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An Introduction to the Outcomes of Children with Hearing Loss Study

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Abstract

The landscape of service provision for young children with hearing loss has shifted in recent years as a result of newborn hearing screening and the early provision of interventions, including hearing technologies. It is expected that early service provision will minimize or prevent linguistic delays that typically accompany untreated permanent childhood hearing loss. The post-newborn hearing screening era has seen a resurgence of interest in empirically examining the outcomes of children with hearing loss to determine if service innovations have resulted in expected improvements in children's functioning. The Outcomes of Children with Hearing Loss (OCHL) project was among these recent research efforts, and this introductory article provides background in the form of literature review and theoretical discussion to support the goals of the study. The OCHL project was designed to examine the language and auditory outcomes of infants and preschool-aged children with permanent, bilateral, mild-to-severe hearing loss and to identify factors that moderate the relationship between hearing loss and longitudinal outcomes. We propose that children who are hard of hearing experience limitations in access to linguistic input, which lead to a decrease in uptake of language exposure and an overall reduction in linguistic experience. We explore this hypothesis in relation to three primary factors that are proposed to influence children's access to linguistic input: aided audibility, duration and consistency of hearing aid (HA) use, and characteristics of caregiver input.

Introduction

The landscape of service provision for young children with hearing loss (HL) has shifted dramatically in recent years as a result of newborn hearing screening and early provision of interventions, including hearing technologies. Early identification and fitting of amplification in infancy is far more common than in the past (Dalzell et al. 2000; Harrison et al. 2003; Spivak et al. 2009), and these contemporary practices are predicated on the belief that they will have long-term positive consequences for children with HL. However, there is a need to gather evidence regarding the benefits of early provision of amplification, and to identify factors that contribute to risk or protection for children who use hearing aids.

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See Supplemental Digital Content, Appendix A List of Acronyms used throughout Outcomes in Children with Hearing Loss Study.

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Fortunately, service delivery changes have occurred in tandem with new opportunities to conduct research on the early outcomes of infants who are deaf or hard of hearing with support from the National Institute on Deafness and Other Communication Disorders (NIDCD) and other federal agencies. A relatively new development in the research domain has been the implementation of longitudinal, multi-center or multi-site studies, which were rare in the past. Multi-site studies are critically needed to overcome issues related to sampling and small sample sizes that limited many previous studies of children with HL, and especially those involving children who are hard of hearing (CHH; Moeller et al. 2007; Tomblin & Hebbeler 2007).

Several contemporary longitudinal research projects are examining the outcomes of children with cochlear implants (CIs) and/or hearing aids (HAs) to gain evidence that will guide clinical practice and future research efforts. Notable examples are a national cohort study in Australia, the *Longitudinal Outcomes of Children with Hearing Impairment* (Ching et al. 2013), and studies with multi-state participation in the U.S., such as the *Early Development of Children with Hearing Loss* project (Nittrouer 2010), and the multi-state assessment system, the *National Early Childhood Assessment Project* (Yoshinaga-Itano 2015). Other recent research projects focus on specific subgroups, either children who are deaf with cochlear implants or children who are hard of hearing (CHH) with hearing aids. Notable examples of studies focused on children with CIs are those conducted by Geers and colleagues (see E & H supplement, 2011), and the *Childhood Development after Cochlear Implantation* project, which is a multi-center national cohort study examining outcomes of children with cochlear implants compared to those with normal hearing (Fink et al. 2007; Niparko et al. 2010).

Multi-center studies focused exclusively on CHH (with mild to severe HL) came about as a result of an NIDCD working group that identified gaps in the scientific knowledge regarding this group of children (Donahue 2007; Eisenberg et al. 2007). The working group acknowledged that, in contrast to research on children with cochlear implants, there was a limited body of research on the developmental outcomes of CHH and the effectiveness of the services provided (Eisenberg et al. 2007). Two research projects were implemented in response: the *Development of Adaptive Behaviors in Children with Hearing Loss* (Stika et al. 2015), and the current project, *Outcomes of Children with Hearing Loss* or *OCHL* (Holte et al. 2012; Tomblin et al. 2014). This monograph describes methods, results, and implications from the 5-year OCHL project.

