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South Dakota Parents' Knowledge of Congenital Cytomegalovirus, Its Long-Term Health Effects, and Methods for Minimizing Exposure

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Abstract

Congenital CMV (cCMV) is acknowledged as one of the most common causes of nonhereditary sensorineural hearing loss and an important cause of neurodevelopmental delay in children. Despite the danger cCMV poses, many parents are unaware of the virus, its sequelae, mode of transmission, and preventative behaviors. The purpose of the study was to determine South Dakota parents' knowledge of cCMV, its sequelae, and ways to minimize exposure.

An electronic survey was used for data collection. Parents of children born in South Dakota from 2011 to 2018 were asked about their knowledge of CMV and cCMV, including common sequelae and ways to minimize exposure. Flyers were sent to randomly selected daycares and the link was posted on social media pages to advertise the electronic survey to South Dakota parents. After completing the survey, participants were directed to cCMV educational resources.

Respondents were more knowledgeable regarding the sequelae of cCMV rather than its transmission process or ways in which viral exposure can be minimized. Results show that there remains a need for cCMV awareness in South Dakota, particularly with a large focus on preventative measures.

Key Words: CMV, cCMV, sensorineural hearing loss, public awareness

Acronyms: CMV = cytomegalovirus, cCMV = congenital cytomegalovirus, CDC = Centers for Disease Control and Prevention, IRB = Institutional Review Board

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Cytomegalovirus (CMV) is common and typically harmless to the general public, but congenital cytomegalovirus (cCMV) poses a danger to babies when contracted in utero. Awareness of cCMV is lower than many other well-known congenital conditions, yet prevalence of cCMV is higher (Doutre et al., 2016). Preventing transmission is paramount and begins with awareness of the virus and how it is transmitted, which is why some states have created legislation aimed at increasing awareness and screening at birth. South Dakota does not have cCMV legislation and has the added challenge of unique rural geography, making dissemination of information at varying levels of healthcare difficult.

Cytomegalovirus belongs to the *Herpesviridae* family of viruses. After initial infections, all viruses of the *Herpesviridae* family remain latent within their host cells, with the possibility of reactivation. In individuals with a latent infection, the virus can reactivate without causing symptoms; however, during the reactivation period, the viral host will be actively shedding the virus, thus transmitting the infection. Initial infection of CMV occurs through direct contact with the virus, typically through saliva and urine, with symptoms that mimic the common cold and likely go unnoticed. Congenital CMV contracted by a baby through the placenta, when the mother is actively infected with the virus during pregnancy, can cause serious and permanent risks.

According to the Centers for Disease Control and Prevention (CDC), the United States' prevalence rate for infants with cCMV is one in 200 children. Of infected newborns, about 10% are born with symptoms at birth, 10–15% are asymptomatic at birth but later develop hearing loss or other neurologic impairments, and the remaining 75%–80% will have no sequelae (Boppana et al., 2013; Kenneson & Cannon, 2007). What many do not realize is all infected newborns, with or without sequelae, will continue to shed and transmit the infection through bodily fluids for 18 to 30 months (Pati et al., 2016).

The most common diagnosis for a newborn infected with cCMV, symptomatic or asymptomatic, is sensorineural hearing loss (Naing et al., 2016). In addition, cCMV is acknowledged as the most common cause of

nonhereditary sensorineural hearing loss and an important cause of neurodevelopmental delay in children (Goderis et al., 2014; Kenneson & Cannon, 2007; Kimberlin et al., 2015). In addition to hearing loss, infants born with symptomatic cCMV often have more severe and permanent sequelae than infants born asymptomatic. Some sequelae can include neurologic delays, microcephaly, intracranial calcification, hyperbilirubinemia, motor defects, chorioretinitis, and seizures.

Despite the prevalence and danger cCMV poses for babies, many parents and medical providers are unaware of the virus, its sequelae, mode of transmission, and ways to minimize exposure. Awareness of cCMV was ranked the lowest by women when compared to other childhood conditions including the following: Spina bifida, Down syndrome, HIV/AIDS, sudden infant death syndrome, fetal alcohol spectrum disorder, congenital toxoplasmosis, congenital rubella syndrome, autism, Parvovirus B19, and Beta strep (Cannon et al., 2012; Doutre et al., 2016). Studies also reveal that medical providers have limited knowledge about cCMV and its dangers. The American College of Obstetricians surveyed a sample of OB/GYNs across the United States. Results revealed about half of the OB/GYNs surveyed were knowledgeable about how CMV is transmitted and preventative measures pregnant women can take, and 44% reported counseling their patients about preventing cCMV infection (Anderson et al., 2007; Fowler & Boppana, 2018).

The public health impact of cCMV is substantial, under-recognized, and is an issue worldwide due to its prevalence and the permanent sequelae (Binda et al., 2016). In an effort to reduce the prevalence and increase awareness, legislation to support cCMV screening and/or education has been enacted in 11 states, with additional states having proposed legislation. Five of the 11 states have specific laws regarding hearing-targeted testing for cCMV in infants (Doutre, 2015; National CMV Foundation, n.d.). South Dakota has no legislation for cCMV, but does have one hospital, Sanford Health, with a hearing-targeted cCMV screening protocol, which started April 29, 2013.

South Dakota is a conservative state that has historically opposed mandated healthcare policy. South Dakota's unique landscape of healthcare services adds to the challenge of disseminating information about cCMV, especially for pregnant mothers and families. Currently in South Dakota there are twenty-two hospitals, located in twenty counties, that offer labor and delivery services, meaning only 30% of counties in the state provide these services (South Dakota Department of Health - Birth report; South Dakota Department of Health - Metabolic screening program report, February 2018). Many mothers living in rural parts of the state must travel to receive services from a specialist in the management and care of pregnant women and babies, such as an obstetrician. However, due to the travel and associated costs, many mothers may seek prenatal care from another qualified medical provider (i.e., family practice physicians, pediatricians, nurse practitioners, or physician assistants), instead of a specialist located farther away in a larger town. Therefore, not only do obstetricians in South Dakota need to be aware and knowledgeable about cCMV, but a wide range of medical professionals need to be able to counsel and educate mothers and families about cCMV.

The purpose of this study was to evaluate the knowledge of South Dakota parents and medical providers with regards to cCMV, its sequelae, and ways to minimize the risk of infection and transmission.

Method and Materials

Participants

Prior to data collection, approval to conduct the study was obtained from the Institutional Review Board (IRB) at the University of South Dakota. The target population included South Dakota parents and licensed medical providers who work with pregnant women and children, including medical doctors specializing in family medicine, obstetricians and gynecologists, pediatricians, nurse practitioners, and physician assistants. Participation was voluntary. Parents choosing to volunteer had to be current residents of South Dakota, and their child must have been born in the state. The total number of parent survey responses was 150. However, 15 respondents did not provide all the demographic information necessary to analyze data, and therefore their responses were not included in data analysis; 135 surveys had complete demographic information necessary for data analysis. Responses from the 135 participants who provided complete demographic information were included in data analysis. To estimate the total sample size needed, a G-power analysis with a moderate effect size of 0.25 and a power of 0.95 was completed a priori. Previous studies with similar focus did not report an effect size; therefore, a moderate effect size was selected. The estimate for total sample size was 210 participants (105 parent participants and 105 medical provider participants).

Participant Recruitment

Participants were recruited from within the state of South Dakota using emails and flyers sent to parent and tot groups, daycares, and various social media platforms. A list of licensed daycares was obtained online from the South Dakota Department of Social Services. For this study, the state of South Dakota was split into four regions as defined by the South Dakota Department of Health Public Health Preparedness and Response Team.

Five counties were randomly selected from within each of the four regions to be included in the survey. The daycares within these counties were then randomly rank ordered and called in order, smallest to largest. During each phone call, daycares were asked for their willingness to pass out information about the survey to the parents of the children in their care. If the daycare initially selected *did not agree to distribute information about the survey,* the daycare associated with the next value in rank order was contacted. In each phone call, the licensed daycare provider listed by the South Dakota Department of Social Services was requested as a point of contact.

The same script was used for each phone call, which

explained the reason for calling, described what would be asked of the daycare provider, and asked about their willingness to participate. If they agreed to participate, cover letters containing the survey link and investigators' contact information were provided and passed on to the parents. The cover letter purposefully did not specify cCMV; rather it generally stated that the study was seeking information from parents and healthcare providers on illness during pregnancy.

Medical providers were recruited via their state organizations, social media, and by contacting all large hospitals, critical access hospitals, rural health clinics, and healthcare networks in South Dakota. These individuals were asked to send out the cover letter (again not specifying cCMV), which contained the link for the survey. They were asked to distribute this cover letter to the providers in their organization or network. The contact information for all hospitals, critical access hospitals, and rural health clinics can be found on the South Dakota Department of Health website and was therefore considered public knowledge. However, not enough medical provider survey responses were obtained to perform statistical analysis for this population.

Survey Administration

A cross-sectional survey was used for this study to collect descriptive data from people across a geographically large and rural target population (see Appendix). The survey was conducted electronically using PsychData. Participants were given a link to this survey through cover letters provided to daycare providers and were to access the link using any internet accessible device. The survey took approximately five to seven minutes to complete. In the first section of the survey, demographic information was collected. The first demographic question for the parents asked for the child's date of birth. This allowed investigators to know whether the child was born before or after a hearing-targeted cCMV screening protocol was implemented by Sanford Health on April 29, 2013. Other demographic information collected included at which hospital the child was born and in which South Dakota county the family resided at the time of their child's birth.

The next section of the survey focused on questions related to the knowledge participants had regarding CMV and cCMV. Knowledge questions focused primarily on sequelae and ways to minimize exposure. Questions about other areas of knowledge, including the definition of cCMV, incident rate, and ways to identify cCMV, were included to look for trends. Participants were asked to answer the questions with respect to their youngest child and to the best of their ability, with all questions presented in closed-set, multiple-choice format. The last section of the survey was educational. Participants were provided links to websites and other materials where they could learn more about cCMV.

Results

A total of seven medical providers were surveyed, which is too small to provide statistically meaningful results. The distribution of respondents across South Dakota, in terms of the county they resided in at the time of their child's birth, the county where birth occurred, and where the birthing hospitals are located, by county, in South Dakota is represented in Figure 1.

Respondents were asked to rank, using a Likert Scale, how knowledgeable they felt about ability to minimize their risk of exposure to CMV, cCMV sequelae (problems associated), and transmission from mother to baby. Responses can be seen in Figure 2. The most frequently selected answer for each of the three questions was v*ery unsure*.

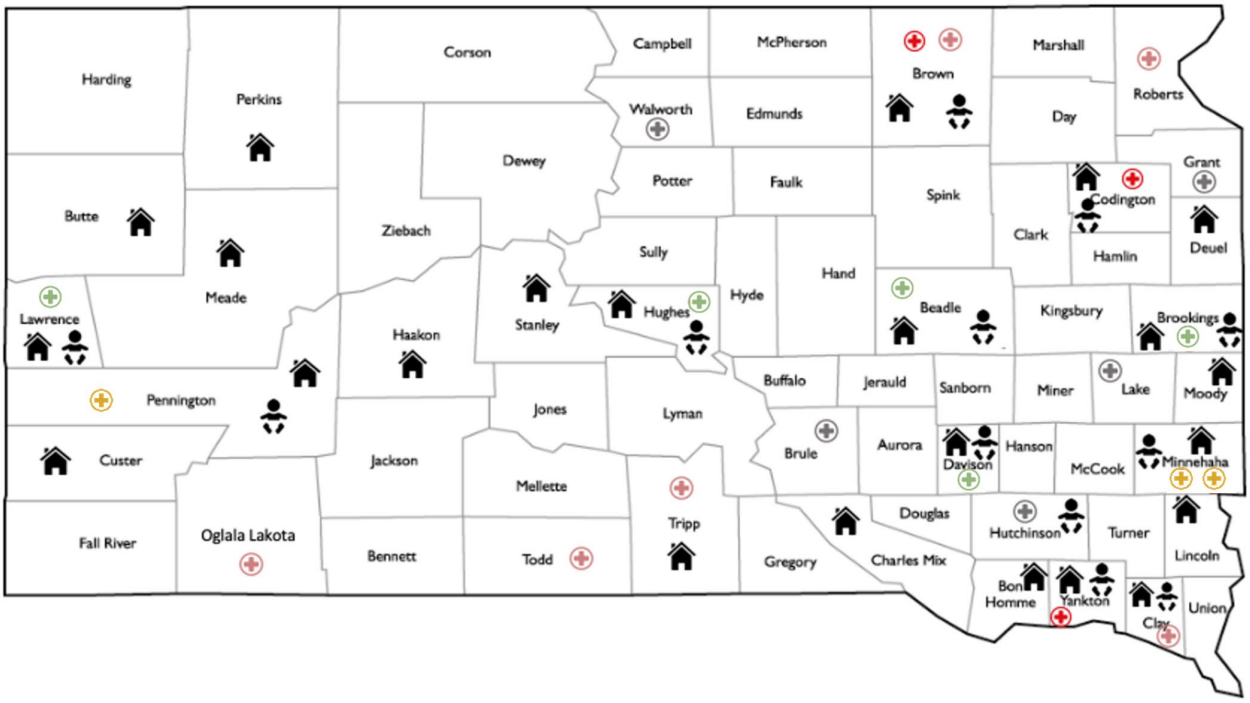
When respondents were asked where they had learned about congenital CMV, they most frequently answered (56%) that they had not learned about cCMV from anyone. The next most frequently answered response (18%) was *other*. Individuals who gave this response were often healthcare professionals from various fields (e.g., nursing, speech language pathology, audiology, physical therapy), a student of one of the aforementioned fields, or someone who worked with mothers and/or infants.

Respondents were asked 10 questions about their knowledge of cCMV and CMV; the first five questions pertained to the sequelae and the last five questions pertained to how to minimize risk of transmission (refer to Figure 3). For ease of analysis, please note that these 10 questions are listed as Questions 9–19 in the Appendix. In response to the questions about sequelae, respondents generally answered three of five questions correctly. Question 2, "Congenital CMV can be diagnosed no later than____," was frequently answered incorrectly, with the most commonly selected incorrect answer being "at birth". Question 3, "What is the most common problem associated with cCMV?" was answered incorrectly 60.0% of the time when the birth of the child was before 4/29/13 and 48.7% of the time when the birth of the child was after 4/29/13. The commonly selected incorrect answers were evenly spread across the following choices: "vision problems" (30% before, 18% after) and "seizures" (10% before, 18% after). The last five questions pertained to how to minimize risk of transmission. Respondents correctly answered question 6 ("All of the following activities are dangerous, as they may expose a mother to CMV and her unborn baby to cCMV, except____") and question 7 ("True or False: Changing a diaper exposes me to CMV through urine and fecal matter."). Question 8, "During which activity below is it most likely for a pregnant mother to be exposed to CMV?" was answered incorrectly about 70.0% of the time for birthdates before and after 4/29/13, with the most frequently selected incorrect answer being "scooping a cat's litter box" (40% before, 48% after). Question 9, "Children who are born with CMV will shed the virus for____," was answered incorrectly about 90% of the time for birthdates before and after 4/29/13, with the most frequently selected incorrect answers being "4–6 weeks (50% before, 44% after) and "6–12 months" (30% before, 35% after). Question 10, "What is the incidence rate of cCMV occurrence each year?" was answered incorrectly 70% of the time before 4/29/13 and 76% after 4/29/13, with the most frequently selected incorrect answers being

Figure 1

Respondent Distribution Across Counties in South Dakota

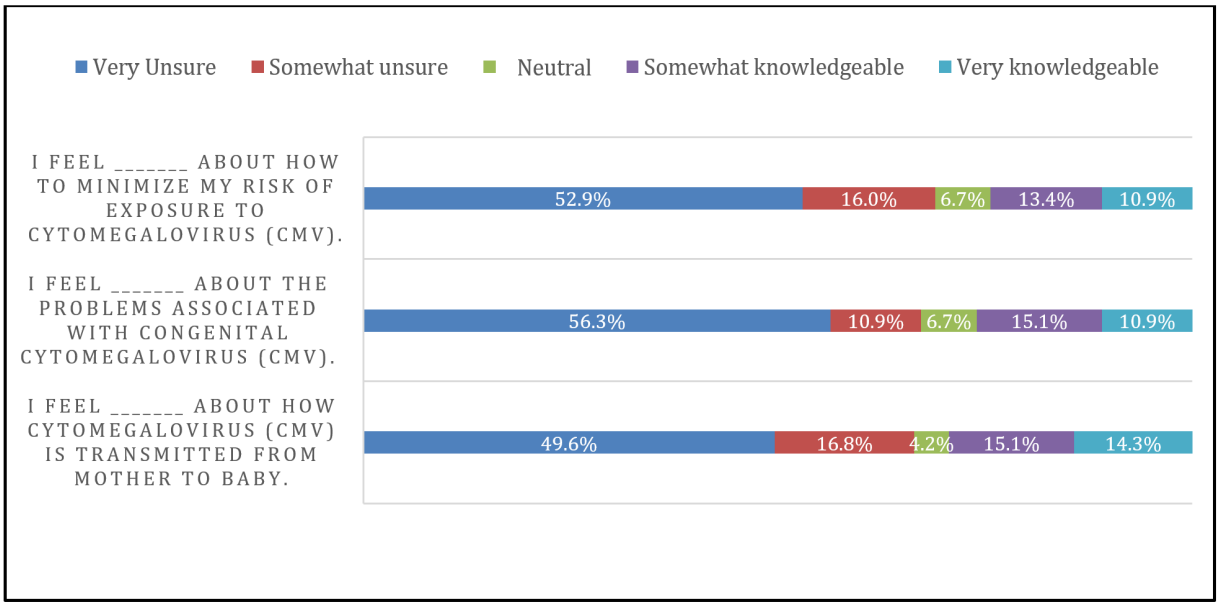
South Dakota County	Number of survey respondents residing in the county at the time of child's birth	Number of survey respondent's babies born in the county	Hospitals with Birthing Services (births per year)
			2000+ 500-1000 300-499 100-299 1-99
Beadle	1	1	
Bon Homme	2	0	None
Brookings	7	4	
Brown	1	1	
Brule	0	0	
Butte	3	0	None
Charles Mix	1	0	None
Clay	11	3	
Codington	3	3	
Custer	3	0	None
Davison	3	2	
Deuel	2	0	None
Grant	0	0	
Haakon	1	0	None
Hughes	8	9	
Hutchinson	0	1	
Lake	0	0	
Lawrence	3	5	
Lincoln	19	0	None
Meade	1	0	None
Minnehaha	28	64	
Moody	1	0	None
Oglala Lakota	0	0	
Pennington	27	34	
Perkins	2	0	None
Roberts	0	0	
Stanley	1	0	None
Todd	0	0	
Tripp	1	0	
Walworth	0	0	
Yankton	2	4	



Note. The distribution of respondents across South Dakota, the county they resided at the time of their child's birth, the county where birth occurred, and where the birthing hospitals are located, by county, in South Dakota.

Figure 2

Participant Responses to Questions about Cytomegalovirus (CMV) Transmission, Sequelae, and Exposure



Note. Respondents were asked how knowledgeable they felt about CMV transmission from mother to baby, congenital CMV sequelae (problems associated), and how to minimize risk of exposure. Respondents most frequently answered "very unsure".

"1 in 550" (40% before, 27% after) and "1 in 1050" (20% before, 21% after).

Size of birthing facility and presence of protocol were analyzed (refer to Figure 4). A *large hospital* was considered any hospital that had 2000+ births a year. It did not appear that size of birthing facility had an effect on knowledge about CMV and cCMV. Next, the knowledge of those respondents whose birthing facility had an established hearing-targeted cCMV screening protocol was compared to the knowledge of those respondents whose birthing facility had no such protocol. Much like the size of the birthing facility, the presence of a protocol had no impact on respondents' knowledge of CMV or cCMV.

Discussion

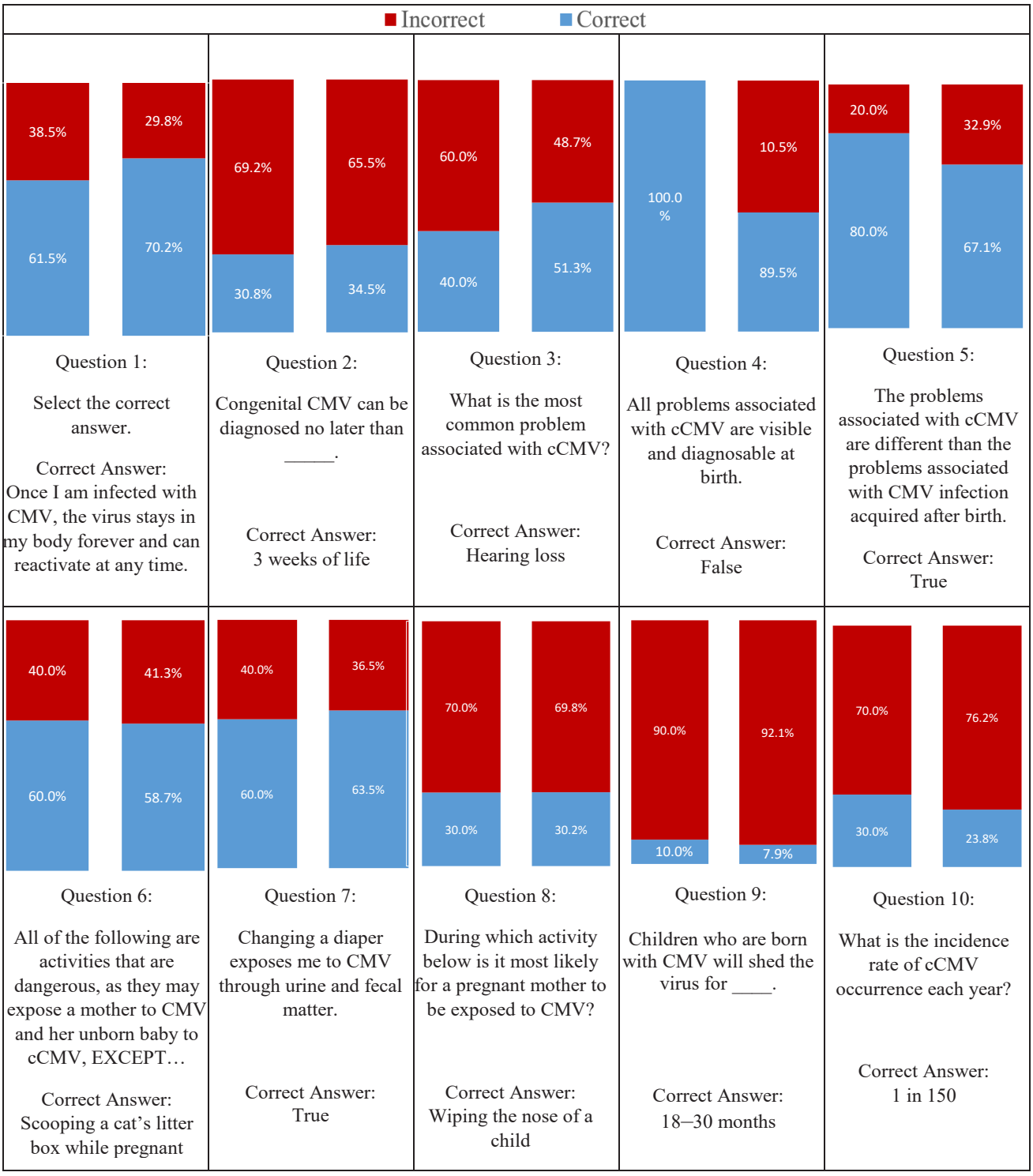
Respondents were able to correctly answer several questions about cCMV sequelae and ways to minimize exposure on the survey, despite overwhelmingly rating their confidence about the subject as *very unsure*. This finding is consistent with 56% of respondents reporting that they had "not learned about cCMV from anyone," which is consistent with data from the 2015–2016 HealthStyles survey, that showed only 9% of women had heard about CMV (Doutre et al., 2016). It is clear that parents lack confidence in their knowledge about CMV, the problems associated with the virus, and how to minimize exposure. It was also interesting to observe that when parents did report having learned about cCMV, 18% reported learning about it from "other" and explained they were an employee or a student of the healthcare field. Therefore, it appears cCMV is important enough to be taught to future healthcare providers; however, it also appears the message is not being relayed to parents.

Respondents showed they were more knowledgeable about sequelae compared to their knowledge about minimizing exposure. When analyzing responses to the questions about minimizing exposure, there was confusion between cCMV and toxoplasmosis. This indicates there is confusion about how CMV is transmitted and therefore how to minimize risk of exposure. In Question 6, respondents were asked about activities that expose a mother to CMV and an unborn baby to cCMV. Approximately 60% of the time, respondents correctly responded that scooping a cat's litter box did not put a mother or her baby at risk for becoming infected with CMV/cCMV. In Question 8, respondents were contradictory in their answer to Question 6; nearly 45% of respondents incorrectly answered that "scooping a cat's litter box" was an activity that was the most likely to cause a pregnant mother to be exposed to CMV. This contradiction reveals there is work to be done when it comes to educating parents about CMV and how to minimize risk of exposure. Marshall and Adler (2009) estimate that every two years, an average of 80,000 pregnant women will be exposed to CMV from an infected child who attends daycare. Without a hygienic intervention, approximately half will become infected during pregnancy, with the majority of the infections being preventable. Educating parents about CMV, how it is transmitted, and ways to minimize exposure is imperative to reducing infection rates.

South Dakota has many small birthing facilities and only a few larger facilities. Results from this study suggest that birthing facility size does not impact parent knowledge of CMV and cCMV sequelae or how to minimize exposure to CMV and cCMV. There was also no difference between

Figure 3

Respondents' Knowledge about Sequelae (Questions 1-5) and How to Minimize Exposure and Transmission (Questions 6-10)

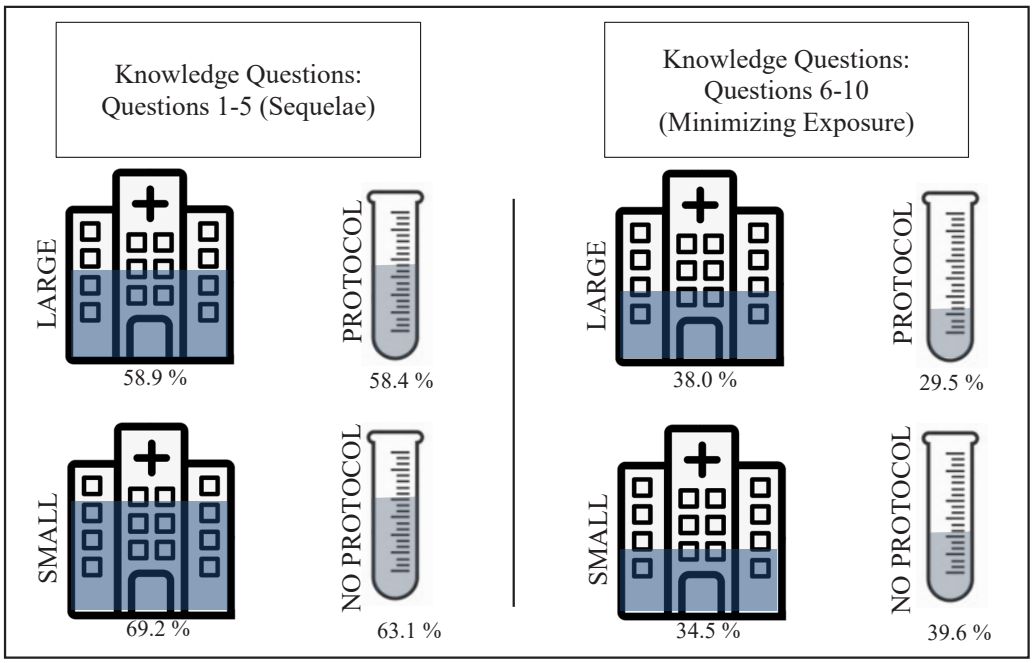


knowledge of sequelae and how to minimize exposure when comparing responses from parents whose infant was born at the hospital that has a hearing-targeted screening protocol to responses from parents whose infant was born at a hospital with no CMV screening protocol. With neither size nor screening protocol having an impact on knowledge, it likely means there will need to be a focused effort on cCMV awareness in South Dakota at all the prenatal clinics and birthing facilities.

Although the results obtained from this study are specific to South Dakota, they are in line with studies across the United States and the world that identify cCMV as having high prevalence yet low awareness and knowledge (Doutre et al., 2016; Marshall & Adler, 2009; Mazzitelli et al., 2017). The need for cCMV awareness is substantial, particularly with a large focus on preventative measures (Thackeray et al., 2017).

Figure 4

Cytomegalovirus (CMV) Knowledge at Large and Small Birthing Hospitals



Note. Knowledge of CMV and congenital CMV (CCMV) was analyzed at both large and small hospitals, along with knowledge at the hospital where there is a hearing targeted cCMV screening protocol.

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Appendix
Parent Survey

***IRB* Approval effective from: 12/19/2017**
***IRB* Approval not valid after: 12/18/2018**
USD IRB

Date: December 12, 2017

Dear Parent:

You are invited to participate in a research study. The purpose of the study is to better understand parent knowledge about different metabolic, inherited, and genetic disorders at birth. We are inviting you to be in this study because you are the parent of a child born in South Dakota.

If you agree to participate, we would like you to complete a survey. The survey can be completed on any computer/device with Internet access and will take approximately 5 to 7 minutes. The types of questions you will be asked include where your child was born, where/if you learned about various metabolic, inherited, and genetic disorders during pregnancy, and your current knowledge of congenital metabolic, inherited, and genetic disorders.

We will keep the information you provide anonymous, however federal regulatory agencies and the University of South Dakota Institutional Review Board (a committee that reviews and approves research studies) may inspect and copy records pertaining to this research.

Your responses will be anonymous to ensure that they cannot be linked to you. If we write a report about this study we will do so in such a way that you cannot be identified.

There are no known risks from being in this study, and you will not benefit personally. However, we hope that others may benefit in the future from what we learn as a result of this study.

All survey responses that we receive will be treated confidentially and stored on a secure server. However, given that the surveys can be completed from any computer (e.g., personal, work, school), we are unable to guarantee the security of the computer on which you choose to enter your responses. As a participant in our study, we want you to be aware that certain “key logging” software programs exist that can be used to track or capture data that you enter and/or websites that you visit.

Your participation in this research study is completely voluntary. If you decide not to be in this study, or if you stop participating at any time, you will not be penalized or lose any benefits for which you are otherwise entitled.

If you have any questions, concerns or complaints now or later, you may contact us at the number below. If you have any questions about your rights as a human subject, complaints, concerns or wish to talk to someone who is independent of the research, contact the Office for Human Subjects Protections at 605/677-6184. Thank you for your time.

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Appendix (cont.)
Parent Survey

- Please answer all questions to the best of your ability
- All questions should be answered in regard to your YOUNGEST child

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1. What is your child's date of birth (month/date/year)?

2. At what facility did you receive your prenatal care? (e.g. Avera Women's Clinic, Sanford Obstetrics and Gynecology Clinic, Black Hills Obstetrics and Gynecology Clinic)

3. In which South Dakota hospital was your child born? (Drop down)
   a. Avera – McKennan (Sioux Falls)
   b. Avera – Sacred Heart (Yankton)
   c. Avera – St. Mary's (Pierre)
   d. Avera – Queen of Peach (Mitchell)
   e. Avera – St. Luke's (Aberdeen)
   f. Avera – St. Benedict (Parkston)
   g. Avera – Milbank Area Hospital
   h. Brookings Hospital
   i. Coteau Des Prairies Hospital (Sisseton)
   j. Huron Regional Medical Center
   k. Madison Community Hospital
   l. Mobridge Regional Hospital
   m. Pine Ridge IHS Hospital
   n. Prairie Lakes Health Care (Watertown)
   o. Sanford Aberdeen Medical Center
   p. Sanford Chamberlin Medical Center
   q. Sanford USD Medical Center – Sioux Falls
   r. Sanford Vermillion Hospital
   s. Rapid City Regional Hospital
   t. Spearfish Regional Hospital
   u. Rosebud IHS Hospital
   v. Winner Regional Health Care Center
   w. Other – Please Specify

4. In what South Dakota county did you reside at the time of your son or daughter's birth?

5. I feel _____ about how congenital CMV is transmitted from mother to baby.
   a. Very Unsure
   b. Somewhat Unsure
   c. Neutral
   d. Somewhat Knowledgeable
   e. Very Knowledgeable

6. I feel _____ about the problems associated with congenital cytomegalovirus (CVM).
   a. Very Unsure
   b. Somewhat Unsure
   c. Neutral
   d. Somewhat Knowledgeable
   e. Very Knowledgeable

7. I feel _____about how to minimize my risk of exposure to cytomegalovirus (CMV).
   a. Very Unsure
   b. Somewhat Unsure
   c. Neutral
   d. Somewhat Knowledgeable
   e. Very Knowledgeable

Appendix (cont.)

