

21st Century Teenagers and Young Adults who are Deaf or Hard of Hearing: Outcomes and Possibilities

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Abstract: The purpose of this study was to document demographics, characteristics, and long-term outcomes of teenagers and young adults who are deaf or hard of hearing (DHH) and who all attended the Moog Center for Deaf Education for preschool and/or a portion of elementary school. Because it is not an experimentally controlled study, it does not establish causal relationships among outcomes and variables describing the intervention program or the participants. It does provide valuable data about the possibilities for children who are deaf or hard of hearing and identifies variables associated with positive outcomes that can be more closely examined in future experimentally controlled studies. Data were obtained via an online survey from 108 individuals who were DHH and had attended the Moog Center for Deaf Education. The survey assessed educational, employment, and personal outcomes of individuals who were currently in high school and beyond (15–32 years of age). Results indicate this group of individuals obtained high levels of achievement in terms of educational attainment, employment experience, social involvement, and communication competence.

Key Words: deaf education, listening and spoken language intervention, long-term outcomes, teenagers and young adults who are deaf or hard of hearing

Acronyms: CART = Communication Access Real-time Translation; CI = cochlear implant; DHH = deaf or hard of hearing; EHDI = early hearing detection and intervention; HA = hearing aid; IDEA = Individuals with Disabilities Education Act; PTA = pure tone average; SLP = speech-language pathologist; TOD = teacher of the deaf

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Over the past 25–30 years, several important changes in technology and public policy have resulted in a monumental shift in the education of children who are deaf or hard of hearing (DHH) and dramatically increased the potential outcomes and opportunities for these children. Changes in technology include advancements in hearing technology, and information and communication technologies. Changes in public policy include federal laws such as the Early Hearing Detection and Intervention (EHDI) Act of 2017 and the Individuals with Disabilities Education Act (IDEA, 2004).

1. Advancements in hearing technology in both hearing aids and cochlear implants have dramatically increased access to sound for individuals who are DHH. In addition, the age at which the FDA approved implantation of cochlear implants has decreased from the initial candidacy

criteria of 18 years in 1984, two years of age in 1989, and one year of age in 2000 (Sorkin, 2016).

2. Computers, captioning, social media, and other internet technology have expanded the ways in which individuals communicate and access information that have affected the lives of all individuals, with potential long-term benefits for individuals who are DHH.

3. Early Hearing Detection and Intervention (EHDI) programs have reduced the average age of identification of hearing loss. Prior to the 21st century, most children who were DHH were not identified until they were two to three years of age when parents noticed they were not talking (Toward Equality, 1988; White, 2014). Earlier identification has resulted in earlier intervention and earlier fitting of hearing aids (Harrison, Rousch, & Wallace, 2003;

Hoffman & Beauchaine, 2007). EHDl programs now exist in all 50 states with the purpose of ensuring that all infants are screened for hearing and that those identified with hearing loss are enrolled in early intervention as soon as possible. This has reduced the average age of identification of hearing loss by more than two years, to an average of three to six months (White, 2014). Additionally, the Centers for Disease Control and Prevention (CDC, 2018) reports 98% of all infants are now screened for hearing loss.

4. Individuals with Disabilities Education Act (IDEA) is a law that ensures that eligible students with a disability are provided with a free appropriate public education and related services that are tailored to their individual needs (IDEA, 2004).

As a result of these changes, opportunities for the current generation of teenagers and young adults who are DHH have exceeded those of past generations. Even as opportunities continue to expand, parents remain concerned about outcomes for their children (Szarkowski & Brice, 2016). Ninety-five percent of children who are DHH have at least one hearing parent (Mitchell & Karchmer, 2004). Research suggests that hearing parents of children who are DHH experience unique concerns (Hintermair, 2006; Pipp-Siegel, Sedey, & Yoshinaga-Itano, 2002). When hearing parents first find out that their child has a hearing loss, they are concerned about their child's future (e.g., Will my baby have friends? Be involved in sports? Go to college? Get a job?). EHDl service providers are often the first points of contact for new parents of children who are DHH, and parents look to them to answer these questions and express what can be expected for their child. Longitudinal outcome data are needed to answer these questions for the current generation of children who are DHH.

The current study begins to address those questions by describing the educational, employment, and related outcomes for 108 alumni from the Moog Center for Deaf Education. Because it is not an experimentally designed study, it does not establish cause and effect relationships among outcomes, children's characteristics, and the type of interventions they received. The study nonetheless provides valuable information about what is possible in the 21st century for children who are DHH.

The Moog Center is a listening and spoken language program for children who are DHH. All participants attended the Moog Center for a portion of their early education, including preschool and/or elementary school. Study participants ranged in age from 15 to 32 years at the time data were collected. To the authors' knowledge, this is the first longitudinal description of outcomes for teenagers and young adults who are DHH, in which all participants had attended the same deaf education program prior to entering a general education setting with their hearing peers. The information in this article helps to fill the gap in the deaf education literature about longitudinal outcomes

for children who are DHH after controlling for educational environment and instructional philosophy.

Method

This study received approval from IntegReview Institutional Review Board, Austin, TX (#201516). All individuals ages 15 and older at the time data were collected and who attended the Moog Center for at least one year were eligible to participate in the study. Data for this study were obtained from two sources: (a) the Moog Center's in-house database, and (b) an online survey created by the Moog Center's founding director. The in-house database contained historical data on each participant, including contact information, demographics, and audiological histories. The online survey, via Survey Gizmo, was designed to collect information about participants' educational, employment, and personal experiences in high school, higher education, and beyond.

Young adult participants, 18 years and older, were contacted via an email invitation. Teen participants were recruited by parental phone call and parental consent to contact the participant via a parent-provided email address. Contact information for alumni and parents of alumni was obtained from the school's database and social media. Email addresses for ten of 132 eligible alumni could not be procured, and four parents of high schoolers declined to consent for their children to participate, resulting in 118 emailed invitations to alumni for participation in the online survey.

The email contained a brief description of the study, including what the researchers hoped to learn, what would be expected for participation, an estimation of how much time the survey would take, and information about a compensation of \$50 for participants who completed the survey. The email also contained a link to the survey, and the first page of the survey contained the consent form for participating. Of the 118 alumni to whom surveys were sent, 108 (92%) consented to participate and completed the survey.

Survey questions inquired about education, employment, communication, use of technology, special recognitions received, and other aspects of the participants' lives after leaving the Moog Center. The survey was composed mostly of multiple-choice questions with a few open-ended questions. The survey used skip logic, a feature that leads participants through the survey based on their previous answers.

Participants

Of the 108 participants, 92% were identified with hearing loss before three years of age, and the remaining 8% were identified before five years of age. All participants met the following criteria: (a) attended the Moog Center program for at least one school year during preschool and/or elementary school, and (b) were above the age of 14 at the time of the study. The 108 respondents were divided

into two groups: (a) 44 high schoolers, henceforth referred to as Teens, and (b) 64 alumni who were beyond high school, henceforth referred to as Young Adults. Table 1 summarizes the characteristics of participants.

Table 1
Participant Characteristics

Attribute	Description	Teens <i>n</i> = 44	Young Adults <i>n</i> = 64	All Participants <i>N</i> = 108
Unaided PTA Better Ear	Pure tone average, mean thresholds in dB and range	90.6 (38.6 – NR)	98.3 (55 – NR)	95.2 (38.6 – NR)
Hearing Technology	Number with:			
	1 CI, no HA	8	32	40
	2 CI	18	17	35
	1 CI, 1 HA	9	7	16
	2 HA	9	8	17
Deaf of Deaf	<i>n</i> and % with parents who are DHH	1, 2%	3, 5%	4, 4%
Mother's Education	<i>n</i> and % with mothers who obtained degrees from higher education institutions	33, 75%	51, 80%	84, 78%
Performance IQ	Mean nonverbal intelligence quotient and range	112 (90–129)	107 (79–133)	109 (79–133)
First Amplification	Mean age in years at first hearing aid fitting and range	1.5 (0.2–4.5)	1.6 (0.1–5.0)	1.6 (0.1–5.0)
First Implant	Mean age in years at first cochlear implant and range	3.0 (1.0–7.8)	5.7 (1.8–25.0)	4.7 (1.0–25.0)

Note. NR = No Response at the limits of the audiometer; PTA = pure tone average; CI = Cochlear Implant, HA = hearing aid; DHH = deaf or hard of hearing.

Hearing Technology

On average, participants first received hearing aids by 18 months of age and 91% were amplified before three years of age. When asked about present-day use of hearing technology, 84% of participants reported use of at least one cochlear implant, and 16% reported wearing bilateral hearing aids. Among CI users, 41% of Teens and 27% of Young Adults reported bilateral implantation. More Teens (41%) than Young Adults (27%) were bilaterally implanted. All but one participant, who received his CI at age 25, responded that device(s) were worn most or all waking hours, excluding inappropriate times such as swimming, taking a shower, and/or sometimes in noisy places.

Preschool and Elementary Education

The Moog Center curriculum is based on a curriculum developed by Jean Moog during the Experimental Project in Instructional Concentration (Moog & Geers, 1985). The teens and young adults surveyed for this article were taught using this curriculum and it is still used today. The Moog Center provides a full-day spoken language program for preschool and elementary school children who are DHH. The program is intensive, focused, and

objective-driven. The two main components of the Moog Center programs are small-group instruction and large group instruction. Additionally, parent informational group meetings, parent support group meetings, and individual parent-child coaching sessions are available. Preschool children spend about half of the day in small groups for individualized therapy and the other half of the day in large groups. Small groups typically consist of two or three children with similar abilities in each spoken language area, including speech, vocabulary, language, and auditory skill development. Small groups allow for explicit instruction in each of these skills. For children in preschool, large groups typically consist of eight to twelve children in a classroom where the focus is on the development of motor skills, social skills, pragmatic skills, and preschool academic skills. The larger preschool classroom setting also provides natural opportunities for children to transfer specific learned spoken language skills to conversational settings in the context of preschool activities. Children in the elementary school program have a similar schedule for small group instruction for spoken language and reading development; medium sized groups of four children for elementary subjects such as written language, math, science, social studies, and critical thinking; and large groups of 8–12 for special activities, computers, centers, and physical education. Throughout the day, children in both the preschool and elementary school programs alternate between small and large group activities. Appendix A details sample daily teacher/learner schedules for both programs. Teaching staff include certified teachers of the deaf, speech-language pathologists, and early educators.

Audiology services are provided onsite by experienced pediatric audiologists for all school children. These services include objective and behavioral hearing evaluations, fitting and programming of hearing aids, cochlear implants, and remote microphone technology. In addition, aided assessments, including speech perception testing, are routinely performed to maximize audibility and ensure consistent, optimized access to sound.

Results

Preschool and Early Elementary Education

Table 2 describes participants' early elementary education. The majority (78%) of participants enrolled in the Moog Center program before age five years. Of these, 50% entered between ages one month and three years and another 28% entered between ages three and five years, with the remaining 22% entering after age five years. Ninety-two participants (85%) entered general education settings with typically hearing peers after leaving the Moog Center. The remaining 15% continued education in other specialized settings, including listening and spoken language programs, special education classrooms, and one in a homeschool setting. The average age upon entering general education settings was significantly different for Teens and Young Adults, with the Teens

entering an average of more than two years earlier than the Young Adults.

Table 2
Description of Early Education

Attribute	Description	Teens n = 44	Young Adults n = 64	All Participants N = 108
Entry to the Moog Center	Mean age and range in years at entry to the Moog Center	2.9 (0.3–5.9)	3.6 (0.4–9.3)	3.4 (0.3–9.3)
Exit from the Moog Center	Mean age and range in years at graduation from the Moog Center	6.8* (3.6–9.6)	8.8* (4.1–12.9)	8.0 (3.8–12.9)
Time spent at the Moog Center	Mean number and range of years spent at the Moog Center	3.8 (1.6–8.6)	5.2 (0.9–10.6)	4.6 (0.9–10.6)
Entry to general education setting	Mean age and range in years at entrance to a general education setting	6.7* (4.2–9.2)	8.9* (5.2–12.9)	8.0 (4.2–12.9)

*p<0.01

High School and Post-Secondary Education

All 64 Young Adults (100%) were high school graduates. Four of these (6%) stopped their formal education after high school and obtained full-time employment. The other 60 (94%) attended a post-secondary education program, as described in Figure 1. Six were currently attending graduate programs, while seven had obtained graduate degrees. Thirty-nine different college and universities were attended (see Appendix B for complete list). One hundred survey respondents (93%) participated in sports and/or clubs during their high school and college years. Forty-three respondents (40%) participated in more than one sport, and 21 (19%) reported being in leadership positions and/or achieving special recognition, such as

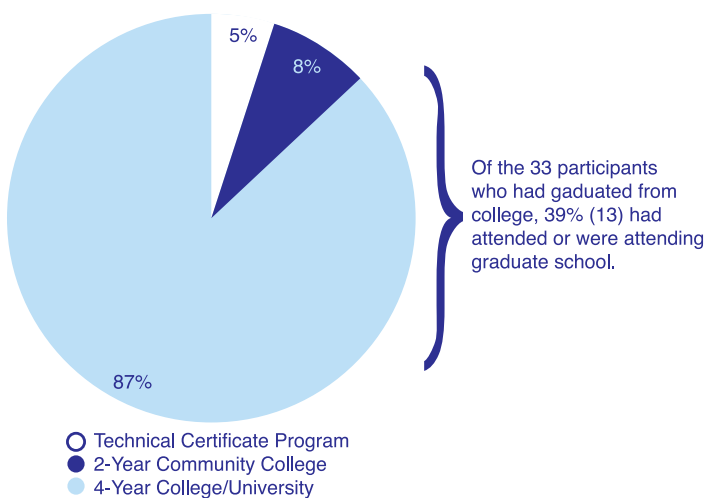


Figure 1. Post-secondary programs attended. Three of the 60 attended a technical certificate program and stopped at that level or were still attending at the time of the survey. Five (8%) attended a 2-year college program and stopped at that level or are still attending. Fifty-two (87%) were currently attending or had graduated from a 4-year college/university. Of the 52, 33 (63%) had graduated, and 19 (37%) were currently attending. Of the 33 college graduates, 13 (39%) went on to attend graduate programs.

being team captains and team managers. Twenty varieties of athletic teams were included among the participants' survey responses. Sixty-four of the respondents participated in organized clubs while attending high school, and 23 varieties of clubs were included among the responses, including social, service, language, STEM, pre-professional, and leadership organizations. In addition to these activities, seventy-two participants (67%) reported receiving awards and special recognition such as prestigious academic awards, athletic recognition, and honors such as valedictorian and commencement speaker. Among Young Adults in college, 12 received academic scholarships, one graduated Cum Laude, one Magna Cum Laude, and one Summa Cum Laude. A full list of awards and clubs can be found in Appendix C.

While attending high school, 101 (94%) participants accessed at least one support service, and of those in post-secondary programs, 100% accessed at least one service. In both high school and post-secondary programs, many students accessed multiple services. Figure 2 details the services accessed by survey respondents during their high school and post-secondary programs.

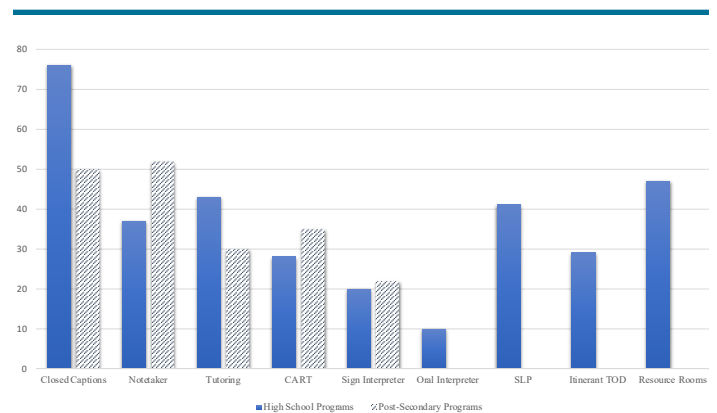


Figure 2. Support services accessed by participants in high school and post-secondary programs. Support services included closed captions, designated notetakers, tutoring services, Communication Access Real-time Translation (CART), sign language interpreters, oral interpreters, speech-language pathologists (SLPs), itinerant teachers of the deaf (TODs), and resource rooms.

Employment

Among the 64 Young Adults (i.e., those beyond high school), 24 were still attending post-secondary programs or graduate schools. Of those, 14 had jobs, including teaching assistant, retail sales positions, child care provider, online boutique entrepreneur, and other jobs typical for students working while in college. Thirty-nine of the Young Adults were no longer in school. Of these, 32 (82%) were employed, 21 in full-time jobs and 11 in part-time jobs. Areas of employment included 18 in business, six self-employed, four in education, one in government, and three in other areas. Salaries were commensurate with salaries of hearing peers.

For those out of school and working full-time, 18 of the 21 respondents (86%) reported being extremely satisfied or very satisfied with their current job. Participants were also

asked to indicate which of the following statements applied to their present employment (numbers in parentheses indicate the percentage of respondents who checked each of the statements):

- My skills are well-utilized in my employment (86%).
- My current employment offers prospects for further advancement (65%).
- Being competent in spoken language is important to my job (60%).
- My employment fits my long-term goals (53%).
- I would like to remain with my current employer for the foreseeable future (53%).
- I plan to remain in my current occupation for the foreseeable future (46%).
- During college, I had an internship, a cooperative education assignment, or field experience (including student teaching) related to my present employment (46%).
- During college, I had a part-time or summer job related to my present employment (37%).
- I supervise two or more people (26%).

Communication

The survey participants were asked to assess their speech intelligibility and comprehension when talking with:

1. Very familiar people, such as immediate family members, teachers, friends at school, and other close friends.
2. Less familiar people, ones you see once or twice a month, such as grandparents, cousins, aunts/uncles, neighbors, friends.
3. Someone who has very little experience talking to people who are DHH, such as a cashier in a store or a waitress at a restaurant.

Possible responses were (a) *completely understood*, (b) *mostly understood*, (c) *barely understood*, or (d) *not understood at all*. Table 3 summarizes the participants' assessment of their success in communicating face-to-face using spoken language.

In response to being understood when talking with very familiar people, 97% of participants responded, completely

or *mostly understood*. In response to being understood when talking with less familiar people, 96% responded *completely understood*. With people who have little interaction with individuals who are DHH, 87% responded *completely or mostly understood*.

Participants were also asked, "How well do you understand when they talk to you?" In relation to very familiar people, 94% responded *completely or mostly understood*. With less familiar people, 88% responded *completely or mostly understood*. When talking to people who have little interaction with individuals who are DHH, 69% responded *completely or mostly understood* and 31% responded they understood about half or less than half of what the speaker said.

In response to the question, "How do you communicate with your friends and family?" participants were provided options and asked to check all that apply. Figure 3 illustrates the options offered and the percentages reported for each.

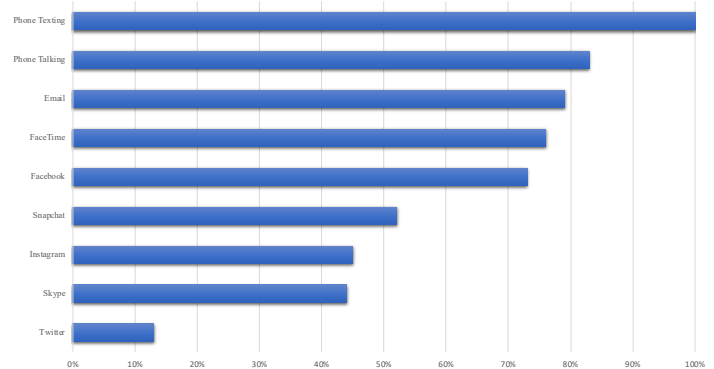


Figure 3. Communication Using Technology. Respondents were asked, "How do you communicate with your friends and family?" The responses are divided into different types of technological communications. Respondents were asked to check all options that apply and percentages are reported for each option used.

Table 3
Spoken Communication Competence

	How well are you understood when you talk?			How well do you understand when others talk to you?		
	With very familiar people	With less familiar people	With people who have little interaction with those who are DHH	With very familiar people	With less familiar people	With people who have little interaction with those who are DHH
(N = 108)						
Completely understood	70	56	54	51	34	22
Mostly understood	35	48	40	51	61	52
About 50% understood	2	1	12	5	10	28
Barely understood	0	2	1	0	1	4
Not at all understood	0	0	0	0	1	1
No survey response	1	1	1	1	1	1

Participant Reflections

Open-ended questions in the survey provided opportunities for participants to express what they considered to be their accomplishments and to reflect on other aspects of their lives. Two of the survey's open-ended items were: (a) *What are you most proud of since you left the Moog Center?* and (b) *Please comment about anything else you would like to share with us.* Major themes that emerged from both Teen and Young Adult responses included accomplishments such as educational attainments (43%), competence in communicating (49%),

community involvement (32%), employment (25%), and academic honors received in high school and college (12%). Other topics included personal competencies that had been important influences in participants' lives, such as self-confidence, motivation, and determination. Participants also reflected on their Moog Center education, support of family and friends, hearing technology, and advice for parents. Verbatim responses from Young Adult participants can be seen in Table 4 and from Teens in Table 5. Additional reflections are presented in Appendix D.

Table 4
Young Adult Reflections

What are you most proud of since you left the Moog Center?	"My college degree, getting the job offer at my dream agency and marrying a wonderful man. All in one year. Besides that, I'm also blessed that I have the ability to advocate for myself, embrace challenges, as well as the motivation to never give up."
	"Becoming one of the first in the country with a cochlear implant to become an Emergency Medical Technician (One of the requirements is that you have to [have] "complete hearing") – and obtaining a 2nd Bachelors' degree in nursing from [X] University."
	"I graduated with a 3.8 GPA from [X] University and have [been] steadily employed and promoted [in my career]. I lead daily and weekly meetings in person and via phone."
	"I am successful in my career as an Account Executive, managing the [X national corporation] e-commerce account. I am very proud of where I am today, and I love teaching my coworkers about what it means to be deaf. Everyone is genuinely curious about my hearing impairment, the cochlear implant, how I was able to learn to speak so well, etc."
Please comment about anything else you would like to share with us.	"If there's anything someone tells you or to your child, "It can't be done." Use that as your motivation and power to drive you or your child to accomplish it. Nothing is impossible and there are no limitations...If there is a will, there is a way."
	"While my speech is not flawless, it is effortless...Moog gave me the tools I needed to succeed in this world."
	"I couldn't have come so far in life without all the help and support you provided early on."
	"The therapy and training I've received at Moog laid down the foundation for the rest of my life. It's made me learn to ask for help, to never stop practicing nor stop learning."

Table 5
Teen Reflections

What are you most proud of since you left the Moog Center?	"Knowing how to talk to people and learning how to keep up in class."
	"My ability to excel in nearly all areas of my life; having close friends and being social, participating in sports and being athletic, all the while being academically one of the top in my class."
	"I have signed to play baseball at [X] University. I have made honor roll every semester. I am at the top of my class."
	"I am most proud of my ability to speak and interact with hearing people. I fit right in at my mainstream school. I can socialize very easily."
Please comment about anything else you would like to share with us.	"My cochlear implant has helped to give me a great amount of success in the world, and I'm very thankful for that!"
	"High school has been difficult for me because of the demands of the classes, but I have really pushed through it and am proud of that. I'm also thankful of the support that my family and friends gave me."
	"If you have a deaf child or hard of hearing child that would like to involve into some kind of sports or any of the activities, then parents should let them do what they love."
	"Being a student at Moog has made a huge impact on my life, not only did I learn to speak clearly, but I gained self-confidence and learned how to advocate for myself. I'll never forget my wonderful teachers, and I love my audiologists."

Discussion

Preschool and Elementary Programs

The Moog Center is a non-profit independent center that provides a full-day listening and spoken language preschool and elementary school program for children who are DHH. On average, tuition for 40–50% of the children is supported by their home school district. For those who do not receive school district support, financial aid is available through the Moog Center's Scholarship Fund. The Scholarship Fund is provided on a sliding scale to all families who qualify, so no family is turned away based on ability to pay.

The daily teacher/learner schedule, a signature element of the Moog Center, was adapted and updated from the program organization and teaching strategies developed during the Experimental Program in Instructional Concentration (EPIC) Project (Moog & Geers, 1985). Modeling and Imitation was the overall teaching strategy used in activities throughout the day, as explained in Appendix E. Sample morning schedules for preschool and elementary school programs, as well as the rationale, are more fully described in Appendix A.

Access to Technology and Entrance to General Education

Advances in hearing technology, early identification, and educational support services provided by IDEA meant that all of the children in the study had access to sound during their preschool years. Access to sound was thought to be an important factor in preparing children to enter general education programs during their elementary school years. The fact that Young Adults (8.9 years) entered general education more than two years later than Teens (6.7 years) may reflect the generational advantage provided to the younger population. Advantages included continuing improvements in hearing aids and cochlear implants, which likely contributed to the development of good spoken communication as reported by participants, documented in Table 3. It is likely that being included in educational settings with hearing children for most of elementary school would have helped prepare all of these individuals to develop strong self-confidence and form friendships with hearing peers.

As depicted in Figure 2, the technology of closed-captions, CART (Communication Access Real-time Translation), and other support services provided through IDEA were accessed to some degree by all participants. Such technological supports probably made accessing the general education curriculum easier and more complete throughout their education and may account, at least in part, for their academic success and high level of educational attainment.

There was virtually no difference between Young Adults and Teens in mean age of receiving their first hearing aids (1.6 years for Young Adults and 1.5 years for Teens). This is surprising since the average age of identification

of hearing loss prior to the 21st century was two to three years (Harrison et al., 2003; Hoffman & Beauchaine, 2007). Young Adults in the current study were born between 1984 and 1998, which was before Congress passed the Newborn and Infant Hearing Screening and Intervention Act of 1999. On the other hand, unsurprisingly, there was a two-year difference between the groups in terms of receiving cochlear implants. The FDA age of approval for cochlear implants decreased from 18 years of age and older in 1984, to two years of age and older in 1989, and finally for children as young as one year of age in 2000. During the time the participants in this study were growing up, improvements in hearing technology provided increased access to sound, resulting in improved ability for perceiving speech and for developing high speech intelligibility. These improvements in hearing technology, as well as the younger age at which Teens received their cochlear implants, could easily have contributed to making it possible for the younger group to join general education settings two years earlier than the older group.

Participation in High School Sports and Other Activities

Several studies of teenagers who have typical hearing have found that being involved in extracurricular activities in high school is beneficial in a variety of ways, such as growing up to be more successful in communication and developing stronger relationships (Mahoney, Cairns, & Farmer, 2003; Guèvremont, Findaly, & Kohen, 2014). Research including students with disabilities involved in extracurricular activities shows that they were more likely to have friends and be engaged in relationships than those who were not (Pence & Dymond, 2016).

An important component of adolescent and young adult development is the degree to which one feels a sense of belonging within a community of peers. In a study using data from the National Longitudinal Study of Adolescent Health, Feldman & Matjasko (2005) reported that 70% of American adolescents were involved in some form of extracurricular activity. In the current study, 93% of respondents reported that they participated in sports and/or clubs in high school and college—a substantially higher rate of participation than that reported for their hearing peers. Not only did almost all Moog Center alumni participate in high school activities, but 18% attained leadership roles as captains and managers of sports teams, leaders in clubs, and elected officers in student government. It is likely that participation in high school activities had a positive impact on their high school experiences, building their self-confidence, developing relationships, learning how to work with others, and feeling comfortable with their hearing peers.

Educational Attainment

According to a recent study of the National Deaf Center (NDC) on Post-Secondary Outcomes of Young Adults 18 to 25 years who identify as DHH, 27% were enrolled in post-secondary education and training programs, compared to 39% of hearing individuals (Garberoglio,

Cawthon, & Sales, 2017). Of the 64 Young Adults in the current study, 100% graduated from high school, 94% of them attended, are attending, or have graduated from post-secondary programs, and 39% of college graduates are attending or have received degrees from graduate programs, as detailed in Figure 1. These high levels of educational attainment of Moog Center alumni exceed the educational attainment for both deaf and hearing individuals as reported by Garberoglio et al. (2017). The 39 diverse college programs attended by these participants are listed in Appendix B.

Employment

The wide areas of employment in which the current study's survey participants were engaged indicated the range of interests, skills, and opportunities that were available to the participants in this study. The majority of those employed full time (89%) reported high satisfaction with their current employment. In addition, over half of the respondents reported that their current employer offers prospects for further advancement, being competent in spoken language is important to their job, and their skills are well-utilized in their employment.

Communication

As detailed in Table 3, participants reported having some difficulty understanding individuals who had little experience talking with people who are DHH. A possible explanation for greater difficulty in understanding speakers, such as clerks in stores, servers in restaurants, and others who rarely interact with people who are DHH, is that these people may talk too fast or not clearly enunciate. Another possible explanation may be that places such as stores, restaurants, and other public places are noisy environments, making hearing and understanding more difficult for individuals who are DHH.

In response to survey questions asking participants to rate their level of success in communicating using spoken language, almost all (96%) rated themselves as being competent when communicating with familiar people, both in being understood and in understanding the speaker. When communicating with familiar people, virtually all (more than 96%) of participants rated themselves as competent in communicating with familiar people with whom they have ongoing contact.

The communication opportunities created by the ever-expanding social media technology, such as email, texting, captioning, Skype, Snapchat, Facebook, and Twitter, have transformed social communication, as documented in Figure 3. These technologies have enabled participants to be in touch with their families and friends, both hearing and deaf, across the country and the world. In addition, the technology of captioning has given people who are DHH, including those that focus on listening and spoken language, better access to TV and movies, which has expanded opportunities to enjoy these activities with both their hearing and deaf friends as well as their families. Many of the participants commented that they use

technology to develop social relationships and to feel and stay connected. This kind of access had become increasingly available as these individuals were growing up in contrast to earlier times when people who were DHH were dependent on Relay, TTYs, and snail mail for communication that was not face-to-face.

Participants' Reflections

In the responses to open-ended questions at the end of the survey, as detailed in Tables 4 and 5 and Appendix D, participants expressed important thoughts about themselves and various other aspects of their life experiences not addressed in the previous multiple-choice survey items. The question *What are you most proud of?* provided an opportunity to reflect on their accomplishments and provided insight about what participants strove for and were proud to have accomplished. Accomplishments cited included levels of educational attainment, academic awards, participation, and leadership in clubs and sports in high school and college, as well as success in employment. Especially enlightening were the responses to the very open prompt, *Comment on anything else you would like to share*. In their comments to this request, it was clear that many had set high expectations for themselves, had learned that hard work pays off, had become self-confident, and had acquired other personal competencies such as high motivation, determination, persistence, and ability to communicate and advocate for themselves. Hintermair and colleagues, in a study of adults who were DHH and who considered themselves successful in their jobs, found that the participants in their study reported similar social and personal competencies as being important contributors to their success in their jobs (Hintermair, Cremer, Gutjahr, Losch, & Strauß, 2018).

Conclusion

The results of this study demonstrate that teens and young adults who are DHH in the 21st century can be very successful with respect to education, employment, and related outcomes—much more so than has historically been the case for individuals who were DHH. Although it is reasonable to conclude that these Young Adults and Teens benefitted from public policy changes, technology advancements, and early education in an intense, focused intervention program, the descriptive nature of the data preclude being able to make such causal conclusions.

Regardless of what factors contributed to the outcomes documented in this study, it is clear that the overall level of achievement in educational attainment, employment, and general satisfaction with their lives is greater for the participants in this study than has been typically reported in previous studies of teenagers and young adults who are DHH (e.g., Dammeyer & Marschark, 2016; Garberoglio, Cawthon, & Bond, 2016; Garberoglio, Cawthon, & Sales, 2017). These achievements, along with participants' reflections, provide evidence of the participants' high expectations of themselves and their ability to meet those expectations.

It should be noted that participants in the current study were an advantaged group within the overall population of individuals who are DHH, and the results reported here may not be generalizable to all individuals of similar ages who were identified with hearing loss during early childhood. Because parents of participants found and chose the Moog Center for their children, they may have been more heavily invested in their children's education than other parents. The Moog Center provided a strong parent component for guiding, educating, and empowering parents in ways to support their children in learning to talk. Parents were supported and guided through transition to general education. Parents of the teens and young adults in this study were also more highly educated than is typical, with 78% of mothers being college educated. In addition, the mean IQ of the participants were all within the normal range, and 52% were above average.

The fact that all participants in this study attended a single program means that results are easier to interpret because all of the children had reasonably similar educational experiences during the early childhood period. At the same time, the absence of children from other programs or who were not in any program (i.e., a control group) means that we do not know whether these very positive outcomes can be attributed to this particular program or to other factors that were not measured such as family background or parent motivation.

For parents of children who have recently been identified as DHH, these results make it clear that children who are DHH can have very high levels of achievement with respect to educational, employment, communication, and related outcomes. In fact, their achievement can be on the same level as their peers with typical hearing. EHDI providers and educators working with young children who are DHH can use the results from this study, to inform parents of what is possible, as well as to calibrate their own expectations about what children who are DHH are able to achieve.

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Appendix A Sample Schedules and Rationale

Sample Preschool Morning Schedule with Individual Children Represented by Alphabet Letters

Time	Teacher 1	Teacher 2	Teacher 3	Teacher 4	Discovery Room, Teacher 5
8:25 – 8:30 Device Check	A, B, C, D	E, F, G, H	I, J, K, L	M, N, O, P	
8:30 – 9:00 Syntax/Vocab	A, B	E, F	I, J	M, N	Circle, Choice C, D, G, H, K, L, O, P
9:00 – 9:30 Syntax/Vocab	C, D	G, H	K, L	O, P	Circle, Choice A, B, E, F, I, J, M, N
9:30 – 10:00 Speech/Aud. Skill	A, B	E, F	I, J	M, N	Music/Movement C, D, G, H, K, L, O, P
10:00-10:10 Snack	A, B, C, D	E, F, G, H	I, J, K, L	M, N, O, P	
10:10-10:30 Recess	Staff time	Staff time	Staff time	Staff time	Recess ALL students
10:30 – 11:00 Speech/Aud. Skill	C, D	G, H	K, L	O, P	Music/Movement A, B, E, F, I, J, M, N
11:00-11:30 Conv. Lang	A, E	B, I	F, J	M, N	Thematic Art C, D, G, H, K, L, O, P
11:30-12:00 Conv. Lang	C, G	D, K	H, L	O, P	Thematic Art A, B, E, F, I, J, M, N

Note. Sample schedules are provided here to help the reader understand the reasoning behind the development of these schedules. The daily schedule was organized to provide opportunities for the continuum of teaching activities from structured lessons to conversational activities. At one end of the continuum is teaching within a lesson, using repetitive, structured activities to practice specific language targets. Further along the continuum is teaching within contrived conversational activities which are designed by the teacher to obligate use of a variety of structures for practice in the context of naturally communicative interactions. At the far end of the continuum is teaching during spontaneous exchanges as the teacher capitalizes on a child's spontaneous language during all communicative interactions throughout the day to help the child improve his or her language.

The framework of the schedule provided opportunities for this continuum of teaching activities from lessons to spontaneous conversation. Children were organized in small groups of two or three for focused spoken language instruction (i.e., syntax, vocabulary, language, speech, and auditory [aud.] skill development). Small groups ensured that the teacher could know precisely each child's skills and could individualize instruction for maximum challenge and maximum success. The larger groups provided opportunities for transferring learned skills to a variety of natural situations and for a variety of purposes. The afternoon schedule for preschool children included instruction in early math, reading readiness, hands-on language experiences, and cognitive activities.

Sample Elementary Morning Schedule with Individual Children Represented By Alphabet Letters

<i>Time</i>	<i>Teacher 1</i>	<i>Teacher 2</i>	<i>Teacher 3</i>	<i>Teacher 4</i>	<i>Learning Center, Teacher 5</i>
8:25 – 8:30 Device Check	Q, R, S, T	U, V, W, X	Y, Z, AA, BB	CC, DD, EE, FF	
8:30 – 9:00 Reading	Q, R	U, V	Y, Z	CC, DD	Special Activities S, T, W, X, Y, Z, CC, DD
9:00 – 9:30 Reading	S, T	W, X	AA, BB	EE, FF	Special Activities Q, R, U, V, AA, BB, EE, FF
9:30 – 10:00 Speech/Aud. Skill	Q, R	U, V	Y, Z	CC, DD	Critical Thinking S, T, W, X, Y, Z, CC, DD
10:00 – 10:30 Speech/Aud. Skill	S, T	W, X	AA, BB	EE, FF	Critical Thinking Q, R, U, V, AA, BB, EE, FF
10:30-11:00 Phys. Ed/Recess	Staff time	Staff time	Staff time	Staff time	Phys. Ed/Recess ALL students
11:00-11:30 Language	Q, R	U, V	Y, Z	CC, DD	Computer S, T, W, X, Y, Z, CC, DD
11:30-12:00 Language	S, T	W, X	AA, BB	EE, FF	Computer Q, R, U, V, AA, BB, EE, FF

Note. In the elementary program, children were organized in small groups of two or three for focused spoken instruction in reading, speech and auditory (aud.) skill development, and language. Large groups included special activities, critical thinking, physical education, and computer. Special activities included Art, Social Skills, Theater Workshop, etc. provided on different days throughout the week. The afternoon schedule for this group of elementary school children was organized in groups of four for social studies, science, math, and written language.

For both preschool and elementary groups, all spoken language instruction was explicitly focused on specific objectives. The Moog Center schedules were designed to provide a balance for children, moving from periods of intense, explicit instruction in small groups to larger group activities in which children had opportunities for natural communicative interactions. The physical movement, alternating from space to space, from intense to less intense, and from small group to larger group activities, provided a good balance for children and enhanced learning.

Appendix B Colleges and Universities Attended

Abilene Christian University	Purdue University
Arizona Christian University	Rochester Institute of Technology (15)
Arizona State University (2)	Southeast Missouri State University
Art Institute of Colorado	St. Louis University
Art Institute of St. Louis	Texas Woman's University
Baylor University	Trevecca Nazarene University
Bradley University	Trinity International University
California State University Northridge (3)	University of Delaware
Christian Life College	University of Denver
Fontbonne University	University of Illinois at Chicago
Gallaudet University	University of Miami
Grand Canyon State University	University of Minnesota Rochester
Lindenwood University	University of Missouri (2)
Longwood University	University of Toledo
Missouri State University (2)	University of Tulsa
Multnomah University	Washington University in St. Louis
National Institute for the Deaf	Yale University
Pennsylvania State University	

Appendix C

Participants' noted awards, achievements, and club participation

Academic Awards: Honor Roll, High honor roll, JCAA Academic Scholarship, 4.0 GPA throughout entire schooling career, National Junior Honor Society, High School Scholastic Achievement Award, Academic Excellence Award, Cum Laude Society, Magna Cum Laude, Summa Cum Laude, Commended National Merit Scholar, A+ Program, Scholar Athlete award, high school commencement speaker, and valedictorian.

Athletic Achievements: Varsity letters in various sports, including baseball, basketball, track, dance, and volleyball; all conference champions, leadership positions and captain of teams, Eagle Scouts, Black Belt in Mixed Martial Arts, CPR certified, and First Aid certified.

Clubs: Student campus activities committee, student campus government, campus ambassadors, literary magazine, reading club, mission trip organizations, historic preservation club, random acts of kindness club, volunteer organizations, social fraternities and sororities, professional and business fraternities, service fraternities, Christian campus ministry organizations, peer educator organizations, professional and major organizations (School of Health Professions, American Advertising Federation, National Student Speech Language Hearing Association, Supply Chain Management Association, Future Farmers of America Lab Science Technology), deaf organizations (National Association of the Deaf, ASL Club, Sign Language Organization, Deaf club), leadership in organizations including events coordinator, secretary, treasurer, executive board member, and vice president roles.

Appendix D

Additional Verbatim Participant Reflections

Additional Young Adult responses to "What are you most proud of since you left the Moog Center?"

- "That I am able to be a part of the hearing world and be successful because I don't think I would have the opportunities I do if my parents hadn't gotten me a cochlear implant."
- "The fact that I know how to talk and most people do not even realize I'm deaf until I tell. I also love how I can be an inspiration to others (parents and kids) who have had the same concerns that my parents and I have had over the years."
- "Graduating from the #1 Journalism school in the country, with honors, and being accepted into that University's Masters' program."
- "Creating an anti-bullying lesson plan that is now taught throughout MN."
- "I am most proud of my independence since leaving the Moog Center. I have gone away to college and even studied abroad for a semester."
- "Making an entire career out of my passion for languages and getting people to pay me to do what I love."
- "Marriage of my wife and I, Bachelors' Degree, Current engineering position...continuing to progress in communicating with others."
- "The most proud moment was when I graduated with my Masters' degree in Deaf Education."
- "Participating fully in the hearing world, being able to speak clearly."
- "Getting an education and a job."
- "That I have managed to retain my speech and continued to use it in my daily life and at work."
- "My ability to excel in the classroom and be an actively involved member outside of the classroom...I work hard to get good grades while at the same time I am very social and involved in my community."
- "I would say the fact that I've been able to make the transition pretty seamlessly from the Moog environment to a normal hearing world and have been able to thrive."
- "My gymnastics career as well being able to communicate well with others!"
- "I can hear well, do well in school, have good speech and grammar. I have been fortunate to be able to succeed at whatever I wanted to try."
- "Finishing my degree at [X University] and found the perfect job at [X University]."

Additional Young Adult responses to "Please comment about anything else you would like to share with us."

- "My instructor told me for my EMT class that he didn't think I was going to be able to be certified by the state because of my hearing deficits.... Not only did I pass my class, I was one of the top of my class and more importantly, my program director who initially doubted me ended up defending and advocated for me to the [state] department of transportation saying that I was fully competent to be certified."
- "I am extremely thankful to the Moog Center for all the time and effort they put in me to help build my confidence and prepare me for the world."

- “Everything that I learned at Moog Center has been contributed to my success in the hearing world. Because of my confidence and determination, I am able to be successful in most things that I attempt.”
- “I’m thankful for my time at the Moog Center. I don’t know where I would be without your tireless teachers.”
- “I am very proud that I can speak very well. I/O this to my cochlear implant, my audiologist, my teachers, my parents, and my own desire to learn to speak. I can’t imagine how my life would be without my implant & if I couldn’t speak. It was very hard for me to start talking and took forever for me to learn talk. My parents & Moog Center never gave up on me. I appreciate my parents & Moog Center. I strongly urge all new parents who have a child who is hard of hearing, please, don’t give up trying make your child learn spoken words. Your child will thank you the rest of his or her life. I know that I do!”
- “I’m proud to have attended Moog School. Without them, I would never have as much success as I have lately. Good group of people and lifetime relationships.”
- “Life is as good as you make it, you can be as miserable in the situation you are in, make the best of what you can, life will treat you well after you enjoy it.”

Additional Teen responses to “What are you most proud of since you left the Moog Center?”

- “Being inducted into Cum Laude Society in my junior year.”
- “Joining my Highschool Robotics team and building successful competitive robots.”
- “I can hear well, do well in school, have good speech and grammar. I have been very fortunate to be able to succeed at whatever I wanted to try.”
- “My ability to play an instrument at a very high level, which I plan to major in college.”
- “Taking 5 AP classes, a math class at the local college and leading 75-member team practices senior year.”
- “Proud of myself for developing more confidence in my Algebra skills. I struggle with Math. Proud of my family for not being too afraid to let me follow my dreams.”
- “I am most proud of reaching the rank of Eagle Scout in Boy Scouts of America. It required me to plan, develop, and carry out a massive community project that required hundreds of hours of work on my part.”
- “Success in school, AB honor roll, being able to play sports with hearing friends/teammates.
- I’m proud of achieving high grades, such as having a current 4.2 GPA. I’m fully confident of myself.”
- “I’m most proud of myself. It took a lot of courage to meet new friends when I left the [Moog] community.”

Additional Teen Responses to “Please comment about anything else you would like to share with us.”

- “Thank you for everything that Moog has done for me from teaching me how to talk, to my implants, etc.”
- “I have cheered at the loudest of basketball/football games with the rest of my cheerleading squad, I have set school records for pole vault, I have taken up playing the piano, and I even joined my school’s diving team this last year.”
- “Ever since I left the Moog all of us that went there are close like peas in a pod.”
- “Thank you for giving me the experience and help that I needed so I could go on to regular hearing schools.”
- “I would like to say that Moog is one of the greatest schools I have ever been to. I still tell my parents how I would love to work there.”
- “I wouldn’t be where I am today without Moog...it enabled me to become the successful and independent man I am today.”

Appendix E

Modeling and Imitation

In interactions with the children throughout the day, teachers strive to help children increase their spoken language competence. Teachers listen not only to what a child says but also to how the child says it and then help the child say it better. This may be by including more words, adding new vocabulary, correcting grammar, increasing the complexity of the syntax, or improving the speech intelligibility. Once the child has succeeded in getting his or her idea across, it is important to help the student express that idea. However, at the Moog Center, teachers believe that it is important to help the child use higher levels of vocabulary and/or longer, more complete phrases and sentences. Teachers use the Modeling and Imitation strategy as a technique to facilitate and accelerate the child’s learning. The words modeled by the teacher are based on both what the child means and what the child actually says. Here’s how it works: (a) the child talks, (b) the teacher listens, (c) the teacher indicates she understands, (d) the teacher selects a target for improvement, (e) the teacher restates what the child has said and highlights the added target word(s) in her model, (f) the child imitates the teacher’s model (Moog & Stein, 2008; Moog, Stein, Biedenstein, & Gustus, 2003).

Imitating the teacher’s model and including the targeted aspect provides the child practice with producing improved language. Imitation is an essential step in the process as it provides practice using the syntactic structure, vocabulary word, or speech sound that was targeted in the model. In addition, imitation helps the child learn to recognize and

understand the new words or sounds the next time he or she hears them and helps the child's development of auditory memory.

A model given by a teacher may serve many purposes, such as correction, expansion, and/or completion. The following are examples of Modeling and Imitation:

Jack comes into class after recess.

Jack: I play tag Suzie!

Teacher: I played tag with Suzie.

Jack: I play tag with Suzie.

Teacher: I played tag with Suzie.

Jack: I played tag with Suzie.

The teacher and child are engaging in a language activity involving cutting and pasting. The teacher is holding a pair of scissors, which the child needs to complete the next step in the activity.

Child: Need scissors cut paper.

Teacher: I need scissors to cut...

Child: I need scissors cut paper.

Teacher: I need scissors to cut the paper.

Child: I need scissors to cut the paper.

Effects of Frequency of Early Intervention on Spoken Language and Literacy Levels of Children Who are Deaf or Hard of Hearing in Preschool and Elementary School

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Abstract: Language delays associated with hearing loss during infancy may have a negative impact on academic development throughout childhood. Early intervention provided by the Moog Center for Deaf Education prior to 36 months of age was quantified, and associations with later outcomes were examined for 50 students who are DHH representing Moog Center alumni. The objective was to determine whether the amount of early intervention (referred to hereafter as dose of early intervention received at the Moog Center during the time children were 0–36 months of age) contributed uniquely to outcomes in preschool (4–6 years) and in elementary school (8–14 years). Analysis of language and reading outcomes concluded that greater doses of early intervention were beneficial, even when other contributing factors such as degree of hearing loss, nonverbal intelligence, and age at first intervention were taken into account. Those children with poor aided speech perception scores in preschool exhibited the most benefit from early intensive intervention. Average language scores were within the expected range in comparison with hearing peers in preschool and remained within expectation when assessed an average of four years later in elementary school. The intensity of early intervention provided at the Moog Center contributed significantly to long-term development of language and literacy over and above the benefits associated with the age at which intervention was delivered.

Key Words: early intervention, language and literacy in deaf and hard of hearing children, listening and spoken language intervention

Acronyms: BKB-SIN = Bamford-Kowal-Bench Speech in Noise; CASL = Comprehensive Assessment of Spoken Language; CELF-P = Clinical Evaluation of Language Function-Preschool; CI = cochlear implant; DHH = deaf or hard of hearing; EI = early intervention; HA = hearing aid; HL = hearing loss; LNT = Lexical Neighborhood Test; LSL = listening and spoken language; mLNT = Multi-syllabic Lexical Neighborhood Test; NVIQ = Nonverbal Intelligence; PPVT = Peabody Picture Vocabulary Test; PTA = pure tone average; SLP = speech-language pathologist; SNR = signal to noise ratio; TORC = Test of Reading Comprehension; UNHS = Universal Newborn Hearing Screening; VIQ = Verbal Reasoning; WISC-V = Wechsler Intelligence Scale for Children; WNL = within normal limits; WRMT = Woodcock Reading Mastery Test

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The adoption of universal newborn hearing screening (UNHS) in the majority of states in the United States has enabled earlier identification of children with congenital hearing loss. The goal of screening by one month, confirmation by three months, and intervention by six months is intended to maximize linguistic and communicative competence, including providing infants with the opportunity for amplification as early as possible

(JCIH, 2000). As a result, programs for children who are deaf or hard of hearing (DHH) have focused on early identification and intervention during the birth to three age range (Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998). Evidence suggests that children who are DHH and are enrolled at younger ages in early intervention (EI) demonstrate better language skills by the end of preschool than do later-enrolled children, regardless of degree of

hearing loss (Moeller, 2000). This EI period is particularly critical to Listening and Spoken Language (LSL) service providers, where the focus is on comprehension and intelligible production of speech (Estabrooks, 2006). The achievement of spoken language skills commensurate with those of hearing age-mates during the preschool years is a primary objective of such EI programs (Moog, 2002). Although research suggests intervention should begin as early as possible, little evidence is available concerning the optimal amount or intensity of EI for reaching this objective for children who are DHH.

Research that is specifically designed to assess the effects of increasing the intensity (dose) of intervention in children with communication disorders has reached mixed conclusions. A greater number of hours of intervention has resulted in improved phoneme production in three to six-year-olds with speech disorders (Cummings, Hallgrimson, & Robinson, 2019) and better spoken vocabulary in children with Down Syndrome (Yoder, Woynaroski, Fey, & Warren, 2014). A meta-analysis of treatment studies of children with developmental speech and language delays found greater expressive language gains for interventions that were longer in duration (Law, Garrett, & Nye, 2004). However, a report by Fey, Yoder, Warren, & Bredin-Oja (2013) of children with delayed vocabulary acquisition and no diagnosis of autism at 18–27 months showed that greater intervention was not necessarily associated with better outcomes. Similar results were reported in a study of five to eight-year-olds diagnosed with language impairment (Schmitt, Justice, & Logan, 2017).

A few studies have addressed the effects of intervention dose on spoken language acquisition in children who are DHH. One nationwide study tested 112 five- and six-year-olds who had used a CI for at least one year and received early LSL intervention (Moog & Geers, 2010). The analysis examined the effects of age and type of intervention on preschool outcomes across a broad battery of standardized spoken language measures including vocabulary, verbal reasoning, and global language skills. Educational interventions included individual parent-child coaching in LSL strategies and preschool classes. These programs differed in their intensity, with classes occurring several times each week for at least two hours, while individual parent-child sessions generally consisted of weekly one-hour sessions. Depending on the specific outcome assessed, between 44% and 65% of the sample scored within normal limits (WNL)—defined as within one standard deviation of hearing age-mates—by the end of preschool. The probability of achieving scores WNL was increased for children who received a CI by 24 months of age. In addition, placement in an LSL-specialized class by two years of age further increased the probability of age-appropriate language scores. More importantly, 71% of those who attended an LSL class from two through four years of age scored WNL compared to only 41% of those who did not start preschool until age three (averaged across tests).

A more recent study examined the effects of specialized preschool education on language and literacy skills in DHH children between three and five years of age by comparing progress during the school year with progress over summer months without formal intervention (Scott, Goldberg, Connor, & Lederberg, 2019). Vocabulary, phonological awareness, and letter-word identification skills all improved during the school year, but not during the summer. This result highlights the importance of preschool for DHH children and argues in favor of increasing the intensity of preschool intervention. Chu and colleagues (2016), on the other hand, reported that greater frequency and dose of individual EI sessions were not related to better receptive communication outcomes in children given a cochlear implant by age 7, even though children with higher doses of EI services tended to be in families who had greater relative socio-economic advantage. Children with earlier access to cochlear implants demonstrated better expressive language with less total EI dose than was documented for children who received a CI later.

The advent of cochlear implantation has brought the goal of normal spoken language within reach for many more children by increasing their early auditory access to speech. Even after appropriate sensory devices are provided, language delays associated with hearing loss during this early formative period may continue to have a negative impact on academic development through elementary grades and high school (Geers, Nicholas, Tobey, & Davidson, 2016; Geers, Strube, Tobey, Pisoni, & Moog, 2011; Moog & Geers, 2010). It is, therefore, important to document the type and dose of EI needed to optimize the chances of achieving age-appropriate spoken language.

The current study examined the outcomes of a specific LSL EI program for children who are DHH, the Moog Center for Deaf Education. The intensity of intervention provided by the Moog Center prior to 36 months of age was quantified, and associations between amount of Moog Center EI and later outcomes in children who are DHH were examined. Outcomes were measured for 50 children at two points in time: the first testing occurred at the end of Moog Center preschool and the second testing occurred, on average, four years later during general education elementary school (here, defined as grades two through eight). The goals of this investigation were as follows:

- To document speech perception, spoken language, cognitive, and reading outcomes in a sample recruited from all eligible alumni of the Moog Center for Deaf Education.
- To quantify the dose of intervention (as measured in number of hours) each child accumulated in the Moog Center EI program between birth and 36 months of age.
- To determine whether dose of EI received at the Moog Center contributed uniquely to language and literacy outcomes in preschool and, later, in elementary school.

Method

Families of all children with a better ear unaided pure tone average (PTA) threshold of 40dB hearing loss (HL) or greater who had attended the Moog Center by 6.5 years of age and were currently between 8.0 and 14.0 years old ($N = 60$) were contacted for follow-up testing. Each child, accompanied by a parent, was invited to attend a one-day testing session, held at the Moog Center, with all travel expenses paid for families living outside of the local (St. Louis) area. The test battery was completed successfully by all but one child, for whom testing was discontinued because the child became ill. Preschool speech perception and language scores were obtained from the Moog Center's files for each of these children from when they were between three and six years old. All testing was conducted at the Moog Center by qualified audiologists, speech-language pathologists (SLPs), psychologists, and

LSL teachers. Parents and children individually consented to participate in data collection, analysis, and reporting. Human Subjects Review for this study was conducted and approved by IntegReview IRB, Austin, TX.

Participants

Fifty of the 60 alumni who qualified (84% of the total qualifying population), returned for a testing session. Table 1 compares mean characteristics of the tested sample with those of the ten qualifying children who did not attend a follow-up session. ANOVAs comparing mean characteristics of the two samples revealed only one statistically significant difference; children who did not return for follow-up assessment had higher average nonverbal intelligence scores than those who did return. Thus, it appears that the tested sample was representative of children attending this program in most characteristics and was not biased toward better-performing subjects.

Table 1
Student Demographics

	<i>Mean</i>	<i>Standard Deviation</i>	<i>Range</i>
Age at Identification (mos)— Test group ($N = 50$)	11.9	13.3	0.5–51
Non-participant group ($n = 10$)	15.2	16.1	0.5–42
Age at First Hearing Aid (mos)— Test group	16.1	14.3	0.75–57
Non-participant group	16.7	15.5	1–46
Better Ear Unaided PTA ¹ (dB HL)— Test group	89.9	28.5	43.3–120.0
Non-participant group	86.8	25.0	46.7–113.3
Preschool mLNTe ² (% correct)— Test group	77.7	23.3	8.3–100
Non-participant group	76.7	23.2	16.7–100
Preschool Celf-P3 ³ (St. Score)— Test group	91.2	19.4	50–125
Non-participant group	84.7	20.0	57–112
*NVIQ ⁴ — Test group	101.3	14.1	74–132
Non-participant group	111.6	6.9	100–127
Maternal Education (yrs)— Test group	15.6	2.3	10–20
Non-participant group	14.8	1.9	11–18
Gender (percent)— Test group	44% Female		56% Male
Non-participant group	40% Female		60% Male

¹PTA – Pure Tone Average (500, 1000, 2000 Hz) in dB, hearing level, better ear unaided

²mLNTe – Multisyllabic Lexical Neighborhood speech perception Test - easy list percent correct

³CELF-P3 – Clinical Evaluation of Language Function – Preschool Level, Standard Score

⁴NVIQ – Nonverbal Intelligence Quotient – Wechsler Intelligence Scale for Children, 5th edition (WISC-V)

*The non-participant group that did not come back for testing had significantly higher Non-Verbal IQ scores.

All but four of the children had documented congenital or pre-lingual (i.e., before 36 months) onset of HL, as well as early identification and early intervention. Although age at onset of HL could not be confirmed for these four children, identification of HL occurred at 24, 44, 49, and 53 months of age, and hearing aids were fit between 50 and 54 months of age.

Table 2 summarizes the intervention and assessment history for the 50 participants in this study. Children ranged from 1 month to 6.5 years old when they entered the Moog Center and were between 4 and 10 years old when they graduated. Children graduated at an average age of 6.4 years, having spent an average of 4.2 years at the Moog Center. Upon graduation, 48 of the children entered general education classes with hearing children and two students were homeschooled. Most of the children received additional support in the general education setting, including services from itinerant teachers of the deaf, SLPs, special educators, and remote microphone technology.

delivery are provided, depending on the child's age. For children younger than 18 months, the program is primarily parent-centered, and for children 18 to 36 months, a child-focused component is also provided. All EI providers are either LSL teachers of the deaf or SLPs. The Moog Center's intervention setting also includes audiologists, so if any problems occur on-site with a child's hearing aid or cochlear implant, a qualified professional will troubleshoot immediately. If the problem cannot be fixed, the child is fitted with a loaner device. Back-up hearing aids and cochlear implants from the three companies that market CIs in the United States are on-hand for loan when needed. The audiologists recognize the importance of access to sound and are available on weekends and holidays to ensure uninterrupted access to sound. In addition, parents are trained on troubleshooting their child's sensory aid.

The program for children under 18 months consists of one-hour home visits by an EI provider at least twice a month and a Center visit once a month. Home visits

Table 2
Intervention and Assessment History

	<i>Mean</i>	<i>Standard Deviation</i>	<i>Range</i>
Age at Identification (mos)	12.1	13.4	0.5–51
Age at First Intervention (mos)	18.5	15.6	1–58
Age Enrolled Moog Center (mos)	26.3	20.5	1–78
Age Graduated Moog Center (yrs)	6.4	1.6	3.7–10.0
Duration of Moog Center Intervention (yrs)	4.2	1.8	1.2–8.1
Age at Preschool Assessment (yrs)	4.5	0.7	3.1–6.5
Age at Elementary Assessment (yrs)	10.5	1.9	8.1–14.0
Duration from Graduation to Elementary Assessment (yrs)	4.0	2.0	0.9–7.8

At the time of preschool testing, 16 of the children used hearing aids (HA), and 34 were cochlear implant (CI) users; 14 children received at least one CI before 18 months of age, and 21 received a CI after 18 months of age. All but one of the children received his or her first CI before age five. All but two of the families reported their child used a sensory aid at least 8 hours daily during the preschool years.

At time of follow-up testing, 35 children used at least one CI (6 bimodal, 28 bilateral, and 1 unilateral). Fifteen children continued using two hearing aids. As expected, PTA threshold average differed significantly among device users (*mean* = 115dB HL for CI-only users, 75dB HL for bimodal users, and 50 dB HL for HA-only users). Almost all (*n* = 49) parents reported sensory aid use during all waking hours, and one reported use 5 days a week during school.

Intervention

The Moog Center EI program serves children from birth to three years of age and their families. Two types of service

include providing parents information about hearing loss and its impact on a child's acquisition of spoken language, importance of amplification, discussion of parents' concerns, activities and strategies to help parents facilitate their child's learning to talk, and other information and topics of interest. All visits also include at least a 20-minute period of an EI provider coaching the parent engaged in an activity with his or her child. The monthly Center visit includes an individual parent-child session and an appointment with one of the Center's pediatric audiologists. Only the parent-child portion of the Center visit was included in the calculation of hours.

Children 18 months and older attend a center-based toddler class, which is offered every day from 8:30 to noon. Children attend two, three, four, or five mornings a week depending on their age, maturity, and family factors such as distance from the Moog Center, jobs, other commitments, and so forth. For children, participation in the toddler class includes three components: (a) one-hour of individual therapy intervention for the child, (b) two

and half hours of group experiences for the child, and (c) weekly 30-minute individual sessions for the parent with his or her child. Individual therapy intervention for the child focuses on the development of spoken language skills including explicit teaching of vocabulary, language, speech, and listening skills. For the group sessions, children are organized in classes of six children, where they engage in circle time, gross motor activities, centers, a variety of fine motor and cognitive activities, and snack time. The weekly 30-minute individual parent-child session includes the EI provider coaching the parent engaging with his or her child and discussion about the child's language development (Brooks, 2016).

To assess the intensity of the program for each child, our goal was to specify dose (number of hours) of participation in the Moog Center EI program. To quantify the dose of intervention, we examined billing and attendance records for each of the 50 Moog Center alumni who returned for testing. The total number of hours attended at the Moog Center prior to 36 months was determined, with individual intervention sessions encompassing home visits, Center

visits, individual child therapy, and individual parent-child sessions. Calculations for group intervention included hours spent in the toddler class between 18 and 36 months of age.

The dose distribution is summarized in Figure 1 for each of the 50 children. The histogram depicts the total number of hours each child had attended the Moog Center between 0–36 months of age by frequency-ordered columns. The first 15 subjects depicted without a frequency column in Figure 1 did not begin attending the Moog Center until after their third birthday and thus showed zero hours of intervention. Ten of these 15 children were enrolled in EI elsewhere before attending the Moog Center. For children who received intervention elsewhere before enrolling in the Moog Center, age at first HA represents age at first intervention. The remaining 35 children in the sample attended both individual and group sessions at the Moog Center. Hours of individual intervention for all 50 children ranged from zero to 279 and group intervention for all children ranged from zero to 482.

