Three Cases of Recovery from Sensorineural Hearing Loss in the First Year of Life: Implications for Monitoring and Management

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J Am Acad Audiol 2021;32:54–68.

Abstract

Background Three infants with different risk factors, behavioral and physiologic audiometric histories, and diagnoses were fit with amplification between 3 and 8 months of age. Two of the three met criteria for cochlear implantation.

Purpose This article aims to heighten awareness of the rare possibility of recovery from sensorineural hearing loss in infants with varying histories and emphasize the importance of a full diagnostic test battery in all infants diagnosed with sensorineural hearing loss every 3 months until objective and subjective thresholds are stable to ensure appropriate intervention.

Research Design Case reports.

Results All three infants demonstrated improvement or full recovery of hearing and cochlear function by approximately 12 months old. Their change in hearing was discovered due to frequent follow-up and/or caregiver report. One of these infants was tentatively scheduled to have cochlear implant surgery 2 months later.

Conclusion Appropriate early intervention for infants with hearing loss is critical to ensure maximum accessibility to speech and language cues. The Federal Drug Administration approves cochlear implantation in infants as young as 12 months. When providing audiometric management of infants with sensorineural hearing loss, it is imperative to conduct a full diagnostic test battery every 3 months (including tympanometric, acoustic reflex, and otoacoustic emission measurement) until objective and subjective thresholds are stable. There was no apparent pattern of factors to predict that the infants highlighted in these cases would recover. Discussion among pediatric audiologists and otologists and comparison of data from clinics across the U.S. is needed to identify predictive patterns and determine appropriate, consistent monitoring of infants with sensorineural hearing loss.

Keywords
- hearing loss
- infants
- early intervention
- assessment
- development

Portions of these data were submitted for presentation at the annual meeting of the American Academy of Audiology, April, 2016.

The impact of sensorineural hearing loss (SNHL) on infant and early childhood development is well documented.[1–7] Early identification, management, and habilitation of childhood hearing loss reduce the associated economic, social,

received
November 26, 2019
accepted after revision
June 23, 2020
published online
February 15, 2021

Thieme Medical Publishers, Inc., 333 Seventh Avenue, 18th Floor, New York, NY 10001, USA

DOI https://doi.org/10.1055/s-0040-1719129.
ISSN 1050-0545.

and educational challenges.[6,8–13] In the most recent position statement from the Joint Committee on Infant Hearing (JCIH),[14] standards for early detection and management are published, including recommendations that screening for hearing loss occur prior to 1 month of age, confirmation of hearing loss via diagnostic testing by 3 months of age, hearing aid fitting within 1 month of diagnosis, and intervention by 6 months of age. Further, the JCIH recommends at least one auditory brainstem response (ABR) evaluation as part of a complete diagnostic test battery for children under 3 years of age for confirmation of permanent hearing loss. With greater than 95% of newborns screened in the U.S., a growing number of infants are diagnosed with hearing loss.

The increased number of newborns receiving objective hearing tests in the U.S. also has illuminated cases of potential recovery from SNHL during infancy. Some reports of improved SNHL are more likely due to misdiagnosis or inaccurate initial results intrinsic to the use of objective tests to estimate behavioral hearing thresholds. However, relatively recent studies report that somewhere between 21 and 64% of high-risk infants demonstrate partial or complete recovery on follow-up testing during their first year.[15–18] Findings have led some to recommend early habilitation decisions be made with caution until reliability and stability of SNHL can be confirmed through objective and/or subjective reexamination.[18] However, most health professionals widely recognize SNHL to be due to irreversible damage to the auditory system. This recognition in combination with the documented benefits of early intervention on development and JCIH recommendations have resulted in most audiologists making a concentrated effort to amplify infants promptly. Currently, cochlear implants are approved by the Food and Drug Administration for children as young as 12 months of age (►**Table 1**).

Evidence that some number of high-risk newborns initially diagnosed with SNHL may recover at least some of their auditory function within their first year has not appeared to have much influence on clinical practice. This may be due to several factors including a lack of knowledge that SNHL may be reversible, the absence of a significant predictor for which infants may recover, and/or reluctance to withhold or reduce effective, evidence-based habilitation strategies in the critical first year. The purpose of the current article is not to delay or modify early intervention protocols. Instead, it is to raise awareness among audiologists of the potential for recovery of infant SNHL by describing, in detail, case studies of three infants with different risk factors, behavioral and physiologic audiometric histories, and diagnoses who were treated at the University of Tennessee audiology clinic in recent years. Results of these case studies, along with previous findings,[15–18] emphasize the need for more frequent and thorough monitoring of hearing loss during the first 12 months of life than is currently recommended by U.S. guidelines in the event that hearing improves or recovers. Changes in hearing (whether an improvement or decline) must be monitored closely in our youngest patients so that intervention decisions are appropriate. Further, sharing these case studies may encourage discussion among pediatric audiologists and otologists and comparison of data from clinics across the U.S. to identify predictive patterns for recovery versus persistence of SNHL in infants.

Table 1 U.S. cochlear implant candidacy guidelines

Patient age	Candidacy guidelines
12 to 24 mo	Profound SNHL
	Limited benefit from binaural amplification
2 to 17 y	Severe to profound SNHL
	Limited benefit from binaural amplification
	≤30% Multisyllabic Lexical Neighborhood Test (MLNT) or Lexical Neighborhood Test (LNT)

Abbreviation: SNHL, sensorineural hearing loss.

Methods

Context of Service Delivery

Since 2008, Tennessee has mandated newborn hearing screening on all babies before discharge from the hospital or no later than 1 month of age. Per Tennessee's Early Hearing Detection and Intervention (EHDI) Program, 98.8% of babies received newborn hearing screens in 2017 (2017 CDC EHDI Hearing Screening & Follow-up Survey).[19] The UT Audiology Clinic is often the referral source of choice for a large number of pediatricians in the East Tennessee region for infants who (1) missed their newborn hearing screen or (2) need a rescreen after failing their hospital hearing screen. There are other pediatric audiology providers in Knoxville and the surrounding areas that see infants for hearing follow-up. In addition, there are several birthing hospitals that have either hospital-based audiology programs or in-house otolaryngology offices where infants may also go for their immediate hearing follow-up needs. The UT Audiology Clinic often becomes the site of amplification management, audiological and vestibular maintenance, and aural/oral habilitation for these infants as they develop. The UT Audiology Clinic provides services to approximately 250 infants per year and the UT Aural Habilitation Clinic works with 90 children per week who are deaf or hard of hearing.

Two of the infants described in this article (Case 1) and (Case 2.) were referred to the UT Audiology Clinic by an otolaryngologist at the East Tennessee Children's Hospital after sedated ABRs were conducted. Subsequently, all audiological management and follow-up testing was performed in the UT Audiology Clinic. One infant (Case 3) failed her newborn hearing screening in Georgia and received immediate follow-up services by a private practice in Georgia and the University of South Carolina School of Medicine before being referred to the UT Audiology Clinic for all remaining audiological testing and management.