Parent Survey

8. I learned about congenital CMV from____?
   a. Social media
   b. A friend or family member
   c. Medical provider
   d. Online resource (not social media)
   e. I have not learned about congenital CMV from anyone
   f. Other (please explain)

   LOGIC (if c was selected)
   8a). Which medical provider educated you the most regarding congenital CMV?
      a. Pediatrician
      b. OB/GYN
      c. Family Medicine
      d. Nurse practitioner
      e. Physician Assistant
      f. Other (please specify)

   8b.) Please specify when the medical provider informed you about congenital CMV.
      a. Pre-pregnancy
      b. First trimester
      c. Second trimester
      d. Third trimester
      e. Post pregnancy
      f. My medical provider never talked with me about congenital CMV.

9. Select the correct answer
   a. Once I am infected with cytomegalovirus (CMV), I have immunity and will not be infected again.
   b. Once I am infected with cytomegalovirus (CMV), the virus stays in my body forever, and can re-activate at any time.
   c. Once I received the vaccine for cytomegalovirus (CMV), I will have immunity towards the virus, and will not be infected.

10. Congenital CMV can be diagnosed no later than ___.
   a. At birth
   b. 3 weeks of life
   c. 3 months of life
   d. 3 years of life

11. What are the known problems associated with congenital cytomegalovirus (CMV)?
Select all that apply.
   a. Small head size
   b. Autism
   c. Vision Problems
   d. Hearing Loss
   e. Lung problems
   f. Jaundice
   g. Mental Disability
   h. Facial abnormalities

12. What is the most common problem associated with congenital cytomegalovirus (CMV)?
   a. Vision Problems
   b. Seizures
   c. Hearing loss
   d. Cerebral Palsy

13. All problems associated with congenital cytomegalovirus (CMV) are visible and diagnosable at birth.
   a. True
   b. False

Appendix (cont.)

Parent Survey

14. The problems associated with congenital cytomegalovirus (CMV) are different than the problems associated with CMV infection acquired after birth.
    a. True
    b. False

15. All of the following activities are dangerous, as they may expose a mother to cytomegalovirus (CMV) and her unborn baby to congenital CMV, EXCEPT ____.
    a. Sharing a cup or straw with a child
    b. Scooping a cat's litter box while pregnant
    c. Sharing a fork with your child
    d. Picking up or playing with children's toys

16. Changing a diaper exposes me to CMV through urine and fecal matter.
    a. True
    b. False

17. During which activity below is it most likely for a pregnant mother to be exposed to CMV?
    a. Scooping a cat's litter box
    b. Touching an infected surface
    c. Wiping the nose of a child
    d. Getting bit by an insect

18. Children who are born with CMV will shed the virus for____.
    a. 4 - 6 weeks
    b. 6 months – 12 months
    c. 18 months – 30 months
    d. 36 months – 42 months

19. What is the incidence rate of congenital CMV occurrence each year?
    a. 1 in 50
    b. 1 in 150
    c. 1 in 550
    d. 1 in 1050
    e. 1 in 10,500
    f. 1 in 30,500
    g. 1 in 50,500

For more information about congenital CMV
Visit the following links:

National CMV Foundation
https://www.nationalcmv.org/home.aspx

Centers for Disease Control and Prevention
https://www.cdc.gov/cmv/overview.html

Thank you!

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Brief History of Auditory-Based Interventions and Related Developments

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Abstract

This is a brief but broad narrative and non-systematic review of developments that led up to how 21st century digital technology and translational research influenced, in particular, cognitive psychology and our improved understanding of mental resources among children who are deaf or hard of hearing (DHH). In turn, systemic multi-disciplinary research findings gave birth to Auditory Cognitive Neuroscience (ACN). Three broad constructs unique to ACN (i.e., auditory attention, effortful listening, and auditory fatigue) are then described in relation to children who are DHH. This review concludes with a brief examination of future opportunities for researchers and clinicians who can ensure that children who are DHH will benefit from cross-disciplinary translational research findings.

Keywords: hearing loss, science, evidence-based, cochlear implants

Acronyms: ACN = Auditory Cognitive Neuroscience; DHH = deaf or hard of hearing; EBP = evidence-based practice; EF = executive functions; LSL = listening and spoken language

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Prior to the mid-19th[th] century, a child who was deaf or hard of hearing (DHH) was typically objectified as a *deaf-mute* or *the deaf and dumb* (e.g., Burnes, 1967; Huizing, 1959). However, educational practices and technological developments of the past century made huge differences in the lives of families and their children diagnosed as either deaf or hard-of-hearing. Person-first language, such as *child who is deaf or hard of hearing* is now standard in medical settings and is becoming more widespread in society (e.g., Rhoades, 2010b).

The evolution of auditory-based interventions for families and their children came about as the result of many helping hands, particularly those in the audiological and otological professions as well as science inventors (for reviews, see Felisata, 2007; Nogueira et al., 2007; Vogel et al., 2007). Wearable electric or vacuum tube hearing aids were used at the outset of the 20[th] century; these devices enabled some children with severe hearing loss to access conversational sound (Howard, 1998). Consequently, some American and European educators, audiologists, and otologists began earnestly advocating for the use of residual hearing (e.g., Ewing et al., 1936; Goldstein, 1928; Kroiss, 1903; Urbantschitsch, 1895; Wright, 1915).

By the mid-20[th] century, portable transistorized hearing aids became widely available (Bello, 1953). Concurrently, aural rehabilitation programs were being developed to include tests of hearing, speech perception, and hearing aid selection while counseling, and placement services were also being developed (e.g., Carhart, 1946; Ross, 1997). These programs included the teaching of speech reading and auditory skills coupled with the use of assistive hearing technology. Early intervention programs were also established for families and their children who are DHH (e.g., Fiedler, 1952). Many of those programs were designed to promote listening and spoken language (LSL) skills (e.g., Beebe, 1953; Griffiths, 1955; Huizing & Pollack, 1951; Wedenberg & Fant, 1949).

Digital Technological Revolution

The advent of digital technology during the latter part of the 20[th] century dramatically changed hearing technology and LSL interventions. The transition from analog to digital hearing aids enabled clinicians to better meet individual needs (e.g., Gustafson et al., 2014; Levitt, 2007; Packer, 2016; Reinhart et al., 2019). Cochlear implants, developed and first worn in 1961 (Eshraghi et al., 2012), were soon followed by other types of auditory implants (Møller, 2006). The same circuitry found in computers and smart phones is now used in hearing devices along with Bluetooth capability. This provides hearing device users with the capability of hearing the sound source as if it were directly in their ear.

In addition to empowering audiologists with more specialized and complex diagnostic equipment to facilitate the selection and programming of hearing devices, digital technology gave rise to the development of equipment that identified the nature and origin of hearing loss (Hoth & Baljić, 2017). It also expanded potential therapeutic or rehabilitation options for hearing device users (e.g., Flynn, 2005; Stagiopoulos et al., 2016; Zeitler et al., 2019).

Moreover, as digital technology has gained worldwide prominence, it facilitated the widespread sharing and management of research data in hearing healthcare. This digital transformation gave rise to early identification and tele-intervention programs for families and their babies who are DHH (e.g., Alam et al., 2016; McCarthy et al., 2010).

By the end of the 20th century, partly due to information technology, the professions of otology, laryngology, and rhinology were dramatically altered. These disciplines combined to form the broader and more complex cross-disciplinary profession now known as otorhinolaryngology; this embraces a multitude of sub-specialties that include pediatric otorhinolaryngology, some of whose physicians may be referred to as Children's Ear, Nose and Throat (ENT) physicians (Weir, 2000). Significant improvements have since been made in identifying and managing hearing-related syndromes (e.g., Hone & Smith, 2003) as well as such common childhood hearing health issues as otitis media (Bluestone & Shurin, 1974).

Simultaneously, the field of psychology was undergoing a metamorphosis (for reviews, see Miller, 2003; Saffran & Kirkham, 2017). Insights into the human brain and mind were flourishing and linguistics was being redefined (e.g., Chomsky, 1965; Pinker, 1994). Teachers of the deaf, audiologists, and speech-language pathologists were directly affected by this cognitively-driven linguistic revolution (e.g. Furth, 1966; Levine, 1960; Myklebust, 1960; Van Uden, 1970; Weikart et al., 1971). Consequent to the considerably expanded knowledge base of how language develops as well as advances in hearing technology, increasingly more programs promoting auditory-verbal practices were established (e.g., Rhoades, 1982).

Cognitive psychologists began integrating information processing models, such as computer science (Aaronson, 1994), into their study of mental resources, that is, the cognitive processes of purposeful goal-directed behaviors as well as hearing and language (Barkley, 2012; Goldstein et al., 2014). Research data gave rise to constructs widely referred to as *statistical learning* and *executive functioning* (e.g., de Boysson-Bardies, 1999; Eisenberg, 1976; Gopnik et al., 1999; Tomasello, 2003; Yang, 2006). The meta-construct referred to as *Executive Functions* (EF) involves those interrelated foundation skills carried out by the prefrontal areas of the brain; those capacities include attention, working memory, fluency or speed of processing information, self-regulation or response inhibition, and cognitive flexibility—all considered essential for learning, creativity, problem-solving, self-regulation, empathy, and socio-emotional behaviors (e.g., Meltzer, 2007; Sarma & Thomas, 2020). Cognitive psychology revealed underlying differences in learning processes and outcomes.

During the latter half of the 20th century, some children with severe-profound deafness learned to listen and use spoken language quite well and were educated within mainstream classrooms (Goldberg & Flexer, 1993; Rhoades & Chisolm, 2000; Robertson & Flexer, 1993; Wray et al., 1997). However, in spite of much-improved technology and interventions, many other children did not perform as well as expected (Lim & Hogan, 2017). Neurobiological findings that informed the research of developmental psychologists, cognitive psychologists, and neuropsychologists also served to inform practitioners from the disciplines of audiology, deaf education, speech pathology, and otolaryngology (e.g., Faulkner & Pisoni, 2013). Digital technology across these disciplines helped give rise to modern neuroscience which further informs practitioners as to why children who are DHH demonstrate tremendous variability in learning how to listen and use spoken language.

During the initial rise of data-driven research findings, clinicians were not integrating the scientific evidence into their practice (Carnine, 1997; Davies, 1999). Near the end of the 20th century, demands were repeatedly made for *evidence-based practice* (EBP; e.g., Davies, 1999; Foster, 1999; Sackett et al., 1996). EBP indicates that well-designed research findings, that is, verifiable scientific evidence, should affect clinical decision-making and how clinicians trained in auditory-verbal therapy should systematically implement carefully designed services for families and their children who are DHH (Rhoades, 2010a).

21st Century Translational Research

The first decade of the 21st century amplified and broadened the call for implementing data-driven evidence (e.g., Eccles & Mittman, 2006; Gallagher, 2004; Odom, 2009). *Implementation science* called for effective strategies that would facilitate clinician learning and behavioral changes, something that had not yet occurred on a wide scale (Burns & Ysseldyke, 2009). However, before scientific evidence can be incorporated into practices, the evidence must be rendered meaningful, that is, the knowledge translated so that clinicians understand it.

For instance, multidisciplinary translation research can be seen in biometrics. This is a branch of computer science and technology that has become part of the broader research currently serving those who are DHH. 3D ear scanners can now be used to provide custom fit ear molds that are of critical importance to young hearing aid wearers (Liu et al., 2015). Currently, the most common way to create ear molds continues to be through the use of ear mold impression materials; however, 3D ear scanners are a new technology that will likely impact future practice. This is an example of data-driven evidence showing how researchers from seemingly disparate disciplines are significantly affecting treatment for children who are DHH.

The integration of data logging into hearing aids is another example of how cross-disciplinary research benefits children who are DHH. The data logging feature can be used to monitor and hopefully increase the time that acoustic accessibility is provided to language learners (Ambrose, 2019). As a valuable early intervention tool, it encourages collaboration between audiologist and therapists to promote increased hearing aid use. Data logging has many uses for improving hearing aid behavior (e.g., McMillan et al., 2018).

Auditory systems are shaped by complex, dynamic, and reciprocal processes between genetics, neurobiology, and experiences (for review, see Kral & O'Donoghue, 2010). Knowledge of brain mechanisms and cognitive functions supporting auditory learning is critical for understanding the enormous variability of outcomes experienced by children who are DHH (see McLachlan & Wilson, 2010 for a review). Disruptions to auditory functioning such as tinnitus (Mohamed et al., 2016) and auditory neuropathy (Zeng et al., 2005) affect a variety of neurocognitive skills such as spoken language, mental resources, socio-emotional growth, and learning (Kral et al., 2016). Moreover, difficulties arising from disruptions occurring during infancy may persist beyond early childhood. Although critical periods for language learning are established, whether we can extend those periods of plasticity remain under investigation (Werker & Hensch, 2015).

Neurocognitive research findings show that: (a) One's mental resources have a saturation level that can be allocated to behavioral or learning tasks (e.g., Bays, 2018). (b) No two children are alike; there are individual differences in cognitive capacities (e.g., Dingemanse & Goedegebure, 2019; Lofkvist et al., 2020). (c) The amount or degree of mental resources allocated a task increases as the task becomes more difficult or demanding. For example, cognitive load increases and comprehension or learning outcomes decrease when listening to speech in difficult listening conditions because the task of processing information is more complex (e.g., Lehmann & Seufert, 2020; Zekveld et al., 2011). (d) Persons with good working memory capacity may have an advantage when learning languages and listening to speech in noisy backgrounds (e.g., Archibald, 2017; Astle et al., 2018; Michalek et al., 2018). (e) Children with early access to spoken or signed language are less likely to have executive deficits than those with late access to language (e.g., Botting et al., 2017; Hall et al., 2018). (f) Many children who are DHH demonstrate deficits in auditory attention, working memory, and processing speed (e.g., AuBuchon et al., 2015; Beer et al., 2014; Faulkner & Pisoni, 2013; Kronenberger & Pisoni, 2019). (g) Children who readily engage in pattern recognition tend to demonstrate good statistical learning skills that, in turn, can promote rapid language learning and more effective auditory perception (e.g., Arciuli & Conway, 2018; Deocampo et al., 2018; Riecke et al., 2020; Saffran & Kirkham, 2018; Studer-Echenberger et al., 2016). (h) Children who are DHH but have better language and working memory skills have better speech recognition scores in noise and reverberation than peers who are DHH but have lower language and working memory skills (e.g., McCreery et al., 2019; Torkildsen et al., 2019). (i) Among children who are DHH, better aided audibility is linked to stronger spoken language skills (e.g., McCreery et al., 2019). (j) Cognitive training may improve young children's core cognitive capacities of attention and working memory as well as other EF skills and speech perception-in-noise (e.g., Di Lieto et al., 2020; Du & Zatorre, 2017; Dubinsky et al., 2019; Koshimori & Thaut, 2019; Scionti et al., 2020). Figure 1 shows a summary of this information (see Figure 1).

Translational research currently promotes the multidirectional and multidisciplinary integration of patient-oriented research and population-based research (Rubio et al., 2010). Although cross-collaborative efforts are challenging, the fields of inquiry are ever-expanding. Science and innovation have become too complex for some audiologists, otolaryngologists, and auditory-based clinicians to fully comprehend and thus implement widely effective interventions (Woolf, 2008). A different type of researcher, such as one whose expertise cuts across

Figure 1

Neurocognitive Research Findings at a Glance (adapted from a variety of sources and discussed throughout this paper)

Neurocognitive Research Findings at a Glance

1. Cognitive capacities exist and can be saturated by specific tasks.
2. Individual differences exist for cognitive capacity.
3. Cognitive load increases as the complexity of the task increases.
4. High working memory capacity may be advantageous when learning in noisy environments.
5. Children with late access to spoken or signed language have increased executive function delays.
6. Children who are deaf or hard of hearing often demonstrate difficulty with auditory attention, working memory, and processing speed.
7. Pattern recognition and statistical learning skills promote language learning and auditory perception.
8. Children who are deaf or hard of hearing who have increased working memory skills have better speech recognition skills in noisy environments.
9. Better aided hearing audibility is linked to improved spoken language skills.
10. Cognitive training may improve cognitive capacity in the areas of attention and working memory as well as executive function skills and speech perception in noise.

many branches of knowledge, is bridging the translational divide. This type of researcher harnesses knowledge from seemingly disparate, complex disciplines to generate new knowledge for the benefit of evidence-based practitioners who, in turn, can implement new treatments (La Velle, 2015; Mitchell, 2016; Rubio et al., 2010).

The effort to build on basic scientific research from multiple fields of study is widespread (Lustig & Akil, 2012; Millett, 2020; Pichora-Fuller, 2014). Researchers are translating knowledge from across varied areas of specialization to inform auditory-based interventions (Butler, 2008). For example, genome sequencing may soon complement universal physiologic newborn screenings so that more children with syndromic and nonhereditary sensorineural hearing loss, such as congenital cytomegalovirus, will benefit from early identification and individualized interventions to meet specific needs (Goderis et al., 2014; Shearer et al., 2019). This will translate into more positive outcomes for children with complex needs.

Auditory Cognitive Neuroscience

Modern neuroscience is evolving to encompass many branches. Cognitive neuroscience is the study of the biological mechanisms underlying cognition. Auditory cognitive neuroscience (ACN) covers all aspects of auditory cognition that include perception of speech, music, and natural sounds to emotion, memory, attention, and production of auditory events as well as assessment of listening difficulties (e.g., Moore, 2015; Roessig & Mücke, 2019).

ACN research methods can include psychophysics or other behavioral paradigms, neurophysiology, anatomy, neuroimaging techniques (including MEG, fMRI, PET, EEG, TMS, and optical imaging), motion capture, modeling, neuropharmacology, and behavioral genetics. ACN scientists are interested in collaborating across disciplines and applying these methods to human development and those with hearing differences and/or disorders (Arlinger et al., 2009; Azhari et al., 2020; Pichora-Fuller, 2014). For example, pupillometry is the study of changes in the diameter of the pupil as a function of cognitive processing. This is now used widely in psychological and neurological research. Use of the pupil dilation response will permit improved understanding of the cognitive processes experienced by infants and older children who are deaf or hard of hearing (Kaldy & Blaser, 2020; Naylor et al., 2018).

Progress in understanding the structure and function of our children's responses to and the production of sounds necessitates crossing many disciplines that include disciplines within psychology as well as neuroscience, neurobiology, computer science, physiology, psychoacoustics, speech and hearing sciences, physics, and between theory and practice. ACN is the forum for such cutting-edge research.

Auditory Attention and Spatial Perception

Sensory attention is important to information processing because it controls finite resources, permitting an overall level of alertness or ability to engage with surroundings (Lindsay, 2020). ACN researchers have considerably broadened our understanding of auditory learning. For example, *auditory attention* is an intricate multi-dimensional construct that includes orienting, selecting, and/or focusing on environmental sound stimuli, like speech, for varying periods of time (Pichora-Fuller et al., 2017). Auditory attention serves as a critical core cognitive capacity underlying auditory learning, working memory, and other executive capacities (Engle, 2018; Kaya & Elhilali, 2017; Stavrinos et al., 2018). This attentional capacity operates as a form of sensory gain control, enhancing the attended stimuli whilst suppressing other stimuli. As such, auditory attention interacts with other sensory, motor, and cognitive systems (Zatorre, 2007).

Relative to those with normal hearing, persons who are DHH tend to demonstrate poorer auditory spatial acuity and weaker suppression of auditory distractors (Dai et al., 2018). This is important because attending to a sound is related to identifying the location of sound source or auditory spatial perception (Letowski & Letowski, 2012). Also of interest is that auditory perception is affected by non-spatial features of acoustic stimuli such as other sensory systems (Recanzone, 2011).

Sustained auditory attention is the prolonged focus on auditory stimuli. The listener's brain tracks attended speech through phase-locking of neural activity to the speech envelope known as the onset of a particular speech stream (Petersen et al., 2016). Sustained auditory attentional focus, then, is the neural tracking of pertinent auditory stimuli (Evans & McGettigan, 2017; Kaya & Elhilali, 2017).

Selective auditory attention is the process of allocating one's cognitive capacity on a specific auditory stimulus to the exclusion of other stimuli; this seems to be significantly affected by one's ability to localize sound (Dai et al., 2018). Moreover, selective attention seems biased by reward cues; that is, motivation is an important factor in directing attention to a particular sound (Asutay & Västfjäll, 2016).

Complex sound fields, such as those in classrooms (Gremp & Easterbrooks, 2018) include background noise and reverberation. These acoustic landscapes affect auditory attention and learning for all children, but more so for those with hearing or learning differences (Bhang et al., 2018). As degree of hearing loss increases, the beneficial effect of reduced noise on the speech envelope seems to decrease (Petersen et al., 2016). Better hearing imposes greater sensitivity to changes in the signal-to-noise ratio (Petersen et al., 2016). Restated, tracking of speech gets worse as the hearing loss becomes more severe. Adding to this issue is the finding that sentence complexity imposes additional demands on the listener (Wendt et al., 2016).

Ultimately, then, sustained selective auditory attention is important for optimal learning. This seems to be both reward-dependent and linked to degree of hearing loss, spatial acuity, and cognitive skills (e.g., resistance to distractors), as well as to the linguistic complexity of

explicit verbal direction, and subjective familiarity (Isbell et al., 2016; Tervaniemi et al., 2009; Wendt et al., 2016).

Effortful Listening and Contributing Factors

Listening is the active counterpart of passive hearing (Moore et al., 2020). The act of listening, aided or unaided, is an effort necessitating auditory attention and other mental resources to understand an auditory message (Gagné et al., 2017; McGarrigle et al., 2014). As evidenced neurobiologically, the more effort one expends in listening, the more one's cognitive skills (e.g., attention, working memory, and academic learning decrease) are taxed (Macpherson et al., 2019; Prodi et al., 2019; Roebuck et al., 2018). When auditory attention decreases, then greater effort is needed to listen, understand, and remember (Peelle, 2018). When one engages in *effortful listening*, one's auditory attention must be both focused and selective, deliberate and purposeful (Pichora-Fuller et al., 2017).

There are many factors that affect effortful listening and those include: (a) room reverberation and background noise which may or may not include music; (b) the listener's quality and levels of unaided and aided hearing as well as level of language comprehension; (c) contextual information within the primary auditory stimuli; (d) clarity of acoustic speech stimuli; and (e) the listener's mental resources (e.g., Dingemanse & Goedegebure, 2019; Mattys et al., 2019; Ohlenforst et al., 2017; Pejovic et al., 2020; Peng & Wang, 2019; Wagner et al., 2015). Researchers are investigating ways to improve speech perception and minimize listening effort (e.g. Barrett et al., 2020; Good et al., 2017; Pejovic, 2020). For example, music-based interventions are being investigated as one way to facilitate speech perception-in-noise, but effectiveness remains debatable (e.g., Akça et al., 2020; Alain et al., 2018; MacCutcheon et al., 2020; Yurgil et al., 2020).

Mental resources that affect one's auditory attention include such psychological issues as the listener's state of mind and mood as well as levels of expectation and motivation (Pichora-Fuller et al., 2017). The listener's processing speed (i.e., reaction time) and working memory are two critical cognitive skills; these mental resources also affect the degree and extent of success at effortful listening (Rudner, 2016). It is uncontested that cognitive capacities influence auditory perception. Noisy situations tend to increase the cognitive demands made of the listener, hence these situations necessitate greater listening effort except, perhaps, when the listener is provided with certain cues, such as those obtained via speech reading (Koelewijn et al., 2015; Newman et al., 2013; Picou et al., 2013).

Auditory Fatigue and Cognitive Capacities

There is substantial evidence that children who are DHH are at risk for difficulties in speech comprehension in adverse environments. Some listeners are unable or unwilling to sustain sufficiently high levels of effort, so they may experience *auditory fatigue* or extreme tiredness (Hornsby et al., 2016; Pichora-Fuller, 2017). This construct is complex and may be best defined by the person experiencing it; this is commonly described as a feeling, mood or state, or demonstrated as a decrement in physical or cognitive performance (Hornsby et al., 2017; Pichora-Fuller et al., 2017).

Relative to those with normal hearing, children who are DHH and have other learning differences must exert greater efforts in the act of listening; thus, when they require more cognitive resources for listening, they may be more prone to listening-related fatigue and irritability (McGarrigle et al., 2014; Taitelbaum-Swead et al., 2019; Werfel & Hendricks, 2016). Additionally, the degree of difficulty involved in understanding a speaker can determine the degree of age-related auditory fatigue experienced by listeners, and this is not necessarily predicted by degree of hearing loss (Alhanbali et al., 2017; Ward et al., 2017).

It is important to avoid making generalizations about effortful listening and listening-related fatigue, since many listener-related factors vary considerably across different situational landscapes (Hornsby et al., 2016). Although fatigue is less likely to occur among listeners with greater cognitive capacities, it potentially compromises classroom learning for all persons who are DHH (Bess et al., 2020; Bess & Hornsby, 2014). If auditory fatigue is severe or recurrent, it may cause undue stress and influence quality of life (Hornsby & Bess, 2016). Conversely, auditory fatigue may decrease with practice in listening over noise (Ayasse & Wingfield, 2020).

Auditory cognitive neuroscientists continue to expand our psychological and physiological knowledge about listening and listening-related issues in adverse listening situations. In doing so, they are paving the way for clinical audiologists to provide many types of signal processing algorithms for hearing device users (e.g., Bierer, 2017; Johnson, 2018). Perhaps, as a result of ongoing multidisciplinary research, hearing technology and interventions will become even more individualized for learners with varied cognitive capacities, thus reducing the current wide variability in developmental outcomes.

The Charge and Challenge: Families & Clinicians

ACN is a highly innovative, multidisciplinary and collaborative approach to the complex scientific challenge of hearing and hearing-related issues. Such an approach necessarily involves research scientists, policymakers, clinicians, and other stakeholders from diverse professions. As such, ACN warrants extensive cross-disciplinary communication and information technology to create a 21st century holistic management of hearing loss. This may require considerable adaptation from clinicians when some intervention strategies warrant modification. However, it will ultimately generate enormous opportunities for persons who are DHH (Dritsakis et al., 2019).

Families as well as LSL early intervention service providers, educators, speech-language pathologists, and audiologists are broadening their perspectives. Hearing, auditory learning, and spoken language are just part of the larger intervention process (Zatorre, 2007). New data

and technologies are informing a wider variety of device programming, assessment, and treatment options for families and their children who are DHH (e.g., Dai et al., 2018; Han et al., 2019). Ultimately, this implies greater potential management options that address the specific needs of children who are DHH.

Cultural differences, not discussed here, certainly contribute to the brain's complexity and how a person behaves, thinks, and feels. Psychology and neuroscience are broad and deep in that each involves many different branches having to do with the mind and behavior. These multi-layered disciplines have much to offer auditory-based clinicians working with families and their children who are deaf or hard of hearing (Pichora-Fuller, 2014). It has been proven that cultural, psychological, and neural processes are interwoven (Ambady & Bharueha, 2009; Edwards & Crocker, 2008; Han & Ma, 2014; Huang et al., 2019). Given that each human being represents a highly organized information processing system, it is imperative that clinicians adopt a *systems approach* in how interventions for families and their children who are DHH are viewed and offered (Faulkner & Pisoni, 2013; Rhoades, 2017).

However, scientific evidence is useless unless clinicians take up the charge by first understanding and then implementing the knowledge that has been synthesized and translated for ease in comprehension by all stakeholders (Cook & Odom, 2013). The assumption that clinicians will automatically implement evidence-based practices is shown to be faulty (Douglas et al., 2015; Odom et al., 2020). Therefore, the challenging task of modifying practices and strategies is largely dependent on *active drivers*—both from within organizational systems and data-driven clinician perspectives (Sugai & Horner, 2020).

Clinicians are no longer alone in providing auditory-based interventions and implementing strategies to improve developmental outcomes. The village has evolved to become a sprawling urban mass. It is imperative that all clinicians embrace the findings from other disciplines, including the many different branches of psychology and neuroscience. Translating their findings into workable strategies can serve to minimize the developmental vulnerabilities often experienced by families and their children who are DHH (Evans-Whipp et al., 2017). Vulnerabilities arise as a result of a mismatch between these families' characteristics and those of treatment providers (Sossauer et al., 2019). To minimize gaps between these families' needs and the means intended to meet them, flexibility in clinician application is critical.

Research findings that inform clinicians serving families and their children who are DHH cannot continue without the involvement and express approval of parents and other caregivers. It is critical that clinicians explicitly support researchers in the quest to better understand all those factors that work for or against children who are DHH. Ways in which parents and other caregivers as well as clinicians can assist in the multi-layered world of auditory cognitive neuroscience are listed in Figure 2.

Clinicians, parents, and other caregivers play a vital role in moving science and evidence-based practices forward. With greater participation in inter-disciplinary and cross-professional collaborative studies as well as greater flexibility in the application of scientific data to auditory-based intervention practices, the outcomes for children

Figure 2

Recommended Practical Steps for Clinicians and Caregivers

Some Practical Steps

Clinicians

1. **Actively support researchers in their quest to involve large numbers of families.**
2. **Encourage parents and other caregivers to express their opinions on matters involving policies, regulatory action, and the trajectories of future research.**
3. **Encourage parents and other caregivers to participate in surveys and other research-based studies that have been approved by such institutions as universities and their school districts.**
4. **Provide parents and other caregivers with appropriate informational counseling pertaining to the implications of peer-reviewed research studies as well as legislation and regulatory policies.**
5. **Provide parents and other caregivers with contact information pertaining to all above sources.**

Caregivers

1. **Document your child's progress in a "progress notebook" or journal that can be shared with the entire care team.**
2. **Participate in research study opportunities (including surveys) to contribute to future policy development and impact future service provision.**
3. **Be consistent in following recommendations provided for your child by your care team.**
4. **Communicate reports and progress as well as concerns with all care team members.**
5. **Encourage care team members to consistently communicate and share reports, clinic notes, and care plans.**

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Getting Started with Home Visits: Recommendations for Serving Families of Children who are Deaf or Hard of Hearing

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Abstract

The successful implementation of newborn hearing screening programs across the United States has facilitated timely diagnosis of hearing loss and referral to early intervention (EI) services for families of children who are deaf or hard of hearing (DHH), thus increasing the potential for improved language development outcomes. As new parents engage in EI services that involve professionals entering their home, the effectiveness of the early interventionists' engagement, knowledge, coaching skills, and ability to provide emotional support can substantially influence families' experiences. This article provides graduate students and new early interventionists an overview of key concepts related to home-based EI services, including (a) establishing the parent-professional partnership, (b) developing the parent coaching model, (c) setting auditory development priorities, and (d) providing goal-oriented services. Tables containing websites, assessments, and other materials and intervention resources are provided to support content depth and service delivery competence in each concept area. The final section outlines the flow of a typical home visit. An example of a completed Family Session Planning Guide and a hypothetical example of dialogue between the parents and the EI provider as they establish the child and family goals and identify strategies for meeting those goals is provided. Also included is a Family Session Planning Guide template.

Acronyms: CDC = Centers for Disease Control and Prevention; DEC = Division for Early Childhood; DHH = deaf or hard of hearing; ECTA = Early Childhood Technical Assistance Center; EI = early intervention; LSL = listening and spoken language; NAEYC = National Association for the Education of Young Children

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Congenital hearing loss affects approximately two to three infants per 1000 live births in the United States and, if undetected or untreated, can result in delayed speech and language development (Centers for Disease Control and Prevention, 2010). However, the successful implementation of newborn hearing screening programs across the United States has facilitated timely diagnosis of hearing loss and referral to early intervention (EI) services for families of children who are deaf or hard of hearing (DHH), thus increasing the potential for improved language development outcomes (Nittrouer & Burton, 2001; Schramm et al., 2010). Children who are identified early and promptly begin EI services have better language skills compared with children who were later-identified or who did not engage in effective EI services (Ching et al., 2017; Decker & Vallotton, 2016). With advances in hearing technology, such as hearing aids and cochlear implants, and appropriate EI services, many young children who are DHH can develop listening and spoken language (LSL) similar to their same-aged hearing peers (Cole & Flexer, 2015; Hayes et al., 2009; Lederberg et al., 2013; Tomblin et al., 2015). Because more than 90% of babies who are DHH are born to hearing parents (Mitchell & Karchmer, 2004), hearing loss is unfamiliar to most new parents and the process of preparing for and effectively participating in EI services can seem daunting. In the United States, EI is most commonly defined as the period between birth and age three, as indicated under Part C of the Individuals with Disabilities Education Act (2004). Consistent with the priorities of EI best practices, services should be provided in the child and family's natural environment (Division for Early Childhood/National Association for the Education of Young Children [DEC/NAEYC] Joint Position Statement, 2009), which is most commonly in the home. In a longitudinal study of 122 EI professionals and 131 parents, Harrison et al. (2016) found that family involvement is highest when EI services are home-based, supporting the need for EI in the home whenever possible. See Nicholson et al., 2016 for a comprehensive review of home-visit models.

