babies with PHL			Ears with PHL			Prevalence of PHL (per 1,000)	Referred for diagnosis		Completed diagnosis	
	SN	PC	Total	SN	PC	Total		%	*n*	%	*n*
1 / 16,608	17	1	18	24	2	26	1.08	1.2	199	82.4	164
2 / 9,393	18	1	19	31	2	33	2.02	1.5	140	95.7	134
3 / 4,509	4	0	4	6	0	6	0.89	0.2	9	88.9	8
4 / 9,252	16	0	16	27	0	27	1.73	0.3	28	96.4	27
5 / 24,032	39	3	42	60	3	63	1.75	0.8	193	87.6	169
6 / 6,217	16	1	17	25	2	27	2.73	0.7	41	65.9	27
7 / 16,623	36	6	42	55	6	61	2.53	0.6	94	79.8	75
Total: 86,634	146	12	158	228	15	243	1.82	0.8	704	85.8	604

Using the same birth cohort from the same sites, 704 infants failed OAE and failed A-ABR. Of these infants, 604 (85.8%) had enough diagnostic evaluation data to make a determination about their hearing status. As shown in Table 6, of the 604 infants evaluated, 158 children with 204 ears were diagnosed as having PHL. This represents a prevalence of 1.82 per 1,000 in this birth cohort. It should be noted that this prevalence is probably a little lower than would be expected in a general population cohort, because two of the seven sites only recruited infants from the well baby nursery (in the data reported in Table 6, only infants from the well baby nursery were included in the comparison group for those sites).

Table 7 shows that 71.4% of the infants with PHL in the study group had mild hearing loss (i.e., less than 40 dB pure-tone average), whereas only 19.6% of the infants with PHL in the comparison group had mild hearing loss. This result is consistent with concerns that led the CDC to fund this study: specifically, that infants with mild hearing loss might not be identified with the OAE/A-ABR protocol. In contrast, infants with moderate to profound hearing loss represent only 28.6% of the study group, but 80.1% of the PHL in the comparison group.

To interpret the significance of finding 21 infants (30 ears) with PHL among those who passed the A-ABR newborn hearing screening test, it is also useful to examine the prevalence of hearing loss among those who passed the OAE newborn hearing screening test. Although the study was not specifically designed to do this, the question can be answered to some degree since there were a number of infants enrolled in the study because one ear failed the initial OAE and the A-ABR while the other ear passed the initial OAE screening in the hospital. When these infants returned for a diagnostic evaluation of the ear targeted by the study, the ear that passed the initial OAE was often tested. If a significant number of the ears that passed the initial OAE had been identified with PHL, it would raise questions about the significance of finding 30 ears with PHL among the ears that failed OAE and passed A-ABR.

As shown in Table 8, none of the ears of infants in the study group who passed the initial OAE screening were identified with PHL. It should be noted that a larger percentage of infants in this group were classified as not having sufficient data to make a determination about hearing status. This may have happened because testers did not try as hard to complete the protocol for a "nonstudy" ear. For the 375 ears for which data were available, no ears were identified with PHL.

Another important question is how many of the ears with PHL that passed the A-ABR during newborn hearing screening were congenital losses and how many were late onset losses? Because the study was not designed to answer this question, it is impossible to provide a definitive answer. However, some information on this important question is available. First, as shown earlier in Table 5, only 8 of the 21 infants had one or more of the risk indicators for late onset or progressive hearing loss identified by the JCIH (2000). Even though most of these risk indicators are not particularly good predictors of late onset hearing loss (Cone-Wesson et al., 2000; JCIH, 2000), the fact that 13 of the 21 infants exhibited none of the risk indicators for either late onset or progressive loss suggests that at least some of these losses were congenital. This interpretation is consistent with the fact that most of the hearing losses in the study group were mild, which is what would have been expected if the two-stage OAE/A-ABR screening protocol were missing mild hearing losses. It is also consistent with the data from the NIH multicenter

Table 7. Degree of hearing loss for infants in study and comparison groups.