Procedure for Chart Review

The cases presented in this article were selected because all three infants, after being referred to the UT Audiology Clinic

Table 2 Summary of major milestones in three cases of recovery from SNHL in first year of life patients

	Patients		
Milestones	**Case 1**	**Case 2**	**Case 3**
Birth history	24 wk gestation 1 lb., 7.3 oz NICU 127 d	28 wk gestation 2 lb., 3 oz. NICU 60 d	36 wk gestation 6 lb., 6.4 oz. NICU 14 d
Newborn screen	Failed A-ABR ABR: RE WNL LE Profound	Failed A-ABR	Failed A-ABR OAE/ABR: ANSD
Ophthalmology	WNL	WNL	Not available
Otolaryngology	6 mo ABR: Profound AU CI surgery scheduled	2.5 mo OAE: absent AU, ABR: absent AU	2 mo MRI: Chiari malformation
Early intervention	6 mo: IFSP for PT, OT, and SLP	3 mo: IFSP	4 mo: IFSP for PT, OT, and SLP
Genetic testing	7 mo: Negative	Not available	4 mo: Negative
Speech	13 mo: < age level	5 mo: mild language	Did not keep any
	TX 1 ×/wk	Delay; TX 1 ×/wk	Appointments
Audiology F/U 1	6–8 mo: OAE absent ABR: mod sev-sev AU HAs AU	2.5–3 mo: ABR RE mod-sev; LE: sev HAs AU	3 mo: HAs AU
Audiology F/U 2	11 mo: mom reports better hearing; OAEs: better than expected SAT: 45 dB HL in SF ABR: 5–25 dB better Discontinued HAs, Postponed CI surgery	12 mo: OAEs WNL 2–8 kHz, AU Behav: WNL RE, Mild SNHL LE Reset HAs	9 mo: Mom reports refusal to wear HAs Felt she could hear better without HAs
Audiology F/U 3	12–13 mo: OAEs: better Behav: mild range, AU	13 mo: AR WNL 500 and 1 kHz; OAEs: WNL 2–8 kHz RE; LE CNT Discontinued HAs, temporarily	9 mo: AR: WNL 1 and 2 kHz; Behav: 20 dB in SF; Discontinued HAs, temporarily
Audiology F/U 4	18 mo: OAEs WNL 2–8 kHz RE; 4–8 kHz LE; Discontinued HAs	Behav: WNL, AU Discontinued HAs	9 mo: AR: OAEs WNL 1–8 kHz, bilaterally ABR to 20 dB HL, AU Discontinued HAs

Abbreviations: A-ABR, automated ABR; ANSD, auditory neuropathy synchrony disorder; AR, acoustic reflexes; ABR, auditory brainstem response; AU, both ears; Behav, behavioral audiometric testing; CI, cochlear implant; HAs, hearing aids; IFSP, Individualized Family Service Plan; lb, Pounds; LE, left ear; mod, Moderately; NICU, neonatal intensive care unit; OAE, otoacoustic emission; OT, occupational therapy; oz, ounces; PT, physical therapy; RE, right ear; SAT, speech awareness threshold; sev, severe; SLP, speech-language therapy; SNHL, sensorineural hearing loss; TX, therapy; WNL, within normal limits.

with physiological responses consistent with severe-profound SNHL, experienced improvement or recovery of hearing function within their first year of life. These three infants were seen by UT audiologists over a span of 8 years. Records were independently reviewed by two of the authors, with the third author reviewing records for two of the three patients. When information in the patients' chart was unclear, the individuals who originally provided the records were contacted for clarification. When information was not reported or could not be corroborated, those data were not included in the article. Therefore, all material presented is valid and has been verified by multiple readers. The order of presentation of the material was aligned as similarly as possible for the three infants and major findings for each patient may be found in ►**Table 2** for ease of comparison. Details are provided in the subsequent sections.

Case Study One

Birth History

A female infant was born at 24 weeks' gestation with a birth weight of 660 g and spent 127 days in the neonatal intensive care unit (NICU). She required ventilator assistance for 2 months and oxygen assistance for 6 weeks. She had hyperbilirubinemia and required four courses of phototherapy. Her peak serum bilirubin level was 10.6 mg/dL at 10 days old. The

pregnancy was complicated by advanced maternal age, depression, anxiety, daily tobacco smoke, and use of marijuana. She was discharged from the hospital at 4 months chronological age.

Initial Hearing Screening (4 months old; prior to discharge):

Prior to discharge, the patient failed a physiologic screening of her auditory pathway via an automated ABR (Natus ALGO), bilaterally. Subsequently, a diagnostic ABR was completed before she left the hospital and results were reported as "within normal limits in the right ear" and "suggesting profound hearing loss" due to the inability to obtain repeatable waveforms in the left ear. (No further information about this diagnostic ABR is available.)

Initial Referrals/Results

- Ophthalmology (4 months old): No ocular abnormality was documented in either eye.
- Otolaryngology (Initial exam at 6 months old; follow-up exam at 7 months old): ABR testing indicated absent responses bilaterally and the patient subsequently was scheduled for cochlear implant surgery.
- Genetic testing (7 months old): Results were negative for the connexin, Pendred, cytomegalovirus, and mitochondrial panels.
- Early intervention services (6 months old): An Individualized Family Service Plan (IFSP) was developed and assistance was provided to help the family coordinate services. The patient also received physical and occupational therapy.
- Speech pathology (13 months old): The family did not keep the initial speech appointment. Thus, she was first seen at the University of Tennessee aural habilitation program for her speech and language evaluation when she was 13 months old. The following were her results:
- *Receptive-Expressive Emergent Language Test*[20]: The patient's Receptive Language Equivalent was 7 months and first percentile rank. Her Expressive Language Equivalent was 8 months and fifth percentile rank.
- *Cottage Acquisition Scales for Listening Language and Speech*[21]: Based on area, results ranged from 6- to 9-month-old age equivalents.
- *Rosetti Infant-Toddler Language Scale*[22]: Based on area, results ranged from 3- to 12-month-old age equivalents.
- *Developmental Observation Checklist System*[23]: Based on area, results ranged from first percentile to fifth percentile.
- *Little Ears Questionnaire*[24]: The patient identified 25/35 items correctly.
- *Infant-Toddler Meaningful Auditory Integration Scale*[25]: The patient scored 33/40 correct (82%).

Overall, results showed her to be below age level on comprehensive language testing. Therapy was recommended for 1 hour, one time a week to address expressive language, receptive language, and auditory skills.

Follow-Up Audiological Testing/Management

Six and 7 months old results:

- Otoscopy: Results of otoscopy were not reported.
- Tympanometry: Tympanograms were type A, bilaterally, using a 226- and 1,000-Hz probe tone (GSI 39 Auto Tympanometer).
- Distortion Product Otoacoustic Emissions (DPOAEs): The patient received an overall refer for 2,000 to 6,000 Hz, bilaterally (Biologic Scout). The criterion for a passing response was a +6 dB signal-to-noise ratio (SNR).
- ABR: ABRs were consistent with moderately severe to severe SNHL from 500 to 4,000 Hz, bilaterally. Replicable responses were obtained with 500, 2,000, and 4,000 Hz toneburst stimuli presented at a rate of 27.7/second, with adequate levels of contralateral masking (►**Fig. 1**). Replicable responses were obtained to click air conduction stimuli at a rate of 27.7/second down to 75 dB nHL in the right ear and 80 dB nHL in the left ear (►**Fig. 2**, top panel). No response was obtained to bone conduction click stimuli bilaterally at 40 dB nHL (►**Fig. 2**, bottom panel). A rate study was normal in the right ear, using air conduction click stimuli at 80 dB nHL with click rates of 27.7 and 57.7/second. A polarity study was normal in the right ear, with repeatable responses that did not invert to both condensation and rarefaction polarity clicks at a rate of 27.7/second and a level of 80 dB nHL. Rate and polarity studies were not conducted in the left ear (Biologic Navigator Pro ABR).
- Management: It was recommended that amplification be pursued and a hearing evaluation be completed when the patient was developmentally capable. A cochlear implant was planned for the right ear.

Audiological Management

Eight months old:

The patient was fit with binaural personal amplification. Hearing aids were set to Desired Sensation Level (DSL) targets utilizing Real Ear to Coupler Difference (RECD) measures (Audioscan Verifit Binaural test box).

Additional pertinent information (11 months old): The family noted that the infant seemed to be hearing better and was "not herself" when she was wearing the hearing aids.