As new parents embark on this unexpected journey of EI services and having professionals enter their home, the effectiveness of the early interventionists' engagement, knowledge, coaching skills, and ability to provide emotional support can substantially influence families' experiences. In a parent survey, Ealy (2013) reported the EI provider was considered the most influential practitioner and is in the best position to influence the EI experience. In a

study exploring parent engagement in EI, Weiber (2015) reported findings from detailed interviews with 10 parents of children with hearing loss. This study highlighted some of the questions parents have when they first learn about EI services. For example, one parent stated, "I didn't even know prior to this that this thing [EI] even existed.… I didn't even know what it was" (p. 131). Another parent stated, "The first thought that came through my mind was a strange person would be coming to my house once a week" (p. 127). During the process of initiating EI services, one parent stated, "I had a really, really wonderful lady come to my house. We sat and had a long conversation. She told me her personal story and it gave me hope. I walked away from that conversation with the hope that at some point I will be able to communicate with her and she, in turn, would be able to communicate with me. So that's kind of where the ball got rolling" (p. 102). Other parents described the desire for their EI provider to be a better listener or the concern their provider may not have conveyed the full range of service delivery options available to them. However, most parents described their EI provider as "being knowledgeable," "a tremendous professional," "providing valuable one-on-one services," and "being a friend" (Weiber, 2015).

An EI provider who seeks to connect with families in a manner most comfortable and culturally appropriate for the family can become a trusted and valuable companion for families as they engage in EI programs and services (Division for Early Childhood [DEC], 2014). The purpose of this article is to provide graduate students and new early interventionists entering the LSL field an overview of key concepts related to home-based EI services, including (a) establishing the parent-professional partnership, (b) developing the parent coaching model, (c) setting auditory development priorities, and (d) providing goal-oriented services. Tables containing websites, assessments, and other materials and intervention resources are provided to support content depth and service delivery competence in each concept area.[1] In the final section, the flow of a typical home visit is provided, including an example of a completed Family Session Planning Guide. This is accompanied by a script that provides a hypothetical example of dialogue between the parents and the EI provider as they establish the child and family goals and identify strategies for meeting those goals. Also included is a Family Session Planning Guide template.

Establishing the Parent-Professional Partnership

An essential priority when serving young children who are DHH in EI is establishing a strong connection and partnership with parents, caregivers, and families.[2] The partnership must be founded on trust and assurance that the EI provider will take the time to learn the parents' priorities for their child and to understand what is important to them and their family (DesJardin, 2009; DEC, 2014; Moeller et al., 2013). The importance of providers developing a trusting relationship with families is recognized as a priority by the Early Childhood Technical Assistance Center (ECTA). The ECTA center is funded by a cooperative agreement with the Department of Education's Office of Special Education Programs and provides technical assistance to state EI agencies to develop high quality EI and preschool special education systems. In partnership with The Center for IDEA Early Childhood Data Systems (DaSy), the ECTA center developed an interactive, four-part web broadcast series aimed at helping EI providers to develop trusting relationships with families (ECTA, 2017). In the broadcast series, the ECTA center emphasizes that the parent-professional partnership lays the foundation for achieving the long-term intended outcomes for the children they serve and provides evidence-based information and materials to support practices that develop parent-professional trust. In addition to the recorded series, written materials and resources are provided.

The initial realization that a hearing loss may be present and the subsequent process of obtaining or confirming the diagnosis is, for most families, a difficult and emotional journey (Scarinci et al., 2018). Professionals involved in this process can contribute to families' experiences both positively and negatively. In a survey study of 445 caregivers of children who are DHH, Scarinci et al. found that approximately 85% of their sample reported they were satisfied with the emotional support and information they received from their providers following their child's hearing loss diagnosis. However, in a follow-up qualitative phase of the study, Scarinci et al. found through in-depth interviews with five families that the diagnostic process, interactions with audiologists, and initiation of EI services was a difficult and emotional experience for parents. In some instances, parents were hurt or confused by the comments of professionals or the manner in which information was provided, however inadvertent. Although most professionals provide caring support and guidance, it is important to ensure implementation of practices that develop trust and that are mindful of parents' needs. Providers should identify practices and behaviors that will minimize the potential for miscommunication and provide the information or supports that align with parents' priorities. As EI services are initiated, providers can help parents understand the construct and purpose of the home visit. EI providers are guides to support parents in skills that can promote their child's language and communication development, whereas the parents are the most important teachers and agents of change for their children as they implement strategies for development throughout the day and across daily routines (Decker & Vallotton, 2016; Roberts, 2019; Roberts et al., 2014; Moeller et al., 2013). Early Intervention providers can guide parents through discussion, coaching, counseling, and listening.

[1] The contents provided in the tables are not inclusive of all available websites, materials, or resources. Further, website URLs can change. Thus, the information provided is the most current at the time of publication.

[2] The definition of parents, caregivers, and families encompasses a rich variety of circumstances, cultures, and individually-specific details. To improve readability, the term "parents" is used throughout the article but is inclusive of all caregivers and family constructs.

The Parent Coaching Model

A central tenet of providing family-centered services is use of *parent coaching* as the service delivery model. The concept of coaching may elicit the image of a sports analogy, in which the coach is in charge of the team, identifies the goals, calls the plays, and motivates the players. However, the sports analogy in EI services does not hold and, in practice, is quite the opposite. An effective EI provider supports parents in meeting *their* goals for their child and family, with the provider offering content knowledge in research-based recommended practices and suggestions for implementing the goals within the families' daily routines in accordance with family needs and preferences. A working definition of the purpose of parent coaching in EI services is provided by Rush and Shelden (2019):

> Coaching is used to acknowledge and perhaps improve existing knowledge and practices, develop new skills, and promote continuous self-assessment and learning on the part of the coachee. The coach's role is to provide a supportive and encouraging environment in which the coach and coachee jointly examine and reflect on current practices, apply new skills and competencies with feedback, and problem solve challenging situations. The coach's ultimate goal is sustained performance in which the coachee has the competence and confidence to engage in self-reflection, self-correction, and the generalization of new skills and strategies to other situations as appropriate. (pp. 3–4)

A coaching model of interaction used in EI home visits requires both planning and flexibility on the part of the EI provider. Planning is essential for the coaching sessions to result in the coachee's desired learning processes or the achievement of a goal or outcome. The act of planning for each EI session can ensure the EI provider is purposeful in guiding developmentally appropriate targets in concert with priorities of the family. Likewise, the EI provider must also be flexible in the home visit plan and be prepared to make on-the-spot adjustments. For example, the provider may have planned to demonstrate joint awareness during book reading but, upon arriving, find the parents and child involved in planting flowers outside. The provider recognizes the rich language opportunities naturally occurring and can engage with the family in this activity. In all sessions, the priorities are determined by the parents, and then the coach can help to identify developmentally appropriate language and auditory perception targets. Together, the provider and the parents can brainstorm ways to implement or reinforce the targets during the family's daily routines and activities.

Reflective Questions

Through open-ended, reflective questions, providers can help parents and caregivers recognize *why* the targeted goals and recommended activities are important to their child's development (Rush & Shelden, 2019). Bruin & Ohna (2015) reported that not all parents understand the purpose of the activities or strategies their EI provider suggests, with one parent who stated, "We really didn't get it. We were supposed to use [it] in everyday situations, which became quite artificial, I felt. It's unnatural!" That artificial feeling happens when situations are contrived to meet strategies instead of strategies to fit everyday situations. When parents understand the purpose of the strategies, they are more effective at determining points in their natural routines where strategies will support their child's targeted goals. Reflective questions are open-ended questions used to drive discussion, review progress, introduce a new strategy, brainstorm ideas, plan for the future, and build the parent-professional relationship (Smith & Cook-Ward, 2020). For example, to build on a topic that was discussed during a previous visit, the EI provider may ask, "What do you remember about...?" By probing for the parent's current level of understanding in an open and non-judgmental way, the parent is more freely able to give an honest answer.

Specific Feedback

In addition to reflective questions, the thoughtful use of specific feedback can guide parents in their daily implementation of the strategies they are using to facilitate their child's progress and development. Parent-directed feedback should be encouraging, informative, and specific. For example, if a parent is reading a book to their child and uses acoustic highlighting, nonspecific feedback would be "I like the way you're reading to her! Great job!" This comment may be encouraging, but it is not particularly meaningful. In comparison, the provider could provide specific feedback, such as, "Using acoustic highlighting to emphasize certain words over others is keeping her engaged and promotes our goals of auditory perception development, great job!" Specific feedback can facilitate parent confidence and knowledge for using the LSL strategies throughout the child and family's daily routines and across a variety of environments.

Another form of specific feedback is to detail the connection between the actions the parent has taken and the child's demonstrated skills. In the same example of the parent reading to a child using acoustic highlighting, the provider could say, "When you used acoustic highlighting in the book, your child was able to notice our target word and find the corresponding picture in the book." Parents will recognize connections between strategies and outcomes as their coach shares his or her own observations.

Implementation of LSL Strategies During Daily Routines

As parents become adept at analyzing the outcomes of their strategies, they will be able to extend strategies into new daily routines. When intervention strategies are applied across normal daily activities, children are more likely to generalize the skills they are practicing (DEC, 2014). Early intervention providers can guide parents by using reflective questions to inquire about the child's participation in daily routines such as mealtimes, bedtime, bath time, getting ready for the day, going to the store,

and even family outings. For example, the provider might inquire about the activities the child most enjoys or how routines change from the week to the weekend. McGinnis (2017) suggests asking parents to write out their schedule to promote discussion that will help them identify effective strategies that target their child's developmental goals as aligned with the family's routines. In addition to speech and language, it is highly beneficial for parents to include music goals into their daily routines and interactions. Both listening to music and singing is fun and age-appropriate, but also promotes auditory perception development. Torppa et al. (2018) reported children who are DHH with cochlear implants who sing regularly have better perception of speech in noise compared to children who are DHH who don't sing. Implementation of auditory, speech, language, and music into daily living activities with specific feedback empowers parents to extend their understanding of their child's goals and how to use the LSL strategies in meaningful and age-appropriate ways.

Adult Learning Styles

To facilitate content knowledge and to help parents be reflective and have discussions that utilize feedback, effective coaches must have an understanding of adult learning and the ability to adapt to a variety of personalities and interpersonal communication styles. The partnership between the EI professional and parents will be more positive and successful when professionals, both verbally and through their body language, convey warmth, empathy, and a sincere desire for a meaningful connection with the family. This is more likely achieved when the provider has an understanding of adult learning styles in general and can specifically apply intervention strategies in harmony with the individual learning styles of parents.

Table 1

Websites and Written Materials to Support Parents and the Parent-Professional Partnership

Websites or Online Resources

Alexander Graham Bell Association
https://www.agbell.org

Baby Hearing
https://www.babyhearing.org/

Early Childhood Technical Assistance Center
https://ectacenter.org

EI Excellence
http://www.eiexcellence.org/

Family Guided Routines Based Intervention and Caregiver Coaching
http://fgrbi.fsu.edu/index.html

Hands and Voices
https://www.handsandvoices.org

Hear to Learn
www.heartolearn.org

Hearing First
www.hearingfirst.org

National Association for the Education of Young Children (NAEYC)
https://www.naeyc.org

National Center for Hearing Assessment and Management (NCHAM)
http://www.infanthearing.org/index.html

Parent Center Hub
https://parentcenterhub.org

Question Prompt List (QPL) (Phonak, 2017)
https://www.phonakpro.com/us/en/resources/counseling-tools/family-centered-care/fcc-children/family-centered-care-qpl.html

Supporting Success for Children with Hearing Loss
https://successforkidswithhearingloss.com

Vroom
https://www.vroom.org/

Zero to Three
https://zerotothree.org

Books and Materials

Agreed Upon Practices for Providing Early Intervention Services in Natural Environments
https://ectacenter.org/~pdfs/topics/families/AgreedUponPractices_FinalDraft2_01_08.pdf

Early Childhood Coaching Handbook, 2nd Ed (Rush & Shelden, 2019)
ISBN: 1681252562

Framework for Reflective Questions (Rush & Shelden, 2019)
https://fipp.org/static/media/uploads/casetools/casetool_vol4_no1.pdf

Routine-Based Early Intervention (Williams, 2010)
ISBN: 1598570625

Seven Key Principles: Looks Like/Doesn't Look Like (Ecta Center)
https://ectacenter.org/~pdfs/topics/families/Principles_LooksLike_DoesntLookLike3_11_08.pdf

Furthermore, professionals must learn to *meet parents where they are*. Parents and family dynamics are unique and their engagement with EI services can be influenced by a variety of factors. Early intervention providers who scaffold their support to match parent readiness provide a better EI experience for families in the short-term and long-term. See Table 1 for websites and written materials to support parents and the parent-professional partnership.

Auditory Development Priorities

Auditory Perception Development

A fundamental difference between serving children who are DHH and children with typical hearing is the accessibility of sound, essential for LSL development. The development of LSL in children who are DHH is founded on principles of early identification of hearing loss, use of appropriately-fitted hearing technology worn during all waking hours, and family-centered, goal-oriented EI services guided by professionals with expertise to meet the needs of children and families (Joint Committee on Infant Hearing [JCIH], 2019). A fundamental premise of speech and language development recognizes the critical window of language acquisition as a neurological emergency (Cole & Flexer, 2015; Livotsky, 2015). In other words, intervention to promote development of the auditory system in children who are DHH who use hearing technology should include systematic application and reinforcement of goals implemented during all waking hours through daily routines and across a variety of environments. Although age-appropriate acquisition of speech and language is a broad priority in many EI services, families of children who are DHH who wish for their child to use LSL will benefit from the guidance of a provider with expertise in LSL development. A provider's guidance can maximize the neurological foundations of auditory perception development. Auditory perception development is the maturation of the brain's ability to process and analyze sound, which requires consistent and meaningful stimulation as a foundation for LSL development (Abdollahi et al., 2017; Werner, 2007). It can be easier for parents to understand and be excited about their child's expressive language targets than the less familiar concept of receptive language and auditory perception development. The understanding and implementation of auditory perception goals may take more guidance and practice so they are not under-emphasized; thus, the importance of parents knowing *why* particular targets are being recommended.

Management of Hearing Technology

At the beginning of every session, providers should consistently prompt and reinforce the importance of parents checking their child's hearing technology (e.g., hearing aids, cochlear implants, or other assistive hearing devices). This ensures the child has functioning hearing technology during the session, but more importantly, it reinforces to parents the need for them to check their child's technology on a daily basis. This also provides an opportunity to troubleshoot any concerns and to answer any questions the parents may have about their child's devices. In a parent-coaching model, the parents perform the listening check with the provider there for support or guidance as needed. A daily listening check is also an opportunity to support parents' understanding and use of the Ling 6/7 sound test (/ah/, /oo/, /ee/, /sh/, /ss/, /mm/, and no sound). Some parents may have confusion between the *learning to listen* sounds that are often included in intervention activities (e.g., "mmmm, I like ice cream" or "shh, the baby is sleeping") with the Ling 6/7 sound test for the purpose of checking the hearing technology. These clarifications are important components of the coaching guidance. The EI provider helps parents know and recognize the type of response from the child that is developmentally appropriate at each stage. This guidance is an opportunity for parents to become more familiar with the development of auditory skills in a hierarchical progression.

Goal-Oriented Services

Assessment and Progress Monitoring

A primary role of the EI provider is to obtain accurate baseline data to establish the child's present levels of performance in their LSL development and then use ongoing assessment data to monitor child progress. Both formal and informal measures can be used to assess young children who are DHH, such as checklists, norm-referenced tests, and language sampling (Neuss et al., 2013; Thomas & Marvin, 2016). Checklists are a common form of assessment for children birth to three years due to their ease of use, although professionals should be mindful of the limitations of checklists given their lack of standardization and potential constraints in their specificity for documenting progress. Most professionals concur that checklists are most valuable when used in conjunction with other measures, such as norm-referenced tests that provide measures of development as compared with a standardization sample of same-aged peers. In fact, use of norm-referenced assessments is specifically recommended by JCIH for baseline and progress monitoring (JCIH, 2019). Use of language samples can also be an effective tool for monitoring both speech and language progress. Providers can track generalization of vocabulary and articulation by transcribing a child's use of their words and word approximations during sessions. Parents can also support data gathering by using video recordings. Video allows providers to observe language use during daily routines that occur when the provider is not present.

The IDEA Part C requires documentation of progress monitoring at least every 6 months (IDEA, 2004). Although the minimum requirements for progress monitoring must be satisfied, the frequency of monitoring should be informed by specific outcomes being targeted and the need for making decisions, rather than the minimum timeline recommendations (Thomas & Marvin, 2016). Overall, the intervention plan and the guidance provided to parents should be data driven to avoid ineffective home visit sessions or misguided goals and targets.

Goal Setting

In a collaborative process, the EI provider and the parents use the assessment data and the developmental hierarchy to identify the speech, language, and auditory perception goals to be implemented until the next home visit. Through discussion and open-ended questions, the provider can offer guidance to combine appropriate goal selection and family priorities (Kahn et al., 2009; Rush & Shelden, 2019). The provider can then formulate the specific wording of the goal that matches the family and child's needs. Early intervention providers who may be tempted to dictate this for the parents fail to realize that the implementation of goals throughout the child's day and during naturally-occurring routines are most effective when the parents are involved in the selection.

As parents consider the family's activities for an upcoming week, the provider and the parents can brainstorm ideas for incorporating the identified goals into those activities in meaningful ways. These may be typical activities such as mealtime or getting dressed, or less-frequent activities such as going camping or an upcoming birthday party. In other words, helping parents to identify the rich language opportunities that are happening all around them can facilitate their consistency and comfort with incorporating their child's goals during nearly any activity. Further, it is ideal when the reinforcement of goals involves the whole family. Through discussion, the provider and parents can identify ways for siblings, grandparents, or others to be involved in supporting the child's LSL goals through natural interactions. See Table 2 for developmental hierarchy guides and checklists, standardized assessments and screening tools, and intervention apps and materials.

Table 2

Developmental Hierarchy Guides and Checklists, Standardized Assessments and Screening Tools, and Intervention Apps and Materials

Developmental Hierarchy Guides and Checklists

Auditory Learning Guide
https://hearingfirst.org (free log-in required)

Auditory Skills Checklist
https://successforkidswithhearingloss.com/wp-content/uploads/2011/12/Auditory-Skills-Checklist-Cincinatti-Childrens-Hosp.pdf

Centers for Disease Control and Prevention - Developmental Milestones
https://www.cdc.gov/ncbddd/actearly/milestones/index.html

Cochlear Integrated Scales of Development
https://www.cochlear.com/us/en/professionals/resources/school-resource-center/rehabilitation-resources/integrated-scales-development

Cottage Acquisition Scales for Listening, Language, and Speech
https://edproducts.sunshinecottage.org/

Early Listening Function
https://successforkidswithhearingloss.com/wp-content/uploads/2011/08/ELF-Oticon-version.pdf

Functional Auditory Performance Index (FAPI)
http://www.tsbvi.edu/attachments/FunctionalAuditoryPerformanceIndicators.pdf

Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS)
https://advancedbionics.com/content/dam/advancedbionics/Documents/libraries/Tools-for-Schools/Educational_Support/Assessment-Tools/ITMAIS-ResourceBrochure.pdf

LittleEARS Auditory Questionnaire
https://www.medel.com/about-hearing/hearing-test/little-ears-auditory-questionnaire

Rosetti Infant-Toddler Language Scale
https://www.therapro.com/Browse-Category/Comprehensive-Language/The-Rossetti-Infant-Toddler-Language-Scale_2.html

Standardized Assessments or Screening Tools

Ages & Stages Questionnaires®, Third Edition (ASQ®-3)
https://agesandstages.com/products-pricing/asq3/

Assessment, Evaluation, and Programming System for Infants and Children, 2nd Ed
https://brookespublishing.com/product/aeps/

MacArthur-Bates Communicative Development Inventories (MB-CDIs)
https://mb-cdi.stanford.edu/

Preschool Language Scales, 5th Ed (PLS-5)
https://www.pearsonassessments.com/store/usassessments/en/Store/Professional-Assessments/Speech-%26-Language/Preschool-Language-Scales-%7C-Fifth-Edition/p/100000233.html

Intervention Apps and Materials

Advanced Bionics - Intervention Apps and Materials
https://advancedbionics.com/in/en/home/support/rehab.html

Cochlear Corporation - The Communication Corner
https://www.cochlear.com/us/communication-corner/program-selection/young-children-families.htm

Listening Room
https://thelisteningroom.com

Med-El - Intervention Apps and Materials
https://www.medel.com/support/rehab/rehabilitation

Components of Home-Visit Sessions

The specific components or flow of the home visit will vary depending on the individual needs and circumstances of the family. For example, some families may be navigating a variety of medical appointments or home visit services from other professionals, particularly if their child has health concerns or additional disabilities. There can be cultural factors as to how home visits are constructed, who is present, or how goals are developed and implemented. Financial worries about meeting basic family needs (e.g., having enough food, daycare costs, paying the rent or mortgage and other monthly bills) can influence parents' ability to focus on their child's hearing-related priorities. It is common for many parents to make substantial adjustments and sacrifices to their work and family routines to meet the needs of their children. For example, Bruin and Ohna (2015) reported one father who stated, "We did a lot. . . it takes a huge effort. I took time off work approximately one day a week for about a year." Another father reported, "We had to work many hours every single day and every single week to teach him to listen and speak, because he had to practice much more than normally hearing children." Overall, the challenges parents face in balancing work and community responsibilities, while also meeting the needs and schedules of other children in the family are substantial. Professionals can be more impactful in their services when they are cognizant of these realities and can effectively meet families where they are in their journey. Families are complex and the implementation of the home visit should be appropriately tailored for each family's unique needs and preferences. Keeping this in mind, home visits may be conceptualized into four segments: (a) greeting and family update, (b) prioritizing session targets, (c) implementation and practice, and (d) reflection and planning.

Greeting and Family Update

Consistent with the priority of developing, maintaining, and enhancing the relationship between the family and the EI provider, the home visit session should begin with inquiries as to how the parents and family are doing (Ekberg et al., 2018; Turan, 2010; Turan, 2012). Although this may seem obvious, most EI providers carry demanding caseloads and follow busy daily schedules. It can be easy to fall into a pattern of entering a home with a pre-determined priority and session plan. However, if the provider comes to the home on a day that has been particularly stressful or challenging for parents or if the provider is not in tune with the needs of the parents or child, this can set the stage for an unproductive, or even counter-productive session. Taking the time to make that initial inquiry can promote empathy, engagement, and positive dialogue between the parents and the provider. This personable interaction reinforces the relationship as one of care for each parent as an individual, and not just as parents of the child whom the EI provider is there to support. The beginning of the session is also an opportunity to get an update on child progress since the last session, to address questions or concerns the parents may have, and to celebrate the progress and accomplishments of the child and family.

Prioritizing Session Targets

The follow-up of the events and progress since the previous session can naturally lead to a discussion of that day's targets. An effective coach can implement the joint plan agreed on in the previous session, while also adapting to address needs a parent expresses in the current session. For example, one of the previous session targets may have included the child following a simple direction through listening with no visual or gestural cues. The provider intends to follow up on that target at the next session; however, upon arriving at the home, finds the parents very excited that their child has begun to put two words together. The parents express the priority of continuing to reinforce these new developments and want to spend substantial time in identifying ways to promote this skill. Rather than maintaining the session emphasis on listening to and following directions, the provider makes the adjustment to emphasize the parents' priorities regarding the two-word utterance, knowing the *following directions* goal can be revisited at a future session.

Implementation and Practice

The main portion of the home visit should consist of implementation and practice of skills that the parents can use to meet their priorities. Providers can model skills and coach parents to try the skills themselves. Modeling can be an effective form of instruction (Roberts et al., 2014), but professionals should be careful not to jump into the activity in a manner that parents could perceive as indicating they are not doing it well enough. This would be counter-productive to the coaching model and would be an unfortunate lost opportunity for parents to gain their own confidence and competence in effectively implementing the strategies. Expert modeling followed by guided practice gives parents an opportunity to analyze the strategy and receive feedback to support their use of the skills.

Reflection and Planning

At the end of the session, a period of reflection provides an opportunity for the parents and provider to blend their expert knowledge, perspectives, and observations to determine what is working well, address questions or challenges, and identify upcoming priorities. Using a Family Session Planning Guide, the parents and provider can collaboratively create a written plan for the upcoming week, with the provider ensuring all goals are developmentally appropriate. As parents' priorities and the associated goals are determined, strategies for implementation during the family's activities and routines can be discussed and identified. Use of the term *lesson plan* is intentionally avoided, as this term can imply a preconstructed plan developed by the provider that is to be closely followed. The term *Family Session Planning Guide* can promote the collaborative nature of the home visit, the parents' role in identifying session priorities, and the importance of flexibility in the EI session details. See Appendix A for an example of a completed Family Session Planning Guide. Appendix B provides a hypothetical example of dialogue between the parents and the EI provider as they establish the child and family goals and

identify strategies for meeting those goals. See Appendix C for a Family Session Planning Guide template.

Summary

Early interventionists who provide home visits for families of children who are DHH can support parents and caregivers in learning skills and strategies to promote their child's auditory perception and language development within their daily routines. Early interventionists should understand the breadth of LSL best practice recommendations that provide the foundations of their services and use the resources available to them to best support the children and families they serve.

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Appendix A
Family Session Planning Guide – Example

Family Session Planning Guide

Child: Jane
Age: 8 months
Date: 3/2/2020

Listening Check: Visual and Listening inspection was completed with the hearing aids. Both hearing aids sounded clear.

Present during session: Mom, Dad, and Jane

Target	Child/Family Update	Needs/Concerns	Reflection/Plan
Increasing Hearing Aid Wear time	The family reported that they are putting the hearing aids on right after Jane wakes up from the night or a nap, but she is pulling the hearing aids off frequently and putting them in her mouth.	How to get Jane to leave the hearing aids in her ears	Implement the "pat, pat, clap clap strategy," when they are sitting close to the baby during play.
Auditory attention to speech	The family has been working on joint attention. The parent reported that Jane has been looking at them while they are playing. Extension of joint attention is to work on auditory attention.	How to get Jane's attention	Use positioning when the baby is upset. Use auditory first while playing with the baby
Vocal turn taking	New target based on parent priority of Jane starting to use words. Create a space for Jane to respond by singing a song they love and stopping before the end of the song. This strategy where we wait for Jane to fill in the space we leave open is called auditory closure.	The parent wondered how long to wait, and how many times to try. Answer: Wait about 10 seconds and try about 3 times in a row before moving on to keep Jane from getting frustrated.	Reflection: The baby started using a sing-song vocalization when we stopped singing. Plan: The parents decided to sing songs with auditory closure before bed at night.

Created by Lauren Smith, MEd, Utah State University

Appendix B

Family Session Dialogue - Example (Reflective Questions marked with RQ)

Professional	Parents
Greeting and Family Update	
Hi Jenna. Hi Brad, it's so nice to see you guys. I am excited to hear about your practice this week.	Hi. We had lots of fun this week. Jane really liked playing together.
What did she do that helped you know she liked it? RQ	She was smiling at me and reaching for the toys.
It sounds like she really let you know that she liked the way you were playing with her.	Yeah it was fun. I felt like we really connected.
Prioritizing Session Targets	
Would it be all right if I shared with you another strategy and goal with you to use while you play?	Sure.
Before we jump into that, what other priorities did you have for our session today? RQ	We've been working really hard at keeping the hearing aids on. We put the hearing aids on right after Jane wakes up like we discussed last week, but I don't know if she is really wearing them more because she pulls them out and puts them in her mouth.
What have you already tried to help her leave her hearing aids on? RQ	We tell her no, and she stops for a minute, but then she sticks them back in her mouth.
What kinds of support from me would be helpful? RQ	I don't really know. What can we do?
We can try to teach Jane a replacement for pulling the hearing aids off or we can try one of the listening and spoken language strategies. Which one do you think would be more effective for your family? RQ	I think a replacement sounds better for us. I don't think she will stop no matter what we say.
Ok, So far today we're planning to add a new strategy to our play to work on Jane's listening skills. We are going to try a replacement behavior to keep Jane's hearing aids on. Last week we also talked about your goal of helping Jane use words to communicate with you. We can start working on that by teaching Jane when it is her turn to talk in communicating. So we have three things we want to target today. What would you like to begin with?	We're feeling really frustrated about the hearing aids, so can we talk about that first?
Implementation and Practice	
Absolutely. So you said you wanted to give Jane a replacement behavior. When we implement a replacement behavior we want to try and catch Jane before she grabs the hearing aid and give her something different to do. A replacement behavior other parents I've worked with before have used is to show Jane how to pat her hearing aids instead of pulling on them. So to teach Jane to pat her hearing aids, say "pat, pat" when she reaches for them, then show her what to do, then cheer for her. That way she is more likely to pull her hands back down to clap along before she can pull on the hearing aids.	Ok. I think we can do that.
If we want to try this out, when does she usually pull her hearing aids out? RQ	She just does it all the time.
OK, would you like to show her what to do now? Or wait for her to reach for them to try it?	Let's wait.
In the meantime, which of our other goals would you like to work on next?	Will you remind me what the other two are?

Appendix B (cont.)

Family Session Dialogue - Example (Reflective Questions marked with RQ)

We talked about working on listening skills and participating in a conversation.	I'm really excited for her to start talking. So let's do that one first.
The strategy we are going to use to help Jane know when it is her turn to talk to us is auditory closure. What do you already know about auditory closure? RQ	I don't know that one yet.
Auditory closure is when we say something Jane is familiar with, but we stop before the end. Today we can try it with a song that Jane knows already. Then before we finish the song we will stop and wait for Jane to say something.	Jane loves singing.
What song does Jane enjoy the most? RQ	She really loves singing "The Itsy-Bitsy Spider."
OK let's sing it to Jane. Since we are going to stop before the end. Let's sing everything except the last word which is "again".	(Both sing the song together stopping at the last word "again".)
Exactly, that is where we are going to stop. Can I give you another strategy to help Jane know it is her turn?	Sure.
I am going to sing the song to Jane again, but this time I am going to wait a little longer and I am going to raise my eyebrows at Jane to show her I am waiting for her.	(Professional sings "Itsy Bitsy Spider" waiting with an expectant look. This time Jane smiled and laughed.)
I really enjoyed her little laugh. As we try this a few more times, we will hope to hear her voice.	(Professional and parents sing the song again using auditory closure and an expectant look.)
Jane is reaching up for her hearing aids, so I'm going to get in her way so she can't grab them and show her what we want her to do instead. Pat pat Jane, Yay you did it! (Professional reached out and patted Jane's hearing aids right after saying "pat pat").	So, I'm supposed to do that every time she reaches for them? I will try that.
Reflection and Planning	
Realistically, when during the day would it make sense for you to use this strategy? RQ	Well, I don't think I can do this strategy when I'm cooking or doing laundry or those things.
What are the barriers for those times? RQ That may help us find a time that makes sense for your family.	Well I already have my hands full with other things at those times.
What times of the day would work better? RQ	What about just when I'm playing with her. Then I'm already sitting close by her and I'll be able to catch her before she pulls them off.
That sounds like a great plan. Is there any other support you need from me to practice this strategy with her? RQ	No, I don't think so.
Ok, now that we have a plan for the hearing aids, let's go back to our auditory closure. What have you seen working for Jane with this strategy? RQ	She is really interested, but she is still just laughing not talking.
What do you think about practicing this during the week and watching for changes? RQ	Yeah, I feel she is bored now, but another time she might do it for me.
When do you think it would work in your day to practice this? RQ	Well we already sing to her at night. Does it need to be separate from that? Or can we do it then?
Auditory closure works really well with a familiar routine, so that sounds perfect. What else do you need to practice this strategy? RQ	I think we just need to keep doing it.

Appendix B (cont.)