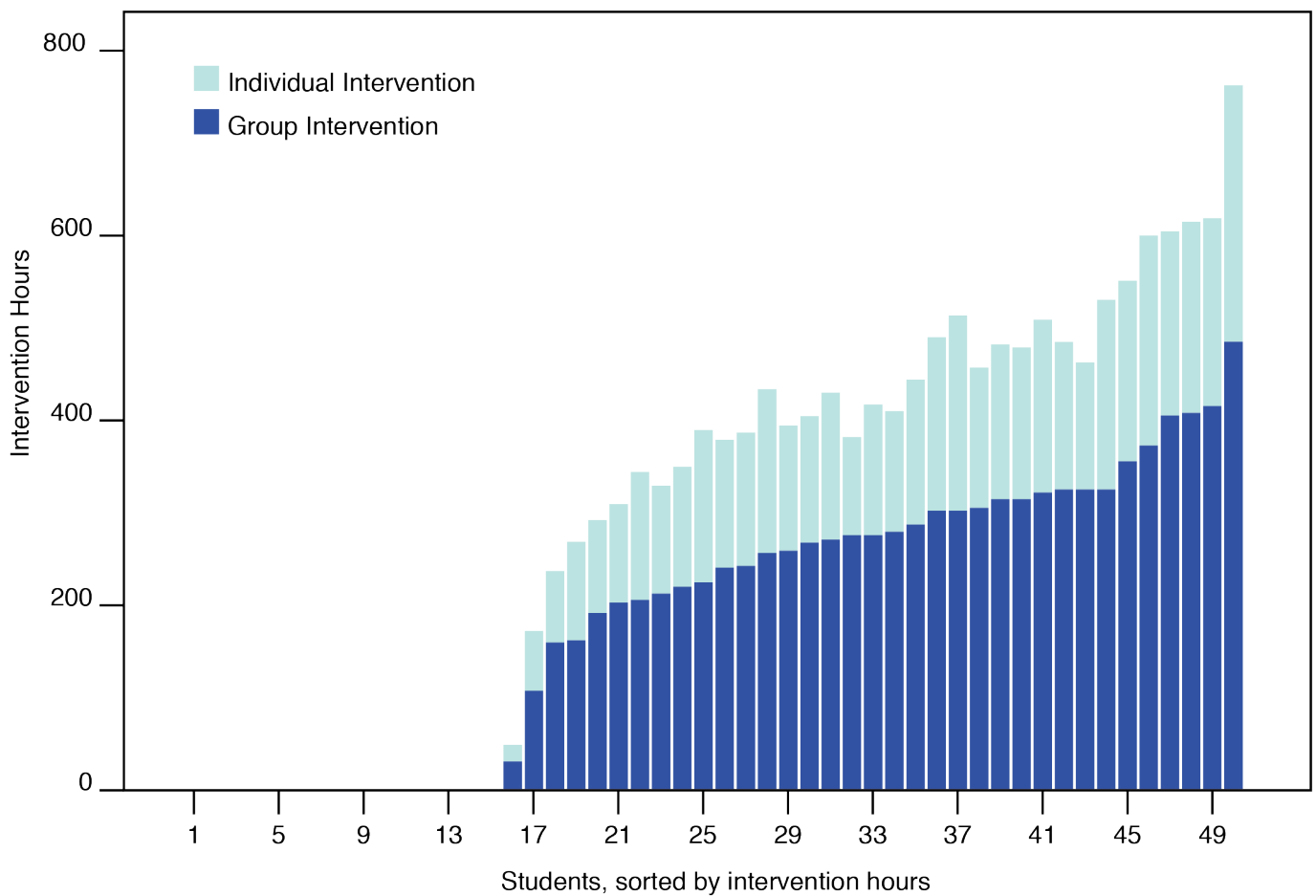


Figure 1. Number of hours of group and individual intervention at the Moog Center between 0 and 36 months of age. Hours and individual intervention are plotted in stacked bars for each of 35 Moog Center alumni. Fifteen subjects did not have any Moog Center intervention in that time frame.

Preschool Assessment

Speech perception. *Multi-syllabic Lexical Neighborhood Test* (mLNT; Kirk, Pisoni, & Osberger, 1995) was designed to measure auditory word recognition in very young children who are DHH. This open-set test consists of 24 multi-syllable words representative of the vocabulary of young children (e.g., *purple, glasses, again, animal*). Two sub-lists within each set contain 12 “easy” words that frequently occur in the English language and are less likely to be confused with other words and 12 “hard” words that occur less frequently and can be easily confused with similar sounding words. Scores were consistently available for all children on the easy list, so only scores on that 12-word list are represented in this report. The target words were presented at 60 dB SPL in quiet, and the children responded by repeating the word they heard. The word was scored as correct if the response was recognizable as the target word.

Spoken language. *Clinical Evaluation of Language Function-Preschool* (CELF-P; Wiig, Secord, & Semel, 2004) is a comprehensive language assessment normed on hearing children between 3.0 to 6.9 years of age. The particular subtests administered varied slightly based on age at test (Basic Concepts, Sentence Structure, Concepts & Following Directions, Word Structure, Expressive Vocabulary, and Recalling Sentences). Subtest scores were combined into a Total Language standard score using age-appropriate norms for hearing children with an average range from 85 to 115.

Receptive vocabulary. *Peabody Picture Vocabulary Test* (PPVT; Dunn & Dunn, 2007) is a receptive vocabulary test standardized on hearing subjects between infancy and adulthood. The examiner provides a spoken label, and the student selects one of four pictures that best represents the label. Testing is discontinued after the student misses 8 out of 12 in a set. Results were expressed as a standard score in relation to hearing age-mates in the normative sample with an average range from 85 to 115.

Elementary School Assessment

Speech perception. *Lexical Neighborhood Test* (LNT; Kirk et al., 1995) measures open-set auditory word recognition in children who are DHH. This open-set test consists of 50 single-syllable words representative of the vocabulary of young children (e.g., *pink, more, hit, juice*). The list contains 25 easy words and 25 hard words as described above for the mLNT. The target words were presented at 60 dB SPL in quiet, and the children responded by repeating the word they heard.

BKB-SIN Speech-in-Noise Test (Etymotic Research, 2005; Bench & Bamford, 1979; Bench, Kowal, & Bamford, 1979) measures a child’s ability to understand speech in background noise. This open-set test consists of lists of sentences, each of which contains three or four keywords. Sixteen or twenty of the sentences were presented in a background of four-talker babble noise (Auditec, 1971)

based on whether the child used cochlear implants or hearing aids. The level of noise increased with each sentence, reflecting easy to difficult listening situations. The target sentences were presented at 65 dB SPL in increasingly difficult signal to noise ratios, and the children responded by repeating each sentence. Based on the number of keywords repeated correctly, a signal to noise ratio (SNR)-50 score is calculated. The SNR-50 score indicates how much louder sentences must be above the noise for a child to understand approximately 50% of spoken words.

Spoken language. *Comprehensive Assessment of Spoken Language* (CASL; Carrow-Woodfolk, 1999) measures spoken language in hearing children between three and 21 years of age across four structural categories: Lexical/Semantic, Syntactic, Supralinguistic, and Pragmatic Language. All children received the core language subtests appropriate for their age: Antonyms, Synonyms, Paragraph Comprehension, Morphemes, Non-literal Language, and Pragmatics. Subtest scores were combined as described in the test manual and results are expressed as standard scores in relation to their hearing age-mates in the normative sample with an average range from 85 to 115.

Vocabulary. *Peabody Picture Vocabulary Test*, 4th edition (PPVT-4; Dunn & Dunn, 2007), described above from the preschool battery, was re-administered at the elementary school assessment.

Reading. *Woodcock Reading Mastery Test*, Revised, 3rd edition (WRMT; Woodcock, 2011) is an individual assessment of reading skills for children and adults. Subtests include Word Identification, Word Attack, Word Comprehension, and Passage Comprehension. Results were expressed as a standard score in relation to hearing age-mates in the normative sample with an average range from 85 to 115.

The Test of Reading Comprehension, 4th edition (TORC-4; Brown, Hammill, & Wiederholt, 2009) assesses silent reading comprehension using five subtests (Relational Vocabulary, Sentence Completion, Paragraph Construction, Text Comprehension, and Contextual Fluency). Results are expressed as a standard score in relation to hearing age-mates in the normative sample with an average range from 85 to 115.

Cognition. *The Wechsler Intelligence Scale for Children*, 5th edition (WISC-V; Weschler, 2014) is an individually administered intelligence test for children between the ages of six and 16 years. The index scores represent a child’s ability in discrete cognitive domains. Non-verbal intelligence (NVIQ) included the following subtests: Block Design and Visual Puzzles (visual spatial skills), Matrix Reasoning and Figure Weight (fluid reasoning skills), Digit Span and Picture Span (working memory), Coding, and Symbol Search (processing speed). Verbal reasoning

(VIQ) included the subtests of Similarities and Vocabulary. Results are expressed as a standard score in relation to hearing age-mates in the normative sample with an average range from 85 to 115.

Objectives

This study addresses both short-term and long-term effectiveness of Moog Center intervention provided to children up to 36 months of age. Short-term outcomes were assessed during preschool (3 to 6 years of age) and long-term outcomes during elementary school grades (8–14 years). Analyses addressed the four following questions.

Question 1: What levels of speech perception, vocabulary, and language are achieved at or near the end of Moog Center EI and preschool intervention?

Question 2: Does intensity of Moog Center intervention between 0–36 months predict children’s language achievement in preschool?

Question 3: What levels of speech perception, vocabulary, language, verbal reasoning, and reading are achieved by Moog Center graduates at or near the end of elementary school?

Question 4: Does intensity of Moog Center intervention between 0–36 months predict children’s language and reading achievement in elementary school?

Results

Question 1: What levels of speech perception, vocabulary and language are achieved at or near the end of Moog Center EI and preschool intervention?

Table 3 summarizes test results gathered when children had completed preschool at the Moog Center or at the point of departure. Out of the 50 children, 25 (50%) scored within one standard deviation of their hearing age-mates (standard score > 85) on the overall language measure (CELF-P) and 82% achieved vocabulary scores on the PPVT within the average range. No statistically significant difference between language standard scores of the 15 children who used hearing aids and those 35 children who used at least one cochlear implant was found. Both device groups achieved average scores within expectation for hearing age-mates (HA = 101 and 92; CI = 95 and 86 for PPVT and CELF-P, respectively) by the time they either reached the end of preschool or exited from the Moog Center program. Aided speech perception scores on the mLNT averaged 78% and did not differ for CI and HA users, although there was large variability in performance. Despite very large differences in unaided PTA thresholds, CI users with severe-profound hearing losses did not differ from HA users with moderate impairment in their ability to understand speech through their devices.

Question 2: Does intensity of Moog Center intervention between 0–36 months predict children’s language achievement in preschool?

The number of intervention hours correlated $r = .348$ ($p = .013$) with speech perception scores on the mLNT, $r = .645$ ($p < .001$) with global language skills measured by the CELF-P, and $r = .537$ ($p < .001$), with receptive vocabulary

Table 3
Preschool Results for Vocabulary, Language, and Speech Perception

	<i>Mean</i>	<i>Standard Deviation</i>	<i>Range</i>	<i>Within Normal Limits (WNL)</i>
Age at Test (years)	4.38	0.66	3.05–6.12	-
Total PPVT ¹	96.7	17.2	46–128	82%
<i>HA users (n = 15)</i>	<i>101.53</i>	<i>20.27</i>	<i>46–128</i>	<i>87%</i>
<i>CI users (n = 35)</i>	<i>94.63</i>	<i>15.63</i>	<i>54–117</i>	<i>80%</i>
Preschool CELF-P2 ²	87.8	18.7	50–125	50%
<i>HA users (n = 15)</i>	<i>91.67</i>	<i>17.63</i>	<i>61–119</i>	<i>60%</i>
<i>CI users (n = 35)</i>	<i>86.20</i>	<i>19.20</i>	<i>50–125</i>	<i>46%</i>
mLNT easy ³	77.7	23.3	8–100	n/a
<i>HA users (n = 15)</i>	<i>78.3</i>	<i>22.7</i>	<i>8–100</i>	<i>n/a</i>
<i>CI users (n = 35)</i>	<i>77.4</i>	<i>23.9</i>	<i>8–100</i>	<i>n/a</i>

Note. HA = Hearing Aid; CI = Cochlear Implant; PPVT = Peabody Picture Vocabulary Test—standard score; CELF-P2 = Clinical Evaluation of Language Function, Preschool Level—standard score; mLNT = Multisyllabic Lexical Neighborhood Test (easy List)—percent correct.

measured by the PPVT. These positive correlations indicate children with more hours of Moog Center intervention between 0 and 36 months of age achieved higher speech perception, language, and vocabulary scores in preschool.

In terms of demographics, correlations between intervention hours over the 0–36 months of age and PTA threshold ($r = -.10$), Mother’s Education ($r = -.08$), and WISC Nonverbal Intelligence ($r = .23$) did not reach statistical significance; however, the correlation with age at first HA was statistically significant ($r = -.584$; $p < .000$). Children who received a HA (and typically began intervention) at younger ages accumulated more hours of Moog Center intervention between 0 and 36 months of age. Thus, it is important to separate the effects of these variables on outcome measures to determine the extent to which age at intervention and amount of early Moog Center intervention independently influence language outcome.

Multiple regression analysis assessed the contribution of intervention hours to preschool CELF scores after accounting for the independent contributions of demographic and child performance characteristics. Table 4 summarizes statistical significance levels for each variable independently. Collectively, the control variables (PTA threshold, age at first HA, mother’s education level, nonverbal intelligence, and mLNT speech perception scores) accounted for 66.72% of the variance in CELF-P scores. Total intervention hours predicted significant added variance above and beyond these control variables, adding 5.85% to the total variance accounted for in CELF-P (total predicted variance = 71.57%). Better preschool language was independently associated with a younger age of fitting a HA, higher nonverbal intelligence, better early speech perception, and more hours of Moog Center intervention between birth and 36 months. Unaided PTA threshold (500, 1K, 2K) and mother’s education level did not contribute statistically significantly to overall variance in CELF-P scores. None of the interactions among predictor variables was statistically significant, and the collective contribution of interactions was not statistically significant.

The regression model coefficients were used to obtain expected CELF-P scores as a function of total intervention hours, and results are plotted in Figure 2. The diagonal solid line represents the mean predicted CELF-P score with the other predictor variables set at their sample means.¹ The function is linear, and the point at which the line crosses the 85 standard score (the cutoff corresponding to one SD below the normative mean) is equal to 187 hours, indicating that half of the cases from any new sample can be expected to achieve a standard score of 85 at 187 hours of intervention. The shaded area around the prediction line is the 95% confidence band, providing an indication of the variability arising from the regression model.

Table 4
Factors Predictive of CELF-P Scores

Predictors	Vocabulary/Language		
	Standard Coefficient	F-ratio	p
PTA Threshold	-0.06	-1.04	0.306
Age at First HA	-0.36	-2.62	0.012
Mother’s Education	0.46	0.65	0.517
Performance IQ	0.38	3.32	0.002
mLNT % Correct	0.25	3.29	0.002
Total Intervention Hours	0.03	2.97	0.005
Explained Variance	71.57%		df = (1,43)

Note. PTA = Pure Tone Average; HA = Hearing Aid; mLNT = Multi-syllabic Lexical Neighborhood Test

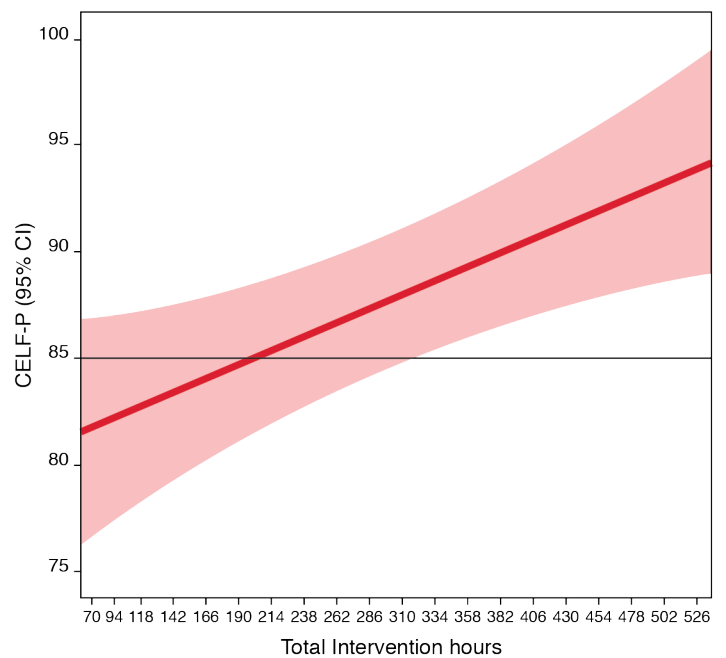


Figure 2. Predicted standard score on the Clinical Evaluation of Language Function-Preschool Test (CELF-P). Control variables (unaided pure tone average threshold, age at first hearing aid, mother’s education level, nonverbal intelligence, and speech perception scores) are set at the sample mean and plotted by total hours of intervention provided at the Moog Center between 0 and 36 months of age. The diagonal line represents the predicted mean and the shaded area around the prediction line is the 95% confidence band, providing an indication of the variability arising from the regression model.

¹Predictors are correlated and some combinations implied in the graph may not be realistic. For example, age at first HA is highly correlated with total intervention hours, which means assuming mean age at first HA at all levels of total intervention hours does not fully match the underlying data. That is one reason the confidence intervals get wider at the extremes; they account for uncertainty in regions for which there is less information

Question 3: What levels of speech perception, vocabulary, language, verbal reasoning, and reading were achieved at or near the end of elementary school?

Table 5 summarizes results obtained on a battery of tests administered to the same 50 children when most were near the end of elementary school (average chronological age = 10.5 years). Both nonverbal (100.3) and verbal (97.2) composite scores on the WISC-V intelligence scale were within the average range, and there was no statistically significant gap between verbal (97) and nonverbal (101) index scores, indicating that these children were realizing their nonverbal potential in verbal reasoning skills. Average scores on the CASL (96.8) and the PPVT (97.5) were within one SD of hearing age-mates (> 85), as were reading scores on both the WRMT (100.2) and the TORC (102.7). Table 5 also summarizes the percent of the sample scoring 85 or higher on each test, ranging from 68% on the CASL global language measure to 92% on nonverbal intelligence. Scores within age-expectation were achieved by more than 75% of the sample for PPVT vocabulary and reading on the WRMT and the TORC.

Average speech perception scores are also presented in Table 5. Mean open-set word recognition on the LNT test was 87%, approaching the ceiling of the test. Scores on the BKB-SIN test indicated that, on average, children understood half of the sentence material when the speech exceeded the noise by 5.3 dB (signal-to-noise ratio). Post-hoc comparisons of speech perception scores for HA ($n = 15$) and CI ($n = 35$) users indicated a statistically significant advantage for HA users in word recognition scores in quiet with LNT mean = 84% for CI and 94% for HA users ($F = 4.25$; $p = .045$). HA users also exhibited statistically significantly lower (i.e., better) SNR ratio on the BKB-SIN (mean = 2.7 dB) compared to CI users (mean = 6.36 dB; $F = 7.46$; $p = .009$).

Mean of subscale score and associated 95% confidence intervals are presented in Figures 3 and 4 for WISC-V and CASL tests, respectively. Average subscale scores were within the average range for hearing age-mates and did not differ statistically significantly from one another except for higher standardized scores for the Visual-Spatial Scale ($M = 105$) than Working Memory Scale ($M = 97$; $F(1,48) = 4.71$, $p = .04$). CASL mean subtest scores were also within normal limits for age, but with statistically significantly lower scores on the Syntax ($F = 12.86$; $p < .0001$) and the Pragmatics ($F = 32.63$; $p < .0001$) subtests.

Average reading subtest scores are presented in Figure 5 for the WRMT and in Figure 6 for the TORC. All of the mean subtest standard scores on the WRMT fell within the average range for hearing age-mates, and no statistically significant differences were observed between decoding skills (word identification, word attack) and comprehension (word comprehension, passage comprehension).

All subtest means on the TORC were within the average range for hearing age-mates, but with higher subtest scores on Text Comprehension and Paragraph Construction compared to Contextual Fluency, Sentence Completion, and Relational Vocabulary. Text Comprehension is a subtest where students are given a list of questions prior to reading a passage, then tasked with answering the questions after silently reading the passage. Paragraph Construction measures the ability to reasonably construct a meaningful paragraph when given a list of sentences in random order. Thus, it appears that these children excel at comprehending connected text.

TORC scaled scores were statistically significantly lower on tasks tapping vocabulary and syntactic knowledge ($F = 58.3$; $p < .0001$). Contextual Fluency is a timed subtest of progressive difficulty, where students are given

Table 5
Average Performance at the End of Elementary School

	<i>Mean</i>	<i>Standard Deviation</i>	<i>Range</i>	<i>% Within Normal Limits (WNL)</i>
Nonverbal Quotient (WISC-V)	100.3	14.1	74–132	92
Verbal Comprehension Index (WISC-V)	97.2	16.4	70–136	72
CASL – Standard Score (SS)	96.8	18.0	66–136	68
PPVT – SS	97.5	17.8	55–132	80
WRMT – Basic Skills SS	97.5	15.1	63–134	84
WRMT – Comprehension SS	100.2	17.4	73–140	76
TORC – SS	102.7	18.4	54–144	86
LNT % Correct	87.1	16.0	22–100	n/a
BKB-SIN (SNR)	5.3	4.6	-1–22	n/a

Note. WISC-V = Wechsler Intelligence Scale for Children; CASL = Comprehensive Assessment of Spoken Language; PPVT = Peabody Picture Vocabulary Test; WRMT = Woodcock Reading Mystery Test; TORC = Test of Reading Comprehension; LNT = Lexical Neighborhood Test; BKB-Sin = Bamford-Kowal-Bench SPeech in Noise.

strings of text containing words in uppercase print without spaces or punctuation. As a measure of their knowledge of words in context, the students must identify as many words as they can by drawing a line between words. Relational Vocabulary measures the student's ability to identify related words using two lists of words. The first list contains three related words and the second list contains

four words with two words related to the first list and two unrelated words. The student must then select the two related words from the second list that relate to the first list of related words. Sentence Completion is a task where the student must fill in a sentence missing two words with the correct word pairs chosen from a list of word pairs.

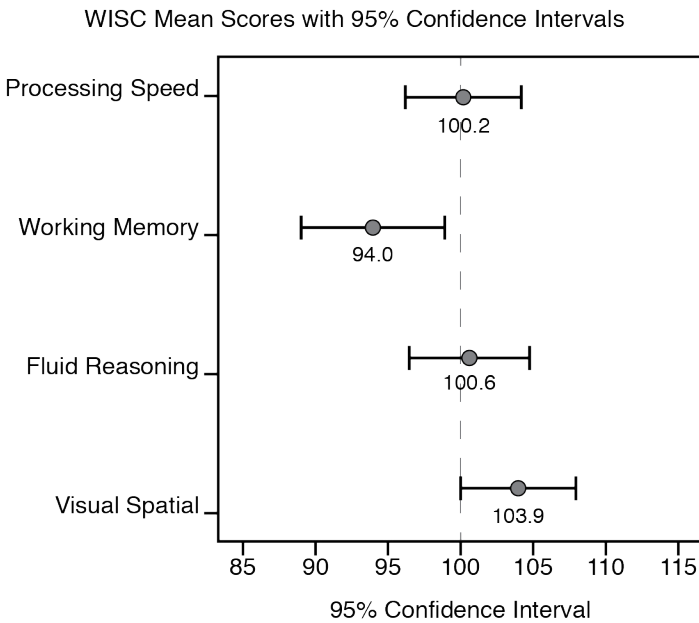


Figure 3. Average subscale standard scores on the Wechsler Intelligence Scale for Children (5th Edition; WISC-V). Scores are plotted for 50 alumni of the Moog Center in elementary grades. Error bars around each mean represent the 95% confidence interval.

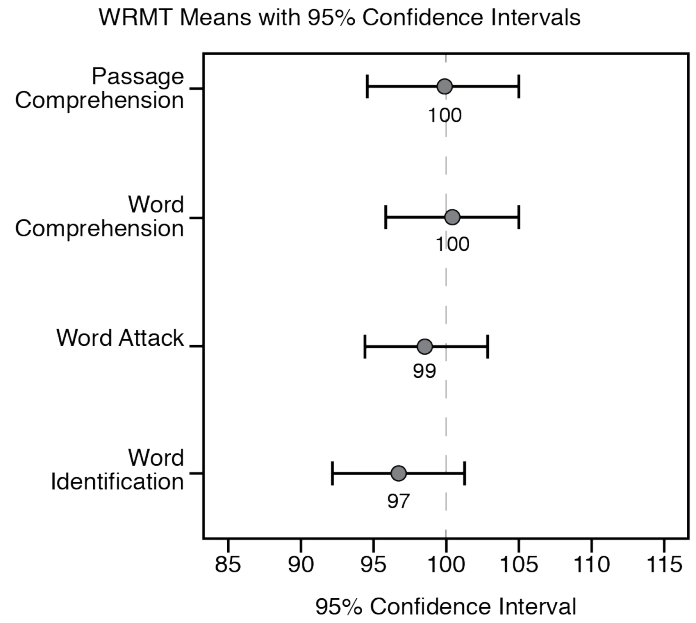


Figure 5. Average subtest standard score on the Woodcock Reading Mastery Test (WRMT). Scores are plotted for 50 alumni of the Moog Center in elementary grades. Error bars represent the 95% confidence interval.

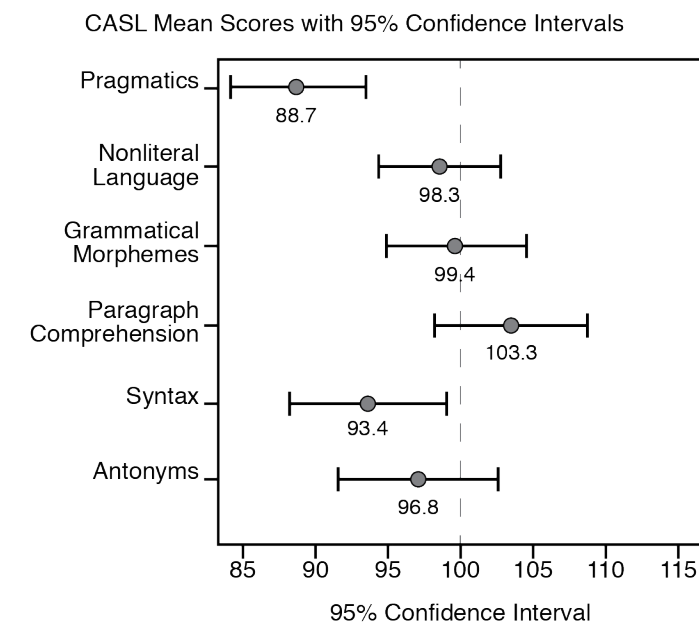


Figure 4. Average subtest standard scores on the Comprehensive Assessment of Spoken Language (CASL). Scores are plotted for 50 alumni of the Moog Center in elementary grades. Error bars represent the 95% confidence interval.

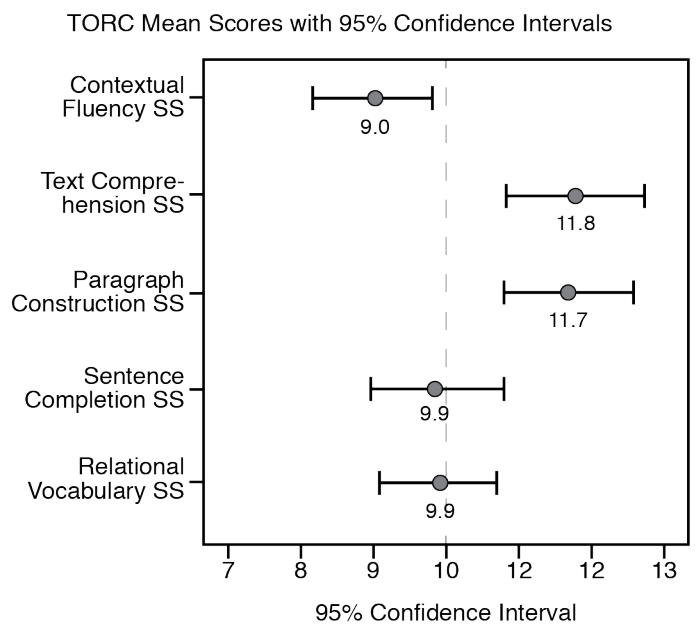


Figure 6. Average scaled standard scores (SS) on the Test of Reading Comprehension (TORC). The average score for each subtest on the TORC is 10, with a range of 7–13. Scores are plotted for 50 alumni of the Moog Center in elementary grades. Error bars represent the 95% confidence interval.

Table 6
Correlations

	LNT in Quiet	BKB-SIN	CASL std Score	WRMT Basic Skill	WRMT Read Comp	TORC Read Comp Index
Intervention Hours	0.131 0.363 50	-0.122 0.399 50	.479** 0.000 50	0.253 0.076 50	.337* 0.017 50	.300* 0.034 50
Multisyllabic LNT easy words	.666** 0.000 50	-.385** 0.006 50	.298* 0.036 50	0.029 0.843 50	0.106 0.463 50	0.146 0.311 50
WISC NVIQ	0.067 0.642 50	0.040 0.782 50	.650** 0.000 50	.554** 0.000 50	.725** 0.000 50	.723** 0.000 50
Age at First HA	-0.033 0.818 50	-0.102 0.483 50	-.491** 0.000 50	-.361** 0.010 50	-.402** 0.004 50	-.371** 0.008 50
Unaided PTA	-.299* 0.035 50	.379** 0.007 50	-0.120 0.407 50	-0.158 0.274 50	-0.046 0.752 50	-0.224 0.117 50

Note. WISC-V = Wechsler Intelligence Scale for Children; CASL = Comprehensive Assessment of Spoken Language; WRMT = Woodcock Reading Mastery Test; TORC = Test of Reading Comprehension; LNT = Lexical Neighborhood Test; BKB-SIN = Bamford-Kowal-Bench Speech in Noise; NVIQ = Nonverbal Intelligence; HA = Hearing Aid; PTA = Pure Tone Average.

*Correlation is significant at the 0.05 level (2-tailed).

**Correlation is significant at the 0.02 level (2-tailed).

Question 4: Does intensity of Moog Center intervention between 0–36 months predict language and reading achievement in elementary school?

Table 6 summarizes correlations between four predictor variables (Age at first HA, Nonverbal IQ, mLNT speech perception score, and Moog Center intervention hours) with the five language and reading outcomes measured in elementary school. Number of hours of Moog Center intervention (0–36 months) correlated $r = .479$ ($p < .001$) with language level, $r = .337$ ($p = .017$) with reading comprehension on the WRMT, and $r = .300$ ($p = .043$) with total score on the TORC.

To establish whether this relation remains strong after other predictor variables are controlled, multiple regression analyses were conducted to predict variance in CASL Total Language standard scores and WRMT total reading scores from four predictor variables: age at first HA, nonverbal IQ, mLNT speech perception scores in preschool, and total intervention hours 0–36 months of age. Results for the CASL appear in Table 7. Together with interactions, predictor variable accounted for 70% of total variance, with nonverbal IQ and total Moog Center intervention hours reaching statistical significance along with the interaction between mLNT speech perception and intervention hours. This result indicates that language scores in elementary school were associated with the child’s cognitive ability and the amount of EI they received at the Moog Center. In addition, the statistically significant interaction between speech perception and intervention reflected the tendency

Table 7
Factors Predictive of CASL Scores

Predictors	Language		
	Standard Coefficient	F-ratio	p
Age at First HA	-0.23	-1.29	0.203
Nonverbal IQ	0.72	5.08	< 0.001
mLNT % Correct	0.04	0.51	0.616
Total Intervention Hours	0.02	2.18	0.035
Age at HA x Nonverbal IQ	0.01	-2.03	0.146
Age at HA x mLNT	-0.02	-2.03	0.050
Age at HA x Inter. Hrs	0.00	-0.17	0.864
NVIQ x mLNT	0.01	1.22	0.230
NVIQ x Interv. Hrs	0.00	1.55	0.128
mLNT x Interv. Hrs	-0.01	-3.59	0.001
Explained Variance	70%		df = 1,39

Note. CASL = Comprehensive Assessment of Spoken Language; HA = Hearing Aid; mLNT = Multi-syllabic Lexical Neighborhood Test; NVIQ = Nonverbal Intelligence. Boldface indicates significance.

for children with the poorest speech perception to benefit the most from intensive EI while those with high preschool speech perception benefitted the least.

Results of regression analysis to predict WRMT total reading scores are summarized in Table 8. Predictors accounted for 65% of the variance in reading scores. Nonverbal IQ was the only statistically significant predictor. In addition, the interaction between preschool speech perception and intervention hours was a statistically significant predictor of reading outcome, indicating that those with the poorest speech perception in preschool showed the most reading benefit from large doses of intervention during the 0 to 36 month period.

Table 8
Factors Predictive of WRMT Scores

Predictors	Language		
	Standard Coefficient	F-ratio	p
Age at First HA	-0.01	-1.00	0.323
Nonverbal IQ	0.05	5.31	< 0.001
mLNT % Correct	0.01	-1.15	0.256
Total Intervention Hours	0.00	1.11	0.275
Age at HA x Nonverbal IQ	0.01	-2.03	0.146
Age at HA x mLNT	0.00	0.56	0.578
Age at HA x Inter. Hrs	0.00	0.68	0.498
NVIQ x mLNT	0.00	0.58	0.568
NVIQ x Interv. Hrs	0.0.00	1.12	0.272
mLNT x Interv. Hrs	-0.00	-2.51	0.016
<i>Explained Variance</i>	65%		<i>df = 1,39</i>

Note. HA = Hearing Aid; mLNT = Multi-syllabic Lexical Neighborhood Test; NVIQ = Nonverbal Intelligence; WRMT = Woodcock Reading Mastery Test. Boldface indicates significance.

Summary

This study documents speech perception and language outcomes in preschool and elementary school and reading outcomes in elementary school for a group of 50 alumni representative of participants in the Moog Center for Deaf Education. The report describes levels of achievement at both ages and examines the effectiveness of the Moog Center EI program between birth and 36 months for later achievement. The following findings were supported by the data examined:

- Average language test scores at the end of preschool (or upon leaving the Moog Center) were within age-appropriate expectations for hearing children and remained at comparable levels when tested in elementary school.
- When the Moog Center alumni were assessed in elementary school, both their basic reading skills and reading comprehension levels were, on average, within age-appropriate expectations for hearing children. Both their verbal and nonverbal cognitive/reasoning abilities averaged within age-appropriate expectations for hearing children, with no statistically significant gap between verbal and nonverbal skill levels.
- HA users with moderate hearing loss did not differ from CI users with severe-profound hearing loss in their vocabulary comprehension, language, or reading scores, despite statistically significantly better unaided hearing thresholds and aided speech perception scores, especially in noise.
- Children with more hours of Moog Center intervention between 0 and 36 months of age achieved higher language scores at the end of preschool and in elementary school than children with less EI, after accounting for the positive effects of younger age at hearing aid fitting/intervention, higher cognitive level, and better speech perception.
- Children with poorer speech perception levels in preschool received more benefit from greater amounts of EI at the Moog Center than did children with better speech perception levels. This benefit was apparent for both language and reading.

Conclusions

For some children who are DHH, particularly those who are slow to develop aided auditory perception of speech, early intervention alone may not be sufficient to ensure age-appropriate spoken language development. For these children, the intensity of early (0–36 months) intervention provided at the Moog Center contributed significantly to long-term development of language and literacy over and above the benefits associated with the age at which intervention was initiated. The large dose of intervention provided by group instruction beginning as young as 18 months of age at the Moog Center is atypical for early intervention programs for children who are DHH, where parents are viewed as the child’s primary teachers and intervention is focused on coaching them in language stimulation techniques. The results of this study are consistent with those reported by Moog and Geers, 2010, showing substantial language benefits from participation in a toddler class. This study extends those findings by (a) quantifying the number of hours of intervention provided and (b) following language outcomes into elementary grades and examining long-term benefits for learning to read. Because early educational intervention plays a vital

role in language and academic success for children who are DHH, it is important to document the effects of the amount and intensity of intervention using a particular instructional approach. Further research is needed to assess the benefits of extending intensive intervention for children whose language delay persists beyond the preschool years, when children in LSL programs are often placed in regular education settings with hearing age-mates.

As in studies with other language-delayed populations, greater intervention intensity was more beneficial for some children than for others. Those children with poor aided speech perception scores in preschool exhibited the most benefit from early intensive intervention. Regardless, for all 50 alumni of the Moog Center, average language scores were within expectation for hearing children their age in preschool and remained within this range when they were assessed an average of four years later in elementary school grades. This longitudinal finding suggests that the early language foundation provided through intensive special education at the Moog Center continued to benefit these children through age-appropriate language and literacy in general classroom placement with their hearing age-mates.

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Development of a State-Wide Database of Early Intervention and Educational Outcomes for Children who are Deaf or Hard of Hearing

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Abstract: Children who are born deaf or hard of hearing (DHH) are at increased risk for delays in language, cognitive and social-emotional development. Early identification through screening and early intervention (EI) can improve outcomes for children who are DHH. However, a need remains to evaluate the effectiveness and practices of statewide programs for children who are DHH. The Ohio Early Hearing Detection and Intervention (EHDI) Data Linkage Project was created as a state-wide collaborative that included multiple Ohio government agencies and an academic institution. The objective of the project was to develop and characterize a population-based, longitudinal database that documents state-level services and outcomes for children who are DHH identified through a state EHDI Program. The database includes information regarding birth data, EHDI program data, early intervention data, and early academic data. Children born in Ohio between 2008 and 2014 identified with permanent hearing loss ($n = 1746$) served as the cohort for this project; 1262 records linked with EI data and 502 records linked with education data. Multi-agency linked databases contain novel combinations of data and can be valuable resources for public health evaluative and epidemiologic research. This resource can expand our understanding of the early predictors of academic success for children who are DHH.

Key Words: deaf/hard of hearing, outcomes, early hearing detection and intervention, EHDI, data linkage

Acronyms: ASQ:SE = Ages and Stages Questionnaire: Social-Emotional; CCHMC = Cincinnati Children's Hospital Medical Center; CDC = Centers for Disease Control and Prevention; DHH = deaf or hard of hearing; DODD = Ohio Department of Developmental Disabilities; ECTA = Early Childhood Technical Assistance Center; EHDI = early hearing detection and intervention; EI = Early Intervention; EMIS = Education Management Information System; IEP = Individualized Education Program; IFSP = Individualized Family Service Plan; JCIH = Joint Committee on Infant Hearing; LDS = Language Development Scale; ODE = Ohio Department of Education; ODH = Ohio Department of Health

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Children who are born deaf or hard of hearing (DHH) are at a significantly higher risk for delays in language, cognitive, and social-emotional development (Ching et al., 2010; Holt, Beer, Kronenberger, Pisoni, & Lalonde, 2012; Lund, 2015; Meinzen-Derr, Wiley, Grether, & Choo, 2011, 2013; Meinzen-Derr et al., 2014; Stevenson et al., 2011; Tomblin et al., 2015; Wiley, Meinzen-Derr, Grether, Choo, & Smith, 2015; Yoshinaga-Itano, 2006). In fact, deficits in language often worsen through the school years (Geers, 2003; Marschark, 2003; Stevenson, McCann, Watkin, Worsfold, & Kennedy, 2010), placing children who are DHH at severe disadvantage in many areas of development and wellness. Additionally, without appropriate interventions, these disparities can extend to adulthood, affecting academics (Luckner, Sebald, Cooney, Young, & Muir, 2005; Traxler, 2000), literacy (Traxler, 2000), and employment opportunities (Van Naarden Braun, Yeargin-Allsopp, & Lollar, 2006). All 50 states and the District of Columbia have established Early Hearing Detection and Intervention (EHDI) systems in order to “maximize linguistic competence and literacy development for children who are deaf or hard of hearing” (Joint Committee on Infant Hearing [JCIH] & Pediatrics, 2007, p. 898). As such the Joint Committee on Infant Hearing recommends infants receive hearing screening by one month of age, have a diagnostic evaluation by three months of age, and if diagnosed with hearing loss, receive appropriate intervention by six months of age (1-3-6).

Early identification through screening and early intervention (EI) can improve language development for children who are DHH and reduce discrepancies in non-verbal cognitive functioning and language development (Yoshinaga-Itano, 2003; Yoshinaga-Itano, Sedey, Wiggin, & Chung, 2017). However, a need remains to evaluate the effectiveness and practices of statewide programs for children who are DHH. Recently, Yoshinaga-Itano et al. (2017) evaluated the EHDI 1-3-6 guidelines as they applied to children with bilateral hearing loss across 12 different states. Investigators assessed the impact of the current EHDI 1-3-6 guidelines and made additional recommendations regarding the evaluation of early intervention services on outcomes (Yoshinaga-Itano et al., 2017). Further large-scale evaluations will enable policy-makers and practitioners to implement improvements to these systems and subsequently, mitigate the developmental disparities that persist for children who are DHH.

Fundamental limitations to large population-based evaluations include the lack of integrated and longitudinal data. Important EI process and outcome measures often exist across disparate state departments and databases. Further, key evaluation measures must be abstracted and integrated from these datasets at multiple intervals including birth (birth records and newborn screening outcomes), birth to 3 years (early intervention services), and school age (preschool and later academic services and outcomes). The ability to leverage multiple sources of population-based data (often stored in public health and

education departments) to support observational research is growing in feasibility. This research includes quasi-experimental studies to examine program effectiveness and epidemiological studies to determine predictors of developmental outcomes. Integrating sources of information through novel data linkages has been used to support similar, yet unrelated efforts (Folger, 2013; Hall et al., 2014). Briefly, the process of data linkage involves deterministic and/or probabilistic algorithms to join databases that contain common individuals (e.g., children who are DHH), and unique measures such as sociodemographic characteristics, service utilization (e.g., types and intensity of preventive services), and health and academic outcomes. These linked databases contain novel combinations of data and can be valuable resources for public health evaluative and epidemiologic research.

The U.S. Department of Education mandates that states evaluate the effectiveness of EI and early childhood special education programs. In the state of Ohio, the following outcomes are priorities and mirror the national outcomes identified by the Early Childhood Technical Assistance Center (ECTA): (a) positive social-emotional skills (including social relationships); (b) acquisition and use of knowledge and skills (including early language/communication); and (c) use of appropriate behaviors to meet their needs (Early Childhood Technical Assistance Center & FPG Child Development Institute of the University of North Carolina at Chapel Hill, 2019). As part of Ohio’s State Systemic Improvement Plan, the EI program has emphasized the acquisition and use of knowledge and skills (including early language/communication) for children who are DHH. However, these outcomes are not available to state EHDI programs, hindering robust evaluation efforts. In Ohio, separate departments manage data that document newborn screening, EI service, and education outcomes. These departments do not currently share a common data system. However, approximately 200 children are identified annually with permanent hearing loss, and these children will cross over departments/programs as they age.

Our objective was to develop a population-based database of linked records across multiple state systems for children identified with permanent hearing loss in the state of Ohio who had been served by the EHDI system. The public data sources included records from the newborn hearing screening program (Ohio’s EHDI program), EI, and educational records. We characterize the process and challenges of developing a state-level, population-based DHH resource and share findings from an initial data linkage.

Method

Participants

The target population included children born in Ohio between January 1, 2008 and December 31, 2014 who were identified with permanent hearing loss through the

EHDI program. A cohort of 1,746 children were born during the study period, identified with permanent hearing loss, and entered into the EHDI tracking and surveillance system for the state of Ohio. These initial records were linked to data available through public health and educational data systems.

Procedures

Partners. A state-wide collaborative was formed under the auspices of an initiative launched by the Centers for Disease Control and Prevention (CDC) EHDI and implemented by Cincinnati Children’s Hospital Medical Center (CCHMC). The Ohio EHDI Data Linkage Project included participation among multiple Ohio government agencies including the Ohio Department of Health (ODH), the Ohio Department of Developmental Disabilities (DODD), and the Ohio Department of Education (ODE). The collaboration among multiple agencies required data sharing agreements between CCHMC and each agency (i.e., ODH, DODD and ODE). In 2017, agreements were executed, and institutional review board approval granted by the CCHMC and ODH. Subsequently, data were provided to integrate multiple sources of data including vital records and hearing screening, EI, and early education (i.e., preschool to 2nd grade) educational records.

Data Linkage. The creation of an integrated database required two distinct interdepartmental data linkages performed across three data systems. The first data linkage was performed between newborn hearing screening/follow-up data and EI records. Newborn screening data were stored in the HiTrack (version 4.6.1) surveillance system and were provided by ODH. HiTrack is an EHDI database for managing EHDI tracking and follow-up (HiTrack EHDI Data Management System). The EI data were collected and managed by the Ohio DODD and stored in the Early Track data system (Early Track Early Intervention Data System). Early Track data contained information on developmental assessments and eligibility, diagnosed conditions, and EI service engagement. Further, Early Track contained a unique student school identification number that served as a unique master student index used to link both EI and Ohio public schools data. The linkage between the HiTrack and Early Track systems was performed onsite at ODH and under supervision of both ODH and Ohio DODD program staff. Following this data linkage described in detail below, all personal identifiers were removed.

The initial data linkage (i.e., HiTrack-Early Track) was a multistep process that required matching records on multiple personal identifiers. The SAS server via Enterprise Guide 7.1 was used to maximize computational resources. The SAS SQL (Structured Query Language) procedure was used to match records with a deterministic algorithm that used child characteristics (i.e., gender, date of birth, first name, and last name) and maternal characteristics (i.e., first name, last name, and date of birth). Prior to running the matching algorithm and classifying the links,

we removed all special characters and spaces from the infant name and mother name fields and converted all characters to uppercase. Matched pairs of records were classified according to the number of shared maternal-child identifiers. This approach was adapted from similar past research that used Ohio data sources (Bowers et al., 2018). The classification methodology is depicted in Figure 1. Records were classified as (a) complete matches on all identifiers, (b) maternal partial matches (complete matches except for mother’s date of birth), and (c) matches of only child’s information. First, we selected records that had a perfect match on all criteria. Next, we selected maternal partial matches. Maternal partial matches were largely due to missing dates of birth. Finally, we selected records that matched only on all infant identifiers; this was the least specific approach, but allowed for manual review of potential matches (where either the mother’s first or last name matched). Following each stage of matching, we manually verified records that linked only using infant characteristics (did not link on mother’s first or last name). Using this linking methodology, nearly 20% of records from HiTrack were successfully linked to Early Track records using all of the mother-infant matching variables. An additional 47.1% of the records were matched using all variables except for mother’s date of birth (Figure 1). Nearly 3% of records were matched using only infant characteristics. Once the linkage between HiTrack and Early Track was complete, a unique identifier was assigned to each individual and the identifiers used in the linkage were removed from the final dataset. A separate dataset was created that contained both the unique identifiers assigned to individuals and the identifiers that were used in the linkage process; ODH maintained the database and served as the gatekeeper. This dataset functioned to verify records for outliers and missing values as necessary. Only ODH and DODD had access to the key identifier. The second data linkage was performed to merge the academic outcomes including early educational

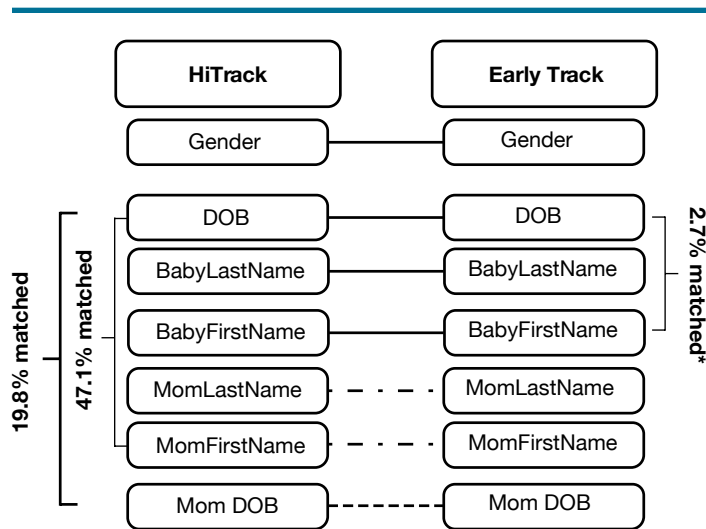


Figure 1. Criteria for the linkage of newborn screening/follow-up data and early intervention data. * Indicates records that required manual verification. DOB = date of birth.

assessments, socio-emotional assessments, and disability codes provided by the ODE through the Education Management Information System (EMIS). EMIS is a statewide data collection system for Ohio's primary and secondary education. The EMIS data were de-identified and provided in Microsoft Excel file format. The unique student identification number was used to perform a simple merge of the HiTrack Early Track combined data to the EMIS data.

Analysis

Simple descriptive analyses were conducted to compare the full cohort of children who were identified as DHH relative to those who enrolled in EI and those with data linked to education outcomes. Because this study was focused on successful data linkages and not the testing of a specific hypothesis, we did not conduct any statistical testing.

Results

There were 1,746 babies identified as DHH through the Ohio EHDI program between January 1, 2008 and December 31, 2014. Among the identified infants, 1,262 (72.3%) were linked to an enrollment record within EI and 502 unique individuals had matched education records (Figure 2). Four hundred eighty-four EHDI records did not have a corresponding Early Track record. Infants who did not have documentation of enrolling into EI would not have data within the Early Track system. Of the 1,262 successfully linked Early Track records, 760 records could not be linked to EMIS (education data). Likely reasons for our inability to link these records to EMIS include a child (a) was not enrolled into preschool, (b) was not of school age, (c) attended a private school but did not have an Individualized Education Program, and/or (d) no longer lived within the state of Ohio. Table 1 describes the characteristics of the infants by linked groupings.

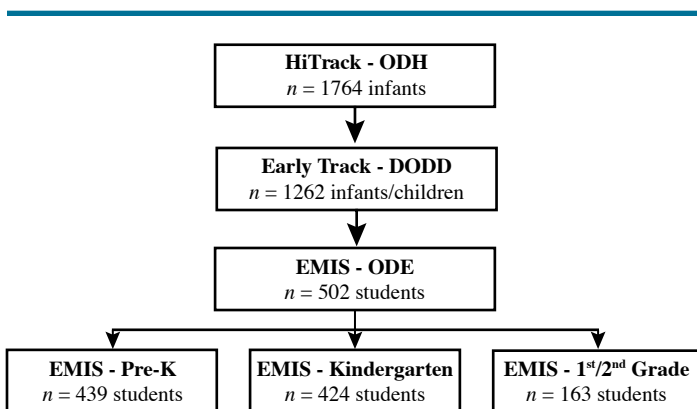


Figure 2. Data Linkage Results: Number of linked individuals with data across three Ohio data systems. DODD = Ohio Department of Developmental Disabilities; EMIS = Education Management Information System; ODE = Ohio Department of Education

Table 1
Characteristics of DHH Infants in Ohio by Data Linkage Status

Characteristics	All infants N = 1746	Linked to EI n = 1262	Linked to EMIS n = 502
Gender- Male	892 (51.1%)	684 (54.2%)	281 (56%)
Race			
Caucasian	1227 (70.3%)	952 (75.4%)	397 (79.1%)
Black/African American	228 (13.1%)	155 (12.3%)	68 (13.6%)
Asian	34 (2%)	24 (1.9)	9 (1.8%)
Other	75 (4.3%)	49 (3.9%)	13 (2.6%)
Unknown	182 (10.4%)	82 (6.5%)	15 (3.0%)
Ethnicity-Hispanic	80 (4.6%)	55 (4.4%)	14 (2.8%)
Gestational age in weeks (SD)	37.3 (3.5)	37.3 (3.5)	37.3 (3.4)
Birth weight in grams (SD)	2952 (836)	2959 (845)	2951 (859)
Born Premature	367 (21.0%)	270 (21.4%)	118 (23.5%)
Maternal Education			
Less than high school	219 (12.5%)	147 (11.7%)	59 (11.8%)
High school	390 (22.3%)	271 (21.5%)	117 (23.3%)
Some college	473 (27.1%)	369 (29.2%)	145 (28.9%)
College graduate	413 (23.7%)	343 (27.2%)	148 (29.5%)
Missing	251 (14.4%)	132 (10.5%)	33 (6.6%)
Median [IQR] age in months of hearing loss confirmed	3.9 [1.9-9.6]	3.9 [1.9-9.0]	4.0 [1.8-9.0]
Has risk indicator for hearing loss	674 (38.6%)	507 (40.2%)	229 (45.6%)
Bilateral hearing loss	1285 (73.6%)	897 (72.5%)	393 (78.3%)
Degree of loss in worse ear			
Slight/Mild	538 (30.8%)	380 (30.1%)	159 (31.7%)
Moderate	263 (15.1%)	194 (15.4%)	80 (15.9%)
Mod-Severe	229 (13.1%)	170 (13.5%)	60 (12.0%)
Severe	117 (6.7%)	84 (6.7%)	40 (8.0%)
Profound	486 (27.8%)	364 (28.8%)	132 (26.3%)
Unknown	113 (6.5%)	70 (5.5%)	31 (6.2%)

Note. DHH = deaf or hard of hearing; EI = Early Intervention; EMIS = Education Management Information System; mod-severe = moderately severe.

Education

Final Linked Database

Birth and screening data. The Ohio EHDI Data Linkage Project resulted in a comprehensive database containing a large number of birth, hearing screening, and EI variables. Demographic fields included maternal age (at child's birth), race and ethnicity, education level of the mother and the father, and insurance status/payer. Fields that characterized the birth included gestational age at birth (weeks), birthweight (grams), Apgar score, risk factors specific for hearing loss, and pregnancy-related risk factors. Hearing-specific information was characterized in

fields including age at screening and diagnosis (screening and diagnosis dates), laterality of hearing loss (unilateral/bilateral), and degree of loss in each ear (e.g., mild, moderate, moderate-severe, severe, and profound).

EI specific data. The EI service fields included dates of evaluation and individualized family service plan (IFSP), documented developmental delays and disabilities, types of services, frequency and duration of services (dates of service), and the presence of diagnosed conditions. The file indicated whether a child scored >1.5 standard deviations below the population mean on standardized assessments in the categories of cognitive, social-emotional, communication and language, and gross and fine motor development. Because of the EI system specific for children who were DHH at the time of data collection, language development was captured within the HiTrack system. At the time, language was assessed using the SKI*HI Language Development Scale (LDS; Tonelson & Watkins, 1979).

Academic data. Data pertaining to the Individualized Education Program (IEP) were obtained from the educational record, such as the disability eligibility category, dates of the IEP, and grade level and age of the child. Multiple outcomes were available for children who were served in a preschool classroom. The Ages and Stages Questionnaire: Social-Emotional (ASQ:SE; Squires, Bricker, & Twombly, 2002) was used to measure the outcome of social-emotional development of children. The ASQ:SE is a well-validated, parent-completed screening tool that contains items to assess the dimensions of self-regulation, compliance, communication, adaptive functioning, autonomy, affect, and interaction with people. The Get it! Got it! Go! is a preschool assessment used to assess critical early literacy skills (i.e., picture naming, rhyming, and alliteration), and is administered multiple times during the academic year after the age of 3 years (Early Childhood Research Institute on Measuring Growth and Development, 1998).

The Early Childhood Outcome Summary assesses social-emotional skills, acquiring and using knowledge and skills, and taking appropriate action to meet needs. The Early Learning Assessment measures awareness & expression of emotion, cooperation with peers, phonological awareness, communication, coordination, safety-injury prevention, relationships with adults, vocabulary, numbers, and personal care in preschool children. Ohio's Kindergarten Readiness Assessment (KRA) measures school readiness aligned to Ohio's Early Learning and Development Standards (birth to kindergarten) and is intended to be used by teachers to improve outcomes for all kindergarten children enrolled in public or community schools. The Language and Literacy area of the KRA may be used for the K diagnostic requirement of the Third Grade Reading Guarantee as it measures students' skills in the areas of early reading, letter recognition and using words in conversations. The KRA includes 50 questions

that address a child's growth and development in four main areas, Language and Literacy, Social Foundations, Mathematics, and Physical Well-Being and Motor Development.

Discussion

The Ohio EHDI Data Linkage Project demonstrates the successful development of an integrated data source to support observational research that is needed to improve outcomes for children who are DHH. The resulting process has established a roadmap for expanding these efforts to states beyond Ohio. The need for evaluation is apparent as deficits in language development persist despite the implementation of newborn screening programs for hearing loss and EI programs for children birth to 3 years of age with the focus of mitigating developmental risks for children who are DHH.

To our knowledge, this is the first study to characterize the successful development of a population-based, longitudinal database that documents state-level services and outcomes for children who are identified as DHH through a state EHDI Program. This new resource can provide novel integrated data to support program evaluation and epidemiologic research with a focus on key child developmental and family outcomes important for EI services (Early Childhood Technical Assistance Center & FPG Child Development Institute of the University of North Carolina at Chapel Hill, 2019). Through this project, we were able to demonstrate the feasibility of developing a resource that could enable Ohio and other states to evaluate the effectiveness of early age EI enrollment (i.e., by six months of age, meeting the EHDI benchmark) to improve language outcomes and early academic outcomes, such as pre-literacy and kindergarten readiness. Such studies can provide evidence for the advent of the 1-3-6 EHDI benchmarks while addressing fundamental questions regarding the types and intensities of different EI services. This resource may also simulate opportunities to measure the successful and unsuccessful connection points between important programs for children who are DHH. Cross-system linkages provide the data that can facilitate system-level quality improvement efforts that promote quality interface between entities such as EI and the education system.

Although many studies address language and communication skills, the literature is lacking in understanding broader domains of development and early predictors of academic success. A comprehensive longitudinal database is an innovative resource that has the potential to address questions about predictors of social-emotional development and academic success in children who are DHH. Because we were able to link to the education system, we have the opportunity to assess outcomes beyond language and beyond the birth to 36 month period; and provide a picture of the educational trajectory for children who are DHH as they grow. Once

this picture is provided, these data can provide powerful evidence in support of state-based EHDI and EI systems.

Project Challenges

We encountered several challenges regarding the project. Although we had established collaborations across the 3 state agencies involved with the project, these agencies were disparate, operating as independent entities in mission, data systems, and policy. This required approximately six months of various approvals for data use agreements and memoranda of understanding between the agencies and our academic institution.

Our linkage methodology was based on an algorithm that required a perfect match of infant records. Employing a strict algorithm potentially misses infants in the linkage process. The solution would require additional manual verification of all infants believed to have received EI services. This activity would have been a large endeavor and would not be a feasible model if this program were to be replicated regularly for state-level program evaluation. An alternative approach would be to use probabilistic algorithms to facilitate additional matches (Mneimneh et al., 2013); however, deterministic methods (as employed in our study using names) likely mitigate misclassification of matches (Kotelchuck et al., 2014). Further, more complex matching algorithms often require special expertise in statistical methodology and would not necessarily preclude manual verification of matches. These more complex methods may not be as accessible or readily adopted by programs, diminishing the overall feasibility of this project in public health practice.

Certain sub-populations such as transient families may require collaboration across state systems to ensure adequate linkage. Although documentation of every record achieves a population-based approach, conditioning the sample on linked individuals represents the vast majority engaged in the system. The linkage to the EI database, Early Track, resulted in 1,252 matches, providing a robust sample available to form important questions regarding the impact of the system on child outcomes. Modest enhancements to state systems could also facilitate these linkages. For example, maternal date of birth, an important linkage variable, was absent in most records within the HiTrack system (i.e., newborn screening program). Although this did not greatly inhibit the linkage process, the same may not be true in other state systems. Additionally, misspellings in the mother's last name required manual verification of "near matches" (matches that were close with the exception of the mother's last name). Creating or adapting current systems to better capture the appropriate spellings would decrease the need for manual verifications. Other reasons for the inability to link records on the mother's identifiers include name changes (e.g., due to marriage or divorce) as well as alternative caregivers (e.g., foster care, in the care of other guardians). Misclassification of true matches as non-matches results in a reduced total sample, but if occurring infrequently and randomly, could still result in

a large, unbiased sample to support evaluation. Manual review would be necessary to ensure appropriate linkage and classification in these instances, but ultimately improved documentation, data exchange, and data archiving within state systems will improve inter-departmental/system linkages. Through collaborations across system and states, independent research efforts could be used to identify the extent and reasons for missing data. Subsequently, quality improvement approaches could be pursued to ensure higher data accuracy at the time of collection; however, we must acknowledge that without shared systems of data capture, the potential for missing records will remain using linkage approaches.

Although novel population-based data may stimulate the evaluation of state systems designed to support DHH children, data are largely collected for administrative purposes and can lack the rigor required within research protocols. Relatedly, the Ohio EHDI Data Linkage Project currently lacks data on certain family characteristics and comprehensive measures of service engagement. Nevertheless, opportunities remain to refine the data capture by programs such as EI to document the quantity and content of service visits relative to expectations. Although some challenges that families face (e.g., poverty and other adversity) can be reasonably identified from existing data (e.g., insurance status), there remains limited information collected on the array of family factors such as involvement and parenting stress. Gaps identified in data may inform states on how to optimize new system-level data collection procedures.

This project has several strengths including (a) the collaboration of multiple state agencies and academic institutions; (b) integration of population-based data on children who are DHH; (c) the development of a roadmap for promoting the necessary inter-agency collaborations and commitments; and (d) demonstration of real-world outcomes data available for both evaluation and epidemiologic analyses.