Follow-up audiological testing/management (11 months old):

- Otoscopy: Otoscopy was unremarkable.
- Tympanometry: Tympanograms were normal type A, bilaterally with a 226-Hz probe tone (Grason-Stadler GSI-39 Auto Tymp).
- Acoustic reflexes: The patient's ipsilateral reflex in the right ear was absent at 1,000 and 4,000 Hz. The left ear could not be tested due to lack of patient compliance (Grason-Stadler GSI-39 Auto Tymp).
- DPOAEs: DPOAEs were present for 2,000 and 4,000 to 8,000 Hz in the right ear and 4,000 to 8,000 Hz in the left ear (Biologic AuDx Pro). Results indicated a significant change from previously absent DPOAEs. (Responses were

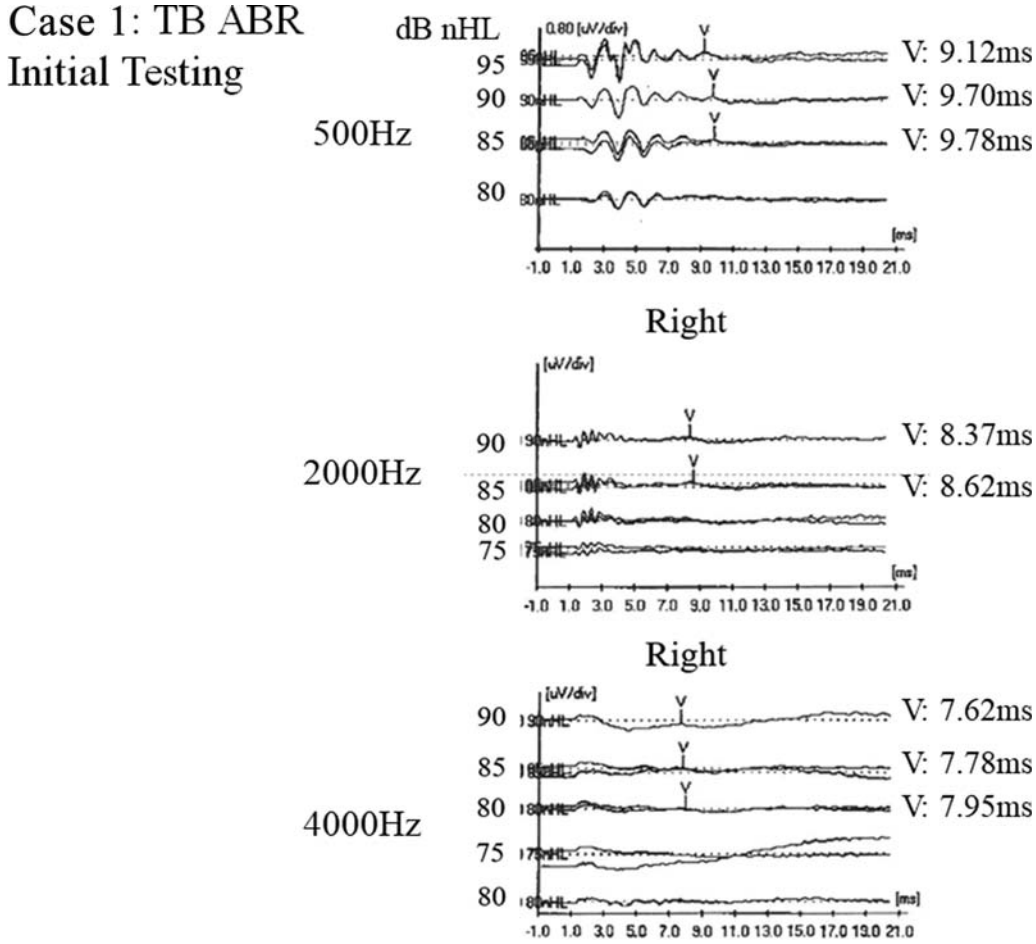

Fig. 1 Initial toneburst auditory brainstem response (ABR) waveforms for Case 1 plotted on the time domain with amplitude (μV) as a function of time (ms). Responses to 500, 2,000, and 4,000 Hz tonebursts presented to the right ear are arranged from top to bottom in the figure panels, respectively. In each panel, wave V is marked at the lowest stimulus level at which it replicated. At each stimulus level for which wave V was present, its latency (ms) is provided.

elicited utilizing 65/55 dB sound pressure level [SPL] L1/L2 probe tones with two points per octave ranging from 2,000 to 8,000 Hz. Criteria for a passing response were a distortion product [DP] ≥ -5 dB, DP-noise floor [NF] ≥ 8 dB, and repeatability between responses of 3 dB.)

- Behavioral testing: Behavioral results were minimal due to lack of patient compliance. However, a speech awareness threshold of 45 dB hearing level (HL) in the left ear was obtained (Grason-Stadler Audiostar Pro).
- ABR: A follow-up ABR was completed to rule out auditory neuropathy and establish thresholds. Replicable results indicated a mild to moderately severe hearing loss in the right ear and a moderately severe hearing loss in the left ear with 500, 1,000, 2,000, and 4,000 Hz rarefaction tonebursts at a rate of 37.7/second (►**Fig. 3**). A polarity study was normal bilaterally, with repeatable responses that did not invert to both condensation and rarefaction polarity clicks at a rate of 11.1/second and a level of 90 dB nHL. All ABRs were recorded using a two-channel vertical montage (Vivosonic Integrity V500 7.1.1).

Overall testing showed an improvement of 5 to 25 dB in ABR thresholds and significant improvement in DPOAEs compared with those obtained at birth. Amplification was temporarily discontinued until further testing could be completed to show stability of these results. Cochlear implantation was postponed.

Follow-Up Audiological Testing/Management

Twelve to 13 months old:

- Otoscopy: Otoscopy demonstrated nonoccluding cerumen.
- Tympanometry: Tympanograms were normal, type A with a 226-Hz probe tone (Grason-Stadler GSI-39 Auto Tymp).
- DPOAEs: DPOAEs were present from 2,000 to 8,000 Hz in the right ear and from 4,000 to 8,000 Hz in the left ear (Biologic AuDx Pro). Responses were elicited utilizing 65/55 dB SPL L1/L2 probe tones with two points per octave ranging from 2,000 to 8,000 Hz. Criteria for a passing response were a DP ≥ -5 dB, DP-NF ≥ 8 dB, and repeatability between responses of 3 dB.
- Behavioral testing: Behavioral thresholds were in the mild range of hearing, bilaterally. Speech awareness thresholds were 45 dB HL, bilaterally (Grason-Stadler Audiostar Pro).
- Hearing aids: Hearing aids were set to DSL targets utilizing RECD measures with slightly conservative thresholds

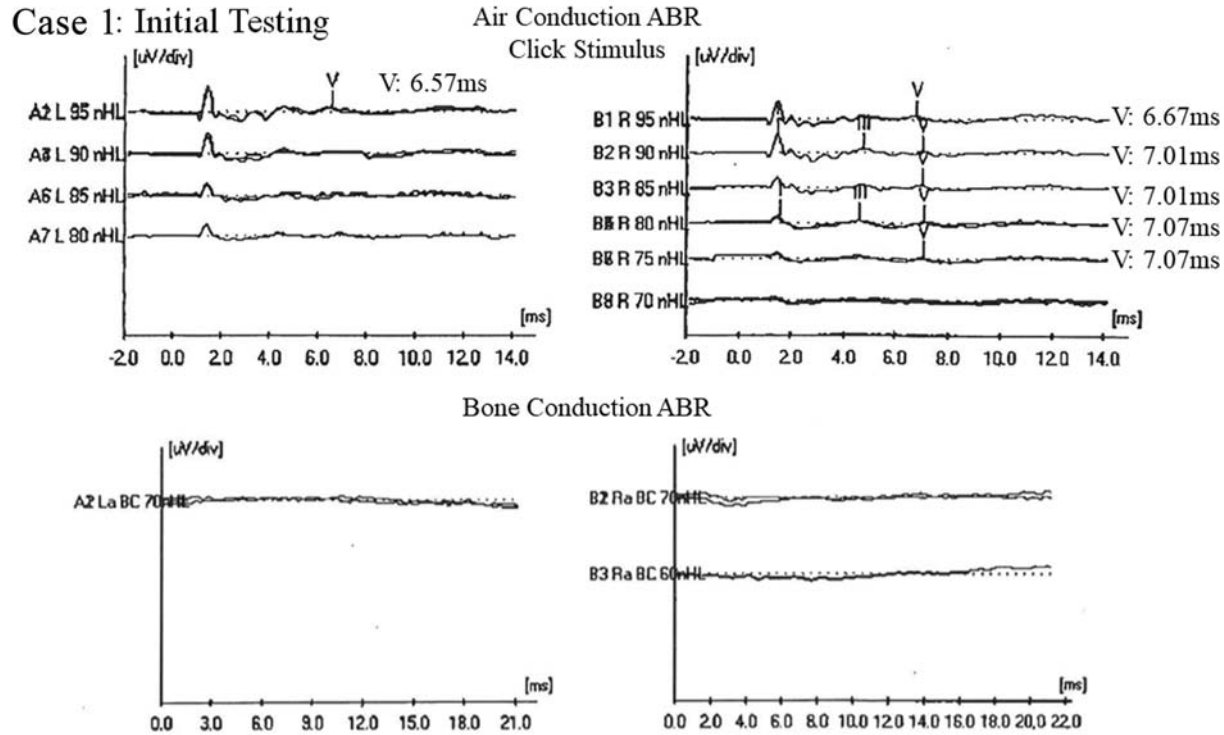

Fig. 2 Initial click auditory brainstem response (ABR) waveforms for Case 1 plotted on the time domain with amplitude (μV) as a function of time (ms). Air-conducted click ABRs are presented in the top row for the left and right ears, respectively. Absent bone-conducted click ABRs are displayed in the bottom row. In each panel, wave V is marked at the lowest stimulus level at which it replicated. At each stimulus level for which wave V was present, its latency (ms) is provided.