Family Session Dialogue - Example (Reflective Questions marked with RQ)

Implementation and Practice	
Let me know if any questions come up when you practice. Our last goal today was to help Jane learn to listen to our voices and to pay attention to what we say. While we are playing today, we can use *auditory first* to help Jane recognize that our voice is important. Auditory first is when we talk before we start playing or trying to get Jane's attention. For example, "Jane do you want to play with the rattle?" Then after I've said that, I'll pick up the rattle and hold it out to Jane. What questions do you have about using auditory first while you play? RQ	I'm not sure I have one. I just talk about what we are doing and then do it?
Yes, exactly. Why don't we see what Jane does as we keep trying it.	OK, Jane do you want to play with your ball? (The parent holds up the ball to the Jane.) Give me the ball Jane.
How do you feel it is going so far? RQ	How do I get her to pay attention to me? I feel like I'm just talking to myself.
Yes, right now she appears to be in her own world. One way we can help her pay more attention to what she is hearing is to make it easier for her to hear it. When we sit close to her and at her level that will help her hear better.	Ok, so I should be closer when I talk?
It is one strategy that we can try with her. How effective do you feel that strategy will be at getting Jane's attention? RQ	She likes it when I lay on my tummy when she is doing tummy time.
That's a great example of using positioning to get her attention. Can we try it now?	(Parents moved closer to Jane). Let's play with the ball. (Jane looked over at the parent).
Wow, This time Jane looked over at you right when you spoke.	Yeah I guess she did. Jane, do you want your rattle? (Jane reached for the rattle and looked at mom).
Jane did it again. You used auditory first and she looked to you for more information. Do you think you can use this strategy during your week?	I am already talking a lot during the day. I think I can do this auditory first with almost everything.
Reflection and Planning	
That is very true. How will you know that you were successful with the strategy this week? RQ	I guess when she looks over just when she hears me instead of when I show her something.
Are there times when you need her to turn to just your voice? RQ	Sometimes she gets upset when I'm busy and I want her to calm down without having to pick her up.
How could the strategy of moving closer to her help when she is upset? RQ	Well she cries really loud. So maybe right now she can't hear me over herself. So maybe when she cries I should go closer.
I'd love to hear how that goes this week. Our time is almost up for today. What concerns would you like to address next time? RQ	I definitely want to talk about hearing aids again next week. Can we keep playing games where she practices talking?
Yes. Anything else I can help you with today? RQ	I want to make sure I know what to expect her to learn next.
Would it be helpful for me to bring the auditory learning guide and the Cochlear Scales of Development to look at together next week?	Yes, I think that would be helpful.
Ok, I'll see you next week.	Thank you, see you next week.

Appendix C
Family Session Planning Guide Template

Family Session Planning Guide			
Child: ***Age:*** ***Date:*** ***Listening Check:*** ***Present during session:***			
Target	***Child/Family Update***	***Needs/Concerns***	***Reflection/Plan***

Created by Lauren Smith, MEd, Utah State University

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Parent Perceptions of Person-Centered Care: A Randomized Controlled Trial of the Childhood Hearing Loss Question Prompt List for Parents

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Abstract

Objective: When children are identified with hearing loss, parents are often unsure about what they need to know. A Childhood Hearing Loss Question Prompt List for Parents (CHLQPL) was recently developed to help parents and providers address questions. This exploratory study investigated if parents who used the CHLQPL in their audiology appointment perceived their appointment as more person-centered than parents who received treatment as usual. Parent perceptions regarding use of the CHLQPL during the audiology appointment was also sought.

Design: Randomized control trial.

Study sample: Parents of children with permanent hearing loss ($N = 50$).

Results: There were no statistically significant differences found between the intervention and control groups in parent perception of person-centeredness. Parents who used the CHLQPL found it useful and would recommend its use to others.

Conclusions: Further research is needed to explore other factors and benefits of including the CHLQPL in supporting parents of children who are deaf or hard of hearing. Audiologists can incorporate the CHLQPL to facilitate communication on topics of importance to parents and to facilitate parent engagement in a shared process.

Keywords: question prompt list; childhood hearing loss; parent

Acronyms: CHLQPL = Childhood Hearing Loss Question Prompt List for Parents; DHH = deaf or hard of hearing; PCC = person-centered care; QPL = question prompt list

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Pediatric hearing loss is one of the most common congenital conditions with approximately three infants identified with permanent hearing loss per every 1000 births (Centers for Disease Control and Prevention, 2017); however, the diagnosis is often unexpected as more than 90% of parents have typical hearing (Mitchell & Karchmer, 2006). Following hearing loss identification, parents must adjust to this information and navigate the intervention process to learn how to meet their child's needs. In healthcare, Question Prompt Lists (QPL) are often used to help patients consider questions to talk about with their provider and to facilitate their ability to raise issues that are on their mind related to the impact of the condition on their life. Recently, a QPL for permanent childhood hearing loss was developed for parents to support person-centered care (PCC) and focus on parents' immediate questions and concerns during audiology sessions (English et al., 2017).

PCC is applicable broadly in healthcare and reflects an approach that embraces a shared process, in contrast to the medical model of service delivery, and includes understanding and addressing client priorities within each session. PCC encourages patients to be active participants by creating an environment that respects their autonomy and supports a shared process (Grenness et al., 2014). Parents of children who are deaf or hard of hearing (DHH) often experience challenges adjusting to and managing needs related to their child's hearing loss, underscoring the need to address issues of importance to parents. For example, parents have reported wanting more information on a range of topics, including how to meet other parents of children who are deaf or hard of hearing, how to keep hearing aids on their child, how to obtain loaner hearing aids, and how to find financial assistance (Muñoz et al., 2016). As parents adjust to the diagnosis they may

experience a range of emotions including but not limited to grief (Kurtzer-White & Luterman, 2003), increased stress levels (Lederberg, 2002), feeling overwhelmed (Lesperance et al., 2018), and shock (Gilbey, 2010). PCC provides a holistic perspective rather than solely focusing on the health condition (Reynolds, 2009), and values active involvement in the treatment process that respects the family's beliefs (Kiwanuka et al., 2019). Through PCC, audiologists target support specific to each family's needs, based on their values, goals, challenges and barriers; thus, helping parents to more effectively meet the needs of their child.

QPLs have been used to aid communication between the patient, their family, and the health care provider. The Childhood Hearing Loss Question Prompt List (CHLQPL) was created by parents of children who are DHH and audiologists with the goal to promote PCC by having conversations on a broader range of topics of importance to parents (English et al., 2017). The CHLQPL provides a list of questions that families may indicate, thus empowering them to raise issues on their mind for inclusion in discussion during their appointment. Through an iterative process, 32 questions represented in four categories were identified for inclusion in the final version: 1) Our Child's Diagnosis; 2) Family Concerns; 3) Management of Devices; 4) Support Systems. The aim of the current exploratory study was two-fold. First, to explore if parents who used the CHLQPL in their audiology appointment perceived their appointment as more person-centered than parents who received treatment as usual. Second, to obtain feedback from parents on their use of the CHLQPL instrument.

Method

Participants and Procedures

Participants were recruited from two audiology clinics in the western and midwestern United States respectively. Study procedures were reviewed and approved by the corresponding institutional review boards. To be included in the study, parents were proficient in English, had no prior experience using the CHLQPL, and their child had been previously fitted with hearing technology. Parents were presented with a study flyer at the time of their scheduled audiology appointment (e.g., hearing monitoring or hearing device follow-up). Those who were interested signed a consent and were enrolled in the study. A sample size of 50 was determined a priori based on an effect size of $d = 0.3$, power of .85, and an alpha level of .05.

Participants were randomly assigned to either the CHLQPL or No CHLQPL condition. Random assignment was conducted using a random number generator with odd and even numbers representing each condition. Due to a communication error, one group began assignments by alternating participants into each group before using the random number generator, resulting in unequal group sizes. A total of 50 parents were enrolled, 22 were allocated to the intervention group and 28 to the control group (see Table 1 for participant demographic information).

Participants assigned to the CHLQPL condition were given a copy of the CHLQPL (available on the Phonak website) on the day of their appointment to review before seeing their audiologist. Audiologists were instructed to inquire about questions participants had from the CHLQPL and to facilitate discussion about parents' concerns using the CHLQPL as a springboard. Participants assigned to the No CHLQPL condition received treatment as usual. At the end of the appointment, participants completed the study survey. The CHLQPL condition survey contained items to obtain their feedback on use of the CHLQPL.

Measures

Basic Information Form

Demographic information (e.g., age, ethnicity, family income) on the parent and child, along with questions about the child's hearing loss and use of hearing technology was gathered using this measure (18 items). Two additional questions explored the extent parents agreed with statements on a six-point scale (1 = *strongly disagree* to 6 = *strongly agree*): (a) the audiologist wanted to know about my priorities for what I felt was important to talk about today, and (b) I had enough time to talk about my questions/concerns with the audiologist.

Parent Perceptions of Audiology Consultation (PPAC)

This is a post-consultation patient-centeredness questionnaire for doctor visits (Little et al., 2001), and was modified for the study with permission. Wording on the questionnaire was changed (i.e., doctor to audiologist; the problem to child's hearing; symptoms to concerns; illness to hearing difficulty) and section headings were modified (i.e., health to hearing; problem to hearing), so the instrument wording would be relevant for audiology services. The questionnaire assesses five aspects of the patient-centered model: communication and partnership (10 items), personal relationship (3 items), hearing promotion (2 items), positive and clear approach to hearing (3 items), and interest in effect on life (2 items). Items were rated from 1 (*very strongly disagree)* to 7 (*very strongly agree)*. This questionnaire has shown convergent validity and its subscales have good to excellent internal reliability (Little et al., 2001). Internal reliability for our sample was excellent (Cronbach's $\alpha = .98$).

Working Alliance Inventory – Short Revised (WAI-SR: Hatcher & Gillaspy, 2006)

The WAI-SR is a 12-item measure of therapeutic alliance (a core aspect of PCC) across three domains: (a) agreement on treatment tasks, (b) agreement on treatment goals, and (c) development of clinician-patient bond. The WAI-SR has demonstrated good to excellent internal reliability, stable factor structure, and convergent validity (Hatcher & Gillaspy, 2006; Munder et al., 2009). Items were rated from 1 to 5 with higher scores indicating stronger working alliance. Internal reliability in our sample was good (Cronbach's $\alpha = .89$). This measure was only administered to a subset of our sample ($n = 18$) due to its later inclusion (see Statistical Analysis section for detail).

Table 1
Child and Family Demographics

Questionnaire Items	QPL (*n* = 22)		No QPL (*n* = 28)	
	M(SD)	%(*n*)	*M(SD)*	%(*n*)
Child's current age (in months)	57(28.23)		45(32.07)	
Age hearing loss identified? (in months)	24(30.07)		18(31.47)	
Unilateral hearing loss		17%(4)		21%(6)
Bilateral hearing loss		78%(18)		79%(22)
Parent reported degree of hearing loss				
Mild		17%(4)		32%(9)
Moderate		57%(13)		46%(13)
Severe		9%(2)		11%(3)
Profound		13%(3)		11%(3)
Hearing technology				
Hearing aid		70%(16)		71%(20)
Cochlear implant		22%(5)		14%(4)
Bone anchored hearing aid		4%(1)		18%(5)
FM system (with hearing device)		30%(7)		3%(1)
Other		4%(1)		
Age fit with hearing technology (in months)	31(30.18)		24(31.31)	
Hours of device use*	9(2.80)		9(4.25)	
Additional disabilities				
Yes	39%(9)			39%(11)
No	52%(12)			61%(17)
Child's racial identification				
Asian	4%(1)			
Black	39%(9)			54%(15)
White	44%(10)			39%(11)
Multiracial	4%(1)			7%(2)
Other family members had a hearing loss since childhood	13%(3)			25%(7)
Primary caregiver's racial identification				
Asian				4%(1)
Black	4%(1)			%(2)
White	87%(20)			89%(25)
Primary caregiver's educational level				
Less than 7th grade				3%(1)
High school graduate				11%(3)
Partial college (at least one year)	4%(1)			7%(2)
College education	35%(8)			43%(12)
Graduate degree	48%(11)			36%(10)
Family annual income				
Less than $20,000				4%(1)
$21-40,000	9%(2)			14%(4)
$41-80,000	26%(6)			21%(6)
More than $81,000	48%(11)			57%(16)
Prefer not to answer	9%(2)			4%(1)

Note. QPL = Question Prompt List; *$n = 21$.

CHLQPL Use

The CHLQPL is a new measure and parent perceptions on use of the instrument has value and can inform audiologists considering incorporating the instrument in their practice. Participants in the CHLQPL condition were asked an additional 6 questions to obtain information on their perceptions, and they were asked to estimate duration spent discussing the CHLQPL in session. Five items measured use of the CHLQPL with item scores ranging from 1 (*strongly disagree)* to 5 *(strongly agree)*. Higher scores reflect more positive perceptions. For one item parents were asked to circle all that applied regarding use of the CHLQPL, with the stem "Using the QPL…" (i.e., was a comfortable experience; helped my discussion with the audiologist; seemed unnecessary; caused some anxiety for me; supported my understanding of my child's hearing loss).

Statistical Analysis

The IBM Statistical Package SPSS v25 was used for data analyses (IBM SPPSS, Statistics for Macintosh, Version 25.0). Descriptives (*e.g.,* means, standard deviations) were calculated for demographic variables and QPL feedback. Between-group comparisons (*t*-tests) were used to determine difference in outcomes of interest: PPAC and WAI-SR.

Preliminary *t*-test analyses ($n = 29$) revealed no differences between conditions on the PPAC ($M_{QPL} = 117.1$, $M_{No\ QPL} = 126.8$, $p = .309$). Because we wanted to examine if the PPAC lacked sensitivity to detect differences in our construct of interest, patient centeredness, or if the CHLQPL simply did not enhance patient centeredness, we later added the WAI-SR to the study.

Results

Parents reported information about their child's condition (see Table 1). There were differences in the demographic make-up between the groups. The children in the QPL group were older compared to the no QPL group ($d = .4$), and they received hearing technology later ($d = .23$). Over one-third of the children had additional disabilities (vision [QPL 26%; no QPL 7%]; intellectual [QPL 9%; no QPL 14%]; autism [QPL 4%; no QPL 4%]; syndromic [QPL 13%; no QPL 7%]; emotional/mental [QPL 9%; no QPL 0%]; physical [QPL 13%; no QPL 14%]; and other [QPL 13%; no QPL 14%]). Some families reported a history of childhood hearing loss (sibling [QPL 9%; no QPL 9%; parent [QPL 4%; no QPL 4%]; and other [QPL 9%; no QPL 9%]).

All parents were asked the extent they agreed with two statements on a six-point scale (1 = *strongly disagree* to 6 = *strongly agree*): (a) the audiologist wanted to know about my priorities for what I felt was important to talk about today, and (b) I had enough time to talk about my questions/concerns with the audiologist. The majority strongly agreed with both statements (a: [QPL 78%; no QPL 82%]; b: [QPL 86%; no QPL 89%]). One person strongly disagreed in the no QPL group that the audiologist wanted to know about their priorities.

Parent Perception Measures

Parents completed two questionnaires regarding their perception of working with the audiologist, the PPAC and the WAI-SR (see Table 2). An independent samples *t*-test was conducted to compare the QPL and no QPL conditions. There was no statistically significant difference between parent perceptions on the PPAC (total scale scores) in the QPL group compared to the no QPL group ($M_{QPL} = 124.09$, SD = 26.55; $M_{No\ QPL} = 124.07$, $SD = 11.97$); $t(49) = -.891$, $p = .101$. Parent responses on the WAI-SR (total scale scores) also revealed no statistically significant differences ($M_{QPL} = 628.8$, $SD = 472.1$; $M_{No\ QPL} = 695.5$, $SD = 449.1$); $t(49) = -.515$, $p = .322$. Results from the WAI-SR and the PPAC suggest that the parents who used the CHLQPL did not perceive their audiology session as more person-centered when compared to parents who did not use the CHLQPL.

Two additional questions were asked to evaluate parent perceptions of the interaction with their audiologists. First, parents were asked if the audiologist wanted to know about their priorities for the appointment. Second, parents were asked if they had enough time to talk about their questions or concerns with the audiologist. Results indicate that the majority of parents in both groups reported the audiologist was interested in their priorities (QPL 100%, $n = 23$; no QPL 96%, $n = 27$) and that they had enough time to address their concerns (QPL 96%, $n = 22$; no QPL 100%, $n = 28$).

CHLQPL Use

Parents assigned to the QPL condition completed the CHLQPL use questionnaire. Parents estimated the amount of time the audiologist spent talking with them about their questions on the CHLQPL. Thirty-five percent ($n = 8$) estimated more than 10 minutes, 26% ($n = 6$) 6–10 minutes, 35% ($n = 8$) less than 5 minutes, and 4% ($n = 1$) reported that questions on the CHLQPL were not discussed. Parents also rated their agreement (*strongly disagree* to *strongly agree*) on five questions regarding use of the CHLQPL. The majority of parents indicated they thought the CHLQPL was easy to understand (100%; $n = 23$), helpful (91%; $n = 21$), relevant (95%; $n = 22$), they would use it again (78%; $n = 18$), and would recommend its use to other families (96%; $n = 22$). Parents selected all that apply for "Using the QPL…" (i.e., was a comfortable experience [83%]; helped my discussion with the audiologist [72%]; seemed unnecessary [70%]; caused some anxiety for me [0%]; supported my understanding of my child's hearing loss [52%]).

Discussion

The purpose of this exploratory study was to investigate if use of the CHLQPL in audiology appointments increased parent perception of person-centeredness compared to treatment as usual, and the secondary purpose was to obtain parent perceptions on use of the CHLQPL. The findings revealed no statistically significant differences in parent perception of patient-centeredness between those who used the CHLQPL in their session and those who did not.

Table 2
Person-centered Measures

Questionnaire and items	QPL *M(SD)*	No QPL *M(SD)*	*t* (DF)	*p*
Parent Perceptions of the Audiology Consultation (PPAC)	*n = 23*	*n= 28*	-.891(49)	.101
Was interested in my worries about my child's hearing	6.48(1.34)	6.75(0.44)		
Was interested when I talked about my concerns**	6.43(1.34)	6.81(0.39)		
Was interested in what I wanted to know	6.48(1.34)	6.86(0.36)		
I felt encouraged to ask questions	6.43(1.34)	6.82(0.39)		
Was careful to explain information so I could understand	6.43(1.34)	6.86(0.36)		
Was sympathetic	6.35(1.34)	6.57(0.79)		
Interested in my thoughts about challenges experienced	6.35(1.34)	6.54(0.69)		
Discussed and agreed together what the problem was	6.30(1.36)	6.61(0.63)		
Was interested in what I wanted done	6.30(1.36)	6.57(0.69)		
Discussed and agreed on a plan for addressing challenges	6.17(1.47)	6.54(0.79)		
Knows me and understands me well	6.04(1.46)	5.93(1.25)		
Understands my emotional needs	6.00(1.48)	6.04(1.17)		
I'm confident the audiologist knows me and my history	6.00(1.45)	6.43(0.88)		
Discussed lowering risk of hearing difficulty for my child	5.96(1.61)	6.07(1.05)		
Discussed preventing future hearing difficulty for my child	5.83(1.61)	6.11(1.07)		
Explained clearly how my child is hearing*	6.09(1.48)	6.50(0.92)		
Was definite about intervention steps	6.22(1.45)	6.29(1.05)		
Was positive about how to monitor my child's hearing**	6.35(1.43)	6.37(0.97)		
Interested in effect of child's hearing loss on family life	6.04(1.49)	6.39(0.96)		
Was interested in the effect of my child's hearing loss on everyday activities	6.09(1.51)	6.50(0.75)		
Working Alliance Inventory-Short Revised (WAI-SR)	*n = 9*	*n= 9*	*-.512*(49)	.322
After sessions I am clearer as to how I might be able to change.	4.56(0.53)	4.11(1.69)		
Today's session gives me new ways of looking at my problem.	4.11(0.60)	3.89(1.83)		
I believe ___ likes me.	4.78(0.44)	4.78(0.44)		
___ and I collaborate on setting goals for my sessions.	4.67(0.50)	4.78(0.67)		
___ and I respect each other.	4.78(0.44)	4.78(0.67)		
___ and I are working towards mutually agreed upon goals.	4.78(0.44)	4.78(0.67)		
I feel that _____ appreciates me. ***	4.63(0.52)	4.44(1.13)		
___and I agree on what is important for me to work on.***	4.88(0.34)	4.89(0.33)		
___ cares about me even when I do things that he/she does not approve of. ***	4.63(0.52)	4.56(0.73)		
I feel that the things I do in sessions will help me to accomplish the changes that I want. ***	4.75(0.46)	4.56(0.88)		
___ and I have established a good understanding of the kind of changes that would be good for me. ***	4.75(0.46)	4.22(1.72)		
I believe the way we are working with my problem is correct.***	4.88(0.35)	4.89(0.33)		

Note. QPL = Question Prompt List; **n* = 22 for item; ***n* = 27 for item; ****n* = 8. For the PPAC, a higher score is consistent with greater perceived person-centered care. For the WAI-SR, a higher score is indicative of a stronger working alliance. Item ratings for the PPAC are on a 1 to 7 scale and items on the WAI-SR are on a 1 to 5 scale.

When interpreting the results, it is important to consider study limitations, including the participant population, the background of the audiologists, and the settings. The sample size was small, was not reflective of the population that makes up the United States (United States Census Bureau, 2018), and parents were recruited at the time of regularly scheduled hearing device monitoring appointments, not based on how recently their child received hearing devices. Furthermore, parents reviewed the questions at the time of their appointment, which may not have provided adequate time for parents to consider their questions. Additionally, the audiologists were experienced in pediatrics and they had established relationships with the participants. The influence of these factors on the results are not known; however, given this composition it is likely parents were more willing to ask their questions, regardless of group assignment. In addition, the study was completed at two settings, a University clinic and a Medical Clinic. The CHLQPL may enhance PCC in other environments and circumstances.

Including the CHLQPL may enhance PCC for audiologists less experienced or confident in working with the pediatric population, as it is a tool audiologists can easily incorporate into their practice to facilitate addressing questions of importance to parents. Furthermore, the CHLQPL can help parents consider questions they may not have thought to ask, prompting a more comprehensive discussion with their audiologist. The parents who used the CHLQPL indicated they would recommend its use to other parents.

A foundational aspect of PCC is understanding and addressing issues of importance through a shared process. This has been found in other areas of healthcare. In a study with cancer patients, 90% found the QPL helpful or useful in aiding communication (Clayton et al., 2007). In a review evaluating various QPLs, findings were mixed related to effectiveness to facilitate communication and encourage patient participation (Dimoska et al., 2008). For example, in the Clayton et al. (2007) study, 85% of respondents indicated the QPL encouraged them to ask more questions and 95% reported they felt the QPL made it easier to ask the physician questions, while in a larger study only 33% felt the QPL helped them ask more questions (Glynne-Jones et al., 2006). Sansoni and colleagues (2015) reviewed the use of QPLs in various health care settings and emphasized that although QPLs can aid communication, they do not replace effective communication or repair poor communication between the provider and patient.

Research in other areas of healthcare has found a range of benefits to using a QPL. For example, a study evaluating the use of a QPL for cardiac patients found that the QPL had a significant impact on patient anxiety. Researchers reported that the reduction in anxiety was likely due to better preparation for the appointment (Martinali et al., 2001). Other benefits of QPL use have included increases in the number of questions patients ask (Kinnersley et al., 2011), increase in patient knowledge (van der Meulen et al., 2008), and a significant increase in the amount of information provided to patients and their families (Brown et al., 2001; Little et al., 2001). Parents of children with attention-deficit/hyperactivity disorder reported use of a QPL helped them ask more questions, that it was helpful for use during the initial visit, and that it would continue to be useful at future follow-up appointments (Ahmed et al., 2017). Furthermore, the pediatricians in the study reported that parents were more likely to initiate discussion of difficult topics with the assistance of the QPL.

Research Implications

Further research is needed with the CHLQPL to better understand potential benefits for parents of children who are deaf or hard of hearing and to improve audiologists' understanding of when and how to use the CHLQPL in practice. For example, it would be beneficial to explore use of the CHLQPL in various clinical settings, with audiologists less familiar with the pediatric population, during transitions (e.g., transition out of early intervention), with parents of recently identified children or who are new to the practice, use with parents over time, and use by other professionals working with the family (e.g., early interventionists). Additionally, studies exploring providers' perceptions regarding addressing the broader range of topics included in the CHLQPL and how to navigate the discussion when they may feel less confident with certain topics would be useful. Comparing the total number of questions asked and the types of questions asked when the CHLQPL is used compared to when it is not used may offer additional insights.

Conclusion

The findings of this exploratory study revealed that there was not a statistically significant difference in parent perception of person-centeredness when parents used the Childhood Hearing Loss Question Prompt List (CHLQPL) compared to appointments when the CHLQPL was not used. Further research is needed to explore other factors and benefits of including the CHLQPL in supporting parents of children who are deaf or hard of hearing.

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Timeliness of EHDI Benchmarks in Infants with a NICU Admission Greater than Five Days: Analysis from a Retrospective Cohort

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Abstract

The purpose of this study was to examine the timeline of early hearing healthcare in infants with a history of lengthy (> 5 days) admission to a neonatal intensive care unit (NICU) compared to non-NICU peers. We compiled four years of state Early Hearing Detection and Intervention (EHDI) records from 156,335 infants using a statewide administrative database. We compared age at the time of newborn hearing screening, diagnostic audiological evaluation, and entry into early intervention in NICU infants and non-NICU infants. We also compared the proportion of NICU and non-NICU infants meeting prescriptive EHDI timing benchmarks based on the Joint Committee on Infant Hearing (2019) position statement. Results indicated that NICU infants experienced delayed newborn hearing screening and diagnostic evaluation compared to non-NICU peers and reached both benchmarks in lower proportions. NICU and non-NICU infants entered early intervention at equivalent ages and met the early intervention benchmark in similar proportions. Considering the important medical factors that drive lengthy NICU admissions, our results suggest that specific clinical guidelines for the timing of early hearing healthcare in NICU infants may be warranted.

Acronyms: ECMO = extracorporeal membrane oxygenation; EHDI = early hearing development and intervention; EI = early intervention; IDPH = Iowa Department of Public Health; JCIH = Joint Committee for Infant Hearing; LTFU/D = loss to follow up/documentation; NICU = neonatal intensive care unit; PCHL = permanent childhood hearing loss

Keywords: EHDI, pediatric audiology, screening, NICU

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The Joint Committee on Infant Hearing (JCIH) position statement is a broad clinical practice guideline for providers and policy-makers about the screening, diagnosis, medical management, intervention, and surveillance of infants with hearing loss (or infants at risk for developing hearing loss; JCIH, 2019). In the United States, individual state early hearing detection and intervention (EHDI) programs integrate JCIH recommendations at the state level. A key feature of EHDI program quality has been the establishment of a timeline for three primary benchmarks: hearing loss screen no later than one month of age, diagnosis no later than three months, and enrollment in early intervention no later than six months. In the most recent JCIH position statement published in 2019, the committee advocated for states that regularly meet the 1-3-6 timeline to now pursue a 1-2-3 timeline. In both cases, meeting timing recommendations may be more challenging for families when infants have additional medical needs in the newborn period and spend time in the neonatal intensive care unit (NICU).

NICU Trends and EHDI Status

Infants spend time in a NICU after delivery for a variety of reasons (e.g., low birth weight, preterm delivery). The rate of admission to the NICU increased 23% from 2007 to 2012 (Harrison & Goodman, 2015) and although long term survival for preterm infants has improved in the past 20 years, the likelihood of additional disabilities is high for preterm, low, and very-low birth weight infants (Chan et al., 2001; Kilbride et al., 2004; Stoll et al., 2015). Program planning for newborn hearing screening must account for an increasing burden of infants with a history of NICU stay.

Across NICU and non-NICU birth settings, hearing loss is the most common medical condition that is currently identified via newborn screening, at 16 infants per 10,000 live births (Williams et al., 2015). For comparison, recent incidence estimates for other serious congenital conditions in the United States were 14.85 cases of Trisomy 21 and 10.25 cases of cleft lip (with and without cleft palate) per 10,000 births (Mai et al., 2019). Other factors suggest that the congenital hearing loss rate of 16/10,000 may

underestimate the true number of infants who are born with developmentally significant hearing loss. Many newborn hearing screening programs experience high rates of loss to follow up or documentation (LTFU/D; or cases where the outcome of a failed screening cannot be confirmed). Across studies, the rates of LTFU/D for diagnostic audiological evaluation after the newborn hearing screening ranged from 9% to 41% (see review in Ravi et al., 2016). This rate does not account for infants who are born with slight and mild hearing loss and may not be detected with current screening approaches.

In NICU infants, incidence rates of hearing loss are higher than in non-NICU infants (Hille et al., 2007; Veen et al., 1993; White et al., 1994). In a 2007 study of early hearing outcomes in Dutch infants, 2.2% of study participants born at less than 32 weeks' gestation exhibited permanent childhood hearing loss (PCHL; van Dommelen et al., 2015). For comparison, similar population-level infant research on PCHL has revealed an overall rate of 0.16% (Williams et al., 2015). Younger gestational ages were associated with higher rates of hearing loss. Among the very earliest preterm births (24–25 weeks' gestation) the observed rate of hearing loss was 7.5% (van Dommelen et al., 2015). Xoinis et al. (2007) reported on both sensorineural hearing loss and auditory neuropathy spectrum disorder in NICU infants and found incidence rates of 2.2% and 0.56%, respectively.

There are many reasons for clinicians and researchers to have special concern regarding the early hearing healthcare of NICU infants. First, their risk of hearing loss is more acute. A NICU stay of greater than five days has been identified as a risk factor for late-onset hearing loss and is sufficient motivation for a follow-up hearing evaluation no later than 9 months of age (JCIH, 2019). Low APGAR scores are associated with both the need for NICU admission (Chu, 2003; Weinberger et al., 2000) and increased risk of infant hearing loss (Hille et al., 2007; Vohr et al., 2000). Infants in NICUs routinely require medical interventions that are associated with increased risk of permanent hearing loss including broad-spectrum IV antibiotics, mechanical ventilation, and extracorporeal membrane oxygenation (ECMO; Coenraad et al., 2010). Second, NICU parents may balance competing health priorities during the neonatal period. Using qualitative research methods with parents of infants with auditory neuropathy spectrum disorder, researchers found that hearing status was a low priority at the point of diagnosis amidst more urgent medical needs in the newborn period (Uus, 2012). Third, many NICU infants who pass the newborn hearing screening before discharge have risk factors that put them at significant risk for developing hearing loss (Dumanch et al., 2017).

Older age at newborn hearing screening has been associated with late follow up and incomplete audiological diagnosis among low birth weight and normal weight infants (Tran et al., 2016). Measuring EHDI follow up in NICU infants is challenging due to their heterogeneous health and developmental outcomes, and there are mixed findings about the impact of NICU status on audiological follow-up. Awad and colleagues (2019) reported ages at diagnosis and hearing aid fitting for ten NICU infants in their analysis of adherence to JCIH benchmarks among infants with bilateral hearing loss in a large metropolitan children's hospital. Of the nine surviving infants, four were diagnosed and fit with hearing aids beyond the 1-3-6 timeline in unadjusted age. However, among their collapsed study cohort of children with PCHL, NICU stay was not associated with an increased risk of delays between diagnosis and hearing aid fitting or age at diagnostic assessment. They did not report the timing of JCIH benchmarks for NICU infants who were ultimately diagnosed as normal hearing after not having passed the newborn hearing screening. In Crouch et al. (2017), investigators found that although low birth weight infants with hearing loss were less likely to access early diagnostic services, they were more likely to be enrolled in early intervention. They did not report the NICU status of their sample, however, we expect that many were NICU graduates based on their low birth weight.

In other studies, NICU status was associated with greater challenges meeting the recommended EHDI timeline. High intensity of neonatal care needs has been associated with lower rates of follow up for diagnostic testing at 3 and 6 months of age (Deem et al., 2012). In that analysis of quality metrics in the Buffalo, New York area newborn hearing screening programs, the highest observed rates of LTFU/D occurred in the region's only level IIIB (more acute) nursery. Others have found that a NICU stay does not contribute to increased risk of LTFU/D among infants who do not pass the initial screening (Spivak et al., 2009). Lieu and colleagues (2006) showed that although follow up in NICU infants has improved over time, it falls behind the recommended EHDI timeline. That investigation followed NICU infants who did not pass the newborn hearing screening between 1999–2002. Researchers followed families for up to four years after a failed newborn hearing screening, but they did not report the timing of follow up services. The authors classified children as having received follow up if parents reported that a hearing evaluation took place at any point in the intervening years, and did not report the timing of follow up.

The challenges that a long-term NICU stay poses for accessing early hearing services on time (diagnosis, fitting of appropriate technology, and enrollment in EI) have not been well characterized in a population-level group of infants. Given the increased risk for hearing loss in this group and the barriers that NICU infants may face, an important first step is to identify practice patterns related to the timing of their early hearing care. Significant public health resources are allocated to EHDI tracking and data management systems and these systems have been identified as the strongest tool to improve rates of follow up (Ravi et al., 2016). The administrative dataset that EHDI tracking programs generate provides a valuable opportunity to assess program quality and ascertain if states are meeting the recommendations laid out in the JCIH (2019) position statement. In the present study, we

use state-level EHDI program data to examine hearing healthcare trajectories in NICU and non-NICU infants.