Group	Mild (20–40 dB)		Moderate (41–70 dB)		Severe through profound (>70 dB)		Total infants with PHL	
	n	%	*n*	%	*n*	%	*n*	%
Study group	15	71.4	5	23.8	1	4.8	21	100
Comparison group	31	19.6	64	40.5	63	39.6	158	100
Total	46	25.7	69	38.5	64	35.8	179	100

Note. As measured in the worse ear.

Table 8. Hearing status for ears of study group infants who passed initial OAE newborn hearing screening.

Site	Total ears	PHL		Increased suspicion of PHL		Not PHL	Probable not PHL	Not sufficient data
		SN	PC	High	Some			
1	13	0	0	0	0	11	0	2
2	112	0	0	1	0	107	3	1
3	25	0	0	1	0	3	15	6
4	53	0	0	2	6	19	15	11
5	53	0	0	1	2	30	5	15
6	127	0	0	3	1	60	38	25
7	113	0	0	0	0	30	22	61
Total	496	0	0	8	9	260	98	121
%	100	0.0	0.0	1.6	1.8	52.4	19.8	24.4

study of hearing screening reported by Cone-Wesson et al. (2000) in which only 1 of 56 children with PHL by 12 months of age was thought to be a late onset loss. Thus, it is very likely that some infants with congenital hearing loss are being missed by the two-stage OAE/A-ABR screening protocol, but it cannot be concluded with certainty exactly how many of the instances of hearing loss in the 21 infants were congenital and how many were late onset.

Estimating the Percentage of Children With PHL at 8–12 Months of Age Who Fail the OAE and Pass the A-ABR Newborn Hearing Screening Test

Based on the results of this study, the question remains, what is the best estimate of the number of infants with PHL at 8–12 months of age who would fail OAE and pass A-ABR newborn screening? The following three issues should be considered in making this estimate:

- First, how lenient or strict were the criteria used for determining whether a child had PHL?
- Second, sites varied considerably in the number of infants identified with PHL. Thus, consideration should be given to whether all sites should be weighted equally in making this estimate.
- Finally, some important characteristics of the study group and the comparison group may warrant adjustments in making the "best" estimate.

Criteria for classifying PHL. As shown earlier in Table 4, 21 infants (30 ears) who failed OAE and passed A-ABR were determined to have a PHL when they returned for audiologic assessment at 8–12 months of age. There were also 19 ears from 12 infants in the study group who were at high increased suspicion for PHL but did not meet the strict criteria used in this study for classifying a child as having a PHL. As shown in Table 9, the estimated percentage of infants with PHL who would pass the A-ABR during newborn hearing screening is quite different based on which of these estimates is used (2.06 vs. 2.49 per 1,000).

It could be argued that the definition for PHL was so strict that it underestimated the true number of children with PHL at 8–12 months of age. Certainly those infants who were at high increased suspicion of PHL were not hearing normally at the time of the audiologic assessment. As illustrated by the earlier example in Table 3, these infants typically had elevated minimum response levels of 25 to 35 dB HL in one or more frequencies between 2.0 and 4.0 kHz, but the quality of testing was only fair and/or there were data from the OAE testing at the frequencies with elevated thresholds that were consistent with normal hearing. Given conflicting information from the VRA testing and the results of the OAE testing, it would have been best if these children had returned for additional testing. Unfortunately, extensive efforts to get these families to return were not successful. If those children who were at high increased suspicion had been included, the prevalence of hearing loss in the group of children who failed OAE but passed A-ABR would have almost doubled. However, the research team concluded that the evidence for hearing loss was not strong enough to justify including these infants in the group judged to have PHL.

Variation across sites. Another important consideration relates to the variation in number of infants in the study group identified with PHL across the participating sites. In conducting such a study, an implicit, but often unexamined, assumption is that study procedures were equally well implemented at all of the sites. To the degree that this is not true, data from some sites may be a better estimate than data from other sites about whether infants with hearing loss could fail the OAE but pass the A-ABR.

