CHAPTER THREE

Children Who Are Hard of Hearing: Still Forgotten?

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Abstract

In the 1970's, Dr. Julia Davis called attention to major gaps in research and service delivery for children who are hard of hearing. She referred to this group as "Our Forgotten Children" and many of the issues she raised then remain true today. The need to address these gaps is paramount in an era where children are identified in infancy and have access to early interventions. Although it is presumed that these service innovations will lead to improved outcomes, there is a critical need for research to determine if earlier interventions are bringing about expected results for the majority of children. In an effort to address research gaps and strengthen the evidence guiding practice, a multi-site, longitudinal study of the outcomes of children with mild to severe hearing loss (Outcomes of Children with Hearing Loss; OCHL) was implemented. The aims of this chapter are to 1) describe the goals and the design of this multi-site project, 2) discuss the research team's working hypothesis that inconsistent access to linguistic input places some children and some aspects of development at increased risk for communication delays, and 3) summarize selected main findings regarding outcomes and influential factors. Implications for service delivery are described.

This article focuses on the needs of children who are hard of hearing (HH); specifically those with bilateral, persistent hearing losses in the mild through moderately-severe range, who are fit with hearing aids (HAs) and rely on spoken language for communication and learning. Historically, HH children have been identified around 2 years of age or later (Halpin, Smith, Widen, & Chertoff, 2010), underrepresented in research (Moeller, Tomblin, Yoshinaga-Itano, Connor, & Jerger, 2007), and underserved in regular educational settings (Davis, 1977). Recent evidence shows that the practice of universal newborn hearing screening (NHS) has been effective in reducing the ages of diagnosis for infants with varying degrees of hearing loss (Durieux-Smith, Fitzpatrick, & Whittingham, 2008). NHS has also led to increased opportunities to study HH children in infancy and renewed interest in addressing existing research gaps. Research specific to HH children is needed to understand ways in which the presence of partial hearing loss may impact early linguistic development. Such work is essential for building an evidence base to guide clinical and educational practices.

Davis and colleagues (1977) used the term "Our Forgotten Children" to describe students who are HH. It is of value to consider the historical concerns in light of current practices to determine which issues remain unresolved. Davis observed that the sole responsibility for educating HH children in the schools typically was assigned to regular education teachers, who frequently lacked specific training or understanding of the subtle consequences of hearing loss on learning. She described a tendency for adults to underestimate HH children's needs. This may occur if teachers experience successful communication during face-to-face interactions with HH students, thus leading the teachers to underestimate the extent to which noise and subtle language gaps may serve as barriers to learning in classroom settings. Consequently, teachers may ascribe children's difficulties to behaviors unrelated to hearing loss (see Lewis, Spaulding, & Valente, this volume) as evidenced by frequent reports to clinicians that a HH child "hears me fine, but just does not pay attention." This attitude fits with previous research documenting limited vigilance in monitoring HH children's language and academic outcomes (Davis, Stelmachowicz, Shepard, & Gorga, 1981) and amplification needs. However, based on a classic study of the outcomes of 40 HH children, it was concluded that

any degree of hearing loss may place a child at risk for language delays (Davis, Elfenbein, Schum, & Bentler, 1986). A recent study showed that 25 4- and 5-year-old HH children were delayed in speech and language skills, on average, compared to peers with normal hearing (Fitzpatrick, Crawford, Ni, & Durieux-Smith, 2011). These collective concerns underscore the importance of prospectively examining the outcomes of a large cohort of preschool-aged HH children to understand sources of risk for language delays as well as protective factors that can be emphasized to prevent later challenges. Without such data, this group may remain "our forgotten children."