Research Questions

This study utilizes a large public health dataset to analyze the timeliness of EHDI benchmarks for infants in the state of Iowa between 2014–2017. It is motivated by the need to establish the baseline characteristics of service delivery to NICU infants in light of expected challenges to meeting benchmarks (e.g., later ages at discharge driving later ages at diagnosis and early intervention, competing health priorities). Infants who had lengthy admissions to a NICU (> 5 days) are compared with non-NICU peers. We designed our research question to make a comparison in terms of their absolute ages at each of three hearing benchmarks and with reference to exogeneous timing benchmarks prescribed by state and national EHDI programs. Our research addresses the following questions:

1. How does the timing of EHDI benchmarks in infants with lengthy NICU stays compare to the timing of EHDI benchmarks in non-NICU infants*? We hypothesize that NICU infants will achieve EHDI benchmarks at later ages than non-NICU peers.*
2. Do lower proportions of NICU infants meet EHDI timing benchmarks compared to non-NICU infants? *We hypothesize that a lower proportion of NICU infants will meet EHDI benchmarks by the recommended ages compared to non-NICU infants.*

Method

Iowa Department of Public Health EHDI Data

To complete this retrospective cohort study on EHDI timing benchmarks in NICU and non-NICU infants, we accessed newborn hearing screening and follow-up records from the state of Iowa gathered between 2014–2017. The Iowa EHDI program tracks screening and follow up using *e-Screener Plus*™ (eSP™) software developed by OZ Systems. As of August 2020, although Iowa has begun educating providers about the 1-2-3 EHDI timeline, its goal remains meeting the 1-3-6 timeline. All EHDI records were extracted from eSP by the Iowa Department of Public Health (IDPH) at the end of March 2019, de-identified, and shared via a secure data transfer. Table 1 lists the variables we extracted from individual records. Iowa's EHDI Coordinator shared the dates of enrollment in early intervention for a sub-set of infants with confirmed hearing loss and linked them with the eSP dataset prior to data transfer. This study was approved by the University of Iowa Institutional Review Board under a data-sharing agreement with IDPH. It was determined that this study did not meet the criteria to be considered human subjects research.

Table 1

List of Extracted Variables from the Oz Database for Infants Included in this Study

Date of Birth
Gender
Race/Ethnicity
City
State
Zip Code
Nursery (well-baby, NICU)
Place of Birth (Hospital/Home/Other)
Birthing Facility
Birth Screen Provider
Outpatient Screen Provider
Assessment Provider
Patient Outcome (e.g., deceased, moved out of state, complete in process)
Hearing Outcome (e.g., bilateral hearing loss complete, unilateral hearing loss-in process, normal hearing)
Birth Screen Date
Birth Screen Outcome (e.g., Bilateral Pass, Unilateral Pass)
Outpatient Screen Outcome (e.g., Bilateral Pass, Unilateral Pass)
Audiological Assessment Outcome (e.g., bilateral hearing loss complete, unilateral hearing loss-in process, normal hearing)
First Test Type
First Diagnostic Session Date
Right and Left Ear Outcomes (e.g., sensorineural, mixed, auditory neuropathy, normal)
Date of HL Confirmation
Date of Early Intervention referral
Risk Factors (e.g., Cranio-facial anomalies, transfusion for elevated bilirubin, assisted ventilation)
Family history of childhood hearing loss
NICU > 5 days
Assisted Ventilation
Bacterial or Viral Meningitis
Congenital CMV confirmed in baby

Note. CMV = cytomegalovirus; HL = hearing loss; NICU = neonatal intensive care unit.

NICU and Non-NICU groups

The initial dataset included records for 156,335 infants. We classified infant records according to their NICU status: Infants with a NICU stay greater than five days (NICU group, *n* = 8,149) and infants without lengthy NICU stays (non-NICU group, *n* = 143,888). Thus, the non-NICU group includes infants with very short NICU admissions in addition to infants with no NICU stay. Given the focus of this investigation on timing aspects, we did not expect shorter stays than 5 days to impact a family's ability to meet EHDI 1-3-6 goals. Iowa tracks infants with a NICU stay of greater than five days to monitor for delayed-onset hearing loss as recommended by JCIH (2019), and newborn hearing screeners check a separate box to indicate that an infant met this criteria. Therefore the five day cutoff was a reliable method for separating our groups. We approached incomplete records (for example, infants whose nursery was reported as the NICU but for whom the hospital screener did not include risk factors) in two ways. If risk factor information was missing, but newborn nursery location was reported as "Well-baby," infants were classified in the non-NICU group. If records were so incomplete that no determination could be made with relation to nursery status, we excluded those infants from further analysis. Table 2 provides demographic characteristics of both groups and sample sizes available during analysis for each of the EHDI benchmarks. Figure 1 illustrates how the data were reduced. If infants were classified as deceased, we did not include their records in any analyses.

Table 2

Summary Statistics and Demographic Characteristics in Infants with Lengthy Stays in the Neonatal Intensive Care Unit (NICU) and Infants without (Non-NICU)

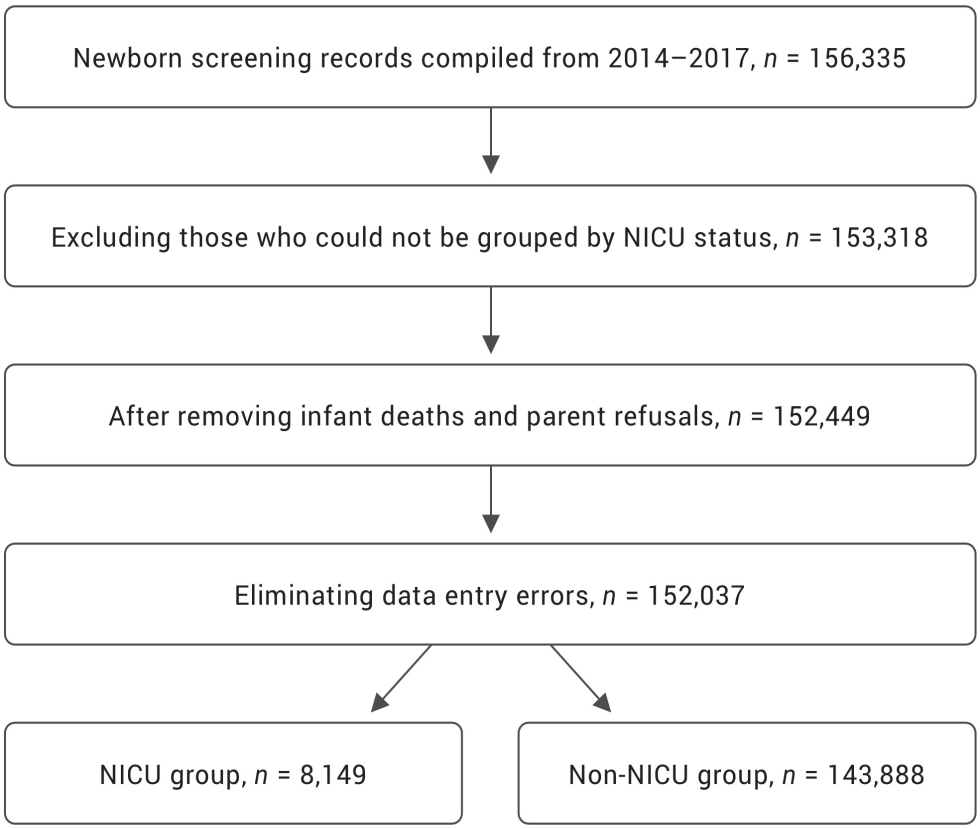
	Non-NICU	NICU	Between Group
Age at screen (days) Median Mean (*SD*) Range (*n*)	 1 1.75 (3.58) 0–249 (*n* = 143,888)	 11 22.01 (28.48) 0–353 (*n* = 8,149)	*p*-value < 0.001*
Age at diagnostic test (days) Median Mean (*SD*) Range (*n*)	 42 73 (85.89) 0–673 (*n* = 1,167)	 89 107.7 (79.59) 8–537 (*n* = 227)	*p*-value < 0.001*
Age at EI enrollment (days) Median Mean (*SD*) Range (*n*)	 118 173.5 (149.97) 35–749 (*n* = 111)	 155.5 188.58 (150.6) 54–629 (*n* = 38)	*p*-value = 0.6
Maternal race: White (*n*; %)	121,752; 84.6%	6,606: 81.1%	*p*-value < 0.001*
Maternal race: Black (*n*; %)	10,196; 7.1%	790; 9.7%	*p*-value < 0.001*
Race: Other/Multi-race (*n*; %)	11,940; 8.3%	753; 9.2%	*p*-value < 0.001*
Lost Contact (*n*; rate)	457; 0.3%		n/a
Moved out of state (*n*; rate)	203; 0.1%		n/a

Note. EI = early intervention

*indicates significance with alpha level = .05 level

To contribute to the larger body of research on hearing outcomes in NICU infants and characterize the representativeness of our dataset, we calculated group-specific incidence rates of hearing loss based on the full set of non-redacted data (with any length of NICU stay included in the NICU group, for incidence calculations only). We classified hearing losses as congenital if they were confirmed as a result of not having passed the newborn hearing screening. Across the four years examined here, the total incidence rate was found to be 1.91/1,000 births. Stratified by NICU status, the NICU-specific incidence rate was 5.27/1,000 births and the well-baby-specific incidence rate was 1.64/1,000 births.

Figure 1
Data Filtering for Each Benchmark from Full 2014–2017 Dataset



Data Analysis

For statistical analysis, we narrowed the four years of data by stage in the EHDI process. We included all infants for the screening benchmark analysis, only infants who did not pass the screening for the diagnostic benchmark analysis, and only infants with confirmed hearing loss for the early intervention benchmark analysis. We performed all data manipulation and analyses in R 2.14.0, using the *epitools*, *dplyr*, *lubridate*, and *ggplot2* packages for analysis and data visualization (Aragon, 2020; Grolemund & Wickham, 2011; Wickham, 2016; Wickham et al., 2020). We generated new variables to represent an infant's age (in days) at each of the primary EHDI benchmarks by comparing appointment dates with the dates of birth. Finally, we created dichotomous variables to classify study participants as having *met* or *not met* timing recommendations. For all analyses, a month was treated as 30 days, three months as 90 days, and six months as 180 days to remain consistent throughout the four years of data. For this study, the early intervention benchmark represented enrollment into IDEA Part C Early Intervention programs, not the date of hearing aid fitting.

For each of the three benchmarks, we first compared the un-adjusted ages at EHDI benchmarks using a Welch's adjusted *t*-test due to unequal variances between groups. We then performed a chi-squared test to assess proportions of each group that met specific EHDI timing benchmarks. Odds ratios and 95% confidence intervals were calculated to characterize the relationship between the exposure of interest (lengthy NICU admission) and the outcome of interest (successfully completing EHDI benchmarks on time).

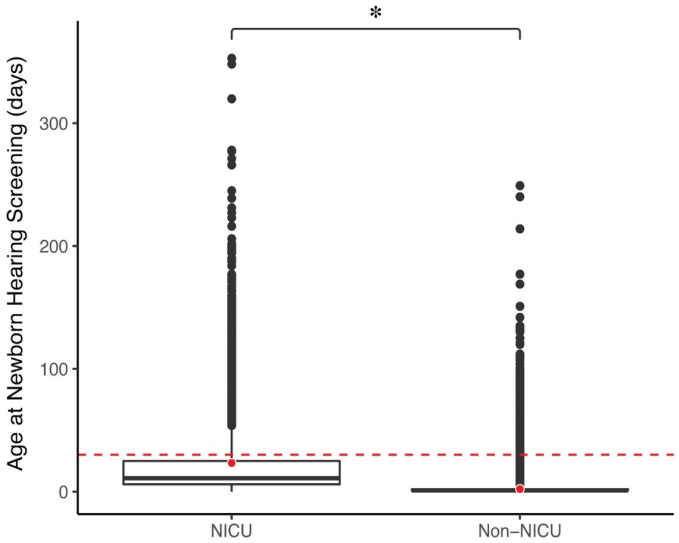
Results

Timing of EHDI Benchmarks in NICU Infants

Table 2 contains descriptive statistics for both groups, including the means, medians, standard deviations, and ranges of ages at each benchmark. Figures 2–4 show ages and distributions for both groups at each EHDI benchmark. On average, NICU infants received the initial screening at 22 days of life (compared to 1 day of life in non-NICU infants), had their first diagnostic assessment at 110 days (compared to 75 days), and enrolled in early intervention at 189 days (compared to 174 days). Although all infants in the NICU group were confirmed to have spent five or more days admitted, our data revealed that some infants in the NICU group received the newborn hearing screening on the first day of life. This could reflect late admission or re-admission to the NICU. We observed wide ranges for all three benchmarks across the full sample. NICU infants were significantly older at the time of hearing screening (p-value < 0.001) and diagnostic evaluation (p-value < 0.001) than non-NICU peers, but both groups enrolled in early intervention at equivalent ages. Fewer records were available for the early intervention benchmark due to both the lower numbers of confirmed hearing loss that required early intervention referral and incomplete records of referral for some cases of PCHL.

Figure 2

Age at EHDI Benchmark for Newborn Hearing Screening in Infants with Lengthy NICU Stay and Non-NICU Infants

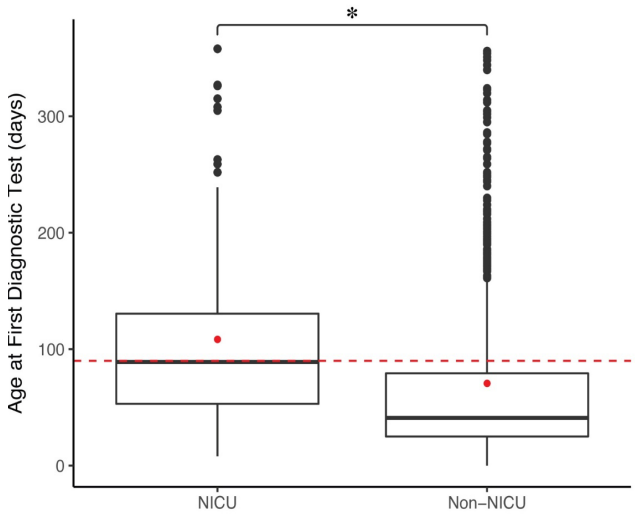


Note. Boxes show lower (Q1) and upper (Q3) quartiles and the median. Whiskers show data points within 1.5 times the interquartile range, and black circles show outliers. Means are plotted in red. For comparison, red hashed lines show the age recommendation in the Joint Committee on Infant Hearing (2019) position statement. EHDI = early hearing detection and intervention.

*indicates significance with alpha level = .05 level

Figure 3

Age at EHDI Benchmark of Diagnostic Evaluation for Infants with Lengthy NICU Stay and Non-NICU Infants

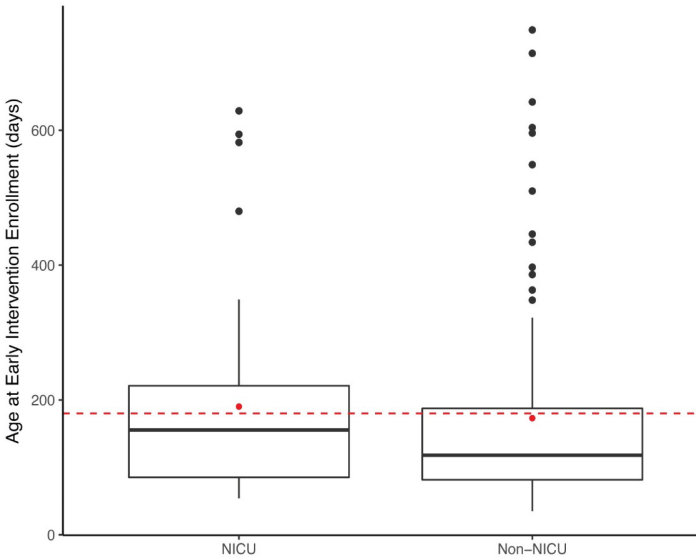


Note. Boxes show lower (Q1) and upper (Q3) quartiles and the median. Whiskers show data points within 1.5 times the interquartile range, and black circles show outliers. Means are plotted in red. For comparison, red hashed lines show the age recommendation in the Joint Committee on Infant Hearing (2019) position statement. EHDI = early hearing detection and intervention.

*indicates significance with alpha level = .05 level

Figure 4

Age at EHDI Benchmark of Enrollment in Early Intervention for Infants with Lengthy NICU Stay and Non-NICU Infants



Note. Boxes show lower (Q1) and upper (Q3) quartiles and the median. Whiskers show data points within 1.5 times the interquartile range, and black circles show outliers. Means are plotted in red. For comparison, red hashed lines show the age recommendation in the Joint Committee on Infant Hearing (2019) position statement. EHDI = early hearing detection and intervention.

Table 3

Odds Ratios and Confidence Intervals Associated with Missing EHDI Timing Benchmarks in Infants with Lengthy Stays in the Neonatal Intensive Care Unit (NICU) and Infants without (Non-NICU)

EHDI Benchmark	Group	Not Met Benchmark	Met Benchmark	Odds Ratio	Confidence Interval (95%)
Screening	NICU (*n* = 8,149)	1,623	6,526	96.47*	(85.9–108.3)
	Non-NICU (*n* = 143,888)	370	143,518		
Diagnosis	NICU (*n* = 227)	110	117	3.17*	(2.36–4.25)
	Non-NICU (*n* = 1,167)	267	900		
Early Intervention Enrollment	NICU (*n* = 38)	16	22	1.88	(0.87–4.04)
	Non-NICU (*n* = 111)	31	80		

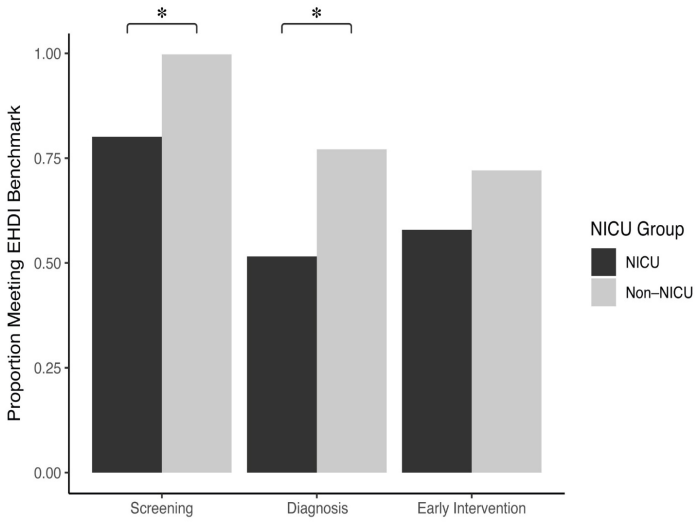
Note. EHDI = early hearing detection and intervention.
*indicates significance with alpha level = .05 level

Benchmark Attainment by NICU Group

Our second research goal was to compare the proportion of infants who met EHDI age recommendations for NICU compared to non-NICU infants. Table 3 presents these results, including odds ratios and confidence intervals.

Figure 5

Proportions Meeting Three EHDI benchmarks in Infants with Lengthy NICU Stays and Non-NICU Infants



Note. EHDI = early hearing detection and intervention.
*indicates significance with alpha level = .05 level

Odds ratios express the likelihood of missing the recommended EHDI timeline for infants with lengthy NICU stays compared to non-NICU infants. For newborn hearing screening by one month of age, the odds of delay in NICU infants was 96.47 times that of non-NICU infants (CI = 85.9–108.3). For diagnostic evaluation by three months of age, the odds of delay in NICU infants was 3.17 times that of non-NICU infants (CI = 2.36–4.25). Both these differences were significant at the alpha = .05 level. There was no significant difference in the likelihood of enrolling in early intervention on time. Figure 5 displays these results.

NICU-Related Delays by Maternal Race

Based on the differences in maternal race between our NICU and non-NICU groups (Table 2), we performed follow-up analyses with racially stratified data for screening and diagnostic benchmarks. Table 4 contains stratified odds of missing EHDI benchmarks in white, black, and other/multiracial NICU infants. Wide, overlapping 95% confidence intervals revealed no large differences in NICU-associated odds of missing either EHDI benchmark among white, black, and other/multiracial infants in our sample. We did not perform a stratified analysis by race for the early intervention benchmark due to low numbers of infants with data for this benchmark.

Table 4

Odds Ratios and Confidence Intervals Associated with Missing EHDI Timing Benchmarks in Infants with Lengthy Stays in the Neonatal Intensive Care Unit (NICU) and Infants Without (Non-NICU), Stratified by Maternal Race

		White		Black		Other/Multirace	
Screening		Not Met Benchmark	Met Benchmark	Not Met Benchmark	Met Benchmark	Not Met Benchmark	Met Benchmark
	NICU	1268	5252	187	603	168	671
	Non-NICU	300	120,221	30	10,166	40	13,131
		OR: 96.75* (CI: 85.06–110.05)		OR: 105.09* (CI: 70.86–155.86)		OR: 82.19* (CI: 57.72–117.04)	
Diagno.	NICU	77	98	17	9	16	10
	Non-NICU	206	730	27	66	34	104
		OR: 2.78* (CI:1.99–3.9)		OR: 4.62* (CI: 1.83–11.63)		OR: 4.89* (CI: 2.03–11.8)	

Note. EHDI = early hearing detection and intervention.
*indicates significance with alpha level = .05 level

Discussion

The findings of this study revealed that infants with a history of lengthy NICU stays access newborn hearing screening and diagnostic evaluation at later ages than non-NICU infants. Further, NICU infants met EHDI benchmarks for newborn hearing screening and diagnostic evaluation in lower proportions than non-NICU infants. On average, NICU infants were screened and seen for diagnostic assessment within the recommended age ranges; however, marked variability was present. This partly confirms the previous findings in Crouch et al. (2017). A discrepancy between the early benchmarks (screening and diagnostic evaluation) and the later enrollment in early intervention benchmark may result from NICU infants being referred for EI services for reasons other than PCHL. This would be consistent with clinical practice patterns observed for NICU graduates with preterm delivery and extremely low birth weight (Verma et al., 2003; Kuppala et al., 2012). However, due to the low number of infants whose enrollment in EI could be confirmed, we had lower power to detect true differences for this benchmark compared to screening and diagnosis benchmarks. Because of the nature of research with administrative data, we were not able to collect additional information that may reveal primary EI referral diagnosis. Thus, while we may find overall age at enrollment and proportions meeting the EHDI goal are equivalent among NICU and non-NICU infants, it remains important to ensure that children with PCHL receive services that address their

auditory and language development needs even in the presence of other qualifying diagnoses.

Our work demonstrates that infants with lengthy NICU stays do not achieve EHDI benchmarks at the same rate as their non-NICU peers. Failure to meet even one benchmark is associated with poorer long-term outcomes for children with PCHL, even if other benchmarks are met (Yoshinaga-Itano et al., 2017). However, this has not yet been examined in NICU infants alone. If delays are caused by lengthy NICU admissions, they may not lead to the same adverse effects on long-term outcomes as delays that stem from LTFU/D and clinical undermanagement.

A strength of this population-based study is that it incorporates the screening and outcomes of a large number of infants who were born in Iowa hospitals, regardless of hearing outcomes. Rather than excluding infants with normal hearing, we have used a winnowing treatment of the dataset. Thus, we were able to include benchmark timing data for the full population of Iowa infants who required care, even if they later went on to receive a diagnosis of normal hearing. A shorter time-to-diagnosis for children with normal hearing means fewer state public health resources tracking progress, shorter windows of parent concern, and an increased likelihood that diagnostic assessment can be completed under natural sleep. In addition, our work documents that although the NICU group defined in our analysis exhibited greater racial diversity than our non-NICU group, the relationship between lengthy NICU admission and risk of missing EHDI benchmarks appeared consistent across racial categories.

Limitations

The results from the first research question were meant to be descriptive in nature and capture the current clinical practice patterns regarding the timing of clinical activities. Our dichotomous categorization strategy pooled the data from infants with any length of NICU stay beyond five days and was not sensitive to discrepencies between intermediate term NICU stays and extended NICU stays. A major limitation of this investigation is the lack of access to gestational age that could be matched with infants in our two groups. Without gestational age, we are not able to characterize delays in NICU infants that stem from prematurity alone compared to infants with complex medical needs. Although the findings explored here are essential to characterize the current screening and follow-up timing trajectory for infants with lengthy NICU stays, a critical next step would be to consider delays in light of their gestationally adjusted age and comorbidities. Specific recommendations regarding gestational age adjustment would be a valuable addition to future JCIH position statements. Our analysis also excluded infants whose data concerning early benchmarks or NICU status could not be confirmed. These were the result of LTFU/D, incomplete data entry (such as missing information about risk factors), and parental withdrawal of consent to share detailed screening records with the IDPH.

A final limitation is that we calculated age at diagnosis using the first diagnostic assessment. Although we can safely assert that a confirmed diagnosis could not have preceded the first diagnostic appointment, we cannot exclude the possibility that this date represents a best-case scenario rather than a true age at confirmation of hearing loss. Holte et al. (2012) showed that, on average, families experienced delays between the initial diagnostic assessment and what they considered the confirmation of hearing loss. Recent EHDI literature suggests that some families go through up to five diagnostic evaluations before receiving a confident diagnosis of PCHL (Awad et al., 2019). If a transient conductive loss is suspected, the process of confirmation can be further delayed if families have long waits for ENT (Ear, Nose, and Throat specialist or otolaryngologist) appointments or if their physician prefers a wait-and-see approach for transient conductive loss. In the Outcomes of Children with Hearing Loss longitudinal study, parents reported reasons for delay included multiple re-screening, equivocal results, and protracted medical management (Holte et al., 2012; Walker et al., 2014). There is also the risk that results reported to EHDI as the first diagnostic assessment consist of repeated screening (i.e., OAEs only) instead of a true diagnostic evaluation. Concurrent quality checks at the IDPH during an overlapping period revealed that among children with hearing loss, in 87 of 299 cases the child's first evaluation with an audiologist consisted of a repeat screening despite being reported as a diagnostic evaluation (A. Hagerman, personal communication, August 12, 2020).

Future Directions

There are significant research opportunities in partnerships between researchers and state EHDI programs to improve service delivery in early hearing healthcare. Access to large public health databases of EHDI tracking results provides a unique opportunity to ask such questions and allows researchers to measure quality changes over time. Our work here examines one narrow piece of the JCIH clinical practice guideline. The data collected and tracked by state EHDI programs is rich with the level of detail necessary to examine other medical and audiological management patterns. Specific to NICU populations, future work should include a population-level assessment of the exclusive use of AABR screening technology. Using eSP records, we can track progress on this goal over time by comparing service dates with discharge dates and potentially address some of the delays revealed by the present research.

Our findings suggest that greater attention to timing benchmarks for NICU infants is needed within EHDI systems. Further research should assess the functional impact of these delays and whether a modified timeline or one executed with respect to gestationally adjusted age results in language and developmental outcomes on par with those of non-NICU peers. Research should also examine length of NICU stay with greater granularity (e.g., NICU stays of less than one month, six months, nine

months, 12+ months) and in the presence or absence of additional medical diagnoses. The JCIH now emphasizes the use of diagnostic ABR services prior to discharge for infants with lengthy admissions (JCIH, 2019), but we do not yet know how this update will change the care trajectories of NICU infants. Widespread access to inpatient diagnosis could remediate the NICU-related effects that we observed for the diagnostic benchmark (for infants born in hospitals with pediatric audiology services). It could also open the door for inpatient fitting of assistive devices when care teams confirm the presence of PCHL and the initiation of early intervention services. The heterogenous patient populations that require protracted NICU admission may not benefit from a one size fits all approach to improving EHDI delays. Expansion of inpatient diagnostic services and the development of other strategies to meet the needs of NICU infants should be family-centered to promote attention to and respect for a family's goals, ensure access to timely and evidence-based care, and provide coordinated services (Moeller et al., 2013). Care coordination would be especially important for families of NICU infants with complicated medical needs and who must balance competing concerns.

Finally, although we analyzed racially stratified odds ratios with respect to missing prescriptive EHDI benchmarks in a sub-set of NICU infants with longer admissions, significant gaps remain in our knowledge about EHDI benchmarks and racial disparities among both NICU and non-NICU infants. Future work may consider examining racial disparities among infants with any length of NICU stay, using more specific categorizations of racial background, including hearing outcomes, and integrating data on LTFU/D.

Conclusion

This work contributes to the epidemiological literature about infant and early childhood hearing loss. Baseline characterization of the current EHDI trajectory for infants with lengthy NICU stays is a necessary step to refining recommendations for this population and if indicated, adapt JCIH recommendations in the future by accounting for gestational age. Our results reveal that overall, NICU babies achieve EHDI benchmarks at lower rates than non-NICU peers, including age at initial screening which has otherwise been considered fully-achieved in the literature. It may be appropriate to consider an alternate EHDI timeline based on gestationally adjusted age in formal clinical guidelines.

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Pathway to Amplification in Children who Passed their Universal Newborn Hearing Screening Bilaterally

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Abstract

Purpose: The aim of this study was to investigate the pathway to amplification technologies for children who passed their universal newborn hearing screening (UNHS) bilaterally with the intent of revealing effective strategies to identify children with acquired or progressive hearing losses. Additionally, the degrees, types, and causes of hearing loss, as well as the types of amplification used by the patients were investigated.

Methodology: Medical records were reviewed for 102 children who passed their UNHS bilaterally and who are enrolled in the Boston Children's Hospital Amplification or Cochlear Implant Programs. Of the 204 total ears, 177 ears were identified with hearing loss and were included in the study.

Conclusion: More than half of new hearing loss identifications in children over 11 years and approximately one third of all new hearing loss identifications resulted from a referred hearing screening. For children under age three, a speech-language delay was the most common reason for referral leading to identification of a permanent, postnatal hearing loss. This study emphasizes the importance of routine hearing screenings in school-aged children as well as highlights the need for audiological evaluations when signs of childhood hearing loss arise, such as a speech-language delay.

Keywords: UNHS, hearing aid, cochlear implant, acquired hearing loss

Acronyms: AABR = automated auditory brainstem response; BCH = Boston Children's Hospital; CMV = cytomegalovirus; cCMV = congenital cytomegalovirus; EVA = enlarged vestibular aqueduct; OAE = otoacoustic emission; UNHS = universal newborn hearing screening

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Universal Newborn Hearing Screening (UNHS) has remarkable value in decreasing the average age of hearing loss identification (Dalzell et al., 2000; Vohr et al., 1998); however, it is possible for a child to pass the newborn hearing screening with a mild, congenital hearing loss. Current automated auditory brainstem response (AABR) testing and otoacoustic emissions (OAEs) screening tools frequently use a 30–35 dB criterion level, which would fail to capture newborns with a slight to mild hearing loss. Johnson et al. (2005) estimates that approximately 23% of newborns who have a permanent hearing loss would pass a UNHS conducted via AABR as a result of the chosen screening level.

In addition, there are many causes of delayed-onset congenital or acquired hearing loss that can occur in childhood, including hearing loss associated with genetic mutations, infectious diseases, anatomic abnormalities, trauma, and ototoxicity (Kenna, 2015). By age nine, 25% of permanent childhood hearing loss is postnatal in nature, suggesting that while the UNHS is playing a significant role in the identification of permanent childhood hearing loss, provisions must also be in place to identify children who acquire hearing loss postnatally (Weichbold et al., 2006; Watkin & Baldwin, 2011). Among the cases of permanent childhood hearing loss identified through post-neonatal care pathways, hearing loss is most commonly identified due to school hearing screenings and parental concerns regarding hearing (Dedhia et al., 2013; Watkin & Baldwin, 2011). Once identified, Walker et al. (2014) observed significantly longer delays from hearing loss identification to intervention for children with postnatal hearing loss compared to children who were identified in the newborn period. The same investigation revealed that degree of hearing loss predicted age at follow-up clinical services

for children with postnatally identified hearing loss, such that children with more severe losses received services at younger ages compared to children with milder hearing loss.

Approximately 40% of patients in the Amplification Program at Boston Children's Hospital (BCH) passed their UNHS bilaterally. In the BCH Cochlear Implant Program, 18% of patients with known UNHS outcomes passed in both ears. We designed this study to investigate the pathway to amplification technologies for children who passed their UNHS with the aim of revealing the factors that led to the later identification of children with hearing loss. This study addresses the average age of hearing loss identification and the average time between hearing loss identification and amplification fittings in this population. Additionally, we describe the degree, type, and causes of hearing loss observed. Based on previous studies described above, we hypothesized that most children would be identified through childhood hearing screening programs and that more severe hearing losses would have a shorter time between identification and intervention.