Conclusion and Implications for Future Work

Through collaborations with state agencies, we were able to demonstrate that an integrated data system is feasible. The availability of such a comprehensive data system can help investigators, whether public health or academic, address relevant and important topics regarding short and long-term outcomes for children served in state EHDI programs. Not only does this project demonstrate that partnerships and innovative data linkages across state information systems can serve as a model for other state EHDI programs, it can also serve as a model for public health programs serving the broader population of children with disabilities. This work has broad implications for public health practice regarding infants who are DHH based on findings showing the positive impact of early entry into EI on language and a possible sustaining effect on early academic outcomes.

In our next phase of data analysis, we will further characterize the population of DHH children in the linked statewide database. This will include describing the observed early social-emotional and literacy skills (preschool), kindergarten readiness, and important early education outcomes (namely, emergent literacy skills). We also plan to use quasi-experimental approaches to evaluate the impact of EI services on key child outcomes.

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Newborn Hearing Screenings for Babies Born at Home: Report from an Initiative in Michigan

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Abstract: Objective: Babies born in an out-of-hospital setting (e.g., homebirth) often do not receive a universal newborn hearing screening (UNHS). The purpose of this study was to evaluate the effect of providing training and equipment for newborn hearing screening to midwives who attend homebirths.

Study Design: Midwives from around the state of Michigan were invited to participate in a two-part UNHS training. Hearing screening data from all midwives who attended homebirths ($N = 112$) during the 2015 and 2016 calendar years were analyzed using a two-level multilevel model. Estimated odds of babies being screened were calculated based on midwife group.

Results: Having a midwife who hosted an Automated Auditory Brainstem Response (AABR) machine at her practice increased the odds of receiving a screening by 39.37 times. Having a midwife who had access to an AABR machine increased the odds of receiving a screening by 8.57 times. Having a midwife who received focused education about the importance of newborn hearing screening increased the odds of receiving a screening by 10.82 times.

Conclusion: Providing UNHS equipment and training to midwives significantly increases the odds that babies born at home will receive a hearing screening at birth. This is evidence for the continued outreach and inclusion of midwives in UNHS programs.

Key Words: newborn hearing screening, homebirth, midwifery

Acronyms: AABR = Automated Auditory Brainstem Response; EHDl = Early Hearing Detection and Intervention; SWOT = Strengths, Weaknesses, Opportunities, and Threats; UNHS = universal newborn hearing screening

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Having a homebirth is a choice that an increasing number of Americans are making (MacDorman, Declercq, & Mathews, 2013; MacDorman, Mathews, & Declercq, 2012). There has been a 39% increase in the overall proportion of out-of-hospital births in the United States from 2004–2010 (MacDorman et al., 2013). Unfortunately, in a Strengths, Weaknesses, Opportunities, and Threats (SWOT) analysis of Early Hearing Detection and Intervention (EHDI) programs across the country, homebirths were listed as the third most reported weakness (12% of respondents; Houston, Bradham, Muñoz, & Guigand, 2011). Concerns included lack of follow-up for homebirths and many EHDI coordinators reported that the majority of babies born at home did not receive a screening (Houston et al., 2011). Many families who choose to have a homebirth face financial, cultural, educational, or logistical barriers when trying to obtain a newborn hearing screening.

Most homebirths (70.1%) in the United States are attended by a midwife (MacDorman et al., 2013), and midwives have professional responsibilities in the newborn hearing screening process. The American College of Nurse-Midwives Core Competencies (2012) indicates that the midwife independently manages and provides care for newborns up to 28 days of life. In addition, according to the Midwives Alliance of North America Core Competencies for Midwifery Practice (2014), the midwife provides postpartum care to the newborn as well as support and information to parents about screening tests and the applicable laws and regulations, including newborn hearing screening. In the state of Michigan (MI), for example, the state guidelines for newborn hearing screening state that, “Health professionals who provide birthing services outside of a hospital will ensure that a newborn hearing screening is completed within one month of the birth” (MI Early Hearing Detection and Intervention System, 2002). In Michigan, the term *health professional* is typically interpreted as a professional who holds a license in their health care field. Midwives are not currently licensed in Michigan, but an amendment to current legislation will require any midwife attending homebirth to be licensed beginning in 2019 (MI Public Health Code, Act 368 of 1978). Although the legal guidelines vary state to state, this specific example suggests that the responsibility is on the midwife attending an out-of-hospital birth to verify that the hearing screening is completed.

Although midwives have a responsibility to provide information to their clients about newborn hearing screening, a survey of 518 practicing midwives showed that 92.9% reported having a lack of knowledge to guide families through the newborn hearing screening process (Goedert, Moeller, & White, 2011). Many midwifery education programs report including some information about newborn hearing screening as part of their curriculum, but this may not be sufficient for midwives to take an active role in a newborn hearing screening program (Palmer, Bednarz, Dilaj, & MacDonald, 2016). The purpose of this study is to determine if a training program, along with providing equipment, improved hearing screening rates for babies born in out-of-hospital

settings. This included an analysis of newborn hearing screening data after implementation of this training program to see if babies born in an out-of-hospital setting were more likely to receive a newborn hearing screening based on their midwife’s participation in the training program and her access to an Automated Auditory Brainstem Response (AABR) screening machine.

Method

Training

In 2014, an initiative spearheaded by the Michigan Coalition for Deaf and Hard of Hearing People, a 501(c)3 organization, in partnership with the Michigan EDHI program and Central Michigan University provided hands-on training and distributed 15 AABR machines to midwives who attend homebirths. This effort was supported by a grant from the Carls Foundation, who only funds 501(c)3 agencies. All midwives in the state of Michigan were invited to participate in a training session. Invitations to participate were distributed through the Michigan Midwives Association, who supported this effort, and direct contact with midwives across the state. In order to participate in the hands-on training and receive access to an AABR machine, the midwives were required to first complete an online educational training. The online training was created by the Michigan EDHI program to train all healthcare professionals who will be doing newborn hearing screening. It consisted of ten modules covering topics such as auditory anatomy, hearing screening methods, risk factors for hearing loss, communicating and reporting screening results, the hearing screening process, and a final assessment. This is the same online training completed by hospital staff. Each participant completed the online training and passed the final assessment with a score of 80% or better prior to attending a hands-on training session.

Hands-on training sessions were conducted in five different locations around Michigan over a four-month period in early 2014. The hands-on training sessions were conducted by a MI EHDI program consultant, a pediatric diagnostic audiologist, an audiology graduate student, and a representative of the equipment distribution company. The equipment representative provided step-by-step instruction and practice using the AABR equipment. The audiologist then led a discussion of the importance of hearing screening, how to communicate screening results to parents, and the process for follow-up after a baby refers on the screening. Challenges specific to homebirth families were addressed. The MI EHDI program consultant then reviewed the Coalition Agreement for using the equipment, the process and paperwork for reporting screening results, and diagnostic sites where families could be referred if additional testing was needed. Finally, each midwife completed a hearing screening using the Baby ISAO (Intelligent Hearing Systems) hearing loss simulator. The training sessions were about 2–3 hours each.

Midwives who participated in this training were either given an AABR machine to host in their practice or provided access to borrow a machine from a host location. The Coalition purchased an additional AABR machine, for a total of 16 and provided an additional hands-on training session in mid-2016. This was a refresher course for most participants who had extremely limited access to a machine, and recruited two new midwives into the program, with one hosting the new machine. The Coalition maintains ownership of the machines, purchases supplies, and arranges calibration and insurance for the equipment. The Coalition also works closely with EHDI and their data to determine best placement of machines on an annual basis. After the second year of the grant (Fall of 2015), midwives were assessed a minimal per baby screening fee, payable to the Coalition, to be able to continue to purchase and ship supplies, as well as provide calibration and insurance on the machines.

Participants

Data for this study were obtained from the state-wide hearing screening data reported to the EHDI program. Data included all midwives from the state of Michigan who reported attending a homebirth in the 2015 and 2016 calendar years and who did a metabolic blood spot screen ($N = 112$). Midwives belonged to four groups including those who hosted a machine for EDHI screening (*host midwives*; $n = 15$; 13.39%), those who had access to a machine (*access midwives*; $n = 25$; 22.32%), those who received educational resources through the free online training provided by EHDI but did not complete the hands-on training and therefore did not have access to a machine (*education midwives*; $n = 4$; 3.57%), and those who did not receive access to screening machines or to educational resources (*non-participants*; $n = 68$; 60.71%). There were no missing data.

Although all midwives in the state were encouraged to participate in the training program, midwives self-selected whether they were interested in the training or not. Any midwife who completed both the online and hands-on training were included as access midwives (excluding those chosen as host midwives). Host midwives were chosen based on geographic location and birth volume to have a distribution across that state that met the needs of the region. Midwives who submitted a metabolic bloodspot screen but did not participate in any part of the training program were included in the non-participant group.

Data Analysis

The purpose of this study was to determine the odds of an infant undergoing hearing screening based on a midwife's access to and experience with AABR screening machines, as well as the total number of homebirths the midwife has attended. Because infants who were delivered by the same midwife do not have independent outcomes from one another (i.e., infants are "clustered" or "nested" within midwives), a two-level multilevel model was used to account for the non-independence of observations (McCoach & Adelson, 2010) and to use a midwife-level

variable (treatment group) to explain variability in our outcome (hearing screening status; McCoach, 2010). The outcome of interest was an indicator of whether or not the infant had been screened (SCREENED; 0 = no, 1 = yes). The level-one, or infant-level, model controlled for YEAR the baby was born (0 = 2015, 1 = 2016). The level-two, or midwife-level, variable of interest was their treatment status, represented by three dummy-coded group variables (HOST, ACCESS, and EDUCATE, with NONE as the reference group) At this level we controlled for the total number of births the midwife attended in 2015 and 2016 combined (TOTBIRTH), which we grand-mean centered so that it would have a meaningful 0 (Enders & Tofighi, 2007).

Given that our outcome (whether an infant was screened) was binary, we specified our model using a Bernoulli distribution, a binomial level-1 sampling model that provides the probability or odds of the desired outcome. Full maximum likelihood (FIML) and EM Laplace iterations were used to produce population-average models. Compared to unit-specific models, "population average models generally will be more useful when the desired inferences focus on the group-level variables, rather than the varying effects of individual level covariates" (O'Connell & McCoach, 2008, p. 218). Additionally, with the population model, random effects are not held constant (O'Connell & McCoach, 2008).

We used a model-building approach, as recommended by Raudenbush and Bryk (2002). First, we used the HLM 7.03 software to estimate an unconditional model with SCREENED as the outcome variable to estimate the average probability that an infant was screened for hearing loss: $\exp(-0.65) / 1 + \exp(-0.65) = 0.52 / (1 + 0.52) = 0.34$. Next, we added the level-one control variable, YEAR, to determine if its slope should be allowed to randomly vary in subsequent models. Although the differential for 2015 and 2016 was not statistically significant ($p = .055$), the slope ($\gamma_{10} = 0.04$) did statistically significantly vary between midwives ($\tau_{11} = 0.13$, $\chi^2_{(91)} = 176.54$, $p < .001$). Based on model fit comparisons ($\chi^2\Delta_{(2)} = 12.59$, $p = 0.002$; $AIC\Delta = 8.58$; $BIC(n)\Delta = -3.13$; $BIC(j)\Delta = 3.15$), we chose to allow the slope to randomly vary and to retain the variable as a covariate in the model. This indicates that although the probability of being screened did not differ on average based on the year of birth, that differential varied across midwives; in other words, babies were more likely to be screened in 2015 for some midwives, more likely to be screened in 2016 for other midwives, and yet for other midwives there was no difference. Next, we added the level-two control variable, TOTBIRTH, as a predictor of the intercept. Although the total number of births a midwife attended did not predict whether an infant was screened ($\gamma_{01} = -0.0001$, $p = .99$), because our model is relatively simple and we identified this as a potential covariate a priori, we opted to leave it in the model. Finally, we added the three dummy-coded group variables of interest, HOST, ACCESS, and EDUCATE, to the intercept. This resulted in our final model:

$$SCREENED_{ij} = \gamma_{00} + \gamma_{01} * TOTBIRTH_{j} + \gamma_{02} * HOST_{j} + \gamma_{03} * ACCESS_{j} + \gamma_{04} * EDUCATE_{j} + \gamma_{10} * YEAR_{ij} + u_{0j} + u_{1j} * YEAR_{ij}$$

where the outcome is whether infant *i* whose birth was attended by midwife *j* was screened and γ_{02} , γ_{03} , and γ_{04} represent the differential in the log-odds of being screened when the attending midwife had hosted a machine, had access to a machine, or were provided with educational resources, respectively, compared to midwives who did not participate in the project at all, after controlling for the year of birth and the total number of births the midwife attended.

Results

For each group of midwives, we examined the number of births and the number of infants who were screened for hearing loss in 2015 and 2016. (The average number of births/infants screened per midwife for each group

is provided in parentheses throughout the current paragraph.) The total number of births (2015–2016) for host midwives was 571 ($M = 38.07$, $SD = 29.64$) with 453 infants screened (79.33%; $M = 30.20$, $SD = 23.82$). Access midwives attended 513 births ($M = 20.52$, $SD = 13.66$) and screened 243 infants (47.37%; $M = 9.72$, $SD = 7.57$). Education midwives assisted with 140 births ($M = 35.00$, $SD = 12.46$) and screened 83 infants (59.29%; $M = 20.75$, $SD = 7.14$). Finally, our largest group, non-participants, assisted with 1,356 births ($M = 19.94$, $SD = 37.03$) and screened 87 infants (6.42%; $M = 1.28$, $SD = 2.53$). The average number of births, infants screened, and percentage of infants screened for each midwife group are provided in Table 1. In comparison with data from the MI EHD database from 2013, prior to the implementation of the training program, the proportion of babies screened increased in all groups except the non-participant group. In 2013, only 14.2% of babies born at home received a hearing screening.

Table 2 reports the results for the final model. Total births

Table 1

Average Number of Births, Infants Screened, and Percentage of Infants Screened Per Midwife for Each Midwife Group 2015–2016

	Average Number of Births		Average Number of Infants Screened		Average Percentage of Infants Screened
	<i>N</i>	Mean (<i>SD</i>)	<i>N</i>	Mean (<i>SD</i>)	Mean (<i>SD</i>)
Host Midwives	571	38.07 (29.64)	453	30.20 (23.82)	82.08 (18.39)
Access Midwives	513	20.52 (13.66)	243	9.72 (7.57)	51.56 (29.68)
Education Midwives	140	35.00 (12.46)	83	20.75 (7.14)	59.86 (7.95)
Non-Participants	1,356	19.94 (37.03)	87	1.28 (2.53)	14.28 (27.27)

Note. Host Midwives: $n = 15$; Access Midwives: $n = 25$; Education Midwives: $n = 4$; Non-Participants: $n = 68$.

($\gamma_{01} = -0.01$; $p = .04$), host ($\gamma_{02} = 3.67$; $p < .001$), access ($\gamma_{03} = 2.15$; $p < .001$), and educate ($\gamma_{04} = 2.38$; $p < .001$) were statistically significant predictors of being screened. The intercept, $\gamma_{00} = -2.14$ ($p < .001$), represents the expected log odds of an infant being screened for hearing loss in 2015 when the midwife did not participate in the hearing screening project, after controlling for number of births she attended. Thus, the estimated odds (or *referent odds*) of being screened for a child with these characteristics is 0.12. Total Births had a negative effect on the log-odds of infant screening ($\gamma_{01} = -0.01$; $p = .04$) when controlling for midwife group and year. The odds of being screened is expected to be lowered by 0.99 as total births increases by one (holding other variables constant). There was not a statistically significant difference in the log-odds of an infant being screened when born in 2015 or 2016 ($\gamma_{10} = 0.22$; $p = .12$).

Having a midwife who hosted a machine for AABR screening had a positive effect on the log-odds of infant screening ($\gamma_{02} = 3.67$; $p < .001$) when controlling for total

births, midwife group, and year. The odds of an infant with a midwife hosting a machine being screened was 39.37 times greater compared to an infant with a midwife in the non-participant group (holding other variables constant). Having a midwife who had access to an AABR machine had a positive effect on the log-odds of infant screening ($\gamma_{03} = 2.15$; $p < .001$) when controlling for total births, midwife group, and year. For infants with midwives in this group, the odds of being screened was 8.57 times greater compared to infants with a midwife in the non-participant group (holding other variables constant). Finally, having a midwife who was provided with educational resources had a positive effect on the log-odds of infant screening ($\gamma_{04} = 2.38$; $p < .001$) when controlling for total births, midwife group, and year. For infants with these midwives, the odds of being screened was 10.82 times greater compared to an infant with a midwife in the non-participant group (holding other variables constant).

Table 2
Fixed Effects from the Final Model of Infant Screening

Fixed Effect	Coefficient	SE	t(df)	p
Model for SCREENED outcome (β_0)				
Intercept (γ_{00})	-2.14	0.22	-9.78 (107)	<.001
Total Births (γ_{01})	-0.01	0.004	-2.10 (107)	.04
Host (γ_{02})	3.67	0.39	9.37 (107)	<.001
Access (γ_{03})	2.15	0.33	6.46 (107)	<.001
Educate (γ_{04})	2.38	0.65	3.68 (107)	<.001
Model for YEAR slope (β_1)				
Intercept (γ_{10})	0.22	0.14	1.56 9111)	.12

Fixed Effect	Coefficient	Odds Ratio	Confidence Interval
Model for SCREENED outcome (β_0)			
Intercept (γ_{00})	-2.14	0.12	(0.08, 0.18)
Total Births (γ_{01})	-0.01	0.99	(0.98, 1.00)
Host (γ_{02})	3.67	39.37	(18.10, 85.64)
Access (γ_{03})	2.15	8.57	(4.34, 16.58)
Educate (γ_{04})	2.38	10.82	(3.00, 39.06)
Model for YEAR slope (β_1)			
Intercept (γ_{10})	0.22	1.25	(0.94, 1.65)

Discussion

The likelihood that an infant would receive a universal newborn hearing screening differed significantly depending on midwives' access to AABR machines and the educational resources that they were provided during their initial trainings. Providing midwives with training and access to newborn hearing screening equipment had a positive effect on the number of babies who received a hearing screening. However, the likelihood that an infant would be screened decreased as the total number of births the midwife attended increased. These results support the need for continued national efforts to include midwives in the universal newborn hearing screening process.

Many practicing midwives do not think that participating in newborn hearing screening is part of their job or feel unprepared to participate in a newborn hearing screening program (Goedert et al., 2011). However, during their care for infants, midwives are expected to develop a plan for care, which includes national and local screening guidelines (ACNM, 2012). This includes newborn hearing screening. By training midwives and providing them access to newborn hearing screening equipment, the rate of newborn screenings increased. Although the number of midwives receiving education only was small ($n = 4$), there was an increase in the odds of screening even for those midwives who only received focused education about the importance and process of newborn hearing screening. This suggests that even if implementing a full screening program for midwives is not financially or logistically feasible, increasing educational outreach to midwives and identifying local community locations where they can refer their families to have the baby's hearing

screened can have a significant positive effect on newborn hearing screening rates. Further research on this as an intervention needs to be conducted.

To date, this is the first study to present outcome data from a program to train midwives to conduct newborn hearing screenings. In a study of the implementation of universal newborn screening in the state of Wisconsin, Kerschner et al. (2004) mentioned that a group of midwives purchased hearing screening equipment and provided screening services for their homebirth clients. Although the midwives who participated had 79% screening rate, there were only three groups of midwives who participated in this program as of 2002 and the efforts were focused on a small geographical region of the state (Kerschner et al., 2004). Although there may be some initial resistance, from either midwives or state agencies, to training midwives, both the midwives in Wisconsin (Kerschner et al., 2004) and the midwives in Michigan who participated in these programs have been supportive of these efforts.

Two populations that traditionally choose homebirth and often are served by midwives are the Amish and Mennonite communities. With the increased likelihood of genetic and congenital conditions in these closed communities, effective newborn screening is extremely important (Morton et al., 2008). In a study of opinions about newborn screening in Amish and Mennonite communities in Wisconsin, Sieren et al. (2016) found that most families reported a positive view of newborn screening but cited lack of knowledge at the time or lack of access as reasons for not having their children screened. Sieren et al.'s (2016) questions focused on the newborn screening program as a whole, not specifically newborn

hearing screening. However, the newborn hearing screening is considered a standard part of the newborn screening process. These data suggest that if midwives serving these communities are able to offer newborn hearing screening as part of their services, the Amish and Mennonite communities would be amenable to increasing their screening rate.

Limitations of this study include the timeframe of data collection, self-report nature of the hearing screening data, and difference in group sizes. Screening rates for this study were only analyzed for the first two years following implementation of this program. Continued training and support may result in further change in screening rates. Therefore, additional analysis over a longer timeframe would be beneficial.

The data for this study was taken from the MI EHDI database for all reported hearing screenings. However, it is possible that there are practicing midwives who chose to not report any screening data or were unable to be tracked with the Michigan data system. This information is not included in this analysis. At the time of this study, Michigan used Perkin & Elmer software to track hearing screenings and they can only be tracked if the baby also has a metabolic blood card screening as well. Midwives who performed hearing screenings, but not the metabolic screenings are not included in this study.

Looking at the size of each subject group, there was a much smaller number for midwives in the education only group ($n = 4$) compared to the other groups. The midwives in this group completed the online training modules but did not attend a hands-on training session. Most of the midwives of the education-only group were recent transplants to the state and learned about the program immediately after all the hands-on training took place. Those midwives worked with the EHDI program consultant to take the on-line training and identify local community resources to direct their families. One of these midwives was from an Amish community. Even with such a small group there was a significant difference between the screening rates of babies born to midwives in this group compared to the non-participant group. Having seen an effect with such a small group could indicate the importance of additional education for midwives.

Distribution of the equipment was a limiting factor for this program. There were certain areas of the state that had higher homebirth rates than other areas, requiring an uneven distribution of the AABR equipment to account for the busier midwifery practices in those areas. Requiring midwives to share equipment was often challenging because several practices may have had conflicting schedules or needs. This necessitated a re-evaluation of the host sites and locations of the equipment annually. Continual monitoring of the birth and screening rates in different regions of the state have been vital to the maintenance of the program. Recognizing that homebirth attendants have a powerful

influence and provide guidance among parents who choose homebirth, it is important for EHDI programs to include this population when considering outreach programs. For programs considering embarking on a similar project, it is important to consider multiple training dates due to the nature of the work of midwives to be on call to deliver babies. In every training session, there was at least one and up to four fewer midwives attending than signed up, due to their unpredictable schedules. Offering multiple trainings in different locations ensures midwives had a chance to attend a later training if circumstances prevent them from attending a training session.

In Michigan EHDI's own homebirth analysis, covering the years 2014–2016, rates of babies identified with hearing loss within this population was statistically larger than expected, which was a revelation. The potential of early identification of babies who are deaf or hard of hearing and ensuring timely intervention services is the ultimate goal of all EHDI programs. Without this program, these babies were unlikely to be diagnosed until they were much older.

Conclusion

Providing midwives with training and education about newborn hearing screening as well as access to equipment increases the odds of a baby receiving a newborn hearing screening. Although midwives who had constant access to screening equipment had the highest odds of screening babies, providing access to equipment, even if not constant, and providing additional education and community resources, but not access to equipment also had a positive effect on the odds of babies being screened. The logistics of completing the trainings, distributing equipment across the state, maintaining equipment, and obtaining insurance for equipment are complicated; however, the outcomes have demonstrated the success of this type of program. Indeed, the results of this study, feedback from the midwives and the EHDI analysis has spurred The Coalition to seek additional funds and extend the partnerships to expand this project to increase the number of AABR machines available for Michigan midwives to be able to offer hearing screenings for their families.

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Family Impact of Pediatric Hearing Loss: Findings from Parent Interviews and a Parent Support Group

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Abstract: Parents experience numerous stressors tied to their child's diagnosis as deaf or hard of hearing (DHH). This study sought to inquire about the lived experiences of parents with children who are DHH to determine the types of supports that should be provided within an audiology care coordination system. Semi-structured phone interviews were conducted with parents of children under the age of five who are DHH and patients of the Division of Audiology at Cincinnati Children's Hospital Medical Center (CCHMC). Interview findings determined focus group questions, which were facilitated in a parent support group with parents of children who are DHH and seen by the division. The results revealed parents' reactions and adaptations to their child's hearing health needs, as well as helpful supports and services. An audiology care coordinator (ACC) and a local parent support group were two of four supports identified as helpful in navigating their child's hearing health care. The findings of this study lend context for the types of support services pediatric institutions can provide to help families when their child is identified as DHH through a care coordination approach.

Key Words: deaf or hard of hearing, care coordination, family support, parent needs, support services

Acronyms: ABR = Auditory Brainstem Response; ACC = audiology care coordinator; BCMH = Bureau of Children with Medical Handicaps; BTE = behind the ear; CCHMC = Cincinnati Children's Hospital Medical Center; CI = cochlear implant; CMV = Cytomegalovirus; DHH = deaf or hard of hearing; FAC = Family Advisory Council; FL3 = Family Leadership in Language and Learning; HL = hearing loss; LSL = listening and spoken language; NICU = neonatal intensive care unit

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Hearing loss is one of the most common congenital birth defects, yet nearly all parents with children who are deaf or hard of hearing (DHH) have typical hearing and no experience with the implications of hearing loss (Centers for Disease Control and Prevention, 2010; Jackson & Turnbull, 2004; Mitchell & Karchmer, 2004). This lack of experience may explain initial feelings of shock and unpreparedness in parents that are cited throughout the literature (Kurtzer-White & Luterman, 2003; Jackson & Turnbull, 2004; Yoshinaga-Itano & Abdala de Uzcatogui, 2001; Young & Tattersall, 2007). Parents with children who are DHH also

experience greater levels of stress around communicating with their child, making decisions about education for their child, selecting hearing devices, and maintaining hearing devices (Dammeyer, Hansen, Crowe, & Marschark, 2019; Dirks, Uilenburg, & Reiffe, 2016; Fitzpatrick et al., 2015; Hintermair, 2000; Lederberg & Golbach, 2002; Quittner, 1991; Quittner et al., 2010; Quittner, Glueckauf, & Jackson, 1990; Ward, Hunting, & Behl, 2018). Since it has been documented that parents' psychological well-being is paramount to cognitive and social-emotional outcomes in children (Calderon, 2000; Hintermair, 2006), it is crucial

to understand the types of support services needed by parents with children who are DHH.

In the literature, parents have noted a desire for more information about their child's hearing loss and social-emotional and cognitive development (Fitzpatrick, Angus, Durieux-Smith, Graham, & Coyle, 2008; Henderson, Johnson, & Moodie, 2014; Jackson, 2011; Jamieson, Zaidman-Zait, & Poon, 2011; Yucel, Derim, & Celik, 2008). In addition, more dissemination of educational, childcare, community, and financial resources is needed (Jackson, 2011; Jamieson et al., 2011; Yucel et al., 2008), as well as comprehensive information about services and support at different points in their child's life (Ward et al., 2018). Further, information about navigating the health care system and building competence in caring for children with hearing loss are parent needs noted in the literature (Henderson et al., 2014). Parents have also requested resources for the well-being of the entire family (Henderson et al., 2014). In a comprehensive literature review, Jackson & Turnbull (2004) found that deafness can have various adverse impacts on the family unit. Family interactions, family resources, parenting, and support services are all domains of family life that are impacted by having a child who is DHH. Fortunately, family involvement in early intervention has been found to promote successful outcomes by the age of five (Moeller, 2000), perhaps because parents can vocalize their needs and gain access to supportive resources.

A quality improvement survey administered by the Division of Audiology at Cincinnati Children's Hospital Medical Center (CCHMC) revealed that families rely heavily on support from the in-house audiology care coordinator (ACC). Audiology care coordination is a relatively new strategy employed by pediatric institutions to provide comprehensive, coordinated care, yet to our knowledge, there is no published research on care coordination in audiology practice. In primary care, care coordination is considered an approach to care that meets patients' needs and enhances the capabilities of care-takers (Council on Children with Disabilities and Medical Home Implementation Project Advisory Committee, 2014). Care plans are determined by family needs, roles, responsibilities, and desired outcomes (Antonelli, McAllister, & Popp, 2009; National Quality Forum, 2010). In the literature, care coordination has been defined as "the deliberate organization of patient care activities between two or more participants involved in a patient's care to facilitate the appropriate delivery of health care services" (McDonald et al., 2007, p. v). Wagner, Gupta, & Coleman (2014) identified the goals and common features of successful programs that use care coordination. Successful care coordination exhibits accountability of the organization in coordinating their patients' care, clear and shared understanding of roles and responsibilities of all parties, support when patients go elsewhere for care, and timely transfer of relevant and understandable information. These findings are extended by Van Houdt, Heyrman, Vanhaecht, Sermeus, & De Lepeleire (2013) who found that clarity of

roles and responsibilities, quality of relationships, mutual respect and collaboration, and information exchange between health care providers and families are key characteristics of care coordination.

In local practice, care is coordinated primarily by the ACC but also in concert with audiologists and staff. Some of the services that are coordinated by our ACC and providers are:

- Providing telehealth services for patients, especially for those who live out-of-state.
- Disseminating a newsletter to inform parents about hearing health and resources.
- Sharing a Facebook group for parents of children with hearing loss.
- Collaborating with an Audiology Family Advisory Council (FAC) to facilitate hearing health care by clarifying and communicating needed areas of support, developing contextualized care plans, and identifying and disseminating resources for families with children who are DHH in ways that are family-accessible and content appropriate.
- Providing binders with written information for all families, including funding/financial resources, helpful websites for learning about hearing loss, contact persons, information about hearing devices, early intervention, and more.

The ACC serves as the primary point of contact for families and is responsible for providing and informing families of all these resources. Practically, the role of the ACC involves acting as a primary messenger of information and source of support. Patients receive a one-on-one experience with the ACC through regular check-ins and correspondence. This ensures that even families who are too overwhelmed to seek advice on their own receive social support. If families come to the ACC with questions, the coordinator is responsible for responding to families in an accurate and timely fashion. Other responsibilities of the care coordinator include connecting families to other specialists and medical staff, sharing written information regarding all sources of support (e.g., funding/financial resources, support groups with other families with children or parents who are DHH, information about hearing devices, and early intervention), and organizing all hearing-related appointments in an efficient manner, especially for traveling and out-of-state patients. The ACC also connects traveling and out-of-state families with resources for support near their hometown.

In the Family Leadership in Language and Learning (FL3) Needs Assessment report, parents indicated that they would benefit from coordinated, trusted resources; contact with and support from other parents who share their lived experiences; access to role models who are DHH; invitations to participate in parent activities; appointment reminders; and connections to early intervention (Ward et al., 2018)—all of which are resources and services provided by our in-house ACC. When asked where they receive these supports, parents responding to the

FL3 Needs Assessment indicated top providers were audiologists, early intervention staff, family support organizations, physicians, and website or social media. However, when asked what would make accessing these supports easier, the most common response was to have one contact such as a family support coordinator. These findings highlight the utility and necessity of an ACC in pediatric practice, as well as further research to define the role and evaluate outcomes of having an ACC.

Due to positive feedback on audiology care coordination at CCHMC and in the literature, this follow-up study takes a deeper look at the needs of families with children who are DHH to maximize support services provided by the care coordinator. We contribute to the literature on audiology care coordination by exploring the impact of having a child identified with hearing loss including sources of support that have facilitated their experience. Our hope is that inquiring about a wide spectrum of experiences, practical and emotional, will provide a broader, more holistic view of the experiences met by families with children who are DHH. Therefore, the purpose of this study is to (a) explore the experiences of parents with children who are DHH, (b) uncover helpful existing and needed support services for families of children who are DHH, and (c) make recommendations for coordinating these supports in pediatric institutions.

Method

Participants

Participants of this study included 13 mothers and one father of children who are DHH under the age of five ($N = 14$). In the first phase of data collection, purposive sampling was employed to identify parents of children under five years old who are DHH and received hearing health care from the Division of Audiology at CCHMC. These parents were selected based on their ability to provide information-rich cases about their experiences. Of the ten parents invited for a phone interview, eight agreed to participate (see Table 1). The eight interview participants were mothers ranging from 20–40 years old and the majority ($n = 7$) identified as Caucasian while one identified as Hispanic and Native American. Three mothers were high school graduates, two held college degrees, and three held graduate (master's) degrees. As a note of interest, two mothers worked in the education field (art teacher and special education teacher) while three held positions in healthcare (RN manager, nurse practitioner, and research administration). Two others were employed by the service industry (clerk and server) and one mother identified as a stay-at-home mom. In the second phase of data collection, an additional five mothers and one father recruited from a parent support group participated in a follow-up focus group to determine and refine the interview themes. All focus group parents had children under the age of five who were DHH and were patients of the Division of Audiology at CCHMC.

Data Collection and Analysis

This study has been granted a Non-Human Subjects Determination by the Cincinnati Children's Hospital Medical Center Institutional Review Board for research conducted by the Division of Audiology with parents of patients as part of an evaluation of the division. Consent for participation in tape-recorded interviews and focus groups was obtained prior to each interview or focus group session.

Semi-structured interviews. Individual phone interviews were conducted using a semi-structured interview guide. The questions related to the overall experience of being the parent of a child who is DHH, barriers and challenges, and helpful resources that assist or would assist in managing their child's hearing impairment. All interviews were conducted by the same interviewer, who is a researcher with a background in community-based and participatory approaches to health research and several years of experience conducting qualitative health research. The interviewer was contracted from a division outside of Audiology (Division of Research at CCHMC), to limit bias and encourage candidness from participants. All interviews were audio-taped and transcribed verbatim by the interviewer directly after each interview.

Interview data was analyzed by the primary contracted researcher using thematic analysis as described by Braun & Clarke (2006). In the first phase, the audio-taped interviews were transcribed and read twice with initial ideas written as notes. Using this initial list of ideas about the data, phase 2 involved the construction of initial codes that appeared important or meaningful to the experience of having a child who is DHH. The literature review assisted in identifying points of interest in the data. Phase 3 involved sorting these initial codes into themes and collating all of the relevant codes within the identified themes. In phase 4, overarching themes were eliminated if there was not enough data to support them, or collapsed if two separate themes related to one another. Other themes were broken down into separate themes as necessary. In phase 5, themes were defined, refined, and given a title by identifying and capturing the essence of each theme's meaning. Two members of the research team (both audiologists, one of which was the division care coordinator) and an expert in parent needs for children who are DHH reviewed the themes independently to enhance the credibility of the study findings. The entire research team discussed their independent reviews and worked together through democratic discussion to establish a final codebook representative of the interview themes.

Focus Group. The interview findings guided the design of a focus group guide which inquired about concepts emerging from the interview data. The contracted interviewer facilitated the focus group, which centered on questions related to thoughts and feelings associated with their child's hearing loss, the family impact of the hearing loss, barriers and challenges regarding their child's hearing

Table 1**Family Impact of Pediatric Hearing Loss: Parent and Child Demographics at Time of Interview**

Age of Patient	Parent Hearing Status	Gender of Parent	Gender of Child	Child's Hearing Loss	Child's Characteristics	Device Use
24 months	Typical hearing	F	F	Fluctuating slight-mild bilateral	<i>Fit at age of 27 months*</i> – Down syndrome; chronic otitis media with effusion; multiple sets of pressure equalizing tubes; speech and language delay.	Bone conduction device; no longer wearing.
11 months	Typical hearing	F	F	Left Unilateral moderate to severe, sensorineural	<i>Fit at 7 months</i> – Typical speech and language development per parent report; recommended speech evaluation to confirm.	Monaural Behind the Ear (BTE) aids; no longer wearing.
30 months	Typical hearing	F	F	Moderate to severe U-shaped bilateral, sensorineural	<i>Late Identification – Fit at 20 months</i> – Delayed receptive and expressive language; enrolled at area school for Listening and Spoken Language (LSL); biological sibling (below) has hearing Loss (HL).	Bilateral BTE aids; wears consistently.
14 months	Typical hearing	F	M	Moderate bilateral, sensorineural	<i>Fit at 4 months</i> – Receptive language within typical limits; expressive language mildly delayed; biological sibling (above) has HL.	Bilateral BTE aides; wears consistently.
10 months	Typical hearing	F	M	Profound bilateral	<i>Implanted at 14 months</i> – Receptive and expressive speech delays; enrolled in area LSL program.	Bilateral Cochlear Implant (CI); wears consistently.
25 months	Typical hearing	F	M	Typical hearing	Initially diagnosed in another state with bilateral hearing loss. Determined to have typical hearing at CCHMC – discharged from speech therapy.	None.
13 months	Typical hearing	F	F	Moderate rising to mild bilateral mixed	<i>Fit at 7 months</i> – Cleft Palate; Stickler Syndrome; delayed speech and language.	Bilateral BTE aids; wears inconsistently.
17 months	Typical hearing	F	M	Profound bilateral	<i>Implanted at 12 months</i> – Cytomegalovirus (CMV); delays in speech and language; enrolled in area LSL program.	Bilateral CI; wears consistently
21 months	Typical hearing	F	F	Moderate to severe, conductive	<i>Fit at 3 months</i> – Bilateral ear canal atresia/stenosis; global developmental delays.	Bilateral BTE aids; wears consistently.

*Trial with a bone conduction device (BCD) due to chronic fluctuations with conductive hearing loss— parent opted to try management with a BCD at 27 months to avoid additional surgeries.

loss, and support services that have been helpful or would be helpful in managing their child's hearing impairment. The focus group discussion was audio-taped and transcribed verbatim by the facilitator shortly after the focus group session concluded. The focus group data was thematically coded by the facilitator using thematic analysis (Braun & Clarke, 2006). In this process, findings were triangulated with the themes from the interview data to further refine the overarching themes and enhance the credibility of the thematic categories. A final codebook was reviewed by the two previously mentioned members of the research team who are experts in parent needs for children who are DHH. Phase 6 of thematic analysis continues in this article as we use our thematic map to tell a story about the burden costs of parenting a child with hearing loss.

Results

Six major themes emerged from the interview and focus group data, falling into three overarching concepts: Reactions and Adaptation to Hearing Loss, Barriers and Challenges, and Supports (see Table 2). Each subtheme within the categorical themes represents individual stressors or strains on the parent that impact their lived experience, and existing or needed support services. This section elaborates on each theme in relation to their subthemes.

Reactions and Adaptation to Hearing Loss

Many parents reported feeling shocked when first learning that their child was identified as DHH, primarily because they held no previous knowledge about hearing loss and

Table 2

Family Impact of Pediatric Hearing Loss: Summary of Interviews and Focus Group Findings

Overarching Concept	Theme	Subtheme	Description	n	Quote as Evidence of Subtheme
Reactions and Adaptation to Hearing Loss	Reactions and Adaptation to Hearing Loss	It Gets Easier	Parent's description of adapting to child's hearing loss—it is difficult at first, but it gets easier.	7	<i>"It's been difficult at times but with the help of my doctors that we have for him, and his therapist and everybody that has helped us through it, it has been a bit easier for us to get through it emotionally and physically."</i>
		Shocking	Specific term used to describe first learning about child's hearing loss and not having previous knowledge about hearing loss, nor knowing anyone who has undergone this experience.	6	<i>"I have two other kids, they're older than my daughter and they have normal hearing. For me, it was a complete shock because neither side has hearing loss."</i>
		Concerns about Child Development	Concern about child's cognitive, physical, and social-emotional development compared to typical hearing children; not wanting child to be bullied or live a difficult life.	7	<i>"I'm afraid of what his life is going to be like. Are people going to pick on him, because not only is he going to have this hearing aid on, he's going to have these little ears? That is what I worry about."</i>
		Social Implications	Parents' desire to expose child to others with hearing impairment, encouraging others to ask about condition instead of judging.	6	<p><u>Quote 1:</u> <i>"I think it's beneficial for her to meet other people, adults or kids, who have the same sort of thing just because she does have two older brothers who don't have the same situation and she will be mainstreamed in school, so she probably won't be around other kids, outside of our friends and seeing them in the audiology department."</i></p> <p><u>Quote 2:</u> <i>"To be honest if I see people looking at them and not asking, I get angry. I want to be like 'Just ask what is up with his ears, because it's rude to stare.'"</i></p>
Barriers and Challenges	Use of Hearing Devices	Financial Coverage	Difficult in obtaining insurance or financial coverage for hearing devices.	6	<i>"That was a shock that insurance didn't cover the ABRs and regular audiology appointments. We had spent several hundreds of dollars before being approved for BCMH and even then still waiting to get reimbursed for some of the costs. I can't believe most insurance companies don't cover hearing, especially in children, yet they cover things like Viagra. So frustrating especially for those of us on very tight budgets."</i>
		Hearing Aid Retention	Difficulty keeping hearing aids on children.	10	<i>"When he first got the CIs, he was a stinker and would just throw them off all the time. He still does that."</i>
		Ear Molds	Excessive time for the remaking of child's ear molds.	5	<i>"We just wish ear molds could be made quickly and on-site so we could get them right away, that would be awesome."</i>
		Managing Devices	Stress associated with managing child's hearing devices.	6	<i>"When we first got them, they would monitor how long he had them on and he would average an hour a day. I would get so frustrated, I'm like 'He needs at least seven to eight hours for a full day, even with naps and stuff like that.' It's getting people to understand they need to stay on even if he goes to bed, that kind of stuff."</i>
	Scheduling	Making Appointments	Not enough time in parent schedule for appointments.	8	<i>"I think it was those kinds of stresses and impact when you're thinking about appointments and who can make the appointments."</i>
		Number of Appointments	Too many appointments in the first year for all service providers related to hearing.	8	<i>"You have to go see a pediatrician, you have to go talk to a geneticist. We did all of our appointments in one day, like eight appointments in one day, trying to get social worker, oral rehab, all that stuff."</i>
	Financial Costs	Overall Cost of Services	Overall cost was considered burdensome.	9	<i>"I was terrified... 'What are we going to do?' We had family members—both sets of parents offered to loan us money, but not everyone has family support where they would just be able to get that money."</i>
		Hearing Devices	Cost of hearing devices was considered burdensome.	6	<i>"My insurance—and I have the insurance for our whole family—said that for his hearing aid, they would cover 100% or a maximum of \$3,000. Well, he has Bahas and has two of them. They are approximately \$10,000."</i>

Table 2 cont.

Family Impact of Pediatric Hearing Loss: Summary of Interviews and Focus Group Findings

Supports	Education /Resources	Support Groups	Sought support from other groups of parents after learning about child’s hearing loss.	6	<i>“There’s huge groups out there if I want to talk to people in other countries or across the country—but to have local parents, seeing the same departments, the same doctors, possibly the same schools, that’s huge.”</i>
		Internet Research	Parents sought to teach themselves about child’s hearing loss through internet research.	7	<i>“I was googling the minute after the NICU staff left the bedside.”</i>
	Healthcare Team	Audiology Care Coordinator	Considered a helpful asset of medical team due to timely communication, having questions answered, connecting to other specialists, organizing appointments in an efficient manner for patients, and overall support.	12	<i>“She actually came when [child] had surgery, she showed up at Children’s downtown. We weren’t expecting her. she stayed for two, three hours and talked to all of us. She just goes above and beyond. If I have a question or concern and she doesn’t know the answer, she’ll talk to whoever she needs to and gets right back to me. She just always takes the time to research and find the correct answer.”</i>
		Audiologists, Doctors, Specialists, Nurses, and Staff	Considered helpful facilitators or care due to timely communication, having questions answered and quality of care.	12	<i>“The one-on-one experience with them, you don’t get that anywhere else. They check on you and make sure you’re doing okay. [Child] has been chewing through his ears [hearing aids], so I called [provider] the other day. She answered my phone call right then and there, took the time to talk to me. You don’t get that anywhere else.”</i>

Note. N = 14. CI = cochlear implant; ABR = Auditory Brainstem Response; BCMH = Bureau of Children with Medical Handicaps; NICU = neonatal intensive care unit.

did not know anyone with the condition ($n = 6$). Several parents also expressed concern for their child’s cognitive, physical, and social-emotional development compared to children with typical hearing, and feared their child would live a difficult life ($n = 7$). One mother expressed anxiety for potential bullying due to physical differences in the appearance of her child’s ears. In the focus group, she said, “I’m afraid of what his life is going to be like. Are people going to pick on him, because not only is he going to have this hearing aid on, he’s going to have these little ears? That is what I worry about.” However, half of the parents also agreed with the sentiment that managing their child’s hearing loss gets easier with time. As stated by a mother, “It’s been difficult at times but with the help of my doctors that we have for him, and his therapist and everybody that has helped us through it, it has been a bit easier for us to get through it emotionally and physically.” To normalize and adapt to the issue, parents expressed a desire to expose their child to other children and adults who are DHH ($n = 6$). Although there were several issues parents grappled with, their child’s hearing loss became more manageable as families adapted to their child’s hearing needs.

Barriers and Challenges

The second overarching concept, Barriers and Challenges, contains three major themes: Use of Hearing Devices, Scheduling, and Financial Costs, which are described further.

Use of hearing devices. A number of parents complained about the excessive time it takes for remakes of their child’s ear molds as well as setting aside time in their personal schedules to pick up the ear molds ($n = 5$). At least half of the parents communicated frustration with the management of their child’s hearing devices ($n = 6$), keeping hearing aids on their child ($n = 10$), and

obtaining insurance or financial coverage for hearing devices and services ($n = 7$). One mother expressed shock and frustration that her insurance didn’t cover Auditory Brainstem Response (ABR) tests and regular audiology appointments. “We had spent several hundreds of dollars before being approved for Bureau of Children with Medical Handicaps (BCMh) and even then, still waiting to get reimbursed for some of the costs. I can’t believe most insurance companies don’t cover hearing [technology], especially in children, yet they cover things like Viagra.” She was just one of many parents who conveyed both shock and frustration at the high cost of hearing devices and limited knowledge about financial assistance to cover them.

Scheduling. A majority of parents ($n = 8$) felt they did not have enough time in their schedules to make or attend appointments. In reference to the challenges she has experienced with her child’s hearing loss, one mother remarked, “I think it was those kinds of stresses and impact when you’re thinking about appointments and who can make the appointments.” Parents pointed out the excessive number of appointments in the first year for all service providers related to managing their child’s hearing loss. “You have to go see a pediatrician, you have to go talk to a geneticist. We did all of our appointments in one day, like eight appointments in one day, trying to get social worker, aural rehab, all that stuff.” Setting aside time in their personal schedules for a large quantity of appointments in the first year after identification of hearing loss and thereafter was a shared struggle among many parents in the interviews and focus group.

Financial costs. According to most parents ($n = 9$), the overall cost of services related to their child’s hearing loss was considered burdensome. One mother conveyed fearfulness in response to the cost of her child’s hearing

services. “I was terrified... ‘What are we going to do?’ We had family members—both sets of parents offered to loan us money, but not everyone has family support where they would just be able to get that money.” The cost of hearing devices specifically was a concern for a subset of parents ($n = 6$). According to one father, “My insurance—and I have the insurance for our whole family—said that for his hearing aid, they would cover 100% or a maximum of \$3,000. Well, he has Bahas and has two of them. They are approximately \$10,000.” Even for parents who have insurance benefits that cover the cost of hearing aids, the entire cost may not be covered, creating a frightening and stressful experience.

Supports

The overarching concept of Supports contains two major themes: Education/Resources and Healthcare Team. The section below elaborates on these themes in relation to their subthemes.

Education/resources. Parents identified the different types of education and resources they used after learning about their child’s hearing loss. Some of the parents ($n = 6$) joined support groups with other parents who have children who are DHH. One mother expressed gratitude for the support group in which this focus group was conducted. “There’s huge groups out there if I want to talk to people in other countries or across the country—but to have local parents, seeing the same departments, the same doctors, possibly the same schools, that’s huge.” A majority of parents ($n = 7$) also claimed to have conducted internet research to learn more about their child’s hearing loss. In the words of one mother, “I was googling the minute after the NICU staff left the bedside.” Other types of support from audiologists and listening and spoken language (LSL) schools or programs, were also mentioned in the focus group discussion—however, support groups and internet research were noted as the most common resources for learning about and coping with their child’s condition.

Healthcare team. The ACC was overwhelmingly noted by parents as an exceptional addition to the medical team ($n = 12$). She was praised for her overall support and timely communication with parents in need. One mother commented “She actually came when [child] had surgery, she showed up at Children’s downtown. We weren’t expecting her. She stayed for two, three hours and talked to all of us. She just goes above and beyond” and that if she has a question or concern, “She just always takes the time to research and find the correct answer.” The ACC was also appreciated for connecting parents to other specialists and organizing appointments in an efficient manner, especially for out-of-state patients. Most parents ($n = 12$) also noted audiologists, doctors, specialists, nurses, and staff in the Division of Audiology at CCHMC as helpful due to their promptness in communication and overall quality of care. One mother noted, “The one-on-one experience with them, you don’t get that anywhere else. They check on you and make sure you’re doing okay.”

Nearly all parents agreed that the entire healthcare team helped improve their experience with their child’s hearing health needs.

Discussion

This study queried parents of children who are DHH about their personal experiences with their child’s hearing loss. The thematic analysis revealed various challenges and supports as they managed, adapted, and coped with their child’s hearing loss. This section will discuss each theme that emerged from parents’ personal stories as they relate to the literature as well as implications for clinical practice and care coordination in pediatric audiology.

Reactions and Adaptation to Hearing Loss

Parents reported feeling shocked and unprepared when their child was identified as DHH, especially because they have typical hearing and do not know any parents with children who are DHH. Feelings of shock and unpreparedness are typical for parents who first learn about their child’s hearing loss (Jackson & Turnbull, 2004; Kurtzer-White & Luterman, 2003; Yoshinaga-Itano & Abdala de Uzcatogui, 2001; Young & Tattersall, 2007), especially because most parents have typical hearing and no prior experience with the implications of hearing loss (Centers for Disease Control and Prevention, 2010; Jackson & Turnbull, 2004; Mitchell & Karchmer, 2004). Additionally, parents in this study expressed concern about the physical, cognitive, and social-emotional development of their children as have other parents throughout the literature (Fitzpatrick et al., 2008; Henderson et al., 2014; Jackson, 2011; Jamieson et al., 2011; Yucel et al., 2008). Though parents in the present study expressed initial shock, their child’s hearing loss became easier to manage over time with consistent communication and support from the care coordinator and providers. Reliable and well-coordinated care systems provide access to resources such as childcare, community, and financial resources that are vital to parents of children who are DHH (Jackson, 2011; Jamieson et al., 2011; Yucel et al., 2008) and can help alleviate the stress around managing hearing loss. ACCs can be essential messengers of information and sources of support for parents who must manage their child’s hearing loss. Pediatric institutions should consider creating care coordinator positions within their audiology practices, or, developing policy that allows for more thorough coordination in practice. The FL3 Needs Assessment supports our finding that a primary contact through which support is coordinated, such as a family support coordinator, would be helpful in managing a child’s hearing health care (Ward et al., 2018).

Use of Hearing Devices

Hearing devices was one of the largest themes that emerged from the interviews and focus group discussion. Similar to parents in the literature, parents in this study expressed stress around communicating with their children, maintaining devices, and making decisions

about hearing devices (Dammeyer et al., 2019; Dirks et al., 2016; Fitzpatrick et al., 2015; Hintermair, 2000; Lederberg & Golbach, 2002; Quittner, 1991; Quittner et al., 2010; Quittner et al., 1990; Ward et al., 2018). Parents were also frustrated with the task of training family and friends in their child's hearing device management. However, parents mentioned the local support group as a safe and resourceful place to learn about hearing devices, especially from one of the members who is deaf herself. Parents became aware of the support group from the care coordinator and audiologists who managed their child's audiology care. The ACC and audiologists also shared written information about hearing devices, early intervention, and resources for support near their hometown area to help families understand their options and how to manage hearing devices. To help parents navigate obstacles related to hearing devices, coordinated care systems can connect parents with resources for teaching the entire family about hearing devices.

Scheduling

Many parents expressed frustration with the number of appointments in the first year for all services related to hearing. The hassle of scheduling and making time for hospital appointments has been mentioned in another study on stressors for mothers of children who are DHH (Jean, Mazlan, Ahmad, & Maamor, 2018). Additionally, taking time off from work and traveling for medical appointments are other barriers related to scheduling for parents of children with hearing loss (Henderson et al., 2014). Parents in the present study discussed juggling their own work schedules with their child's medical appointments, especially those who were traveling far distances. Though scheduling barriers are sometimes inevitable, one helpful resource parents identified for navigating scheduling barriers was the ACC. Parents were pleased with how she organized appointments in an efficient manner, particularly for traveling parents. Although healthcare systems can be rigid in operating structure, this is one demonstration of how care coordination can alleviate the burdens of parents.

Financial Costs

Parents felt a great deal of fear regarding finding ways to afford their child's hearing care. This is an area where parents can use assistance with resource and health care system navigation (Dammeyer et al., 2019; Dirks et al., 2016; Fitzpatrick et al., 2015; Hintermair, 2000; Lederberg & Golbach, 2002; Quittner, 1991; Quittner et al., 2010; Quittner et al., 1990; Ward et al., 2018) as well as dissemination of community and financial resources (Jackson, 2011; Jamieson et al., 2011; Yucel et al., 2008). In the focus group, parents mentioned that they learned about financial resources from other parents in the support group, which helped ease their fears. As mentioned previously, many of the group members became connected with the support group by the ACC and other providers in the Division of Audiology at CCHMC. The division also offers a parent binder to all families of children. It includes written information about funding

and financial assistance, as well as resources to support parents during early intervention and beyond. Coordination in pediatric institutions can help ensure all families receive information about the different supports available. This study showed that having personnel for care coordination facilitates comprehensive support to all families who receive treatment in our division.

Education/Resources

Support groups and internet research were the most highly discussed educational resources in this study. It is no wonder that parents considered the support group helpful. Social support is one of the most important mediators of parental stress (Asberg, Vogel, & Bowers, 2008; Lederberg & Mobley, 1990; Sarant & Garrard, 2013) and recommended for inclusion in care models for children who are hearing-impaired (Dirks et al., 2016). Support from other families with children who are DHH was noted as a valuable resource in the FL3 Needs Assessment (Ward et al., 2018). Support groups allow parents to share educational, childcare, community, and financial resources which are needed by the community of parents with children who are DHH (Jackson, 2011; Jamieson et al., 2011; Yucel et al., 2008). Support groups also advocate for hearing-related issues and may build parental empowerment, confidence, and competence in caring for a child with hearing loss (Henderson et al., 2014). Parents confirmed these findings in their discussions within the focus group. Audiology practices should consider identifying parents who may be interested in starting a support group, or providing information to patients about current support groups. Formal systems or positions for care coordination can help disseminate this information to families.

Although there is scarce literature on the role of parent internet research on child hearing health, one study found that the most searches for hearing loss related information are conducted by mothers (Porter & Edirippulige, 2007). However, the study found that parents did not always visit the most reliable websites. It may be helpful for practitioners to be aware of parents' tendencies to conduct internet research and offer reliable sources for them to peruse at home. The FL3 Needs Assessment indicated that parents desire online resources for learning about and managing their child's hearing loss (Ward et al., 2018). The ACC in our division is responsible for providing helpful websites for parents to read about their child's condition. This ensures that parents are receiving accurate information to make informed decisions for their child's hearing health.

Healthcare Team

Parents in this study spoke at length about the ACC as one of the most helpful supports. They repeatedly commented on how she goes "above and beyond" to provide social support, communicate in a timely fashion, answer questions, connect them to other specialists, and organize appointments in an efficient manner. Parents also identified other personnel in the Division of

Audiology at CCHMC as helpful (audiologists, doctors, specialists, nurses, and staff) due to the “one-on-one” and interpersonal care they received from these practitioners and staff. This type of support is in accordance with research that finds parents need health care systems with strong service coordination (Fitzpatrick et al., 2008; Jackson, 2011; Yucel et al., 2008) and a highly integrated and coordinated health care model in general (Hintermair, 2006; Fitzpatrick et al., 2008; Ward et al., 2018).

Implications for Clinical Practice and Care Coordination

Families need access to social support, financial assistance, and information about hearing devices, education, and communication with providers. In local practice, the Division of Audiology at CCHMC provides these services by emphasizing dual ownership between the family and service provider. Patients are encouraged to consistently attend appointments and come prepared to ask any and all questions that come to mind. Asking questions and having access to the appropriate contacts is a vital aspect of family involvement in hearing health care. The ACC serves as a primary contact that addresses concerns and connects patients with specialists and medical staff. The coordinator also regularly contacts parents in case they are too overwhelmed to seek advice on their own. Parents appreciate the open lines of communication and personal care they receive from the coordinator, as noted in the interviews and focus group. The coordinator also shares written information with parents regarding support groups, financial assistance, hearing device assistance, early intervention, and more. As mentioned in the introduction, the Division of Audiology at CCHMC also provides telehealth services, a newsletter, information about a Facebook and parent support group, and an FAC.

In 2015, audiologists at each CCHMC audiology location were asked to nominate potential parents to join the FAC with the goal of bringing together a diverse group of parents to help guide audiology practice from a patient experience perspective. Parents are from different locations around Cincinnati; have children with different types and degrees of hearing loss; use varying types of technology to assist with hearing; and communicate with their children via sign language, listening and spoken language, or a combined approach. Their children attend private or public schools. The FAC has suggested many changes to improve the patient experience in audiology such as updating the web page to make it easier to navigate, online scheduling for audiology appointments, a Facebook group, and changes to the cochlear implant program initial appointment paperwork. The FAC has also shared their experiences in learning their child was DHH and how they were impacted by the news. One parent shared that she appreciated how the audiologist told her that her daughter was deaf. The audiologist said that she had concerns about hearing and would need to do more testing. This allowed the parent to slowly come to terms with the fact that her daughter’s hearing may

not be typical. All parents agreed that they wanted to interact more with families with children who are DHH. They suggested an annual event, such as a picnic, where their children could interact with others with hearing differences and the creation of Facebook group exclusively for parents of children who are DHH. They also wanted to create a road map for new parents to educate them on the appointments that their child may need and why they were needed during the first years after diagnosis of hearing differences. The FAC also suggested the creation of a parent manual containing information about types of hearing loss, assistive technology, communication modes, and education choices.

Although hiring care coordinators to facilitate these services may not be feasible in some pediatric institutions, care coordination can still be integrated in hearing health care. We encourage further research on audiology coordination to develop a consistent coordination system across pediatric institutions. As the literature grows, evaluation of care coordination practices could help measure the benefits of care coordination. We recommend parent partnership in the design of care models and support services to ensure hearing health care is tailored to family needs. At a minimum, this can be accomplished through parent engagement and surveys for program improvements. In our personal experience, the FAC has been instrumental in collaborating with parents to improve care delivery. Support groups are also a resourceful place to learn about parents’ experiences and encourage parent engagement in hearing health care. Pediatric institutions may consider partnering with schools to disseminate support services and improve existing services. Future research should explore additional ways to obtain parent and stakeholder perspective and feedback.

Limitations and Future Directions

The Division of Audiology at CCHMC was limited in the number of patients who fit the criteria for the study, resulting in a smaller sample size than desired. Although demographic information about focus group participants was unavailable, all are patients of CCHMC with what appeared to be similar backgrounds to our interview participants. We plan to conduct more focus groups with our support group network in the future, which will allow us to better coordinate collection of demographic information without sacrificing anonymity. Although this study would have benefited from more data, a strength of this study was the robust information we received from focus group interaction that augmented the themes we had collected through the individual interviews. The findings from the focus group validated the themes we had already identified through the interviews and expanded our understanding of parent needs and supports. Though the focus group had an ideal number of participants, future studies should seek to attend multiple support groups to capture different voices and life experiences. Most parents in this study were Caucasian middle-class mothers. Attending various support groups and recruiting from other institutions may help capture the different life experiences of parents of

patients who are DHH. Finally, because some participants had children who were identified with hearing loss up to five years prior, there may have been inaccuracies in recollection of memory. Future studies should record parent experiences after identification of hearing loss and several other time points in the child's development, as these experiences likely differ at different stages of hearing intervention.

Conclusion

Hearing loss comes with many challenges for families who must accommodate their child's new hearing health needs. This study investigated the impact and experience of parenting a child who is DHH, as well as supportive resources for successful early hearing intervention and family well-being. Although parents struggled with using hearing devices, affording services, and adapting to their child's hearing loss, care coordination provided by an ACC and providers at CCHMC made a positive impact on the overall family experience. The consistency of the study's themes with the literature provides the opportunity to focus improvements in care coordination for families with children who are DHH. Audiology institutions should continue contributing to the growing literature on audiology care coordination by detailing and evaluating how family support services are coordinated within their own audiology care systems.

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Involvement of Adults Who Are Deaf or Hard of Hearing in EHDI Programs

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Abstract: Consistent with a position statement of the Joint Committee on Infant Hearing (JCIH, 2007), several key organizations and groups have supported involving adults who are deaf or hard of hearing (DHH) in Early Hearing Detection and Intervention (EHDI) systems, including providing families of children who are DHH with opportunities to interact with adults who are DHH. This article reviews the available data on the involvement of adults who are DHH in EHDI systems to determine the availability of opportunities for families who have children who are DHH to interact with adults who are DHH, how families feel about these experiences, and describe the programs that exist to provide these experiences. The article is based on results from three separate national surveys which included responses from parents and from EHDI related programs and organizations. Results showed that about half of parents with children who are DHH wanted opportunities to interact with adults who are DHH, but often experienced difficulty accessing these connections. Also, the variety of these services were too limited, and programs that promote involvement of adults who are DHH need more funding to provide these services to families.

Key Words: Deaf, Children, Adults Who Are Deaf or Hard of Hearing, Deaf Mentor

Acronyms: ASL = American Sign Language; DHH = deaf or hard of hearing; EHDI = early hearing detection and intervention; FL3 = Family Leadership in Language and Learning; GBYS = Guide By Your Side; HRSA = Health Resources and Services Administration; JCIH = Joint Committee on Infant Health; LSL = Listening and Spoken Language; MCHB = Maternal Child Health Bureau; NCHAM = The National Center for Hearing Assessment and Management

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Several key stakeholders in early hearing detection and intervention (EHDI) systems have made statements recommending what they consider to be best practice, ensuring opportunities for families with children who are deaf or hard of hearing (DHH) have opportunities to interact with adults who are DHH. This article summarizes a review of these statements which is crucial to understanding the current landscape of how adults who are DHH are involved in EHDI systems.