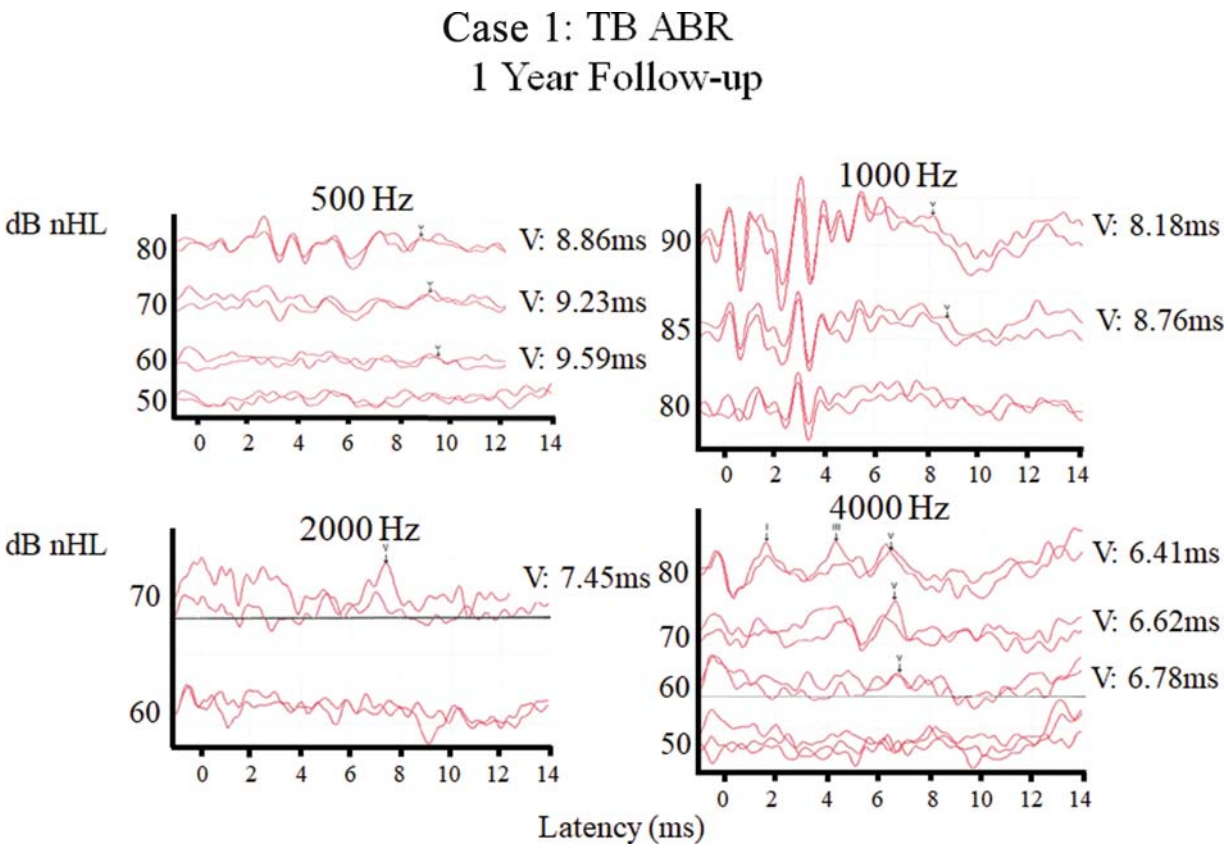

Fig. 3 Follow-up auditory brainstem response (ABR) waveforms for Case 1 at age 12 months plotted on the time domain with amplitude (μV) as a function of time (ms). Responses to 500, 1,000, 2,000, and 4,000 Hz tonebursts presented to the right ear are displayed. In each panel, wave V is marked at the lowest stimulus level at which it replicated. At each stimulus level for which wave V was present, its latency (ms) is provided.

(35 dB), in the event that her thresholds continued to improve (Audioscan Verifit Binaural test box).

Follow-Up Audiological Testing/Management

Eighteen months old:

- Otoscopy: Otoscopy was unremarkable.
- Tympanometry: Tympanograms were normal, type A with a 226-Hz probe tone (Grason-Stadler GSI-39 Auto Tymp).
- DPOAEs: DPOAEs were present from 2,000 to 8,000 Hz in the right ear for half replications and from 4,000 to 8,000 Hz in the left ear (Biologic AuDx Pro). Responses were elicited utilizing 65/55 dB SPL L1/L2 probe tones with two points per octave ranging from 2,000 to 8,000 Hz. Criteria for a passing response was a DP $\geq$ –5 dB, DP-NF $\geq$ 8 dB, and repeatability between responses of 3 dB.
- Behavioral testing: Behavioral thresholds were normal, bilaterally (Grason-Stadler Audiostar Pro).
- Hearing aids: Hearing aids were discontinued.

Follow-Up Audiological Testing/Management

Twenty-four – 25 months old:

- Otoscopy: Otoscopy was unremarkable.
- Tympanometry: Tympanograms were normal, type A with a 226-Hz probe tone (Grason-Stadler GSI-39 Auto Tymp).
- DPOAEs: The patient would not accept insert earphones.
- Behavioral testing: The patient would not accept insert earphones at 24 months old. Sound field testing for tones and speech was normal. At 25 months old, ear-specific speech awareness thresholds were obtained at 15 dB bilaterally and sound field testing for tones was normal (Grason-Stadler Audiostar Pro).

Case Study 2

Birth History

A female infant was born at 28 weeks gestation with a birth weight of 1,050 g and spent a prolonged period in the NICU. She required ventilator assistance, phototherapy for hyperbilirubinemia, and two rounds of gentamycin antibiotics. The pregnancy was complicated by advanced maternal age, daily tobacco smoke, marijuana and cocaine use, and no prenatal care.

Initial hearing screening (during NICU stay):

The patient failed a physiologic screening of her auditory pathway via an automated ABR (Natus ALGO), bilaterally. She was referred to otolaryngology upon discharge for follow-up testing.

Initial Referrals/Results

- Ophthalmology (during NICU stay): No ocular abnormality was reported in either eye.
- Otolaryngology (~2.5 months; postdischarge): Tympanometry was normal, bilaterally. DPOAEs were absent at all frequencies and stimulus levels in both ears. The patient failed an ABR screen at 40 dB nHL, bilaterally. No further information was reported.
- Genetic testing: Information was not available.
- Early intervention services (3 months): An IFSP was developed and assistance provided to help the family coordinate services.
- Speech pathology (5 months): The patient underwent a speech and language evaluation at the University of Tennessee aural habilitation clinic. She was identified with a mild language delay. To ensure that her speech, language and auditory skills continued to develop, therapy was recommended for 1 hour, one time a week.