Methods

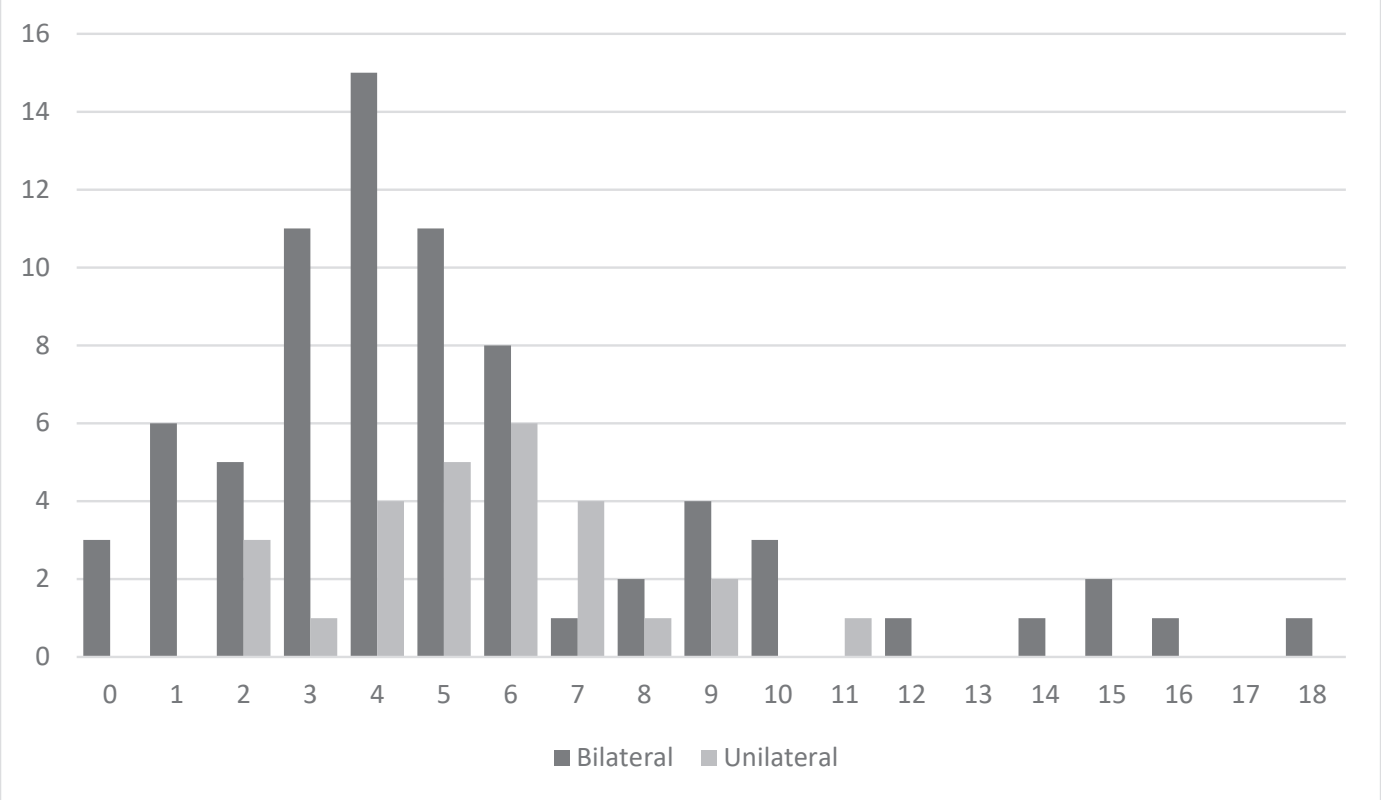
We reviewed medical records of 102 children who passed their UNHS and who are enrolled in the BCH Amplification and/or Cochlear Implant Programs. Medical records were included for review from July 1999 through July 2018. Participants were included in this study if they were (a) identified with hearing loss between 0–22 years of age, (b) had known outcomes of their UNHS, and (c) were users of amplification technologies including hearing aids, cochlear implants, or bone anchored hearing systems. Of the 204 ears, 177 ears were identified with permanent hearing loss and were included in the study. Table 1 indicates the breakdown of participants by sex and by whether the hearing loss was unilateral or bilateral at initial diagnosis.

Table 1
Participant Breakdown by Sex and Number of Ears with Hearing Loss (Unilateral vs. Bilateral)

	Male	Female	Total
Bilateral	37 (36.3%)	38 (37.3%)	75 (73.5%)
Unilateral	16 (15.7%)	11 (10.8%)	27 (26.5%)
Total	53 (52.0%)	49 (48.0%)	102 (100%)

Figures 1 and 2 respectively display the age of identification broken down by laterality of hearing loss and by sex. Note that race/ethnicity data are not reliably coded in the hospital medical record and are not included.

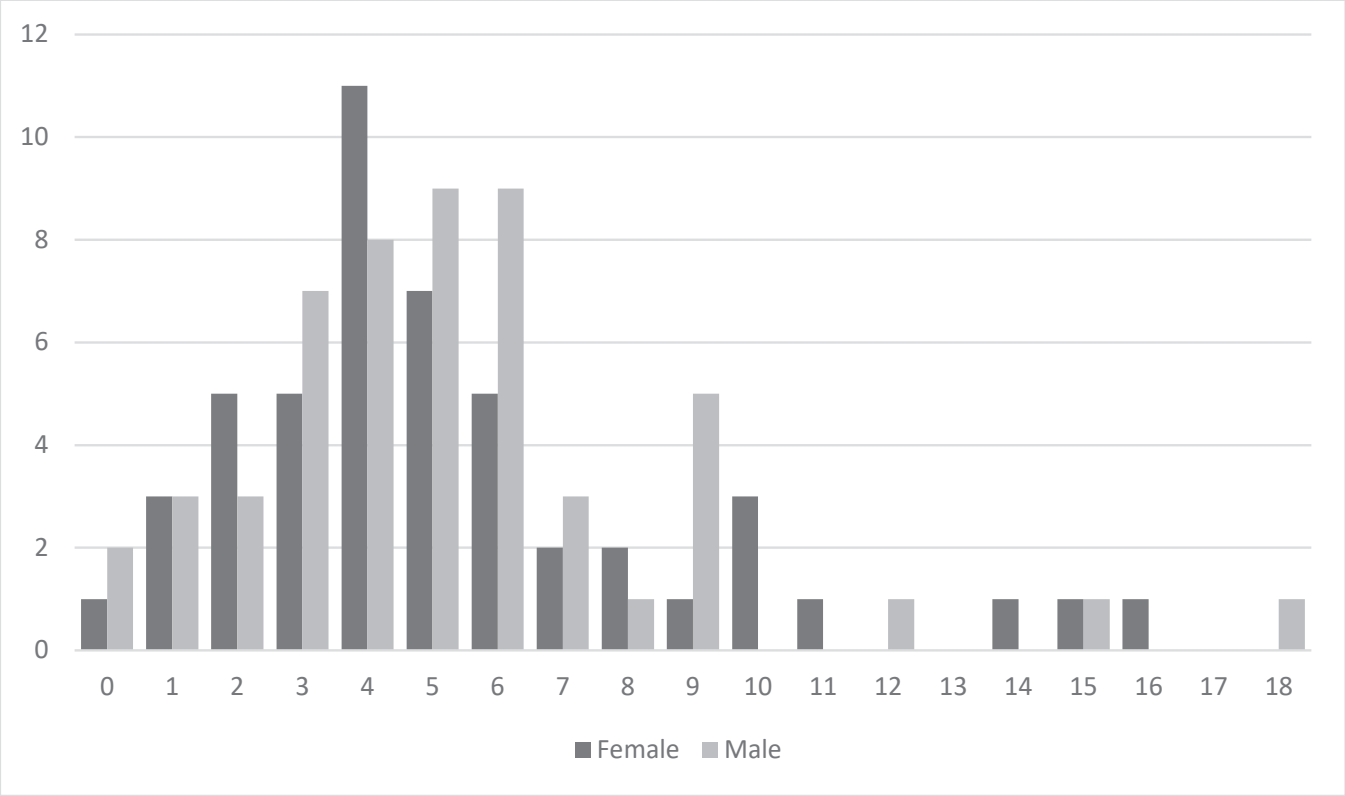
Figure 1
Histogram of Age (in Years) of Identification of Hearing Loss Split by Unilateral Versus Bilateral Hearing Loss



Note. Age of identification was not significantly different between these two groups (t_{78} = -0.6, p = 0.5).

Figure 2

Histogram of Age (in years) of Identification of Hearing Loss Split by Sex of Participant



Note. Age of identification was not significantly different between these two groups (t_{98} = -0.08, p = 0.9).

When reviewing the medical records, we investigated certain criteria to describe the type, degree, and configuration of the hearing losses. The types of hearing loss were determined to be sensorineural, conductive, or mixed. We categorized the patients' hearing loss configurations using the terms flat, rising, sloping, cookie bite, reverse cookie bite, notched, or unconventional. The patients' best threshold degrees and worst threshold degrees were documented to fully capture their hearing loss and to not exclude those with irregular configurations. Additionally, the patients' 2000 Hz pure tone threshold degrees were documented due to the importance of 2000 Hz in speech recognition.

To capture the patients' timeline to amplification technologies, we looked at the month and year of initial hearing loss diagnosis and calculated the years between birth and hearing loss identification to find the average age of identification. We then investigated the month and year of initial hearing aid fitting and calculated the years between hearing loss identification and hearing aid fitting. For patients who use cochlear implants, we documented the date of initiation for their pre-surgical hearing aid trial, if known; if there was no documented hearing aid trial, the date of intervention was marked as the implant surgery date. With this information we were able to calculate the average amount of time between initial hearing loss diagnosis and amplification fitting across all of our patients.

We were also interested in capturing identifiable reasons for the referral for audiological evaluations. These fields included a referred hearing screening at the pediatrician or school, speech-language delay, pediatrician concern, parental concern, suspected or confirmed diagnosis of autism spectrum disorder, or other medical referral from a specialist. These reasons were not mutually exclusive, and, for some patients, more than one reason was selected. In our records, it was not always clear whether the hearing screening was performed at the doctor's office or the school; hence these are combined. Tier 1 and 2 risk factors for childhood hearing loss outlined by the Massachusetts Department of Public Health (Table 2) were investigated as possible predictors for late-onset childhood hearing loss. The risk factor tier indicates when an infant would receive follow-up hearing testing. Infants born with Tier 1 risk factors are recommended to receive a diagnostic ABR by 3 months of age. This appointment is scheduled by the birth hospital prior to discharge. Infants born with Tier 2 risk factors are recommended to receive a diagnostic hearing assessment at 6–9 months of age (Stewart, 2017). This is coordinated by the medical home.

Knowing the etiology of hearing loss was important in the determination of whether the participant's hearing loss was acquired, presumably congenital missed by the UNHS, or delayed-onset congenital. If the etiology of the hearing loss was known, we categorized them as

Table 2

Massachusetts Department of Public Health Tier 1 and Tier 2 Risk Factors for Hearing Loss

Tier 1	Tier 2
• Maternal CMV • Down Syndrome • Cleft lip/palate • Bacterial meningitis • Craniofacial anomalies • Syndromes associated with hearing loss • Perinatal asphyxia • ECMO • Hyperbilirubinemia (> 20 mg/dL bilirubin) • Permanent hearing loss in immediate family • Parental or medical provider concern	• > 10 days mechanical ventilation • ≤ 32 weeks gestational age • < 1500 grams birth weight • Permanent hearing loss in extended family • Herpes, rubella, syphilis, or toxoplasmosis • Head trauma • Ear pits with preauricular tags • Ototoxic medications (> 7 day course in conjunction with loop diuretics) • NICU stay > 5 days

Note. CMV = cytomegalovirus, ECMO = Extracorporeal membrane oxygenation, NICU = Newborn Intensive Care Unit.

genetic, such as connexin-26 or related with a syndrome; anatomical, such as enlarged vestibular aqueduct (EVA); caused by infection, such as cytomegalovirus (CMV); caused by ototoxic medications, such as chemotherapy; or due to another cause. We further wanted to investigate whether the patients had a coexisting diagnosis related to neurologic status, such as Autism Spectrum Disorder or intellectual disability.

Results

Approximately half (52.0%) of hearing losses were sensorineural in nature. Conductive hearing loss comprised 30.5% of hearing losses in our cohort and the remaining 17.5% of hearing losses were mixed in nature. Figure 3 illustrates degree of hearing loss for the 177 ears in the study based on the 2000 Hz threshold, the best threshold, and the poorest threshold. For 63.8% of ears, the 2000 Hz threshold at hearing loss identification was in the normal hearing or mild hearing loss range. 2000 Hz thresholds were observed in the moderate or moderately-severe hearing loss range for 26.0% of ears and in the severe to profound range for the remaining 10.1% of ears. At initial identification, more than 80% of ears had at least one pure-tone threshold in the normal to mild loss range and more than 60% of ears had at least one pure-tone threshold in the moderate to profound range. The majority (90.2%) of participants wore hearing aids; 8.8% used cochlear implants exclusively or as a bimodal solution. The rest of the participants (1%) used a bone-anchored device.

Etiologies of hearing loss varied greatly across participants. Unknown etiology accounted for 37.3% of participants, often despite the use of temporal bone imaging and genetic testing under management by an otolaryngologist. Acquired conditions accounted for 31.4% of hearing loss, including conditions such as chronic otitis media (53.1%), cholesteatoma (25.0%), or ototoxicity (18.8%). Syndrome related losses accounted for 16.7% of participants, of which the most common was Down syndrome (58.8%). Enlarged vestibular aqueducts accounted for 10.8% of participants. Connexin-26 genetic mutations accounted for 3.9% of participants. Congenital CMV (cCMV) accounted for 2.9% of participants. Incidentally, 5.9% of participants had a comorbid diagnosis of Autism Spectrum Disorder and 4.9% of participants had a comorbid diagnosis of intellectual disability.

Table 2 summarizes hearing loss identification and amplification fitting timelines by type of hearing loss. The average age of hearing loss identification was 5.7 years (SD = 3.6 years). Group means for type of hearing loss were evaluated for differences using one-way Analysis of Variance (ANOVA) testing. No significant difference for age of hearing loss identification was observed based on hearing loss type, $F(2, 174) = 2.79$, $p = 0.06$. Once identified with hearing loss, the average time from diagnosis to amplification fitting was 2.0 years (SD = 2.8 years). A significant main effect of type of hearing loss was observed for the time from hearing loss diagnosis to amplification fitting, $F(2, 174) = 6.45$, $p < 0.01$. A Tukey test for multiple comparison of means, using a 99% confidence level, revealed that children with sensorineural hearing loss had a significantly shorter time from hearing loss diagnosis to amplification fitting than children with conductive hearing loss ($p < 0.01$). No difference was observed when comparing children with mixed hearing loss to those with either sensorineural ($p = 0.15$) or conductive hearing losses ($p = 0.63$).

Tier 1 and 2 risk factors for hearing loss were investigated as possible predictors for late-onset childhood hearing

loss. At least one Tier 1 or 2 risk factor for hearing loss in the neonatal period was present for 40.2% of our cohort. The average age of hearing loss identification for those with at least one risk factor was 5.6 years (*SD* = 4.2 years) compared to 5.8 years (*SD* = 2.8 years) for those without a risk factor. A Tier 1 risk factor for hearing loss was present in 24.5% of participants. The most frequent Tier 1 risk factor was an immediate family history of hearing loss (*n* = 9) followed by cCMV (*n* = 3). 19.6% of participants had a Tier 2 risk factor for hearing loss. Among Tier 2 risk factors, the most commonly observed was a neonatal intensive care unit stay of greater than 5 days (*n* = 11). Six participants had an extended family history of hearing loss. Five participants were given ototoxic medication in the neonatal period. Five participants had a gestational age of less than 32 weeks.

Table 3 shows reasons for audiological referral by age group. Approximately 1 in 4 patients did not have an identifiable reason for audiological evaluation. For children older than 3, a hearing screening was the primary reason for referral for diagnostic hearing testing. For children under age three, a speech-language delay was the most common reason for referral leading to identification of a permanent, postnatal hearing loss. A referral from a specialist (e.g., geneticist, developmental pediatrician, cardiologist) led to diagnosis for 22.1% of patients.

Table 3

Reason for Referral for Audiological Evaluation by Age Group (Age of Diagnosis)

	Infant/Toddler (0-3 Years)	Preschool (4-5 Years)	Early School (6-10 Years)	Later School (11+ Years)	All Ages
n	25	31	39	7	102
Referred Screening	0.0%	38.1%	35.5%	57.1%	27.5%
Speech-Language Delay	37.9%	22.9%	16.1%	14.3%	25.5%
Referral from Specialist	27.6%	17.1%	22.6%	0.0%	20.6%
Parent Concern	17.2%	22.9%	9.7%	14.3%	16.7%
Primary Care Provider Referral	0.0%	5.7%	6.5%	14.3%	4.9%
No Known Reason	34.5%	14.3%	25.8%	14.3%	23.5%

ANOVA was performed to determine whether severity of hearing loss was related to identification of hearing loss. The analysis indicated no significant relationship between severity of hearing loss, either based on best hearing threshold or best threshold at 2 kHz, and number of months between identification of hearing loss and first fitting with amplification. The average time between identification and fitting was 25.5 months (*SD* = 34.9 months).

Discussion

The implementation of the UNHS has made a significant impact on early hearing detection and intervention. However, UNHS cannot stand alone in detection of childhood hearing loss. As observed by Walker et al. (2014), this study indicates that children identified with hearing loss through post-natal pathways experience long delays between hearing loss identification and the implementation of hearing loss interventions.

Documented risk factors for hearing loss fall into two tiers, which then determines the timeline for initial diagnostic testing. Children with a Tier 1 or Tier 2 risk factor requiring diagnostic testing may have not developed hearing loss by the time of initial appointment despite the possibility of later-onset hearing loss. This supports routine monitoring and screening of hearing to document any changes in a prompt manner. However, the risk factors do not capture every child who may develop a delayed-onset congenital or acquired hearing loss. The list of risk factors increases the number of children being diagnostically monitored for potential hearing loss in childhood but cannot encompass or predict all children that will require audiological evaluations. This is supported by our cohort as children with and without risk factors were included.

Children who pass their UNHS, but experience signs of hearing loss during childhood must be appropriately referred to an audiologist trained to evaluate hearing in pediatric patients. The most frequent catalyst for hearing loss identification in our cohort was referring on a routine hearing screening, consistent with published data (Dedhia et al., 2013; Watkin & Baldwin, 2011). Our data demonstrate the importance and necessity of school- and primary care provider-based hearing screenings in the process of identifying and treating children with hearing loss. There may have been delays that we could not capture in this study. For instance, if a patient referred their

school screening and then went to their physician for a repeat screening and then was referred to our clinic, this may have caused added delay to the time of diagnosis.

Additionally, our data show the importance of referring children with speech delays for hearing evaluations, even if they passed the newborn hearing screening. This was the primary route to identification for children under 3 years of age. Speech-language pathologists and Early Intervention staff should not assume hearing is normal if a child passed their newborn hearing screening and should include a hearing test as part of the work-up when a child is exhibiting speech and language delays.

We found that the average duration between diagnosis and fitting is greater than one year. This suggests there is a lesser sense of urgency for these older children than there is for children who refer newborn hearing screening and are fit with amplification by 6 months. Boston Children's Hospital does abide by the EHDI 1-3-6 guideline for newborns, it being tied to a state mandate. These data suggest that Boston Children's may benefit from an initiative to fit later-diagnosed children with hearing aids within 3 months of diagnosis.

Our data also demonstrate a relative greater average time from diagnosis to fitting of children with conductive hearing losses. This is not surprising given the time it takes to evaluate candidacy for the greater number of medical and surgical treatments available for conductive hearing loss. Future research may evaluate whether efforts to quickly determine the etiology of conductive hearing loss may lead to earlier fitting of amplification. Future research may evaluate whether there are benefits to fitting amplification synchronously with the medical evaluation process instead of waiting for the physicians to complete their assessments prior to fitting amplification. This finding raises the question as to whether the addition of new options for medically treating sensorineural hearing loss (e.g., gene therapy) may increase time between diagnosis and fitting in the coming years.

Conclusions

It is critical to reinforce the importance of regular childhood hearing screenings through later school-age years. These efforts provide opportunities for earlier identification of childhood hearing loss allowing for earlier intervention options. Family members, educational professionals and clinicians alike should be aware of and pay attention to signs of childhood hearing loss, such as speech-language delay, academic difficulties, and increased exhaustion at the end of a school day to ensure proper referrals lead to early diagnosis. Pediatric medical centers should ensure that, once diagnosed with hearing loss, older children are being fit with amplification with as little delay as possible, similar to the 1-3-6 guidelines for newborns.

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A Survey on the Global Status of Newborn and Infant Hearing Screening

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Abstract

Objective: Assess the global status of newborn/infant hearing screening (NIHS) and its effectiveness in early detection and intervention of permanent childhood hearing loss (PCHL).

Design: Individuals potentially involved with NIHS in 196 countries/territories (in the following text referred to as countries) received a questionnaire about coverage, strategies, and outcomes of country-specific NIHS programs.

Study Sample: Questionnaires from 158 countries were returned.

Results: Thirty-eight percent of the world's population were reported to have no/minimal screening, 33% reported screening more than above 85% of the babies (hereafter referred to as universal newborn hearing screening [UNHS]). Mean living standard of countries with UNHS was 10 times higher than in countries with NIHS coverage that was less than 10%. Average age at diagnosis of PCHL was 4.6 months for screened children and 34.9 months for non-screened children. Average age at start of intervention was 6.9 months for screened children and 35.2 months for non-screened children. Methods used for screening included otoacoustic emissions (OAE) in 57% of countries, automated auditory brainstem response (AABR) in 11%, and two-step OAE-AABR in 30%. On average, 4.5% of the infants failed the screening and 17.2% of those children were reported as lost-to-follow-up. The prevalence of PCHL identified in NIHS programs ranged from 0.3–15.0 per 1,000 infants with a median of 1.70.

Conclusions: Newborns with PCHL are more likely to benefit from early identification and intervention in countries where NIHS is done. There is a need to invest in NIHS programs, including data collection, in low-income countries.

Keywords: hearing loss, children, newborn hearing screening, neonatal hearing screening, infant hearing screening

Acronyms: AABR = automated auditory brainstem response; EHDI = Early Hearing Detection and Intervention; HL = hearing loss; JCIH = Joint Committee on Infant Hearing; NIHS = Newborn and Infant Hearing Screening; OAE = otoacoustic emissions; PCHL = permanent childhood hearing loss; UNHS = universal newborn hearing screening; WHO = World Health Organization

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Recent estimates by the World Health Organization (WHO) indicated growing absolute numbers and prevalences of people with disabling hearing loss (Olusanya et al., 2019; WHO, 2018a). For children, too, the absolute numbers are rising as the world population grows. An estimated 34 million children currently have disabling hearing loss, most of them living in South Asia, Asia Pacific, and Sub-Saharan Africa (Vos et al., 2016; WHO, 2018a, 2018b, 2018c; Wilson et al., 2017). These children are in danger of impaired language, social, emotional, and academic development (Ching et al., 2010; Ching et al., 2018; Neumann et al., 2006; Vohr et al., 2011; WHO, 2016; Yoshinaga-Itano et al., 1998; Yoshinaga-Itano et al., 2018).

Universal newborn hearing screening (UNHS) and prevention of permanent childhood hearing loss (PCHL) are the most effective measures to reduce both the prevalence and negative consequences of PCHL, with UNHS being very effective for high-income countries (Joint Committee on Infant Hearing [JCIH], 2013; Pimperton et al., 2016; WHO, 2010; Wilson et al., 2017), and prevention expected to show higher relative effects for low-income countries (Ching et al., 2010; Ching et al., 2018; Neumann et al, 2006; Vohr et al., 2011; Vos et al., 2016; WHO, 2016, 2020a; Wilson et al., 2017).

According to the recommendations of the Joint Committee on Infant Hearing (JCIH, 2007, 2019), babies should undergo UNHS before one month of age, those who fail the screening should get an audiological diagnosis before 3 months, and those with PCHL should be enrolled in early intervention before 6 months of age (EHDI 1-3-6 guidelines). If a country is already accomplishing this goal, it is advised that this country should strive to achieve the new goal of undergoing UNHS by 1 month of age, getting an audiological diagnosis before 2 months of age, and enrolling in early intervention by 3 months of age (JCIH, 2019).

Many studies convincingly demonstrate that children with PCHL who were identified and treated early have better language and academic outcomes than those with late-treated hearing loss. This has been shown for general language development (Ching et al., 2018, Neumann et al., 2006; Yoshinaga-Itano et al., 1998), vocabulary (Yoshinaga-Itano et al., 2017), developmental scores, and quality-of-life (Korver et al., 2010) for children whose hearing loss was identified by NIHS, who were fitted early with hearing aids (Tomblin et al., 2015) or cochlear implants (Yoshinaga-Itano et al., 2018), or who were enrolled in early intervention services (Vohr et al., 2011) compared to children without UNHS. Recent large-scale epidemiological studies in Australia and Great Britain have provided strong evidence of the positive long-term outcomes of earlier treatment of infant hearing loss that can be achieved through UNHS programs, compared to later treatment in terms of language, cognitive, reading, and general academic development of hearing impaired children and adoslescents (Ching et al., 2018; Kennedy et al., 2006; Pimperton et al., 2016; Wake et al., 2016). Although the direct and indirect costs and some potential negative consequences of UNHS programs have to be taken into consideration (Kemper et al., 2000; Zhao et al., 2003), studies on parents' perspectives (Fitzpatrick et al., 2007; van der Ploeg et al., 2008; Young & Tattersall, 2007) and cost-benefit analyses of unaddressed hearing loss (WHO, 2017a) showed that advantages of early hearing detection and intervention (EHDI) outweigh the disadvantages.

In 1995 a WHO resolution called on member states to prepare national plans for the prevention and control of major causes of avoidable hearing loss, and for early detection of hearing loss in babies, toddlers, and children (WHO, 1995). Yet, in 2012 only 32 countries reported the implementation of such policies, and the WHO bemoaned an overall scarcity of epidemiological evidence regarding prevalence of hearing loss and ear diseases (WHO, 2013). A second resolution by WHO, adopted in 2017, reaffirmed the goals of the first and urged member states to collect high-quality population-based data on ear diseases and hearing loss (WHO, 2017b). So far, no information has been gathered about the global situation of NIHS.

The international study presented here aimed to assess the global status of coverage, strategies, and results of NIHS programs and child audiology services in as many countries or territories (referred to hereafter as countries) as possible to serve as a baseline for further evaluation and improvement of NIHS effectiveness. In addition, the study explores the relation between national economical indices and key screening parameters.

Materials and Method

Questionnaire

A 19-item questionnaire, based on an Italian NIHS questionnaire (Bubbico et al., 2008), was modified to investigate the country-specific status of NIHS. It requested information for a reference year about (a) percentage of babies who either underwent a newborn hearing screening or a screening later in the first year of life, relative to the number of live births in the reporting year; (b) target population of the screening as either covering all babies in a country, state, region, or institution (universal screening) or restricted to babies at risk for a PCHL (targeted screening); (c) screening method: otoacoustic emissions (OAE) or automated auditory brainstem response (AABR) alone, or two-stage OAE-AABR screening (AABR follows immediately if a baby has failed an OAE screening), questionnaire-based screening, or other screening method; (d) percent of all newborns and of screened babies who would have needed an audiological assessment because they were suspected for a hearing loss, and the percent of babies who received such an assessment; (e) number per 1,000 infants identified with PCHL; (f) the percentage of these infants identified with PCHL who had undergone a hearing screening; (g) median or mean ages and age ranges of diagnosis and onset of early intervention for infants with PCHL who either had been screened or not; (h) percent of all babies with PCHL who needed an intervention for their PCHL, percent of babies with PCHL (both screened

and unscreened) who received early intervention, and percentage of all babies with PCHL and of screened babies with PCHL whose intervention started before 6 months of age; (i) whether and when a hearing screening has been mandated; (j) when mandated, for which type of screening; (k) where the screenings were done and who performed them; and (l) percent of all birth institutions in a country which have NIHS programs.

PCHL was defined as unilateral or bilateral permanent hearing loss of > 20 dB HL (hearing loss) in the better hearing ear, averaged over frequencies 0.5, 1, 2, and 4 kHz. These criteria meet more recent evidence for significant risks children with untreated minimal hearing loss face for their speech-language and academic development (Olusanya et al., 2019, Winiger et al., 2016). Intervention may include (but is not limited to) fitting with hearing devices, speech-language therapy, early intervention programing by a parent-infant specialist, or medical or surgical treatment. In cases where it is unclear whether treatment is required, further monitoring also counts as intervention.

Participants

The questionnaire was e-mailed to persons thought to be involved in NIHS programs of as many countries as possible. Much effort was devoted to identifying these key individuals, but for some countries it was futile, especially for those with little or no audiological services. Many key persons were identified through personal knowledge of the authors Katrin Neumann, Shelly Chadha, and Karl R. White, all of whom are well-known in the hearing screening community and who are or have been active in it themselves. As Medical Officer of the WHO Program for Prevention of Deafness and Hearing Loss, Chadha had contact with many national programs and key persons. A letter of invitation signed by the chairs of several international organizations concerned with hearing care (Coalition for Global Hearing Health—CGHH, International Association of Logopedics and Phoniatrics—IALP, International Society of Audiology—ISA, International Working Group on Childhood Hearing, Hearing International) accompanied the questionnaire. Activities of the Executive Committees and members of the above and other organizations such as the American Academy of Audiology (AAA), and also of non-governmental organizations (NGOs), such as Soundseekers and the Christoffel Blindenmission (CBM), helped to identify key persons. In French-speaking Africa, the network of the Société Oto-rhino-laryngologie (ORL) des pays francophones d'Afrique (SORLAF) helped. Where available, national or state Newborn Hearing Screening centers were contacted. All inquiries were made via e-mail by the first author, by telephone, or by addressing contact or key persons directly at international conferences. For some countries the authors of publications on audiological or pediatric topics were contacted with a request for contact details of key people in ear and hearing care. Various key persons also contacted the first author after she had asked professional colleagues to pass on the request. Ministries of Health and regional WHO offices were also requested.

The distribution of the questionnaires started in the autumn of 2014, and updates were received until January 2019. The originally proposed reference year was 2013, but for some countries only information from earlier periods was available. Because the recruitment of informants lasted unexpectedly long, some respondents reported or updated their information up to 2018. The returned questionnaires were proofed for reliability, completeness, and plausibility. To this end, the authors checked whether the responses were within the probable range and, where applicable, percentages added up to 100%. If study-, hospital-, city-, or region-based information was provided additionally or alternatively to country-wide data, Appendix B indicates the population for which the information was given. In cases where data were only available for a subset of a country, the respondent was asked how representative the data were for the entire country. All questionnaires were returned to the respondents with comments and questions as needed. For most countries, one or two revisions were necessary. To eliminate potential biases, attempts were made to have the information confirmed by a second person whenever possible.

Statistical Analysis

In addition to descriptive analyses of the questionnaire responses, we investigated the correlations between the national average nominal gross domestic product per capita (GDP per 1000; the United Nations Statistics Division (https://en.wikipedia.org/wiki/List_of_countries_by_GDP_(nominal)_per_capita); the total health expenditure per capita (HE; Global Health Expenditure Database [GHED]; https://apps.who.int/nha/database); and screening coverage, fail rate, prevalence, mean age at diagnosis, and mean age at treatment start.

Because the distributions of fail rate, prevalence, mean age at diagnosis, and treatment start were positively skewed, as were GDP and HE, they were normalized by a log10-transformation. The normalized distributions of GDP and HE correlated very highly ($r = .96$; $N = 182$). Because both variables were nearly identical, further analyses were performed only with GDP.

Results

Of the 196 contacted countries (192 UN Member States and Kosovo, Macedonia, Palestine, and Puerto Rico), 158 provided information. The country-specific results of the survey are presented in Appendix B. Because some of the information requested in the questionnaire (see Materials and Method, subsection Questionnaire) was difficult for respondents to access, Appendix B presents only the variables which appeared to be the least biased ones, leaving out the items (b), (d), (f), (h), (j), (l). Nonetheless, for several of these items some

justifiable results are shown and discussed in the following text. For some countries, study-, hospital-, city-, or region-based information was provided, either exclusively or in addition to nationwide information. Appendix B indicates the population groups for which information was provided on a particular item, and participants were asked about the representativeness of their information for the country as a whole. Therefore, in several cases, a country-specific row in Appendix B contains both—study-, hospital-, or region-related information, marked with a superscript index, and country-wide information. In most cases, country-specific information was included in the statistical analysis, and regional information was only included if representativeness was ensured, sample sizes were large enough, and selection bias was judged by the authors to be small.