It has been estimated by the National Institutes on Deafness and Other Communication Disorders (NIDCD) that 30,000 children in the U.S. under the age of 6 years have persistent, mild-to-severe hearing loss. In spite of this fairly high prevalence, there has been limited investment in research that provides a clear description of the unique needs of this group (Eisenberg et al., 2007). Because of research gaps, there remains disagreement and skepticism in the professional community about the needs of HH children, especially those on the mildest end of the continuum. In response to these issues, the NIDCD funded two prospective, longitudinal studies focused on understanding the outcomes of young HH children. The Outcomes of Children with Hearing Loss project, described in this chapter, was one of the funded multi-site studies. It is a collaborative effort of researchers at the University of Iowa, Boys Town National Research Hospital, and the University of North Carolina at Chapel Hill. There are three primary aims guiding this multidisciplinary research endeavor:

1. Describe the characteristics of the children and families, their intervention receipt, and factors associated with variations in services.

2. Measure a range of child & family outcomes and compare child outcomes to a well-matched group of children with normal hearing (NH).

3. Explore how variations in child, family, and intervention factors influence children's outcomes, using multivariate analyses.

Design and Methods

The OCHL study design and methods have been described elsewhere (Holte et al., 2012), and therefore will only be summarized briefly in this chapter. The research goals were addressed through an accelerated longitudinal design. This design served the project goals well, allowing the team to maximize the amount of longitudinal and cross-sectional data accrued during the 5-year grant period. If the team had started all children in infancy and followed them prospectively, the sample size would have been limited and insufficient to address multivariate analyses. Instead, the team worked in the first two years of the grant to identify all children who met the following criteria 1) current age within the range of 6 months to 6 years, 11 months, 2) persistent, bilateral hearing loss, 3) better-ear pure tone average (BEPTA at .5, 1, 2, and 4 kHz) ranging from 25 to 75 dB HL, 4) no significant secondary disabilities that would preclude participation in testing, and 5) at least one primary caregiver using spoken English in the home setting. Children were recruited with assistance from state EHDI coordinators, early intervention specialists, audiologists, and educators. Children who met the criteria entered the study for a baseline visit and then were followed prospectively on an annual basis for at least 3 years. Those who were enrolled as infants or toddlers were seen every 6 months until 24 months of age and annually thereafter. A comprehensive set of audiological, speech, language, pre-literacy, cognitive, and psychosocial outcome measures were collected at each visit. Relevant retrospective records were collected to complement the prospective data. The research team maintained regular contact with parents and established community-based collaborations to minimize attrition rates (8.1% for the 5-year project).

The team was successful in enrolling 316 HH children from 13 states. The mean BEPTA for the HH group at the baseline visit was 48.61 dB HL (SD = 13.52). A comparison group of 115 NH children were enrolled. The two groups (NH, HH) were matched on age, cognitive abilities (both groups within the average range), and home characteristics (i.e., use of spoken English, maternal education/socioeconomic status). The research team made a concerted effort to recruit samples that reflected a typical distribution of socioeconomic levels. In the end, the NH and HH samples in the OCHL study are well matched on this dimension, but are more advantaged than a typical U.S. sample. This is a common challenge in longitudinal studies, but needs to be taken into consideration when interpreting the results. Selected results will be highlighted in this chapter, and the sample sizes will vary, depending on the research question and age group(s) analyzed.

A Hypothesis About Factors Influencing Outcomes

A positive finding from the OCHL project to date is that many HH children achieve better speech and language outcomes than were demonstrated by previous generations of children who were HH. Depending on the measure administered, as many as 70% of 5 year olds presented with speech and language scores with the average range compared to test standardization samples (Tomblin, Oleson, Ambrose, Walker, & Moeller, 2014). An important caveat, however, is that on some of these same measures, the average score for HH children was significantly lower than that of the matched comparison group of NH children, who came from families with similar backgrounds and resources. This fits with findings of previous research showing that children with mild hearing loss fell behind their NH classmates, who presumably came from similar family backgrounds (Blair, Peterson, & Viehweg, 1985). Of more concern is the finding that 25-30% of the HH children in the OCHL study performed one standard deviation or more below the level of the standardization sample and their matched peers. This is higher than the 7% rate of language problems that would be expected in a NH group (Tomblin et al., 1997). There is also considerable variation in the outcomes of HH children in the lower-performing group, suggesting that selected children are particularly vulnerable to the effects of hearing loss. Furthermore, the data reveal that selected aspects of linguistic development, such as articulation and morphology, seem to be more susceptible than others to the influence of hearing loss. Thus, the challenge ahead is to determine what factors explain individual differences in outcomes and why some aspects of language learning may represent vulnerable domains.