In the next section, we discuss several theories of typical language development, and the ways in which they emphasize the importance of children's access to linguistic input. This serves as theoretical background to the hypothesis guiding the work in this monograph; that CHH experience limitations in access to linguistic input, which leads to an overall reduction in their linguistic experience. We return to this hypothesis later in this supplement.

Importance of Access to Input for Language Development

Early accounts of child language development downplayed the role of the input to which infants were exposed. Nativist theories, for example, emphasized that input to a language acquisition system did not need to be of particular fidelity or regularity. Instead, it was

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posited that most of language acquisition, particularly phonology and grammar, consisted of a hypothesis-testing process where innate knowledge was used to provide possible alternatives for what language the child was hearing, and the input was only needed for confirmation or rejection of hypotheses. Theorists noted that young children developed linguistic proficiency in spite of the fact the language input they received contained errors and often lacked examples of language forms that appear to be learned by the child (Pullum & Schloz 2002). This view was summarized as the "poverty of the stimulus" notion (Chomsky 1980), and it contended that children's innate linguistic capacities were necessary to supplement the input. We should emphasize that within this perspective, some access to features of the ambient language was considered necessary. For example, vocabulary development was considered to be driven by frequency-dependent learning processes (i.e., how often the word occurred in the input). Over the past two decades, several lines of research on child language development have come to place a greater importance on the child's access to input in shaping what the child learns and how rapidly the child learns language.

Evidence that children are affected by the nature of language input can be found in research concerning caregiver input. Research by Hart and Risley (1995) and Huttenlocher et al. (1991) has shown that the simple amount of words the child was exposed to in the home was associated with subsequent individual differences in language development. Hoff and Naigles (2002) pointed out that rich language input affected children's growth in language in two key ways, (1) by increasing the frequency and variety of words, and (2) by increasing the likelihood that words would be used with different referents and in different sentence contexts. Each of these data-providing features would enhance the child's depth of semantic knowledge. Hoff (2003) has also shown that the length and complexity of utterances provided to children influences their overall rates of language development. Thus, the quantity and quality of the language provided to the child has been shown to be important in influencing the child's language abilities throughout development. Within this research stream there is considerable emphasis on the effects of parent talk on the child's development; however, it is well understood that the child's behavior is also likely to influence the parents' engagement. In this respect, any factor, such as HL, that leads to a potential limitation of a child's communication function and engagement is likely to lead to adjustments in the parent's behavior, including the amount and richness of language they provide to the child.

Research showing that characteristics of language input were associated with individual differences in language growth has been complemented by extensive research suggesting that language learning may draw heavily on statistical learning processes (Elman 1990; Christiansen & Chater 2001; Saffran 2003). Much of this research shows that language is fundamentally structured by probabilistic properties of fine phonetic cues, phonetic segments, syllables, words, and grammatical relations among words. The statistics available in the input involve at least frequency, variance, covariance and conditional probabilities structured over time and sequence. Aslin and colleagues (1998) demonstrated that infants could find patterns in the input and learn the conditional probabilities of phoneme sequences to identify word boundaries. Farmer and colleagues (2006) showed there are a number of subtle acoustic cues in the input that allow children to discover the word classes of nouns

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and verbs. Furthermore, variability in the input (e.g., in phonemes, words, and word forms in sentences) supports children's acquisition of phonetic categories, word types, and grammatical relations (Maye et al. 2002; Rost & McMurray 2009; Richtsmeier et al. 2011). Learning language ultimately involves the acquisition of abstract knowledge about syntactic structure, but we are learning that these abstractions can emerge from concrete acoustic-phonetic properties in the signal. Thus, access to the acoustic-phonetic properties in the input is essential for spoken language learning and a hearing loss could result in reduced learning both with respect to what is learned and how rapidly it is learned.