Table 10 shows that there was substantial variation across the seven sites related to the quality of implementation. Site 2 enrolled the highest percentage of eligible

Table 9. Prevalence of PHL in study and comparison groups based on different criteria.

	Comparison group	Study group	Total
Based only on those meeting criteria for PHL	1.82 (158 infants)	.24 (21 infants)	2.06 (179 infants)
Including those categorized as high suspicion	1.82 (158 infants)	.43 (33 infants)	2.49 (191 infants)

Table 10. Quality of implementation and number of infants with PHL identified at various sites.

Site	No. of study group infants with PHL	Births during enrollment period	Eligible infants enrolled		Refusals during recruitment		% returning for diagnostic evaluations	% with "not sufficient data"	Average rank for implementation quality of site[a]
			%	*n*	%	*n*			
1	0	16,608	18.3	191	41.7	435	42.4	7.4	5.8
2	5	9,393	87.9	370	7.8	33	80.8	0.8	1.3
3	0	4,509	29.5	84	5.3	15	50.0	18.6	4.8
4	10	9,252	70.3	147	15.3	32	74.1	9.1	2.8
5	2	24,032	37.3	170	60.3	275	50.6	27.0	6.0
6	3	6,217	61.4	266	27.0	117	69.2	7.1	3.0
7	1	16,623	48.2	296	40.2	247	58.1	11.7	4.5

[a]1 = best.

infants, had the highest percentage who returned for diagnostic evaluations, and had the lowest percentage of infants for whom there were insufficient data to make a classification. Site 3 had the lowest percentage of infants who refused to participate during the enrollment process. Based on the data summarized in Table 10, Sites 2 and 4 were the two sites that best implemented the study. These two sites also identified the highest number of infants in the study with PHL (15 of 21 infants). It is noteworthy that only 33.9% of the enrolled infants but 71.4% of the infants with hearing loss came from these two sites. The fact that most of the infants with hearing loss who failed OAE and passed A-ABR screening came from the best implemented sites suggests that the estimates based on the entire sample are probably conservative and may underestimate the true percentage of infants with PHL who fail OAE and pass A-ABR newborn hearing screening.

Comparability of study group and comparison group. In estimating the percentage of infants with PHL at 8–12 months of age who are likely to pass the A-ABR after failing the OAE, it is important to account for several other characteristics of this data set. Because the infants in the study group and the comparison group came from the same hospitals during the same time period, they were quite similar with respect to variables such as ethnicity, family composition, family income, and type of insurance coverage. However, on two other variables that could substantially affect the estimated prevalence of PHL, the study group and the comparison group were quite different:

- First, in the study group, only 44% of the infants who failed the OAE and passed the A-ABR were actually recruited for the study and, consequently, invited to return for a diagnostic assessment at 8–12 months of age. Thus, the estimated prevalence for the study group is based on only 44% of the population. But in the comparison group, 100% of the infants who failed the OAE and failed the A-ABR were invited to come back for a diagnostic evaluation.
- Second, a different percentage of infants in each of these groups returned for a diagnostic evaluation. It should be noted that parents in the study group were given approximately $20 to pay for incidentals such as travel, parking, and child care each time they came for a diagnostic assessment, but parents in the comparison group were not given this inducement to return. Only 64% of the infants in the study group returned, whereas 87% of the infants who failed the OAE and failed the A-ABR returned for a diagnostic evaluation.

It is important to consider how these differences affected estimates of the prevalence of PHL.

There is no reason to believe that parents of infants who were not invited to participate in the study were any different than those who were invited, and little reason to think that the prevalence of hearing loss would be lower among infants of parents who declined to participate in the study than among those who agreed to participate. Thus, had 100% of the eligible infants been enrolled in the study, it is almost certain that additional infants with PHL would have been identified. Given that the increased prevalence of PHL was 0.24 per 1,000, based on the 44% who were enrolled and invited to return for a diagnostic evaluation, it is estimated that the prevalence of hearing loss would have been 0.55 per 1,000 if 100% of the eligible infants had been enrolled and invited to return (0.24/0.44 = 0.55).