Follow-Up Audiological Testing/Management

Approximately 2.5 months old:

- Otoscopy: Otoscopy was unremarkable.
- Tympanometry: Tympanograms were normal.
- ABR: Toneburst (2,000 Hz) and click-evoked ABRs (►**Fig. 4**) estimated a moderate to severe hearing loss in the right ear and a severe hearing loss in the left ear. For each ear and stimulus type, rarefaction stimuli were presented at a rate of 29.1/second. Initial test levels were at 70 dB nHL. Replication of wave V was required at each level with a minimum of 2,000 accepted sweeps. Intensity levels were reduced by 10 dB until wave V no longer replicated. A single-channel vertical montage was used (Grason-Stadler Audera).
- Auditory steady state responses: Auditory steady state responses to 500, 1,000, and 2,000 Hz stimuli were consistent with ABR findings (Grason-Stadler Audera). The "Estimated Audiogram" program relying on "Default Asleep" protocol was used for testing.
- Management: Pursuit of amplification was recommended.

Audiological Management

Three months old:

The patient was fit with binaural personal amplification. Hearing aids were set to DSL targets utilizing RECD measures (Audioscan Verifit Binaural test box). She tolerated the hearing aids and wore them during all waking hours for 8 months. The family felt she was doing well, but was exploring a cochlear implant.

Follow-Up Audiological Testing/Management

Twelve months old:

- Otoscopy: Otoscopy was unremarkable in the right ear and demonstrated moderate amounts of cerumen in the left ear.
- Tympanometry: Tympanograms were normal type A in the right ear and type B in the left ear with a 226-Hz probe tone (Grason-Stadler GSI-39 Auto Tymp).
- DPOAEs: DPOAEs were present from 2,000 to 8,000 Hz in the right ear. Responses were elicited utilizing a 65/55 dB SPL L1/L2 probe tones with two points per octave, ranging from 2,000 to 8,000 Hz. Criteria for a passing response were a DP $\geq$ –5 dB, DP-NF $\geq$ 8 dB, and repeatability

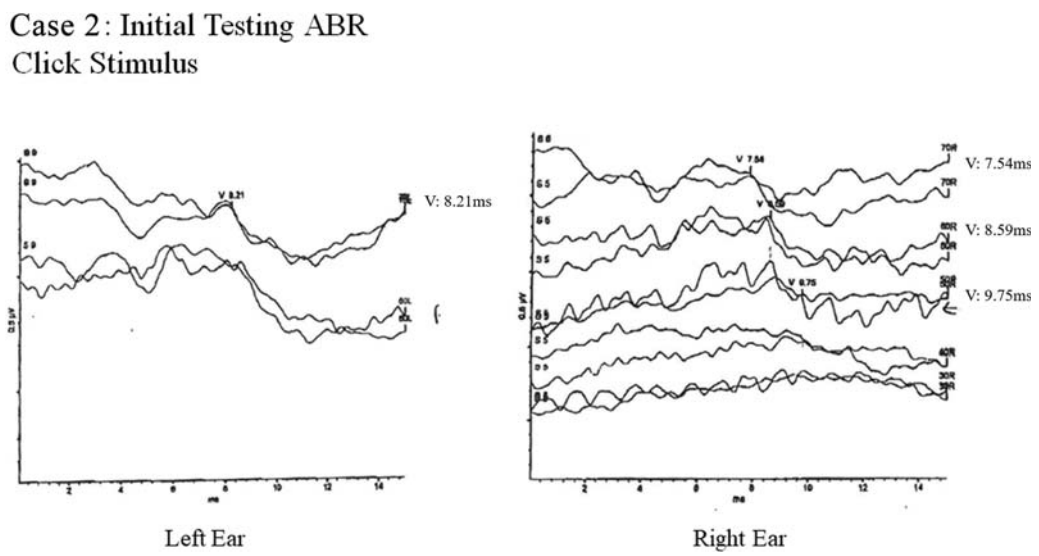

Fig. 4 Initial air-conducted click auditory brainstem response (ABR) waveforms for Case 2 plotted on the time domain with amplitude (μV) as a function of time (ms). ABRs are plotted from top to bottom in response to 70 and 60 dB nHL clicks to the left ear (left column) and to 70, 60, 50, 40, and 30 dB nHL clicks to the right ear (right column). In each column, wave V is marked down to the lowest stimulus level at which it replicated (70 dB nHL for the left ear, 50 dB nHL for the right ear) and its latency is provided.

between responses of 3 dB (Biologic AuDx Pro). Results in the left ear were not obtained based on tympanometric results and lack of patient compliance.

- Behavioral testing: Behavioral thresholds were in the mild range of hearing in at least the better ear (sound field). Speech awareness thresholds were 30 dB HL, bilaterally (Grason-Stadler Audiostar Pro).

Overall, testing showed improvement in hearing. Based on changes in audiological results and the family reports that the patient was still accepting and wearing the hearing aids, amplification was readjusted to the new behavioral thresholds.

Follow-Up Audiological Testing/Management

Thirteen months old (two separate evaluation days):

- Otoscopy: Otoscopy was unremarkable.
- Tympanometry: Tympanograms were normal type A with a 226-Hz probe tone, bilaterally (Grason-Stadler GSI-39 Auto Tymp).
- Acoustic reflexes: Ipsilateral reflexes were obtained at normal sensation levels at 500 and 1,000 Hz, bilaterally (Grason-Stadler GSI-39 Auto Tymp).
- DPOAEs: DPOAEs were present from 2,000 to 8,000 Hz, bilaterally (►**Fig. 5**).

Responses were elicited utilizing a 65/55 dB SPL L1/L2 probe tones with two points per octave, ranging from 2,000 to 8,000 Hz. Criteria for a passing response were a DP > –5 dB, DP-NF > 8 dB, and repeatability between responses of 3 dB (Biologic AuDx Pro).

- Behavioral testing: Behavioral thresholds in the right ear were normal, with a mild hearing loss at 500 Hz rising to

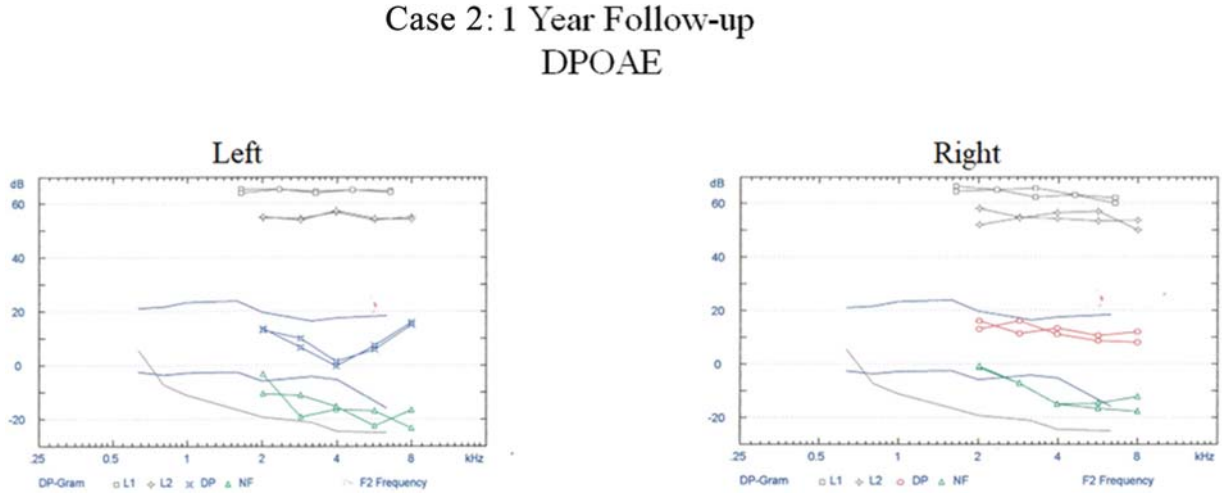

Fig. 5 Distortion Product Otoacoustic Emissions (DPOAEs) from the left and right ears of Case 2 at age 12 months. Responses are within normal limits at all frequencies tested, bilaterally. Squares = stimulus level of the L1 probe tone. Diamonds = stimulus level of the L2 probe tone. X and circles = DPOAE response for left and right ear, respectively. Triangles = the level of the noise floor.