Finally, one additional stream of research in the last two and a half decades can be found within usage-based accounts of language development (Tomasello 2003; Ibbotson 2013). These theories contend that language is about mappings between forms and meaning. Such mappings involve language forms that often contain multiple words that frequently occur together in association with a particular referent or context. At least initially, the child learns these as *meaning units* without acquiring abstract notions like subjects or infinitives. Children gradually build their knowledge of particular language forms over the course of many exposures. Usage-based accounts, then, assume that much of what the young language learner acquires is available in the input, and frequency of exposure to constructions in communicative contexts drives the child's learning. The usage-based account also emphasizes social aspects of early language learning, including the need for the child to figure out the adult's communicative intentions. The claim is that the child relies on social skills of *intention-reading* and cognitive skills of *pattern-finding* in order to learn. As such, the most potent input for language learning will occur in the context of social engagement and interaction with caregivers and playmates. The usage-based approach is consistent with research emphasizing the importance of a rearing environment that provides frequent opportunities for engagement and child-directed language experience. In summary, both access to input and quality interaction matter for language development. As we noted earlier a hearing loss could reduce both the amount and regularity of language input, but also alter the patterns of communication interactions between the child and caregiver.

We can see that in the last 25 years, there have been at least three important lines of research in child language development that argue that the language input to the child is central to the process of language development and that variations in the quantity and quality of language input should account for a substantial fraction of individual differences in language development. Since these accounts were largely concerned with children with normal hearing (CNH), they have assumed that if input is provided, then it will be used by the child for language learning. Harris (1992) distinguished between *input* versus *uptake* with regard to language experience. CNH could have variable input; however it is assumed in this research that they have normal uptake. That is, if the input is available, the child is likely to take up and use the input for communication and learning. We say likely to take up, because even CNH may not always do so for a variety of reasons. For children with HL, even in the best of circumstances where the environment does provide a rich linguistic experience, they are at greater risk for missing out on some of these learning opportunities as the HL reduces the overall level of input. Clearly, when the input is more limited due to HL, thus limiting

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the potential for uptake, we can envision that the feedstock for language learning becomes even more constrained.

Inconsistent access and variable uptake of language in CHH

This leads to the central hypothesis of much of our research. CHH experience limitations in access to and perception of linguistic input, which leads to a decrease in uptake and an overall reduction in language experience. This is important developmentally, because deficits in language experience have been shown to alter the efficiency of language and auditory-perceptual processes that support early learning (Nittrouer & Burton 2001, 2005; Weisleder & Fernald 2013). Weisleder and Fernald (2013) found that richer language experience (i.e., more exposure to child-directed speech) positively influenced the efficiency of children's language-processing skills that promote language growth. We suggest that CHH encounter a number of potential barriers to linguistic access which can impact the quantity and nature of their language experience. When barriers are minimized or removed (i.e., through effective interventions), CHH may experience protection, demonstrating resilience in language development. In contrast, when barriers to access persist over time, risk for delayed language increases. We contend that factors that influence the child's access to linguistic input may explain some of the extreme variation in outcomes commonly observed in CHH.

Multiple factors that reduce auditory-linguistic access relate to the HL itself and the intervention with amplification, such as audibility, low pass filtering, and spectral degradation. These can alter the nature of the linguistic input received by CHH. For example, the limited bandwidth provided by amplification can render final -s nearly inaudible (Stelmachowicz et al. 2001; 2002). In this circumstance, for example, a child will fail to perceive a subset of grammatical morphemes in the input, and it is as if the talker never used the morpheme. Thus, the input is siphoned by the HL leading to reduced linguistic experience. Key to these notions is that input opportunities are *probabilistic*. Any child is likely to take up only a fraction of available input, and HL has the effect of reducing this probability. HA use or non-use and characteristics of the auditory environment can further impact access to and uptake of the input in CHH at any moment in time. At one moment, the child is in close proximity to the parent in a quiet environment, wearing optimally fit HAs, and engaged in joint attention. In this circumstance, the child is likely to access the input quite well. At another moment, the child is not wearing the HAs, the parent speaks at a distance, there is noise in the environment, and exposure to the input is distorted or missed entirely. Although such "misses" also happen in typical development, we contend that they are far more frequent in the context of childhood HL. Furthermore, we expect that individual CHH will vary in how often and in what ways their access is disrupted, leading to individual differences in outcomes. This supports the need to identify sources of ***inconsistent access*** to linguistic input. Over time, the moments with limited or distorted access accumulate and have the effect of reducing the child's ***cumulative linguistic experience***, which places the child at risk for reductions in language learning efficiency and for language delays.