For all items of the questionnaire, respondents were asked to state whether replies were data-based or estimated. Reporting years (with the number of countries or territories using that year in parentheses) were 2008 (1), 2009 (1); 2010 (2), 2011 (6), 2012 (14), 2013 (67), 2014 (75), 2015 (11), 2016 (15), 2017 (2), and 2018 (3). The number adds up to 197 because information was received from some countries from several regions, sources, or studies, as shown in Appendix B. Number of live births were obtained from the national statistic institutions, the United Nations (UN) Population Department, UN Demographic Yearbooks, or other demographic sources (sources not explicitly named in Appendix B; only NIHS-related data sources are referenced). When country-wide information was not available, fragmentary (or implausible, regional, hospital-based, or study-based) data were reported separately in Appendix B if the samples seemed to be representative for specific items.

Appendix B and Figure 1 demonstrate the worldwide coverage rates of NIHS programs as reported by participants for their country. In Table 1 and Figure 1 the coverage of screening within a country was classified into five categories, from no or minimal screening (0% to < 1% of newborns were screened), over three middle-range categories, to near/full UNHS (more than 85% of newborns were screened). The coverage of screening was bimodally distributed, with approximately one third of countries (38% of the world's population) having no/minimal screening and another third (33% of the world's population) having near/fully implemented UNHS programs.

Screening coverage is closely associated with average living standards and economic well-being, as measured

Figure 1
Country Specific Coverage of NIHS Programs

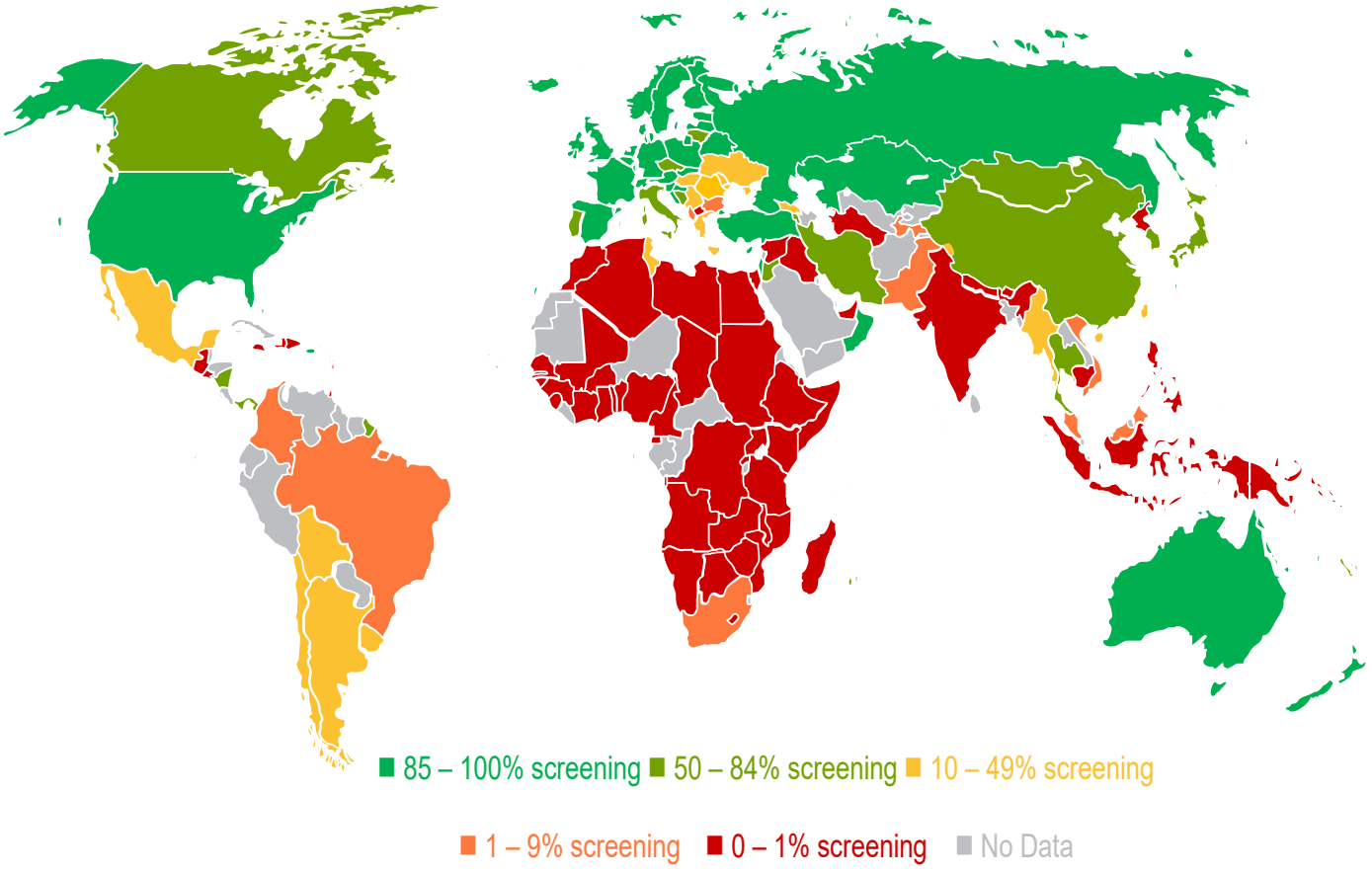


Table 1

Global Coverage of Newborn and Infant Hearing Screening

Coverage of Screenings	Number of Countries	Percent of Countries	Percent of World Population	GDP (nominal) per capita, average
0% to < 1%	64	32.7	37.63	3.7
1% to 9%	14	7.1	7.42	3.9
10% to 49%	19	9.7	8.33	10.7
50% to 84%	17	8.7	6.72	14.4
85% to 100%	41	20.9	32.59	40.4
No/ insufficient data	41	20.9	6.09	8.6
Sum	196	100	98.78	

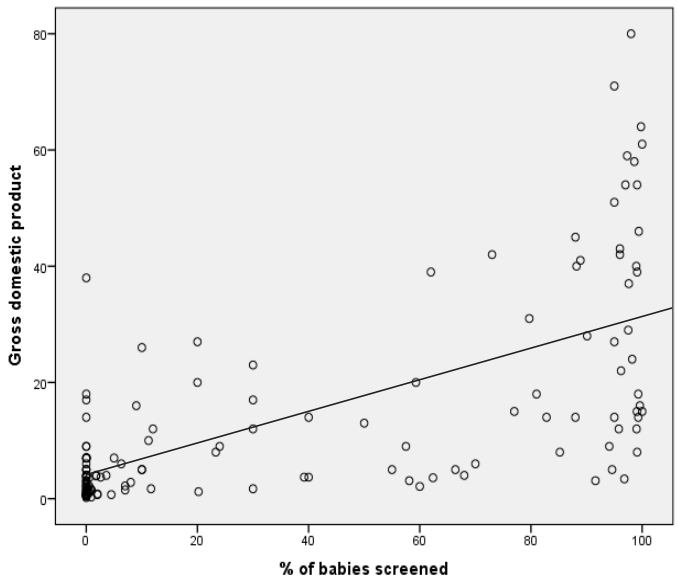
Note. The entries do not add up to exactly 100% because of not listed dependent and disputed territories. GDP = gross domestic product.

by the GDP. Countries with a near/full screening enjoy an average of living standard which is 10 times higher than that of countries with a screening coverage of < 10%.

Of the more than 32 million babies of the participating countries who were screened with the standard screening methods (OAE alone, AABR alone, OAE-AABR) within a reporting year, more than 21 million (66.5%) were screened with OAE alone, about 4.6 million (14.3%) with AABR alone, and about 6.2 million (19.2%) with OAE-AABR (i.e., only if OAE failed was AABR recorded).

Figure 2

Association Between Gross Domestic Product of Countries and Coverage of Newborn Infant Hearing Screening



Note. Gross domestic product (GDP) not log 10-normalized for illustrative reasons.

Behavioral tests were reported as used in only 6 countries, maternal questionnaires or tympanometry were seldom used. OAE was the favored method in 57% of the countries and exclusively used in 29%, followed by OAE-AABR (30% and 21%, respectively), and AABR (11% and 4%, respectively).

The association between coverage of screening and GDP (Pearson's $r = .68$, GDP not log-transformed for sake of clarity) is illustrated in Figure 2 which shows that most data points of the scatter plot cluster at the far right or the far left side. Countries with a moderate coverage of screening (20%–80%) are in the minority. The countries with a relatively high coverage show a large variance in GDP, and they include countries with a low GDP of < 10 (Belarus, China, Kazakhstan, Marshall Islands, Micronesia, and Russia).

The log10-normalized GDP correlated negatively with all normalized screening parameters, that is with fail rate ($r = -.30$, $p = .031$), prevalence ($r = -.43$, $p < .001$), mean age at diagnosis for screened ($r = -.30$, $p = .018$) and non-screened hearing-impaired babies ($r = -.43$, $p = .012$), and mean age at treatment start for screened ($r = -.34$, $p = .016$) and non-screened ($r = -.54$, $p = .003$) babies.

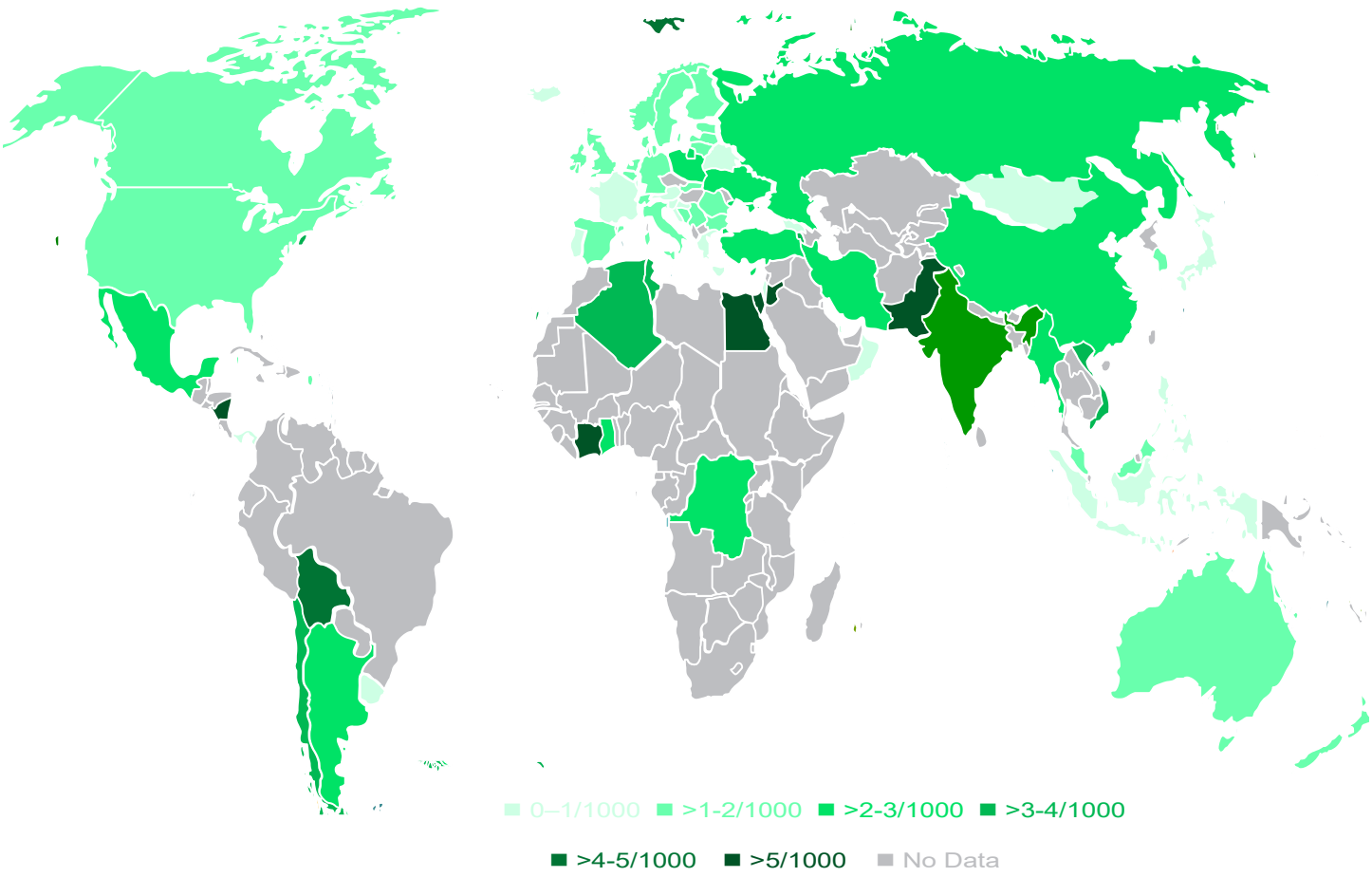
The mean fail rate of the NIHS programs of all 55 reporting countries was 4.5% (*SD* 5.1, range 0.2–30.8). It was statisticially significantly lower ($p = .007$, Mann-Whitney U-test) in the countries with high NIHS coverage of ≥ 85% ($M = 3.1\%$, *SD* 2.6, $n = 33$) and its range was narrower (0.3–11.6), compared to countries with lower screening coverage ($M = 6.5\%$, *SD* 7.0, range 0–98.2, $n = 22$; all *n*s denote number of countries). The mean lost-to-follow-up rate was 17.2% (*SD* 25.6, range 0–98.2, $n = 51$). This rate was numerically but not statistically significantly lower for countries with high screening coverage (14.3%, *SD* 24.9, $n = 30$) compared to countries with lower coverage (22.3%, *SD* 27.0, $n = 20$).

The prevalences of PCHL (Appendix B, column 5; Figure 3) ranged between 0.3 and 15.0 per 1,000 (median: 1.70, $n = 75$).

The average age at diagnosis of PCHL for children who had undergone hearing screening was 4.6 months (*SD* 3.4, range 0.1–18, $n = 61$) and the average age for non-screened children was 34.9 months (*SD* 20.4, range 12–120, $n = 34$). The average age at start of early intervention for screened children was 6.9 months (*SD* 4.0, range 1.6–24, $n = 49$), compared to that for non-screened children of 35.2 months (*SD* 18.8, range 12–88.1, $n = 28$). There were large and statistically significant standardized mean difference effect sizes (SMDES) between screened and non-screened children in age at diagnosis ($p < .001$, SMDES = 7.98 months) and age at therapy start ($p < .001$, SMDES = 7.10 months).

Of the babies identified with PCHL in their birth year for this study, 82% on average ($n = 59$) were identified by

Figure 3
Country Specific Prevalance Figures of Infant Hearing Loss



a hearing screening. From 39 reporting countries, 57% of screened babies received early intervention before 6 months of age.

Whether hearing screening was mandated was answered by 98 informants. Of these, 46 reported the presence of a mandate and 52 replied there was none. Of the countries with mandates, 38 required UNHS, 6 required targeted screening, and 2 did not specify. A mandate for hearing screening was first legislated in the United States (1992), followed by Oman (1996), and China (1999). Although governmental mandates seem to be associated with screening coverage (Spearman correlation between the existence of a mandate and categories of screening coverage was *rho* = .51), there are noticeable exceptions. For example, of the 38 high-coverage countries with available information, nine had no mandate.

The screening was carried out in birth facilities in 93% of the countries. Screening occurred in other places such as pediatric, hearing care, immunization, or well-baby clinics in 51% of the countries, and in the homes in 14% of the countries (percentages sum to > 100% because a single country could have screening done in multiple places).

Professions involved in NIHS were physicians (26% of the countries, in 5% exclusively), audiologists, audiological staff, or technicians (69% and 16%, respectively), nurses, midwives, and nonprofessionals such as trained screeners, auxiliary staff, or community health workers (69% and 24%, respectively).

Discussion

This survey provides the first data-based information about the global status of NIHS programs. According to the summarized information provided by respondents, which must admittedly be treated with caution because it is unverified self-report data, 38% of the world's population had no or minimal screening, and 33% had near/fully implemented UNHS programs, that is above 85% coverage. Because the participating countries represent 94.8% of the world population, the results of the study are a reasonable approximation of the global situation.

Worldwide, OAE is the most commonly used screening method, followed by NIHS programs that use a two-step OAE-AABR protocol or those that use AABR alone. This finding is consistent with the results of a 2015 study involving 39 predominantly European countries (Sloot et al., 2015), but differs from a more recent systematic review in which the OAE-AABR procedure was the method most frequently cited in the published literature (Kanji et al., 2018). Yet, our results may be more precise because they summarize information from many countries with low screening coverage, from which there is little

published literature and in which OAE is mainly used as a screening method. In our study, countries with more resources often used a combination of procedures, while low-income countries applied predominantly single-step procedures, mostly using OAE. Almost all participants in our study, who provided separate information on screening in neonatal intensive care units (NICUs), used AABR, which is consistent with international standards (JCIH, 2019) to identify babies with auditory neuropathy spectrum disorders, who are more commonly found in NICUs (Neumann et al., 2006; White et al., 2005). The advantages and disadvantages of the different screening methods are not discussed in detail here, as there is extensive literature on this subject (Kanji et al., 2018; Nennstiel-Ratzel et al., 2017; Neumann et al., 2006; Sloot et al., 2015; White et al., 2005; Wroblewska-Seniuk et al., 2017). With an average failure rate of 4.5 in our study, most screening programs appear to require rescreening after a failed screening, as recommended by the JCIH (2007, 2019) otherwise the failure rate would be higher (Nennstiel-Ratzel et al., 2017).

NIHS is performed mostly in birthing facilities, less in out-patient clinics and homes. The survey also provides region-specific figures on the prevalences of PCHL, with a median of 1.70 per 1,000 infants.

NIHS is associated with a lower average age at which a PCHL is diagnosed and treated, at a time when the brain structures are still physiologically well accessible for treatment (Sloot et al., 2015). The remarkable discrepancy between the average ages of screened and non-screened children at diagnosis of a PCHL and of onset of intervention is a strong argument for the implementation of NIHS programs. It overrides the counterarguments regarding higher direct and indirect costs of a UNHS, compared with a targeted screening for children at risk (Kemper & Downs, 2000), and of potential negative consequences of false positive screenings (Zhau et al., 2003).

The discrepancy in age at diagnosis and onset of treatment of infant hearing loss between countries with and without UNHS programs is reminiscent of the history of implementation of UNHS programs in high-income countries when screened and non-screened populations could be compared. For example, the average age in months at diagnosis of hearing loss for screened children was 4.2 in the United States, 3.1 in Germany, and 3.9 in Austria. However, for children not screened the average age in months for diagnosis was 17.5 in the United States, 39.0 in Germany, and 37.6 in Austria (Harrison et al., 2003; Neumann et al., 2006; Weichbold et al., 2005). The age in months at the start of treatment was 6.8 in the United States, 3.5 in Germany, and 9.4 in Israel for screened children and 19.8 in the United States, 39.0 in Germany, and 19.0 in Israel for non-screened children (Harrison et al., 2003; Neumann et al., 2006; Wasser et al., 2019).

The average age at diagnosis of 4.6 months and start of early intervention of 6.9 months in the present study approaches the goals of the EHDI 1-3-6 guidelines, but does not quite reach them. The implementation of NIHS programs and the associated reduction in the average age of diagnosis and therapy start in many countries around the world can be seen as positive and can be expected to produce significantly better language and academic outcome and benefit from hearing devices of the screened children with PCHL than for later ages (JCIH 2007, Kral & Sharma, 2012; Yoshinaga-Itano et al., 2017; Yoshinaga-Itano et al., 2018). However, the fact that only just over half of all babies with PCHL examined in our study received early intervention before the age of 6 months remains a critical point. Yoshinago-Itano et al. (2017, 2018) have shown that young children with hearing loss who are identified before 3 months of age and begin receiving early intervention before 6 months of age have better outcomes than similar children who are identified and begin intervention at later ages. NIHS programs should thus work toward meeting these critical time marks, which cast a new light on the crucial length of the sensitive periods of auditory pathway maturation for the development of speech understanding and acquisition of spoken language (Kral et al., 2019).

For 38 of 196 contacted countries (mostly countries with very limited audiological services) no key informant could be identified. For low-income countries, responses frequently came from non-governmental organizations or from domestic/foreign researchers who had conducted studies or provided services in that country.

The lack of regional or national NIHS databases and regular data collection impacts the quality of many screening programs. Such a lack is associated with a dearth of tracking programs to refer babies who have failed the screening to audiological diagnostic and treatment services. Without tracking, the lost-to-follow-up rate is usually high or simply unknown. This is also illustrated by the finding that in countries with near/fully functioning UNHS programs lost-to-follow-up rates are on average 7% lower than in countries with lower UNHS coverage. Of the 27 countries that reported trustworthy lost-to-follow-up rates, 13 (48%) were above 30%, the criterion based on the recommendations of the JCIH (American Speech-Language-Hearing Association, 2019; JCIH, 2007) to achieve a return-for-follow-up of 70% of infants or more. This result is in close agreement with that of the meta-analysis of Bussé et al. (2020), where lost-to-follow-up rates from 18 out of 41 (44%) studies were above 30%, which means that nearly half of NIHS programs lose too many children with suspected hearing loss for diagnosis. In a systematic review by Ravi et al. (2016), educational disparity and parents' lack of knowledge were associated with high lost-to-follow-up rates, and the most commonly used strategy to overcome the latter was to use appropriate data management systems.

It is impossible to know how extensive or complete an NIHS program is without a database. For example, several Arab, South East Asian, and Latin American countries do hearing screening in many institutions, but were unable to provide information about relevant screening parameters due to the lack of data. Respondents from such countries

often reported only hospital-based data that may or may not be representative for the whole country. The lack of data becomes even more serious when there is no hearing screening at all. Usually in such cases no information is available on how many children in a birth cohort would have needed diagnosis, how many are suspected of having a hearing loss, how many actually have one, how many have received diagnosis or therapy (therefore these items of the questionnaire are not shown in Appendix B), and at what age (Appendix B, columns 6 and 7).

A full or nearly full UNHS was implemented in the USA, Uruguay, most European countries, Israel, Kazakhstan, Oman, Qatar, South Korea, Seychelles, Australia, New Zealand, and in Pacific Island nations that are territories of the USA. Other countries such as Canada, Mongolia, Panama, and China have implemented large-scale NIHS programs even though they are not universal in the entire country. Interestingly, these countries are by no means all high-income countries. We therefore assume that the implementation of NIHS depends not only on national wealth but also on other factors such as awareness and attention to the problem of infant hearing health among policy makers and health care professionals in a country. The importance of such factors is supported by the fact that some countries with high coverage rates do not have a governmental mandate for screening, indicating that a mandate might be helpful for the implementation of a nationwide NIHS program, but is not necessary.

The dramatically lower living standard of countries with low screening coverage, compared to countries with a high coverage, is aggravated by the fact that 80% of people with disabling hearing loss live in low- or middle-income countries (Lancet, 2017), where poor birth conditions and lack of vaccination programs contribute substantially to the incidence of PCHL (WHO, 2018b). This imbalance is also reflected in the fact that the global production of hearing aids covers less than 3% of the needs in these countries (WHO, 2011).

Less developed NIHS programs often have relatively high lost-to-follow-up rates. Reasons often mentioned for not coming to follow-up appointments include distance from the hospital, difficulties with transport, fear and uncertainty about the referral hospital, procedural problems, lack of awareness and understanding about hearing loss, and inadequate visibility and availability of services (WHO, 2011). The statistically significant negative correlation of GDP, and thus HE, with the prevalence of early childhood hearing loss may be related to the fact that its prevention is not sufficiently effective in low income countries. For example, infections of pregnant women and newborns are more common, education about the consequences of parental consanguinity is less frequent, and the quality of obstetrics and care for premature babies is not as developed as in high-income countries (WHO, 2016). The negative correlation with the mean age of diagnosis and treatment start of non-screened hearing impaired children may be due to the low political will, limited public awareness, low prioritization of childhood hearing loss as a hidden disease, and the low or simply impossible allocation of resources to this condition in low-income countries (WHO, 2016; 2017a). The negative association with the mean age of diagnosis and treatment start of screened infants with hearing loss may be explained by the low standards of NIHS that is mostly sporadic in low resource countries where audiological diagnostic and treatment services are often lacking (Bright et al., 2017; Olusanya, 2012; Olusanya et al., 2009; Olusanya et al., 2014).

The prevalence reports of PCHL were of the expected magnitude (median 1.70 per 1000; range 0.3–15; 75 countries) and are close to those of a recent systematic review and meta-analysis (overall prevalence 2.21 per 1000; range 1–6; 35 included studies; Bussé et al., 2020). The highest prevalences were reported from regions where the proportion of inherited forms of sensorineural hearing loss is relatively high due to traditional high parental consanguinity (e.g., Pakistan, Egypt, Algeria, Jordan, Turkey). This is in line with the findings of the UK Millennium Cohort Study, where the risks of having a parent-reported PCHL at the age of 11 was increased, among others, in children of Pakistani or Bangladeshi ethnicity (Butcher et al., 2019). Genetic counseling and health education is important to reduce these numbers (WHO, 2016; Smith et al., 2005).

In high-income countries NIHS is predominantly hospital-based. But other settings have been shown to be successful, too, such as screenings performed in the parents' homes or in well-baby clinics (van der Ploeg et al., 2012), or as community-based programs in primary health care clinics (Friderichs et al., 2012), screening camps (Bright et al., 2017), or in conjunction with childhood vaccination programs (Friderichs et al., 2012; Olusanya et al., 2009). Community-based infant hearing screening models may be even more cost-effective than hospital-based models, as shown for a program in Nigeria (Olusanya et al., 2009). Worldwide, screening is mostly performed by nurses and trained non-professionals.

To make a UNHS program effective and to enable inter-program comparability, it is necessary to apply quality indicators and benchmarks of the screening, which have been published in position papers of the JCIH (2007, 2019) and the WHO (2010) and are specified in a recently published checklist (Mincarone et al., 2015). Critical points that contribute to the quality of a program are: Definition of the screening targets (e.g., bilateral or unilateral hearing loss, detection threshold); unified definition of risk populations and classification of hearing loss; high coverage rate of the screening; keeping lost-to-follow-up rates low; timely completion of screening, diagnosis, and start of intervention, and continuous quality control and monitoring of the screening process.

Limitations

Because this is the first time a global assessment of NIHS programs has been done, there are understandably a number of limitations that need to be addressed in future efforts of a similar nature. The reporting period was originally planned for 2013. However, the time consuming

data collection prolonged this period and several countries updated their information, whereas others did not. Hence, for some countries recent developments are not taken into account, which may have caused biases. The reported data were often self-reported estimates that occasionally tended to be optimistic. For example, from the reported data a lost-to-follow-up rate of 0% was calculated for 24 out of 51 countries, which is highly unlikely. Where data seemed implausible to the authors after checking with the respondents, they were omitted from calculations. Therefore, the number of reported data points per item shown in Appendix B frequently differs from the sample size of the calculated overall outcomes. Moreover, it has been difficult to collect data from some large countries, such as India, which is a subcontinent in itself, and the data often refer to local screening programs or extrapolations from them. The authors have tried to extract information from the available data that

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Appendix A

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Appendix B

Country-Specific Information on Newborn Infant Hearing Screening (NIHS) as Retrieved by Informant Replies

Region / Country	Reporting year*: # of live births / Data source**	% of newborns screened / Method used (%)	Fail rate / Lost-to-follow-up	Babies with PCHL[1] per 1000	Mean age in months (range) at diagnosis of screened / and non-screened babies	Mean age in months at treatment start of screened / and non-screened babies	Screening mandated? / Year	Where was screening done? / Who screened?
Europe								
Albania	2013: 35,750 MH[2]	3.6% UNHS 100% OAE					no	2 private hospitals 20% audiologists, 80% nurses
Austria	2012: 78,952 SA[3], TCHR[4]	88% UNHS 90% OAE, 10% OAE-AABR	2.3 0.0	1.0	8.7 (0.2–145) 37.1 (0.9–188)	8.7 (0.2–145) 46 (0.9–188)	no	95% birth facilities, 5% ENT clinics/practice 10% physicians, 20% audiologists, 65% nurses, 5% midwives
Belarus	2013: 116,073 MH[2], NCORL[5]	94.6% UNHS 42.9% OAE		0.7	6 (1–17) 12 (9–19)	6 18 (9–19)	yes, UNHS/2008	94.6% birth facilities, 4% other medical institutions 10% physicians, 10% audiologists, 80% nurses
Belgium	2010: 129,173 KG[6], DSN[7], ZOG[8]	88.9% UNHS 61.4% AABR, 38.6% OAE-AABR	2.8 42.3	1.7	1.8 (0.4–6.1) (Flanders)	1.8 (0.4–6.1) (Flanders)	yes, targeted/not reported	3.6% birth facilities, 29.9% homes, 51,6% welfare baby clinics, 14,9% local districts houses 3.6% audiologists, 96.4% nurses/CHW[9]
Bosnia and Herzegovina (Rep of Srpska)	2013: 3.883[10] BR[11], ENT Dept[12]	80.9% UNHS 100% OAE-AABR	0.2 0.0	1.3	7.5 (6-9) 30 (24–36)	10 33 (29–39)	no	100% birth facilities 100% nurses
Bulgaria	2013: 66,578	5% UNHS 90% OAE, 10% OAE-AABR		2.0			no	
Croatia	2011: 41,200 CAEHD[13], CUHZ[14]	99% UNHS 97% OAE, 3% OAE-AABR	0.6 16.7	1.0	5 (1–19)	5.2 (1–19)	yes, UNHS/ 2006	100% birth facilities 100% nurses
Cyprus	2013: 9,341 NGO[15], CUNHS[16]		0.4	2.3	1 (0.5-2)	4	no	10% birth facilities, 90% outpatient (community centers) 20% physicians, 10% audiologists, 70% nurses
Czech Republic	2013: 106,751 CSO[17]	81% UNHS 90% OAE, 10% OAE-AABR					yes, UNHS/ 2012	90% birth facilities, 10% ENT & pediatric departments 20% physicians, 10% audiologists, 70% nurses
Denmark, CDR[18]	2013: 12,604 EPF[19]	99.1% UNHS 88% AABR, 12% OAE-AABR	1.7 0.0	1.7	0.7 (0–2)	7.6	yes, UNHS/ 2005	100% birth facilities 30% nurses, 70% midwives
Estonia	2012: 14,056 TUC[20]	99.3% UNHS 100% OAE-AABR	0.2 0.0	1.7	6.7 (0–10)	6.7	yes, UNHS/ 2004	87.5% birth facilities, 12.5% child hospitals 100% midwives
Finland	2013: 59,856 NIHW[21], ES[22], MHHUCH[23]	96% UNHS 100% OAE-AABR	1.2 0.0	1.2	6 (3–36)		yes, UNHS/not reported	100% birth facilities 100% nurses
France	2016: 744,296 AFDPHE[24], ARS[25]	97.6% UNHS 36% OAE, 64% OAE-AABR	1.5 37.1	1.0			yes, UNHS 2012	100% birth facilities 10% audiologists, 30% midwives, 60% auxiliary nurses
Germany	2013: 682,069 REGNHS[26], SoH[27]	96% UNHS 38% OAE, 42% OAE-AABR & 20% AABR, NICUs[28]: 83.8% OAE-AABR or AABR[26]	5.5 20.8	1.3–1.7 (bilat.)[26]; 2.7 (uni- and bilat)[26]	4 (0–46)[26] 25	5 (1–46)[26] 28	yes, UNHS/2008	76% birth facilities, 4% pedaudiology institution, ENT & pediatric practice, 20% NICUs[28] 1% physicians, 5% audiologists, 92% nurses, 2% midwives
Greece	2013: 94,134	30% UNHS 95% OAE, 5% OAE-AABR	4.0 87.5	0.5	5 (2–11) 25 (12–42)	5 25 (12–42)	no	100% birth facilities 100% physicians
Hungary	2013: 88,689	>50% UNHS 100% OAE					yes, UNHS/2009	100% birth facilities 30% audiologists, 70% nurses
Iceland	2013: 4,323 NHSI[29], DH[30]	92% UNHS 100% OAE	0.4 0.0	0.9	1.2 (0–3)	2	no	89% birth facilities, 11% NHSI[29] 21% audiologists, 78% nurses, 1% midwives
Ireland	2013: 68,930 ESP[31]	99.8% UNHS 84.6% OAE, 15.4% OAE-AABR (NICU[28])	3.1 3.2	1.2	0.13 (0–12)		no, NHS/2011	99.4% birth facilities, 0.6% homes 21% audiologists, 78% nurses, 1% midwives
Italy	2011: 527,308 NSNHS[32]	79.7% UNHS 91% OAE, 8% OAE-AABR, 1% AABR	3.8 0.0	2.0	5 (3–7) 14 (9–18)	12 18 (12–24)	yes, UNHS/not reported	100% birth facilities 11% pediatricians, 2% audiologists, 72% nurses, 11% technicians, 4% CHW[9]
Latvia	2013: 20,251 NIS[33], LCHC[34]	95% UNHS 95% OAE, 5% OAE-AABR	5.3 0.0	2.0	11.5 (1–24) 36 (12–48)	8 22 (3–60)	yes, UNHS/not reported	100% birth facilities 100% nurses
Liechtenstein	2013: 339	no newborn hearing screening						
Lithuania	2013: 3,977[35] KCP[36]	77% UNHS 100% OAE	5.6 0.0	2.9	2 (1–8)	nd	yes, UNHS/2014	100% birth facilities 100% nurses
Luxembourg	2013: 6,889 MH[2]	98.6% UNHS 96.8% OAE, 3.2% AABR (NICU[28])	4.2 22.0	0.7	8.25 (4–18) 3 (3–6)	12.75 (6–21) 6 (3–18)	yes, UNHS 2007	85% birth facilities, 15% MH[2] 100% audiologists
Macedonia	2013: 23,138	no screening						
Malta	2013: 4,127	10% UNHS 100% OAE		1.5	0.5 (0.1–1.0) 18 (screen+non-screen)	20 (screen+non-screen)	no	10% birth facilities, 90% ENTOP[37] 10% physicians, 50% audiologists, 40% auxiliary nurses
Moldova	2017: 34,060	11.70% 65% OAE-AABR or questionnaires			5 (1–9) 55 (26–84)	10 (4–16)[38] 72 (24–84)[39]	no	70% birth facilities, 15% homes, 15% other outpatient places 90% audiologists, 5% nurses, 1% midwives, 4% CHW[9]
Monaco	2013: 964	95% 90% OAE, 5% AABR, 5% OAE–AABR					yes, UNHS/2014	100% birth facilities 10% nurses, 90% midwives
Netherlands	2013: 171,341, 167,490[a,39] CBS[40], NSDSK[41], TNO[42], RIVM[43]	99.4%[39] 99.7%[39] OAE-AABR, 0.3%[39] AABR	0.3 0.0	1.2[39,44]	1.7	5.3	yes UNHS/2015 (revised version)	75% at homes, 25% well baby clinics 100% CHW[9]
Norway	2014: 59.084, 4.024[a,47]	95% 95–97% TEOAE, 3–5% TEOAE-AABR	5.3 0.0	2.0	5 (0.5–10)	6	yes, UNHS/2008	100% birth facilities 5% audiologists, 95% nurses
Poland	2013: 368,576 PUNHSP[46]	95.80% 100% OAE	7.5 9.7	3.0	3 (0–17)	4.2 6.4	yes, UNHS/not reported	98% birth facilities, 2% audiology centers 100% audiologist/nurses/midwives

Appendix B (cont.)