The likely consequence of having a substantially different percentage of infants in the two groups who completed the diagnostic evaluation is less clear. Sixty-four percent of the infants in the study group and 87% of the infants in the comparison group returned for diagnostic evaluation. Even though 23% more parents returned for diagnostic evaluation in the comparison group than in the study group, we cannot be confident about why this happened. It may be that families who think their child has a hearing loss are more likely to return. If that happened, there would probably be a higher prevalence of hearing loss among those who returned than there was among those who did not return. Higher return rates for children in the comparison group may also be because parents of these children received more definitive information at the time their infant left the hospital about the fact that their child had failed a hearing screening and needed to come back for a diagnostic evaluation. There was also greater effort from the public health system and probably more encouragement from health care providers for the parents in the

Table 11. Various estimates of the percentage of 9-month-old children with PHL who failed OAE and passed A-ABR.

	Infants who failed OAE/failed A-ABR	Infants who failed OAE/passed A-ABR	
		Based on 44% who participated	Adjusted for those who did not participate
All sites	1.82 per 1,000	2.06 per 1,000; 0.24 per 1,000 increase; 12% of PHL in cohort	2.37 per 1,000; 0.55 per 1,000 increase; 23% of PHL in cohort
Sites with best implementation	2.27 per 1,000	2.75 per 1,000; 0.48 per 1,000 increase; 17% of PHL in cohort	2.95 per 1,000; 0.68 per 1,000 increase; 23% of PHL in cohort

comparison group to bring their infants back for diagnostic evaluations. Thus, the lower percentage of returning parents in the study group may have been because they were told that their infant had passed the screening and, consequently, they might not have been very motivated to return (in which case, returning for a diagnostic evaluation would probably be uncorrelated with whether the child had a PHL).

Other factors could also have contributed to a lower prevalence of PHL among those who returned than among those who did not return. For example, the factors contributing to low follow-up rates may be the same factors that increase the risk of an infant having a hearing loss. For example, it is plausible that families who are poor, have single heads of household, are transient, or have poor health are less likely to return for an evaluation appointment and also more likely to have an infant with hearing loss. To the degree this is true, the observed prevalence of PHL would be artificially low in the study group because this group had a lower return rate than the comparison group.

Return rates in the two groups may also have been affected by the fact that parents in the study group were given a monetary incentive to return ($20 per visit), but parents in the comparison group were not. This probably increased the return rate in the study group, but it is very unlikely that it created any bias regarding the prevalence of children identified with hearing loss.

Because some variables associated with a lower return rate for diagnostic evaluations among infants in the study group would most likely lead to an *overestimate* of hearing loss, and some variables would most likely lead to an *underestimate* of hearing loss, it seems unwise to make any adjustments for the differences in "return rates" between the groups.

Making the "best estimate." Table 11 summarizes how the preceding variables would lead to different estimates of what percentage of 8–12-month-old children who passed the A-ABR newborn hearing screening test would have PHL. Basing the estimate only on the 44% of eligible infants who were invited to return for a diagnostic evaluation gives a substantially lower estimate than if the estimate is adjusted for what it would have been if all eligible infants had been invited to return for a diagnostic evaluation. However, basing the estimate only on the sites with the best implementation, versus basing it on all sites, makes very little difference because even though the best implemented sites identified the most infants with hearing loss, they also had the highest percentage of eligible infants enrolled. Taking all of the data together, the best estimate is that 23% of children with PHL at 8–12 months of age will have failed the OAE and passed the A-ABR during newborn hearing screening.