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A model of factors influencing outcomes in CHH—The OCHL team sought to ***identify factors that influence children's access to linguistic input***, and to determine how these factors may interact over time to exacerbate risk or provide protection. Our proposal has been consolidated into an overall conceptual framework, which is illustrated in Figure 1. The model includes a representation of the child's HL (box on far left) and the child's outcomes (triangle on far right). In the past, researchers have explored the impact of degree of HL on outcomes, and several studies did not find influences of this factor in groups that included only CHH (Davis et al. 1986; Gilbertson & Kamhi 1995; Norbury et al. 2001). This led some to suggest that *any* degree of HL puts a child at risk (Blair et al. 1985; Davis et al. 1986), while others suggested a high incidence of co-occurring language disorders in CHH because degree of HL was not an explanatory factor (Gilbertson & Kamhi 1995). It is important to consider that degree of HL is unlikely to operate alone to influence outcomes, and a number of potentially influential variables such as the audibility provided by hearing aids or the extent of hearing aid use have not been systematically explored in the extant literature. We propose that three factors influence a child's access to linguistic input and subsequent cumulative linguistic experience, including (1) aided audibility, (2) HA use (age at fitting, duration and consistency of use), and (3) quantity and quality of linguistic input provided by caregivers. This aspect of the model is conceptualized as the child's cumulative linguistic experience (represented by the center circle in Figure 1). Although we conceptualize the influence of interventions at three distinct points in the model [signified by (1), (2), (3)], our primary emphasis in the current set of studies is the impact of the provision of hearing aids through audiological intervention. The literature review that follows provides support for the OCHL study goals and the constructs incorporated in this model. Before turning to the literature review, we provide background on the audiological details of the children participating in the OCHL project to orient the reader to the sample.

CHH in the OCHL study—The OCHL participants (317 CHH and 117 CNH) are described in detail in the Methods section of this supplement (Tomblin et al. this issue, XXXX) and subsequent articles. However, some specific audiological details about the CHH are provided here as background. The total number of participant numbers cited above reflects all children who contributed data to the OCHL project. These numbers will vary in subsequent articles in this issue, depending on the focus of the article and inclusion of subgroups relevant to the research questions. The OCHL study focused exclusively on children with bilateral, permanent, mild to severe hearing loss (better-ear pure-tone averages between 25–75 dB HL; with a few exceptions, see Tomblin et al., this volume, pp. XXX[METHOD]) who used HAs (or no amplification in the case of 7 children with mild bilateral HL) rather than cochlear implants. Thirty-eight percent of the children had slight-mild hearing loss, 40% had moderate losses, and 22% had moderately-severe or severe losses. The incidence of progressive HL in the OCHL group was approximately 16% (based on a criterion of a 10 dB shift in pure tone average between at least one visit that was not related to middle ear status changes). Thirteen children (4%) received cochlear implants subsequent to study enrollment, and only their pre-implantation scores are included in analyses reported in this volume. Finally, if we consider types of HL represented at the baseline visit, 5.5% of the children had permanent conductive loss, 68.7% had confirmed sensorineural losses, 22.1% were undetermined (due to lack of bone conduction results), and

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3.6% had mixed HL. This study did not include children whose HL was transient (i.e., due to otitis media only), or children with unilateral HL. Given this sample, our literature review focuses primarily on CHH rather than those with CIs. While children using CIs or HAs are likely to share common features related to developmental risk, communicative challenges, and outcomes, their early auditory-linguistic experiences differ in potentially important ways. In concert with this concept and the NIDCD research request, the OCHL project focused selectively on CHH in order to gain a better understanding of the outcomes and unique needs of this subgroup (Donahue, 2007).

Outcomes of children who are hard of hearing

Children's language outcomes in a contemporary era—One of the primary goals of the OCHL study was to determine the degree to which spoken language is vulnerable or resilient to the effects of HL in an era of early identification and early provision of HAs. Studies prior to the newborn hearing screening era documented that CHH were at risk for delayed spoken language and literacy outcomes (Blair et al. 1985; Davis et al. 1986; Elfenbein et al. 1994; Bess et al. 1998). Davis and colleagues (1986) documented deficits in vocabulary and social skills in school-age students with mild to severe HL. In a related study, Elfenbein et al. (1994) identified a profile of deficits in CHH that also included morphology, advanced syntax, and articulation. This pattern of errors was attributed to the *impact of reductions in acoustic input* experienced by CHH. Children in this earlier era, however, were typically late-identified with later provision of amplification (after 5 years of age in Davis et al. 1986), and may or may not have been fit with hearing aids (Blair et al. 1985). Details regarding the boost in audibility provided by hearing aids were unavailable, and this factor is particularly relevant to a child's access to linguistic input.