The OCHL research team has hypothesized that hearing loss may interfere with the consistency and quality of children's access to linguistic exposure in the environment. Access to input is regarded as a critical factor influencing acquisition both for typical children (Slobin, 1985) and for children with specific language impairment (SLI; Leonard, 1989). An inconsistent access hypothesis leads to some specific predictions about language development. First, inconsistent or reduced access would contribute to a reduction in overall input frequency. It is known that input frequency influences how early certain linguistic forms are acquired in NH children (Hsieh, Leonard & Swanson, 1999). Thus, a reduction in the frequency of exposure to certain forms might slow the rate of language development. Second, the inconsistent access hypothesis suggests that aspects of development that are most dependent on the fidelity of the speech signal (e.g., articulation, morphology) may be most vulnerable to the effects of hearing loss on language development. The current work seeks to identify variables beyond the hearing loss that impact audibility and access.

Severity of hearing loss would be expected to influence linguistic access. Studies examining outcomes in groups comprising only HH children do not always find an effect of degree of hearing loss (Davis, et al., 1986; Gilbertson & Kamhi, 1995). However, to adequately explore this question there is a need for a sample of sufficient size with adequate variance in degrees of loss, and not inclusive of children with severe and profound hearing loss. The influence of BEPTA on articulation skills was examined in a subset of OCHL participants; 3 year olds who were HH (n = 110) or NH (n = 45). Results suggested strong and systematic effects of degree of hearing loss on speech outcomes (see Figure 1). Greater degrees of hearing loss were associated with poorer speech outcomes on the Goldman-Fristoe Test of Articulation (Goldman & Fristoe, 2000). Note that the effect sizes (bolded squares in Figure 1) increase as degree of hearing loss increases.

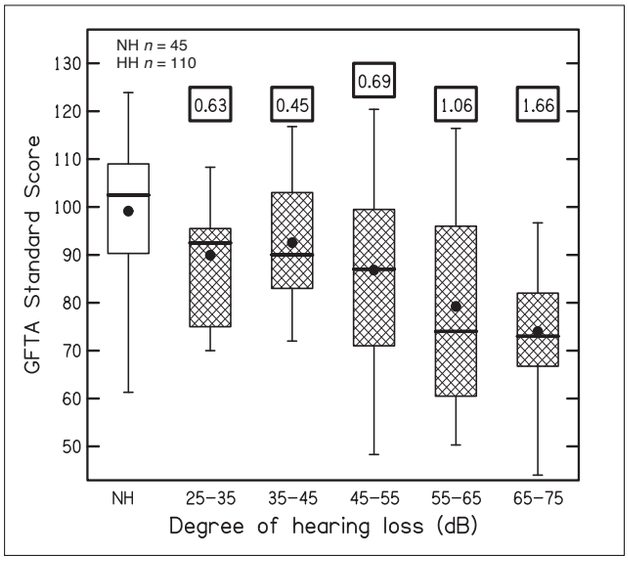

Figure 1. Goldman-Fristoe Test of Articulation (GFTA) standard scores for 3 year olds plotted as a function of hearing level in 10 dB increments compared to age-matched children with NH. Lines in the box plots signify median score, whereas filled circles represent means. Effect sizes (Cohen's d) are shown in the bolded rectangles above the distributions.

Results also suggested that children with hearing loss greater than 45 dB HL were at greater risk for delays in speech than children with hearing loss less than 45 dB HL. Examination of the interquartile ranges in Figure 1 reveals considerable overlap across the categories, however, with some children functioning within normal limits and others well below. This suggests that other influential factors are at play and further supports the need to go beyond BEPTA to understand what may be impacting children's access to the input.