The Joint Committee on Infant Hearing (JCIH) was established in 1969 for the purpose of gathering professionals in the fields of audiology, otolaryngology, pediatrics, and nursing to discuss and summarize the best practices for early intervention programs for infants who are deaf or hard of hearing (DHH). Today, the committee is comprised of representatives from the Alexander Graham Bell Association for the Deaf and Hard of Hearing,

American Academy of Pediatrics, American Academy of Audiology, American Academy of Otolaryngology-Head and Neck Surgery, American Speech-Language-Hearing Association, Council of Education of the Deaf, and Directors of Speech and Hearing Programs in State Health and Welfare Agencies. In 2007, the JCIH published a position statement summarizing research and making recommendations to ensure high-quality Early Hearing Detection and Intervention (EHDI) programs for children who are DHH.

The 2007 JCIH Position Statement strongly endorsed having adults who are DHH play “an integral part in the EHDI program” (p. 903). The statement concluded that connecting parents with adults who are deaf or hard of hearing is a critical part of ensuring parents have the opportunity to make informed decisions (JCIH, 2007). JCIH suggested that connecting parents with adults who

are DHH is a good first step and added that adults who are DHH should be included in all aspects of EHDI programs, including serving on state EHDI advisory boards.

Other groups have also advocated for the inclusion of adults who are DHH in providing support to families of children who are DHH. For example, Moeller, Carr, Seaver, Stredler-Brown, and Holzinger (2012), described the conclusions of an international consensus panel about *Best Practices in Family-Centered Early Intervention for Children Who are Deaf or Hard of Hearing*. The panel noted that “Families [should be] connected to support systems so they can accrue the necessary knowledge and experiences that can enable them to function effectively on behalf of their DHH children ... [including supporting] connections between families and adult role models who are DHH” (p.435). Moeller et al. encouraged early intervention programs to provide families with opportunities for “meaningful interactions” (p. 441) with adults who are DHH including involving them on early intervention teams as role models, mentors, and/or consultants, who can offer information and resources, and “demonstrate enriching language experiences” (p. 441).

Providing families of children who are DHH with opportunities to interact with adults who are DHH is also encouraged in federal legislation that provides funding for all of the state-based EHDI programs. As noted in the Early Hearing Detection and Intervention Act at 42 USC 280g-1(a)(1)(c), “Programs and systems under this paragraph shall offer mechanisms that foster family-to-family and deaf and hard-of-hearing consumer-to-family supports.”

Additionally, the U.S. Department of Health and Human Services, Maternal Child Health Bureau/Health Resources and Services Administration (MCHB/HRSA) issued guidance for funding the “Family Leadership in Language and Learning (FL3)” program in 2017. HRSA noted one goal of the program was “[t]o increase by 30 percent from baseline, the number of families that are offered support from Deaf Mentors by the end of the three-year project period” (p.1). The term Deaf Mentors was later clarified to include “[m]entoring by a variety of DHH adults including those who use ASL [American Sign Language], Cued Speech, Listening and Spoken Language (LSL), and combinations of modalities” (Hands & Voices, 2017b).

Given the widespread support for programs to provide opportunities for families of children who are DHH to interact with adults who are DHH, this article summarizes the available data to paint a picture of what is known about the programs that offer these opportunities to families of children who are DHH.

Data Collection Methods

Data were collected from the following sources to draw conclusions about how many families of young children

who are DHH have opportunities to interact with adults who are DHH. Data also indicated how they felt about those interactions if they had them.

- The National Center for Hearing Assessment and Management (NCHAM) at Utah State University surveyed the coordinators and state-based EHDI programs in all states and territories in 2010 and updated that information via a similar survey and telephone interviews in 2017. State EHDI coordinators were asked to provide information about programs in their state or to recommend other people in the state who might have better information. Data were collected from people in 49 states and territories (Shuler-Krause, 2018). Details about the data collection methods and a report on the findings are available at <https://tinyurl.com/dhhadultinvolvement>
- In 2018, NCHAM published the results of a national study entitled EI SNAPSHOT (*Early Intervention for Children who are Deaf or Hard of Hearing: Systematic Nationwide Analysis of Program Strengths, Hurdles, Opportunities, and Trends*). A part of the EI SNAPSHOT study included data collected from a national sample of 303 families with 2–6 year-old children who were DHH. Details about the data collection methods as well as results, conclusions, and recommendations of the larger study are available at <https://infantheating.org/ei-snapshot/>
- During 2017–2018, the newly funded FL3 program conducted a national needs assessment to take an “in-depth look at the needs of families, family-based support organizations (FBOs), and U.S. state and jurisdiction EHDI programs with the purpose of ensuring that the FL3 is helping to meet the needs of all families of children who are or are at risk for being DHH” (p.5). One part of this Needs Assessment collected information from a national sample of 458 families of 0–6 year-old children who were DHH. Information from these families included their responses to questions about the extent to which they had had interactions with adults who were DHH and, for those who had had such interactions, their perceptions about benefits, challenges, and opportunities for improvement. Details about the data collection methods as well as results, conclusions, and recommendations of the complete Needs Assessment study are available at <https://handsandvoices.org/fl3/resources/needs-assessment.html>

Summary of Available Data

To provide context for the results about the availability of programs that provide opportunities for families of young children who are DHH to interact with adults who are DHH,

The approximate number of families served in each of the programs is shown in Figure 2. Most of the programs serve less than 25 families at any point in time. Combining the results across programs, we can estimate that less than 1,000 families were receiving services from DHH Mentors/Guides/Role Models at the time these data were collected. Table 1 shows that 45% of families reported that they wanted opportunities to interact with adults who are DHH and 22% had no problems accessing such services. Slightly more families (59%) reported that they wanted to meet with other families who had children who were DHH and 34% were able to access these experiences.

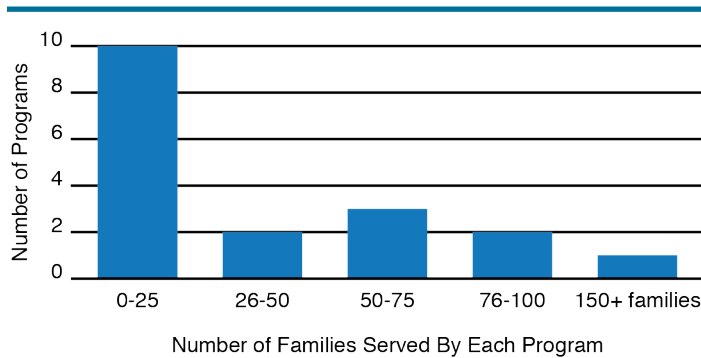


Figure 2. Approximate number of families served programs that offer families of children who are deaf or hard of hearing (DHH) opportunities to interact with adults who are DHH.

Table 1
Data from EI SNAPSHOT Study (2018): Percentage of Families of Children who are Deaf or Hard of Hearing (DHH) who Wanted and were Able to Access Opportunities to Interact with Adults who are DHH and Families who had Children who are DHH

Type of service	We Needed and received it with no problems	We Needed, but had trouble getting it
Opportunities to interact with adults who are deaf or hard of hearing	22%	23%
Meeting with other families with children who are DHH	34%	25%

As shown in Figure 3, the FL3 Needs Assessment had similar findings with 27% of families reporting that they had access to a DHH Role Model. Interestingly, of those families offered the opportunity to interact with an adult who is DHH, only 69% actually met with an adult who is DHH. Families in the FL3 Needs Assessment who had an opportunity to meet with an adult who was DHH but opted not to do so reported being too busy, feeling that it did not meet their needs, or they already had contact with adults who are DHH (see Figure 4).

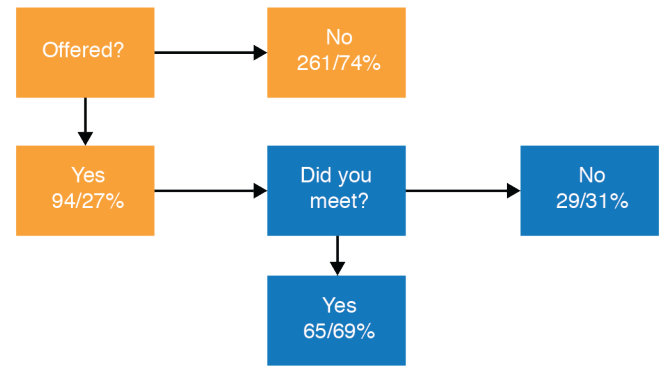


Figure 3. Data from Family Leadership in Language and Learning (FL3) Needs Assessment (2017). Percentage of families with children who were offered and met with Role Models who are deaf or hard of hearing.

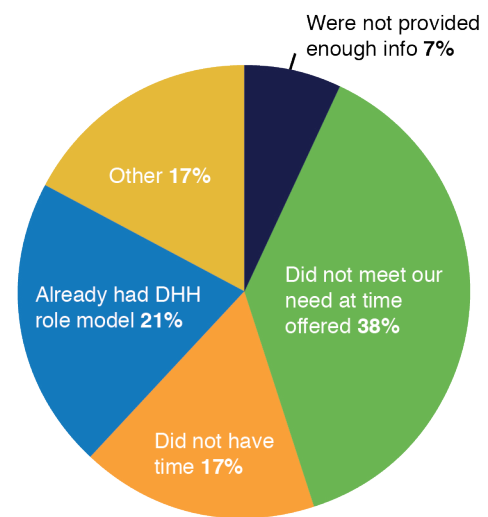


Figure 4. Reasons families in the Family Leadership in Language and Learning (FL3) Needs Assessment did not meet with the Role Models who are deaf or hard of hearing.

Families in the FL3 Needs Assessment who met with an adult who was DHH were asked what they perceived as benefits of these interactions. As shown in Figure 5, the most frequently cited benefits were that the DHH Role Model provided information about communication in different situations and helped increase the family's confidence in deciding how they would communicate with their child who was DHH.

The FL3 Needs Assessment (Hands & Voices, 2017a) also included focus groups in which participants were asked "How would a DHH role model be valuable to your family's experience?" (p.33). Participants noted that mentors, role models, and guides were helpful because "They could help you ask the questions you don't know you have yet," (p. 33) and "They could answer questions about the future, things she can do, things she can't, and the best way to teach her about hearing loss" (p. 33). Other parents noted

that adults who are DHH helped them with issues like “How to access interpreters,” (p. 33) and “How to connect more with kids my son’s age who have cochlear implants” (p. 33).

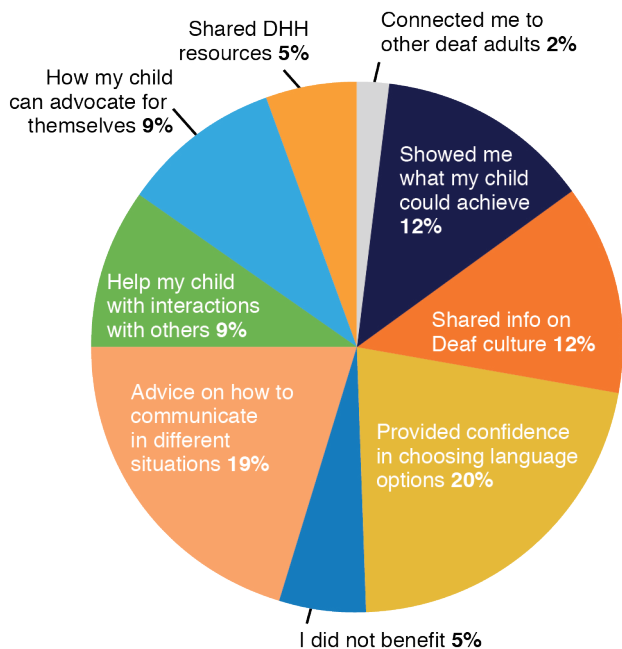


Figure 5. Benefits of meeting with a Role Model who is deaf or hard of hearing reported in the Family Leadership Language and Learning (FL3) Needs Assessment.

Program Administration and Funding

Shuler-Krause (2018) also gathered information about the programs’ structures including administrative affiliations, annual budgets, and funding sources. As shown in Figure 6, most programs that involve adults who are DHH are administered by non-profit organizations, family-based organizations, and state schools for the deaf.

Programs that systematically involve adults who are DHH use a variety of funding sources including private grants, early intervention/Part C funding, state EHDI/HRSA federal funding, Medicaid billing, and state or federal grants (see Figure 7).

Many programs (41%) used other sources of funding which included State Department of Education, State Schools for the Deaf, Deaf and Hard of Hearing Resource Centers, School Districts, State Association of the Deaf, and State Department of Health and Human Services. Almost all programs (21 of 22) reported using multiple sources to fund their programs.

Annual budgets for programs involving adults who were DHH ranged from under \$10,000 to over \$150,000 each year as shown in Figure 8. The amount of budget for the program was positively correlated with the number of families receiving services.

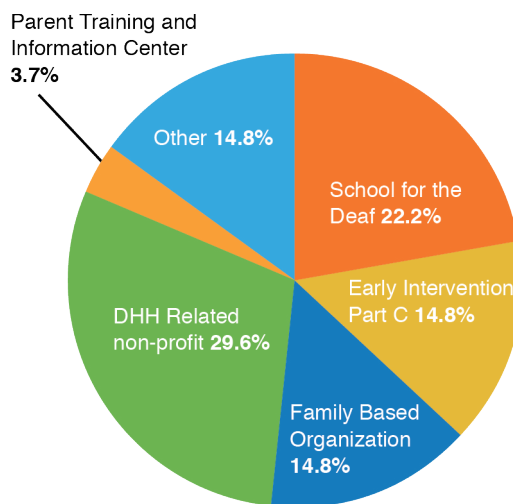


Figure 6. Affiliation of programs that provide opportunities for families of children who are deaf or hard of hearing (DHH) to interact with adults who are DHH.

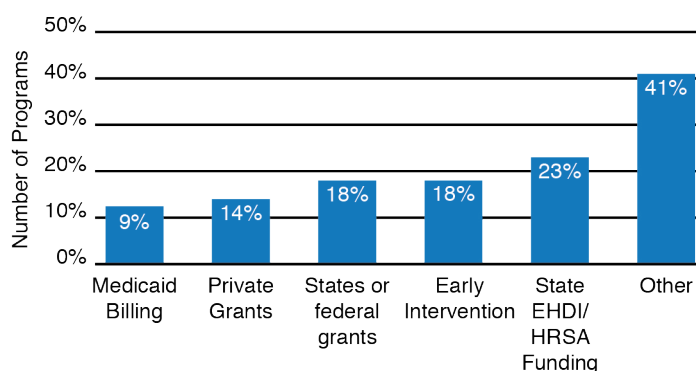


Figure 7. Funding sources of programs that offer opportunities for families of children who are deaf or hard of hearing (DHH) to interact with Adults who are DHH. EHDI = Early Hearing Detection and Intervention; HRSA = Health Resources and Services Administration.

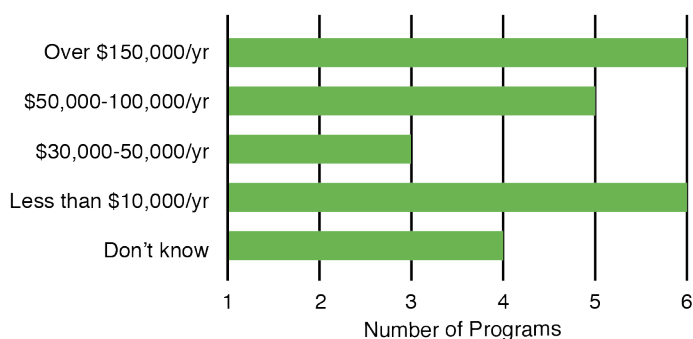


Figure 8. Annual budgets of programs that offer opportunities for families of children who are deaf or hard of hearing (DHH) to interact with adults who are DHH.

Focus and Frequency of Visits

Most programs reported that a majority of the families served by their programs had children who were DHH in the 13 to 24-month age range, and 70% of programs responded that families were provided opportunities to interact with adults who are DHH on a weekly basis, as shown in Figure 9.

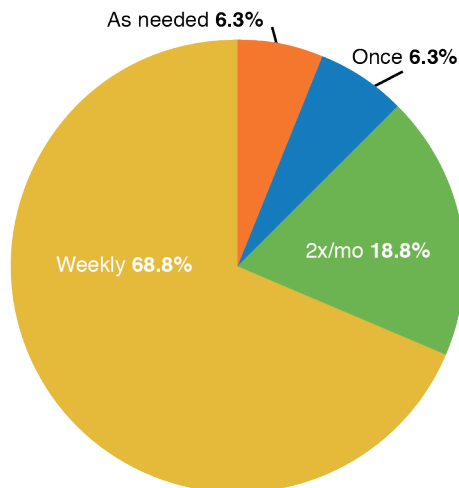


Figure 9. Frequency of Deaf Mentor services.

Programs were asked about the emphasis placed on teaching children and families a specific language or modality. As shown in Figure 10, 16 of the 24 programs (67%) reported that their programs had a moderate-major emphasis on teaching children and families a specific language or communication modality, with all of these focusing on ASL or sign language.

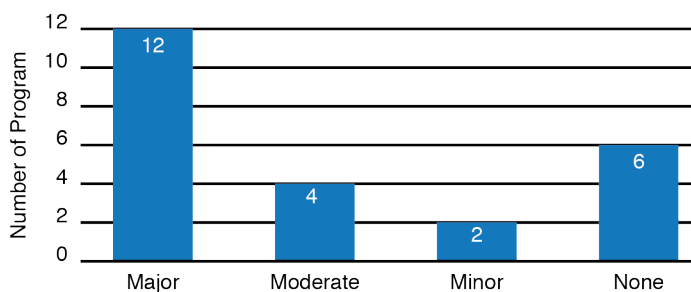


Figure 10. Emphasis placed on teaching families a specific language or communication method.

When asked if their program used an established curriculum or training, 13 programs responded that they used the SKI-HI curriculum (SKI-HI), two programs used the Hand & Voices (2017c) Guide By Your Side training, and one program used the Shared Reading Project curriculum (Clerc Center, 2015).

Challenges

As shown in Figure 11, programs cited securing and maintaining funding as the number one challenge they faced. Other commonly reported challenges were in the areas of recruitment of skilled/qualified staff and lack of training opportunities. Some programs mentioned difficulty finding racially, linguistically, culturally, and hearing level diverse DHH individuals in their state, as well as challenges in receiving timely referrals to their programs.

Discussion and Conclusions

Support for including adults who are deaf or hard of hearing in EHDI systems and ensuring that parents of children who are DHH have the opportunity to connect and interact with adults who are DHH is not new, but has gained momentum following the 2007 JCIH position statement, the signing of the EHDI Reauthorization Act, and the initiation of the FL3 project. Widespread support for including adults who are DHH in EHDI systems is undeniable, yet less than half of states report having a systematic program for ensuring these connections and 45% of families who have children who are DHH report that they would like to have such interactions. Of the families who wanted these opportunities, 22% reported that they had difficulty accessing them. However, of the parents who were offered the opportunity to meet with adults who are DHH, only 69% actually met. The parents that did not take advantage of these offerings said they were too busy or felt that the program did not meet their needs at the time. It is also important to note that in answering this question in the SNAPSHOT study, a slightly higher percentage (59%) of families reported being interested in meeting with other parents of children who are DHH, and 25% of these families had difficulty accessing these experiences. Although many families are interested in opportunities to interact with adults and have difficulty accessing these experiences, an even higher percentage are interested in meeting other parents of children who are DHH and these families have even more challenges finding these opportunities.

Although there is a strong desire for opportunities to interact with adults who are DHH, the availability of these programs is only one factor to consider in providing support from adults who are DHH to parents. EHDI systems should also consider other factors that influence parent engagement in these systems and ensure that their programs provide these services in a way that meets the needs of each family. These factors include consideration of what stage in the journey families most benefit from these services, scheduling opportunities at a time that works for families, and ensuring diversity of the adults who are both DHH and available to meet with families. Over 67% of programs reported a moderate to major emphasis on instruction of a specific language or modality, and of those, 100% of the programs reported a focus on sign language or ASL instruction. This is significant because

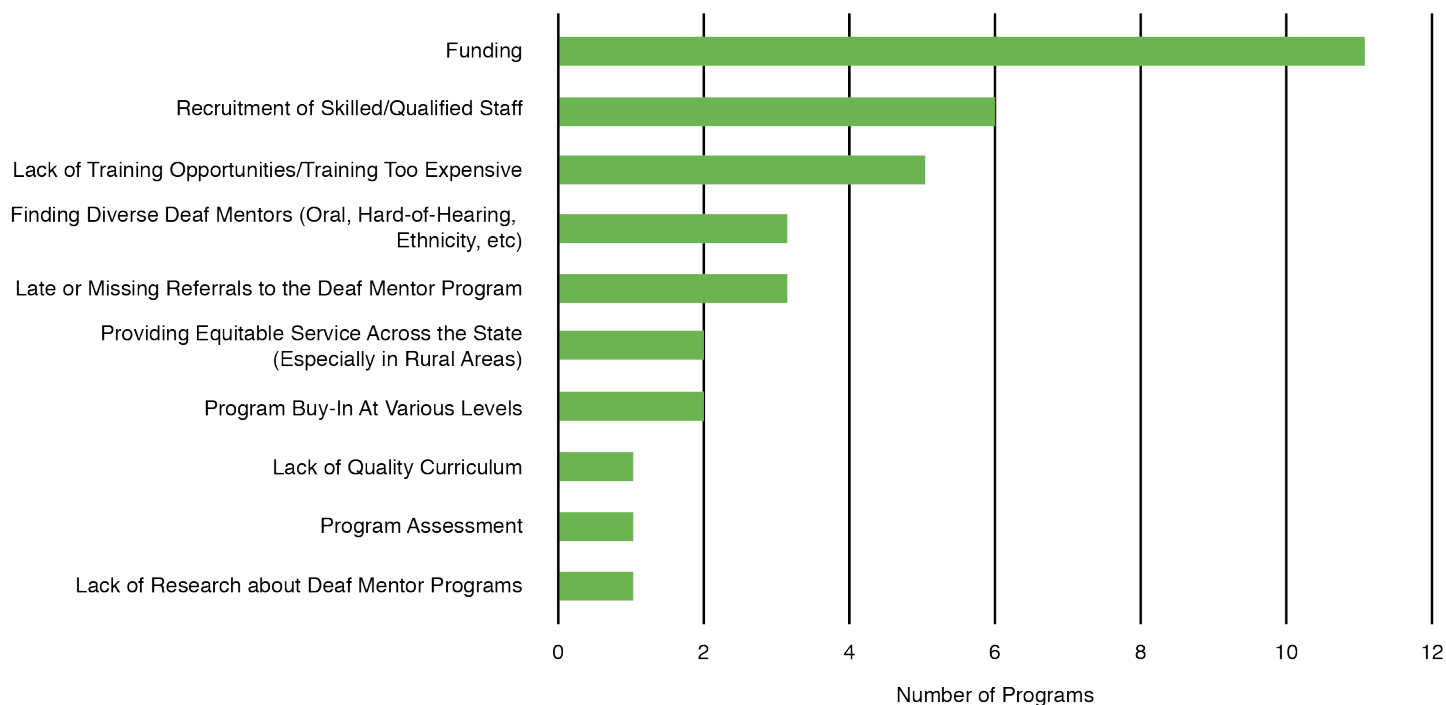


Figure 11. Challenges faced by Deaf Mentor programs.

children who are DHH and their families use a variety of different methods and languages to communicate with each other. Regardless of the way the child or family communicates, all families can potentially benefit from interacting with adults who are DHH. This finding suggests that EHDI systems should also consider how to provide a wider range of opportunities to interact with adults who are DHH. EHDI systems should work to reflect the diverse communication preferences of children who are DHH and their families including ASL and sign language instruction as well as a focus on English language acquisition, listening and spoken language skills, cued speech, and other communication modalities.

When families did meet with adults who are DHH, they touted many benefits which echoed some of the findings of the Deaf Mentor Experimental Project conducted more than 20 years ago (Watkins et al., 1998). These benefits included increased parental confidence in deciding how to communicate with their child and increased parental understanding of different ways to communicate with their child in different situations. Additionally, parents reported benefiting from information the adult who is DHH shared with them about Deaf culture and from seeing a model of what their child is capable of achieving in the future.

Data available about programs that offer opportunities for adults who are DHH to interact with families do not address whether parents see any disadvantages of such interactions. This would be important information in definitively determining the overall effect these experiences have on parents. However, it appears that parents receive significant benefits from these experiences.

Surprisingly, programs designed to offer opportunities for families of children who are DHH to interact with adults who are DHH varied significantly in how they are administered. Programs are run by state schools for the deaf, Part C services, parent support groups, and non-profit organizations among other types of groups. Many programs are quite small, serving less than 25 families, although some of the larger ones serve at least 50 families each year. Annual program budgets also reflect this with a range from less than \$10,000 to over \$150,000 per year with funding coming from a variety of sources including private grants, early intervention/Part C funding, state EHDI/HRSA federal funding, Medicaid billing, and state or federal grants. Almost all programs reported that they get funding from more than one of these sources. The fact that these programs are administered by so many different groups and in so many different ways, may contribute to the low availability of these opportunities for families depending on which part of the country they live in, what programs they are aware of, and how eligibility criteria differ.

Results also pointed to other barriers in providing these services to families. Not surprisingly, funding was the biggest challenge faced by programs. Programs also reported challenges with recruitment of diverse, skilled, and qualified individuals who are DHH. Finally, programs struggled with finding affordable training and appropriate curricula. Funding, staff, training, and curriculum are all vital components of programs that offer families support from adults who are DHH. These challenges are yet another reason for a low availability of these opportunities for families of children who are DHH.

Results indicate that many families would like opportunities to interact with adults who are DHH, and that families who forge these connections benefit from them, but these opportunities are too limited in availability and scope within EHDI systems. Additionally, increased funding is needed to administer these programs alongside other EHDI services.

As these programs continue to expand, it is important that research and evaluation data be collected to determine the costs, benefits, and challenges associated with programs that provide opportunities for families to interact with adults who are DHH. Future research should focus on the following questions:

- What kinds of interactions with adults who are DHH most benefit families?
- At what stage in the family's journey do they most benefit from these interactions?
- What kinds of outcomes do these interactions produce for the child's social/emotional development and/or educational attainment?
- Do these opportunities have an impact on family engagement in the EHDI system?

This article drew data from surveys completed by parents, state EHDI coordinators, service providers, and family based organizations. More research outside of surveys is also needed to determine the answers to the questions asked above, as well as to provide evidence to encourage public health agendas in regards to funding these kinds of programs.

Although there is widespread agreement about the positive benefits of families of children who are DHH interacting with adults who are DHH, there is little data available on this topic. As these programs become more widely available, it is important this data is collected and carefully considered to ensure that resources are expended in ways that will be most beneficial to families.

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Listen to Us: Dad-Endorsed Strategies for EHDI Professionals

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Abstract: Although fathers play an increasingly important role in today’s families, there is an imbalance of research regarding fathers of children with disabilities compared to mothers, including research about fathers of children who are deaf or hard of hearing. With the advent of newborn hearing screening, the number of children who have hearing loss being identified within the first six months of life has significantly increased. Thus, the number of fathers participating in EHDI services has also increased. This article seeks to answer the question, “What do fathers’ experiences in the early intervention process tell professionals about family-centered practice?” The authors have synthesized the available research into strategies that EHDI professionals can take to foster more effective parent-professional relationships with fathers of children with hearing loss. Discussion is offered regarding implementation of these action steps as part of family-centered early intervention that considers the needs of fathers in the context of the whole family system.

Key Words: family-centered, fathers, deaf, early intervention, EHDI

Acronyms: DHH = deaf or hard of hearing; EHDI = early hearing detection and intervention; IEP = Individual Education Plan; IFSP = Individual Family Service Plan; NFI = National Fatherhood Initiative

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Whenever it was a home visit day, I always thought of an excuse to not be there. I didn't really know what I was supposed to do. My wife seemed to have such a good rapport with [the early interventionists] and I felt self-conscious—kind of like a third wheel on a date. It wasn't that I didn't care about [my daughter], it was just really uncomfortable so I found other things to do during that time. I had the cleanest garage in town during those early months!

At no other time in the history of the education of students who are deaf or hard of hearing (DHH), has the opportunity for communication access and optimal educational and career outcomes been so great (Strickland, Eichwald, Cooper, & White, 2011). Advances in technology now allow for identification of hearing loss in infancy and pave the way for timely early intervention for children who are DHH and their families.

Essential in the early intervention process are family-centered practices, which reflect an equal partnership between parents and professionals rather than an approach where professionals are viewed as the experts. Placing the family at the center of the early intervention process is based on overwhelming research demonstrating that when all members of the child’s family are involved and empowered, child outcomes are positively impacted. A meta-analysis conducted more than 30 years ago

by Shonkoff and Hauser-Cram (1987) found that early intervention was most successful for infants and toddlers with disabilities when family members were involved. A wide range of child outcomes are associated with family involvement and parental self-efficacy, including social development, cognitive skills, school readiness, emotional well-being, decreased problem behaviors, and later academic achievement (Turnbull, Turnbull, Erwin, Soodak, & Shogren, 2015). Research with children who are DHH shows higher levels of family involvement in early hearing detection and intervention (EHDI) are associated with better child language and literacy development (Calderon, 2000; Moeller, 2000; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998). Healthy family functioning, parental involvement, empowerment, and engagement comprise the foundation for positive child outcomes for all children, including those who are DHH. The question arises, though, are all members of the child’s family truly being included in the family-centered equation?

Research on the evolution of gender roles and the make-up of the Western family recognizes that fathers are increasingly taking on child care-giving responsibilities once reserved only for mothers. Thus, the unique contributions of fathers to the healthy development of their children is receiving national attention (Chelsey, 2017; Valiquette-Tessier, Gosselin, Young, & Thomassin, 2018). A meta-analysis of father involvement (Sarkadi,

Kristiansson, Oberklaid, & Bremberg, 2008) revealed a positive association with child outcomes of cognitive and language skills, decreased problem behavior in boys, and fewer mental health issues in girls across factors such as socio-economic status and family structure (Figure 1). A look at the impact on child outcomes when fathers are not involved is more startling. The National Fatherhood Initiative (NFI) reports father absence is associated with higher risk of poverty, teen pregnancy, behavioral problems, incarceration, substance abuse, child neglect, and school failure (NFI, 2016).



Figure 1. Father involvement is associated with improved child outcomes.

Despite these data, research in family-centered early intervention is heavily reflective of mothers. This is problematic because, although EHDI professionals are uniquely poised to support healthy family functioning from the start of a child's life by supporting all members of the family, professionals may not be equipped with knowledge and skills to attend to the unique aspects of father involvement. In a profession where the representation is predominantly female, it is important for EHDI professionals to be aware of any unconscious bias that may potentially interfere with equal engagement by both mothers and fathers in the services provided. A cultural competence model of intervention warns that when professionals are unaware of their own potential biases, they may often default to their own world view (Lynch & Hanson, 2011). Professionals should examine any potential unconscious biases they may hold associated with parenting roles and leave them at the door.

The purpose of this article is to leverage the influence of EHDI professionals on establishing empowered and engaged families by building awareness of the available research on fathers relative to early intervention and by offering strategies for family-centered services that include fathers of children who are DHH. The term father is used here as inclusive of biological, adoptive, foster, traditional

marriage, custodial and non-custodial, and other males serving as a substantial and consistent influence in the life of a young child.

Fatherhood Culture

A large body of literature exists regarding traditional and evolving gender differences, including learning preferences, parenting approaches, parent-child interaction styles, and social-support needs. This research has yielded varying results, especially as concepts of gender in our society become more fluid (Majdandžić, de Vente, Colonnaesi, & Bögels, 2018). Yet, there is recognition that support needs of men can be different than those of women. For the first time, the American Psychological Association (APA) issued a guidance document for practitioners when working with boys and men (APA, 2018). The existence of a culture of fatherhood has been increasingly recognized and researched in the sociology and gender fields since the turn of the 21st century; this research has also been applied in working with fathers in human services fields such as Social Work (Dermott, 2014; Wall & Arnold, 2007).

Bodner-Johnson (2001) recommends that EHDI professionals adopt an adult learning perspective that seeks to know parents as individuals to form better partnerships. The following list summarizes some general differences between mothers and fathers that may be relevant for consideration by professionals as they approach the process of getting to know individual family members (Lamb & Lewis, 2010; National Family Preservation Network, 2012; Pelchat, Lefebvre, & Perreault, 2003; Pruett, 1998).

- Whereas mothers tend to be first focused on day to day care tasks, fathers tend to focus on outer-world and future aspects.
- Whereas mothers tend to excel at interpersonal and group communication, fathers are often less likely to independently seek social support.
- Whereas mothers' interactions tend to focus more on care-taking than play, play is the prominent factor in father-child interactions. Fathers' play is more physical and unpredictable than is mothers'.
- Whereas mothers' interaction style tends to be predictable and safe, fathers tend to build confidence by allowing more freedom to explore and encourage risk-taking.
- Whereas mothers' discipline tends to stress sympathy, care, and problem-solving, fathers' discipline tends to focus on justice, fairness, and explanation of rules.
- Whereas mothers tend to modify their language in communicating with their child, fathers tend to use shorter utterances but are less likely to modify their language.
- Whereas mothers tend to be more comfortable learning through listening and talking, fathers tend

to prefer kinesthetic, tactile, and visual learning strategies in an informal environment.

- Whereas mothers tend to be comfortable discussing personal relationships and sharing self, fathers are more task-oriented, and less likely to talk about relationships without support.

The composition of the American family continues to change and become more diverse. Cultural norms must be considered within the culture of fatherhood as gender roles are often dictated or influenced by the family's cultural affiliation. Professionals must keep in mind that descriptions of gender roles and their associated recommendations for interaction are helpful in a broad context; however, careful attention to the individuality of each family member and the family system is paramount.

Family-Centered EHDI

The Division for Early Childhood at the Council for Exceptional Children¹ (2014) defines family-centered practices as

Practices that treat families with dignity and respect; are individualized, flexible, and responsive to each family's unique circumstances; provide family members complete and unbiased information to make informed decisions; and involve family members in acting on choices to strengthen child, parent, and family functioning. (p. 10)

Much has been written regarding family-centered EHDI practices since the turn of the 21st century, such as the Supplement to the JCIH 2007 Position Statement outlining best practices in early intervention after diagnosis of hearing loss (Muse et al., 2013). The ability of the early interventionist to establish a trusting relationship with the family is vital to the implementation of family-centered practices in EHDI. That relationship can be used as a foundation to support families in discovering their strengths and needed resources to parent their child who is DHH (Stredler Brown, 2005). Best practice recommendations for building effective family-centered parent-professional relationships in EHDI include (a) focusing on strengthening competence and self-efficacy, (b) using a non-judgmental approach, (c) asking families what information and resources they need rather than assuming, (d) using active listening and supported problem-solving, (e) offering both social and emotional support opportunities, and (f) providing support for self-determination (Ingber & Dromi, 2009; Sass-Lehrer, 2004).

Establishing and maintaining relationships with families requires early interventionists to strategically select and employ strategies that are a match to the unique needs of a particular family. Just as a one-size fits all approach is ineffective when teaching children, failing to meet the unique needs of parents, including fathers, can result in less than optimal parent-professional relationships.

Dads of Children who are Deaf or Hard of Hearing

Not surprisingly, literature addressing fathers of children who are DHH is limited. This limited research does appear to align with two important findings of research conducted with fathers whose children have other types of disabilities: (a) that father role-identity and parenting self-efficacy are positively associated with involvement in their child's programming, and (b) that father involvement was a mediator in mothers' stress and led to increased family harmony (Hintermair & Saramski, 2019; Ingber & Most, 2012; Zaidman-Zait, Most, Tarrasch, & Haddad, 2018). Further, the barriers to father involvement identified in other disability areas may also occur in EHDI programs. Muñoz, Nelson, Blaiser, Price, and Twohig (2015) surveyed 45 professionals providing services to families of young children who were DHH. The professionals reported teaching skills directly to mothers 91% of the time, while teaching to fathers only 19% of the time. Muñoz et al. also describe the practice of EHDI professionals focusing on child skills and lacking in their attention to the emotional and learning needs of parents. There is also evidence to suggest that fathers of children who are DHH process the parenting experience differently, and therefore, may require different types of support (Hintermair & Saramski, 2019; Zaidman-Zait et al., 2018). Fathers' own input on how they can be involved in the parenting experience is equally scarce in the literature. Table 1 displays five peer reviewed studies examining the recommendations of fathers to facilitate their own involvement.

Strategies for Professionals to Offer Family-Centered Services that Include Dads

Findings from the studies described in Table 1 have been synthesized into seven strategies that EHDI professionals can use when seeking to provide family-centered services that consider the needs of fathers. Given the diverse and evolving nature of gender roles and varying family compositions, these strategies may also be applied to other family members, in addition to fathers, who face similar potential barriers to involvement in EHDI services. The seven strategies for EHDI professionals are:

- Offer flexible options for fathers.
- Be patient, persistent, and proactive.
- Treat fathers as equal partners in parenting when sharing information.
- Build a team that is knowledgeable, current, and unbiased about ALL aspects of the child.
- Remember different isn't wrong.
- Build confidence and competence.
- Facilitate novice to expert father support.

To further add father voice, fathers' quotes from Pedersen and MacIver's (2013) study relevant to each of the seven strategies is offered.

¹The Council for Exceptional Children is the premiere professional organization devoted to quality education for children and youth with disabilities. The Division for Early Childhood is one of 17 specialty divisions and focuses on children ages birth to eight years with disabilities or those at risk for disabilities.

Table 1**Peer Reviewed Research on Fathers of Children who are Deaf or Hard of Hearing**

Study	Design	Population	Relevant Findings
Houston, 2012	Quantitative survey	262 American fathers	Fathers wanted professionals to be up-to-date with latest information, share information and options without bias, and be more proactive about including fathers.
Ingber & Most, 2012	Correlation with control group	38 Israeli fathers	Fathers of preschool children with and without hearing loss self-reported similar levels of involvement. Involvement correlated positively with parenting self-efficacy, family cohesion, and adaptability.
Pedersen & MacIver, 2013	Qualitative focus group	10 American fathers	Fathers value service options, including those specifically designed for children who are DHH, and highly trained EHDI professionals. Identified themes in narratives were Memories, Relationships, Unique Dad Characteristics, and Advice.
Hintermair & Saramski, 2018	Quantitative survey with correlation	92 European fathers	A strong relationship between parenting self-efficacy and perceived EHDI support programs and activities. Impact of a child's hearing loss on parenting may be unique in fathers.
Zaidman-Zait, Most, Tarrasch, & Haddad, 2018	Quantitative survey with correlation	30 Israeli-Arab fathers	Mothers were more involved than fathers in intervention activities. Parental self-efficacy, informal and formal social support was associated with father involvement. Only formal social support was associated with involvement for mothers.

Note. DHH = deaf or hard of hearing.

Offer flexible options for fathers.

These kids are expensive so both of us can't always be taking off work for appointments and home visits, someone's gotta work.

In a survey of over 700 fathers conducted by the National Fatherhood Initiative (2016), fathers cited work responsibilities as the number one obstacle to being a good father and financial problems as the third. Although the American family increasingly has both parents working, working mothers are often afforded more flexibility in work hours and work absences than working fathers (Harrington, Van Deusen, & Humberd, 2011; Rehel & Baxter, 2015). Additionally, care must be taken to consider the inclusion of fathers who do not live with their children, military fathers, and fathers whose jobs take them away from home for long periods of time.

Family life in general is becoming increasingly busy and all families struggle with time demands. Families who have children with disabilities specifically report struggling with the sheer number of appointments and tasks they are asked to complete and express frustration with a lack of flexibility in scheduling family-centered services (Brotherson & Goldstein, 1992).

For EHDI providers, accommodating the wide variety of differences in fathers' schedules may seem overwhelming. To combat this, maximizing the time fathers are available for services is key (also known as getting the most bang for one's buck). Coordination among appointments is especially helpful to allow fathers a chance to form relationships with other EHDI providers they may not see as often as mothers. Embedding intervention strategies into daily routine is a fundamental strategy in family-centered services; however, EHDI providers

must recognize that daily routine interactions between fathers and children are unique. Professionals also need to consider alternate options for father participation other than the typical home visit format. Many organizations of parents of children who are DHH host a variety of family events such as barbeques, carnivals, and even ice fishing in North Dakota! Since fathers tend to be task-oriented, these family activities may offer opportunities for fathers to be assigned specific responsibilities that may lend themselves to more natural involvement such as grilling hotdogs or coaching the softball game at a picnic. Results of father involvement efforts in other contexts, such as Head Start, early literacy programs, and social welfare interventions indicate fathers are more satisfied with activities that provide information on how to support their child's development through active participation in general activities like running errands, cooking, games, and sporting events (Cullen, Cullen, Band, Davis, & Lindsay, 2011; Fabiano et al., 2009; Maxwell, Scourfield, Featherstone, Holland, & Tolman, 2012; National Deaf Children's Society, 2006; Raikes & Bellotti, 2006).

Be patient, persistent, and proactive.

When we are at appointments with our wives and they are crying, we are supposed to be the rock and support her, not be the one crying. So, in order be the 'man', we close ourselves off from saying anything to avoid letting the emotion out.

Some evidence suggests that fathers may initially be reluctant participants in the early intervention process for a variety of reasons; for example, they may view themselves as inadequate parents (Maxwell et al., 2012). This is particularly true when the first child born to a couple is DHH and the first-time learning of parenting skills is further complicated with extra visits, technology, and

communication choices, etc. However, even if the parents have other children, the experience of parenting a child with hearing loss is unique and challenging.

Maxwell et al. (2012) suggest that providers be persistent, even when initially brushed off, in consulting fathers about what type of supports they need. A study of fathers in a parent support program also noted this.

You do have to do the drip-drip approach because a lot of fathers will say initially, 'I leave her (the mother) to deal with all that type of thing. What you put on is important, and it's a matter of consulting with them to find out what they want. It's no good just thinking of an idea and then just expecting them to come in. If you put something on that's a kind of like a hook, then they'll come in. If you put something on that dads would never dream of doing, then they'll not come in. (Cullen et al., 2011, p. 493)

Treat fathers as equal partners in parenting when sharing information.

They see me in the grocery store and say, 'Oh, you two are on your own this week? Well, don't worry, you'll make it until mom's back in town', like I can't take care of my own kid by myself. It's really kind of sexist.

Often doctors' attention and eye contact is given directly to the wife during appointments. We might as well go sit out in the waiting room.

Recent national dialogues in the United States challenge the concept of reverse-sexism and question whether men can experience it (Fabello, 2015). However, when it comes to parenting, there is still a tendency for professionals to enter the early intervention process with pre-conceived notions about fathers and their role. This may impact how professionals interact with families (Maxwell et al., 2012; Muñoz et al., 2015).

In *Men are from Mars, Women are from Venus: The Classic Guide to Understanding the Opposite Sex*, Gray (2004) asserts that men and women are so different in their communication needs that they are on different planets. He noted that men cope with stressful situations by withdrawing from conversations while women prefer to talk about the sources of their stress. Also, mothers and fathers of children who are DHH may not navigate the grief cycle in the same manner or on the same timeline (Luterman, 2006). Consequently, EHDI providers must consider their communication approach when sharing information with fathers. In addition to cultural influences on men's communication, a substantial amount of research has identified the concept of mothers functioning as gatekeepers of information and access to children for fathers (Allen & Hawkins, 1999; De Luccie, 1995; Sano, Richards, & Zvonkovic, 2008). Professionals must consider methods for sharing information directly with fathers rather than relying on mothers to convey it.

In addition to assumptions and biases being a potential barrier to sharing information with fathers and involving them in the decision-making process, communication logistics can play a role (Ancell, Bruns, & Chitiyo, 2018). EHDI providers should consider alternate forms of direct communication with fathers such as texts and email. When it comes to sharing coaching and intervention strategies, video modeling and interactive remote technologies such as Skype or Facebook Live can be helpful tools to allow fathers to interact directly with providers and receive information that is not filtered through the child's mother.

Build a team that is knowledgeable, current, and unbiased about ALL aspects of the child.

She (the professional) said our baby may never talk and would probably have a very limited capability in life because she was deaf. That was devastating and we have never forgotten it. I'd love to introduce her to [our daughter] now.

This dad-endorsed strategy is consistent with recommended EHDI practices. The EHDI family-centered early intervention literature strongly advocates that the team include professionals with expertise in all aspects related to deafness; in particular, the potential impact of childhood hearing loss on all aspects of child development. Additionally, because a large number of infants and toddlers who are DHH have additional disabilities, the need for specialized personnel with expertise that matches the child's potential challenge areas is key (Moeller, Carr, Seaver, Stredler-Brown, & Holzinger, 2013; Muse et al., 2013; Sass-Lehrer, 2004). It is interesting that fathers of children who are DHH appear to clearly understand this need. One study of father involvement found that fathers of children in an early intervention program indicated knowing what was involved in the program and knowing that the interventionist is trained were the two most important factors in participating (Tully et al., 2017). It is possible that fathers' involvement may be influenced by being clear about the qualifications of their child's team and what expertise each member has to offer.

Remember different isn't wrong.

I think I scared the early intervention team with how physical I was with [our daughter]. Now we play Monkeys' Jumping on the Bed and keep it a secret from Mom—it's our game.

As long as she's still breathing when mom comes home, I've done my job.

Maxwell et al. (2012) noted that fathers may be concerned that early intervention programs may dictate how they should parent and fathers feared they would not be able to live up to these expectations; they were intimidated. One father in Pedersen and MacIver's (2013) study recalled a memory of walking into his home during an early intervention visit where his wife and three female providers were present. He enthusiastically greeted his infant and tossed her up in the air, as was his practice. There was

a collective gasp from the female providers. The father remarked, “I knew I had done something wrong, so I just went out in the kitchen and tried to look busy.”

Family-centered services must consider how the professionals can leverage each unique family system and individual family member strengths. EHDI professionals should challenge their assumptions about what good parenting is and examine whether their views may be biased toward behaviors that mothers typically exhibit.

Build confidence and competence.

You don't need to know everything and don't be afraid to ask questions.

As mentioned above, fathers may tend to feel inadequate in parenting their child who is DHH. Sass-Lehrer (2004) recommends that a goal of family-centered EHDI services should be to support both confidence and competence in parents. Self-efficacy theory (Bandura, 1997) supports the premise that the more an individual believes they can successfully accomplish a task, the greater the likelihood that he will want to engage in the task and persist in its execution. When professionals facilitate fathers' enjoyment of father-child relationships, it leads to increased father engagement in early intervention (Anderson, Aller, Piercy, & Roggman, 2015). There is also evidence indicating that fathers of children who are DHH who have higher levels of self-efficacy also have higher levels of involvement in their child's early intervention program (Ingber & Most, 2012).

How to build confidence and competence in fathers begins with the previous strategy of remembering that different isn't wrong, and by viewing fathers' styles as unique and complimentary to mothers' rather than opposing or contradictory.

It seems that in the context of family support work, the most effective interventions adopt a strengths-based approach which focus upon the important contributions fathers make to their children's lives, where workers are positive about the father's ability...emphasize the father's existing skills and use solution-focused thinking to develop their skills and build confidence. (Maxwell et al., 2012, p. 165–166)

One example from an early intervention home visit comes from the author's experience as an EHDI professional (Pedersen, personal communication, January 29, 2019).

One mother showed me a 2-minute video taken on her cell phone of a father just playing with his infant daughter who has a dual-sensory disability. Within that two minutes, the father used several instances of evidence-based communication strategies: proximity, turn taking, waiting, reinforcement, and multiple means of sensory input. While he was not yet comfortable interacting with his child like this in front of me, I was able to use this video to

show the father each of these instances and build his confidence and competence by illustrating how his natural interactions were exactly what his daughter needed.

Facilitate novice to expert father support.

It's the fear of the unknown that is the biggest thing.

Probably one of the biggest differences is I was worried if she would ever be a country music star or how would she go waterskiing or play sports with hearing aids. I don't think (my wife) worried about those things as much.

Although offering a combination of whole family and father-specific activities is recommended, one thing is clear—fathers of children with disabilities benefit from accessing peer support (Konstantareas & Homatidis, 1992). In the 2013 International Consensus Statement on best practices in family-centered early intervention for children who are deaf or hard of hearing, Moeller et al. (2013) state that parent to parent support is essential for family well-being. Many early intervention programs offer parent to parent connections, but those specific to fathers are rare. Fathers may again be reluctant to reach out to another father in the same way that mothers do (Pelchat et al., 2003) and the type of social support needed may also be different than mothers (Zaidman-Zait, et al., 2018). For example, in a case study of two fathers of children with hearing loss, laughter was frequently used to characterize the fathers' parenting experiences and humor appeared to mediate stress and support the fathers' transition to confident parent (Pedersen & Spooner, 2017). Recognizing gender differences can be helpful in implementing this strategy as well; while women tend to connect with others simply by talking, men develop relationships with each other through activities (Tannen, 1990). Providers should consider this and be intentional and creative when planning group family activities and support opportunities for fathers to connect organically, rather than through a traditional support group approach.

Tools for Implementing These Strategies

Avoiding subconscious bias necessitates EHDI providers be intentional about the inclusiveness of their communication and addressing both mothers and fathers equally. Three tools are offered here to assist providers and agencies in self-assessing their practices relative to attending to fathers.

- The Checklist for Assessing Adherence to Family-Centered Practices (Wilson & Dunst, 2005) has been adapted with permission to include a focus on fathers in Appendix A.
- The Dakota Father Friendly Assessment (DFFA) tool was developed for use in Head Start programs. The DFFA (White, Brotherson, Galovan, Holmes, & Kampmann, 2011) consists of 33 self-report items designed to measure the constructs of staff biases, staff attitudes, staff behaviors, organizational attitudes, and organizational

behaviors. A list of the items has been reproduced with permission in Appendix B.

- The Father Friendly Check-Up™ (NFI, 2016) is designed for organizations and programs serving fathers to assess their efforts in the four categories of Leadership Development, Organizational Development, Program Development, and Community Engagement. The checklist is available for free download at <https://www.fatherhood.org/ffcu>.

Conclusion

This article has offered considerations and practical strategies EHDI professionals can use to refine their family-centered practices to better include fathers. In seeking to meet the needs of families with many different characteristics, one simple constant must be at the forefront of the EHDI provider's mind: A child cannot have too many people equipped and empowered to support their healthy development. Working to support all members of the child's family increases the odds that children who are DHH and their families can enjoy every opportunity to achieve their desired outcomes.

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Appendix A

Family-Centered/Father-Focused Practices Checklist

Linda L. Wilson & Carl J. Dunst

Staff Member _____

Context _____

Observer/Coach _____

Date(s) _____

Rating Scale				
		1 = Yes, practice was used	3 = Practice not used, opportunity missed	
		2 = Practice was partially, sometimes done	4 = NA, no opportunity to observe the practice	
		In what way was each practice used?	Rating	Example/Comment/Reflection
Interpersonal Skills	Fater-Focused Interpersonal Skills	Communicate clear and complete information in a manner that matches the family's style and level of understanding.		
		Communicate clear and complete information in a manner that matches the father's style and level of understanding.		
		Interact with the family in a warm, caring, and empathetic manner.		
		Interact with the father considering the unique communication preferences of fathers.		
		Treat the family with dignity and respect without judgement.		
		Treat the father with dignity and respect and without judgement—remember different isn't wrong.		
Asset-Based Attitudes	Father-Focused Asset-Based Attitudes	Communicate to and about the family in a positive way.		
		Communicate with a focus on the father about the family in a positive way.		
		Honor and respect the family's personal and cultural beliefs and values.		
		Honor and respect the father's personal and cultural beliefs and values.		
		Focus on individual and family strenghts and values.		
		Focus on individual father and family strengths and values.		
		Acknowledge the family's ability to achieve desired outcomes.		
		Acknowledge the father's value in the ability to achieve desired outcomes.		
Family Choice and Action	Father-Focused Choice and Action	Work in partnership with parents/family members to identify and address family-identified desires.		
		Work in partnership with fathers to identify and address family-identified desires.		
		Encourage and assist the family to make decisions about and evaluate the resources best suited for achieving the desired outcomes.		
		Encourage and assist the father to contribute in decisions and to evaluate the resources best suited for him to participate in achieving desired outcomes.		
		Seek and promote ongoing parent/family input and active participation regarding desired outcomes.		
		Seek and promote ongoing father input and active participation regarding desired outcomes.		
		Encourage and assist the family to use existing strengths and assets as a way of achieving desired outcomes.		
		Encourage and assist the father to use existing strengths and assets as a way of achieving desired outcomes.		
		Provide family participatory opportunities to learn and develop new skills.		
		Provide father participatory opportunities to learn and develop new skills.		

Family-Focused Practitioner Responsiveness	Father-Focused Practitioner Responsiveness	Assist the family to consider solutions for desired outcomes that include a broad range of family and community supports and resources.		
		Assist the father to consider solutions for desired outcomes that include a broad range of family and community supports and resources.		
		Support and respect family member's decisions.		
		Support and respect father's decision		
		Work with the family in a flexible and individualized manner.		
		Work with a father in a flexible and individualized manner.		
		Offer help that is responsive to and matches the family's interests and priorities.		
		Offer help that is responsive to and matches the father's interests and priorities.		
		Assist the family to take a positive, planful approach to achieving desired outcomes.		
		Assist the father to take a positive, planful approach to achieving desired outcomes.		

Note. Adapted with permission from Wilson, L. L., & Dunst, C. J. (2005). Checklist for assessing adherence to family-centered practices. *CASE Tools: Instruments and Procedures for Implementing Early Childhood and Family Support Practices*, 1(1), 1–6.

Appendix B
The Dakota Father Friendly Assessment
(White, Brotherson, Galovan, Holmes, & Kampmann, 2011)

SA = Strongly agree; A = Agree; N = Neither agree nor disagree; D = Disagree; SD = Strongly disagree

1. Our program's mission statement should include services to fathers/father figures SAANDSD
2. Fathers should be involved in the orientation and enrollment process SAANDSD
3. It is important that fathers attend school functions SAANDSD
4. It is important to have program activities for the whole family SAANDSD
5. Mothers are more committed to the care and well-being of their children than most fathers SAANDSD
6. Fathers bring unique strengths to parenting that meet a child's growth and development needs SAANDSD
7. Mothers put more thought into program projects and activities SAANDSD
8. I find it hard to let fathers be in charge after assigning them a task SAANDSD
9. Fathers not living in the home should also be sent announcements of program activities SAANDSD
10. My feelings about the value of fathering has been influenced by negative experiences with men SAANDSD
11. I encourage mothers to support fathers, even if involvement isn't desired (abuse cases omitted) SAANDSD
12. I actively recruit fathers for assistance with program services SAANDSD
13. I usually don't interact with fathers who come with mothers SAANDSD
14. I make an effort to have fathers sign family partnership agreements SAANDSD
15. I make an effort to have fathers take part in the IEP or IFSP process SAANDSD
16. I try to schedule home visits when both parents are available SAANDSD
17. The message I give to fathers in that their role is critical to their child's development SAANDSD
18. Partnership agreements reflect the father's interests & concerns as well as the mother's SAANDSD
19. During program projects, I tend to assist fathers more so they get things done the way I want them SAANDSD
20. I tend to judge how good a father is by his child's appearance SAANDSD
21. All Head Start staff at our center believe in the need for a positive attitude toward working with fathers SAANDSD
22. All Head Start staff at our center believe they should provide the same support for fathers as mothers SAANDSD
23. All staff at our center believe they should provide recognition for fathers' efforts and successes SAANDSD
24. All of our staff believe it is important to facilitate interaction with fathers SAANDSD
25. All of our staff believe fathers should participate in scheduled parent-teacher meetings SAANDSD
26. All of our staff believe input should be sought from fathers about what they want from Head Start SAANDSD
27. Our Head Start center provides regular training on father involvement (at least semiannually) SAANDSD
28. Our Head Start center provides staff with books and resources for and about fathers SAANDSD
29. All of our staff are knowledgeable about fathering behaviors and attitudes SAANDSD
30. Our staff actively recruit male staff members and facilitators for father's events/groups SAANDSD
31. Our staff actively recruit fathers for the parent advisory board, board of directors, etc. SAANDSD
32. Our center's approach to father involvement has tried to engage most fathers in program activities SAANDSD
33. All staff try to identify a primary father figure to encourage involvement in the child's life SAANDSD

Note. Adapted with permission from: White, J. M., Brotherson, S. E., Galovan, A. M., Holmes, E. K., & Kampmann, J. A. (2011). The Dakota father friendly assessment: Measuring father friendliness in head start and similar settings. *Fathering, 9*(1), 22–44

Pediatric Amplification Management: Parent Experiences Monitoring Children’s Aided Hearing

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Abstract: Objective: Investigate parents’ experiences monitoring aided hearing for children who use hearing aids, bone conduction hearing aids, and cochlear implants.

Design: A cross-sectional survey design, using three survey instruments, was used to collect parent data.

Study Sample: A total of 178 parents of children birth to six years were included in the analysis (81 hearing aid; 61 cochlear implant; 36 bone conduction hearing aid).

Results: Surveys explored hearing device use and monitoring. Variability was found for hearing aid use and many parents reported being unaware if their child’s device had data logging capability. Parents varied widely in how often they checked hearing device function, and approximately half did not have access to loaner hearing devices when repairs were required. Variance was observed in how often professionals explored how children are hearing at home through use of parent-report questionnaires, and related to audiology-specific services aimed at monitoring and maintaining audibility during routine appointments (e.g., checking program settings when new earmolds are received, frequency of earmold replacement, checking data logging).

Conclusion: This study revealed variability in hearing device use and monitoring for audibility by professionals and parents. Implications from this study suggest parent-professional partnerships would benefit from better understanding of barriers/facilitators for parent learning and implementation of key monitoring tasks.

Acronyms: AAA = American Academy of Audiology; BCHA = bone conduction hearing aids; CI = cochlear implant; HA = hearing aid; FM = frequency modulation; RECD = real-ear-to-coupler-difference

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Early identification of hearing loss through newborn screening has become a standard of care in the United States (Centers of Disease Control and Prevention, 2017). Early screening allows for intervention within the first few months of life (Joint Committee on Infant Hearing, 2007), giving parents an opportunity to access needed services. For children learning to communicate using spoken language, consistent auditory access to speech sounds using hearing technology is necessary to achieve optimal language outcomes (Tomblin et al., 2015). Both audiologists and parents play critical roles in monitoring aided hearing and when there are gaps in managing hearing care, audibility is inconsistent.

Appropriate hearing device programming is fundamental for audibility. Audiologists program hearing devices specifically for each child based on their individual hearing

needs and it is necessary to monitor device settings over time. For example, children who use hearing aids are fit with new earmolds as they grow because the size of their ear canal increases. To accommodate for physical changes, a measurement (called real-ear-to-coupler-difference [RECD]) should be completed when new earmolds are fit to the child. Hearing aid programming adjustments, based on the child’s current hearing thresholds and RECD, are then made to maintain sufficient sound pressure levels for audibility (American Academy of Audiology [AAA], 2013; Seewald & Scollie, 2003). Even when hearing devices are programmed appropriately, hearing in noisy environments can be challenging. The use of a personal frequency modulation (FM) system in conjunction with hearing devices improves audibility by helping children access speech when listening in more adverse environments (AAA, 2008).

Daily hearing device management is also fundamental for audibility. Parents are responsible for having their children wear their devices and for checking that devices are functioning. Young children are in a critical language learning period and device use of less than 10 hours per day has been found to negatively affect language development (Tomblin et al., 2015). Parents have reported that various child factors (e.g., child behavior) and parent factors (e.g., frustration, depression) interfere with how often children wear their hearing devices (Caballero et al., 2017; Isarin et al., 2015; Muñoz et al., 2016; Walker et al., 2013), and wide variability has been found in average hours of use (Muñoz et al., 2015; Walker et al., 2013). Data logging is a feature built into most hearing devices. Parents and audiologists can use data logging to routinely monitor hours of use and to help recognize when device problems occur. Data logging allows the audiologist to view the average amount of time the child is wearing the device. Even when children wear their hearing devices consistently, however, audibility is compromised if the devices are not functioning. Parents have reported a lack of training in how to check devices and/or not having needed tools (Muñoz, Blaiser, & Barwick, 2013; Muñoz, et al., 2015), and this can result in infrequent monitoring of device function (Burkhalter, Blalock, Herring, & Skaar, 2011; Isarin et al., 2015; Muñoz et al., 2013; Watermeyer, Kanji, & Sarvan, 2017).

Routine monitoring by audiologists and parents is necessary to determine hearing device benefit and to identify changes or problems in audibility that need attention. Parents' observations of how their child is functioning at home and in other environments can be obtained by using questionnaires, and audiologists can assess aided speech perception during monitoring appointments (AAA, 2008, 2013). Parents can also use the Ling-Six sound test every day to check that their child is perceiving speech sounds represented across the frequency range (AAA, 2008). When device malfunctions occur, loaner hearing devices can be provided while the child's device is out for repair, so audibility is not compromised. Given that audibility can be affected by multiple factors (e.g., device use, device function) that ultimately influence child outcomes, understanding parents' experiences can provide important insights about how audiologists and parents can more effectively partner in this journey. The purpose of this study was to investigate parents' experiences monitoring aided hearing for children who use hearing aids, bone conduction hearing aids, and cochlear implants.

Method

This study used a cross-sectional survey design to explore parent experiences monitoring aided hearing. Survey responses were anonymous, and Institutional Review Board approval at Utah State University was obtained prior to conducting this study.