Conclusions

A substantial number of infants who pass the A-ABR portion of a two-stage newborn hearing screening OAE/A-ABR protocol will have PHL in one or more ears and at one or more frequencies when they are 8–12 months old. The best estimate is that this will be about 0.55 infants per 1,000, or 23%, of all infants with PHL in the cohort. Most of these will be infants with mild sensorineural hearing loss, and it is impossible to determine with the data from this study exactly how many of these losses are congenital versus late onset hearing losses. However, the fact that 62% of the infants who passed the newborn hearing screening A-ABR did not have any of the JCIH risk indicators for either late onset or progressive hearing loss suggests that a substantial number of the "missed" infants had congenital hearing loss.

It would be inappropriate, and often incorrect, to conclude from these data that a two-stage OAE/A-ABR newborn hearing screening protocol should never be used because a substantial number of infants with congenital hearing loss will be missed. First, this study was not designed to determine how many infants who pass the newborn hearing screening with OAE will have PHL at 8–12 months of age. That is an equally important question as the one addressed by this study. Second, there are many programs in which even a completely accurate OAE newborn hearing screening protocol (if in fact one existed) would miss more infants with congenital hearing loss than those who passed the A-ABR in this study. To illustrate, assume that an OAE newborn hearing screening protocol is implemented with 86,000 babies, and assume that the referral rate at time of hospital discharge is 7%. Further, assume very optimistically that all of these infants return for an outpatient rescreen at 1–3 weeks of age (such rescreens are common in OAE-based programs) and that 10% of these infants fail and are referred for a diagnostic evaluation. If the prevalence of hearing loss is 2.4 per 1,000 (as was the case for the cohort of infants evaluated in this study), 206 infants with PHL would be present in the group of 602 infants who are referred for a diagnostic evaluation from the rescreen. However, many newborn hearing screening programs, particularly those located in inner city areas, have a very difficult time getting parents to return

for diagnostic evaluations. In such situations, it is not unusual for 50% of the infants who failed the rescreen to not return for a diagnostic evaluation. If 50% did not return, at least half the infants with PHL, or 103 infants, would be "missed." This is many more infants with PHL than would pass the A-ABR in the type of two-stage newborn hearing screening protocol used in this study.

One of the primary advantages of using a two-stage OAE/A-ABR protocol is a reduction in the number of infants who need to be followed for further screening or diagnostic testing. Thus, in situations where loss to follow-up is likely to be high, a two-stage OAE/A-ABR newborn hearing screening protocol will miss fewer infants than an OAE-based protocol that requires many more infants to return for follow-up. Such issues must be considered before deciding which protocol is most sensible for a particular hospital.

The fact that a significant number of infants who pass an A-ABR newborn hearing screening test will be diagnosed with PHL at 8–12 months of age has a number of other important implications, such as the following:

- Administrators of newborn hearing screening programs should carefully evaluate what screening protocol and equipment is best for their situation and objectives. In particular, those responsible for the implementation of public health newborn hearing screening programs should be explicit about whether they want to detect mild hearing loss. In making such decisions, it is important to remember that there is nothing about A-ABR hearing screening equipment that requires a 35-dB nHL click stimulus to be used (as was the case in this study). If a different intensity stimulus had been used (e.g., 25 dB nHL), the results would almost certainly have been quite different.
- Parents and health care providers need to be reminded that passing a newborn hearing screening test does not guarantee that an infant does not have, or will not acquire, PHL. Thus, as noted sometime ago by Mason, Davis, Wood, and Farnsworth (1998, p. 91), "Passing a neonatal screening test, therefore, does not exclude the possibility of subsequent [PHL] and highlights the need for further surveillance" (see also Lutman, Davis, Fortnum, & Wood, 1997).
- In addition to hospital-based newborn hearing screening, public health programs should consider the pros and cons of doing systematic hearing screening during the early childhood years. Such screening may be a useful tool for detecting late onset PHL, as well as PHL that is missed during newborn hearing screening in day care, preschool programs, or well child visits in physician offices (Foust, Winston, Eiserman, Buhrmann, & Shisler, 2005; Winston, Eiserman, & Shisler, 2005).

These, and other implications of this study, are discussed in more detail by Gravel et al. (2005).

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