Country-Specific Information on Newborn Infant Hearing Screening (NIHS) as Retrieved by Informant Replies

Region / Country	Reporting year*: # of live births / Data source**	% of newborns screened / Method used (%)	Fail rate / Lost-to-follow-up	Babies with PCHL[1] per 1000	Mean age in months (range) at diagnosis of screened / and non-screened babies	Mean age in months at treatment start of screened / and non-screened babies	Screening mandated? / Year	Where was screening done? / Who screened?
Portugal	2013: 82,787	59.30%	2.5	0.7	7 (3–36)		no	100% birth facilities
	45,750[a,47]	95% OAE, 0.5% AABR, 4.5% OAE-AABR	0.7		14 (12–36)			10% physicians, 60% audiologists, 30% nurses
Romania	2013: 192,547	57.50%	3.6		5 (3–10)	9	no[49]	85% birth facilities, 15% audiology centers
	UNHS NP[48]	95% OAE, 5% OAE-AABR	27.7		30 (18–48)	30 (20–48)		40% physicians, 10% audiology staff, 50% nurses
Russia	2013: 1,896,000	94.10%	2.4	3.0	3.6 (1.1–7)	6.8	yes, UNHS/2008	87.5% birth facilities, 22.5% outpatient pediatric units
	MH[2], audiology centers	78% OAE-AABR, 1.6% ASSR[50], 4.3% questionnaires	0.4		26 (8–49)	27 (8–56)		5% physicians, 6% audiologists, 89% nurses
Serbia	2013: 65,554	10% sporadic, institute based		1.6	12	13	no	100% birth facilities
		100% OAE						100% nurses
Slovakia	2013: 54,986	99.60%	6.5	1.2	9 (4–26)		yes, UNHS/2006	100% birth facilities
	DPENTD[51]	98% OAE, 2% OAE-AABR	0.0					20% audiologists, 80% nurses
Slovenia	2013: 20,593	96.20%	2.1	0.8	5 (3–17)	5	yes, UNHS/2005	100% birth facilities
	NIS[33]	100% OAE	0.0					100% nurses
Spain	2012: 445,648	>95%	3.7	1.7[53]	4 (3–6)[53]	5[53]	yes, UNHS/2010	90% birth facilities, 10% outpatient consults
	PAGR[52]	50% AABR, 50% OAE-AABR	0.0		15.3[53]	17[53]		100% nurses
Sweden	2014: 114,907	>95%	2.1	1.8	6 (1–14)	6 (2–14)	no	98% birth facilities, 2% ENT/audiology clinics
		95% OAE-AABR, 5% AABR	0.0		38	38		30 audiologists, 30% nurses, 40% midwives
Switzerland	2012: 80,363	98%	2.1	2.0	6 (1–18)	6	no	95% birth facilities, 5% homes
	NS[54], SUS[55]	99% OAE, 1% OAE-AABR	14.3		12 (6–36)	12		1% physicians, 1% audiologists, 88% nurses, 10% midwives
Turkey	2012: 1,292,380	98%	5.1	3.0			yes, UNHS/not reported	
	CSH[56]	OAE, AABR, OAE-AABR[56]						
Ukraine	2016: 397,039	no UNHS, 10% sporadic		2.8	12 (screen+non-screen)	13 (screen+non-screen)	no	
		100% OAE-AABR						
United Kingdom (England only)	2013: 778,803, 685,100[d,57]	98.95%	2.6	1.6	1.6	1.64	yes, UNHS/2001	81% birth facilities, 19% homes
	NSIS[58]	100% OAE-AABR	10.4					25% CHW[9], 75% specially trained healthcare assistants
Africa								
Algeria	2014: 1,014,248	no screening		3.2	3 (3–3)[61]	461	no	
	RNCSTO[59], RNWTO[60]	1.77%[61] OAE, OAE-AABR[61]						
Benin	2014: 376,439 (2012)	no screening			143 (60–216)[65]		no	
	HS[62], ESC[63], CNHU-HKM[64]				84 (12–216)[6]			
Botswana	2012: 40,856	no UNHS, some targeted preterm/ low birthweight from 3 months on					no	2 maternity hospitals
		100% OAE						100% audiologists
Cameroon	2011: 716,000	no UNHS, later<1%, targeted<5%					no	100% ENT services
		20% OAE, 80% OAE-AABR						100% physicians
DR Congo	2015: 2,486,485	no UNHS, 0.07% later		3.0			no	hospitals with ENT/audiology services
		60% OAE, 40% distraction test			36 in cities, 60 rural	48–72		75% physicians, 25% audiologists
Côte d'Ivoire	2011–2012: 738,800,	no UNHS, 0.2% NHS		6.0		5.5 (3.5–7.5)	no	
	1306[a,66]	100 OAE-AABR						
Djibouti	2016: 25,000 (2005–2010)	no UNHS, but in preparation						
Egypt	2012: 2,629,769	8%, no systematic UNHS, 8% targeted[67]		9.0	6 (3–18)	6	yes targeted/2012	40% birth facilities, 60% NICUs[28]
	1200[a,67]	100% AABR			12 (6–24)	12		80% audiologists, 20% nurses
Ghana	2013: 463,409,	0.83% sporadic	16.9	3.0	4 (2–8)	18	no	HACKBTH[68]
	3,893[a,68], HACKBTH 0[68]	100% OAE	0[68]		36 (18–60)	72 (18–144)		30% audiologists, 70% trained national service staff
Guinea	2014: 380,000 (2005–2010)	no UNHS, 2% targeted			67.5 (screen+non-screen)	88.1 (screen+non-screen)	no	1 ENT clinic (Conakry)
	MH[2]	100% AABR						100% physicians
Kenya	2014: 1,534,900 (2012)	no UNHS, some screening of older children					no	3 hospitals
		100% OAE						100% nurses
Madagascar	2014: 698,000	no UNHS[69]					yes, targeted/2011	
	(2005–2010)							
Malawi	2014: 516,529 (2008)	<1%, sporadic			3 (0–5)			some baby screenings by 2 clinics with outreach programs
		100% OAE			120			100% audiologists
Namibia	2015: 60,000,	1.8%[70] hospital-based					no	2 private hospitals
	1,077[a,70] (2005–2010)	100% OAE[70]						100% audiologists[70]
Nigeria	2014: 7,117,000	<1% UNHS			36 (screen+non-screen)	84 (screen+non-screen)	no	
		65% OAE, 35% AABR						1% physicians, 99% audiologists
São Tomé and Príncipe	2016: 5,000 (2005–2010)	Portuguese NGO does screening 3–4 times per year, mostly targeted						
	IMVF[71]	100% OAE						
Seychelles	2014: 1,577	100% UNHS	0.6	2.0	9 (6–15)	9	yes UNHS/2014	95% birth facilities, 5% outpatient
	MH[2]	100% OAE-AABR	0.0					2% audiologists, 58% nurses, 40% midwives
Sierra Leone	2013: 224,000 (2005–2010)	0.01% targeted[72]					no	2 centers
		100% OAE-AABR						
South Africa	2013: 1,084,397	10% UNHS					no	80% birth facilities, 20% other clinics
	JA[73]	81% OAE, 9% AABR, 10% OAE-AABR			42 (2.2–128.2)[74]			80% audiologists, 15% nurses, 5% trained screeners
Tunisia	2013: 221,147	30–40% UNHS, 60% later		3.7		24	yes, targeted/not reported	100% birth facilities
	CNHT[75]	60–80% OAE, 10–20% AABR						70–80% CHW[9]

Appendix B (cont.)

Country-Specific Information on Newborn Infant Hearing Screening (NIHS) as Retrieved by Informant Replies

Region / Country	Reporting year*: # of live births / Data source**	% of newborns screened / Method used (%)	Fail rate / Lost-to-follow-up	Babies with PCHL[1] per 1000	Mean age in months (range) at diagnosis of screened / and non-screened babies	Mean age in months at treatment start of screened / and non-screened babies	Screening mandated? / Year	Where was screening done? / Who screened?
Zimbabwe	2016: 370,000 (2005–2010) AM[76]	sporadic					no	2 audiology practices
		100% OAE						100% audiologists
Asia								
Armenia	2017: 37,699	62.40%	3.2	1.2	6	6	yes, UNHS/not reported	100% birth facilities
		100% OAE	17.1		14	16		20% audiologists, 80% nurses
Bahrain	2014: 21,037 (2015)	Hospital-based						
Cambodia	2013: 330,000 (2010)	sporadic					no	
	AEC[77]	100% OAE			36 (6–216)	36 (6–216)		100 mid-level primary ear and hearing care clinicians
China	2013: 16,400,000	69% UNHS, 30% later, 0.1% targeted	8.7	2			yes/1999	100% birth facilities
	NRMCH[78]	90% OAE, 5% AABR, 5% OAE-AABR						100% nurses
Georgia	2013: 57,878	39.2% UNHS	8.9	0.3				
	NSOG[79]	100% OAE	0.0					100% researchers
India	2014: 27,271,000 (2005–2010)	<1%, regional UNHS[80]	10.24[80]	5–6			no	
	SIDK[80]	100% OAE[80]						5% physicians, 50% audiologists, 10% nurses, 15% trained screeners, 20% CHW[9], junior public health nurses[80]
Indonesia	2012: 4,464,000 (2005–2010)	sporadic		1.0	3 (6–9)	6	no	100% birth facilities
	MH[2], IORLS[81]	30% OAE, 10% OAE-AABR, 60% OAE-ABR click	19.6		28 (18–36)	36 (18–48)		90% physicians, 10% audioogists
Iraq	2015 & 2018 about 1,079,000 7326 (2 studies)	sporadic	9.5	0.5 (2 studies)			yes, UNHS/2017	100% audiology centers
		100% OAE						100% physicians
Iran	2013: 1,427,653	66.4%, 63% UNHS, 3.4% targeted		2.6	1 (1–3)	6	no	95% birth facilities, 5% outpatient centers
	SWOI[82], ENTHNRC[83]	20% OAE, 80% OAE-AABR			42 (24–60)	46 (28–64)		65% audiologists, 10% nurses, 5% midwives, 20% CHW[9]
Israel	2012: 170,940	99.1% UNHS		0.8[85]	3.7 (0–39.3)[86]	9.4[86]	2010 directive given for UNHS	100% birth facilities
	NSI[84]	100% OAE-AABR			9.5[87]	19.3		20% audiologists, 80% trained screeners & biotechnic
Japan	2013: 1,029,816	62% UNHS	1.5	1	1–3 (0.25–14.0)	6	no	98% birth facilities, 2% pediatric clinics
	SORLJ[88], SJAOG[89], SOP[90]	28% OAE, 63% AABR, 9% OAE-AABR	0.0		48 (4-nd)			8% physicians, 1% audiologists, 39% nurses, 35% midwives, 17% technicians
Jordan	2013: 178,000	68%, 67% UNHS, 1% targeted	11.8	6.0	4.3 (0.2–0.9)	4.6	no	90% birth facilities, 10% other
	MH[2], UNHSPJ[91], SACJ[92]	99% OAE, 1% AABR	37.5		1.6 (0.6–5.4)	1.8 (0.6–8)		30% audiologists, 70% nurses
Kazakhstan	2013: 363,123	85.2% UNHS					yes, UNHS/2009	100% birth facilities
	MH[2]	100% OAE						
Korea, North	2013: 356,000	no screening					no	
	AEI[93]							
Korea, South	2014: 435,435	90.1%, 86.7 UNHS, 3.4% targeted[94,95]	1.7	7.1[94]	3(1.5–6)		directive given for UNHS	98% birth facilities, 2% homes/other places
		9.8% OAE, 90.2 AABR, 72% later[94]	80.7					19.3% physicians, 29% audiologists, 51.7% speech pathologists
Kuwait	2014: 61,313	UNHS started in 2013 in 5 hospitals					not reported	
Malaysia	2013: 571,000 (2005–2010) 34,884 UNHS, 53,647 UNHS+targeted[a,96]	3.6% UNHS, 7.6% targeted		1.4			no	100% hospitals[96]
		52.4% AABR, 47.6% OAE-AABR[96]	97.4					25% audiologists, 75% nurses[96]
Mongolia	2018: 78,444	Mongolia total 2013: 60% UNHS, 100% AABR; Ulaanbatar 2018: 100% UNHS, OAE-AABR		Mongolia: 1, Ulaanbatar: 3	Mongolia: nd (6–24); Ulaanbatar 3.5		yes, for Ulaanbatar City/2009	100% birth facilities
	NCMCH[97]	100% AABR			30			100% audiologists
Myanmar	2014: 836,961	20.2%, 14.8% UNHS[98], 5.4% targeted[98]	3.4	35.0[98]	2 (1–3)	2	no	100% birth facilities
	HARNUCWHM[98]	100% OAE-AABR	0.0		36	36		50% physicians, 50% audiologists
Nepal	2012: 593,300	sporadic, 4% at risk			12 (3–36)		no	birth facilities, outpatient, homes
	IOMK[99]	100% OAE-AABR[99]			36 (12–60)[99]	12 (nd)[99]		100% audiologists[99]
Oman	2014: 71,650	96,6% UNHS, targeted 0.04%	1.7	1.0	6 (1–12)	nd (6–36)	yes, UNHS/1996	99% birth facilities, 1% mother–child clinics
	OECP[100]	100% OAE	0.0					nurses, 90% midwives
Pakistan	2013: 4,666,000 (2005–2010)	2% UNHS, 3% later, 2% targeted	2.9	15.0	18 (3–24)	24	no	10% birth facilities, 90% audiology clinics
	PDHS[101]	70% OAE, 5% AABR, 25% OAE-AABR			30 (3–5)	36		100% audiologists
Palestine	2011: 121,493	no UNHS, 3% later	4.7				no	birth facilities, outpatient
	MH[2], CBHB[102]	97% OAE, 3% AABR						100% nurses
Philippines	2013: 2,318,000 (2005–2010) 2,562[a,103]	no UNHS		0.0[103]			yes, UNHS/not reported	
		100% OAE[103]						
Qatar	2012: 18,067	97% UNHS	3.1	1.8	0.5 (0.3–2)	2–3	yes, UNHS/2003	100% birth facilities, private clinics
	HMC[104]	100% OAE-AABR	0.0		3 (3–8)	3 (3–8)		100% audio-physicians
Saudi Arabia	2014: 569,000 (2005–2010)	hospital-based UNHS or targeted		1.8[105]			not reported	
Tajikistan	2014: 187,000 (2005–2010)	1% UNHS, 0.5% later, 0.5% targeted			2 (1–12)	2	no	33% physicians, 33% audiologists, 33% nurses
	NMCT[106]	100% OAE-AABR			1 (1–1)	2 (1–24)		
Thailand	2014: 748,081 (2013)	estimated 70% hospital-based UNHS					no	most provincial/regional hospitals
		100% OAE						nurses, technicians

Appendix B (cont.)

Country-Specific Information on Newborn Infant Hearing Screening (NIHS) as Retrieved by Informant Replies

Region / Country	Reporting year*: # of live births / Data source**	% of newborns screened / Method used (%)	Fail rate / Lost-to-follow-up	Babies with PCHL[1] per 1000	Mean age in months (range) at diagnosis of screened and non-screened babies	Mean age in months at treatment start of screened and non-screened babies	Screening mandated? / Year	Where was screening done? / Who screened?
Uzbekistan	2013: 679,519				1.5–1.8(1–36)		yes, UNHS/2013	48% neonatologists, 42% audiologists, 8.5% nurses, 1.5% others
	IHU[107]							
Vietnam	2013: 1,500,000	1–3% UNHS, 5% targeted		4	24 (3.15)	12	no	10% birth facilities
	CHHCMC[108]	100% OAE, 100% behavioral test			24 (3–40)			25% physicians, 75% nurses
North America								
Canada	2013: 383,800	64% UNHS, 5% later, 4% targeted	1.9	2.0	3 (1–12)	5	no	85% birth facilities, 15% community health centers
	PLPD[109]	3% OAE, 9% AABR, 88% OAE-AABR	8.3					5% audiologists, 10% nurses, 1% midwives, 84% CHW[9]
Mexico	2013: 2,243,352	23.3% UNHS[110]		2.5[111]	12 (6–20)[111]		yes, targeted/not reported	100% birth facilities
	CONADIS[110]	100% OAE			35[112]			33% physicians, 67% nurses
United States	2016: 3,830,526	94.8% UNHS, 3.8% later	1.6	1.7			yes, UNHS in 43 of 50 states	98% birth facilities, 2% homes
	CDC[113], USDHHS[114]	40% OAE, 50% AABR, 10% OAE-AABR	53.3					
Central & South America								
Argentina	2013: 754,603	12% (Arg), 89% UNHS, 20% later, 1% targeted[115]	2.8	3[115]	8 (2–36)[115]	8[115]	yes, UNHS/2008 (Province de Cordoba)[115], 2011 (Argentina)	80% birth facilities, 20% community & private centers
	3,983[a,115]	100% OAE-AABR (well babies), 100% AABR (at-risk babies)[115]	0.0					100% audiologists
Barbados	2014: 2,902	targeted						pediatricians, nurses
		100% OAE						
Bolivia	2013: 313,638	58.1%, 23.3% UNHS, 31.7% later, 3.1% targeted		4.1			not reported	100% birth facilities
		100% OAE (NHS), 57% questionaires						56% physicians, 44% audiologists
Brazil	2012: 3,073,000 (2010–2015) 2,190,398[a,116]	Brazil total: 24%[116] UNHS; São Paulo: 97.7%[117] UNHS	0.2[117]				yes, UNHS/2010	hospitals, maternity wards, primary health care centers, hearing health care units
	MH[2,116], MHSP[117]	Brazil total: OAE, AABR, OAE-AABR; São Paulo: 100% OAE-AABR (well babies 87.9%), 100% AABR (at-risk babies 9.8%)[117]	43.5[117]					100% audiologists
Chile	2016: 231,749 MH[2], HPH[118]	39% UNHS, 1% targeted	5.0	3.0	1 (0.7–3)[118]	4.4 (max. 11.2)[118]	yes, targeted /2005	100% hospitals
		80% OAE, 20% AABR	0.0		24	30		94% audiologists, 6% nurses
Colombia	2013: 764,000 (2010–2015)	2.3% UNHS, 2% later, 2% targeted[119]	15.9[119]	4.0[119]	2 (0–36)	12	yes, UNHS/not reported	10% birth facilities, 90% outpatient places
	FSFB[119]	8.1% OAE, 86.5% AABR, 5.4% OAE-AABR, pediatric clinic child history 10%[119]	40.0[119]		48 (48–72)	36 (48–72)		1% physicians, 99% audiologists
Costa Rica	2013: 70,550	Not reported					yes, UNHS/2013	80% birth facilities, 20% outpatient places
	CCSS[120]	100% OAE						10% physicians, 90% audiologists
Dominican Republic	2012: 150,581	0.09% targeted (preterms)					no	
		100% OAE						audiologists
Guatemala	2013: 387,342	0.09% targeted[121]	5.6	2.9[121]	12	12–24	no	1% birth facilities, 99% outpatient (2 audiology clinics, 1 pediatric clinic)
	FSQE[121]	100% OAE	40.0		36 (1–96)	48 (18–72)		1% pediatricians, 99% audiologists
Jamaica	2012: 39,553	sporadic					no	various places
		100% OAE-AABR						mainly audiologists
Nicaragua	2013: 132,165 (2010)	65%, 40% UNHS, 20% later, 5% targeted[122]	30.8	6.0[122]	1 (0–10)	48[122]	no	20% birth facilities, 80% diagnostic centers
	private hospital[122]	100% OAE (NHS), 50% pediatr[122]	50.0		60 (36–96)	30[122]		10% physicians, 1% audiologists, 89% trained screeners
Panama	2013: 73,804	82.5%, 66% UNHS, 15% later, 1,5% targeted[123]	4.9	1.0[123]	6 (2–12)[123]	7.9[123]	no	93.5% birth facilities, 6.5% private clinics
	SDHNPC[123]	92% OAE, 4% AABR, 4% OAE-AABR[123] (NHS), 2% tymp & AABR	75.0		90 (24–156)[123]	36 (24–156)[123]		96% audiologists, 4% neurophysiological technicians
Puerto Rico	2013: 38,986	97.5% UNHS	2.8	1.4[124]			yes UNHS/2003	98% birth facilities, 2% extramural
	PRDH[124]	100% OAE-AABR (well babies), 100% AABR (NICUs[28])						70% audiologists, 30% trained nurses
Trinidad and Tobago	2010–2015: 619,000	0.005% targeted					no	
		70% OAE, 30% OAE-AABR						
Uruguay	2010–2015: 49,000	99% UNHS, 0.06% targeted	0.5	0.7	6 (4–8)	6	yes UNHS/not reported	99.9% birth facilities, 0.1% homes
		97% OAE, 3% OAE-AABR	0.0					10% physicians, 25% audiologists, 65% nurses
Australia & Oceania								
Australia	2017: 309,142 (2013)	97% UNHS		0.9	1–1.8 (medians according to states)		no, but UNHS is fully funded government policy	>90% birth facilities, <1% homes, <10% outpatient clinics & community health centers
	various sources[125]	100% AABR						state-employed hearing screeners, nurses, or midwives
Marshall Islands	2011: 977[126]	96.8% UNHS	11.6	0.0			no	
	CDC & USDHH[126]		98.2					
Micronesia, Federated States of	2013: 2,555 (2018)	91.60%		1.4			no	
		100% OAE-AABR						
New Zealand	2013: 59,245	88.2% UNHS	2.2	1.1	2.1 (<1–11)/nd		no, but nationally managed UNHS & Early Intervention Program	Maternity hospitals, outpatient clinics, homes (rarely)
	NNHSDB[127]	84% OAE-AABR (well babies), 16% AABR (at-risk)	15.8					Certified newborn hearing screeners
Palau	2013: 229	99.30%					no	
		100% OAE-AABR						

Appendix B (cont.)

Country-Specific Information on Newborn Infant Hearing Screening (NIHS) as Retrieved by Informant Replies

Note. No information obtained from the following countries: Andorra, Azerbaijan, San Marino, United Kingdom (Scotland, Wales, Northern Ireland), Burundi, Cape Verde, Central African Republic, Comoros, Congo (Brazzaville), Eritrea, Gabon, Liberia, Mauritania, Mauritius, Niger, Sudan, Kingdom of Eswatini, Afghanistan, Brunei, Kyrgyzstan, Laos, Lebanon, Maledives, Singapore, Sri Lanka, Yemen, Bahamas, Belize, Cuba, Dominica, Ecuador, Grenada, Guyana, Paraguay, Peru, Saint Lucia, Saint Vincent and the Grenadines, Suriname, Venezuela. NIHS was not yet established in the reporting period in the following countries: Kosovo, Montenegro, Angola, Benin, Burkina Faso, Chad, Djibouti, Equatorial Guinea, Ethiopia, Gambia, Guinea-Bissau, Lesotho, Libya, Mali, Morocco, Mozambique, Rwanda, Senegal, Somalia, South Sudan, Tanzania, Togo, Uganda, Zambia, Bangladesh, Bhutan, Iraq, North Korea, Syria, Timor-Leste, Turkmenistan, United Arab Emirates, Antigua and Barbuda, El Salvador, Haiti, Honduras, Saint Kitts and Nevis, Fiji, Kiribati, Nauru, Papua New Guinea, Samoa, Solomon Islands, Tonga, Tuvalu, Vanuatu.
ABR = automated auditory brainstem response; NHS = newborn hearing screening; OAE = otoacoustic emissions; TEOAE = transitory evoked otoacoustic emissions; UNHS = universal newborn hearing screening

*Pertains to all questionnaire data; reporting year for # of life births is frequently different; **data on life births from national statistical institutes, UN Population Department, UN Demographic Yearbooks, or GeoStat; only NIHS-related data sources indicated by superscript; [a]this birth number refers to a subpopulation, upon which the following data about this country are based.

[1]permanent childhood hearing loss, [2]Ministry of Health/Healthcare, [3]Statistik Austria (www.statistik.at), [4]Tyrolean Childhood Hearing Loss Register, [5]National Centre of Otorhinolaryngology, [6]Kind & Gezin, [7]Dépistage de la surdité chez les noveau-nés, [8]Zorg en Gezondheid, [9]community health workers, [10]data from two of seven screening maternity clinics of a total of 13 maternity clinics, [11]birth registry, [12]ENT department University Hospital Banja Luka, [13]Croatian Association for Early Hearing Diagnostics, [14]Children's University Hospital Zagreb, [15]non-governmental organization responsible for all pre-and neonatal screenings, [16]Cyprus UNHS, [17]Czech Statistical Office, [18]Central Denmark Region (1.25 mio inhabitants, 20% of Danish population), [19]electronic patient file system EPF, used to register all patients in a region including screening and diagnostics, [20]Tartu University Clinic, [21]The National Institute for Health and Welfare, [22]etiology study, [23]Maternity Hospital of the Helsinki University Central Hospital, [24]Association Française pour le Dépistage et la Prévention des Handicaps de l'Enfant, [25]Agences Régionales de Santé, [26]Report on evaluation of the German newborn hearing screening 2011/2012, [27]data from a population-based study with 17,439 screened newborns, performed in the Federal State of Hesse (Neumann et al. 2006), [28]neonatal intensive care unit, [29]The National Hearing and Speech Institute of Iceland, [30]Directorate of Health, [31]E-Screener Plus, [32]nationwide surveys on NHS, [33]National Institute of Statistics, [34]Latvia Children Hearing Center, [35]data available only of the Kaunas region, but representative, [36]Kaunas Center of Perinatology, data only for Kaunas region, [37]Ear, Nose, and Throat outpatient, [38]data from Republican Center of Audiology , [39]data without NICU babies, [40]Centraal Bureau voor de Statistiek, [41]Nederlandse Stichting voor het Dove en Slechthorende Kind, [42]Netherlands Organisation for Applied Scientific Research, [43]Rijksinstituut voor Volksgezondheid en Milieu, [44]detection threshold NHS 40 dB, [45]Statistics Norway, [46]Polish Universal Neonatal Hearing Screening Program, [47]data from 27 out of all 50 maternity hospitals, [48]UNHS National Program, [49]screening obligatory only for institutions included in the pilot National UNHS Program, [50]auditory steady-state response, [51]database of pediatric Ear, Nose and Throat Department, [52]Principado de Asturias government registry, [53]data from Asturias region (7445 babies), [54]national survey 2012, [55]study of the University Hospital Zurich 2005-2010, [56]data from Çorlu State Hospital region of 11,575 neonates screened between Sept. 2009 and November 2012, [57]data from April 1, 2012, to March 31, 2013, national screening IT system, [58]national screening database, [59]Registre des naissance de la clinique Sbihi de Tizi Ouzou and [60]Registre des naissances de la wilaya de Tizi Ouzou, [61]Farid Boudjenah doctoral thesis "Dépistage et réhabilitation de la surdité et néonatal au CHU de Tizi-Ouzou: stratégies et résultats", [62]hospital statistics, [63]Ecoles de sourds de Cotonou, [64]Centre National Hospitalier Universitaire Hubert K. Maga Cotonou, [65]data from a thesis, [66]data from study, [67]data from Ain Shams University Hospital, [68]data from Hearing assessment center Korle Bu Teaching Hospital Accra, the only institution that performs UNHS in Ghana, [69]only screenings for 321 children (0.5-0.6yrs) from 1 clinic reported, [70]data from Namibia Hearing Care Institution, [71]Instituto Marques de Valle Flor, [72]of babies and young children who received ototoxic drugs, [73]journal articles, [74]Swanepoel et al. 2013, [75]study for Charles Nicolles Hospital Tunis with 3260 babies, [76]AudioMax Zimbabwe, [77]All Ears Cambodia,[78]National Report of Maternal and Childrens' Health (2008-2015), [79]National Statistics Office of Georgia, [80]data from State Initiative on Disabilities (SID), Kerala, Social Security Mission, Govt. of Kerala: UNHS program with 40 involved governmental maternity hospitals with ≥100 deliveries per year and 412,164 newborns screened between Oct. 2014 and Aug. 2018, [81]Indonesian ORL Societies, [82]State Welfare Organization of Iran, [83]ENT and Head & Neck Research Center, Iran University of Medical Sciences, Tehran, [84]National Survey Israel data from January 1, 2012, to September 30, 2012, [85]data from 2010-2011 National Survey for children enrolled in rehabilitation centers, [86]data for children enrolled in rehabilitation centers, [87]data from 2007-2009 before UNHS was established, [88]Survey by the Oto-Rhino-Laryngology Society of Japan, [89]Survey by Japan Association of Obstetricians and Gynecologists, [90]Statistics of Okayama Prefecture, [91]UNHS Program Jordan, [92]survey from the main audiology clinics in Jordan, [93]All Ears International DPRK program, [94]NHS results from nation-wide low-income class newborns, [95]data from national infant health examination 2014, [96]data from 4 government hospitals which run UNHS and 26 hospitals which run targeted screening, [97]National Center for Maternal and Child Health, [98]Hospital admission registry, Neonate Unit, Central Women Hospital, Mandalay, data of 6876 newborns, [99]data from hospital records of Institute of Medicine Kathmandu, [100]Oman Ear Care Program, [101]Pakistan Demographic and Health Survey, [102]Caritas Baby Hospital Bethlehem, internal data, [103]pilot study in selected hearing screening centers in Metro Manila, Bulacan, and Pampanga, [104]Hamad Medical Corporation, [105]Habib HS, Abdelgaffar H. Neonatal hearing screening with transient evoked otoacoustic emissions in Western Saudi Arabia. Int J Pediatr Otorhinolaryngol. 2005 Jun;69(6):839-42. doi: 10.1016/j.ijporl.2005.01.018 PMID: 15885338: data from 11986 neonates, [106]National Medical Center Tajikistan, [107]Institute of Health Uzbekistan, [108]Children Hospital No1 HochiMinh City, [109]provincial & local program databases, [110]Consejo Nacional para el Desarrollo y la Inclusión de las Personas con Discapacidad, [111]data from General Hospital of Mexico, México City, DF (based on 5,000 newborns/year), [112]López VMM, Chamlati, E, & Berruecos VP (1997) Hearing loss prevention levels in Mexico; a multicenter study. 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