Participants and Procedures

Parents of young children birth to six years of age who use hearing devices (i.e., hearing aids, bone conduction hearing aids, cochlear implants) and who were proficient in English were recruited to participate in the study from February to November 2017 through parent support websites and social media (e.g., heartolearn.org, handsandvoices.org, agbell.org, Facebook groups). Data collection was completed using Qualtrics, an online survey software tool. Because this distribution method was designed to target the population of interest broadly, it was not possible to estimate the number of people reached to calculate a response rate. Completed surveys were received from 210 parents in 37 states and 8 countries. Thirty-two surveys were excluded (30 children were older than six years; 2 children were not using hearing devices [1 hearing aid, mild degree; 1 cochlear implant]); 178 surveys were analyzed. Participant demographic information can be seen in Table 1. Responses were primarily received from mothers (93%, 166/178) and few reported that their children have a caregiver who has had a hearing loss since childhood (9%, 16/178).

Survey Instruments

Three survey instruments (Hearing Aid [HA; 25 items]; Cochlear Implant [CI; 24 items]; Bone Conduction Hearing Aid [BCHA; 23 items]) were developed by the first and third authors. Items were developed based on professional guidelines (e.g., AAA, 2013) to capture fundamental practices for hearing technology monitoring. Each survey had four sections: Information About Your Child, Information About You, Device Use, and Device Monitoring.

Data Analysis

Descriptive data analysis was completed using SPSS (Version 25), including measures of central tendency to identify variance in parent experiences. Analysis of variance was used to investigate factors that may be associated with parent-reported typical hours of daily hearing device use: length of time with hearing device (i.e., 12 months or less, 13 to 24 months, more than 24 months); device type (i.e., hearing aid, bone conduction hearing aid, cochlear implant), and child age (i.e., early intervention age [0 to 36 months]; preschool age [37 to 60 months]; early elementary age [61 months and older]). Child age groupings reflected systems in the United States that support children and families based on chronological age. Two parents reported 24 hours per day of device use (HA = 1, CI = 1). Although some pediatric patients sleep with their devices on for safety or comfort, this is not common; therefore these responses were not included in hearing aid use analyses to better observe trends. The data were split for analysis (i.e., hearing aids, cochlear implants, bone conduction hearing aids) to explore differences among parents on items that may be related to device type. The sample size varies by survey item as parents were allowed to skip questions. Content analysis was completed for the open-ended questions to identify emergent themes. Appendix A details the number of

Table 1
Participant Demographics

Child and Caregiver Information	HA (<i>n</i> = 81)		CI (<i>n</i> = 61)		BCHA (<i>n</i> = 36)	
	% (<i>n</i>)	<i>M</i> (<i>SD</i>)	% (<i>n</i>)	<i>M</i> (<i>SD</i>)	% (<i>n</i>)	<i>M</i> (<i>SD</i>)
Child						
Age in months		41 (23.81)		47 (18.76)		44 (23.40)
Months since fitting		20 (18.50)		27 (15.89)		25 (18.11)
Typical hours of use per day		10 (02.52)		11 (02.49)		10 (02.92)
Uses hearing aids in both ears	78 (63)		89 (54)		42 (15)	
Degree of hearing loss*						
Mild	12 (10)					
Moderate	49 (40)					
Severe	24 (20)					
Profound	12 (10)					
Unsure	1 (1)					
Has additional disabilities	27 (22)		16 (10)		31 (11)	
Caregiver						
Age in years		35 (5.35)		35 (5.12)		36 (06.62)
Relationship to child – mother	90 (73)		95 (58)		97 (35)	
Child has a caregiver with hearing loss since childhood	11 (9)		7 (4)		8 (3)	
Race						
White	88 (71)		85 (52)		78 (28)	
Prefer not to answer	5 (4)		3 (2)		3 (1)	
Educational level						
High school diploma	6 (5)		8 (5)		3 (1)	
Some college	12 (10)		10 (6)		17 (6)	
Associates degree	12 (9)		8 (5)		22 (8)	
Bachelor's/graduate degree	70 (57)		74 (45)		58 (21)	

Note. HA = hearing aid; CI = cochlear implant; BCHA = bone conduction hearing aid. *Question only in HA survey

participants who responded, the number of statements per question, and provides examples of challenges parents experience while monitoring aided hearing. Appendix B contains advice for professionals that emerged from our findings.

Results

Parent experiences reported were for children from 3 to 83 months of age (HA [Mdn = 44, range: 3–83], BCHA [Mdn = 49, range: 3–76], CI [Mdn = 48, range: 14–78]). The children had been wearing their hearing devices for 1 to 68 months (HA [M = 20, Mdn = 15, range: 1–68], BCHA [M = 25, Mdn = 23, range: 2–68], CI [M = 27, Mdn = 24, range: 1–64]).

Hearing Device Use

Parent-reported typical hours of daily hearing device use varied for all devices (HA [Mdn = 10, range: 4–14], CI [Mdn = 12, range: 5–16], and BCHA [Mdn = 10, range: 4–16]).

Analysis of variance was used to explore three factors (i.e., length of time with device, child age groups, device type) to determine their association with hearing device use (see Table 2). All three factors had statistically significant main effects on parent-reported typical hours of daily hearing device use. First, children who have had their devices more than two years used them, on average, 1.5 hours more per day than children who have had them less than two years; length of time with device $F(2, 171) = 7.053, p = .001$. Second, children in preschool and early elementary school used their hearing devices, on average, 1.68 hours more per day than early intervention age children; for child age $F(2, 171) = 9.888, p = .000$. Third, children who use cochlear implants used their hearing devices, on average, 1.4 hours more per day than children who use hearing aids or bone conduction hearing aids; device type $F(2, 171) = 5.662, p = .004$.

There were not statistically significant main effects on parent-reported typical hours of daily device use for degree

Table 2
Effect of Child Age, Device Type, and Length of Time with Device on Parent-Reported Typical Hours Hearing Device Use

Device Use Factors	<i>n</i>	<i>M (SD)</i>	95% CI	<i>p</i>
Child Age				< 0.001*
Early Intervention Age (0–35 months)	73	9.58 (2.59)	8.97, 10.18	
Preschool Age (36–60 months)	46	11.00 (2.53)	10.25, 11.75	
Early Elementary Age (> 60 months)	53	11.51 (2.47)	10.83, 12.19	
Device Type				0.004*
Hearing Aid	78	10.08 (2.52)	9.51, 10.65	
Bone Conduction Hearing Aid	35	10.06 (2.92)	9.05, 11.06	
Cochlear Implant	59	11.47 (2.49)	10.82, 12.12	
Length of Time with Device				0.001*
12 months or less	58	10.09 (2.50)	9.43, 10.74	
13–24 months	49	9.86 (2.59)	9.11, 10.60	
More than 24 months	65	11.49 (2.64)	10.84, 12.15	

* statistical significance

of hearing loss for children who use hearing aids $F(5, 171) = 1.258, p = .284$, or for children who have additional disabilities $F(2, 171) = .517, p = .597$.

Data logging provides a means for audiologists and parents to monitor hearing device use. Parents were asked if their child’s device had data logging capabilities. Many parents did not know if their child’s device had data logging (HA [30%, $n = 23$]; BCHA [43%, $n = 15$]; CI [19%,

$n = 11$]). Parents of children with CIs indicated devices had data logging (70%, $n = 41$) more often than parents of children with HAs (37%, $n = 29$) and BCHA (26%, $n = 9$). For children that have hearing devices with data logging, parents were asked how often (i.e., *never, sometimes, often, always*) data logging is discussed; often and always were combined to better see trends. Less than half of the parents of children who use HAs or BCHA reported that audiologists often or always talk about hours of use recorded by data logging (HA: [45%, 14/31]; BCHA: [33%, 3/9]); parents of children who use CIs reported more frequent discussions (60%, 25/42).

Parents reported how often each professional, when applicable, talked with them about hearing device use (i.e., *never, sometimes, often, always*). *Often* and *always* were combined to better see trends. For each device type and for all professionals listed, there was variability in frequency, with many parents reporting device use is only discussed *sometimes* or *not at all* (see Table 3).

Loaner hearing device. Parents reported whether or not their child has received a loaner hearing device to use when their device was being repaired. For children who have had their device repaired, half of the parents or more reported never receiving a loaner (HA: [52%, 16/31]; BCHA: [68%, 15/22]; CI: [50%, 13/26]).

Replacement equipment/earmolds. Hearing device use can be affected when custom earmolds do not fit properly and when equipment needed for device function needs to be replaced. Parents of children who use hearing aids and have had them for more than a year were asked how

Table 3
Frequency Professionals Talk with Parents about Device Use

Professional	% (<i>n</i>)									
	HA	Never			Sometimes			Often/Always		
		BCHA	CI		HA	BCHA	CI	HA	BCHA	CI
Audiologist	7 (6)	--	5 (3)	38 (31)	24 (8)	33 (20)	54 (44)	77 (26)	62 (38)	
SLP	23 (14)	17 (5)	8 (4)	18 (11)	28 (8)	49 (25)	60 (37)	55 (16)	43 (22)	
Teacher	38 (21)	30 (7)	33 (14)	29 (16)	39 (9)	40 (17)	34 (19)	30 (7)	27 (12)	
EI	22 (11)	16 (4)	15 (6)	14 (7)	16 (4)	23 (9)	64 (32)	68 (17)	63 (25)	
Physician	38 (31)	37 (13)	25 (15)	42 (34)	40 (14)	9 (5)	20 (16)	23 (8)	24 (14)	

Note. HA = hearing aids; BCHA = bone conduction hearing aids; CI = cochlear implant; SLP = speech-language pathologist; EI = early interventionist

Table 4
Frequency of Earmold Replacement During Previous Year and Shipping Time in Weeks

Age Groups	Earmold Replacement % (<i>n</i>)					Shipping Time % (<i>n</i>)			
	Never	Once	Twice	3 times	4 times or more	1 week	2 weeks	3 weeks	4 weeks
EI	10 (3)	14 (4)	24 (7)	24 (7)	28 (8)	14 (4)	45 (13)	14 (4)	17 (5)
Preschool	13 (2)	40 (6)	40 (6)	7 (1)	--	7 (1)	60 (9)	27 (4)	--
EE	6 (1)	56 (9)	6 (1)	19 (3)	13 (2)	31 (5)	31 (5)	31 (5)	--

Note. EI = early intervention; EE = early elementary

many times during the past year their child's earmolds were replaced (see Table 4). All parents were asked about the typical shipping time to get the new earmolds and replacement components for devices (see Table 4). Five parents indicated replacement earmolds were not needed or their child does not use earmolds.

Hearing Device Monitoring

Parent confidence. Parents reported how confident they felt monitoring the hearing devices (0 = *not confident at all*; 100 = *completely confident*). For HAs, confidence was variable among parents, with the lowest confidence reported for knowing the HA settings are appropriate ($M = 47$, $SD = 32.85$); more parents were confident in determining when to replace earmolds ($M = 72$, $SD = 26.68$) and batteries ($M = 73$, $SD = 26.57$). For BCHA and CIs, most parents reported confidence for items queried: when to replace batteries (BCHA [$M = 78$, $SD = 22.78$]; CI [$M = 89$, $SD = 13.73$]); knowing device is functioning properly (BCHA [$M = 96$, $SD = 14.33$]; CI [$M = 99$, $SD = 3.77$]); interpreting indicator lights (BCHA [$M = 73$, $SD = 30.83$]; CI [$M = 88$, $SD = 18$]); and monitoring external equipment (BCHA [$M = 79$, $SD = 27.71$]; CI [$M = 88$, $SD = 19.85$]). For CI parents, there was more variability for confidence in listening to the microphone ($M = 75$, $SD = 31.31$).

All parents were asked about their confidence related to performing a speech sound check (i.e., Ling-Six sound). Parents of children who use CIs were more confident than parents of children who use HAs and BCHA (see Figure 1).

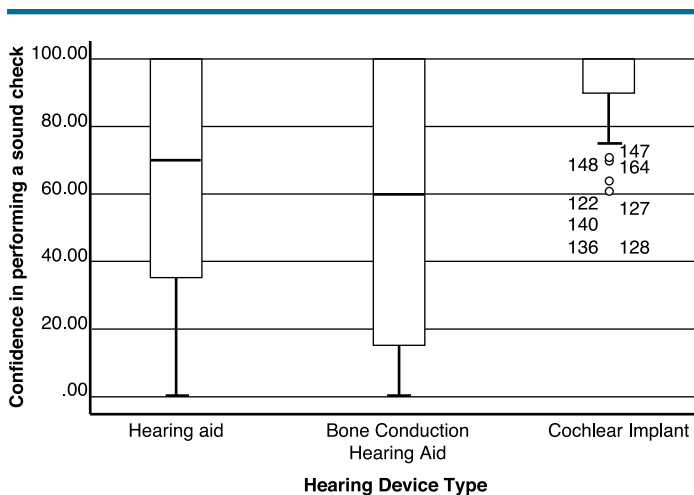


Figure 1. Parent confidence in performing a speech sound check (median and interquartile ranges [IQR]). Median confidence for parents of children who use hearing aids was 70 ($n = 70$), bone conduction hearing aids was 60 ($n = 30$), and cochlear implants was 100 ($n = 56$). The thick horizontal line within the box represents the median, the vertical lines above and below the box represent the IQR, and the circles and asterisks below the vertical line represent the outliers or the cases that were less confident.

Frequency of parent monitoring. Table 5 shows the frequency parents reported monitoring the condition and function of hearing devices (i.e., *when needed*, *never*, *every few weeks*, *weekly*, *daily*). Frequency of parent monitoring for all items varied for all devices.

Frequency of professional monitoring. Parents reported how often each professional, when applicable, asked them to complete a questionnaire to explore aided benefit in daily life (i.e., *never*, *sometimes*, *often*); see Table 6. Few parents indicated that professionals often ask them to complete questionnaires for any device.

For children who use hearing aids, device settings need to be monitored and adjusted when new earmolds are received. Parents reported how often hearing aid settings were checked when their child was fit with new earmolds (i.e., *never*, *sometimes*, *often*, *always*); *often* and *always* were combined for ease in observing trends. The majority reported this *often* or *always* occurs (71%, $n = 56$), some reported it *sometimes* occurs (12%, $n = 9$) or *never* occurs (4%, $n = 3$), and some parents *did not know* (13%, $n = 10$).

Table 5
How Often Parents Check Hearing Device Function

Device	Component Checked	% (n)				
		When needed	Never	Every few weeks	Weekly	Daily
HA (n = 75)						
	Batteries	23 (17)	--	1 (1)	35 (26)	41 (31)
	Sound Quality	15 (11)	21 (16)	16 (12)	15 (11)	33 (25)
	Wax Blockage	4 (3)	4 (3)	4 (3)	15 (11)	73 (55)
	Physical Condition	7 (5)	4 (3)	3 (2)	16 (12)	71 (53)
	Speech sound check	17 (13)	32 (24)	13 (10)	20 (15)	17 (13)
BCHA (n = 35)						
	Batteries	37 (13)	--	6 (2)	17 (6)	40 (14)
	Microphone Quality (n = 34)	29 (10)	44 (15)	12 (4)	9 (3)	6 (2)
	External Equipment	20 (7)	--	9 (3)	9 (3)	63 (22)
	Speech Sound Check	17 (6)	54 (19)	11 (4)	11 (4)	6 (2)
CI (n = 56)						
	Batteries	25 (14)	2 (1)	2 (1)	2 (1)	70 (39)
	Microphone Quality	20 (11)	30 (17)	30 (17)	11 (6)	9 (5)
	External Equipment (n = 55)	29 (16)	--	2 (1)	22 (12)	47 (26)
	Speech Sound Check	27 (15)	11 (6)	14 (8)	25 (14)	23 (13)

Note. HA = hearing aids; BCHA = bone conduction hearing aids; CI = cochlear implant; SLP = speech-language pathologist; EI = early interventionist.

Table 6
Frequency Professionals Asked Parents to Complete a Questionnaire to Explore Benefit

Professional	% (n)								
	Never			Sometimes			Often		
	HA	BCHA	CI	HA	BCHA	CI	HA	BCHA	CI
Audiologist	69 (52)	89 (31)	56 (31)	23 (17)	3 (1)	22 (12)	8 (6)	9 (3)	22 (12)
SLP	67 (39)	68 (21)	49 (24)	28 (16)	16 (5)	31 (15)	5 (3)	16 (5)	20 (10)
Teacher	76 (41)	81 (21)	72 (28)	20 (11)	4 (1)	23 (9)	4 (2)	15 (4)	5 (2)
EI	56 (28)	68 (17)	43 (16)	34 (17)	12 (3)	41 (15)	10 (5)	20 (5)	16 (6)

Note. HA = hearing aids; BCHA = bone conduction hearing aids; CI = cochlear implant; SLP = speech-language pathologist; EI = early interventionist.

For children who use hearing aids, device settings need to be monitored and adjusted when new earmolds are received. Parents reported how often hearing aid settings were checked when their child was fit with new earmolds (i.e., *never*, *sometimes*, *often*, *always*); *often* and *always* were combined for ease in observing trends. The majority reported this *often* or *always* occurs (71%, $n = 56$), some reported it sometimes occurs (12%, $n = 9$) or *never* occurs (4%, $n = 3$), and some parents did not know (13%, $n = 10$).

Personal FM System

Parents were asked if their child has a personal FM system. The majority of children who use CIs had an FM system (79%, 44/56), approximately half with BCHAs (57%, 20/35), and one-third with HAs (39%, 29/75). For those who have an FM system, when applicable, parents indicated how often (i.e., *never*, *sometimes*, *often*) the device is used in different locations (see Table 7); parents reported using FM systems infrequently for all locations queried.

Table 7
Frequency of FM Use in Different Locations

Location	% (n)								
	Never			Sometimes			Often		
	HA	BCHA	CI	HA	BCHA	CI	HA	BCHA	CI
Home	63 (19)	65 (17)	69 (33)	30 (9)	23 (6)	29 (14)	7 (2)	12 (3)	2 (1)
School	44 (4)	64 (7)	56 (9)	22 (2)	9 (1)	--	33 (3)	27 (3)	44 (7)
Daycare	100 (8)	100 (7)	87 (13)	--	--	13 (2)	--	--	--
Car	67 (18)	76 (19)	71 (32)	26 (7)	24 (6)	27 (12)	7 (2)	--	2 (1)
Events	46 (12)	74 (17)	54 (23)	15 (4)	17 (4)	40 (17)	39 (10)	9 (2)	7 (3)

Note. HA = hearing aids; BCHA = bone conduction hearing aids; CI = cochlear implant.

Challenges and Advice

Parents responded to two open ended questions that queried challenges they experience and advice they have for professionals. For parent challenges, three primary themes emerged for all devices (see Appendix A). Child-related challenges were most commonly reported (HA 40%, BCHA 42%, CI 47%), although parent-related challenges (HA 35%, BCHA 29%, CI 27%) and device-related challenges (HA 19%, BCHA 25%, CI 24%) were also raised. The most frequently reported child-related challenge was the inability of the child to tell their parents when there was a problem (e.g., due to young age, non-verbal, multiple disability). A common parent-related challenge reported for HA and BCHA was difficulty knowing if their child was receiving benefit from the device, and for CI parents teaching others and getting enough support from others with management (e.g., teachers, other family members) was raised. The most common device-related challenge for all device types was not knowing if the device was working properly.

Three main themes emerged from parent advice offered for all devices (see Appendix B). Parent education and

support was the most common theme (HA 41%, BCHA 45%, CI 47%). Relationship with parents (HA 37%, BCHA 38%, CI 35%) and professional practices (HA 21%; BCHA 17%; CI 18%) were also themes addressed by parents. The most frequently reported aspect of parent education and support was to provide parents with detailed information. Parents want the professionals to be patient with them, trust them, and to listen to their thoughts and concerns. Parents also offered advice related to professionals' practice, suggesting that providers have information about support (e.g., parent groups), pediatric physicians, and routine data logging. They also want professionals to be patient and have fun with their children.

Discussion

Children who are using hearing technology to learn spoken language need consistent auditory access to speech sounds. Audibility is achieved by wearing appropriately functioning hearing devices during all waking hours. Parents play a central role in monitoring audibility for their children when they are young, and they rely on professionals to support and guide them in knowing how to effectively manage the devices on a daily basis. This study explored parent experiences monitoring aided hearing (i.e., hearing aids, bone conduction hearing aids, cochlear implants) for their children birth through six years of age. Important insights emerged from this study related to consistent audibility, and parent-professional partnerships for monitoring and managing audibility, for young children with hearing loss.

Consistent Audibility

How often children wear their devices has been found to affect language development, with children who use their devices more than 10 hours per day showing better language outcomes than children who use them less (Tomblin et al., 2015). Studies have found variability in hours of use particularly for young children, based on hearing aid data logging, and that parent report often overestimates hours of use (Walker et al., 2013; Muñoz et al., 2014). Parents of young children may have difficulty monitoring and reporting on typical hours of use for young children. For example, Caballero et al. (2017) found parents reported greater hours of use on "good" days. Parents may recall "good" days when they talk about hearing aid use with their audiologist. Device data logging is a tool that can help parents and audiologists identify when there is a problem with use that needs attention. In this study, many parents either did not know if their child's device had data logging or they reported this was not something the audiologist discusses with them.

Monitoring device function is also critical for consistent audibility. Hearing devices malfunction, and young children may not be able to report problems or may inconsistently report problems. As expressed by parents in this study "She is not quite old enough to articulate when there is a problem." For this reason, daily monitoring of the physical condition and sound quality is needed. To monitor device

function parents need special monitoring tools, as well as instruction and support to integrate this habit into their daily routine. Parents in this study generally reported confidence in monitoring tasks; however, the responses varied widely in how often they monitor device function. When devices do malfunction and need repair, children need loaners to maintain audibility while repairs are done; however, in this study only about half of the parents reported receiving a loaner for their child.

Parent-Professional Partnerships

The majority of parents of children with hearing loss have normal hearing (Mitchell & Karchmer, 2004) and are likely unfamiliar with childhood hearing loss, hearing devices, or management issues. In this study, few parents (7–11% based on device type) reported that their child has a caregiver with hearing loss since childhood. Parents have much to learn, and need guidance from professionals to attend to key monitoring practices, as expressed by these parents: “*Explain as much as possible in terms parents understand. For many parents this is a new journey and we are trying to learn what we can to make the best decisions possible and support our child,*” and “*Always share info with parents on how they can help their child. Write it down for them!*” Parents have also reported wanting professionals to check on them more often and to give them support in between clinical appointments (Caballero et al. 2017).

Monitoring audibility for children with hearing loss requires a team effort and collaboration among parents and professionals is critical. In this study, variance was observed in how often professionals talk with parents about hearing device use and explore how children are hearing at home through use of parent-report questionnaires. Variance was also observed related to audiology-specific services aimed at monitoring and maintaining audibility during routine appointments (e.g., checking program settings when new earmolds are received, frequency of earmold replacement, checking data logging). It is important for parents to have confidence in how their audiologist is monitoring their child, and to be aware of best practices so they can appropriately advocate for their child. As expressed by parents in this study: “*Data log even good wearers! We found a faulty cable that way,*” and “*Be as detailed as possible in your exams/ appointment.*” Hearing in a noisy environment is a known challenge for children with hearing loss, yet few children in this study have a personal FM system, and those that do, use it infrequently.

Parents need the support from professionals to help build confidence in their abilities, particularly as they adjust and learn new monitoring tasks. Professionals can develop and nurture a working alliance with parents to support effective device management by (a) assessing and addressing parent barriers, (b) jointly setting specific device management goals, (c) exploring anticipated challenges and potential solutions, and (d) providing accountability by checking in with parents and extending support as needed.

Parent-to-parent support can be another important mechanism for parents to help build their confidence and competence in monitoring aided hearing through compassion and understanding from others who have had similar experiences with their children. Collaboration among professionals on key monitoring components can support continuity of care and parent learning.

Research Implications

Findings from this study revealed important implications for future research. Better understanding of barriers/facilitators for parent learning and implementation of key monitoring tasks as well as educational and support delivery options could inform professional practices. Further research is needed to understand barriers, for professionals and parents, that exist related to personal FM/remote microphone use with young children. Furthermore, more research is needed that focuses on critical elements of implementation of patient-centered care for monitoring aided audibility for children using hearing devices.

Limitations

There were limitations to this study that should be noted. Even though the parent needs from this study reflected response from parents of young children, the majority of parents who responded were mothers who are White with a college education. The responses are self-report and may reflect bias that overestimates hearing aid use and monitoring practices.

Conclusions

This study investigated parents’ experiences monitoring aided hearing for children who use hearing aids, bone conduction hearing aids, and cochlear implants. Findings revealed variability in hearing device use, and monitoring for audibility by professionals and parents. Implications from this study suggest parent-professional partnerships would benefit from better understanding of barriers/facilitations for parent learning and implementation of key monitoring tasks.

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APPENDIX A
Parent Responses to Open Question about Their Challenges Monitoring Aided Audibility

Responses % (n)	Themes % (n)	Quotes
Hearing Aid 62% (53)	Child-related 40% (25/62)	She is not quite old enough to articulate when there is a problem. At 3 years old it's tough to tell if there is a problem with the aid settings or if my child just doesn't want to wear them. My child has been turning hearing aids off whenever they feel like it without anyone's knowledge. It's been a challenge to help them understand how important keeping aids turned on & in ears. He's at an age where he can help monitor/share some responsibility for his hearing aids and he doesn't always tell me right away if one stops working.
	Parent-related 35% (22/62)	Knowing what he's hearing without the hearing aids in and what is developing because of the hearing aids. Being unilateral it is difficult to know whether he is only hearing because of his good ear. It's challenging not knowing if hearing aids are enough. Not knowing if there's more I could be doing to help my child succeed in life. I would like to have a basic understanding of how to trouble shoot problems. I really didn't have any information regarding what to do if they aren't working correctly.
	Device-related 19% (12/62)	I wish there was an app on my phone that I could monitor the battery life or get a notification that something might not be working properly. Even better would be if the hearings could be GPS tracked by a phone app if lost. If the aids are providing adequate sounds. Also, when the battery is dead, not a big indicator... only a small light on the aid to show battery is dead. If I'm not paying attention she could wear for hours without being able to hear.
	Environment-related 5% (3/62)	My challenge is in cafeterias and gyms. My son does not hear what is going on and does not like the noise from so many other kids in those environments.
Bone Conduction Hearing Aid 61% (23)	Child-related 42% (10/24)	She is too young to let me know if there are any issues. He don't want to use.
	Parent-related 29% (7/24)	My child seems to hear very well without the device so it's hard to monitor the difference between his hearing with the device in verses when he isn't wearing it. I don't get to talk to the audiologist enough to ask questions. I assume it helps her but honestly she acts the same with or without it.
	Device-related 25% (6/24)	Hard to know when battery fails. Only by touching the BAHA and listening for feedback do I know it's working. Processor often falls off the band during playtime. I just have to assume that it is programmed correctly and is not too loud for my son.
Cochlear Implant 66% (45)	Child-related 47% (21/45)	She is still very young, so it isn't always easy for me to hold her attention in order to tell how well she is hearing certain speech sounds. Trying to figure out when he is ignoring us because that is what kids do or when he genuinely cannot hear us.
	Parent-related 27% (12/45)	Teaching his PreK teachers who have never worked with a deaf kid before how to best approach different situations with him. Remembering to do equipment checks in all the craziness. My child is 8, and has 2 younger siblings, and both my husband and I work full time. That I am the only family member that does everything because everyone else is afraid to "break them"
	Device-related 24% (11/45)	Battery life especially with disposables. I'd like easier access to the microphone port to more easily listen to the sound quality. They just fall off a lot because she's a toddler and busy! Using the assistive technology is a challenge for me. We do not use it much at this time so I forget to use it and don't remember how to link it with my phone and things like that.

Note. Some responses included more than one challenge.

APPENDIX B
Parent Responses to Open Question Offering Advice for Professionals

Responses % (n)	Themes % (n)	Quotes
Hearing Aid 55% (47/85)	Parent Education & Support 41% (31/75)	Explain as much as possible in terms parents understand. For many parents this is a new journey and we are trying to learn what we can to make the best decisions possible and support our child.
		Give the parents as much info, paper handouts, as possible. Most of the time parents are extremely overwhelmed and forget what was said, this way they would have a reference to look back on.
		Help the parents to become advocates for their children.
Relationship with Parents 37% (28/75)		Be patient with the kids. Be patient with the parents. Let them be involved in what you're doing and trust their feelings too. Don't brush off parent's concerns and questions.
		Be compassionate and acknowledge the difficulty of finding your child has hearing loss.
		Take the time to explain things to parents of young ones.
		Parents know their child best, so always listen to what they have to say when they think something is not right.
Professional Practices 21% (16/75)		Suggest places for support.
		Have on hand many recommendations for pediatric physicians who are familiar with hearing loss.
		I wish my audiologist would be more proactive about my daughter's hearing loss.
Bone Conduction Hearing Aid 55% (21/38)	Parent Education & Support 45% (13/29)	Be as detailed as possible in your exams/appointment.
		I think it's important to really explain and continue to reiterate the importance of consistency when it comes to having a child with hearing loss wear their BCHAs. Helping parents to understand the difference in the child's hearing when not wearing their device verses when they don't hear them. Doing hearing test with and without to show those differences. Especially for children who have hearing loss in only one ear.
		Please understand that parental comprehension may be low, even if they are well educated; it's often the first time we've heard many of these items... Also, "routine" visits/tests/equipment checks can be emotional experiences, depending on the day and parent, which makes comprehension that much more difficult. But these are our babies! We WANT to know all the information.
Relationship with Parents 38% (11/29)		Listen to the parent's gut. Often, they are seeing more than what you see. When a child has multiple developmental struggles, don't group all their speech struggles into a hearing related box; there can often be other struggles such as apraxia, aphasia, autism, or auditory processing that affect speech production, understanding, and development.
		Be honest and up front with parents.
		Be patient.
Professional Practices 17% (5/29)		Offer options and trials...have loaners available.
		Assume they are intelligent. Don't always think not responding in the booth means they simply didn't hear. Assume they are all intelligent enough to give you a hard time, even the ones with extra disabilities.
Cochlear Implant 63% (43/68)	Parent Education & Support 47% (23/49)	Give parents more time to talk and understand equipment and how to help child.
		Always share info with parents on how they can help their child. Write it down for them!
		Remember that parents are processing so much new information and it is super overwhelming, especially finding out their child isn't "perfect". Take the time to sit with parents and let them ask all the questions they have and make sure they understand how to operate and troubleshoot the equipment.
		Explain absolutely everything in great detail. Don't think that because a parent doesn't educate themselves they don't care. I spent endless hours researching hearing loss and cochlear implants but not all parents do that so you need to help bridge that gap. I wish I would have been provided with contact information to other parents of CI kids so if you're able to do that, please do. Lastly, don't become complacent in your job and forget how scary this can be for parents, especially at first. Be sympathetic and understanding.
Relationship with Parents 35% (17/49)		See the children and parents often and don't assume the child is caught up with their hearing peers and dismiss them from therapy. Stick with them and make sure they are comprehending and expanding vocabulary and understanding so they don't fall behind a couple of years later. Also, parents can get overwhelmed with information and it's hard to know what to say when working with your child. Keeping it simple and giving a simple summary of what to be working on and expecting from your child with some examples is a great resource for us to look at throughout the week for guidance.
		Stay patient and flexible!
		Don't discount a parent's concern.
Professional Practices 18% (9/49)		Trust the Mom! They know their child best.
		Be patient. Be positive. Be open to our wants and needs.
		Do not shame parents who use sign language along with cochlear implants. Allow it to be their choice and keep biased opinions to yourself. I do not think my son would understand/hear correctly a new word without sign support. Sign support can benefit cochlear implant children with similar sounding words, and so much more!
		There are tough stages and ages, eventually kids grow up and out of it, but you will always have a younger one coming. Breathe and have fun with them for the short time they are in your office.
		The European healthcare system (NHS) actually requires all pediatric audiologists to provide a retention accessory with every new fitting of a hearing aid or cochlear implant. That's something America does not practice.
		Data log even good wearers! We found a faulty cable that way.

Note. Some responses included advice in multiple areas.

Others' Publications About EHDl: October 2018 through April 2019

The *Journal of Early Hearing Detection and Intervention (JEHDI)* publishes peer-reviewed articles that describe current research, evidence-based practice, and standards of care that are relevant for newborn and early childhood hearing screening, diagnosis, support, early intervention, the medical home, information management, financing, and quality improvement. The aim of the journal is to improve Early Hearing Detection and Intervention (EHDl) systems.

Although JEHDI is the only journal that focuses exclusively on improving EHDl systems, many other journals include articles relevant to JEHDI's aim as a part their journal's broader focus. To help JEHDI readers stay up-to-date about current research and practices related to improving EHDl programs, we provide titles and abstracts of recent publications that are relevant to improving EHDl programs. Titles of all articles are hyperlinked to the source.

EHDl continues to be a global phenomenon. Of the 118 abstracts of articles included in the following abstracts, almost 50% are from authors in low and middle income countries. Many of the abstracts listed below focus on the basic components of EHDl systems (e.g., screening, diagnosis, early intervention), suggesting that there are still areas in the basic EHDl system that need to be improved. Other publications report studies about how to best incorporate detection of hearing loss in screening programs designed to detect conditions such as congenital cytomegalovirus and newborn genetic screening. There are also a number of studies about what causes hearing loss. For example Brennan-Jones et al. did a comprehensive review showing that children treated for childhood cancer using platinum analogues had more hearing loss than other children. The topic of childhood cancer treatment and hearing loss was also addressed by Clemens et al., Robertson et al., and Weiss et al. A number of studies from around the world also examined comorbidities of childhood hearing loss. For example, there were five articles that examined the incidence of childhood hearing loss among children diagnosed with sickle cell disease (Farrell et al., Lago et al., Rissatto-Lago et al., Schopper et al., and Towerman et al.) and De Schrijver et al. looked at the incidence of hearing loss among children with Down syndrome and concluded that it is not as prevalent as many people think. Knowing more about the conditions that cause childhood hearing loss and what other groups of children are affected with hearing loss, will help to improve the efficiency and effectiveness of EHDl systems.

Below are examples of other interesting findings of recently reported studies from around the world.

- Bartlett et al., based on a newborn screening program for congenital cytomegalovirus (CMV) that identified 214 cases of symptomatic and 88 cases of asymptomatic CMV, concluded that universal newborn CMV screening should be considered for implementation.
- Cedars et al., in a study with 3,257 children concluded that hearing screening using a combination of conditioned play audiometry and otoacoustic emissions testing in a preschool setting reduced referral rates, increased identification of hearing loss, reduced outcome disparities, and improved follow-up rates.
- Fitzpatrick et al. studied a total of 120 children (38 with unilateral hearing loss, 31 with bilateral mild hearing loss, and 51 with normal hearing) and concluded that even when they are identified during the first few months of life, children with unilateral hearing loss tend to lag behind their peers in receptive and expressive language development.
- Puia-Dumitrescu et al., in a study of gentamicin use in neonatal intensive care units that involved over 80,000 children concluded that use of gentamicin, regardless of dose and length of treatment was not associated with increased odds of failing the newborn hearing screen.
- Ramkumar et al., concluded that community-based pediatric screening in rural parts of India could be done more effectively using a telepractice model for diagnostic follow-up with auditory brainstem response compared to an in-person evaluation at a tertiary care hospital with auditory brainstem response testing.
- Sözen et al., in a study of the effect of a national pneumococcal vaccination program done in Turkey found that the incidence of meningitis-induced hearing loss had been reduced by more than ten-fold since the implementation of the program.
- Walker et al. found that only about one-third of preschool-aged children who are hard of hearing have access to a remote microphone system for home use, and about one-half for school use. For those children who have access to a remote microphone system, average use was only about 1–2 hours at home and 2–4 hours in school.

Abstracts of many more articles with results that are important for continuing to improve EHCI systems are listed below.

Abulebda K, Patel VJ, Ahmed SS, Tori AJ, Lutfi R, Abu-Sultaneh S.

[Comparison between chloral hydrate and propofol-ketamine as sedation regimens for pediatric auditory brainstem response testing.](#)

Braz J Otorhinolaryngol. 2019 Jan - Feb;85(1):32-36. doi: 10.1016/j.bjorl.2017.10.003. Epub 2017 Oct 28.

INTRODUCTION: The use of diagnostic auditory brainstem response testing under sedation is currently the “gold standard” in infants and young children who are not developmentally capable of completing the test.

OBJECTIVE: The aim of the study is to compare a propofol-ketamine regimen to an oral chloral hydrate regimen for sedating children undergoing auditory brainstem response testing.

METHODS: Patients between 4 months and 6 years who required sedation for auditory brainstem response testing were included in this retrospective study. Drugs doses, adverse effects, sedation times, and the effectiveness of the sedative regimens were reviewed.

RESULTS: 73 patients underwent oral chloral hydrate sedation, while 117 received propofol-ketamine sedation. 12% of the patients in the chloral hydrate group failed to achieve desired sedation level. The average procedure, recovery and total nursing times were significantly lower in the propofol-ketamine group. Propofol-ketamine group experienced higher incidence of transient hypoxemia.

CONCLUSION: Both sedation regimens can be successfully used for sedating children undergoing auditory brainstem response testing. While deep sedation using propofol-ketamine regimen offers more efficiency than moderate sedation using chloral hydrate, it does carry a higher incidence of transient hypoxemia, which warrants the use of a highly skilled team trained in pediatric cardio-respiratory monitoring and airway management.

Aloqaili Y, Arafat AS, Almarzoug A, Alalula LS, Hakami A, Almalki M, Alhuwaimel L.

[Knowledge about cochlear implantation: A parental perspective.](#)

Cochlear Implants Int. 2018 Nov 22:1-6. doi: 10.1080/14670100.2018.1548076.

OBJECTIVES: Cochlear implantation (CI) is used for children with severe to profound hearing loss who show little or no improvement using hearing aids. This study explored parental knowledge of their children’s CI.

METHODS: A cross-sectional study involving the parents of 115 pediatric CI patients was conducted at King Abdullah Specialized Children’s Hospital in Riyadh, Saudi Arabia. Parents were interviewed by telephone using a 50-question validated questionnaire.

RESULTS: Most parents of children with CI reported being comfortable in using the internet (68.7%) and social media (40.9%) to obtain information regarding CI. Although most parents of children with CI relied on health professionals and websites as their main sources of information, they were also able to obtain necessary information at meetings for CI patients and health professionals. Parents of children with CI felt they had sufficient information regarding the impact of hearing loss (78%) and CI (71%) on speech understanding and language development; however, they had insufficient information regarding criteria for CI candidacy, available brands of CI devices, and the advantages and disadvantages of each.

CONCLUSION: Parents reported that health professionals were the ideal source of information regarding hearing loss and CI. Moreover, our study showed that parents should learn more about cochlear implant devices, the post-implantation process, and candidacy criteria.

Ameyaw GA, Ribera J, Anim-Sampong S.

[Interregional Newborn Hearing Screening via Telehealth in Ghana.](#)

J Am Acad Audiol. 2019 Mar;30(3):178-186. doi: 10.3766/jaaa.17059. Epub 2018 Feb 7.

BACKGROUND: Newborn hearing screening is a vital aspect of the Early Hearing Detection and Intervention program, aimed at detecting hearing loss in children for prompt treatment. In Ghana, this kind of pediatric hearing service is available at only one health care facility located in the Greater Accra Region. The current practice in effect has virtually cut-off infants in the other regions from accessing hearing screening and other pediatric audiological services. This has prompted a study into alternative methodologies to expand the reach of such services in Ghana. The present study was designed to assess the feasibility of using telehealth to deliver newborn hearing screening across Ghana.

PURPOSE: To assess the feasibility of using telehealth to extend newborn hearing screening services across the ten regions of Ghana.

RESEARCH DESIGN: A correlational study was designed to determine the extent of association between test results of telehealth and the conventional on-site methods (COMs) for conducting newborn hearing screening. The design also allowed for testing duration between the two methods to be compared.

STUDY SAMPLE: Fifty infants from the Brong-Ahafo Regional Hospital (BARH) were enrolled. The infants aged between 2 and 90 days were selected through convenience sampling. There were 30 males and 20 females.

PROCEDURE: Newborn hearing screening using distortion product otoacoustic emissions were performed via telehealth. By adopting the synchronous telehealth model, an audiologist located at the Korle-Bu Teaching Hospital conducted real-time hearing screening tests over the internet on infants who were at the BARH. The former and latter hospitals are located in the Greater Accra and the Brong-Ahafo Regions, respectively. As a control, similar hearing screening tests were conducted on the same infants at BARH using the conventional face-to-face on-site hearing screening method.

DATA COLLECTION AND ANALYSIS: The test results and testing duration of the telehealth method and the conventional on-site approach were compared and subjected to statistical analysis. Here, the Spearman's correlation coefficient (r_s) was used to determine the level of correlation between the test results, whereas the paired t-test statistic was used to test the level of significance between the testing duration of the two methods.

RESULTS: Analysis of the test results showed a significantly high positive correlation between the telehealth and the COMs ($r_s = 0.778, 0.878, 0.857, 0.823, p < 0.05 @ 2.0, 3.0, 4.0, \text{ and } 5.0 \text{ kHz respectively}$). Also, the difference in testing duration of the two methods was not statistically significant [$t_{(99)} = 1.309, p > 0.05$]. The mean testing duration (in seconds) of telehealth was 27.287 (standard deviation = 27.373) and that of the COM was 24.689 (standard deviation = 27.169).

CONCLUSION: The study showed the feasibility of establishing an interregional network of newborn hearing screening services across Ghana using telehealth. It is more efficient to deploy telehealth for pediatric hearing services than to have patients travel many hours to the Greater Accra Region for similar services. Poor road network, high transportation costs, and bad weather conditions are a few of the reasons for avoiding long distance travel in Ghana.

Bartlett AW, Hall BM, Palasanthiran P, McMullan B, Shand AW, Rawlinson WD.

[Recognition, treatment, and sequelae of congenital cytomegalovirus in Australia: An observational study.](#)

J Clin Virol. 2018 Nov;108:121-125. doi: 10.1016/j.jcv.2018.09.017. Epub 2018 Sep 27.

BACKGROUND AND OBJECTIVES: Australian national surveillance data was used to assess recognition, sequelae, and antiviral therapy for congenital cytomegalovirus (CMV) cases.

STUDY DESIGN: Data from congenital CMV cases reported through the Australian Paediatric Surveillance Unit born January 1999 to December 2016 were described and Chi-square tests used to characterise trends and associations in case reporting, maternal CMV serology testing, and antiviral therapy. Descriptive analyses for hearing loss and developmental delay were reported for cases born ≥ 2004 , following introduction of universal neonatal hearing screening.

RESULTS: There were 302 congenital CMV cases (214 symptomatic, 88 asymptomatic). Congenital CMV was suspected in 70.6% by 30 days of age, with no differences across birth cohorts. Maternal CMV serology testing was associated with maternal illness during pregnancy but not birth cohort. There was increasing antiviral use for symptomatic cases, being used in 14% born 1999-2004, 19.6% born 2005-2010, and 44.4% born 2011-2016 ($p < 0.001$). For those born ≥ 2004 , hearing loss was reported in 42.1% of symptomatic and 26.6% of asymptomatic cases; while developmental delay was reported in 16.9% of symptomatic and 1.3% of asymptomatic cases.

CONCLUSION: There appears to be under-reporting and under-recognition of congenital CMV despite increasing use of antiviral therapy. Universal newborn CMV screening should be considered to facilitate follow-up of affected children and targeted linkage into hearing and developmental services, and to provide population-level infant CMV epidemiology to support research and evaluation of antiviral and adjunctive therapies.

Beaula Vincy VK, Seethapathy J, Boominathan P.

[Parental anxiety towards 'refer' results in newborn hearing screening \(NHS\) in south India: A hospital based study.](#)

Int J Pediatr Otorhinolaryngol. 2019 Jan;116:25-29. doi: 10.1016/j.ijporl.2018.10.021. Epub 2018 Oct 13.

BACKGROUND: Newborn Hearing Screening (NHS) aims at the early detection and intervention for children with congenital hearing loss. In developing countries like India, not all hospitals and birthing suites are equipped with NHS unit but there are few well established and emerging NHS programs that are operating in many parts of India. However, these screening procedures sometimes result in high false positive rates.

METHOD: This was a prospective cross sectional study. A total of 140 parents (parents of 70 well babies & parents of 70 NICU babies) of babies who underwent NHS between June, 2014 and December, 2014 at Sri Ramachandra Medical Centre (SRMC) were recruited for the study. Written parent consent was obtained prior to hearing screening. Parents of infants were counselled regarding the benefits of hearing screening, procedure of the screening test and need for follow-up testing if the neonate did not pass the screening test. Majority of the parents of infants (75%) were college graduates, 13% and 12% of parents had an educational level of high school and middle school respectively. Based on the Kuppuswamy's socioeconomic status scale, 69% of the parents were from upper middle class, 26% were from upper class and 5% were from lower middle class.

RESULTS: The present study aimed to identify parental anxiety towards 'refer' results of infants in the initial

NHS. Mean and standard deviation were used to find the state and trait anxiety levels in parents of each group. **CONCLUSION:** Refer' results in NHS lead to increased anxiety levels in parents of both well babies, and NICU babies. The increased anxiety levels may have greater impact on the parent's emotional status. Educating parents about screening procedures, possible causes for 'refer' results prior to screening, and also efforts to minimize false positive results in NHS can minimize unwanted anxiety in parents. At the same time, it is important that 'refer' results should be clearly explained and not minimized to ensure effective follow up. The audiologist dealing with NHS should take all attempts to alleviate anxiety in parents through public education, counseling and assertion.

Beswick R, David M, Higashi H, Thomas D, Nourse C, Koh G, Koorts P, Jardine L, Clark JE.

[Integration of congenital cytomegalovirus screening within a newborn hearing screening programme.](#)

J Paediatr Child Health. 2019 Mar 27. doi: 10.1111/jpc.14428. [Epub ahead of print]

AIM: Targeted screening by a salivary cytomegalovirus (CMV) polymerase chain reaction (PCR) of infants who 'refer' on their newborn hearing screen has been suggested as an easy, reliable and cost-effective approach to identify and treat babies with congenital CMV (cCMV) to improve hearing outcomes. This study aimed to investigate the feasibility and cost-effectiveness of introducing targeted salivary cCMV testing into a newborn hearing screening programme.

METHODS: The study included three tertiary maternity hospitals in Queensland, Australia between August 2014 and April 2016. Infants who 'referred' on the newborn hearing screen were offered a salivary swab for CMV PCR at the point of referral to audiology. Swabs were routinely processed and tested for CMV DNA by real-time quantitative PCR. Parents of babies with a positive CMV PCR were notified, and the babies were medically assessed and, where appropriate, were offered treatment (oral valganciclovir).

RESULTS: Of eligible infants, the parents of 83.0% (234/283) consented to the cCMV screen. Of these, 96.6% returned a negative result (226/234), and 3.4% (8/234) returned a positive result (three true positive; five false positive). The prevalence of cCMV for infants with confirmed hearing loss was 3.64% ($P = 2/55$; confidence interval = 0.44-12.53%). The cost comparison suggests the cost implementation of cCMV screening (and subsequent potential treatment benefits and management over time), compared to non-screening (and subsequent management), to be negligible.

CONCLUSION: Incorporating cCMV testing into Universal Newborn Hearing Screening within Queensland is realistic and achievable, both practically and financially.

Bezdjian A, Smith RA, Thomeer HGXM, Willie BM, Daniel SJ.

[A Systematic Review on Factors Associated With Percutaneous Bone Anchored Hearing Implants Loss.](#)

Otol Neurotol. 2018 Dec;39(10):e897-e906. doi:10.1097/MAO.0000000000002041.

OBJECTIVE: To investigate factors associated with percutaneous bone anchored hearing implant (BAHI) loss.

DATA SOURCES: Africa-Wide, Biosis, Cochrane, Embase, Global Health, LILACs, Medline, Pubmed, and Web of Science electronic databases.

STUDY SELECTION: All studies reporting on adult and/or pediatric patients with a BAHI loss were identified. Retrieved articles were screened using predefined inclusion criteria. Eligible studies underwent critical appraisal for directness of evidence and risk of bias. Studies that successfully passed critical appraisal were included for data extraction.

DATA EXTRACTION: Extracted data included study characteristics (study design, number of total implants and implant losses, follow-up), patient characteristics (sex, age, comorbidities, previous therapies), and information regarding BAHI loss (etiology of loss, timing of occurrence).

DATA SYNTHESIS: From the 5,151 articles identified at the initial search, 847 remained after title and abstract screening. After full text review, 96 articles were eligible. Fifty-one articles passed quality assessment, however, due to overlapping study population, 48 articles reporting on 34 separate populations were chosen for data extraction. Three hundred one implant losses occurred out of 4,116 implants placed, resulting in an overall implant loss occurrence rate of 7.3%. Failed osseointegration was responsible for most implant losses (74.2%), followed by fixture trauma (25.7%). Most losses due to failed osseointegration occurred within 6 months of the implantation. BAHI implant loss occurred more frequently in pediatric patients ($p < 0.005$).

CONCLUSION: The current systematic review identified factors associated with BAHI loss. These factors should be considered when assessing patients' candidacy and when investigating reasons for impeded implant stability and loss.

Blankenship CM, Hunter LL, Keefe DH, Feeney MP, Brown DK, McCune A, Fitzpatrick DF, Lin L.

[Optimizing Clinical Interpretation of Distortion Product Otoacoustic Emissions in Infants.](#)

Ear Hear. 2018 Nov/Dec;39(6):1075-1090. doi: 10.1097/AUD.0000000000000562.

OBJECTIVES: The purpose of this study was to analyze distortion product otoacoustic emission (DPOAE) level and signal to noise ratio in a group of infants from birth to 4 months of age to optimize prediction of hearing

status. DPOAEs from infants with normal hearing (NH) and hearing loss (HL) were used to predict the presence of conductive HL (CHL), sensorineural HL (SNHL), and mixed HL (MHL). Wideband ambient absorbance was also measured and compared among the HL types.

DESIGN: This is a prospective, longitudinal study of 279 infants with verified NH and HL, including conductive, sensorineural, and mixed types that were enrolled from a well-baby nursery and two neonatal intensive care units in Cincinnati, Ohio. At approximately 1 month of age, DPOAEs (1-8 kHz), wideband absorbance (0.25-8 kHz), and air and bone conduction diagnostic tone burst auditory brainstem response (0.5-4 kHz) thresholds were measured. Hearing status was verified at approximately 9 months of age with visual reinforcement audiometry (0.5-4 kHz). Auditory brainstem response air conduction thresholds were used to assign infants to an NH or HL group, and the efficacy of DPOAE data to classify ears as NH or HL was analyzed using receiver operating characteristic (ROC) curves. Two summary statistics of the ROC curve were calculated: the area under the ROC curve and the point of symmetry on the curve at which the sensitivity and specificity were equal. DPOAE level and signal to noise ratio cutoff values were defined at each frequency as the symmetry point on their respective ROC curve, and DPOAE results were combined across frequency in a multifrequency analysis to predict the presence of HL.

RESULTS: Single-frequency test performance of DPOAEs was best at mid to high frequencies (3-8 kHz) with intermediate performance at 1.5 and 2 kHz and chance performance at 1 kHz. Infants with a conductive component to their HL (CHL and MHL combined) displayed significantly lower ambient absorbance values than the NH group. No differences in ambient absorbance were found between the NH and SNHL groups. Multifrequency analysis resulted in the best prediction of HL for the SNHL/MHL group with poorer sensitivity values when infants with CHL were included.

CONCLUSIONS: Clinical interpretation of DPOAEs in infants can be improved by using age-appropriate normative ranges and optimized cutoff values. DPOAE interpretation is most predictive at higher F2 test frequencies in young infants (2-8 kHz) due to poor test performance at 1 to 1.5 kHz. Multifrequency rules can be used to improve sensitivity while balancing specificity. Last, a sensitive middle ear measure such as wideband absorbance should be included in the test battery to assess possibility of a conductive component to the HL.

Bouillot L1, Vercherat , Durand C.

[Implementing universal newborn hearing screening in the French Rhône-Alpes region. State of affairs in 2016 and the 1st half of 2017.](#)

Int J Pediatr Otorhinolaryngol. 2019 Feb;117:30-36. doi: 10.1016/j.ijporl.2018.11.011. Epub 2018 Nov 10.

INTRODUCTION: Universal newborn hearing screening (UNHS) started as public health policy in 2015 in the French Rhône-Alpes region, aiming to screen for unilateral and bilateral hearing loss. After a first and second screening (retest) in the maternity hospital, the diagnostic process occurred at a limited number of specialist centers. A deferred preliminary screening (T3) was proposed before the age of 1 month. The aims of this study were to assess implementation of the program, impact of T3, and present the incidence of hearing loss in this population.

MATERIALS AND METHODS: The retrospective observational study was based on data transmitted routinely by the 51 maternities to the regional organization responsible for newborn screening, in 2016 and first half of 2017.

RESULTS: All the facilities implemented the UNHS protocol, with 47 out of 51 using the recommended techniques. 99.7% of the 115,435 newborns were screened (excluding 0.2% of parental refusals). A retest was required for 10.2% of the babies. Among babies who didn't pass retest, 7.7% were lost to follow-up. 2.2% of the newborns were referred to diagnostic centers. The rate of T3 was 31.3% of newborns who did not pass retest. 88.6% of the infants passed T3. In the perinatal network making extensive use of T3 (75.8% versus 14.9% elsewhere), 0.6% of the infants were referred to a diagnostic center, versus 2.9% in the rest of the region (2016, $p < 0.001$). For 2016, the outcomes at 6 months revealed an overall hearing loss rate of 1.7‰ (4.7‰ for neonatal care unit babies), and bilateral hearing loss in 1.2‰.

CONCLUSION: In Rhône-Alpes, the national and regional objectives for UNHS were exceeded, although limiting the number of infants lost to follow-up remains essential. Repeating an automated test around 2-4 weeks after birth improves the program by decreasing the false positives of the screening. It considerably limits the number of infants referred to specialist centers, without increasing the number of patients lost to follow-up.

Brennan-Jones CG, McMahan C, Van Dalen EC.

[Cochrane corner: platinum-induced hearing loss after treatment for childhood cancer.](#)

Int J Audiol. 2019. Apr;58(4):181-184. doi: 10.1080/14992027.2018.1539808. Epub 2018 Dec 13.

ABSTRACT: This Cochrane Corner features the review entitled "Platinum-induced hearing loss after treatment for childhood cancer" published in 2016. In their review, van As et al. identified 13 cohort studies including 2837 participants with a hearing test after treatment with a platinum-based therapy for different types of childhood cancers. All studies had problems related to quality of the evidence. The reported frequency of hearing loss varied between 1.7% and 90.1% for studies that included a definition of hearing loss; none of the studies

provided data on tinnitus. Only two studies evaluated possible risk factors. One study found a higher risk of hearing loss in people treated with the combination of cisplatin plus carboplatin compared to treatment with cisplatin only and for exposure to aminoglycosides. The other found that age at treatment (lower risk in older children) and single maximum cisplatin dose (higher risk with an increasing dose) were significant predictors for hearing loss, while gender was not. This systematic review shows that children treated with platinum analogues are at risk of developing hearing loss, but the exact prevalence and risk factors remain unclear.

Butcher E, Dezateux C, Knowles RL.

[Risk factors for permanent childhood hearing impairment.](#)

Arch Dis Child. 2018 Nov 28. pii: archdischild-2018-315866. doi: 10.1136/archdischild-2018-315866. [Epub ahead of print]

OBJECTIVE: While several perinatal risk factors for permanent childhood hearing impairment (PCHI) are known, association with gestational length remains unclear. We hypothesised that shorter gestational length predicts higher PCHI risk.

DESIGN: 19 504 participants from the UK Millennium Cohort Study (born 2000-2002, prior to newborn screening).

METHODS: Multivariable discrete-time survival analysis to examine associations between parent-reported PCHI by age 11 years and gestational length, plus other prespecified factors.

RESULTS: PCHI affected 2.1 per 1000 children (95% CI 1.5 to 3.0) by age 11; however, gestational length did not predict PCHI risk (HR, 95% CI 1.00, 0.98 to 1.03 per day increase). Risk was increased in those with neonatal illness, with or without admission to neonatal care (6.33, 2.27 to 17.63 and 2.62, 1.15 to 5.97, respectively), of Bangladeshi or Pakistani ethnicity (2.78, 1.06 to 7.31) or born to younger mothers (0.92, 0.87 to 0.97 per year).

CONCLUSION: Neonatal illness, rather than gestational length, predicts PCHI risk. Further research should explore associations with ethnicity.

Cedars E, Kriss H, Lazar AA, Chan C, Chan DK.

[Use of otoacoustic emissions to improve outcomes and reduce disparities in a community preschool hearing screening program.](#)

PLoS One. 2018 Dec 10;13(12):e0208050. doi: 10.1371/journal.pone.0208050. eCollection 2018.

INTRODUCTION: Hearing loss substantially impacts pediatric development, and early identification improves outcomes. While intervening before school-entry is critical to optimize learning, early-childhood hearing screening practices are highly variable. Conditioned play audiometry (CPA) is the gold standard for preschool hearing screening, but otoacoustic emission (OAE) testing provides objective data that may improve screening outcomes.

OBJECTIVES: To compare outcomes of a community-based low-income preschool hearing program before and after implementation of OAE in a single-visit, two-tiered paradigm. We hypothesized that this intervention would reduce referral rates and improve follow-up while maintaining stable rates of diagnosed sensorineural hearing loss.

METHODS: We performed a cohort study of 3257 children screened from July 2014-June 2016. Department of Public Health data were analyzed pre- and post-implementation of second-line OAE testing for children referred on CPA screening with targeted follow-up by DPH staff. Primary outcomes included referral rates, follow-up rates, and diagnosis of sensorineural hearing loss.

RESULTS: Demographics, pure-tone pass rates, and incidence of newly-diagnosed permanent hearing loss were similar across years. After intervention, overall pass rates increased from 92% to 95% ($P = 0.0014$), while only 0.7% remained unable to be tested ($P < 0.0001$). 5% of children were unable to be tested by CPA screening but passed OAE testing, obviating further evaluation. Referral rate decreased from 8% to 5% ($P = 0.0014$), and follow-up improved from 36% to 91% ($P < 0.0001$). Identification of pathology in children with follow-up increased from 19% to over 50%. Further, disparities in pass rates and ability to test seen in Year 1 were eliminated in Year 2.

CONCLUSION AND RELEVANCE: In a community setting, implementation of second-line OAE screening for CPA referrals reduced referral rates, increased identification of hearing loss, reduced outcome disparities, and improved follow-up rates. This study provides lessons in how to improve outcomes and reduce disparities in early-childhood hearing screening.

Cetin SY, Erel S, Bas Aslan U.

[The effect of Tai Chi on balance and functional mobility in children with congenital sensorineural hearing loss.](#)

Disabil Rehabil. 2019 Jan 9:1-8. doi: 10.1080/09638288.2018.1535629. [Epub ahead of print]

BACKGROUND: The aim of the study was to examine the effect of Tai Chi on balance and functional mobility in children with congenital sensorineural hearing loss.

METHODS: The study included 39 children, aged 10-14 years, with congenital sensorineural hearing loss. The

participants were divided into three groups as the Tai Chi group, conventional exercise group, and control group. The Tai Chi group and the conventional exercise group received a 1-h exercise program twice a week for 10 weeks. The balance function of the children was assessed using the Pediatric Balance Scale, the balance subtest of Bruininks-Oseretsky Test 2-Short Form, and the Functional Reach Test. The Timed Up and Go Test and the Timed Up and Down Stairs Test were used to assess functional mobility. The Wilcoxon rank, Kruskal-Wallis, and Mann-Whitney U-tests were used for statistical analyses.

RESULTS: When the pre-training values of the groups were compared, with the exception of the Timed Up and Go test, there was no statistically significant difference with respect to demographic data, balance, and functional mobility parameters ($p > 0.05$). After training, the overall balance and functional mobility tests improved compared to pre-training values in both the Tai Chi and conventional exercise groups ($p < 0.05$). When the post-training values were compared between the groups, with the exception of the Functional Reach Test and the Timed Up and Down Stairs Test, the results of both exercise groups were superior to those of the control group ($p < 0.05$).

CONCLUSIONS: The results of this study indicate that Tai Chi and conventional exercise programs have positive effects on balance and functional mobility in children with congenital sensorineural hearing loss. However, no superiority of Tai Chi or the conventional exercise programs was determined over the other. Both Tai Chi and conventional exercise programs could be used to improve balance and functional mobility in children with congenital sensorineural hearing loss. Implications for rehabilitation Tai Chi and conventional exercises are effective on balance in children with congenital sensorineural hearing loss. Tai Chi and conventional exercises are effective on functional mobility in children with congenital sensorineural hearing loss. Tai Chi may be added to the rehabilitation program for children with congenital sensorineural hearing loss.

Chan KH, Dreith S, Uhler KM, Tallo V, Lucero M, De Jesus J, Simões EA.

[Large-scale otoscopic and audiometric population assessment: A pilot study.](#)

Int J Pediatr Otorhinolaryngol. 2019 Feb;117:148-152. doi: 10.1016/j.ijporl.2018.11.033. Epub 2018 Nov 30.

OBJECTIVE: Large-scale otoscopic and audiometric assessment of populations is difficult due to logistic impracticalities, particularly in low- and middle-income countries (LMIC). We report a novel assessment methodology based on training local field workers, advances in audiometric testing equipment and cloud-based technology.

METHODS: Prospective observational study in Bohol, Philippines. A U.S. otolaryngologist/audiologist team trained 5 local nurses on all procedures in a didactic and hands-on process. An operating otoscope (Welch-Allyn[®]) was used to clear cerumen and view the tympanic membrane, images of which were recorded using a video otoscope (JedMed[®]). Subjects underwent tympanometry and distortion product otoacoustic emission (DPOAE) (Path Sentiero[®]), and underwent screening audiometry using noise cancelling headphones and a handheld Android device (HearScreen[®]). Sound-booth audiometry was reserved for failed subjects. Data were uploaded to a REDCap database. Teenage children previously enrolled in a 2000-2004 Phase 3 pneumococcal conjugate vaccine trial, were the subjects of the trainees.