Reduced or inconsistent access to input may occur for a number of reasons beyond severity of loss. First, infants who are HH experience periods without amplification. Even in an era of newborn hearing screening, the average age of HA fitting for infants with mild to severe hearing loss is approximately 6 to 11 months of age (Spivak, Sokol, Auerbach, & Gershkovich, 2009; Walker et al., 2013). This suggests that infants who are HH have a period of restricted auditory access prior to HA fitting. Once HAs are fit, infants experience variations in their daily use of the technology, especially during the second year of life (Walker, et al., 2013). Additionally, HAs themselves have limitations, including restricted bandwidth that limits access to consonants with energy in the high frequency range, especially for child or female talkers (Stelmachowicz, Pittman, Hoover, & Lewis, 2001). Furthermore, young children do not live in well-controlled acoustic environments. Rather, they go in and out of noisy and reverberant settings. Noise and reverberation levels present in typical settings are known to negatively affect speech perception even in children with NH (Klatte, Lachmann, & Meis, 2010), representing a further source of limited access to input for HH children. The OCHL research team suspects that these multiple factors combine to impact the child's access to input and, ultimately, the child's cumulative auditory experience. These issues, in turn, have consequences for child language development.

A global profile of HH children's outcomes at 3 years of age (see Figure 2) lends support to the proposal that selected domains of language learning may be especially vulnerable to the effects of hearing loss. This profile was generated based on work by Tomblin et al. (2014) that explored the contributions of aided hearing to children's speech and language outcomes. Compared to the standardization sample for selected tests, children with moderate to moderately-severe hearing loss demonstrated relative weaknesses in syntax (M = 84.48; SD = 13.89) and articulation skills (M = 83.20; SD = 19.04), two areas that would appear to be dependent on processing fine details in the input. Although the performance of children with mild hearing loss compares more favorably to the standardization samples note that their standard scores trended toward the lower end of the average range for syntax (M = 90.84; SD = 12.65) and articulation (M = 93.00; SD = 12.97). In contrast, performance of both HH groups was within the average range on basic concepts and pragmatics.

The OCHL team has recently been exploring whether these early-observed vulnerabilities resolve with age or whether they persist into the school-age years. Recent research from the OCHL team suggests that some children demonstrate persistent vulnerability in grammatical morphology (Koehlinger, Owen Van Horne, & Moeller, 2013). Accuracy of production of grammatical markers on verbs was evaluated by coding the use of grammatical word endings in spontaneous language samples from 145 HH children and 40 NH children who were 3 or 6 years of age. Significant between-group differences were found with HH children making more errors than the NH children at both ages. Although many children in the HH group performed like their NH peers, some HH children (38%-63%) fell more than 1 standard deviation below the NH group at both ages. The HH children in the 6-year-old group were identified as having hearing loss on average two years later than the 3 year olds, which is important to consider. The question about whether early deficits resolve needs to be addressed using longitudinal rather than cross sectional data, which is a future goal

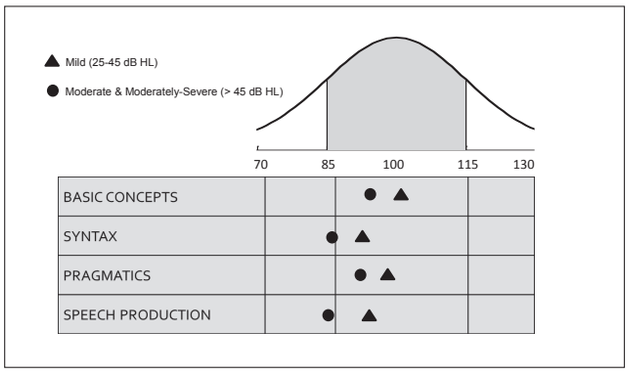

Figure 2. Standard scores for HH children on the Comprehensive Assessment of Spoken Language (basic concepts [n = 72], syntax [n = 70], and pragmatics [n=70]) and the Goldman-Fristoe Test of Articulation [n = 74] at 3 years of age plotted as a profile on the normal distribution. Mean scores for children with mild hearing loss (triangles) are differentiated from those with moderate and moderately-severe hearing loss (circles).