In the newborn hearing screening era, the expectation is that early service provision will lead to normalized language abilities in CHH. Several research teams, including OCHL, have been exploring whether the expected benefits from early intervention are being realized. In general, results of several recent studies are mixed, with some large-scale studies suggesting that CHH continue to be at risk for delayed language, while smaller-scale studies report normalization of language abilities in preschoolers. Ching and colleagues (2013) found that 3-year-old children with a broad range in HL severities (mild to profound) demonstrated spoken language outcomes that were more than one standard deviation below age expectations. This finding, however, was based on pooled data for CHH with HAs, children with CIs, children whose native language was other than English, and children with multiple disabilities. Severity of HL, maternal education, gender, and presence of additional disabilities were predictive factors, but age at device fitting was not. Other population-based studies also reported poorer than expected language outcomes (1–2 standard deviations below the mean) for 7 to 8-year-old students with mild to profound HL, and greater severity of HL was a predictor (Wake et al. 2004; Wake et al. 2005). The children in these latter studies were relatively late-identified (M = 21.6 months), and it is not known how this factor may have contributed to poor outcomes.

Some recent smaller scale studies examined the outcomes of children with early access to HAs. Their results suggest that early-identified CHH are resilient to the effects of mild to

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severe HL and demonstrate outcomes that are comparable to age-matched peers. For example, Stika et al. (2015) reported that a majority of CHH performed within the average range compared to CNH on measures of language development at 12 to 18 months of age. It remains an empirical question whether or not these early benefits are sustained in CHH as language demands advance, although a large scale study by Pimperton and colleagues (2014) suggested long-term benefits for reading outcomes when confirmation of HL occurs by 9 months of age. Fitzpatrick and colleagues (2011) reported that a group of children with HL less than 70 dB HL who received the same spoken language intervention program demonstrated normalized language outcomes by 4 to 5 years of age. In a separate study, school-aged CHH demonstrated outcomes within the average range for their age and outperformed children with CIs (Fitzpatrick et al. 2012).

These studies suggest a positive impact of early service provision on CHH, but there is need for replication of these results in larger longitudinal samples. The OCHL sample purposely excluded children with severe to profound HL in order to draw clear conclusions about the outcomes and needs of CHH who use HAs. The OCHL research team sought to address the impact of early provision of well-fit and consistently worn HAs on a range of outcomes measured longitudinally in a large group of children from ages 2 to 6 years.

Consequences of milder degrees of hearing loss on children's outcomes— Another motivation for the OCHL study was the lack of a clear consensus as to whether milder degrees of HL have consequences for children's language and literacy outcomes. A classic, large-scale study by Bess et al. (1998) found that 37% of students identified with minimal sensorineural HL (mild, high frequency, or unilateral) failed a grade in school. This report underscored the potential language and learning consequences of HL of even the mildest degrees. More pertinent to the OCHL study sample are studies that examined the consequences of mild *bilateral* sensorineural HL. Dokovic et al. (2014) identified 144 school-aged children with mild bilateral HL who had not previously received HAs or other services. These students demonstrated deficits in phonological memory and morphosyntactic skills, suggesting consequences for structural aspects of language when mild HL goes untreated. In contrast, other studies suggest that slight/mild HL has little or no impact on language and academic outcomes (Kiese-Himmel & Ohlwein 2003; Wake et al. 2006; Porter et al. 2013). For example, Wake et al. (2006) reported that school-age children with slight/mild bilateral HL showed deficits in short term phonological memory but were not different than matched controls on language, learning, or social-behavioral measures. Similar findings of phonological processing deficits without associated language and learning consequences were reported for children with mild-moderate HL (Briscoe et al. 2001). Collectively, these studies present an unclear picture about the nature or extent of risk, if any, presented by mild or mild-moderate HL, which complicates clinical decision making. Little insight has been gained from these studies about the role of amplification in preventing subtle deficits for language and phonological sensitivity that have been observed in untreated children. Interpretation of the needs of children with milder bilateral HL will be more straightforward if their skills are examined separately from children with unilateral losses and in relation to HA use, as is done in the OCHL study.