RESULTS: During 4 days of training, 47 Filipino children (M/F = 28/19; mean/median age = 14.6/14.6 years) were the subjects of the trainee nurses. After the training, all nurses could perform all procedures independently. Otoscope findings by ears included: normal (N = 77), otitis media with effusion (N = 2), myringosclerosis (N = 5), healed perforation (N = 6), perforation (N = 2) and retraction pocket/cholesteatoma (N = 2). Abnormal audiometric findings included: tympanogram (N = 4), DPOAE (N = 4) and screening audiometry (N = 0).

CONCLUSION: Training of local nurses has been shown to be robust and this methodology overcomes challenges of distant large-scale population otologic/audiometric assessment.

Chang YS, Ryu G, Kim K, Cho YS.

[Normative wideband absorbance measures in healthy neonates in Korea: A preliminary study.](#)

Int J Pediatr Otorhinolaryngol. 2019 Feb;117:6-11. doi: 10.1016/j.ijporl.2018.11.012. Epub 2018 Nov 11.

INTRODUCTION: The usefulness of wideband absorbance (WBA) in newborns is well-demonstrated. However, it is still not clear whether there might be a difference according to ethnicity with respect to ambient WBA; therefore, further investigation is necessary to evaluate ethnic-specific normative WBA values in newborns.

METHODS: Twenty-one newborns (41 ears) were recruited from the well-baby nursery at a tertiary referral center. All newborn infants who were born at 38 weeks' to 41 weeks' gestation with a normal birth weight (range: 2.5-4.5 kg) and who passed a newborn hearing screening test with distortion product otoacoustic emissions were enrolled. Ambient absorbance values were measured on frequencies ranging from 226 Hz to 6300 Hz (i.e., 250 Hz, 315 Hz, 400 Hz, 500 Hz, 620 Hz, 800 Hz, 1000 Hz, 1250 Hz, 1600 Hz, 2000 Hz, 2500 Hz, 3150 Hz, 4000 Hz, 5000 Hz, and 6300 Hz). The results of median absorbance were compared with the WBA values of Caucasian infants and Korean adults.

RESULTS: The gestational age of the study group was 38 weeks \pm 6.67 days. In a gender comparison, absorbance of female neonate was significantly higher at 3150 Hz, 4000 Hz, and 5000 Hz than in male. Based on the

test frequencies, the medians of the Korean infant WBA values and Caucasian infants are significantly different from one another, except at 1600 Hz, 3150 Hz, and 4000 Hz. The results of a median absorbance comparison between Korean infant and adults WBA values showed that the medians of the two studies were significantly different except at 1250 Hz.

CONCLUSION: We analyzed the normative WBA values measured at ambient pressures in Korean newborns. The comparative analysis between the normative values of two different ethnic groups may infer a possible difference in the normative WBA values. The absorbance from Korean infant ears is substantially different from that from adult's ears. A large-scale study is required to establish normative WBA values to be used for the screening of outer and middle ear status in newborns.

Chen K, Jiang H, Zong L, Wu X.

[Side-related differences in sudden sensorineural hearing loss in children.](#)

Int J Pediatr Otorhinolaryngol. 2018 Nov;114:5-8. doi: 10.1016/j.ijporl.2018.08.022. Epub 2018 Aug 22.

OBJECTIVE: Most studies on sudden sensorineural hearing loss (SSNHL) do not differentiate the outcomes within varied affected ears in children. The present study was designed to determine the clinical differences between unilateral and bilateral SSNHL in children.

METHODS: The clinical data, from a total of 101 pediatric patients with SSNHL, was retrospectively analyzed from January 2003 to December 2016. The main outcome measures included basic characteristics, etiology, clinical symptoms and treatment courses.

RESULTS: When the bilateral group (n = 28) was compared to the unilateral group (n = 73), neither gender nor onset of SSNHL was significantly different (p > 0.05 each); However, bilateral SSNHL tended to occur in younger ages (8.1 ± 4.0 yrs), with higher percentages of suspected etiologies (50%) and proportion of profound deafness (55.4%, p < 0.05 each). The short-term recovery rate was superior in the unilateral cases over the bilateral cases (37.0% vs. 12.5%, p < 0.05). Milder initial hearing threshold, early onset of treatment (5.6 ± 4.8 days) with unilateral involvement and an older age (11.3 ± 3.0 yrs) in bilaterally affected cases were associated with a better prognosis in this cohort. In addition, the unilateral group showed comparable outcomes, when sub-analyzed by comparison to that in either left- (n = 42) or right-sided (n = 31) SSNHL.

CONCLUSION: Although bilateral and unilateral pediatric SSNHL could cause partial to complete cochlear lesion, they may be relevant to distinct backgrounds. Our data also provides valuable information about demographics and outcomes of SSNHL in children.

Clemens E, Brooks B, de Vries ACH, van Grotel M, van den Heuvel-Eibrink MM, Carleton B.

[A comparison of the Muenster, SIOP Boston, Brock, Chang and CTCAEv4.03 ototoxicity grading scales applied to 3,799 audiograms of childhood cancer patients treated with platinum-based chemotherapy.](#)

PLoS One. 2019 Feb 14;14(2):e0210646. doi: 10.1371/journal.pone.0210646. eCollection 2019.

ABSTRACT: Childhood cancer patients treated with platinum often develop hearing loss and the degree is classified according to different scales globally. Our objective was to compare concordance between five well-known ototoxicity scales used for childhood cancer patients. Audiometric test results (n = 654) were evaluated longitudinally and graded according Brock, Chang, International Society of Pediatric Oncology (SIOP) Boston, Muenster scales and the U.S. National Cancer Institute Common Technology Criteria for Adverse Events (CTCAE) version 4.03. Adverse effects of grade 2, 3 and 4 are considered to reflect a degree of hearing loss sufficient to interfere with day-to-day communication (> = Chang grade 2a; > = Muenster grade 2b). We term this "deleterious hearing loss". A total number of 3,799 audiograms were evaluated. The prevalence of deleterious hearing loss according to the last available audiogram of each patient was 59.3% (388/654) according to Muenster, 48.2% (315/653) according to SIOP, 40.5% (265/652) according to Brock, 40.3% (263/652) according to Chang, and 57.5% (300/522) according to CTCAEv4.03. Overall concordance between the scales ranged from $\kappa = 0.636$ (Muenster vs. Chang) to $\kappa = 0.975$ (Brock vs. Chang). Muenster detected hearing loss the earliest in time, followed by Chang, SIOP and Brock. Generally good concordance between the scales was observed but there is still diversity in definitions of functional outcomes, such as differences in distribution levels of severity of hearing loss, and additional intermediate scales taking into account losses <40 dB as well. Regardless of the scale used, hearing function decreases over time and therefore, close monitoring of hearing function at baseline and with each cycle of platinum therapy should be conducted.

Coleman A, Cervin A.

[Probiotics in the treatment of otitis media. The past, the present and the future.](#)

Int J Pediatr Otorhinolaryngol. 2019 Jan;116:135-140. doi: 10.1016/j.ijporl.2018.10.023. Epub 2018 Oct 19.

ABSTRACT: Otitis media (OM) is one of the most common infectious diseases in children and the leading cause for medical consultations and antibiotic prescription in this population. The burden of disease associated with OM is greater in developing nations and indigenous populations where the associated hearing loss contributes to poor education and employment outcomes. Current treatment and prevention is largely focused on

vaccination and antibiotics. However, rates of OM, particularly in indigenous populations, remain high. With growing concerns regarding antibiotic resistance and antibiotic-associated complications, an alternative, more effective treatment is required. Administration of probiotics, both locally and systemically have been investigated for their ability to treat and prevent OM in children. This review explores the theoretical bases of probiotics, successful application of probiotics in medicine, and their use in the treatment and prevention of OM. We conclude that local administration of niche-specific probiotic bacteria that demonstrates the ability to inhibit the growth of otopathogens in vitro shows promise in the prevention and treatment of OM and warrants further investigation.

De Schrijver L, Topsakal V, Wojciechowski M, Van de Heyning P, Boudewyns A.

[Prevalence and etiology of sensorineural hearing loss in children with down syndrome: A cross-sectional study.](#)

Int J Pediatr Otorhinolaryngol. 2019 Jan;116:168-172. doi: 10.1016/j.ijporl.2018.10.048. Epub 2018 Nov 3.

BACKGROUND: The prevalence and causes of sensorineural hearing loss (SNHL) in children with Down syndrome (DS) are poorly delineated.

OBJECTIVE: To describe the prevalence, severity, laterality and underlying etiology of SNHL in a cohort of children with DS.

METHODS: A cross-sectional study was performed among all children with DS followed at the multidisciplinary Downteam of the Antwerp University Hospital. Patients' characteristics, risk factors for hearing loss, audiometric data and results of an etiological work-up were collected.

RESULTS: Among 291 patients in follow-up, 138 patients (47.4%) presented with hearing loss. In the majority this was caused by middle ear effusion and only 13 patients (4.5%) had sensorineural hearing loss, 7 boys and 6 girls with a mean age of 14.4 ± 7.4 years. Hearing loss was bilateral in 8 cases. Hearing loss severity was graded as mild in 38.5%, moderate in 30.8% and profound in 30.8% of the patients. An etiological work-up was completed in 9 children. Four patients presented with single sided deafness due to cochlear nerve deficiency. One patient had a genetic cause and in 2 patients the hearing loss was attributed to excessive noise exposure. The etiology of hearing loss was unknown in 6 patients.

CONCLUSION: Sensorineural hearing loss is uncommon in children with DS with a prevalence of 4.5%. Etiological work-up may allow identifying a specific underlying cause. Cochlear nerve deficiency was found in 4 children with DS and single sided deafness.

Dedhia K, Graham E, Park A.

[Hearing Loss and Failed Newborn Hearing Screen.](#)

Clin Perinatol. 2018 Dec;45(4):629-643. doi: 10.1016/j.clp.2018.07.004. Epub 2018 Sep 24.

ABSTRACT: Hearing loss is the most common congenital defect. With early diagnosis and intervention, we are able to improve speech and language outcomes in this population. In this article, we discuss the implications of the newborn hearing screen, as well as diagnostic interventions, management, and intervention, and the increasing role of congenital cytomegalovirus screening.

Deng Y, Sang S, Wen J, Liu Y, Ling J, Chen H, Cai X, Mei L, Chen X, Li M, Li W, Li T, He C, Feng Y.

[Reproductive guidance through prenatal diagnosis and genetic counseling for recessive hereditary hearing loss in high-risk families.](#)

Int J Pediatr Otorhinolaryngol. 2018 Dec;115:114-119. doi: 10.1016/j.ijporl.2018.08.026. Epub 2018 Sep 12.

OBJECTIVE: To evaluate the accuracy and validity of our protocol for prenatal diagnosis and genetic counseling in high-risk families at a clinic.

METHODS: Fifteen unrelated families with recessive nonsyndromic hearing loss (NSHL) in their family history and a positive attitude towards prenatal diagnosis were recruited in the present study. According to genetic information for each family, Sanger sequencing, fluorescence polymerase chain reaction (PCR)-based congenital deafness gene detection kit and multiple PCR-based target gene capture and high-throughput sequencing were used. Genetic counseling was offered to all participating families by genetic counselors and otologists. Prenatal diagnosis was provided to families with detected pathogenic mutations and who were expected to participate in subsequent prenatal diagnosis.

RESULTS: In this study, confirmed pathogenic mutations were detected in eight families, who were defined as high-risk families. These families all participated in prenatal diagnosis with positive attitudes. One novel variant (c.1687dupA) in the SLC26A4 gene was detected in a family. Through genetic counseling, the recurrence probability of NSHL in fetuses was 25% in six families, 0% in one family, and 50% in one family. The results of fetal DNA detection showed that one fetal variant was wild type, three were heterozygous mutations in SLC26A4, and one was a compound heterozygous mutation in SLC26A4. Two variants were heterozygous mutations in GJB2, and one was a homozygous mutation in GJB2. According to the test results for fetal DNA, prenatal diagnosis found that six fetuses had normal hearing, whereas two fetuses suffered from NSHL. After birth, six infants predicted to have normal hearing passed a newborn hearing screening test and two infants predicted to

have NSHL were diagnosed with NSHL and received cochlear implants.

CONCLUSION: Our protocol for prenatal diagnosis and genetic counseling provides detailed information that can assist couples in high-risk families in preparing for infant arrival and future family planning. For the affected neonates, prenatal diagnosis and genetic counseling achieve an “early screening, early diagnosis, early intervention” strategy.

Dev AN, Lohith U, Pascal B, Dutt CS, Dutt SN.

[A questionnaire-based analysis of parental perspectives on pediatric cochlear implant \(CI\) re/habilitation services: a pilot study from a developing CI service in India.](#)

Cochlear Implants Int. 2018 Nov;19(6):338-349. doi: 10.1080/14670100.2018.1489937. Epub 2018 Jun 29.

OBJECTIVE: To study parental perspectives on re/habilitation services offered for pediatric cochlear implant (CI) users at a non-profit organization in India.

METHODOLOGY: A non-standardized questionnaire comprising 46 items was created to understand perspectives of parents of pediatric CI users. Questions were designed to examine re/habilitation services from the angles of service delivery, parental stress levels, reasons for delay in obtaining services, sources of emotional support, concerns, and fears during each stage starting from diagnosis of hearing loss to CI surgery, re/habilitation services and parents' views of their children post-CI. The questionnaire was posed to 30 parents and responses were recorded and coded.

RESULTS AND DISCUSSION: Qualitative and quantitative analyses based on parents' responses identified several factors that significantly influenced parental perspectives during each stage. The major factors delaying the decision to go for CI included a fear of surgery, lack of funds for CI and the subsequent re/habilitation process, and limited knowledge. Key concerns were the child's academic performance and social acceptance. Familial support played an important role during each stage. A significant reduction in the parental stress levels was observed following CI surgery. Parents indicated that local support for therapy, financial assistance and better guidance at each stage would substantially help in lowering stress levels.

CONCLUSIONS: The parental perspectives analyzed in this study can be utilized towards improving the quality of service delivery in terms of parental satisfaction and outcomes post-CI. Efforts should be taken to improve parental awareness, funding options, and access to re/habilitation services and social networks connecting similar parents.

DiNino M, O'Brien G, Bierer SM, Jahn KN, Arenberg JG.

[The Estimated Electrode-Neuron Interface in Cochlear Implant Listeners Is Different for Early-Implanted Children and Late-Implanted Adults.](#)

J Assoc Res Otolaryngol. 2019 Mar 25. doi: 10.1007/s10162-019-00716-4. [Epub ahead of print]

ABSTRACT: Cochlear implant (CI) programming is similar for all CI users despite limited understanding of the electrode-neuron interface (ENI). The ENI refers to the ability of each CI electrode to effectively stimulate target auditory neurons and is influenced by electrode position, neural health, cochlear geometry, and bone and tissue growth in the cochlea. Hearing history likely affects these variables, suggesting that the efficacy of each channel of stimulation differs between children who were implanted at young ages and adults who lost hearing and received a CI later in life. This study examined whether ENI quality differed between early-implanted children and late-implanted adults. Auditory detection thresholds and most comfortable levels (MCLs) were obtained with monopolar and focused electrode configurations. Channel-to-channel variability and dynamic range were calculated for both types of stimulation. Electrical field imaging data were also acquired to estimate levels of intracochlear resistance. Children exhibited lower average auditory perception thresholds and MCLs compared with adults, particularly with focused stimulation. However, neither dynamic range nor channel-to-channel threshold variability differed between groups, suggesting that children's range of perceptible current was shifted downward. Children also demonstrated increased intracochlear resistance levels relative to the adult group, possibly reflecting greater ossification or tissue growth after CI surgery. These results illustrate physical and perceptual differences related to the ENI of early-implanted children compared with late-implanted adults. Evidence from this study demonstrates a need for further investigation of the ENI in CI users with varying hearing histories.

Dumont J, Abouzayd M, Le Louarn A, Pondaven S, Bakhos D, Lescanne E.

[Total and partial ossiculoplasty in children: Audiological results and predictive factors.](#)

Eur Ann Otorhinolaryngol Head Neck Dis. 2019 Mar 14. pii: S1879-7296(19)30037-7. doi: 10.1016/j.anorl.2019.02.012. [Epub ahead of print]

OBJECTIVE: To assess ossiculoplasty results in children and screen for predictive factors of efficacy.

PATIENTS AND METHODS: Seventy five children undergoing ossiculoplasty between 2001 and 2014 in a pediatric ENT department were included. The following data were collected and analyzed: demographic data, surgical indication, history of tympanoplasty, contralateral ear status (healthy, affected), preoperative hearing

thresholds, surgical technique, intraoperative findings, and ossicular chain status at eardrum opening. Audiological results were reported according to American Academy of Otolaryngology-Head and Neck Surgery guidelines.

RESULTS: Forty eight patients were included in the total ossicular reconstruction prosthesis (TORP) group. Mean age at surgery was 9.9years. Mean follow up was 2.7years. Mean air-bone gap (ABG) closure to within 20dB was achieved in 40% of cases at medium term (12 to 18 months after surgery). Air conduction (AC) threshold ≤ 30 dB was achieved in 68% of cases. AC threshold improved by 14.6dB and 8.7dB at medium and long-term follow-up, respectively. A significant correlation was found between success rate and absence of history of tympanoplasty. The success rate was higher for primary than for revision procedures. Twenty seven children were included in the partial ossicular reconstruction prosthesis (PORP) group. Mean age was 9.5years, and mean follow-up 2.6years. Mean air-bone gap (ABG) closure to within 20dB was achieved in 75% of cases at medium term. AC threshold ≤ 30 dB was achieved in 75% of cases AC threshold improved by 9.3dB and 5dB at medium and long-term follow-up, respectively. No predictive factors for success were found in the PORP group.

CONCLUSION: The present study suggested that total ossiculoplasty leads to better results when performed in first-line. It also confirmed that functional outcome is better in partial than total ossicular reconstruction prosthesis.

EI-Dessouky HM, Aziz AA, Sheikhany AR, ElMeshmeshy LM.

[Validation of the Egyptian Arabic Assessment of Auditory Skills development using children with Cochlear Implants.](#)

Int J Pediatr Otorhinolaryngol. 2019 Apr 2;122:52-59. doi: 10.1016/j.ijporl.2019.03.033. [Epub ahead of print]

INTRODUCTION: Audition is the gateway to spoken language, and infants' early accomplishments in acquiring the sound structure of their native language lays a critical ground work for subsequent learning. The development of pre-lingual auditory perceptual skills for cochlear implanted children is crucial for initial development of oral language.

OBJECTIVE: The aims of the present study were to validate the Egyptian Arabic Assessment of Auditory Skills, and to track the development of auditory skills in Egyptian children fitted with CI during the first three years post implantation.

METHODS: The study included 90 Arabic Egyptian children attending the phoniatic unit, Kasr El Aini hospital. Their chronological age ranged from 36 to 72 months. The study lasted for 18 months from July 2015 to January 2017. The children were divided into six groups according to their cochlear age i.e., amount of implant experience. An Arabic assessment chart of auditory skills was tailored that included six auditory skills' domains; detection, identification, short term auditory memory, supra-segmental discrimination, segmental discrimination and linguistic auditory processing. This chart was then used to develop an assessment tool which was then applied to all the study participants. All children had bilateral Sensorineural Hearing Loss (SNHL) since birth. None of the participants had prior Cochlear Implant (CI), but all had tried conventional hearing aids. All participants were implanted unilateral, with CI devices. All met selection criteria applied in the Egyptian national insurance committee for cochlear implantation.

RESULTS: All auditory skills domains improved with cochlear age. There was significant improvement between 1-6 and 7-12 months in the scores of the Detection (DET) domain. There was significant difference between 1-6 and 7-12 months, 7-12 and 13-18 months, 19-24 and 25-30 months in the scores of the Identification (IDENT) domain. Regarding the Short Term Auditory Memory (STAM) domain scores and the Supra-segmental Discrimination (SSD) domain scores there was significant difference between all the groups. Regarding the Segmental Discrimination (SGD) domain scores, there was significant difference between group 1-6 and 7-12 months, 7-12 and 13-18 months, 19-24 and 25-30 months, 25-30 and 31-36 months. Regarding the Linguistic Auditory Processing (LAP) domain, there was significant difference between group 1-6 and 7-12 months, 7-12 and 13-18 months, 25-30 and 31-36 months.

CONCLUSIONS: Children fitted with Cochlear Implants (CIs) appeared to show improvement in acquisition of auditory skills over a period of three years that followed a hierarchy of development dependent on the cochlear age.

Faes J, Gillis S.

[Auditory brainstem implantation in children with hearing loss: Effect on speech production.](#)

Int J Pediatr Otorhinolaryngol. 2019 Apr;119:103-112. doi: 10.1016/j.ijporl.2019.01.014. Epub 2019 Jan 14.

ABSTRACT: Auditory brainstem implantation (ABI) is a recent technique in children's hearing restoration. Up till now the focus in the literature has mainly been the perceptual outcomes after implantation, whereas the effect of ABI on spoken language is still an almost unexplored area of research. This study presents a one-year follow-up of the volubility of two children with ABI. The volubility of signed and oral productions is investigated and oral productions are examined in more detail. Results show clear developmental trends in both children, indicating a beneficial effect of ABI on spoken language development.

Farrell AN, Landry AM, Yee ME, Leu RM, Goudy SL.

[Sensorineural hearing loss in children with sickle cell disease.](#)

Int J Pediatr Otorhinolaryngol. 2019 Mar;118:110-114. doi: 10.1016/j.ijporl.2018.12.002. Epub 2018 Dec 5.

INTRODUCTION: Sensorineural hearing loss (SNHL) has been reported to occur at increased frequency in the pediatric sickle cell disease (SCD) population, likely secondary to ototoxic medication regimens and repeat sickling events that lead to end organ damage. Risk and protective factors of SNHL in this population are not fully characterized. The objective of this study was to describe audiology results in children with SCD and the prevalence and sequelae of SNHL.

METHODS: A comprehensive clinical database of 2600 pediatric SCD patients treated at 1 institution from 2010-16 was retrospectively reviewed to identify all patients who were referred for audiological testing. Audiologic test results, patient characteristics, and SCD treatments were reviewed.

RESULTS: 181 SCD children (97 male, 153 HbSS) underwent audiological testing, with 276 total audiology encounters, ranging 1-9 per patient. Mean age at first audiogram was 8.9 ± 5.2 years. 29.8% had prior cerebrovascular infarct and an additional 25.4% had prior abnormal transcranial Doppler screens documented at time of first audiogram. Overall, 13.3% had documented hearing loss, with 6.6% SNHL. Mean pure tone average (PTA) among patients with SNHL ranged from mild to profound hearing loss (Right: 43.3 ± 28.9 , Left: 40.8 ± 29.7), sloping to more severe hearing loss at higher frequencies.

CONCLUSIONS: Hearing loss was identified in a significant subset of children with SCD and the hearing loss ranged from normal to profound. Though the overall prevalence of SNHL in SCD patients was low, baseline audiology screening should be considered.

Fitzgerald MP, Reynolds A, Garvey CM, Norman G, King MD, Hayes BC.

[Hearing impairment and hypoxia ischaemic encephalopathy: Incidence and associated factors.](#)

Eur J Paediatr Neurol. 2019 Jan;23(1):81-86. doi: 10.1016/j.ejpn.2018.10.002. Epub 2018 Oct 10.

OBJECTIVE: To establish the local incidence of hearing loss in newborns with Hypoxic Ischaemic Encephalopathy (HIE) and to identify associated risk factors.

STUDY DESIGN: Retrospective Cohort Study. Neonatal Intensive Care Unit (NICU) dual stage hearing screening protocol, including automated otoacoustic emissions (AOAE) and automated auditory brainstem response (AABR) testing.

RESULTS: 57 newborns received therapeutic hypothermia for HIE. Twelve babies (21%) died. Audiology data was incomplete in 3 babies. Complete data was available for 42 babies (male $n = 24$), 4 (9.5%) of whom had hearing impairment. The development of hearing loss was associated with abnormal blood glucose levels ($p = 0.006$), low Apgar score at 1 min ($p = 0.0219$) and evidence of multi organ dysfunction [high creatinine ($p = 0.0172$ and 0.0198) and raised liver transaminases (aspartate aminotransferase (AST) $p = 0.0012$, alanine aminotransferase (ALT) $p = 0.0037$)]. An association with gentamicin was not found.

CONCLUSION: This study confirms that hearing impairment is common in term infants who have undergone therapeutic hypothermia for moderate/severe HIE. Blood glucose should be monitored carefully in these infants and developmental surveillance should include formal audiology. Further larger studies are needed to clarify the role, if any, of hypothermia per se in causation of hearing loss and to fully identify risk factors for hearing impairment in this population.

WHAT IS NEW: The current study confirms that hearing impairment is common in term infants who have undergone therapeutic hypothermia for moderate/severe HIE. No association between gentamicin use and the development of hearing impairment was found however initial blood glucose outside the normal range was of significance. Other factors associated with hearing impairment were low Apgar scores, greater need for resuscitation and evidence of multi organ dysfunction (renal and liver failure).

Fitzpatrick EM, Gaboury I, Durieux-Smith A, Coyle D, Whittingham J, Salamatmanesh M, Lee R, Fitzpatrick J.

[Parent Report of Amplification Use in Children with Mild Bilateral or Unilateral Hearing Loss.](#)

J Am Acad Audiol. 2019 Feb;30(2):93-102. doi: 10.3766/jaaa.17020. Epub 2018 Jan 15.

BACKGROUND: Amplification is considered to be one of the most important interventions for children with hearing loss. However, achieving consistent use of hearing technology in young children is an important problem, particularly when hearing loss is of mild degree. Little information is available about amplification use specifically for children with mild bilateral or unilateral hearing loss when such losses are targeted and identified early because of the availability of newborn hearing screening.

PURPOSE: We examined amplification use in a contemporary cohort of early-identified children with mild bilateral and unilateral hearing loss.

RESEARCH DESIGN: As part of the Mild and Unilateral Hearing Loss in Children Study, we collected parent reports on their child's use of amplification during the preschool years.

STUDY SAMPLE: A total of 69 children (38 unilateral and 31 bilateral mild) enrolled in the study from 2010 to 2015. Children entered the study at various ages between 12 and 36 mo of age and were followed up to age 48

mo. The median age of the children at enrollment was 16.5 mo (interquartile range [IQR] = 9.5, 26.8). Hearing loss was confirmed in these children at a median age of 3.6 mo (IQR = 2.4, 5.7).

DATA COLLECTION AND ANALYSIS: Baseline characteristics related to the child and family were collected through an intake form at study enrollment. Data on amplification fitting and use were collected via parent questionnaires at each assessment interval. Information from parent questionnaires was summarized descriptively and amplification use was grouped into categories. Through logistic regression, we examined the relationship between amplification use and laterality of hearing loss, sex, and maternal education.

RESULTS: Amplification was recommended for 59 (85.5%) children at a median age of 6.5 mo (IQR = 3.6, 21.2) and children were fitted at a median age of 10.9 mo (IQR = 6.0, 22.1). Based on parent report, hearing aid use was consistent for 39 (66.1%) of 59 children who had amplification recommended. Parent questionnaires showed very little change in use for most of the children over the study period. More children with bilateral hearing loss used their amplification consistently than those with unilateral hearing loss. After adjusting for maternal education and sex of the child, the odds for consistent use in children with mild bilateral loss was almost seven times higher (odds ratio = 6.75; 95% confidence interval = 1.84, 24.8) than for those with unilateral loss.

CONCLUSIONS: Although 85.5% of children with mild bilateral or unilateral hearing loss received amplification recommendations, only two-thirds achieved consistent use by age 3–4 yr based on parent report. Children with mild bilateral loss were more likely to use amplification during the preschool years than those with unilateral loss.

Fitzpatrick EM, Gaboury I, Durieux-Smith A, Coyle D, Whittingham J, Nassrallah F.

Auditory and language outcomes in children with unilateral hearing loss.

Hear Res. 2019 Feb;372:42-51. doi: 10.1016/j.heares.2018.03.015. Epub 2018 Mar 13.

OBJECTIVES: Children with unilateral hearing loss (UHL) are being diagnosed at younger ages because of newborn hearing screening. Historically, they have been considered at risk for difficulties in listening and language development. Little information is available on contemporary cohorts of children identified in the early months of life. We examined auditory and language acquisition outcomes in a contemporary cohort of early-identified children with UHL and compared their outcomes at preschool age with peers with mild bilateral loss and with normal hearing.

DESIGN: As part of the Mild and Unilateral Hearing Loss in Children Study, we collected auditory and spoken language outcomes on children with unilateral, bilateral hearing loss and with normal hearing over a four-year period. This report provides a cross-sectional analysis of results at age 48 months. A total of 120 children (38 unilateral and 31 bilateral mild, 51 normal hearing) were enrolled in the study from 2010 to 2015. Children started the study at varying ages between 12 and 36 months of age and were followed until age 36–48 months. The median age of identification of hearing loss was 3.4 months (IQR: 2.0, 5.5) for unilateral and 3.6 months (IQR: 2.7, 5.9) for the mild bilateral group. Families completed an intake form at enrolment to provide baseline child and family-related characteristics. Data on amplification fitting and use were collected via parent questionnaires at each annual assessment interval. This study involved a range of auditory development and language measures. For this report, we focus on the end of follow-up results from two auditory development questionnaires and three standardized speech-language assessments. Assessments included in this report were completed at a median age of 47.8 months (IQR: 38.8, 48.5). Using ANOVA, we examined auditory and language outcomes in children with UHL and compared their scores to children with mild bilateral hearing loss and those with normal hearing.

RESULTS: On most measures, children with UHL performed poorer than those in the mild bilateral and normal hearing study groups. All children with hearing loss performed at lower levels compared to the normal hearing control group. However, mean standard scores for the normal hearing group in this study were above normative means for the language measures. In particular, children with UHL showed gaps compared to the normal hearing control group in functional auditory listening and in receptive and expressive language skills (three quarters of one standard deviation below) at age 48 months. Their performance in receptive vocabulary and speech production was not significantly different from that of their hearing peers.

CONCLUSIONS: Even when identified in the first months of life, children with UHL show a tendency to lag behind their normal hearing peers in functional auditory listening and in receptive and expressive language development.

Fu Y, Zha S, Lü N, Xu H, Zhang X, Shi W, Zha J.

Carrier frequencies of hearing loss variants in newborns of China: A meta-analysis.

J Evid Based Med. 2019 Feb;12(1):40-50. doi: 10.1111/jebm.12305. Epub 2018 Jul 2.

OBJECTIVE: The objective of this study was to review the carrier frequencies of hearing loss gene variants, such as GJB2, SLC26A4, and MT-RNR1 in newborns of China.

DESIGN: PubMed, Embase, BioCentral, CNKI, WanFang, and VIP databases were used for searching relevant literature studies published during the period of January 2007 and January 2016. Meta-analysis was performed

by using the R software. The estimated rate and its 95% confidence intervals (CI) of the relevant indexes in newborns were collected and calculated using a fixed-effects model or a random-effects model when appropriate.

RESULTS: In total, 35 of 958 published literature studies in Chinese and English were selected. The overall results showed that in newborns of China, the carrier frequencies of GJB2 variants (235 delC, 299 delAT) were 1.64% (95% CI 1.52% to 1.77%) and 0.33% (95% CI 0.19% to 0.51%); SLC26A4 variants (IVS7-2 A > G, 2168 A > G) were 1.02% (95% CI 0.91% to 1.15%) and 0.14% (95% CI 0.06% to 0.25%); MT-RNR1 variants (1555 A > G, 1449 C > T) were 0.20% (95% CI 0.17% to 0.23%) and 0.03% (95% CI 0.02% to 0.05%).

CONCLUSIONS: There are high carrier frequencies of GJB2 variants among newborns in China, followed by SLC26A4 and MT-RNR1 variants. In order to achieve “early detection, early diagnosis and early treatment” and reduce the incidence of hereditary hearing loss in offspring, a comprehensive combination of neonatal hearing screening and deafness gene detection should be recommended and implemented in China.

Funamura JL, Lee JW, McKinney S, Bayoumi AG, Senders CW, Tollefson TT.

[Children with Cleft Palate: Predictors of Otologic Issues in the First 10 Years.](#)

Otolaryngol Head Neck Surg. 2019 Jan 22;194599818825461. doi: 10.1177/0194599818825461. [Epub ahead of print]

OBJECTIVE: To evaluate the characteristics of children with cleft palate associated with persistent otologic issues in the first 10 years of life.

STUDY DESIGN: Case series with chart review.

SETTING: Single academic center.

SUBJECTS AND METHODS: Children born with cleft palate from 2003 to 2007 and treated by the UC Davis Cleft and Craniofacial Team between January 2003 and December 2017 were included in the study. Data from 143 patients were analyzed via Wilcoxon rank sum and Fisher exact tests for univariate analysis and logistic regression to determine adjusted odds ratios.

RESULTS: The median length of follow-up was 9.9 years, and the age at last ear examination was 10.7 years. At the last evaluation, unresolved otologic issues were common, with at least 1 ear having a tympanic membrane (TM) perforation (16.1%), a tympanostomy tube (36.2%), or conductive hearing loss (23.1%). After adjusting for demographic and clinical characteristics, history of palate revision or speech surgery was associated with having a TM perforation ($P = .02$). The only clinical variables associated with conductive hearing loss was the presence of a TM perforation ($P < .01$) or a genetic abnormality ($P = .02$). Severity of palatal clefting was not associated with specific otologic or audiologic outcomes after adjusting for other characteristics.

CONCLUSION: A large proportion of children with cleft palate have persistent otologic issues at age 10 years and would benefit from continued close monitoring well after the age when most children have normalized eustachian tube function. Prolonged otologic issues were not found to be associated with cleft type.

Goldsworthy RL, Markle KL.

[Pediatric Hearing Loss and Speech Recognition in Quiet and in Different Types of Background Noise.](#)

J Speech Lang Hear Res. 2019 Mar 25;62(3):758-767. doi: 10.1044/2018_JSLHR-H-17-0389.

Purpose: Speech recognition deteriorates with hearing loss, particularly in fluctuating background noise. This study examined how hearing loss affects speech recognition in different types of noise to clarify how characteristics of the noise interact with the benefits listeners receive when listening in fluctuating compared to steady-state noise.

Method: Speech reception thresholds were measured for a closed set of spondee words in children (ages 5-17 years) in quiet, speech-spectrum noise, 2-talker babble, and instrumental music. Twenty children with normal hearing and 43 children with hearing loss participated; children with hearing loss were subdivided into groups with cochlear implant (18 children) and hearing aid (25 children) groups. A cohort of adults with normal hearing was included for comparison.

Results: Hearing loss had a large effect on speech recognition for each condition, but the effect of hearing loss was largest in 2-talker babble and smallest in speech-spectrum noise. Children with normal hearing had better speech recognition in 2-talker babble than in speech-spectrum noise, whereas children with hearing loss had worse recognition in 2-talker babble than in speech-spectrum noise. Almost all subjects had better speech recognition in instrumental music compared to speech-spectrum noise, but with less of a difference observed for children with hearing loss.

Conclusions: Speech recognition is more sensitive to the effects of hearing loss when measured in fluctuating compared to steady-state noise. Speech recognition measured in fluctuating noise depends on an interaction of hearing loss with characteristics of the background noise; specifically, children with hearing loss were able to derive a substantial benefit for listening in fluctuating noise when measured in instrumental music compared to 2-talker babble.

Graham C, Seeley J, Gina A, Saman Y.

[Mapping the content of mothers' knowledge, attitude and practice towards universal newborn hearing screening for development of a KAP survey tool.](#)

PLoS One. 2019 Feb 20;14(2):e0210764. doi: 10.1371/journal.pone.0210764. eCollection 2019.

ABSTRACT: Understanding mother's knowledge, attitude and practice (KAP) of permanent childhood hearing impairment (PCHI) is essential for the success of universal newborn hearing screening (UNHS) as poor compliance and follow-up remains a global challenge. To determine content area for a questionnaire that measures PCHI-related KAP in rural mothers, we trained moderators who interviewed 145 pregnant women (17 groups) from 5 ante-natal clinics. Interviews were recorded, transcribed, summarised and analysed using thematic framework analysis. Four knowledge themes were identified: 1) PCHI was perceived as the malfunction of hearing leading to disability; 2) a poorly-responsive/communicative child may have PCHI; 3) lifestyle, hereditary and environmental factors are significant causes of PCHI; 4) medical management of PCHI was doubted, with some advocating birth and ancestral rituals. Two themes were identified for attitude: 1) beliefs that PCHI was emotionalised due to the negative lifelong impact on the child and family; 2) UNHS processes were favourable though some preferred other belief systems. Three themes were identified for practice: 1) doctors were the first choice followed by traditional healers; 2) willingness to continue follow-up although challenges exist; 3) minimal family support during consultation. The contextualised KAP of women regarding UNHS processes and PCHI provided content area for the design of a KAP tool.

Han JJ, Nguyen PD, Oh DY, Han JH, Kim AR, Kim MY, Park HR, Tran LH, Dung NH, Koo JW, Lee JH, Oh SH, Anh Vu H, Choi BY.

[Elucidation of the unique mutation spectrum of severe hearing loss in a Vietnamese pediatric population.](#)

Sci Rep. 2019 Feb 7;9(1):1604. doi: 10.1038/s41598-018-38245-4.

ABSTRACT: The mutational spectrum of deafness in Indochina Peninsula, including Vietnam, remains mostly undetermined. This significantly hampers the progress toward establishing an effective genetic screening method and early customized rehabilitation modalities for hearing loss. In this study, we evaluated the genetic profile of severe-to-profound hearing loss in a Vietnamese pediatric population using a hierarchical genetic analysis protocol that screened 11 known deafness-causing variants, followed by massively parallel sequencing targeting 129 deafness-associated genes. Eighty-seven children with isolated severe-to-profound non-syndromic hearing loss without family history were included. The overall molecular diagnostic yield was estimated to be 31.7%. The mutational spectrum for severe-to-profound non-syndromic hearing loss in our Vietnamese population was unique: The most prevalent variants resided in the MYO15A gene (7.2%), followed by GJB2 (6.9%), MYO7A (5.5%), SLC26A4 (4.6%), TMC1 (1.8%), ESPN (1.8%), POU3F4 (1.8%), MYH14 (1.8%), EYA1 (1.8%), and MR-RNR1 (1.1%). The unique spectrum of causative genes in the Vietnamese deaf population was similar to that in the southern Chinese deaf population. It is our hope that the mutation spectrum provided here could aid in establishing an efficient protocol for genetic analysis of severe-to-profound hearing loss and a customized screening kit for the Vietnamese population.

Hilditch C, Liersch B, Spurrier N, Callander EJ, Cooper C, Keir AK.

[Does screening for congenital cytomegalovirus at birth improve longer term hearing outcomes?](#)

Arch Dis Child. 2018 Oct;103(10):988-992. doi: 10.1136/archdischild-2017-314404. Epub 2018 Apr 28.

ABSTRACT: Currently, the diagnosis of congenital cytomegalovirus (cCMV) infection in most highly resourced countries is based on clinical suspicion alone. This means only a small proportion of cCMV infections are diagnosed. Identification, through either universal or targeted screening of asymptomatic newborns with cCMV, who would previously have gone undiagnosed, would allow for potential early treatment with antiviral therapy, ongoing audiological surveillance and early intervention if sensorineural hearing loss (SNHL) is identified. This paper systematically reviews published papers examining the potential benefits of targeted and universal screening for newborn infants with cCMV. We found that the treatment of these infants with antiviral therapy remains controversial, and clinical trials are currently underway to provide further answers. The potential benefit of earlier identification and intervention (eg, amplification and speech therapy) of children at risk of later-onset SNHL identified through universal screening is, however, clearer.

Hoffman HJ, Dobie RA, Losonczy KG, Themann CL, Flamme GA.

[Kids Nowadays Hear Better Than We Did: Declining Prevalence of Hearing Loss in US Youth, 1966-2010.](#)

Laryngoscope. 2018 Oct 5. doi: 10.1002/lary.27419. [Epub ahead of print]

OBJECTIVES/HYPOTHESIS: To investigate factors associated with hearing impairment (HI) in adolescent youths during the period 1966-2010.

STUDY DESIGN: Cross-sectional analyses of US sociodemographic, health, and audiometric data spanning 5 decades.

METHODS: Subjects were youths aged 12 to 17 years who participated in the National Health Examination

Survey (NHES Cycle 3, 1966-1970; n=6,768) and youths aged 12 to 19 years in the Third National Health and Nutrition Examination Survey (NHANES III, 1988-1994; n=3,057) and NHANES (2005-2010; n=4,374). HI prevalence was defined by pure-tone average (PTA) ≥ 20 dB HL for speech frequencies (0.5, 1, 2, and 4 kHz) and high frequencies (3, 4, and 6 kHz). Multivariable logistic models were used to estimate the odds ratio (OR) and 95% confidence interval (CI).

RESULTS: Overall speech-frequency HI prevalence was 10.6% (95% CI: 9.7%-11.6%) in NHES, 3.9% (95% CI: 2.8%-5.5%) in NHANES III, and 4.5% (95% CI: 3.7%-5.4%) in NHANES 2005 to 2010. The corresponding high-frequency HI prevalences were 32.8% (95% CI: 30.8%-34.9%), 7.3% (95% CI: 5.9%-9.0%), and 7.9% (95% CI: 6.8%-9.2%). After adjusting for sociodemographic factors, overall high-frequency HI was increased twofold for males and cigarette smoking. Other significant risk factors in NHANES 2005 to 2010 included very low birth weight, history of ear infections/otitis media, ear tubes, fair/poor general health, and firearms use.

CONCLUSIONS: HI declined considerably between 1966 to 1970 and 1988 to 1994, with no additional decline between 1988 to 1994 and 2005 to 2010. Otitis media history was a significant HI risk factor each period, whereas very low birth weight emerged as an important risk factor after survival chances improved. Reductions in smoking, job-related noise, and firearms use may partially explain the reduction in high-frequency HI. Loud music exposure may have increased, but does not account for HI differences.

Hollanders JJ, Schaëfer N, van der Pal SM, Oosterlaan J, Rotteveel J, Finken MJJ; on behalf of the Dutch POPS-19 Collaborative Study Group.

[Long-Term Neurodevelopmental and Functional Outcomes of Infants Born Very Preterm and/or with a Very Low Birth Weight.](#)

Neonatology. 2019 Mar 5;115(4):310-319. doi: 10.1159/000495133. [Epub ahead of print]

BACKGROUND: Birth weight (BW) is often used as a proxy for gestational age (GA) in studies on preterm birth. Recent findings indicate that, in addition to perinatal outcomes, subjects born very preterm (VP; GA < 32 weeks) differ from those with a very low birth weight (VLBW; BW < 1,500 g) in postnatal growth up to their final height.

OBJECTIVE: To study whether neurodevelopmental and functional outcomes at the age of 19 years differ in VP and/or VLBW subjects.

METHODS: 705 19-year-old subjects from the Project on Preterm and Small-for-Gestational-Age Infants (POPS) cohort were classified as (1) VP+/VLBW+ (n = 354), (2) VP+/VLBW- (n = 144), or (3) VP-/VLBW+ (n = 207), and compared with regard to IQ as assessed with the Multicultural Capacity Test-intermediate level; neuromotor function using Touwen's examination of mild neurologic dysfunction; hearing loss; self- and parent-reported behavioral and emotional functioning; educational achievement and occupation; and self-assessed health using the Health Utilities Index and the London Handicap Scale.

RESULTS: VP+/VLBW- infants, on average, had 3.8-point higher IQ scores (95% confidence interval [CI] 0.5-7.1), a trend towards higher educational achievement, 3.3-dB better hearing (95% CI 1.2-5.4), and less anxious behavior, attention problems, and internalizing behavior than to VP+/VLBW+ subjects. VP-/VLBW+ infants reported 1.8 increased odds (95% CI 1.2-2.6) of poor health compared to VP+/VLBW+ subjects.

CONCLUSIONS: At the age of 19 years, subjects born VP+/VLBW+, VP+/VLBW-, and VP-/VLBW+ have different neurodevelopmental and functional outcomes, although effect sizes are small. Hence, the terms VP and VLBW are not interchangeable. We recommend, at least for industrialized countries, to base inclusion in future studies on preterm populations on GA instead of on BW.

Howell JB, Appelbaum EN, Armstrong MF, Chapman D, Dodson KM.

[An Analysis of Risk Factors in Unilateral Versus Bilateral Hearing Loss.](#)

Ear Nose Throat J. 2019 Apr 15;145561319840578. doi: 10.1177/0145561319840578. [Epub ahead of print]

ABSTRACT: A retrospective review of children with confirmed hearing loss identified through universal newborn hearing screening (UNHS) in Virginia from 2010 to 2014 was conducted in order to compare the incidence of Joint Committee on Infant Hearing (JCIH) risk factors in children with unilateral hearing loss (UHL) to bilateral hearing loss (BHL). Over the 5-year study period, 1004 children (0.20% of all births) developed a confirmed hearing loss, with 544 (51%) children having at least one JCIH risk factor. Overall, 18% of children with confirmed hearing loss initially passed UNHS. Of all children with risk factors, 226 (42%) demonstrated UHL and 318 (58%) had BHL. The most common risk factors for UHL were neonatal indicators (69%), craniofacial anomalies (30%), stigmata of HL syndromes (14%), and family history (14%). The most common risk factors in BHL were neonatal indicators (49%), family history (27%), stigmata of HL syndromes (19%), and craniofacial anomalies (16%). Children with the risk factor for positive family history were more likely to have BHL, while those with craniofacial anomalies were more likely to have UHL (P < .001). Neonatal indicators were the most commonly identified risk factor in both UHL and BHL populations. Children with UHL were significantly more likely to have craniofacial anomalies, while children with BHL were more likely to have a family history of hearing loss. Further studies, assessing the etiology underlying the hearing loss and risk factor associations are warranted.

Iwanicka-Pronicka K, Ciara E, Piekutowska-Abramczuk D, Halat P, Pajdowska M, Pronicki M.

[Congenital cochlear deafness in mitochondrial diseases related to RRM2B and SERAC1 gene defects. A study of the mitochondrial patients of the CMHI hospital in Warsaw, Poland.](#)

Int J Pediatr Otorhinolaryngol. 2019 Mar 16;121:143-149. doi: 10.1016/j.ijporl.2019.03.015. [Epub ahead of print]

OBJECTIVES: Although hearing loss is a well-known symptom of mitochondria-related disorders, it is not clear how often it is a congenital and cochlear impairment. The Newborn Hearing Screening Program (NHSP) enables to distinguish congenital cochlear deafness from an acquired hearing deficit. The initial aim of the study was to research the frequency of the congenital cochlear hearing loss among patients with various gene defects resulting in mitochondrial disorders. The research process brought on an additional gain: basing on our preliminary study group of 80 patients, in 12 patients altogether we identified two defected genes responsible for mitochondrial disorders, whose carriers did not pass the NHSP. Finally, these patients were diagnosed with the congenital cochlear deafness.

MATERIAL AND METHODS: The results of the NHSP in the patients with mitochondrial disorders diagnosed in our tertiary reference center were analyzed. Only the cases with confirmed mutations were qualified for the study group. The NHSP database included 80 patients with mutations in 31 different genes: 25 nuclear-encoded and 6 mtDNA-encoded. We searched the literature for the presence of a congenital hearing impairment (CHI) in mitochondrial disorders caused by changes in 278 already known genes.

RESULTS: For 68 patients from the study group the NHSP test indicated a proper cochlear function and thus suggested normal hearing. For 12 mitochondrial patients, the NHSP test indicated the requirement for the further audiological diagnosis, and finally CHI was confirmed in 8 of them. This latter subset included patients with pathogenic variants in RRM2B and SERAC1, known as “deafness-causing genes”. Contrary to our initial expectations, the patients carrying mutations in other “deafness-causing genes”: MPV17, POLG, COX10, as well as other mitochondria-related genes, all reported in literature, did not indicate any CHI following the NHSP test.

CONCLUSION: Our study indicates that the cochlear CHI is a phenotypic feature of the RRM2B and SERAC1 related defects. The diagnosis of the CHI following the NHSP allows to early distinguish those defects from other mitochondria-related disorders in which the NHSP test result is correct. Wider studies are needed to assess the significance of this observation.

Jabbour N, Weinreich HM, Owusu J, Lehn M, Yueh B, Levine S.

[Hazardous noise exposure from noisy toys may increase after purchase and removal from packaging: A call for advocacy.](#)

Int J Pediatr Otorhinolaryngol. 2019 Jan;116:84-87. doi: 10.1016/j.ijporl.2018.10.028. Epub 2018 Oct 22.

OBJECTIVE: Previous studies identified hazardous noise levels from packaged toys. Sound levels may increase when packaging is removed and therefore, complicate the ability to accurately assess noise levels before purchase. The goal of this study was to evaluate how packaging affects the decibel (dB) level of toys by: 1) Assessing dB level of toys with and without packaging. 2) Evaluating the percentage of packaged and unpackaged toys that exceed a safety limit of 85 dB.

METHODS: Thirty-five toys were selected from the 2009-2011 Sight and Hearing Association (SHA) based on availability for purchase. Toys' speakers were categorized as Exposed, Partially Exposed, or Covered, based on its packaging. The dB level of each toy was tested at 0 cm and 25 cm from the speaker using a handheld digital sound meter in a standard audiometric booth. T tests and ANOVA were performed to assess mean change in sound level before and after packaging removal.

RESULTS: Significant dB increases were noted after packaging was removed (mean change 11.9 dB at 0 cm; and 2.5 dB at 25 cm, $p < 0.001$). Sixty-four percentage of Covered toys ($n = 14$) had dB greater than 85 dB when packaged and this increased to 100% when unpackaged.

CONCLUSION: Many manufactured toys have hazardous sound levels. Caregivers and healthcare providers should be aware that toys tested in the store may actually be louder when brought home and removed from their packaging. Limits on and disclosure of dB level of toys should be considered nationally.

Jackson W, Taylor G, Selewski D, Smith PB, Tolleson-Rinehart S, Laughon MM.

[Association between furosemide in premature infants and sensorineural hearing loss and nephrocalcinosis: a systematic review.](#)

Matern Health Neonatol Perinatol. 2018 Nov 19;4:23. doi: 10.1186/s40748-018-0092-2. eCollection 2018.

ABSTRACT: Furosemide is a potent loop diuretic commonly and variably used by neonatologists to improve oxygenation and lung compliance in premature infants. There are several safety concerns with use of furosemide in premature infants, specifically the risk of sensorineural hearing loss (SNHL), and nephrocalcinosis/nephrolithiasis (NC/NL). We conducted a systematic review of all trials and observational studies examining the association between these outcomes with exposure to furosemide in premature infants. We searched MEDLINE, EMBASE, CINAHL, and clinicaltrials.gov. We included studies reporting either SNHL or NC/NL in premature

infants (<37 weeks completed gestational age) who received at least one dose of enteral or intravenous furosemide. Thirty-two studies met full inclusion criteria for the review, including 12 studies examining SNHL and 20 studies examining NC/NL. Only one randomized controlled trial was identified in this review. We found no evidence that furosemide exposure increases the risk of SNHL or NC/NL in premature infants, with varying quality of studies and found the strength of evidence for both outcomes to be low. The most common limitation in these studies was the lack of control for confounding factors. The evidence for the risk of SNHL and NC/NL in premature infants exposed to furosemide is low. Further randomized controlled trials of furosemide in premature infants are urgently needed to adequately assess the risk of SNHL and NC/NL, provide evidence for improved FDA labeling, and promote safer prescribing practices.

Judge PD, Jorgensen E, Lopez-Vazquez M, Roush P, Page TA, Moeller MP, Tomblin JB, Holte L, Buchman C.
[Medical Referral Patterns and Etiologies for Children With Mild-to-Severe Hearing Loss.](#)

Ear Hear. 2018 Dec 6. doi: 10.1097/AUD.0000000000000682. [Epub ahead of print]

OBJECTIVES: To (1) identify the etiologies and risk factors of the patient cohort and determine the degree to which they reflected the incidence for children with hearing loss and (2) quantify practice management patterns in three catchment areas of the United States with available centers of excellence in pediatric hearing loss.

DESIGN: Medical information for 307 children with bilateral, mild-to-severe hearing loss was examined retrospectively. Children were participants in the Outcomes of Children with Hearing Loss (OCHL) study, a 5-year longitudinal study that recruited subjects at three different sites. Children aged 6 months to 7 years at time of OCHL enrollment were participants in this study. Children with cochlear implants, children with severe or profound hearing loss, and children with significant cognitive or motor delays were excluded from the OCHL study and, by extension, from this analysis. Medical information was gathered using medical records and participant intake forms, the latter reflecting a caregiver's report. A comparison group included 134 children with normal hearing. A Chi-square test on two-way tables was used to assess for differences in referral patterns by site for the children who are hard of hearing (CHH). Linear regression was performed on gestational age and birth weight as continuous variables. Risk factors were assessed using t tests. The alpha value was set at $p < 0.05$.

RESULTS: Neonatal intensive care unit stay, mechanical ventilation, oxygen requirement, aminoglycoside exposure, and family history were correlated with hearing loss. For this study cohort, congenital cytomegalovirus, strep positivity, bacterial meningitis, extracorporeal membrane oxygenation, and loop diuretic exposure were not associated with hearing loss. Less than 50% of children underwent imaging, although 34.2% of those scanned had abnormalities identified. No single imaging modality was preferred. Differences in referral rates were apparent for neurology, radiology, genetics, and ophthalmology.

CONCLUSIONS: The OCHL cohort reflects known etiologies of CHH. Despite available guidelines, centers of excellence, and high-yield rates for imaging, the medical workup for children with hearing loss remains inconsistently implemented and widely variable. There remains limited awareness as to what constitutes appropriate medical assessment for CHH.

Kanabur P, Hubbard C, Jeyakumar A.

[Clinical Guidelines in Pediatric Hearing Loss: Systemic Review Using the Appraisal of Guidelines for Research and Evaluation II Instrument.](#)

Laryngoscope. 2018 Dec 8. doi: 10.1002/lary.27722. [Epub ahead of print]

OBJECTIVES: Despite the importance, impact, and prevalence of pediatric hearing loss (HL), there are very few published clinical practice guidelines (CPG) supporting the evaluation and management of pediatric patients with HL. Our objective was to appraise existing CPGs to ensure safe and effective practices.

METHODS: A literature search was conducted in PubMed, Google Scholar, EBSCO, as well as a manual Google search. Three independent assessors using the Appraisal of Guidelines for Research and Evaluation II (AGREE II) instrument evaluated CPGs related to HL in children. Standardized domain scores were calculated for each guideline.

RESULTS: A total of four guidelines met the inclusion criteria and were appraised. Scope and purpose achieved a high median score of 83%. Stakeholder involvement, clarity of presentation, and editorial independence achieved intermediate scores of 67%, 54%, and 50%, respectively. The areas that required most improvement and achieved low scores were rigor of development and applicability, with scores of 22% and 38%, respectively. Based on the AGREE II measures, the four guidelines had domain scores less than 60% for each domain, and without modification no guideline could be recommended.

CONCLUSIONS: Based on the AGREE II, the qualities of CPGs for pediatric HL have several shortcomings, and the need for a comprehensive CPG remains. Rigor of development and applicability present the greatest opportunities for improvement of these CPGs.

Kanji A, Krabbenhoft K.

[Audiological follow-up in a risk-based newborn hearing screening programme: An exploratory study of the influencing factors.](#)

S Afr J Commun Disord. 2018 Oct 25;65(1):e1-e7. doi: 10.4102/sajcd.v65i1.587.

BACKGROUND: Follow-up return rate in Early Hearing Detection and Intervention (EHDI) programmes is of specific importance as it ensures that benchmarks are met and that no child with suspected hearing loss is left unidentified.

OBJECTIVES: The aim of this study was to determine the factors influencing audiological follow-up of high-risk infants in a risk-based newborn hearing screening programme.

METHOD: A non-experimental, exploratory, qualitative research design was employed. Purposive sampling was used. The study was conducted at a secondary level hospital in the public health care sector in South Africa. Participants comprised 10 caregivers (age range 26-40 years) of infants who had been enrolled in a risk-based newborn hearing screening programme, and returned for follow-up appointments. Data were collected using semi-structured interviews. Responses were recorded by the researcher and a colleague to ensure rigour and trustworthiness of findings. Data were analysed using thematic analysis for open-ended questions and descriptive statistics for the closed-ended questions.

RESULTS: The most common positive contributors that facilitated participants' attendance at follow-up appointments were: having friendly audiologists; a clear line of communication between caregiver and audiologist and a reminder of the appointment. The most significant perceived challenge that participants described in returning for the follow-up appointment was living in far proximity from the hospital.

CONCLUSION: Findings of the study revealed that influencing factors on follow-up return rate are demographic, socio-economic, and interpersonal in nature and further suggested the need for an all-inclusive appointment day. It may be of importance to not only look at what is being done to improve the follow-up return rate but also how it should be done in terms of professional-to-patient communication and interactions.

Kanji A, Khoza-Shangase K, Moroe N.

[Newborn hearing screening protocols and their outcomes: A systematic review.](#)

Int J Pediatr Otorhinolaryngol. 2018 Dec;115:104-109. doi: 10.1016/j.ijporl.2018.09.026. Epub 2018 Sep 25.

OBJECTIVE: To conduct a review of the most current research in objective measures used within newborn hearing screening protocols with the aim of exploring the actual protocols in terms of the types of measures used and their frequency of use within a protocol, as well as their outcomes in terms of sensitivity, specificity, false positives, and false negatives in different countries worldwide.

METHODS: A systematic literature review was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis. Electronic databases such as PubMed, Google Scholar and Science Direct were used for the literature search. A total of 422 articles were identified, of which only 15 formed part of the current study. The 15 articles that met the study's criteria were reviewed. Pertinent data and findings from the review were tabulated and qualitatively analysed under the following headings: country; objective screening and/or diagnostic measures; details of screening protocol; results (including false positive and negative findings, sensitivity and/or specificity), conclusion and/or recommendations. These tabulated findings were then discussed with conclusions and recommendations offered.

RESULTS: Findings reported in this paper are based on a qualitative rather than a quantitative analysis of the reviewed data. Generally, findings in this review revealed firstly, that there is a lack of uniformity in protocols adopted within newborn hearing screening. Secondly, many of the screening protocols reviewed consist of two or more tiers or stages, with transient evoked otoacoustic emissions (TEOAEs) and automated auditory brainstem response (AABR) being most commonly used. Thirdly, DPOAEs appear to be less commonly used when compared to TEOAEs. Lastly, a question around routine inclusion of AABR as part of the NHS protocol remains inconclusively answered.

CONCLUSIONS: There is sufficient evidence to suggest that the inclusion of AABR within a NHS programme is effective in achieving better hearing screening outcomes. The use of AABR in combination with OAEs within a test-battery approach or cross-check principle to screening is appropriate, but the inclusion of AABR to facilitate appropriate referral for diagnostic assessment needs to be systematically studied.

Kapitanova M, Knebel JF, El Ezzi O, Artaz M, de Buys Roessingh AS, Richard C.

[Influence of infancy care strategy on hearing in children and adolescents: A longitudinal study of children with unilateral lip and /or cleft palate.](#)

Int J Pediatr Otorhinolaryngol. 2018 Nov;114:80-86. doi: 10.1016/j.ijporl.2018.08.031. Epub 2018 Aug 27.

OBJECTIVES: To evaluate the relation between ventilation tube insertion, otitis media with effusion duration and otologic outcomes in unilateral cleft lip and/or cleft palate children from infancy to teenage age.

DESIGN AND POPULATION: Retrospective longitudinal charts review of patients from the multidisciplinary cleft team of the University Hospital of Lausanne over a 30-year period. 146 charts from consecutive patients with

non-syndromic unilateral cleft lip and/or cleft palate who were born between January 1986 and January 2003 were included.

RESULTS: The earlier in life a cleft child experience his first otitis media with effusion (OME), the worse his long-term hearing will be. Along with the age of onset of OME, we disclosed an influence of the duration of OME without ventilation tube (VT) insertion on short and long-term hearing outcomes. Different patterns were observed between cleft palate (CP) and cleft lip palate children (CLP), with a higher incidence of otitis media with effusion for the CLP group than the CP group. Direct positive relationship between VT insertion and hearing were disclosed and evaluation of long-term complications did not reveal significant relation with VT insertion. Of note, OME in CLP children led to a higher rate (but not statistically significant) of chronic ear complications than in the CP group, that may indicate more persistent OME or different adverse effect on the middle ear mucosa between CP and CLP children.

CONCLUSIONS: Individualized counseling should take into account different factors such as the type of cleft, the age of onset of OME and duration of OME, keeping in mind the adverse effect of persistent middle ear fluid. In the present report, results prone an early ventilation tube insertion to prevent short and long-term injury to the middle ear homeostasis, hearing loss and related issues.

Karantk TK, Whittemore KR.

Middle-ear disease in children with cleft palate.

Auris Nasus Larynx. 2018 Dec;45(6):1143-1151. doi: 10.1016/j.anl.2018.04.012.

OBJECTIVE: The objective of this review is to summarize all aspects of middle ear diseases in children with cleft palate (CP).

METHODS: PubMed, Scopus, The Cumulative Index to Nursing and Allied Health Literature (CINAHL) and The Cochrane Library were searched for English-language randomized control trials (RCTs), meta-analyses, systematic reviews and observational studies published through 31st July 2017.

RESULTS: Epidemiology and pathogenesis of middle ear diseases in children with cleft palate have been discussed in this review. Methods of Evaluation, CP surgeries, complications and follow up have been detailed for the same.