of the OCHL team. In this same study, after controlling for age, auditory variables (BEPTA and audibility measures) contributed to morpheme production accuracy. It may be that children who are HH are at higher risk than NH children for delays in morphology due to reduced or inconsistent exposure to the grammatical morphemes; HH children may sometimes hear morphemes and at other times do not (McGuckian & Henry, 2007). In addition, the fricatives /s/ and /z/ have a prominent role in spoken English grammar, and are challenging for HH children to perceive (Stelmachowicz, Pittman, Hoover, & Lewis, 2002) and produce (Elfenbein, Hardin-Jones, & Davis, 1994).

Similar concern for persistent vulnerability in specific domains is seen in preliminary cross-sectional data from the Goldman-Fristoe Test of Articulation. As shown in Figure 3, significant between-group differences were observed in articulation skills at 3, 5, and 7 years of age (p values < .001). It is clear from examining the distribution of scores at age seven that some children are at particular risk for persistent delays in speech production. These data support the view that some HH children and selected aspects of development are particularly vulnerable in the context of early hearing loss.

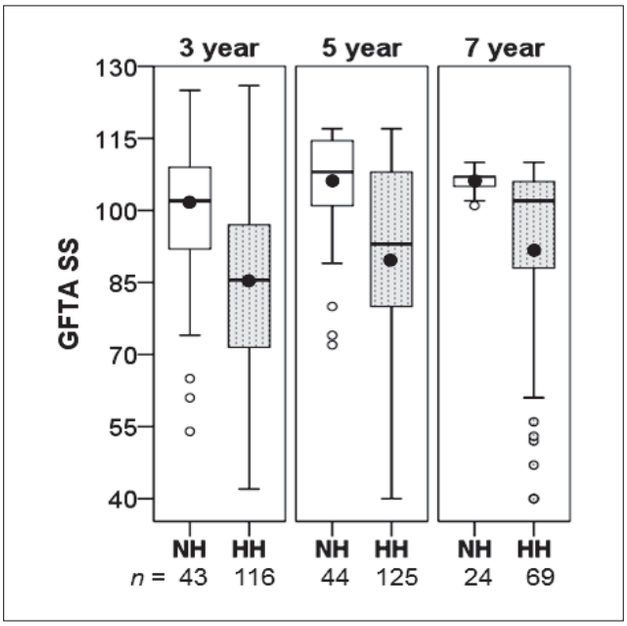

Figure 3. Standard scores on the Goldman-Fristoe Test of Articulation (GFTA) at 3, 5, and 7 years of age for both NH and HH groups.

Modeling Factors That Impact Access to Linguistic Input

The OCHL research team is working to develop a model to guide the identification of factors that link the presence of hearing loss to poor linguistic outcomes in some cases and not others. Although the model continues to evolve, three factors have garnered attention from the research team: 1) audibility of the speech spectrum with amplification, 2) consistency and duration of HA use, and 3) quantity and quality of linguistic input in the home environment.

The OCHL team was interested in studying audibility, but wanted to specifically isolate the contribution of children's hearing aids to outcomes in 180 HH preschoolers (Tomblin, et al., 2014). The contribution of the hearing aids is evaluated using the Speech Intelligibility Index (SII), which is a weighted measure of the proportion of the speech spectrum that is audible to the child with or without hearing aids (ANSI, 1997). It is measured on a scale of 0 (meaning nothing is audible) to 1.0 (fully audible). Isolating the unique contribution of the hearing aids is challenging, however, because the degree to which they can boost the audibility is constrained by the degree of hearing loss; less audibility is possible on the more severe end of the degree of hearing loss continuum. In response to this confound, Tomblin and colleagues derived a solution involving a new variable called the residualized SII (rSII). They removed the variance of the unaided SII that was shared with the aided SII using linear regressions. The residual variance produced by two linear functions in the regression formed the rSII. This new rSII measure reflects the audible hearing provided by the HA independent of unaided hearing. Tomblin et al. found that audibility provided by the HAs was significantly associated with speech and language outcomes and that benefits were inclusive of children with mild and moderate-to-severe hearing loss. These results extend the findings of Stiles, Bentler, and McGregor (2012) who found associations of SII with language outcomes in school-age HH children.