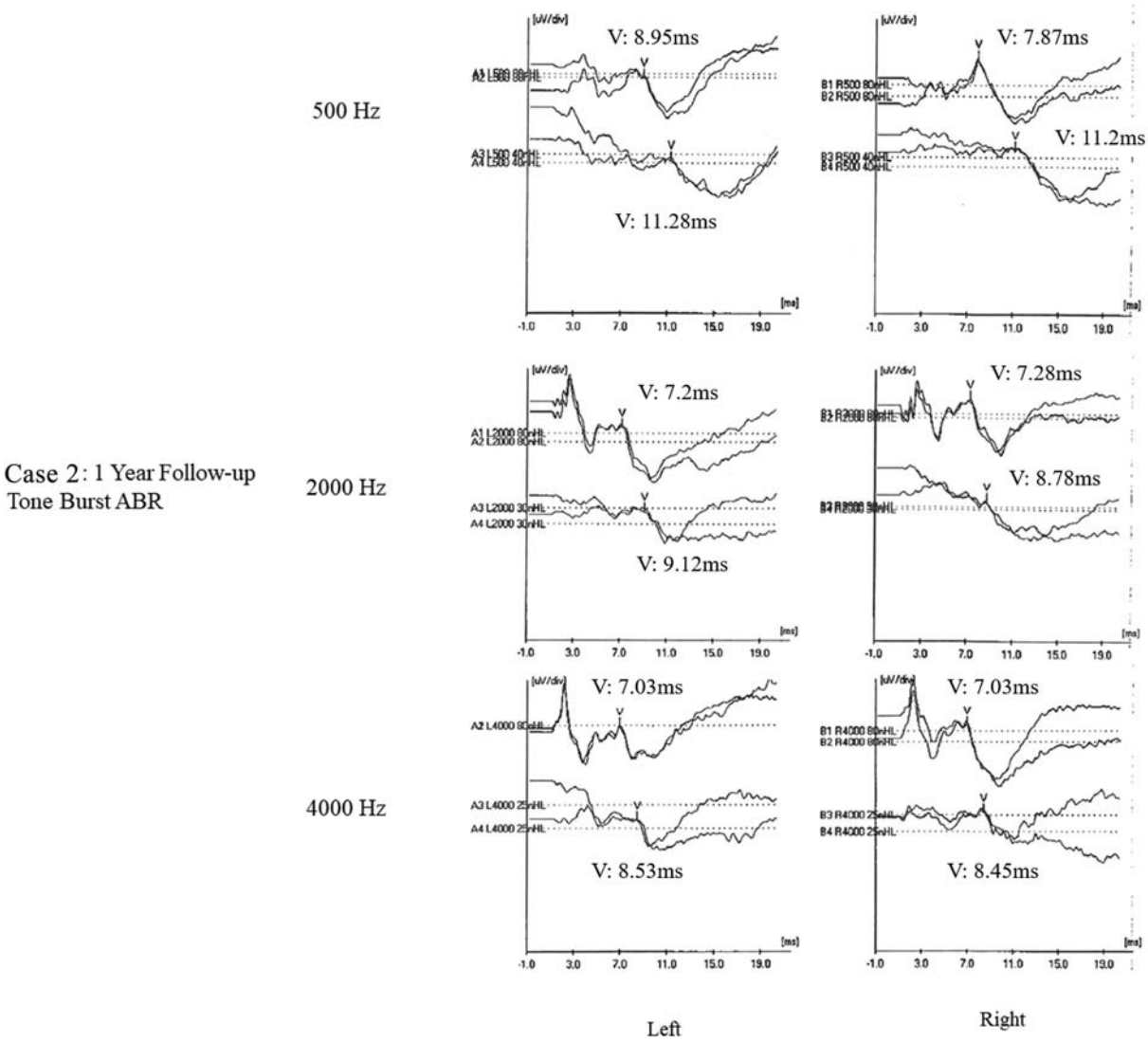

Fig. 6 Follow-up auditory brainstem response (ABR) waveforms for Case 2 at age 12 months plotted on the time domain with amplitude (μV) as a function of time (ms). ABRs are plotted from top to bottom in response to 500 Hz tone bursts at 80 and 40 dB nHL, 2,000 Hz tone bursts at 80 and 30 dB nHL, and 4,000 tonebursts at 80 and 25 dB nHL to the left and right ears. At each stimulus level, wave V was replicated and its latency (ms) is provided.

normal hearing in the left ear. A speech awareness threshold was obtained at 25 dB HL in the sound field (Grason-Stadler Audiostar Pro).

- Hearing aids: Hearing aids were discontinued pending ABR testing.

Follow-up audiological ABR testing (13 months old):

- Otoscopy: Otoscopy was unremarkable.
- Tympanometry: Tympanograms were normal type A with a 226-Hz probe tone, bilaterally (GSI 39 Auto Tympanometer).
- DPOAEs: DPOAEs were within normal limits for 2,000 to 6,000 Hz, bilaterally. Responses were elicited utilizing 65/55 dB SPL L1/L2 probe tones with two points per octave, ranging from 2,000 to 6,000 Hz. Criterion for a passing response was a + 6 dB SNR (Biologic Scout).
- ABR: Replicable waveforms were elicited to click stimuli down to 15 dB nHL, bilaterally. Responses evoked by click and toneburst stimuli at 500, 2,000, and 4,000 Hz were normal bilaterally (►**Fig. 6**). Rate and polarity studies were normal. For each ear and stimulus type, rarefaction stimuli were presented at a rate of 29.1/second and an initial stimulus level of 80 dB nHL. Replication of wave V was required with a minimum of 2,000 accepted sweeps per replication. For 500 Hz, stimulus levels were reduced to 40 dB nHL and replicated. For 2,000 Hz, they were reduced to 30 dB nHL and replicated. For 4,000 Hz and click stimuli, they were reduced to 25 dB nHL and replicated. For the rate study, 80 dB nHL rarefaction click stimuli were increased from 29.1 to 89.1/second. For the polarity study, 80 dB nHL click stimuli at a rate of 29.1/second were presented in rarefaction and

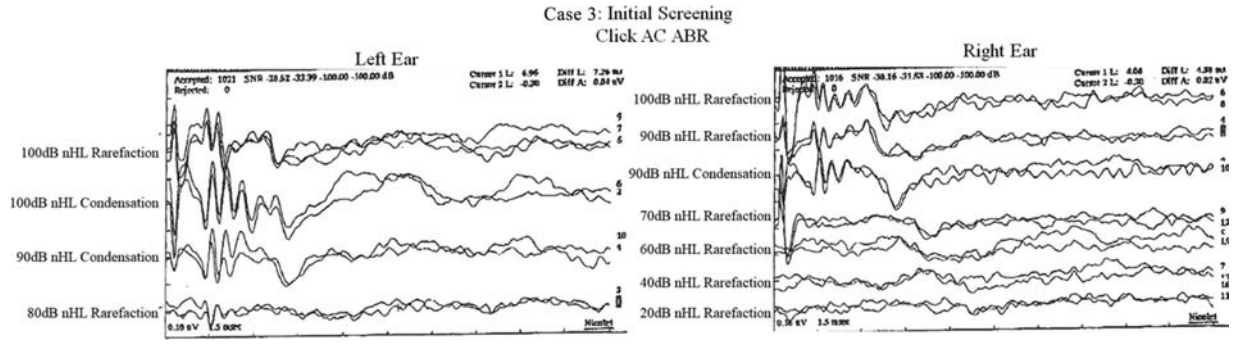

Fig. 7 Initial newborn hearing screening auditory brainstem responses (ABRs) plotted on the time domain with amplitude (μV) as a function of time (ms) in response to click stimuli presented to the left and right ears for Case 3. Reversible cochlear microphonic and absent ABRs were noted, bilaterally. The *X*-axis interval is 1.5 ms. The *Y*-axis interval is 0.16 μV.

condensation phases. A single-channel vertical montage was used for all ABRs (Biologic Navigator Pro).
- Hearing aids: Hearing aids were discontinued.

Case Study 3

Birth History

A female infant was born at 36 weeks' gestation with a birth weight of 3,105 g and spent 2 weeks in the intermediate care nursery secondary to myelomeningocele. She was born with spina bifida and severe hydrocephalus required placement of a shunt at 2 days old.