CONCLUSION: Evaluation of middle-ear disease in children with CP begins at birth by a newborn hearing screen. Tympanometry and otoscopy helps screen for middle-ear disease during follow-up visits. Ventilation tube may be placed when indicated based on the patient's clinical course and presentation. Long-term follow up should be provided to look for the development of cholesteatoma.

Khoza-Shangase K.

Early hearing detection and intervention in South Africa: Exploring factors compromising service delivery as expressed by caregivers.

Int J Pediatr Otorhinolaryngol. 2019 Mar;118:73-78. doi: 10.1016/j.ijporl.2018.12.021. Epub 2018 Dec 18.

AIM: The main aim of this study was to explore factors compromising early intervention (EI) service delivery to hearing impaired children in South Africa, as expressed by their caregivers.

METHODS: Within a qualitative survey design, a sample of 19 hearing impaired children's caregivers completed structured self-administered questionnaires on factors that they perceive compromise EI for their children. These caregivers included mothers, fathers, grandparents, and legal guardians or adoptive parents of children with hearing impairment. Descriptive analysis of the data was undertaken.

RESULTS: Findings indicated various factors compromising EI as reported by caregivers. These included limited availability of appropriate schools and health care facilities for their hearing impaired children; long distances between the few services that are available and the places of residence of the service users; significant costs linked to the services (such as medical expenses, boarding school facilities costs); limited skills and knowledge of professionals and teachers regarding hearing impairment; inconsistent and conflicting professional opinions about the child's diagnosis and treatment; as well as limited community awareness about hearing impairment along with services available for hearing impaired children.

CONCLUSION: These findings have important clinical, training, policy, and advocacy implications within the South African context; if both access to and success within the EI services will be successful.

Kim SY, Choi BY, Jung EY, Park H, Yoo HN, Park KH.

Risk factors for failure in the newborn hearing screen test in very preterm twins.

Pediatr Neonatol. 2018 Dec;59(6):586-594. doi: 10.1016/j.pedneo.2018.01.014. Epub 2018 Jan 31.

BACKGROUND: We aimed to identify prenatal and postnatal risk factors associated with abnormal newborn hearing screen (NHS) results and subsequently confirmed sensorineural hearing loss (SNHL) in preterm twin neonates.

METHODS: Electronic medical records of 159 twin neonates who were born alive after ≤ 32 weeks were retrospectively reviewed for hearing loss in both ears. Histopathologic examination of the placenta was

performed and clinical data, including method of conception and factors specific to twins, were retrieved from a computerized perinatal database. The main outcome measure was failure to pass the NHS test. The generalized estimation equations model was used for twins.

RESULTS: Thirty-two neonates (20.1%) had a “refer” result, and, on the confirmation test, permanent SNHL was identified in 4.4% (7/159) of all neonates. Neonates who had a “refer” result on the NHS test were more likely to be of lower birth weight, more likely to have been conceived with the use of in vitro fertilization (IVF), and more likely to have higher rates of intraventricular hemorrhage (IVH) and bronchopulmonary dysplasia. However, monochorionic placentation, death of the co-twin, or being born first was not associated with a “refer” result on the NHS test. Multivariable logistic regression revealed that conception after IVF and the presence of IVH were the only variables to be statistically significantly associated with “refer” on the NHS test. No parameters studied were found to be significantly different between the SNHL and no SNHL groups, probably because of the relatively small number of cases of SNHL.

CONCLUSION: In preterm twin newborns, IVF and the presence of IVH were independently associated with an increased risk of abnormal NHS results, whereas the factors specific to twins were not associated with abnormal NHS results.

Kitao K, Mutai H, Namba K, Morimoto N, Nakano A, Arimoto Y, Sugiuchi T, Masuda S, Okamoto Y, Morita N, Sakamoto H, Shintani T, Fukuda S, Kaga K, Matsunaga T.

[Deterioration in Distortion Product Otoacoustic Emissions in Auditory Neuropathy Patients With Distinct Clinical and Genetic Backgrounds.](#)

Ear Hear. 2019 Jan/Feb;40(1):184-191. doi: 10.1097/AUD.0000000000000586.

OBJECTIVES: Auditory neuropathy (AN) is a clinical disorder characterized by the absence of auditory brainstem response and presence of otoacoustic emissions. A gradual loss of otoacoustic emissions has been reported for some cases of AN. Such cases could be diagnosed as cochlear hearing loss and lead to misunderstanding of the pathology when patients first visit clinics after the loss of otoacoustic emissions. The purpose of this study was to investigate the time course of changes in distortion product otoacoustic emissions (DPOAEs) in association with patients’ genetic and clinical backgrounds, including the use of hearing aids.

DESIGN: DPOAE measurements from 31 patients with AN were assessed. Genetic analyses for GJB2, OTOF, and mitochondrial m.1555A>G and m.3243A>G mutations were conducted for all cases, and the analyses for CDH23 and OPA1 were conducted for the selected cases. Patients who were younger than 10 years of age at the time of AN diagnosis were designated as the pediatric AN group (22 cases), and those who were 18 years of age or older were designated as the adult AN group (9 cases). DPOAE was measured at least twice in all patients. The response rate for DPOAEs was defined and analyzed.

RESULTS: The pediatric AN group comprised 10 patients with OTOF mutations, 1 with GJB2 mutations, 1 with OPA1 mutation, and 10 with indefinite causes. Twelve ears (27%) showed no change in DPOAE, 20 ears (46%) showed a decrease in DPOAE, and 12 ears (27%) lost DPOAE. Loss of DPOAE occurred in one ear (2%) at 0 years of age and four ears (9%) at 1 year of age. The time courses of DPOAEs in patients with OTOF mutations were divided into those with early loss and those with no change, indicating that the mechanism for deterioration of DPOAEs includes not only the OTOF mutations but also other common modifier factors. Most, but not all, AN patients who used hearing aids showed deterioration of DPOAEs after the start of using hearing aids. A few AN patients also showed deterioration of DPOAEs before using hearing aids. The adult AN group comprised 2 patients with OPA1 mutations, 2 with OTOF mutations, and 5 with indefinite causes. Four ears (22%) showed no change in DPOAE, 13 ears (72%) showed a decrease, and one ear (6%) showed a loss of DPOAE. Although the ratio of DPOAE decrease was higher in the adult AN group than in the pediatric AN group, the ratio of DPOAE loss was lower in the adult AN group. DPOAE was not lost in all four ears with OPA1 mutations and in all four ears with OTOF mutations in the adult group.

CONCLUSIONS: DPOAE was decreased or lost in approximately 70% of pediatric and about 80% of adult AN patients. Eleven percent of pediatric AN patients lost DPOAEs by 1 year of age. Genetic factors were thought to have influenced the time course of DPOAEs in the pediatric AN group. In most adult AN patients, DPOAE was rarely lost regardless of the genetic cause.

Komori K, Komori M, Eitoku M, Joelle Muchanga SM, Ninomiya H, Kobayashi T, Sukanuma N; Japan Environment and Children’s Study (JECS) Group.

[Verbal abuse during pregnancy increases frequency of newborn hearing screening referral: The Japan Environment and Children’s Study.](#)

Child Abuse Negl. 2019 Apr;90:193-201. doi: 10.1016/j.chiabu.2019.01.025. Epub 2019 Feb 23.

BACKGROUND: Verbal abuse during pregnancy has a greater impact than physical and sexual violence on the incidence of postnatal depression and maternal abuse behavior towards their children. In addition, exposure of children (aged 12 months to adolescence) to verbal abuse from their parents exerts an adverse impact to the children’s auditory function. However, the effect of verbal abuse during pregnancy on fetal auditory function has

not yet been thoroughly investigated.

OBJECTIVE: The objective of the study was to examine the relationship between intimate partner verbal abuse during pregnancy and newborn hearing screening (NHS) referral, which indicates immature or impaired auditory function.

PARTICIPANTS AND SETTING: The Japan Environment and Children's Study is an ongoing nationwide population-based birth-cohort study designed to determine environmental factors during and after pregnancy that affect the development, health, or wellbeing of children. Pregnant women living in 15 areas of Japan were recruited between January 2011 and March 2014.

METHODS: Multiple imputation for missing data was performed, followed by multiple logistic regression using 16 confounding variables.

RESULTS: Of 104,102 records in the dataset, 79,985 mother-infant pairs submitted complete data for questions related to verbal and physical abuse and the results of NHS. Of 79,985 pregnant women, 10,786 (13.5%) experienced verbal abuse and 978 (1.2%) experienced physical abuse. Of 79,985 newborns, 787 (0.98%) received a NHS referral. Verbal abuse was significantly associated with NHS referral (adjusted odds ratio: 1.44; 95% confidence interval: 1.05-1.98).

CONCLUSIONS: Verbal abuse should be avoided during pregnancy to preserve the newborn's auditory function.

Lago MRR, Fernandes LDC, Lyra IM, Ramos RT, Teixeira R, Salles C, Ladeia AMT.

[Sensorineural hearing loss in children with sickle cell anemia and its association with endothelial dysfunction.](#)

Hematology. 2018 Dec;23(10):849-855. doi: 10.1080/10245332.2018.1478494. Epub 2018 May 28.

OBJECTIVES: To investigate the prevalence of sensorineural hearing loss (SNHL) in children and adolescents with sickle cell anemia (SCA) and its association with endothelial dysfunction (ED).

METHODS: Fifty-two participants with stable SCA and 44 apparently healthy (AA genotype) participants aged 6-18 years were evaluated for pure tone audiometry and endothelial function using ultrasonographic imaging of the brachial artery to assess flow-mediated dilation (FMD). Laboratory analysis of the lipid profile and C-reactive protein levels was performed.

RESULTS: In the SCA group, 15 (28.8%) patients presented with SNHL. The FMD values were reduced in the SCA with SNHL group compared with the SCA without SNHL and healthy groups. Logistic regression analysis showed that FMD was associated with SNHL independent of the lipid profile and SCA characteristics (odds ratio [95% confidence interval] = 0.614 [0.440-0.858]; $p = 0.004$).

DISCUSSION: SNHL is a common complication in SCA; furthermore, this study identified a significant association between ED and SNHL. Damage to the vascular endothelium because of inflammation in SCA reduced blood flow in the inner ear. Thus, this circulatory disorder culminates in vaso-occlusive process and induces auditory disorders, such as SNHL.

Lee ER, Chan DK.

[Implications of dried blood spot testing for congenital CMV on management of children with hearing loss: A preliminary report.](#)

Int J Pediatr Otorhinolaryngol. 2019 Apr;119:10-14. doi: 10.1016/j.ijporl.2018.12.029. Epub 2018 Dec 21.

INTRODUCTION: Non-genetic, congenital sensorineural hearing loss (cSNHL) is commonly caused by congenital CMV infection (cCMV). Hearing loss related to cCMV is variable in degree, often progressive, and can affect one or both ears.

OBJECTIVES: We sought to examine the outcomes of DBS testing in California, and the hearing outcomes of cCMV-positive children.

METHODS: This is a retrospective study of patients with SNHL of unknown etiology aged 6 months to 17 years old presenting to a tertiary care pediatric center and evaluated for cCMV by DBS testing.

RESULTS: 14 children (228 ears) with SNHL of unknown origin were included. 6/114 (5.3%) tested positive for cCMV versus 108/114 (94.7%), who tested negative. None of the cCMV-positive children had symmetric bilateral hearing loss, compared with 56.5% (61/108) of cCMV-negative children ($p < 0.05$). cCMV-positive children were more likely to have profound SNHL in the worse-hearing ear (5/6 (83%) vs 16/108 (14.9%) of cCMV-negative children, $p < 0.001$). 86% (5/6) exhibited progressive hearing loss, including progression or new-onset hearing loss in the previously better hearing ear. 3 of the 6 children with cCMV underwent CI.

CONCLUSION: A small proportion of patients presenting with SNHL tested positive on DBS. Of cCMV-positive children, most presented with profound hearing loss in the worse-hearing ear, and 50% of cCMV-positive children developed progressive hearing loss in the initially better-hearing ear. Prognostic information afforded by etiologic confirmation of cCMV infection informed decision-making concerning cochlear implantation in these cases.

Lee H, Lee H, Noh H.

[Prediction of uptake and retention of conventional hearing aids in Korean pediatric patients with unilateral hearing loss.](#)

OBJECTIVE: The purpose of this study was to describe and predict hearing aid uptake and retention in Korean pediatric patients with unilateral hearing loss (UHL) in a secondary referral hospital.

METHODS: This was a retrospective study using clinical data collected at the time of UHL diagnosis. The study included data collected from 2009 to 2016. Serial audiograms were extracted from clinical charts, and follow-up status and rehabilitation decisions were analyzed.

RESULTS: Of 102 children and adolescents (9.5 ± 5.1 years, 64 male), 52.9% followed a check-up schedule, and 31 (30.4%) obtained a hearing aid. Hearing threshold and speech discrimination scores were predictive parameters of hearing aid uptake. Among those who used a hearing aid, 17 (56.7%) subjects used it successfully based on significant predictive parameters of channel number.

CONCLUSION: Hearing aid retention in pediatric patients seems less predictable than in adults with UHL. No good predictable parameter for hearing aid retention was identified except channel number for pediatric UHL cases. Regular monitoring of hearing and selection of a multi-channel hearing aid are crucial to minimize the potential negative effects of UHL.

Lee JM, Nozu K, Choi DE, Kang HG, Ha IS, Cheong HI.

[Features of Autosomal Recessive Alport Syndrome: A Systematic Review.](#)

J Clin Med. 2019 Feb 3;8(2). pii: E178. doi: 10.3390/jcm8020178.

ABSTRACT: Alport syndrome (AS) is one of the most frequent hereditary nephritis leading to end-stage renal disease (ESRD). Although X-linked (XLAS) inheritance is the most common form, cases with autosomal recessive inheritance with mutations in *COL4A3* or *COL4A4* are being increasingly recognized. A systematic review was conducted on autosomal recessive Alport syndrome (ARAS). Electronic databases were searched using related terms (until Oct 10th, 2018). From 1601 articles searched, there were 26 eligible studies with 148 patients. Female and male patients were equally affected. About 62% of patients had ESRD, 64% had sensorineural hearing loss (SNHL) and 17% had ocular manifestation. The median at onset was 2.5 years for hematuria (HU), 21 years for ESRD, and 13 years for SNHL. Patients without missense mutations had more severe outcomes at earlier ages, while those who had one or two missense mutations had delayed onset and lower prevalence of extrarenal manifestations. Of 49 patients with kidney biopsy available for electron microscopy (EM) pathology, 42 (86%) had typical glomerular basement membrane (GBM) changes, while 5 (10%) patients showed GBM thinning only. SNHL developed earlier than previously reported. There was a genotype phenotype correlation according to the number of missense mutations. Patients with missense mutations had delayed onset of hematuria, ESRD, and SNHL and lower prevalence of extrarenal manifestations.

Leigh J, Farrell R, Courtenay D, Dowell R, Briggs R.

[Relationship Between Objective and Behavioral Audiology for Young Children Being Assessed for Cochlear Implantation: Implications for CI Candidacy Assessment.](#)

Otol Neurotol. 2019 Mar;40(3):e252-e259. doi: 10.1097/MAO.0000000000002125.

OBJECTIVE: This study aimed to evaluate the feasibility of making cochlear implant recommendations based on diagnostic ABR and ASSR results. The goal was to challenge the need for behavioral audiometry as part of the standard cochlear implant assessment battery for infants with profound hearing loss and to reduce the age at which cochlear implant recommendation was made.

STUDY DESIGN: A retrospective review of 123 patient files for children referred to the pediatric cochlear implant service before 3 years of age over a 3-year period was undertaken. Results for click-ABR, ASSR, and behavioral audiology at 500, 1k, 2k and 4k Hz, and tympanometry were collected and relationships were investigated for 64 children who met the inclusion criteria. Data were excluded for 59 children due to the presence of auditory neuropathy findings, middle ear pathology at the time of testing, if ASSR was not assessed at intensity levels >85dB, and/or behavioral testing was judged to be unreliable by two experienced clinicians.

SETTING: Primary care pediatric cochlear implant program located within a hospital setting.

PATIENTS: Pediatric patients referred for cochlear implant evaluation before 3 years of age.

INTERVENTIONS(S): Children were assessed using ABR, ASSR, and behavioral audiometry for identification and confirmation of hearing loss.

MAIN OUTCOME MEASURES(S): Correlation between diagnostic click-ABR and ASSR thresholds and subsequently obtained behavioral hearing thresholds.

RESULTS: Results for objective measures (click-ABR and ASSR) were significantly correlated with behavioral results. The correlations, however, were poorer than expected with limited predictive value. For 6 of the 64 children click-ABR and/or ASSR suggested profound hearing loss and corresponding behavioral hearing threshold was found to be in the severe hearing loss range.

CONCLUSIONS: Findings of this study do not support making cochlear implant recommendations based on the findings of diagnostic click-ABR and ASSR alone. Investigating ways to reduce the average age children with severe-to-profound hearing loss receive a cochlear implant is a priority for the study institution. An alternate

evaluation pathway for infants which incorporates a multifaceted assessment is warranted and will be the focus of future work at the study institution.

Li Y, Shen M, Long M.

[A preliminary study of auditory mismatch response on the day of cochlear implant activation in children with hearing aids prior implantation.](#)

PLoS One. 2019 Jan 7;14(1):e0210457. doi: 10.1371/journal.pone.0210457. eCollection 2019.

OBJECTIVE: The study aimed to explore the characteristics of auditory mismatch response (MMR) in hearing-impaired children on the day when the cochlear implant (CI) was started (power-up) and the speech processor was programmed, and to investigate the effects of wearing hearing aids (HAs) before cochlear implantation on the early stage of postoperative auditory cortex plasticity, providing some demonstrative data for the objective evaluation of postoperative early auditory ability in children who underwent cochlear implantation.

METHODS: The participants were 34 children with profound sensorineural hearing loss, who underwent cochlear implantation. The classical passive Oddball paradigm was adopted, using a pair of vowels which only have different lexical tones. The standard stimulus was /a2/ and the devious stimulus was /a4/.

RESULTS: 1) On the day of CI activation, the auditory MMR has been elicited in 30 children; the MMR incidence was 88%. 2) We observed both positive and negative auditory MMR waveforms. And logistic regression analysis showed that it was influenced by the age at cochlear implantation. 3) The duration with HA before surgery significantly influenced the MMR latency. The children with longer duration of HA use have much earlier latency of MMR. 4) There was a significant positive correlation between the age at HA use initiation and MMR amplitude. Earlier initial HA use was associated with smaller amplitude.

CONCLUSIONS: MMR in response to Mandarin lexical tone can be recorded in most pediatric patients who had experience with HA on the day of CI power up. MMR is closely associated with the age at cochlear implantation, duration of HA use, and the age at HA use initiation. Hearing-impaired children should wear HA as early as possible and ensure consistent usage.

Liu Y, Ye L, Zhu P, Wu J, Tan S, Chen J, Wu C, Zhong Y, Wang Y, Li X, Liu H.

[Genetic screening involving 101 hot spots for neonates not passing newborn hearing screening and those random recruited in Dongguan.](#)

Int J Pediatr Otorhinolaryngol. 2019 Feb;117:82-87. doi: 10.1016/j.ijporl.2018.11.008. Epub 2018 Nov 22.

ABSTRACT: In order to investigate essential molecular causes for hearing loss and mutation frequency of deafness-related genes, 1315 newborns who did not pass the Newborn Hearing Screening (NHS) (audio-no-pass) and 1000 random-selected infants were subjected to detection for 101 hotspot mutations in 18 common deafness-related genes. Totally, 23 alleles of 7 deafness genes were detected out. Significant difference ($\chi^2 = 25.320$, $p = 0.000$) existed in causative mutation frequency between audio-no-pass group (81/1315, 6.160%) and random-selected cohort (18/1000, 1.80%). Of the genes detected out, GJB2 gene mutation was with significant difference ($\chi^2 = 75.132$, $p = 0.000$) between audio-no-pass group (417/1315, 31.711%) and random-selected cohort (159/1000, 15.900%); c.109G > A was the most common allele, as well as the only one with significantly different allele frequency ($\chi^2 = 79.327$, $p = 0.000$) between audio-no-pass group (392/1315, 16.84%) and random-selected cohort (140/1000, 7.55%), which suggested c.109G > A mutation was critical for newborns' hearing loss. This study performed detection for such a large scale of deafness-associated genes and for the first time compared mutations between audio-no-pass and random-recruited neonates, which not only provided more reliable DNA diagnosis result for medical practioners and enhanced clinical care for the newborns, but gave more accurate estimation for mutation frequency.

Liu Y, Hu C, Liu C, Liu D, Mei L, He C, Jiang L, Wu H, Chen H, Feng Y.

[A rapid improved multiplex ligation detection reaction method for the identification of gene mutations in hereditary hearing loss.](#)

PLoS One. 2019 Apr 11;14(4):e0215212. doi: 10.1371/journal.pone.0215212. eCollection 2019.

ABSTRACT: Hearing loss (HL) is a common sensory disorder. More than half of HL cases can be attributed to genetic causes. There is no effective therapy for genetic HL at present, early diagnosis to reduce the incidence of genetic HL is important for clinical intervention in genetic HL. Previous studies have identified 111 nonsyndromic hearing loss genes. The most frequently mutated genes identified in NSHL patients in China include GJB2, SLC26A4, and the mitochondrial gene MT-RNR1. It is important to develop HL gene panels in Chinese population, which allow for etiologic diagnosis of both SHL and NSHL. In this study, a total of 220 unrelated Han Chinese patients with bilateral progressive SNHL and 50 unrelated healthy controls were performed Single nucleotide polymorphism (SNP) genotyping using an improved multiplex ligation detection reaction (iMLDR) technique, is to simultaneously detect a total of 32 mutations in ten HL genes, covering all currently characterized mutations involved in the etiology of nonsyndromic or syndromic hearing loss in the Chinese population. The 49 positive samples with known mutations were successfully detected using the iMLDR

Technique. For 171 SNHL patients, gene variants were found in 57 cases (33.33%), among which, 30 patients carried mutations in GJB2, 14 patients carried mutations in SLC26A4, seven patients carried mutations in GJB3, and six patients carried mutations in MT-RNR1. The molecular etiology of deafness was confirmed in 12.9% (22/171) of patients carried homozygous variants. These results were verified by Sanger sequencing, indicating that the sensitivity and specificity of the iMLDR technique was 100%. We believe that the implementation of this population-specific technology at an efficient clinical level would have great value in HL diagnosis and treatment.

Lu Y1, Zhou L, Imrit TS, Liu A.

Sudden Sensorineural Hearing Loss in Children: Clinical Characteristics, Etiology, Treatment Outcomes, and Prognostic Factors.

Otol Neurotol. 2019 Apr;40(4):446-453. doi: 10.1097/MAO.0000000000002190.

OBJECTIVE: To investigate the clinical characteristics, etiology, treatment outcomes, and prognostic factors of sudden sensorineural hearing loss (SSNHL) in children to guide the clinical diagnosis and treatment of SSNHL in the pediatric population.

STUDY DESIGN: Retrospective case review.

SETTING: Tertiary referral center.

PATIENTS: Patients diagnosed with SSNHL from November 2011 to December 2017 with relatively complete clinical data.

INTERVENTION: Diagnosis and systemic treatment of SSNHL.

MAIN OUTCOME MEASURES: Patients' clinical characteristics, etiology, laboratory tests, imaging, pure-tone audiometry at admission, and discharge were analyzed.

RESULTS: A total of 25 children and 149 adults with SSNHL were included. Recent or previous viral infection rates (81.8%) and fasting blood glucose level (5.23+1.47mmol/L) in children with SSNHL were lower than those in adult SSNHL patients ($p=0.033$, $p=0.033$). Autoimmune abnormalities (90.0%) and plasma fibrinogen abnormalities (27.3%) were higher in children with SSNHL than those in adult SSNHL patients (40.0%, 8.8%, respectively, $p<0.05$). The recovery rate in children (38.4%) with SSNHL is comparable to that in adults (22.6%), but children have a higher complete rate compared to adults (26.9%, 11.3%, respectively, $p<0.05$). Children with a profound audiometric curve had a worse prognosis in comparison to other types of audiometric curves ($p=0.041$).

CONCLUSIONS: Children with SSNHL have a lower rate of viral infection in comparison to adults with SSNHL. Fasting blood glucose levels, complement C3, C4, and fibrinogen may be closely related to childhood SSNHL. The recovery rate in children with SSNHL is comparable to that in adults, but children have a higher complete rate compared to adults. A profound hearing curve is an unfavorable prognostic factor in both children and adults with SSNHL.

Macielak RJ, Mattingly JK, Findlen UM, Moberly AC, Malhotra PS, Adunka OF.

Audiometric findings in children with unilateral enlarged vestibular aqueduct.

Int J Pediatr Otorhinolaryngol. 2019 May;120:25-29. doi: 10.1016/j.ijporl.2019.01.034. Epub 2019 Jan 25.

OBJECTIVE: To evaluate the prevalence of bilateral hearing loss in children with unilateral enlarged vestibular aqueduct (EVA) at a single institution.

METHODS: A retrospective case review was performed at a tertiary care pediatric referral center involving children with radiologic findings of unilateral EVA and normal labyrinthine anatomy of the contralateral ear diagnosed via CT and/or MRI. The main outcome measure of interest is the number of patients with unilateral EVA who were diagnosed with bilateral hearing loss.

RESULTS: Sixty-one pediatric patients were identified. The mean audiometric follow-up was 48.2 months (0-150). Three (4.9%) patients with unilateral EVA were noted to have bilateral hearing loss, and this rate was not significantly different ($p = 1.0$) from the rate reported in a comparison group of patients with contralateral hearing loss (6.0%) without an EVA. The pure-tone average (defined as the average dB HL at 500, 1000, 2000, and 4000 Hz) in the group with bilateral hearing loss was 31.3 dB HL in the better hearing ear and 79.6 dB HL in the worse hearing ear, with the difference being statistically significant ($p = 0.02$). In the unilateral EVA patients without contralateral hearing loss ($n = 56$, 91.8%), the PTA was 9.4 dB HL in the better hearing ear and 51.9 dB HL in the worse hearing ear, with the difference being statistically significant ($p < 0.001$). Two patients (3.3%) with unilateral EVA were found to have hearing within normal limits bilaterally. The EVA was ipsilateral to the worse hearing ear in all cases.

CONCLUSION: The prevalence of bilateral hearing loss in children with unilateral EVA appears to be low. Specifically, it may be no different than the rate of contralateral hearing loss in children with unilateral hearing loss without an EVA. The present report is somewhat different than the previously described prevalence in the literature. This difference could be related to the imaging type and diagnostic criteria used, the patients included, the source of the identified patents, and the overall population of patients studied.

Maluleke NP, Khoza-Shangase K, Kanji A.

[Communication and school readiness abilities of children with hearing impairment in South Africa: A retrospective review of early intervention preschool records.](#)

S Afr J Commun Disord. 2019 Feb 28;66(1):e1-e7. doi: 10.4102/sajcd.v66i1.604.

BACKGROUND: The national prevalence of hearing impairment in South Africa is estimated to be four to six in every 1000 live births in the public health care sector. An undetected hearing impairment in childhood can lead to delayed speech and language development as well as put the child at risk of not achieving the necessary school readiness abilities that will enable them to achieve academic success. However, through early hearing detection and intervention services, children with hearing impairment can develop communication and school readiness abilities on par with children with normal hearing.

OBJECTIVE: The aim of the study was to describe communication and school readiness abilities of children who were identified with hearing impairment and enrolled in early intervention (EI) preschools in Gauteng.

METHODS: Within a descriptive research study design, a retrospective record review was conducted on files of eight children, ranging in age from 9 years and 7 months to 12 years and 7 months, identified with a hearing impairment and enrolled in EI preschools in Gauteng, South Africa. Descriptive statistics were used to analyse the data, using frequency distribution and measures of central tendency.

RESULTS: Current findings revealed that children with hearing impairment who were enrolled in EI preschools in Gauteng were identified late. This consequently led to delayed ages at initiation of EI services when compared to international benchmarks and the Health Professions Council of South Africa's (HPCSA) guidelines of 2018. Consequently, participants presented with below average communication and school readiness abilities, which are characteristic of hearing impairment that is identified late.

CONCLUSIONS: Transference of current contextually relevant research findings into practice by both the Department of Health and the Department of Basic Education forms part of future directions from this study. This conversion of research findings into service delivery must be conducted in a systematic manner at all levels in these two sectors to facilitate achievement of Early Hearing Detection and Intervention (EHDI), resulting in better communication and school readiness outcomes.

Manjaly JG, Nash R, Ellis W, Britz A, Lavy JA, Shaida A, Saeed SR, Khalil SS.

[Hearing Preservation With Standard Length Electrodes in Pediatric Cochlear Implantation.](#)

Otol Neurotol. 2018 Oct;39(9):1109-1114. doi: 10.1097/MAO.0000000000001917.

OBJECTIVE: Preserving low frequencies following cochlear implantation improves outcomes and allows patients to use a combination of electrical and acoustic stimulation. This importance has been reflected in advances in electrode design and refined surgical techniques. Full insertion of standard length electrodes may be advantageous over shortened electrodes because more electrodes can be activated over time if low frequency hearing loss progresses. Surgeons must counsel patients over this choice but data is lacking regarding the degree and likelihood of hearing preservation achievable with standard length electrodes in children. We report our experience using standard length cochlear implant arrays for hearing preservation in children.

METHODS: Retrospective case series.

INCLUSION CRITERIA: preoperative hearing ≤ 85 dB HL at 250Hz and aged ≤ 18 years. Hearing preservation percentages are calculated using the HEARRING group formula. (Equation is included in full-text article.) Preservation of $> 75\%$ was considered complete, 25 to 75% partial, and 1 to 25% minimal. Patients were implanted with either MED-EL FLEX28 or Cochlear Nucleus CI522. Standardized operative technique with facial recess approach, posterior tympanotomy and minimally traumatic round window insertion.

RESULTS: Fifty-two implantations in 27 pediatric patients met inclusion criteria. Mean age at implantation: 9.8 years. Average latest audiogram: 8 months. Mean total pre- and postoperative pure-tone averages were 82.8 and 92.6dB. Seventeen (33%) ears demonstrated complete hearing preservation, 22 (42%) ears partial hearing preservation, 7 (13%) minimal hearing preservation, and 6 (12%) exhibited no acoustic hearing postoperatively. Mean hearing preservation was 55.5%.

CONCLUSION: Hearing preservation is achievable to varying degrees in pediatric cochlear implantation using standard length electrodes though it is difficult to predict preoperatively which children may benefit. This study is among the largest additions to the knowledge base for this patient group.

Mauldin L.

[Don't look at it as a miracle cure: Contested notions of success and failure in family narratives of pediatric cochlear implantation.](#)

Soc Sci Med. 2019 May;228:117-125. doi: 10.1016/j.socscimed.2019.03.021. Epub 2019 Mar 16.

ABSTRACT: Cochlear implants (CIs) are a routine treatment for children identified with a qualifying hearing loss. The CI, however, must be accompanied by a long-term and intense auditory training regimen in order to possibly acquire spoken language with the device. This research investigates families' experiences when they opted for the CI and undertook the task of auditory training, but the child failed to achieve what might be clinically

considered “success” - the ability to function solely using spoken language. Using a science and technology studies informed approach that places the CI within a complex sociotechnical system, this research shows the uncertain trajectory of the CI, as well as the contingency of the very notions of success and failure. To do so, data from in-depth interviews with a diverse sample of parents (n = 11) were collected. Results show the shifting definitions of failure and success within families, as well as suggest areas for further exploration regarding clinical practice and pediatric CIs. First, professionals’ messaging often conveyed to parents a belief in the infallibility of the CI, this potentially caused “soft failure” to go undetected and unmitigated. Second, speech assessments used in clinical measurements of outcomes did not capture a holistic understanding of a child’s identity and social integration, leaving out an important component for consideration of what a ‘good outcome’ is. Third, minority parents experience structural racism and clinical attitudes that may render “failure” more likely to be identified and expected in these children, an individualizing process that allows structural failures to go uncriticized.

McCrary H, Sheng X, Greene T, Park A.

[Long-term hearing outcomes of children with symptomatic congenital CMV treated with valganciclovir.](#)

Int J Pediatr Otorhinolaryngol. 2019 Mar;118:124-127. doi: 10.1016/j.ijporl.2018.12.027. Epub 2018 Dec 21.

OBJECTIVES: Congenital human cytomegalovirus (cCMV) is a leading cause of pediatric hearing loss. Recent literature has suggested that valganciclovir (VGCV) therapy can improve hearing outcomes. The objective of this study was to evaluate the long-term hearing outcomes among symptomatic CMV patients treated with VGCV.

METHODS: A retrospective chart review of symptomatic CMV patients treated with VGCV was completed. The primary endpoint was the change in best ear hearing scores prior to treatment and after follow-up audiograms. A paired-sample t-test was used to evaluate the data.

RESULTS: A total of 16 children were included in the study and participants were followed for an average of 3.2 years. There was a measurable worsening, but not a statistically significant change in the best ear hearing scores, where the mean change was 11.9 dB (p-value = 0.070). However, 14/16 patients (87.5%, p-value<0.001) were found to have clinically significant worsening of hearing. The mean change in hearing scores for the left and right ear was 14.2 dB (p-value = 0.023) and 15.5 dB (p-value = 0.032), respectively. Mean elapsed time for progressive loss was 2.6 ± 0.2 years. When comparing the better or worse ear, there was no pattern for which ear deteriorated earlier or more frequently.

CONCLUSIONS: Our data did show a measurable, but not a statistically significant worsening outcome in best ear hearing. There was a significant change in both left and right ear hearing. Our results suggest that VGCV may provide only a short-term improvement in hearing outcomes; however, these preliminary post-hoc findings suggest the need for a more rigorous evaluation.

McDaniel J, Camarata S, Yoder P.

[Comparing Auditory-Only and Audiovisual Word Learning for Children With Hearing Loss.](#)

J Deaf Stud Deaf Educ. 2018 Oct 1;23(4):382-398. doi: 10.1093/deafed/eny016.

ABSTRACT: Although reducing visual input to emphasize auditory cues is a common practice in pediatric auditory (re)habilitation, the extant literature offers minimal empirical evidence for whether unisensory auditory-only (AO) or multisensory audiovisual (AV) input is more beneficial to children with hearing loss for developing spoken language skills. Using an adapted alternating treatments single case research design, we evaluated the effectiveness and efficiency of a receptive word learning intervention with and without access to visual speechreading cues. Four preschool children with prelingual hearing loss participated. Based on probes without visual cues, three participants demonstrated strong evidence for learning in the AO and AV conditions relative to a control (no-teaching) condition. No participants demonstrated a differential rate of learning between AO and AV conditions. Neither an inhibitory effect predicted by a unisensory theory nor a beneficial effect predicted by a multisensory theory for providing visual cues was identified. Clinical implications are discussed.

McKearney RM, MacKinnon RC.

[Objective auditory brainstem response classification using machine learning.](#)

Int J Audiol. 2019 Apr;58(4):224-230. doi: 10.1080/14992027.2018.1551633. Epub 2019 Jan 21.

OBJECTIVE: The objective of this study was to use machine learning in the form of a deep neural network to objectively classify paired auditory brainstem response waveforms into either: ‘clear response’, ‘inconclusive’ or ‘response absent’.

DESIGN: A deep convolutional neural network was constructed and fine-tuned using stratified 10-fold cross-validation on 190 paired ABR waveforms. The final model was evaluated on a test set of 42 paired waveforms.

STUDY SAMPLE: The full dataset comprised 232 paired ABR waveforms recorded from eight normal-hearing individuals. The dataset was obtained from the PhysioBank database. The paired waveforms were independently labelled by two audiological scientists in order to train the network and evaluate its performance.

RESULTS: The trained neural network was able to classify paired ABR waveforms with 92.9% accuracy. The

sensitivity and the specificity were 92.9% and 96.4%, respectively.

CONCLUSIONS: This neural network may have clinical utility in assisting clinicians with waveform classification for the purpose of hearing threshold estimation. Further evaluation using a large clinically obtained dataset would provide further validation with regard to the clinical potential of the neural network in diagnostic adult testing, newborn testing and in automated newborn hearing screening.

Mishra S, Pandey H, Srivastava P, Mandal K, Phadke SR.

Connexin 26 (GJB2) Mutations Associated with Non-Syndromic Hearing Loss (NSHL).

Indian J Pediatr. 2018 Dec;85(12):1061-1066. doi: 10.1007/s12098-018-2654-8. Epub 2018 Mar 15.

OBJECTIVE: To determine the prevalence and spectrum of Connexin 26 (GJB2) mutations in pre-lingual non-syndromic hearing loss (NSHL) patients in authors' centre and to review the data of Indian patients from the literature.

METHODS: Sanger sequencing of entire coding region contained in single exon (Exon 2) of GJB2 gene in 15 patients of NSHL.

RESULTS: GJB2 mutations were found in 40% (6/15) of NSHL patients, out of which mono-allelic were 33.3% (2/6). Bi-allelic GJB2 mutations were identified in 4 of 6 patients. Most common GJB2 mutation identified was c.71G>A(p.W24X), comprising 30% of the total GJB2 mutant alleles. Six studies involving 1119 patients with NSHL were reviewed and 4 of them have reported c.71G>A(p.W24X) as the commonest mutation while 2 studies found c.35delG as the commonest. GJB2 mutations accounted for 10.9%-36% cases of NSHL. Sixteen other mutations in GJB2 gene were reported in Indian patients out of which 6 mutations other than c.71G>A(p.W24X) viz., c.35delG, c.1A>G(p.M1V), c.127G>A(p.V43 M), c.204C>G(p.Y86X), c.231G>A(p.W77X) and c.439G>A(p.E147K) were identified in the present study.

CONCLUSIONS: Connexin 26 (GJB2) mutations are responsible for 19.4% of NSHL in Indian population. The c.71G>A(W24X) and c.35delG were the most prevalent GJB2 mutations accounting for 72.2% (234 of 324 total mutated alleles from 7 studies) and 15.4% (50 of 324 total mutated alleles from 7 studies) respectively. Thus, screening of these two common mutations in GJB2 gene by polymerase chain reaction and restriction fragment length polymorphism (PCR-RFLP) would greatly help in providing easy genetic diagnosis and help in genetic counseling of the families with NSHL.

Muñoz K, Price T, Nelson L, Twohig M.

Counseling in Pediatric Audiology: Audiologists' Perceptions, Confidence, and Training.

J Am Acad Audiol. 2019 Jan;30(1):66-77. doi: 10.3766/jaaa.17087. Epub 2017 Dec 21.

BACKGROUND: Pediatric audiologists are an important source of support for parents when a child is identified with hearing loss. As parents learn how to manage their child's hearing loss they often need help navigating challenges that arise; however, audiologists may experience a variety of barriers implementing effective counseling strategies. Many internal and external barriers experienced by parents can be appropriately supported and navigated within audiology services.

PURPOSE: To investigate audiologists' perceptions, training, and confidence related to counseling and to explore the influence of years practicing audiology and taking a counseling course on perceptions and confidence.

RESEARCH DESIGN: A cross-sectional, population-based survey.

STUDY SAMPLE: Three hundred and fifty surveys were analyzed from pediatric audiologists across the U.S. Responses were received from 26 states and one U.S. territory.

DATA COLLECTION AND ANALYSIS: Data were collected through the mail and online. Descriptive and comparative statistics were used to analyze the information. Content analysis was performed to identify emergent themes from the responses to open-ended questions.

RESULTS: Pediatric audiologists reported their perceptions about importance of counseling skills, challenges they encounter, their confidence in counseling, and how often they use the skills when needed in practice. Most audiologists ($\geq 75\%$) felt it was very or extremely important to talk with parents about nine of the ten items (e.g., their [parents'] expectations). Three-fourth of the audiologists reported experiencing a moderate challenge or greater in knowing how to assess the presence of psychosocial challenges and in having enough time to address emotional needs. Many of the audiologists felt very or extremely confident in guiding parents in the development of an action plan (62%) and determining if parents have external barriers (60%). Approximately one-third or less of the participants reported performing any of the skills (e.g., determining if parent has external or internal barriers) $\geq 75\%$ of the time, and a statistically significant difference was found with participants practicing ≤ 10 yr using the skills more frequently than participants practicing for ≥ 11 yr. In addition, there was a statistically significant difference between participants who had taken an audiology-specific counseling course and those who had not; those who had reported being more confident and using counseling skills more often than audiologists did not have a counseling course.

CONCLUSIONS: This study found strong support for audiologist perceived importance of counseling; however, **110**

fewer audiologists reported confidence in their counseling skills and in using counseling skills. Counseling training was variable; audiologists would benefit from a more systematic approach to counseling instruction within graduate training.

Myers J, Kei J, Aithal S, Aithal V, Driscoll C, Khan A, Manuel A, Joseph A, Malicka AN.

[Development of a Diagnostic Prediction Model for Conductive Conditions in Neonates Using Wideband Acoustic Immittance.](#)

Ear Hear. 2018 Nov/Dec;39(6):1116-1135. doi: 10.1097/AUD.0000000000000565.

OBJECTIVES: Wideband acoustic immittance (WAI) is an emerging test of middle-ear function with potential applications for neonates in screening and diagnostic settings. Previous large-scale diagnostic accuracy studies have assessed the performance of WAI against evoked otoacoustic emissions, but further research is needed using a more stringent reference standard. Research into suitable quantitative techniques to analyze the large volume of data produced by WAI is still in its infancy. Prediction models are an attractive method for analysis of multivariate data because they provide individualized probabilities that a subject has the condition. A clinically useful prediction model must accurately discriminate between normal and abnormal cases and be well calibrated (i.e., give accurate predictions). The present study aimed to develop a diagnostic prediction model for detecting conductive conditions in neonates using WAI. A stringent reference standard was created by combining results of high-frequency tympanometry and distortion product otoacoustic emissions.

DESIGN: High-frequency tympanometry and distortion product otoacoustic emissions were performed on both ears of 629 healthy neonates to assess outer- and middle-ear function. Wideband absorbance and complex admittance (magnitude and phase) were measured at frequencies ranging from 226 to 8000 Hz in each neonate at ambient pressure using a click stimulus. Results from one ear of each neonate were used to develop the prediction model. WAI results were used as logistic regression predictors to model the probability that an ear had outer/middle-ear dysfunction. WAI variables were modeled both linearly and nonlinearly, to test whether allowing nonlinearity improved model fit and thus calibration. The best-fitting model was validated using the opposite ears and with bootstrap resampling.

RESULTS: The best-fitting model used absorbance at 1000 and 2000 Hz, admittance magnitude at 1000 and 2000 Hz, and admittance phase at 1000 and 4000 Hz modeled as nonlinear variables. The model accurately discriminated between normal and abnormal ears, with an area under the receiver-operating characteristic curve (AUC) of 0.88. It effectively generalized to the opposite ears (AUC = 0.90) and with bootstrap resampling (AUC = 0.85). The model was well calibrated, with predicted probabilities aligning closely to observed results.

CONCLUSIONS: The developed prediction model accurately discriminated between normal and dysfunctional ears and was well calibrated. The model has potential applications in screening or diagnostic contexts. In a screening context, probabilities could be used to set a referral threshold that is intuitive, easy to apply, and sensitive to the costs associated with true- and false-positive referrals. In a clinical setting, using predicted probabilities in conjunction with graphical displays of WAI could be used for individualized diagnoses. Future research investigating the use of the model in diagnostic or screening settings is warranted.

Nada DW, El Khouly RM, Gadow SE, Hablas SA, Aboelhawa MA, Al Ashkar DS, El Barbary AM, Hussein MS, Rageh E, Elsalawy AM, Abo-Zaid MH, Elshweikh S, El Gharib AM.

[The role of auditory evoked potentials and otoacoustic emissions in early detection of hearing abnormalities in Behçet's disease patients. A case control study.](#)

Clin Exp Rheumatol. 2018 Nov-Dec;36(6 Suppl 115):45-52. Epub 2018 May 10.

OBJECTIVES: To determine the types and to assess the role of auditory evoked potentials and otoacoustic emissions in early detection of hearing abnormalities in Behçet's disease (BD) patients. Their correlations with disease activity were also considered.

METHODS: Thirty patients with BD and thirty apparently sex- and age-matched healthy volunteers were included in this study. Auditory evaluation included pure tone audiometry (PTA), otoacoustic emissions (TEOAEs, DPOAE), auditory brainstem response test (ABR) and cortical auditory evoked potentials (tone and speech CAEPs) for all patients and control.

RESULTS: The highest abnormality of CAEP latencies elicited by (500Hz and 1000 Hz) as well as speech stimuli (da and ga) among our BD patients was delayed P1 and N1 waves at 80 dB with greater bilateral affection, as well as significant differences between patients and controls. All our BD patients had a smaller amplitude of distortion product OAE (DPOAE) and S/N ratio at 1, 2, 4, 6 kHz compared with controls and the differences were highly statistically significant ($p=0.0001$).

CONCLUSIONS: Being one of the autoimmune inner ear diseases (AIED), BD has a definite hearing impairment, even in the presence of normal hearing sensitivity, as evidenced by PTA. BD patients had a sub-clinical cochlear pathology which was not affected by disease activity or different organ affection. DPOAE (S/N ratio) proved to be a sensitive test in detecting minimal changes in cochlear pathology and the latencies of CAEPs

(tone and speech) measures were considered as sensitive indicators (100%) of early detection of hearing impairment in BD patients.

Nam GS, Kwak SH, Bae SH, Kim SH, Jung J, Choi JY.

[Hyperbilirubinemia and Follow-up Auditory Brainstem Responses in Preterm Infants.](#)

Clin Exp Otorhinolaryngol. 2019 May;12(2):163-168. doi: 10.21053/ceo.2018.00899. Epub 2018 Nov 9.

OBJECTIVES: Neonatal hyperbilirubinemia is considered one of the most common causative factors of hearing loss. Preterm infants are more vulnerable to neuronal damage caused by hyperbilirubinemia. This study aimed to evaluate the effect of hyperbilirubinemia on hearing threshold and auditory pathway in preterm infants by serial auditory brainstem response (ABR). In addition, we evaluate the usefulness of the unconjugated bilirubin (UCB) level compared with total serum bilirubin (TSB) on bilirubin-induced hearing loss.

METHODS: This study was conducted on 70 preterm infants with hyperbilirubinemia who failed universal newborn hearing screening by automated ABR. The diagnostic ABR was performed within 3 months after birth. Follow-up ABR was conducted in patients with abnormal results (30 cases). TSB and UCB concentration were compared according to hearing threshold by ABR.

RESULTS: The initial and maximal measured UCB concentration for the preterm infants of diagnostic ABR ≥ 40 dB nHL group (n=30) were statistically higher compared with ABR ≤ 35 dB nHL group (n=40) (P=0.031 and P=0.003, respectively). In follow-up ABR examination, 13 of the ABR ≥ 40 dB nHL group showed complete recovery, but 17 had no change or worsened. There was no difference in bilirubin level between the recovery group and non-recovery group.

CONCLUSION: UCB is a better predictor of bilirubin-induced hearing loss than TSB in preterm infants as evaluated by serial ABR. Serial ABR testing can be a useful, noninvasive methods to evaluate early reversible bilirubin-induced hearing loss in preterm infants.

Neumann K, Thomas JP, Voelter C, Dazert S.

[A new adhesive bone conduction hearing system effectively treats conductive hearing loss in children.](#)

Int J Pediatr Otorhinolaryngol. 2019 Apr 3;122:117-125. doi: 10.1016/j.ijporl.2019.03.014. [Epub ahead of print]

OBJECTIVES: Bone conduction hearing devices integrated in softbands (BCDSs) are frequently not well accepted by children with conductive hearing loss due to pressure on the head, sweating, or cosmetic stigma. A non-surgical hearing system (ADHEAR) uses a new bone conduction concept consisting of an audio processor connected to an adhesive adapter fixed behind the ear. This study is the first to evaluate the audiological and clinical outcome of this novel system, comparing it with conventional BCDSs in a short- and mid-term follow-up in children under 10 years of age.

METHODS: The ADHEAR was compared to a BCDS in 10 children with conductive hearing loss (age: 0.7-9.7 years). Aided and unaided pure tone/behavioral observational audiometry and, if applicable, speech audiometry in quiet and noise were performed initially with both devices and after 8 weeks with the ADHEAR alone. The subjective hearing gain and usage of the new hearing system, as well as patients' and parents' satisfaction were assessed using questionnaires.

RESULTS: The functional gain with the ADHEAR averaged over 0.5, 1, 2, and 4 kHz exceeded that of the conventional BCDS (35.6 dB \pm 15.1 vs. 29.9 dB \pm 14.6, p = .001, n = 9 ears). Speech perception in quiet and noise (n = 8) improved in the aided situation similarly for both hearing devices. The parents of 8 of 10 children evaluated the ADHEAR system as being useful. Minor wearing problems occurred occasionally. Eight children continued using the ADHEAR after the study, one received an active middle ear implant and one continued to use a BCDS.

CONCLUSION: The ADHEAR system is a promising solution for children with conductive hearing loss or chronically draining ears.

Ngui LX, Tang IP, Prepageran N, Lai ZW.

[Comparison of distortion product otoacoustic emission \(DPOAE\) and automated auditory brainstem response \(AABR\) for neonatal hearing screening in a hospital with high delivery rate.](#)

Int J Pediatr Otorhinolaryngol. 2019 May;120:184-188. doi: 10.1016/j.ijporl.2019.02.045. Epub 2019 Feb 27.

INTRODUCTION: Congenital hearing loss is one of the commonest congenital anomalies. Neonatal hearing screening aims to detect congenital hearing loss early and provide prompt intervention for better speech and language development. The two recommended methods for neonatal hearing screening are otoacoustic emission (OAE) and automated auditory brainstem response (AABR).

OBJECTIVE: To study the effectiveness of distortion product otoacoustic emission (DPOAE) and automated auditory brainstem response (AABR) as first screening tool among non-risk newborns in a hospital with high delivery rate.

METHOD: A total of 722 non-risk newborns (1444 ears) were screened with both DPOAE and AABR prior to discharge within one month. Babies who failed AABR were rescreened with AABR \pm diagnostic auditory

brainstem response tests within one month of age.

RESULTS: The pass rate for AABR (67.9%) was higher than DPOAE (50.1%). Both DPOAE and AABR pass rates improved significantly with increasing age (p -value <0.001). The highest pass rate for both DPOAE and AABR were between the age of 36-48 h, 73.1% and 84.2% respectively. The mean testing time for AABR (13.54 min \pm 7.47) was significantly longer than DPOAE (3.52 min \pm 1.87), with a p -value of <0.001 .

CONCLUSIONS: OAE test is faster and easier than AABR, but with higher false positive rate. The most ideal hearing screening protocol should be tailored according to different centre.

Nunes ADDS, Silva CRL, Balen SA, Souza DLB, Barbosa IR.

Prevalence of hearing impairment and associated factors in school-aged children and adolescents: a systematic review.

Braz J Otorhinolaryngol. 2019 Mar - Apr;85(2):244-253. doi: 10.1016/j.bjorl.2018.10.009. Epub 2018 Dec 1.

INTRODUCTION: Hearing impairment is one of the communication disorders of the 21st century, constituting a public health issue as it affects communication, academic success, and life quality of students. Most cases of hearing loss before 15 years of age are avoidable, and early detection can help prevent academic delays and minimize other consequences.

OBJECTIVE: This study researched scientific literature for the prevalence of hearing impairment in school-aged children and adolescents, with its associated factors. This was accomplished by asking the defining question: "What is the prevalence of hearing impairment and its associated factors in school-aged children and adolescents?"

METHODS: Research included the databases PubMed/MEDLINE, LILACS, Web of Science, Scopus and SciELO, and was carried out by two researchers, independently. The selected papers were analyzed on the basis of the checklist provided by the report Strengthening the Reporting of Observational Studies in Epidemiology.

RESULTS: From the 463 papers analyzed, 26 fulfilled the criteria and were included in the review presented herein. The detection methods, as well as prevalence and associated factors, varied across studies. The prevalence reported by the studies varied between 0.88% and 46.70%. Otologic and non-otologic factors were associated with hearing impairment, such as middle ear and air passage infections, neo- and post-natal icterus, accumulation of cerumen, family history, suspicion of parents, use of earphones, age and income.

CONCLUSION: There is heterogeneity regarding methodology, normality criteria, and prevalence and risk factors of studies about hearing loss in adolescents and school-aged children. Nevertheless, the relevance of the subject and the necessity of early interventions are unanimous across studies.

Núñez-Batalla F, Jáudenes-Casaubón C, Sequí-Canet JM, Vivanco-Allende A, Zubicaray-Ugarteche J.

Early diagnosis and treatment of unilateral or asymmetrical hearing loss in children: CODEPEH recommendations.

Acta Otorrinolaringol Esp. 2018 Dec 19. pii: S0001-6519(18)30178-X. doi: 10.1016/j.otorri.2018.09.004. [Epub ahead of print]

ABSTRACT: The aim of this document is to improve the management and the treatment of unilateral or asymmetrical hearing loss in children. One in one thousand newborn infants has unilateral hearing loss and this prevalence increases with age, due to cases of acquired and delayed-onset hearing loss. Although the impact on the development and learning processes of children of these kinds of hearing loss have usually been minimized, if they are not treated they will impact on language and speech development, as well as overall development, affecting the quality of life of the child and his/her family. The outcomes of the review are expressed as recommendations aimed at clinical diagnosis and therapeutic improvement for unilateral or asymmetrical hearing loss.

Olarte M, Bermúdez Rey MC, Beltran AP, Guerrero D, Suárez-Obando F, López G, García M, Ospina JC, Fonseca C, Bertolotto AM, Aldana N, Gelvez N, Tamayo ML.

Detection of hearing loss in newborns: Definition of a screening strategy in Bogotá, Colombia.

Int J Pediatr Otorhinolaryngol. 2019 Mar 26;122:76-81. doi: 10.1016/j.ijporl.2019.03.016. [Epub ahead of print]

OBJECTIVE: To describe the results from the hearing screening protocol adopted in a Hospital in Colombia emphasizing the importance of performing screening on an outpatient basis, when the newborn is more than 24 h old.

METHODS: A prospective study at Hospital Universitario San Ignacio in Bogota, Colombia was carried out, from May 1st, 2016 to Nov 30th, 2017, the study sample included 2.088 newborns examined using transient otoacoustic emissions.

RESULTS: We obtained written consent from the parents of 1.523 newborns and 24 individuals (1.6%) failed the first stage of the screening, nine cases unilateral and 15 bilateral. A total of nine neonates (0,6%) failed the second screening test, six cases unilateral and three bilateral. Four (0,3%) did not return to the second test. Our false altered screening rate was 0.7%.

CONCLUSIONS: In a developing country with limited human and economic resources, in which newborn early discharge is the norm, a newborn hearing screening program linked to infants' check-ups, that uses otoacoustic emissions after 48 h of life, seems a feasible option compare to the standard US protocol aiming to conduct hearing screening prior to discharge.

Palma S, Roversi MF, Bettini M, Mazzoni S, Pietrosemoli P, Lucaccioni L, Berardi A, Genovese E.
[Hearing loss in children with congenital cytomegalovirus infection: an 11-year retrospective study based on laboratory database of a tertiary paediatric hospital.](#)

Acta Otorhinolaryngol Ital. 2019 Feb;39(1):40-45. doi: 10.14639/0392-100X-2020.

ABSTRACT: Congenital cytomegalovirus infection is considered the main cause of infantile non-genetic neurosensory hearing loss. Although this correlation was described more than 50 years ago, the natural history of internal ear involvement has not yet been fully defined. Hearing loss is the most frequent sequela and is seen in a variable percentage up to 30%; the hearing threshold is characterised by fluctuations or progressive deterioration. The purpose of this study was to evaluate the prevalence of hearing loss in cases of congenital CMV infection from Modena county, starting from the database of the microbiology and virology reference laboratory. All children undergoing urine testing for suspected CMV infection or viral DNA testing on Guthrie Card in the period between January 2004 and December 2014 were enrolled in the study. Family paediatricians were contacted and asked about clinical information on the possible presence at birth or subsequent occurrence of hearing loss, excluding cases where this was not possible. The results showed an annual prevalence of congenital cytomegalovirus infection among suspected cases that was stable over time despite the progressive increase in subjects tested. The prevalence of hearing loss was in line with the literature, whereas in long-term follow-up cases of moderate, medium-to-severe hearing loss with late onset were not detected. The introduction of newborn hearing screening in the county has allowed early diagnosis of hearing loss at birth as non-TEOAE-born births underwent a urine virus test. Moreover, despite all the limitations of the study, we can conclude that European epidemiological studies are needed to better define the relationship between congenital CMV infection and internal ear disease as the impact of environmental and genetic factors is still not entirely clarified.

Pasternak Y, Attias J, Ely N, Amir J, Bilavsky E.
[No risk factors for late onset hearing loss in asymptomatic congenital cytomegalovirus infants - close monitoring is needed.](#)

Acta Paediatr. 2019 Apr 12. doi: 10.1111/apa.14814. [Epub ahead of print]

ABSTRACT: Congenital cytomegalovirus (cCMV) is the leading cause of congenital infections, affecting approximately 0.7% of live births worldwide. Although, 85%-90% of infected children are asymptomatic at birth, 10%-15% will develop late onset hearing impairment (1). The appropriate management of cCMV is controversial and data are needed to estimate the cost-effectiveness of universal versus targeted newborn screening. Many risk factors for late onset hearing deterioration have been proposed, but not confirmed.

Pourreza MR, Mohammadi H, Sadeghian L, Asgharzadeh S, Sehhati M, Tabatabaiefar MA.
[Applying Two Different Bioinformatic Approaches to Discover Novel Genes Associated with Hereditary Hearing Loss via Whole-Exome Sequencing: ENDEAVOUR and HomozygosityMapper.](#)

Adv Biomed Res. 2018 Oct 31;7:141. doi: 10.4103/abr.abr_80_18. eCollection 2018.

BACKGROUND: Hearing loss (HL) is a highly prevalent heterogeneous deficiency of sensory-neural system with involvement of several dozen genes. Whole-exome sequencing (WES) is capable of discovering known and novel genes involved with HL.

MATERIALS AND METHODS: Two pedigrees with HL background from Khuzestan province of Iran were selected. Polymerase chain reaction-sequencing of *GJB2* and homozygosity mapping of 16 DFNB loci were performed. One patient of the first and two affected individuals from the second pedigree were subjected to WES. The result files were analyzed using tools on Ubuntu 16.04. Short reads were mapped to reference genome (hg19, NCBI Build 37). Sorting and duplication removals were done. Variants were obtained and annotated by an online software tool. Variant filtration was performed. In the first family, ENDEAVOUR was applied to prioritize candidate genes. In the second family, a combination of shared variants, homozygosity mapping, and gene expression were implemented to launch the disease-causing gene.

RESULTS: *GJB2* sequencing and linkage analysis established no homozygosity-by-descent at any DFNB loci. Utilizing ENDEAVOUR, *BBX*: C.C857G (P.A286G), and *MYH15*: C.C5557T (P.R1853C) were put forward, but none of the variants co-segregated with the phenotype. Two genes, *UNC13B* and *TRAK1*, were prioritized in the homozygous regions detected by HomozygosityMapper.

CONCLUSION: WES is regarded a powerful approach to discover molecular etiology of Mendelian inherited disorders, but as it fails to enrich GC-rich regions, incapability of capturing noncoding regulatory regions and limited specificity and accuracy of copy number variations detection tools from exome data, it is assumed an

insufficient procedure.

Puia-Dumitrescu M, Bretzius OM, Brown N, Fitz-Henley JA, Ssengonzi R, Wechsler CS, Gray KD, Benjamin DK Sr, Smith PB, Clark RH, Gonzalez D, Hornik CP.

[Evaluation of Gentamicin Exposure in the Neonatal Intensive Care Unit and Hearing Function at Discharge.](#)

J Pediatr. 2018 Dec;203:131-136. doi: 10.1016/j.jpeds.2018.07.101. Epub 2018 Sep 21.

OBJECTIVE: To characterize the association between gentamicin dosing, duration of treatment, and ototoxicity in hospitalized infants.

STUDY DESIGN: This retrospective cohort study conducted at 330 neonatal intensive care units (2002-2014) included inborn infants exposed to gentamicin with available hearing screen results, and excluded infants with incomplete dosing data and major congenital anomalies. Our primary outcome was the final hearing screen result performed during hospitalization: abnormal (failed or referred for further testing in one or both ears) or normal (bilateral passed). The 4 measures of gentamicin exposure were highest daily dose, average daily dose, cumulative dose, and cumulative duration of exposure. We fitted separate multivariable logistic regression models adjusted for demographics, comorbidities, and other clinical events.

RESULTS: A total of 84 808 infants met inclusion/exclusion criteria; median (25th, 75th percentile) gestational age and birth weight were 35 weeks (33, 38) and 2480 g (1890, 3184), respectively. Failed hearing screens occurred in 3238 (3.8%) infants; failed screens were more likely in infants of lower gestational age and birth weight, who had longer hospital lengths of stay, higher rates of morbidities, and were small for gestational age. Median highest daily dose, average daily dose, and cumulative dose were 4.0 mg/kg/day (3.0, 4.0), 3.8 mg/kg/day (3.0, 4.0), and 12.1 mg/kg (9.1, 20.5), respectively. Median cumulative duration of exposure was 3 days (3, 6). In adjusted analysis, gentamicin dose and duration of therapy were not associated with hearing screen failure.

CONCLUSIONS: Gentamicin dosing and duration of treatment were not associated with increased odds of failed hearing screen at the time of discharge from initial neonatal intensive care unit stay.

Ramkumar V, Nagarajan R, Shankarnarayan VC, Kumaravelu S, Hall JW.