Inconsistent HA use would also be expected to impact children's audibility and access to linguistic input. Consistency of hearing aid use was found to be more challenging at younger ages (i.e., toddlers), with milder degrees of hearing loss, and in families with lower levels of education (Walker, et al., 2013). These findings also speak to the need to provide unique supports to families with children who experience challenges with hearing aid use (Moeller, Hoover, Peterson, & Stelmachowicz,

2009). A related variable exerting influence on audibility benefits appears to be the duration of HA experience. The reader is referred to Tomblin et al. (2014), who demonstrated that the amount of benefit from aided hearing interacted with the duration of the child's hearing aid experience. With longer periods of hearing aid use, audibility provided by the hearing aid became more reliably associated with better outcomes. This finding fits with the view that duration of auditory experience influences language development.

If HH children have inconsistent access to linguistic input, a way to buffer the effects is to provide rich language exposure in the home environment. In fact, many practices in early intervention strive to achieve this very goal. As part of the OCHL project, a subset of families participated in a project where the children wore a digital language processor, the Language ENvironment Analysis (LENA) device, which gathered full day recordings in the home environment. Characteristics of the child's home language environment were examined using LENA automated analyses. Families of 22 HH children, aged 24 – 36 months were compared to eight families with NH children on measures of amount of talk. The groups did not differ in the numbers of words that adults used or in the number of conversational turns. For the HH group, the numbers of conversational turns taken by adults with the HH child, but not adult word counts, were associated with children's receptive language outcomes (VanDam, Ambrose, & Moeller, 2012). The results underscore the importance of qualitative aspects of parent-child interaction in promoting language development. In a more recent examination of data from 28 of the HH toddlers who used the LENA device, the research team found further support for the influence of conversational turns on children's language outcomes. They also found that higher amounts of television time (electronic media) were associated with a reduction in the conversational exchanges known to promote language (Ambrose, VanDam, & Moeller, 2013). These findings suggest the value of cautioning parents about the potentially negative effects of television exposure on the frequency of parent-child interactions. Qualitative aspects of parent-child interactions are currently being further explored by OCHL researchers using video recorded samples.

Summary of Protective Factors

The findings to date have led the researchers to consider factors that provide protection for children against the risks associated with early and persistent bilateral hearing loss. To summarize, better speech and language outcomes are associated with the following factors: 1) milder degrees of hearing loss, 2) better aided audibility, 3) well-fit hearing aids (see McCreery, Walker, & Spratford in this volume), 4) longer duration of HA fitting, 5) more consistent use of amplification, 6) higher quantity and quality of language interactions, and 7) higher levels of maternal education. Several of these factors can be addressed by adhering to best practices.

Clinical Implications

Several clinical implications can be derived from the results to date. Importantly, OCHL results support the audiological practice of optimizing children's audibility with HAs. It is beneficial to provide children with the best possible access to the speech spectrum to support speech and language development. It appears from this work that the benefits of audibility will be increasingly apparent as children gain auditory experience from longer durations of device use, and this supports the practice of early hearing aid fitting. Audiologists routinely promote consistent HA use with families, but OCHL results suggest that families with toddlers, less educated parents, and parents of children with mild hearing loss encounter unique barriers to regular use. Novel counseling strategies that are adapted to the specific needs of these families are required. Audiologists are encouraged to ask questions that are tailored to identifying these unique needs and then provide responsive resources for families. Finally, audiologists are encouraged to support families in their provision of nurturing language interactions, which includes reducing auditory distractions to enhance the child's access to parental-child conversations. Through such practices, children who are HH may no longer be described as "forgotten."

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