Initial hearing screening (prior to discharge):

The patient failed a physiologic screening of her auditory pathway via an automated ABR, bilaterally. Follow-up diagnostic testing (2 weeks old) showed present DPOAEs for 2,000 through 5,000 Hz, present and reversible cochlear microphonics, and absent air-conducted and bone-conducted click ABRs (at limits of the equipment), bilaterally (►**Fig. 7**). She was identified with auditory neuropathy spectrum disorder.

Initial Referrals/Results

- Ophthalmology: There is no mention of an ophthalmology exam in her records.
- Otolaryngology (initial evaluation at 2 months; follow-up imaging at 5 months): Magnetic resonance imaging results were consistent with Chiari malformation and thinning of the corpus callosum. The 8th cranial nerve appeared to be present.
- Genetic testing (4 months): Results were negative for the cytomegalovirus and mitochondrial panels.
- Early intervention services (4 months): An IFSP was developed and help coordinating services for speech, physical, and occupational therapy provided to the family.
- Speech pathology: The patient was referred for speech pathology services after her initial hearing loss diagnosis. However, the family did not keep any of her appointments.

Audiological Management

Three months old:

The patient was fit with binaural personal amplification. Hearing aids were set conservatively (based on present otoacoustic emissions [OAEs]) to DSL targets utilizing RECD measures (Audioscan Verifit Binaural test box). Pediatric Amplification Practice Guidelines from the American Academy of Audiology[26] state that children with auditory neuropathy should have a trial with amplification. However, the ABR and OAEs are not a valid measure of thresholds in these children and amplification is provided based on behavioral observation from the clinician and family. Continued observation is essential to adjust the amplification as necessary. The patient wore the hearing aid intermittently. The mother reported that she was "more focused" with amplification.

Additional pertinent information (9 months old):

The patient's mother reported that she had not been wearing the hearing aid and felt that she could hear without them.

Follow-up audiological testing/management (9 months old):

- Otoscopy: Otoscopy was unremarkable.
- Tympanometry: Tympanograms were normal type A with a 226-Hz probe tone, bilaterally (Grason-Stadler GSI-39 Auto Tymp).
- Acoustic reflexes: Ipsilateral reflexes were obtained at normal levels for 1,000 and 2,000 Hz, bilaterally (Grason-Stadler GSI-39 Auto Tymp).
- Behavioral testing: Speech awareness in the sound field was obtained at 20 dB HL (Grason-Stadler Audiostar Pro).
- Management: Based on changes in audiological results and the mother's report that the patient seemed to be hearing better, amplification was temporarily discontinued until further testing could be completed.

Follow-Up Audiological Testing/Management

Twelve months old:

- Otoscopy: Otoscopy was not reported.

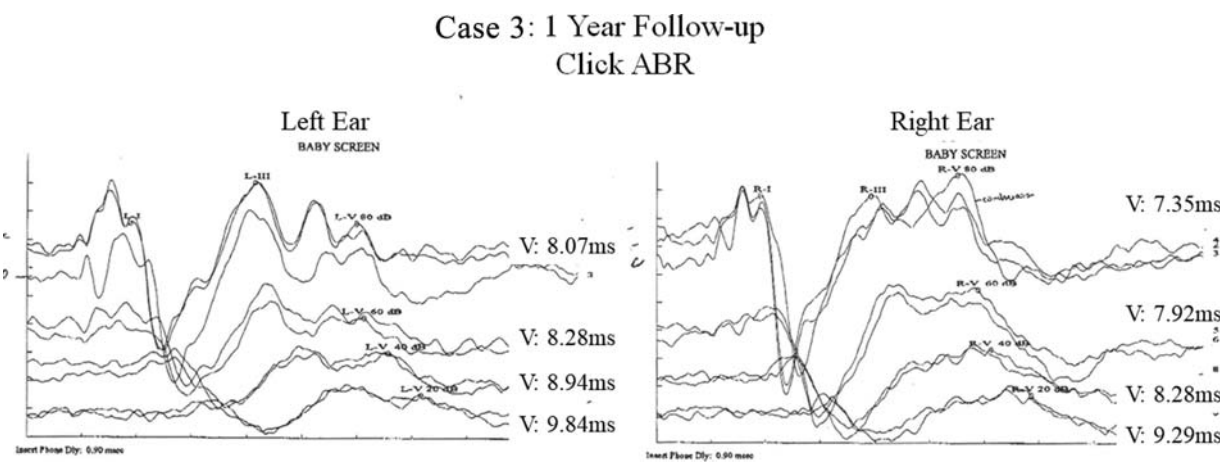

Fig. 8 Follow-up auditory brainstem responses (ABRs) for Case 3 at age 12 months plotted on the time domain with amplitude (μV) as a function of time (ms) in response to click stimuli presented at 80, 60, 40, and 20 dB nHL to the left and right ears. At each stimulus level (highest to lowest = top to bottom), wave V was replicated and its latency (ms) is provided. The *X*-axis interval is 1.5 ms. The *Y*-axis interval is 0.16 μV.

- Tympanometry: Type A tympanograms were obtained in both ears (equipment unknown).
- DPOAEs: DPOAEs were normal for 1,000 to 8,000 Hz, bilaterally (equipment unknown).
- ABR: At 12 months chronological age, clear and repeatable ABRs to click stimuli were obtained down to 20 dB nHL and a polarity study was normal with repeatable responses that did not invert in both ears (►**Fig. 8**). Responses to 500, 2,000, and 4,000 Hz toneburst stimuli were present down to 20 dB nHL in both ears (►**Fig. 9**). For each ear and stimulus type, rarefaction stimuli were presented at a rate of 29.1/second and an initial stimulus level of 50 dB nHL. Replication of wave V was required with a minimum of 2,000 accepted sweeps per replication. For the polarity study, 80 dB nHL click stimuli at a rate of 29.1/second were presented in rarefaction and condensation phases. A single-channel vertical montage was used for all ABRs (Natus Nicolet).
- Hearing aids: Hearing aids were discontinued. Ongoing audiological behavioral testing continues to be consistent with normal hearing.

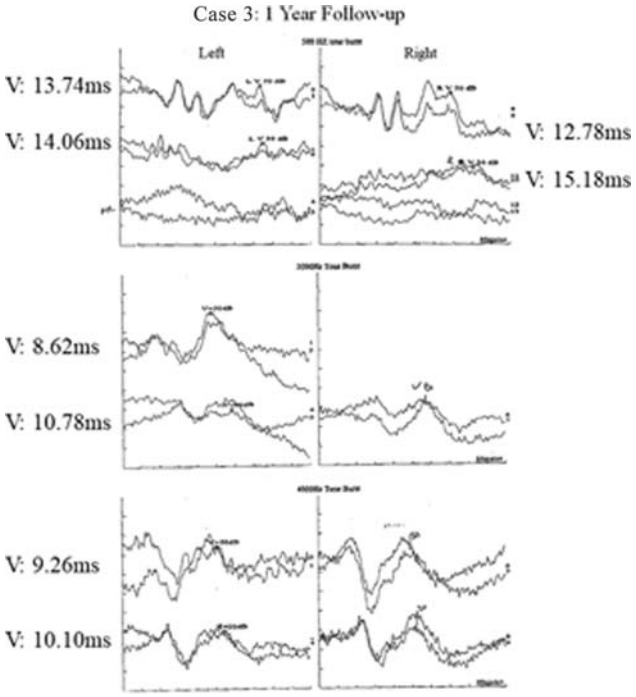

Fig. 9 Follow-up auditory brainstem responses (ABRs) for Case 3 at age 12 months plotted on the time domain with amplitude (μV) as a function of time (ms). Waveforms are displayed from top (highest stimulus level) to bottom (lowest stimulus level) to 500 Hz tonebursts at 50 and 30 dB nHL (top row), 2,000 Hz tonebursts at 50 and/or 20 dB nHL (middle row), and 4,000 Hz tonebursts at 50 and 20 dB nHL (bottom row) presented to the left (left column) and right (right column) ears. At each stimulus level for which wave V was replicable during left ear testing, its latency (ms) is provided. Wave V latencies during right ear testing were available for 500 Hz only. The *X*-axis interval is 2.0 ms. The *Y*-axis interval is 0.16 μV.