[Implementation and evaluation of a rural community-based pediatric hearing screening program integrating in-person and tele-diagnostic auditory brainstem response \(ABR\).](#)

BMC Health Serv Res. 2019 Jan 3;19(1):1. doi: 10.1186/s12913-018-3827-x.

BACKGROUND: In an attempt to reach remote rural areas, this study explores a community-based, pediatric hearing screening program in villages, integrating two models of diagnostic ABR testing; one using a tele-medicine approach and the other a traditional in-person testing at a tertiary care hospital.

METHODS: Village health workers (VHWs) underwent a five day training program on conducting Distortion Product Oto Acoustic Emissions (DPOAE) screening and assisting in tele-ABR. VHWs conducted DPOAE screening in 91 villages and hamlets in two administrative units (blocks) of a district in South India. A two-step DPOAE screening was carried out by VHWs in the homes of infants and children under five years of age in the selected villages. Those with 'refer' results in 2nd screening were recommended for a follow-up diagnostic ABR testing in person (Group A) at the tertiary care hospital or via tele-medicine (Group B). The overall outcome of the community-based hearing screening program was analyzed with respect to coverage, refer rate, follow-up rate for 2nd screenings and diagnostic testing. A comparison of the outcomes of tele-versus in-person diagnostic ABR follow-up was carried out.

RESULTS: Six VHWs who fulfilled the post training evaluation criteria were recruited for the screening program. VHWs screened 1335 children in Group A and 1480 children in Group B. The refer rate for 2nd screening was very low (0.8%); the follow-up rate for 2nd screening was between 80 and 97% across the different age groups. Integration of tele-ABR resulted in 11% improvement in follow-up compared to in-person ABR at a tertiary care hospital.

CONCLUSIONS: Non-availability of audiologists and limited infrastructure in rural areas has prevented the establishment of large scale hearing screening programs. In existing programs, considerable challenges with respect to follow-up for diagnostic testing was reported, due to patients being submitted to traveling long distance to access services and potential wage losses during that time. In this program model, integration of a tele-ABR diagnostic follow-up improved follow-up in comparison to in-person follow-up. VHWs were successfully trained to conduct accurate screenings in rural communities. The very low refer rate, and improved follow-up rate reflect the success of this community-based hearing screening program.

Rashid SMU, Mukherjee D, Ahmmed AU.

[Auditory processing and neuropsychological profiles of children with functional hearing loss.](#)

Int J Pediatr Otorhinolaryngol. 2018 Nov;114:51-60. doi: 10.1016/j.ijporl.2018.07.054. Epub 2018 Jul 31.

OBJECTIVES: This paper compares structured history, auditory processing abilities and neuropsychological findings of children with functional hearing loss (FHL) to those with suspected auditory processing disorder without FHL (control). The main aim was to evaluate the value of a holistic assessment protocol for FHL used in

a routine pediatric audiology clinic. The protocol incorporated a commercially available test battery for auditory processing disorder (APD), non-verbal intelligence (NVIQ) and tools to screen for common co-existing neurodevelopmental conditions such as attention deficit hyperactivity disorder (ADHD), language impairment (LI) and developmental coordination disorder (DCD). The outcome of such holistic assessment was expected to help in understanding the nature of FHL and to provide individualized support to mitigate their difficulties.

METHODS: This retrospective study compared two groups, 40 children (M = 17, F = 23) in each group between seven and sixteen years of age, one group with a history of FHL and the other with suspected APD without FHL (control). The groups were matched against age, gender, hand use, diagnosis of APD or non-APD (31 with APD and 9 without APD in each group) and non-verbal intelligence. All the children were healthy English speaking children attending mainstream schools with no middle or inner ear abnormalities. Structured history was obtained from parents regarding different nonacademic and academic concerns. The SCAN-3:C and SCAN-3:A test batteries were used to assess auditory processing abilities; Lucid Ability test for NVIQ; Children's Communication Checklist-2 (CCC-2) for language ability; Swanson Nolan and Pelham-IV Rating Scale (SNAP-IV) for ADHD; and the manual dexterity components of the Movement Assessment Battery for Children-2 (MABC-2) as a screening tool for DCD.

RESULTS: About 60% of children in both the groups had concerns regarding listening in noisy background. In the history, poor attention was reported in 45% of children in the FHL group compared to 82.5% in the control group ($p < 0.01$). Hyperacusis was present in 35% of children in the FHL group and in 62% of children in the control group ($p < 0.05$). Concerns about overall academic abilities were present in 59% of children in the FHL group and 75% of the controls ($p > 0.05$). Only 15% of children in the FHL group had concerns with numeracy skills in contrast to 41% of the controls ($p < 0.05$). Significantly fewer ($p < 0.01$) children in the FHL group (41%) received additional support at school than the controls (75%). Fewer children performed poorly in Filtered Words (FW) test of the SCAN-3 batteries, 30% in the FHL group and 17.5% in the control group, in contrast to Auditory Figure Ground 0 (AFG0), 85% in FHL and 80% in the control group. The number of children performing poorly in AFG0 was significantly higher compared to all the other SCAN-3 tests in FHL ($P < 0.05$), in contrast to FW and Competing Sentences (CS) only in the control group ($p < 0.05$). The control group had higher prevalence of atypical ear advantage (AEA) in left directed Competing Words (CW) (32.5%) and Time Compressed Sentences (TCS) (32.5%) compared to FW (7.5%). In contrast, FHL group had higher prevalence of AEA in AFG0 (48.7%) compared to CS (21%). High proportions of children in both the groups had LI (80% in FHL and 82.5% in the control group), with significantly lower ($p < 0.05$) levels of ADHD symptoms in the FHL group (39.5%) compared to the control group (72.5%). Impaired manual dexterity was present in 30.7% of children in FHL group and 47.5% in the controls.

CONCLUSIONS: The prevalences of APD and language impairment are high compared to ADHD symptoms in children with FHL, and holistic assessment is recommended. Despite some similarities in the auditory and neuropsychological profiles between children with FHL and those with suspected APD without FHL some differences were noted. The results suggest that children with FHL have genuine difficulties that need to be identified and addressed. Future research is required to identify the neural pathways which could explain the similarities and dissimilarities between the two groups.

Reis FMFDS, Gonçalves CGO, Conto J, Iantas M, Lüders D, Marques J.

[Hearing Assessment of Neonates at Risk for Hearing Loss at a Hearing Health High Complexity Service: An Electrophysiological Assessment.](#)

Int Arch Otorhinolaryngol. 2019 Apr;23(2):157-164. doi: 10.1055/s-0038-1648217. Epub 2018 May 9.

INTRODUCTION: Hearing is the main sensory access in the first years of life. Therefore, early detection and intervention of hearing impairment must begin before the first year of age.

OBJECTIVE: To analyze the results of the electrophysiological hearing assessment of children at risk for hearing loss as part of the newborn hearing screening (NHS).

METHODS: This is a cross-sectional study held at a hearing health public service clinic located in Brazil, with 104 babies at risks factors for hearing loss referred by public hospitals. A questionnaire was applied to parents, and the auditory brainstem response (ABR) test was held, identifying those with alterations in the results. The outcome of the NHS was also analyzed regarding risk factor, gestational age and gender.

RESULTS: Among the 104 subjects, most of them were male (53.85%), and the main risk factor found was the admission to the neonatal intensive care unit (NICU) for a period longer than 5 days (50.93%). Eighty-five (81.73%) subjects were screened by NHS at the maternity and 40% of them failed the test. Through the ABR test, 6 (5.77%) infants evidenced sensorineural hearing loss, 4 of them being diagnosed at 4 months, and 2 at 6 months of age; all of them failed the NHS and had family history and admission at NICU for over 5 days as the most prevalent hearing risks; in addition, family members of all children perceived their hearing impairment.

CONCLUSION: Advances could be observed regarding the age of the diagnosis after the implementation of the NHS held at the analyzed public service clinic.

Riga M, Korres G, Chouridis P, Naxakis S, Danielides V.

[Congenital cytomegalovirus infection inducing non-congenital sensorineural hearing loss during childhood; A systematic review.](#)

Int J Pediatr Otorhinolaryngol. 2018 Dec;115:156-164. doi: 10.1016/j.ijporl.2018.10.005. Epub 2018 Oct 4.

BACKGROUND: Congenital cytomegalovirus (CMV) infection is one of the most important risk factors for delayed onset and progressive hearing loss in children. However, the relevant literature is limited, heterogeneous and currently insufficient to provide guidance toward the effective monitoring of hearing acuity in these children.

OBJECTIVES: The aim of this study was to provide a systematic review focused on types of hearing loss that may escape diagnosis through universal neonatal hearing screening and/or present significant changes during childhood, such as progressive, fluctuating and late-onset hearing loss.

DATA SOURCES: A review of the present literature was conducted via the PubMed database of the US National Library of Medicine (www.pubmed.org) and Scopus database (www.scopus.com) with the search terms “late-onset hearing loss cytomegalovirus”, “progressive hearing loss cytomegalovirus” and “fluctuating hearing loss cytomegalovirus”.

STUDY ELIGIBILITY CRITERIA: Prospective or retrospective clinical studies were included if they presented a detailed audiological assessment, for a follow-up period of >2years.

METHODS: The prevalence and time of diagnosis of progressive, fluctuating and late-onset hearing loss were considered as primary outcomes. Results were recorded separately for symptomatic and asymptomatic children, when possible.

RESULTS: This analysis refers to a population of 181 children with CMV-induced hearing loss, who were diagnosed among 1089 with congenital CMV infection. The prevalence of CMV-induced hearing loss was significantly higher among symptomatic children ($p < 0.0001$), who were also significantly more likely to develop bilateral hearing loss ($p = 0.001$). There was not sufficient information on the prevalence, laterality, degree and time of diagnosis of progressive, fluctuating and late-onset hearing loss that could constitute the basis toward the report of specific follow-up guidelines.

CONCLUSIONS: Further studies are needed in order to understand and quantify the potential effects of congenital CMV infection in the inner ear and hearing acuity. The results presented in the relative studies should be very carefully evaluated and compared to each other, since they correspond to substantially different cohorts, study designs, and result elaboration. Infants with congenital CMV infection should be closely monitored, regarding their hearing acuity at least during their preschool years, although substantial changes in hearing thresholds have been reported as late as the 16th year of age. Parental counseling is of outmost importance in order to minimize the numbers of children lost to follow-up.

Rissatto-Lago MR, da Cruz Fernandes L, Lyra IM, Terse-Ramos R, Teixeira R, Salles C, Teixeira Ladeia AM.

[Hidden hearing loss in children and adolescents with sickle cell anemia.](#)

Int J Pediatr Otorhinolaryngol. 2019 Jan;116:186-191. doi: 10.1016/j.ijporl.2018.10.042. Epub 2018 Nov 2.

OBJECTIVE: To evaluate the auditory system for hidden hearing loss (HHL) and its association with clinical variables and endothelial dysfunction (ED) in children and adolescents with sickle cell anemia (SCA).

METHODS: Participants included 37 patients with stable SCA and 44 healthy controls (HC group) (aged 6-18 years) with hearing thresholds ≤ 20 dB (dB) were evaluated for pure tone audiometry, tympanometry, acoustic reflex, otoacoustic emission, and auditory evoked potentials. Laboratory analysis of the lipid profile, and C-reactive protein levels and endothelial function using ultrasonographic imaging of the brachial artery to assess flow-mediated dilation were performed.

RESULTS: The SCA group presented with a higher rate of increased contralateral acoustic reflex thresholds, compared to those in the HC group at all frequencies and in both ears ($p < 0.05$). There were significant differences in the brainstem auditory evoked potentials between the SCA and HC groups. In the SCA group, the waves III and V latencies were increased ($p = 0.006$ and 0.004 respectively), and the I-III and I-V interpeak intervals were longer ($p = 0.015$ and 0.018 respectively) than those in the HC group. There was no association between the audiological measures and clinical and metabolic variables and sickle cell anemia complications including endothelial function and therapy.

CONCLUSION: In conclusion, our findings suggest that damage in the auditory system in SCA patients can be present involving retrocochlear structures, causing functional deficits without deterioration of auditory sensitivity.

Robertson MS, Hayashi SS, Camet ML, Trinkaus K, Henry J, Hayashi RJ.

[Asymmetric sensorineural hearing loss is a risk factor for late-onset hearing loss in pediatric cancer survivors following cisplatin treatment.](#)

Pediatr Blood Cancer. 2019 Jan;66(1):e27494. doi: 10.1002/pbc.27494. Epub 2018 Oct 18.

BACKGROUND: Ototoxicity is a significant complication of cisplatin treatment. Hearing loss can be symmetric or asymmetric, and may decline after therapy. This study examined the risks of asymmetric and late-onset hearing loss (LOHL) in cisplatin-treated pediatric patients with cancer.

METHODS: A retrospective review of 993 patients' medical and audiological charts from August 1990 to March 2015 was conducted using stringent criteria to characterize patients with asymmetric hearing loss (AHL) or LOHL. Audiologic data were reviewed for 248 patients that received cisplatin to assess cisplatin-induced sensorineural hearing loss and its associated risk factors.

RESULTS: Of the patients evaluable for AHL, 26% exhibited this finding. Of those evaluable for LOHL, 42% of the patients' hearing worsened more than 6 months after therapy completion. Radiation and type of cancer diagnosis were major risk factors for both AHL and LOHL. Furthermore, LOHL was linked to age of diagnosis, noncranial radiation, and longer audiologic follow-up. AHL was strongly associated with LOHL—60% of patients with AHL also had LOHL. Logistic regression analysis revealed that patients with AHL (OR 6.3, 95% CI: 2.2-17.8, $P = 0.0005$) or those receiving radiation (OR 3.2, 95% CI: 1.2-8.6, $P = 0.02$) were at greatest risk for LOHL.

CONCLUSION: Children receiving cisplatin therapy are at risk for developing AHL and LOHL. Those that have received radiation and/or with AHL are at increased risk for further hearing decline. Long-term monitoring of these patients is important for early intervention as hearing diminishes.

Ropers FG, Pham ENB, Kant SG, Rotteveel LJC, Rings EHHM, Verbist BM, Dekkers OM.

[Assessment of the Clinical Benefit of Imaging in Children With Unilateral Sensorineural Hearing Loss: A Systematic Review and Meta-analysis.](#)

JAMA Otolaryngol Head Neck Surg. 2019 Apr 4. doi: 10.1001/jamaoto.2019.0121. [Epub ahead of print]

IMPORTANCE: Imaging used to determine the cause of unilateral sensorineural hearing loss (USNHL) in children is often justified by the high likelihood of detecting abnormalities, which implies that these abnormalities are associated with hearing loss and that imaging has a positive contribution to patient outcome or well-being by providing information on the prognosis, hereditary factors, or cause of hearing loss.

OBJECTIVES: To evaluate the diagnostic yield of computed tomography (CT) and magnetic resonance imaging (MRI) in children with isolated unexplained USNHL and investigate the clinical relevance of these findings.

EVIDENCE REVIEW: Cochrane Library, Embase, PubMed, and Web of Science databases were searched for articles published from 1978 to 2017 on studies of children with USNHL who underwent CT and/or MRI of the temporal bone. Two authors (F.G.R. and E.N.B.P.) independently extracted information on population characteristics, imaging modality, and the prevalence of abnormalities and assessed the studies for risk of bias. Eligibility criteria included studies with 20 or more patients with USNHL who had CT and/or MRI scans, a population younger than 18 years, and those published in English.

MAIN OUTCOMES AND MEASURES: The pooled prevalence with 95% CI of inner ear abnormalities grouped according to finding and imaging modality.

FINDINGS: Of 1562 studies, 18 were included with a total of 1504 participants included in the analysis. Fifteen studies were consecutive case studies and 3 were retrospective cohort studies. The pooled diagnostic yield for pathophysiologic relevant findings in patients with unexplained USNHL was 37% for CT (95% CI, 25%-48%) and 35% for MRI (95% CI, 22%-49%). Cochleovestibular abnormalities were found with a pooled frequency of 19% for CT (95% CI, 14%-25%) and 16% for MRI (95% CI, 7%-25%). Cochlear nerve deficiency and associated cochlear aperture stenosis had a pooled frequency of 16% for MRI (95% CI, 3%-29%) and 44% for CT (95% CI, 36%-53%), respectively. Enlarged vestibular aqueduct (EVA) was detected with a pooled frequency of 7% for CT and 12% for MRI in children with USNHL.

CONCLUSIONS AND RELEVANCE: Imaging provided insight into the cause of hearing loss in a pooled frequency of about 35% to 37% in children with isolated unexplained USNHL. However, none of these findings had therapeutic consequences, and imaging provided information on prognosis and hereditary factors only in a small proportion of children, namely those with EVA. Thus, there is currently no convincing evidence supporting a strong recommendation for imaging in children who present with USNHL. The advantages of imaging should be carefully balanced against the drawbacks during shared decision making.

Schaefer K, Coninx F, Fischbach T.

[LittLEARS auditory questionnaire as an infant hearing screening in Germany after the newborn hearing screening.](#)

Int J Audiol. 2019 Apr 23;1-8. doi: 10.1080/14992027.2019.1597287. [Epub ahead of print]

OBJECTIVE: To investigate the feasibility of using the LittLEARS® Auditory Questionnaire (LEAQ®) as part of the infant hearing screening programme in Germany.

DESIGN: LEAQ®s were distributed to 47 paediatric practices and were completed by the parents/guardians of the infants (aged between 9-14 months) involved in the study (= LEAQ® screening). The infants who failed the LEAQ® screening were invited to a LEAQ rescreening. Infants who failed the LEAQ® rescreening were sent to a paediatric ENT specialist. After 3 years, a follow-up was performed on two groups: the first group comprised infants who failed the LEAQ screening; the second group (control group) comprised 200 infants who passed the LEAQ screening.

STUDY SAMPLE: 5316 questionnaires were returned.

RESULTS: Six infants with permanent hearing loss were identified using the LEAQ® as a screening tool.

CONCLUSIONS: An infant hearing screening using the LEAQ® is easily implementable in paediatric practices and may be a good alternative in countries where no objective screening instruments are available. The LEAQ® was suitable for monitoring hearing development in infants in general and could help to identify a late-onset or progressive hearing loss in infants.

Schopper HK, D'Esposito CF, Muus JS, Kanter J, Meyer TA.

[Childhood Hearing Loss in Patients With Sickle Cell Disease in the United States.](#)

J Pediatr Hematol Oncol. 2019 Mar;41(2):124-128. doi: 10.1097/MPH.0000000000001373.

ABSTRACT: This study sought to examine if modern medical evaluations including newborn screening and early diagnosis along with better methods of disease control have improved rates of hearing loss in children with sickle cell disease (SCD). Audiometric and medical data for patients with SCD was obtained from the AudGen Database and analyzed for the presence of hearing loss, type of hearing loss, severity of hearing loss, and correlation with comorbid conditions. Children with sickle cell trait (SCT) were used as a comparison group. A total of 189 patients with SCD and 244 patients with SCT had sufficient audiologic data available. Hearing loss was present in 62% of children with SCD and 50% of children with SCT in the study population. Patients with SCD were significantly more likely than those with SCT to have a sensorineural component to their hearing loss ($P < 0.001$, odds ratio: 2.41 [1.53 to 3.79]) and to have severe or profound hearing loss ($P = 0.02$, odds ratio: 4.00 [1.14 to 14.04]). The true prevalence of hearing loss in children with SCD has not been established as routine screening is not being performed. Routine auditory testing should be done for these children to detect this loss before it impacts development.

Shah J, Pham GN, Zhang J, Pakanati K, Raol N, Ongkasuwan J, Hopkins B, Anne S.

[Evaluating diagnostic yield of computed tomography \(CT\) and magnetic resonance imaging \(MRI\) in pediatric unilateral sensorineural hearing loss.](#)

Int J Pediatr Otorhinolaryngol. 2018 Dec;115:41-44. doi: 10.1016/j.ijporl.2018.09.003. Epub 2018 Sep 11.

INTRODUCTION: Options for imaging for evaluation of pediatric patients with unilateral sensorineural hearing loss (USNHL) include computed tomography (CT) and magnetic resonance imaging (MRI). Although both CT and MR imaging provide valuable information in the evaluation of pediatric patients with USNHL, debate remains regarding which imaging modality is most ideal and should be the preferred study for these children. The objective of this study is to evaluate and compare the diagnostic yield of CT versus MRI in children with USNHL.

METHODS: A multi-institutional retrospective chart review was conducted. Pediatric patients with hearing loss (diagnosis codes 389.00-389.22) seen between 2010 and 2012 at three tertiary care centers were identified. Only patients with USNHL and imaging studies were reviewed and results of CT and MRI for each patient were examined and compared. Cochleovestibular or central nervous system findings known to directly correlate to SNHL were noted as positive findings on imaging. McNemar's test was used to compare patients with positive CT and MRI results.

RESULTS: A total of 219 patients between the ages of 0-18 years with USNHL who underwent CT and/or MRI were identified. Imaging abnormalities were found in 41/96 patients who underwent MRI with overall diagnostic yield of 42.7% and 69 of 188 patients who underwent CT with overall diagnostic yield of 36.7%. For patients who underwent both imaging modalities ($n = 65$), there was no statistically significant difference in positive findings detected by CT vs MRI ($p > 0.05$).

CONCLUSIONS: Both CT and MR imaging have similar overall diagnostic yield when used to evaluate children with USNHL. Parents and patients should be counseled regarding cost, test duration, radiation exposure, need for sedation, and diagnostic accuracy associated with each imaging modality and these factors should be considered to select the appropriate diagnostic study.

Sharma R, Gu Y, Ching TYC, Marnane V, Parkinson B.

[Economic Evaluations of Childhood Hearing Loss Screening Programmes: A Systematic Review and Critique.](#)

Appl Health Econ Health Policy. 2019 Jan 25. doi: 10.1007/s40258-018-00456-1. [Epub ahead of print]

BACKGROUND: Permanent childhood hearing loss is one of the most common birth conditions associated with speech and language delay. A hearing screening can result in early detection and intervention for hearing loss.

OBJECTIVES: To update and expand previous systematic reviews of economic evaluations of childhood hearing screening strategies, and explore the methodological differences.

DATA SOURCES: MEDLINE, Embase, the Cochrane database, National Health Services Economic Evaluation Database (NHS EED), the Health Technology Assessment (HTA) database, and Canadian Agency for Drugs and Technologies in Health's (CADTH) Grey matters.

STUDY ELIGIBILITY CRITERIA, PARTICIPANTS AND INTERVENTIONS: Economic evaluations reporting costs and outcomes for both the intervention and comparator arms related to childhood hearing screening strategies.

RESULTS: Thirty evaluations (from 29 articles) were included for review. Several methodological issues were identified, including: few evaluations reported outcomes in terms of quality-adjusted life years (QALYs); none

estimated utilities directly from surveying children; none included disutilities and costs associated with adverse events; few included costs and outcomes that differed by severity; few included long-term estimates; none considered acquired hearing loss; some did not present incremental results; and few conducted comprehensive univariate or probabilistic sensitivity analysis. Evaluations published post-2011 were more likely to report QALYs and disability-adjusted life years (DALYs) as outcome measures, include long-term treatment and productivity costs, and present incremental results.

LIMITATIONS: We were unable to access the economic models and, although we employed an extensive search strategy, potentially not all relevant economic evaluations were identified.

CONCLUSIONS AND IMPLICATIONS: Most economic evaluations concluded that childhood hearing screening is value for money. However, there were significant methodological limitations with the evaluations.

Shekdar KV, Bilaniuk LT.

[Imaging of Pediatric Hearing Loss.](#)

Neuroimaging Clin N Am. 2019 Feb;29(1):103-115. doi: 10.1016/j.nic.2018.09.011. Epub 2018 Oct 31.

ABSTRACT: Temporal bone high-resolution computed tomography (HRCT) and magnetic resonance (MR) imaging are valuable tools in the evaluation of pediatric hearing loss. Computed tomography is important in the evaluation of pediatric conductive hearing loss and is the imaging modality of choice for evaluation of osseous abnormalities. MR imaging is the modality of choice for evaluation of sensorineural hearing loss. A broad spectrum of imaging findings can be seen with hearing loss in children. HRCT and MR imaging provide complementary information and are often used in conjunction in the preoperative evaluation of pediatric candidates for cochlear implantation.

Sheppard S, Biswas S, Li MH, Jayaraman V, Slack I, Romasko EJ, Sasson A, Brunton J, Rajagopalan R, Sarmady M, Abrudan JL, Jairam S, DeChene ET, Ying X, Choi J, Wilkens A, Raible SE, Scarano MI, Santani A, Pennington JW, Luo M, Conlin LK, Devkota B, Dulik MC, Spinner NB, Krantz ID.

[Utility and limitations of exome sequencing as a genetic diagnostic tool for children with hearing loss.](#)

Genet Med. 2018 Dec;20(12):1663-1676. doi: 10.1038/s41436-018-0004-x. Epub 2018 Jun 15.

PURPOSE: Hearing loss (HL) is the most common sensory disorder in children. Prompt molecular diagnosis may guide screening and management, especially in syndromic cases when HL is the single presenting feature. Exome sequencing (ES) is an appealing diagnostic tool for HL as the genetic causes are highly heterogeneous.

METHODS: ES was performed on a prospective cohort of 43 probands with HL. Sequence data were analyzed for primary and secondary findings. Capture and coverage analysis was performed for genes and variants associated with HL.

RESULTS: The diagnostic rate using ES was 37.2%, compared with 15.8% for the clinical HL panel. Secondary findings were discovered in three patients. For 247 genes associated with HL, 94.7% of the exons were targeted for capture and 81.7% of these exons were covered at 20x or greater. Further analysis of 454 randomly selected HL-associated variants showed that 89% were targeted for capture and 75% were covered at a read depth of at least 20x.

CONCLUSION: ES has an improved yield compared with clinical testing and may capture diagnoses not initially considered due to subtle clinical phenotypes. Technical challenges were identified, including inadequate capture and coverage of HL genes. Additional considerations of ES include secondary findings, cost, and turnaround time.

Siu JM, Blaser SI, Gordon KA, Papsin BC, Cushing SL.

[Efficacy of a selective imaging paradigm prior to pediatric cochlear implantation.](#)

Laryngoscope. 2019 Jan 6. doi: 10.1002/lary.27666. [Epub ahead of print]

OBJECTIVES/HYPOTHESIS: There is no consensus on the necessary preoperative imaging in children being evaluated for cochlear implantation (CI). Dual-imaging protocols that implement both magnetic resonance imaging (MRI) and high resolution computed tomography (HRCT) create diagnostic redundancy in the face of potentially unnecessary radiation and anaesthetic exposure. The objectives of the current study were to examine the efficacy of an MRI-predominant with selective HRCT imaging protocol.

STUDY DESIGN: Retrospective review.

METHODS: The protocol was implemented over a 4-year period, during which HRCT was obtained in addition to MRI only if specific risk factors on clinical assessment were identified or if imaging findings in need of further evaluation were detected on initial MRI evaluation. Retrospective review of operative reports and prospective review of imaging were performed; anesthetic exposure and costing information were also obtained.

RESULTS: Of the 240 patients who underwent assessment, seven (2.9%) had combined HRCT and MRI performed concurrently based on initial clinical assessment, 15 (6.3%) underwent HRCT based on imaging anomalies found on MRI, and MRI alone was ordered for the remaining 218 (90.1%). All patients were implanted without complication. Overall, radiation exposure, general anesthesia (GA), and healthcare costs were reduced.

CONCLUSIONS: MRI alone can be used in the vast majority of cases for preoperative evaluation of pediatric CI candidates resulting in a significant reduction in healthcare costs, radiation, and GA exposure in children. The additional need for HRCT occurs in a small proportion and can be predicted up front on clinical assessment or on initial MRI.

Skou AS, Olsen SØ, Nielsen LH, Glosli H, Jahnukainen K, Jarfelt M, Jónmundsson GK, Malmros J, Nysom K, Hasle H; Nordic Society of Pediatric Hematology and Oncology (NOPHO).

[Hearing Status in Survivors of Childhood Acute Myeloid Leukemia Treated With Chemotherapy Only: A NO-PHO-AML Study.](#)

J Pediatr Hematol Oncol. 2019 Jan;41(1):e12-e17. doi: 10.1097/MPH.0000000000001302.

BACKGROUND: As more children survive acute myeloid leukemia (AML) it is increasingly important to assess possible late effects of the intensive treatment. Hearing loss has only sporadically been reported in survivors of childhood AML. We assessed hearing status in survivors of childhood AML treated with chemotherapy alone according to 3 consecutive NOPHO-AML trials.

PROCEDURE: A population-based cohort of children treated according to the NOPHO-AML-84, NO-PHO-AML-88, and NOPHO-AML-93 trials included 137 eligible survivors among whom 101 (74%) completed a questionnaire and 99 (72%) had otologic and audiologic examination performed including otoscopy (72%), pure tone audiometry (70%), and tympanometry (60%). Eighty-four of 93 (90%) eligible sibling controls completed a similar questionnaire.

RESULTS: At a median of 11 years (range, 4 to 25) after diagnosis, hearing disorders were rare in survivors of childhood AML and in sibling controls, with no significant differences. None had severe or profound hearing loss diagnosed at audiometry. Audiometry detected a subclinical hearing loss ranging from slight to moderate in 19% of the survivors, 5% had low-frequency hearing loss, and 17% had high-frequency hearing loss.

CONCLUSIONS: The frequency of hearing disorders was low, and hearing thresholds in survivors of childhood AML were similar to background populations of comparable age.

Sokolov M, Gordon KA, Polonenko M, Blaser SI, Papsin BC, Cushing SL.

[Vestibular and balance function is often impaired in children with profound unilateral sensorineural hearing loss.](#)

Hear Res. 2019 Feb;372:52-61. doi: 10.1016/j.heares.2018.03.032. Epub 2018 Apr 3.

RATIONALE: Children with unilateral deafness could have concurrent vestibular dysfunction which would be associated with balance deficits and potentially impair overall development. The prevalence of vestibular and balance deficits remains to be defined in these children.

METHODS: Twenty children with unilateral deafness underwent comprehensive vestibular and balance evaluation.

RESULTS: Retrospective review revealed that more than half of the cohort demonstrated some abnormality of the vestibular end organs (otoliths and horizontal canal), with the prevalence of end organ specific dysfunction ranging from 17 to 48% depending on organ tested and method used. In most children, impairment occurred only on the deaf side. Children with unilateral deafness also displayed significantly poorer balance function than their normal hearing peers.

CONCLUSIONS: The prevalence of vestibular dysfunction in children with unilateral deafness is high and similar to that of children with bilateral deafness. Vestibular and balance evaluation should be routine and the functional impact of combined vestibulo-cochlear sensory deficits considered.

Soylomez E, Ertugrul S, Dogan E.

[Assessment of balance skills and falling risk in children with congenital bilateral profound sensorineural hearing loss.](#)

Int J Pediatr Otorhinolaryngol. 2019 Jan;116:75-78. doi: 10.1016/j.ijporl.2018.10.034. Epub 2018 Oct 23.

OBJECTIVE: To evaluate the balance skills and falling risk in children with a congenital bilateral profound sensorineural hearing loss (CBPSNHL).

METHODS: 25 children with CBPSNHL and healthy 25 children with similar age and gender were included in the study. The flamingo balance test, the tandem stance test, and the one-leg standing test were performed to assess the patients' static balance skills. The pediatric balance scale (PBS) was used to evaluate the dynamic balance. Visual analog scale (VAS) was applied to the patients assess the frequency of falls.

RESULTS: The flamingo balance test, the tandem stance test, and the one-leg standing test in the children with CBPSNHL were all significantly worse than the control group. Although the scores of PBS in patients with CBPSNHL were significantly lower than the control group ($p < 0.001$), the results of both groups were consistent with a low risk of falls. There was no significant difference between the VAS scores indicating the frequency of falls among the groups ($p = 0.552$).

CONCLUSION: Static and dynamic balance skills of the children with CBPSNHL are significantly impaired compared to their healthy peers. Children with CBPSNHL also have a lower risk of falling just like their healthy

peers and there is no significant difference between their falling frequencies. Balance skills of children with CBPSNHL can be assessed quickly and effectively on a hard floor (eyes closed), with a tandem standing test or a one-leg standing test.

Sözen T, Bajin MD1, Kara A, Sennaroğlu L.

[The Effect of National Pneumococcal Vaccination Program on Incidence of Postmeningitis Sensorineural Hearing Loss and Current Treatment Modalities.](#)

J Int Adv Otol. 2018 Dec;14(3):443-446. doi: 10.5152/iao.2018.6169.

OBJECTIVES: The aim of the present study was to investigate the effect of the national pneumococcal vaccination program on postmeningitis sensorineural hearing loss (SNHL).

MATERIALS AND METHODS: Overall, 2751 patients (2615 cochlear implantation and 136 auditory brainstem implantation) who underwent cochlear implantation (CI) and auditory brainstem implantation (ABI) at a tertiary referral hospital otolaryngology clinic were retrospectively analyzed. One hundred sixteen patients with a history of meningitis were included in the study. Patients were evaluated for their age at the time of surgery, gender, computerized tomography (CT) and magnetic resonance imaging (MRI) findings, implant type, side, and incidence before and after the vaccination program.

RESULTS: When patients with cochlear implants or ABI were examined, the incidence of meningitis-induced hearing loss was 6.2% in the pre-vaccination period and 0.6% in the post-vaccination period. There is a significant difference between them when compared by chi-square test ($p < 0.001$).

CONCLUSION: The most important finding of the present study is the dramatic decrease in the number of CI and ABI surgeries performed in patients with SNHL due to meningitis. This shows the effectivity of pneumococcal vaccination in this special group of patients. If total ossification is detected on CT of patients with postmeningitis, ABI should be preferred to CI.

Steuerwald W, Windmill I, Scott M, Evans T, Kramer K.

[Stories From the Webcams: Cincinnati Children's Hospital Medical Center Audiology Telehealth and Pediatric Auditory Device Services.](#)

Am J Audiol. 2018 Nov 19;27(3S):391-402. doi: 10.1044/2018_AJA-IMIA3-18-0010.

PURPOSE: The purpose of this manuscript is to describe the regulatory, technological, and training considerations for audiologists investigating telehealth and to offer some examples of audiology services provided through telehealth.

METHOD: The authors presented the regulatory components, the technology required for audiology staff and patients, and staff training for the audiology telehealth program at Cincinnati Children's Hospital Medical Center. Four case studies highlighting the successful use of telehealth in providing auditory device services to patients were also presented.

RESULTS AND CONCLUSION: The described regulatory, technological, and training hierarchy provides a framework for audiologists interested in starting a telehealth program. The cases presented illustrate that telehealth can be used to provide some auditory device services, such as troubleshooting, mapping, and parent consulting.

Stewart JE, Bentley JE.

[Hearing Loss in Pediatrics: What the Medical Home Needs to Know.](#)

Pediatr Clin North Am. 2019 Apr;66(2):425-436. doi: 10.1016/j.pcl.2018.12.010.

ABSTRACT: Screening infants for hearing loss at birth is a standard in most states in the United States, but follow-up continues to warrant improvement. Understanding the definition of hearing loss, its etiology, appropriate intervention options, and knowledge of methods to optimize an infant's outcomes through the medical home can help to maximize speech and language skills.

Tang K, Gao Z, Han C, Zhao S, Du X, Wang W.

[Screening of mitochondrial tRNA mutations in 300 infants with hearing loss.](#)

Mitochondrial DNA A DNA Mapp Seq Anal. 2019 Mar;30(2):345-350. doi: 10.1080/24701394.2018.1527910. Epub 2018 Nov 19.

ABSTRACT: Mitochondrial DNA (MtDNA) mutations are the important causes for hearing loss. To see the contribution of mtDNA to deafness, we screened for mutations in mt-tRNA genes from 300 deaf infants and 200 healthy subjects. Moreover, we analyzed the mtDNA copy number and ROS levels in patients carrying the mt-tRNA mutations. Consequently, 3 mt-tRNA mutations: tRNA^{Leu(UUR)} A3243G; tRNA^{Ala} T5655C and tRNA^{Glu} A14692G were identified, however, these mutations were not detected in controls. Of these, the A3243G mutation created a novel base-pairing (13G-23A) in the D-stem of tRNA^{Leu(UUR)}; while the T5655C mutation occurred at the very conserved acceptor arm of tRNA^{Ala}; in addition, the A14692G mutation was located at position 55 in the TΨC loop of tRNA^{Glu}. Molecular analysis showed that patients harbouring the A3243G, T5655C

and A14692G mutations had a lower level of mtDNA copy number, while ROS level increased significantly when compared with controls. Through the application of the pathogenicity scoring system, we noticed that the A3243G, T5655C and A14692G should be regarded as 'definitely pathogenic' mutations associated with deafness. Thus, our study provided novel insight into the pathophysiology, early detection of mitochondrial deafness.

Towerman AS, Hayashi SS, Hayashi RJ, Hulbert ML.

[Prevalence and nature of hearing loss in a cohort of children with sickle cell disease.](#)

Pediatr Blood Cancer. 2019 Jan;66(1):e27457. doi: 10.1002/pbc.27457. Epub 2018 Sep 11.

BACKGROUND: Sickle cell disease (SCD) may cause injury to any organ, including the auditory system.

Although the association of SCD and hearing loss has been described, the nature of this complication is unknown. We sought to establish the prevalence and nature of hearing loss in a referred cohort of children with SCD and to identify correlating disease- or treatment-associated factors.

PROCEDURE: We conducted a retrospective review of patients with SCD < 22 years of age who had hearing evaluations between August 1990 and December 2014. Demographics, audiograms, and disease and treatment variables were analyzed.

RESULTS: Two hundred and ten audiograms among 81 patients were reviewed, and 189 were evaluable. Seventy-two children constituted the referred cohort. Fourteen (19.4%) had hearing loss documented on at least one audiogram. Seven (9.7%) patients had only conductive hearing loss, and the loss persisted for up to 10.3 years. The median age of first identification was eight years. Six (8.3%) patients had hearing loss that was at least partially sensorineural. One patient's hearing loss was ambiguous. All sensorineural hearing losses were unilateral and 4/6 patients had prior documented normal hearing, indicating acquired loss. No correlations were identified.

CONCLUSIONS: Both conductive and sensorineural hearing losses are more prevalent in our study population than those observed in the general pediatric population. In children with SCD, sensorineural hearing loss appears to be acquired and unilateral. Conductive hearing loss was identified in older children and can persist. Serial screening is needed for early detection and more prompt intervention in this population.

Tsai YT, Fang KH, Yang YH, Lin MH, Chen PC, Tsai MS, Hsu CM.

[Risk of developing sudden sensorineural hearing loss in patients with hepatitis B virus infection: A population-based study.](#)

Ear Nose Throat J. 2018 Oct-Nov;97(10-11):E19-E27.

ABSTRACT: Sudden sensorineural hearing loss (SSNHL) has significant impact on quality of life. It may result from viral infection, but the relationship between hepatitis B virus (HBV) infection and SSNHL remains uncertain. To investigate the risk of developing SSNHL in patients with HBV, we conducted a nationwide, population-based, retrospective cohort study from the Taiwan National Health Insurance Research Database. A total of 33,234 patients diagnosed with HBV infection and 132,936 control subjects without viral hepatitis were selected from claims made from 2000 to 2008. Each patient was followed for at least 5 years to identify new-onset SSNHL. Among the 166,170 patients, 279 patients (303,793 person-years) from the HBV cohort and 845 patients (1,225,622 person-years) from the control cohort were diagnosed with SSNHL. The incidence of SSNHL was 1.33-fold higher in the HBV group than in the control group (0.92 vs. 0.69 per 10,000 person-years), with an adjusted hazard ratio (HR) of 1.315 (95% confidence interval [CI] = 1.148 to 1.506) calculated using a Cox proportional hazard regression model. We also observed that HBV patients in the 50 to 64 years of age subgroup showed the highest incidence of SSNHL and the highest adjusted hazard ratio (HR = 2.367; 95% CI = 1.958 to 2.861). Patients with HBV infection had a higher risk of acquiring SSNHL than patients without viral hepatitis. For the early detection and timely treatment of SSNHL, clinicians should be aware of the increased risk of SSNHL in HBV patients and arrange auditory examinations for those complaining about acute hearing change.

PMID:

van Hövell Tot Westerfliet CVA, van Heteren JAA, Breugem CC, Smit AL, Stegeman I.

[Impact of unilateral congenital aural atresia on academic Performance: A systematic review.](#)

Int J Pediatr Otorhinolaryngol. 2018 Nov;114:175-179. doi: 10.1016/j.ijporl.2018.09.002. Epub 2018 Sep 8.

BACKGROUND: Little is known about the academic performance of children with unilateral congenital aural atresia (CAA).

OBJECTIVE: of review: Our objective was to summarize what is known about the academic performance of children with hearing loss by unilateral congenital aural atresia, in order to provide pragmatic recommendations to clinicians who see children with this entity.

TYPE OF REVIEW: Systematic review.

SEARCH STRATEGY: We conducted a systematic search in PubMed Medline, EMBASE, and Cochrane Library combining the terms "atresia" and synonyms with "unilateral hearing loss" and synonyms. Date of the most

recent search was 16 May 2018.

EVALUATION METHOD: Two independent authors identified studies, extracted data, and assessed risk of bias. This review was reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA). Observational studies on the academic achievements of patients of any age with unilateral conductive hearing loss of any level due to congenital aural atresia were included. We considered grade retention, special education, individualized education plans, and parental report of school performance as outcome measures for academic achievement.

RESULTS: Two studies reporting on academic performance of patients with unilateral CAA, which both had a significant risk of bias. One study (n = 140) showed a grade retention rate of 3.6% (n = 5) in total. 15.7% (n = 22) needed special education, and 36.4% (n = 51) used an individualized education program. The second study, reporting on 67 patients with unilateral CAA, showed that 29.9% (n = 20) of the patients received school intervention, and 25.4% (n = 17) had learning problems.

CONCLUSION: Current evidence regarding the effect of unilateral congenital aural atresia on academic performance is sparse, inconclusive and has a significant risk of bias. High quality observational studies assessing the effects of aural atresia on academic performance in these patients should be initiated.

Vancor E, Shapiro ED, Loyal J.

[Results of a Targeted Screening Program for Congenital Cytomegalovirus Infection in Infants Who Fail Newborn Hearing Screening.](#)

J Pediatr Infect Dis Soc. 2019 Mar 28;8(1):55-59. doi: 10.1093/jpids/pix105.

BACKGROUND: Congenital cytomegalovirus (CMV) infection is a major cause of sensorineural hearing loss. By law, newborns in Connecticut who fail newborn hearing screening are tested for infection with CMV. This targeted screening is controversial, because most children with congenital CMV infection are asymptomatic, and CMV-related hearing loss can have a delayed onset. Our hospital uses a saliva polymerase chain reaction (PCR) assay (confirmed by a urine PCR assay) to detect CMV. Here, we report the results of the first year of our screening program.

METHODS: We reviewed the medical records of newborns in the Yale New Haven Health System who failed the newborn hearing screening test between January 1 and December 31, 2016.

RESULTS: Of 10964 newborns, 171 failed newborn hearing screening, and 3 of these newborns had positive saliva CMV PCR test results. Of these 3 newborns, 2 had positive results on the confirmatory test (for 1 of them the confirmatory test was not performed until the infant was 10 weeks old), and 1 had a negative result on the confirmatory test. Three additional newborns with congenital CMV infection were tested because of clinical indications (1 for ventriculomegaly on prenatal ultrasound and 2 for CMV infection of the mother). Results of audiology follow-up were available for 149 (87.1%) of the 171 newborns who failed newborn hearing screening; 127 (85.2%) had normal results.

CONCLUSION: Our targeted screening program for congenital CMV infection had a low yield. Consideration should be given to other strategies for identifying children at risk of hearing loss as a result of congenital CMV infection.

Vukkadala N, Giridhar SBP, Okumura MJ, Chan DK.

[Seeking equilibrium: The experiences of parents of infants and toddlers who are deaf/hard-of-hearing.](#)

J Pediatr Rehabil Med. 2019;12(1):11-20. doi: 10.3233/PRM-170528.

PURPOSE: To identify key determinants of the quality of life of caregivers of infants and toddlers (< 3 years) who are deaf/hard-of-hearing (DHH).

METHODS: We conducted focus groups with providers for children who are DHH as well as interviews with hearing parents of infants and toddlers who are DHH. A multi-step qualitative analysis on interview data using grounded theory was performed, and an iterative analysis to investigate codes to characterize specific topics in caring for deaf infants and toddlers was conducted.

RESULTS: Four focus groups (n= 33) and six semi-structured interviews (n= 7) were conducted. The major theoretical code found was the "Search for Equilibrium" in parenting which arose from the three main categories of the caregiver role/experience: (1) being a parent - modifying parenting style as a result of their child's hearing loss, (2) being a mediator - modulating and filtering interactions between their child and their child's environment, and (3) being a navigator - managing the logistics of the medical and educational system.

CONCLUSIONS: For hearing parents, the diagnosis of hearing loss requires changes in multiple domains of parenting. Support in each of these areas is critical for parents to restore a sense of equilibrium that is central to their quality of life. This framework provides a way to categorize parent experiences and may act as a template for focused interventions in the three identified domains.

Walker EA, Curran M, Spratford M, Roush P.

[Remote microphone systems for preschool-age children who are hard of hearing: access and utilization.](#)

Int J Audiol. 2019 Apr;58(4):200-207. doi: 10.1080/14992027.2018.1537523. Epub 2019 Jan 5.

OBJECTIVES: Children who are hard of hearing (CHH) have restricted access to auditory-linguistic information. Remote-microphone (RM) systems reduce the negative consequences of limited auditory access. The purpose of this study was to characterise receipt and use of RM systems in young CHH in home and school settings.

DESIGN: Through a combination of parent, teacher, and audiologist report, we identified children who received RM systems for home and/or school use by 4 years of age or younger. With cross-sectional surveys, parents estimated the amount of time the child used RM systems at home and school per day.

STUDY SAMPLE: The participants included 217 CHH.

RESULTS: Thirty-six percent of the children had personal RMs for home use and 50% had RM systems for school. Approximately, half of the parents reported that their children used RM systems for home use for 1-2 hours per use and RM systems for school use for 2-4 hours per day.

CONCLUSIONS: Results indicated that the majority of the CHH in the current study did not receive RM systems for home use in early childhood, but half had access to RM technology in the educational setting. High-quality research studies are needed to determine ways in which RM systems benefit pre-school-age CHH.

Wang LA, Smith PB, Laughon M, Goldberg RN, Ku LC, Zimmerman KO, Balevic S, Clark RH, Benjamin DK, Greenberg RG; Best Pharmaceuticals for Children Act – Pediatric Trials Network Steering Committee.

[Prolonged furosemide exposure and risk of abnormal newborn hearing screen in premature infants.](#)

Early Hum Dev. 2018 Oct;125:26-30. doi: 10.1016/j.earlhumdev.2018.08.009. Epub 2018 Sep 4.

BACKGROUND: At very high doses, furosemide is linked to ototoxicity in adults, but little is known about the risk of hearing loss in premature infants exposed to furosemide.

AIMS: Evaluate the association between prolonged furosemide exposure and abnormal hearing screening in premature infants.

STUDY DESIGN: Using propensity scoring, infants with prolonged (≥ 28 days) exposure to furosemide were matched to infants never exposed. The matched sample was used to estimate the impact of prolonged furosemide exposure on the probability of an abnormal hearing screen prior to hospital discharge.

SUBJECTS: A cohort of infants 501-1250 g birth weight and 23-29 weeks gestational age discharged home from 210 neonatal intensive care units in the United States (2004-2013).

OUTCOME MEASURES: We defined abnormal hearing screen as a result of either “fail” or “refer” for either ear.

RESULTS: Altogether, 1020 infants exposed to furosemide for ≥ 28 days were matched to 790 unique infants never exposed, yielding a total of 1042 matches due to sampling with replacement and propensity score ties. Matching resulted in a population similar in baseline characteristics. After adjusting for covariates, the proportion of infants with an abnormal hearing screen in the furosemide-exposed group was not significantly higher than the never-exposed group (absolute difference 3.0% [95% CI -0.2-6.2%], $P = 0.07$).

CONCLUSIONS: Prolonged furosemide exposure was associated with a positive, but not statistically significant, difference in abnormal hearing screening in premature infants. Additional studies with post-hospital discharge audiology follow-up are needed to further evaluate the safety of furosemide in this population.

Wang Q, Xiang J, Sun J, Yang Y, Guan J, Wang D, Song C, Guo L, Wang H, Chen Y, Leng J, Wang X, Zhang J, Han B, Zou J, Yan C, Zhao L, Luo H, Han Y, Yuan W, Zhang H, Wang W, Wang J, Yang H, Xu X, Yin Y, Morton CC, Zhao L1, Zhu S, Shen J, Peng Z.

[Nationwide population genetic screening improves outcomes of newborn screening for hearing loss in China.](#)

Genet Med. 2019 Mar 20. doi: 10.1038/s41436-019-0481-6. [Epub ahead of print]

PURPOSE: The benefits of concurrent newborn hearing and genetic screening have not been statistically proven due to limited sample sizes and outcome data. To fill this gap, we analyzed outcomes of newborns with genetic screening results.

METHODS: Newborns in China were screened for 20 hearing-loss-related genetic variants from 2012 to 2017. Genetic results were categorized as positive, at-risk, inconclusive, or negative. Hearing screening results, risk factors, and up-to-date hearing status were followed up via phone interviews.

RESULTS: Following up 12,778 of 1.2 million genetically screened newborns revealed a higher rate of hearing loss by three months of age among referrals from the initial hearing screening (60% vs. 5.0%, $P < 0.001$) and a lower rate of lost-to-follow-up/documentation (5% vs. 22%, $P < 0.001$) in the positive group than in the inconclusive group. Importantly, genetic screening detected 13% more hearing-impaired infants than hearing screening alone and identified 2,638 (0.23% of total) newborns predisposed to preventable ototoxicity undetectable by hearing screening.

CONCLUSION: Incorporating genetic screening improves the effectiveness of newborn hearing screening programs by elucidating etiologies, discerning high-risk subgroups for vigilant management, identifying additional children who may benefit from early intervention, and informing at-risk newborns and their maternal relatives of increased susceptibility to ototoxicity.

Wasser J, Ari-Even Roth D, Herzberg O, Lerner-Geva L, Rubin L.

[Assessing and monitoring the impact of the national newborn hearing screening program in Israel.](#)

Isr J Health Policy Res. 2019 Mar 11;8(1):30. doi: 10.1186/s13584-019-0296-6.

BACKGROUND: The Israeli Newborn Hearing Screening Program (NHSP) began operating nationally in January 2010. The program includes the Otoacoustic Emissions (OAE) test for all newborns and Automated Auditory Brainstem Response (A-ABR) test for failed OAE and infants at risk for auditory neuropathy spectrum disorders. NHSP targets are diagnosis of hearing impairment by age three months and initiation of habilitation by six months.

OBJECTIVES: (1) Review NHSP coverage; (2) Assess NHSP impact on age at diagnosis for hearing impairment and age at initiation of habilitation; (3) Identify contributing factors and barriers to NHSP success.

METHODS: (1) Analysis of screening coverage and referral rates for the NHSP; (2) Analysis of demographic data, results of coverage, age at diagnosis and initiation of habilitation for hearing impaired infants pre-implementation and post-implementation of NHSP from 10 habilitation centers; (3) Telephone interviews with parents whose infants failed the screening and were referred for further testing.

RESULTS: The NHSP coverage was 98.7% (95.1 to 100%) for approximately 179,000 live births per year for 2014-2016 and average referral rates were under 3%. After three years of program implementation, median age at diagnosis was 3.7 months compared to 9.5 months prior to NHSP. The median age at initiation of habilitation after three years of NHSP was 9.4 months compared to 19.0 prior to NHSP. Parents (84% of 483 sampled) with infants aged 4-6 months participated in the telephone survey. While 84% of parents reported receiving a verbal explanation of the screening results, more than half of the parents reported not receiving written material. Parental report of understanding the test results and a heightened level of concern over the failed screen were associated with timely follow-up.

CONCLUSIONS: The findings indicate high screening coverage. The program reduced ages at diagnosis and initiation of habilitation for hearing impaired infants. Further steps needed to streamline the NHSP are improving communication among caregivers to parents to reduce anxiety; increasing efficiency in transferring information between service providers using advanced technology while ensuring continuum of care; reducing wait time for follow-up testing in order to meet program objectives. Establishment of a routine monitoring system is underway.

Weiss A, Sommer G, Schindera C, Wengenroth L, Karow A, Diezi M, Michel G, Kuehni CE; Swiss Paediatric Oncology Group (SPOG).

[Hearing loss and quality of life in survivors of paediatric CNS tumours and other cancers.](#)

Qual Life Res. 2019 Feb;28(2):515-521. doi: 10.1007/s11136-018-2021-2. Epub 2018 Oct 10.

PURPOSE: Hearing loss, a complication of cancer treatment, may reduce health-related quality of life (HRQoL), especially in childhood cancer survivors of central nervous system (CNS) tumours who often have multiple late effects. We examined the effect of hearing loss on HRQoL in young survivors of CNS and other childhood cancers.

METHODS: Within the Swiss Childhood Cancer Survivor Study, we sent questionnaires about hearing loss and HRQoL (KIDSCREEN-27) to parents of survivors aged 8-15 years. We stratified the effect of hearing loss on HRQoL by cancer diagnosis, using multivariable logistic regression and adjusting for sociodemographic and clinical factors.

RESULTS: Hearing loss was associated with impaired physical well-being [unadjusted estimated differences -4.6 (CI -9.2, -0.1); adjusted -4.0 (CI -7.6, -0.3)] and peers and social support [unadjusted -6.7 (CI -13.0, -0.3); adjusted -5.0 (CI -10.5, 0.9)] scores in survivors of CNS tumours (n=123), but not in children diagnosed with other cancers (all p-values >0.20, n=577).

CONCLUSION: Clinicians should be alert to signs of reduced physical well-being and impaired relationships with peers. Especially survivors of CNS tumours may benefit most from strict audiological monitoring and timely intervention to mitigate secondary consequences of hearing loss on HRQoL.

Yazici A, Coskun ME.

[The effect of ventilation tube insertion to the health-related quality of life in a group of children in Southeast Anatolia.](#)

Clin Otolaryngol. 2018 Dec;43(6):1578-1582. doi: 10.1111/coa.13220. Epub 2018 Sep 17.

OBJECTIVE: To demonstrate the influence of ventilation tube insertion to the quality of life in a group of children in Southeast Anatolia by Otitis Media 6-item (OM6) questionnaire.

DESIGN: Patients who underwent ventilation tube insertion due to otitis media with effusion (OME) at Otorhinolaryngology Department of Gaziantep University between December 2016 and April 2017 were enrolled in this prospective study. All patients were evaluated with the OM-6 survey before operation and 6 weeks after surgery.

RESULTS: The mean age of 45 patients out of 50 accounted for 67.64 ± 42.89 months with 27 (60%) males and

18 (40%) females. The numbers of preoperative and postoperative overall OM6 scores represented a significant improvement with 4.34 and 2.16, respectively. Moreover, each domain of OM6 (physical suffering, hearing loss, speech impairment, emotional distress, activity limitations and caregiver concerns) showed statistically significant difference.

CONCLUSION: Ventilation tube insertion procedure provided a significant improvement in a group of children in Southeast Anatolia suffering from chronic OME in terms of Quality of Life (QOL) assessed by OM6. We believe that OM6 is a useful tool for evaluating the patients' health-related quality of life and for providing additional information to the caregivers' or families' enquiries regarding the consequences of surgical intervention.

Yimtae K, Israsena P, Thanawirattananit P, Seesutas S, Saibua S, Kasemsiri P, Noymai A, Soonrach T.

[A Tablet-Based Mobile Hearing Screening System for Preschoolers: Design and Validation Study.](#)

JMIR Mhealth Uhealth. 2018 Oct 23;6(10):e186. doi: 10.2196/mhealth.9560.

BACKGROUND: Hearing ability is important for children to develop speech and language skills as they grow. After a mandatory newborn hearing screening, group or mass screening of children at later ages, such as at preschool age, is often practiced. For this practice to be effective and accessible in low-resource countries such as Thailand, innovative enabling tools that make use of pervasive mobile and smartphone technology should be considered.

OBJECTIVE: This study aims to develop a cost-effective, tablet-based hearing screening system that can perform a rapid minimal speech recognition level test.

METHODS: An Android-based screening app was developed. The screening protocol involved asking children to choose pictures corresponding to a set of predefined words heard at various sound levels offered in a specifically designed sequence. For the app, the set of words was validated, and their corresponding speech power levels were calibrated. We recruited 122 children, aged 4-5 years, during the development phase. Another 63 children of the same age were screened for their hearing abilities using the app in version 2. The results in terms of the sensitivity and specificity were compared with those measured using the conventional audiometric equipment.

RESULTS: For screening purposes, the sensitivity of the developed screening system version 2 was 76.67% (95% CI 59.07-88.21), and the specificity was 95.83% (95% CI 89.77-98.37) for screening children with mild hearing loss (pure-tone average threshold at 1, 2, and 4 kHz, >20 dB). The time taken for the screening of each child was 150.52 (SD 19.07) seconds (95% CI 145.71-155.32 seconds). The average time used for conventional play audiometry was 11.79 (SD 3.66) minutes (95% CI 10.85-12.71 minutes).

CONCLUSIONS: This study shows the potential use of a tablet-based system for rapid and mobile hearing screening. The system was shown to have good overall sensitivity and specificity. Overall, the idea can be easily adopted for systems based on other languages.

Yoshinaga-Itano C, Sedey AL, Wiggin M, Mason CA.

[Language Outcomes Improved Through Early Hearing Detection and Earlier Cochlear Implantation.](#)

Otol Neurotol. 2018 Dec;39(10):1256-1263. doi: 10.1097/MAO.0000000000001976.

HYPOTHESIS: Early identification and intervention, earlier cochlear implantation, and mother's level of education will directly and/or indirectly impact the language outcomes of children with cochlear implants (CIs).

BACKGROUND: Identifying factors that contribute to the wide range of language outcomes in children who use CIs will assist healthcare and rehabilitation professionals in optimizing service delivery for this population. Universal newborn hearing screening provides an opportunity to examine the relationship between meeting the early hearing detection and intervention (EHDI) 1-3-6 guidelines and child language outcomes. These guidelines recommend screening by 1 month, confirmation of hearing loss by 3 months, and intervention by 6 months of age.

METHODS: Participants were 125 children with CIs ranging from 13 to 39 months of age. Language ability was measured using the Child Development Inventory and MacArthur-Bates Communicative Development Inventories.

RESULTS: Meeting EHDI 1-3-6, higher levels of maternal education and earlier cochlear implant activation had a direct, positive impact on language outcomes. Meeting the EHDI 1-3-6 guidelines also had an indirect positive effect on language outcomes via increasing the probability that the children's CIs would be activated earlier. Maternal education did not significantly predict age of cochlear implant activation nor whether a child met EHDI 1-3-6.

CONCLUSION: Ensuring families meet the EHDI 1-3-6 guidelines is an early step that can lead to higher language outcomes and also earlier cochlear implantation.

Zeitler DM, Sladen DP, DeJong MD, Torres JH, Dorman MF, Carlson ML.

[Cochlear implantation for single-sided deafness in children and adolescents.](#)

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OBJECTIVE: To evaluate outcomes in pediatric and adolescent patients with single-sided deafness (SSD) undergoing cochlear implantation.

METHODS: A retrospective cohort design at two tertiary level academic cochlear implant centers. The subjects included nine children ages 1.5 to 15 years-old with single-sided deafness (SSD) who had undergone cochlear implantation in the affected ear. Objective outcome measures included were speech reception testing in quiet and noise, bimodal speech reception threshold testing in noise, tinnitus suppression, and device usage.

RESULTS: Nine pediatric and adolescent patients with SSD were implanted between 2011 and 2017. The median age at implantation was 8.9 years (range, 1.5-15.1) and the children had a median duration of deafness 2.9 years (range, 0.8-9.5). There was variability in testing measures due to patient age. Median pre-operative aided word recognition scores on the affected side were <30% regardless of the testing paradigm used. Six patients had pre-operative word testing (4 CNC, median score 25%; 2 MLNT, 8% and 17%). Four patients had pre-operative sentence testing (3 AzBio, median score 44%; 1 HINT-C, 57%). Median post-implantation follow-up interval was 12.3 months (range, 3-27.6 months). Six subjects had post-operative word recognition testing (CNC median, 70%; MLNT 50%, 92%) with a median improvement of 45.5% points. Five subjects had post-operative sentence testing (AzBio, median 82%; HINT, median 76%), with a median improvement of 40.5% points. Eight patients are full time users of their device. Tinnitus and bimodal speech reception thresholds in noise were improved.

CONCLUSION: Pediatric subjects with SSD benefit substantially from cochlear implantation. Objective speech outcome measures are improved in both quiet and noise, and bimodal speech reception thresholds in noise are greatly improved. There is a low rate of device non-use.

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[Cochlear Implantation for Single-Sided Deafness: A New Treatment Paradigm.](#)

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ABSTRACT: Unilateral severe-to-profound sensorineural hearing loss (SNHL), also known as single sided deafness (SSD), is a problem that affects both children and adults, and can have severe and detrimental effects on multiple aspects of life including music appreciation, speech understanding in noise, speech and language acquisition, performance in the classroom and/or the workplace, and quality of life. Additionally, the loss of binaural hearing in SSD patients affects those processes that rely on two functional ears including sound localization, binaural squelch and summation, and the head shadow effect. Over the last decade, there has been increasing interest in cochlear implantation for SSD to restore binaural hearing. Early data are promising that cochlear implantation for SSD can help to restore binaural functionality, improve quality of life, and may facilitate reversal of neuroplasticity related to auditory deprivation in the pediatric population. Additionally, this new patient population has allowed researchers the opportunity to investigate the age-old question “what does a cochlear implant (CI) sound like?.”