Discussion

Hearing loss is the fourth most common developmental disorder in the U.S. and the most common sensory disorder.[27] Progressive SNHL is widespread in infants diagnosed at birth, especially those with a history of anoxia and/or low birth weight.[28] Data indicating the likelihood of, or predictive factors in, recovery from newborn SNHL are sparser resulting in a general lack of awareness that some SNHL may not be permanent.

Three infants with a different set of risk factors who failed newborn hearing screenings and presented with physiological responses consistent with SNHL during follow-up diagnostic testing are detailed in the current paper. Each infant has a different hearing loss configuration, one was diagnosed with auditory neuropathy spectrum disorder, and all were fit with amplification between three and eight months of age. Two of the three were approved for cochlear implantation using U.S. candidacy guidelines.

Other than verifiable SNHL at birth, the similarity in these infants is significant improvement in hearing prior to their first birthdays. There is no apparent pattern of factors to predict that these infants would recover hearing. Other studies with larger numbers of infants have highlighted similar issues. For example, Psarommatis et al[18] report

significant improvement in ABRs in approximately 42% of high-risk infants diagnosed at birth with SNHL. Other studies demonstrate between 21[15] and 64%[16] of high risk infants present with reversible ABR abnormalities in their first year. None of these studies could determine an underlying etiology or predictor (e.g., asphyxia, jaundice, infection) of recovery from SNHL. In fact, present OAEs in newborns with abnormal ABRs provided the best (and only) significant predictor of ABR improvement post-initial screening.[18] This profile, typical of auditory neuropathy, is similar to Case 3 in the current article but does not explain the improvements documented in the other two infants. Taken together, clinical reports enhance the need for additional infant data sets to determine the trajectory and predictive factors, if any, associated with reversible SNHL. Further, they necessitate review of current U.S. guidelines for monitoring SNHL in infants.

Reversible Hearing Loss or Late Onset of Hearing

It is possible the case studies herein may reflect late onset of hearing, rather than reversible SNHL. There is documented, significant improvement in behavioral auditory thresholds in quiet for high-frequency tones in typical infants between birth and early school age when examining both longitudinal, individual,[29] and cross-sectional group[30,31] data. Results suggest the most rapid auditory behavioral development occurs from birth to approximately 6 months of age.[29]

Because behavioral data are absent or limited for most infants less than 6 months of age, it is probably more relevant to consider whether the patterns of recovery in ABRs and OAEs reported in these case studies (and others) are associated with the developmental trajectories of ABRs and OAEs. In the case of ABR, the most significant and rapid changes (predominately in latency and threshold values) occur from birth to approximately 3 months of age. Up to approximately 6 months old, ABR thresholds are better than behavioral thresholds.[32,33] Additionally, less remarkable changes occur to ABRs until at least 18 months old,[34,35] with recent data suggesting continuing development of the speech-evoked ABR beyond adolescence.[36,37] OAEs reflect cochlear mechanics and, since the inner ear is mature at birth, OAEs are believed to be well developed and mostly adult-like in newborns.[38] Further, many infants in the same age range pass ABR/OAE testing with normal estimated thresholds. Based on these data, it seems safe to assume that the infants reported herein (and most infants reported with SNHL based on ABR/OAE data) do in fact have SNHL at the time of ABR/OAE testing.

Whether the patients highlighted in the current article are demonstrating recovery from SNHL or late onset of hearing may not matter when considering how to manage the patient and ensure maximum accessibility to speech and language cues. Changes in hearing (whether an improvement or decline) must be monitored closely in our youngest patients so that intervention decisions are appropriate. Caregivers need to be educated to signs of deviations in hearing. Pediatric audiologists should follow a consistent, comprehensive test battery for monitoring and data should be shared to a centralized depository in an attempt to identify predictive factors in maternal/birth history and/or audiometric data to help determine whose hearing might improve, whose might remain stable, and whose might decline. Based on the cases presented herein and data from others,[15–18] it seems that current U.S. guidelines on hearing in infants less than 12 months of age may warrant review.

Current Guidelines for Monitoring Hearing in Infants Less Than 12 Months of Age

The 2007 JCIH has separate hearing screening protocols for well-baby nurseries and NICUs. It is recommended that well-baby infants are screened once and, if needed, rescreened prior to discharge. Protocols state that OAE or automatic ABR is appropriate. For babies admitted to the NICU for greater than 5 days, it is recommended that they have a screening ABR. If they fail the screening ABR, they should be referred to an audiologist with skills and expertise in evaluating newborn and young infants for a diagnostic ABR on each ear. Infants who pass the hearing screening but have a risk factor (►**Table 3**) should have at least one diagnostic audiology assessment by 24 to 30 months of age. However, the timing and number of hearing reevaluations should be patient-specific depending on their personal history and level of risk factors. A repeat hearing screening is recommended prior to discharge for hospital readmissions during the first month of life when there are conditions associated with the development of hearing loss. JCIH recommends fitting amplification within 1 month of diagnosis and states intervention services should begin as soon as possible but no later than 6 months old.

Given current and previous documentation of recovery from SNHL at or around 12 months of age, the recommendations by the JCIH outlined above may need to be supplemented, addressing procedures to be followed after intervention services have begun no later than 6 months old.[18] To assess hearing and appropriateness of intervention strategies during infancy, we would argue it is critical to conduct a full diagnostic test battery (procedures standardized across pediatric audiologists) in all infants diagnosed with SNHL, including tympanometry, reflexes, OAEs, and behavioral testing every 3 months until at least 18 months of age. Tympanometry should be conducted with the appropriate probe tone frequency given an infant's age to better monitor for conductive components that may overlay SNHL. Given the finding that normal OAEs were an important predictor of ABR recovery in a large group of high risk infants,[18] these measures also seem crucial. For those infants for whom cochlear implantation is recommended, it may be critical to conduct this full diagnostic test battery every 3 months until reliable and stable hearing thresholds are obtained both objectively and subjectively before proceeding with implantation.[18] Standardization of procedures and systematic data collection would allow pediatric clinics around the country to share their data, enhancing the possibility of discovering predictive patterns in case history or audiometric data that may identify infants early in the intervention process who are likely to experience

Table 3 JCIH high risk factors for SNHL

Caregiver concern
Family history of permanent childhood hearing loss
Extracorporeal membrane oxygenation (ECMO) use
In utero infections such as CMV, herpes, rubella, syphilis, toxoplasmosis
Syndromes associated with progressive hearing loss
Neurodegenerative disorders
Postnatal infections associated with SNHL
Head trauma
Chemotherapy
NICU < 5 days
Assisted ventilation
Ototoxic medication or loop diuretics
Hyperbilirubinemia with transfusion
Physical findings associated with syndromes with hearing loss
Low birth weight

Abbreviations: CMV, cytomegalovirus; JCIH, Joint Committee on Infant Hearing; NICU, neonatal intensive care unit; SNHL, sensorineural hearing loss.

improvement in hearing. This would ensure intervention strategies remain appropriate for the infant during this critical time for speech and language development.

Conclusion

To better identify infant patients whose hearing (conductive and/or sensorineural) improves during their first year of life, supplemental guidelines for standardized, comprehensive, and frequent audiologic testing may be warranted. Comprehensive, frequent monitoring (including regular tympanometric, acoustic reflex, and OAE measurement) will ensure that hearing aid, cochlear implantation, and therapy recommendations are appropriate over time. A large-scale study of infants diagnosed with SNHL at birth is warranted to determine if common factors arise that could predict likelihood of improved hearing in some infants. This would require a centralized mechanism for pediatric audiologists to report maternal/birth history and audiologic responses of infants diagnosed with SNHL during the first year to 18 months of age. It is critical to educate health care providers and families of infants diagnosed early with hearing loss to be aware of any perceived changes in their child's hearing and to share those perceptions of improvement or decline with the team overseeing their hearing health care.

Conflict of Interest
None declared.

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