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Explaining individual differences in outcomes—A general theme from the literature is that many CHH achieve outcomes that are comparable to peers with normal hearing (Sikora & Plapinger 1994; Gilbertson & Kamhi 1995; Wolgemuth et al. 1998; Briscoe et al. 2001; Norbury et al. 2001), while some participants in these same studies fall significantly below age expectations. Importantly, individual differences in outcomes are not fully understood, and several potential sources of variance remain unexamined. This led to the OCHL study team to explore some unique factors that were expected to lead to variations in children's cumulative auditory-linguistic experience. This brings us back to the model in Figure 1 and a review of selected evidence that supports the notion that three proposed factors (audibility, HA use, language input) moderate the influence of HL on children's outcomes.

Hearing status—It is evident from the literature review above that there are discrepant conclusions about the influence of degree of HL within the subgroup of CHH. Larger scale studies find a role for degree of HL, but most have included a range in degrees from mild to profound, which makes it challenging to draw conclusions about the hard of hearing group in its own right. Several studies of smaller groups of CHH have not found a contribution of degree of HL, but their samples have generally been restricted in size and range. A sufficiently large sample was recruited in the OCHL study to allow the team to re-examine the influence of degree of HL in a sample comprised only of CHH. Our model of inconsistent access predicts that greater severity of HL will place more limits on audibility and access, resulting in poorer auditory and linguistic outcomes. Furthermore, rather than consider hearing status as a single time point variable, we explore the natural history of the child's HL over time and potential effects on outcomes (McCreery et al., this issue, pp. XXXX, pp. XXXX).

Audibility—Few previous studies have examined the degree to which aided hearing influences outcomes in CHH. A notable exception is the work of Stiles et al. (2012) who demonstrated that higher levels of aided audibility were associated with better language outcomes in school-aged CHH. An earlier study from the OCHL project (Tomblin et al. 2014) found that preschool-aged CHH with stronger aided audibility had better speech production and language skills than those with less aided audibility. Audibility was also found to be associated with accuracy of verb morphology in CHH, as reported by the OCHL team (Koehlinger et al. 2013). Studies have not yet explored the impact of audibility on language and auditory skills using longitudinal designs with preschool-aged children, which is a central goal of the OCHL study. This question is relevant to exploring the ways in which provision of HAs may protect children from reduced linguistic experience throughout early development (McCreery et al., this issue, pp. XXXX).

HA use—Studies also report diverging conclusions regarding the degree to which age at HA fitting influences language development in CHH. Some studies find significant contributions to language (Sininger et al., 2010), while others do not (Ching et al. 2013; Fitzpatrick et al. 2011). Tomblin and colleagues (2014) in an earlier cross-sectional report from the OCHL study found that the contributions of HAs to language development were more readily identifiable with longer durations of HA use. Longitudinal analyses are needed

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to confirm this finding. The impact of duration of HA use is also likely to be influenced by the consistency of children's device use. Recent studies suggest that device use is variable in CHH, particularly for the youngest children, those with the mildest degree of HL, and in homes with lower levels of maternal education (Moeller et al. 2009; Walker et al. 2013; Munoz et al. 2015). DesJardin (2005) demonstrated that parental report of self-efficacy around managing and promoting device use was stronger in families of children with CIs than in those whose children had HAs, which suggests the need for further research regarding HA use and strategies for promoting use in CHH. A recent study by Marnane and Ching (2015) examined longitudinal trends in device use, and did not find it to be a significant predictor of outcomes after controlling for background variables. There is need for additional research documenting how HA use varies over time for CHH and how this variation may influence longitudinal trajectories of language development. Within the framework of the inconsistent access hypothesis, we predict that children with limited hearing aid use will experience greater reductions in language experience and will demonstrate poorer outcomes than those who regularly use amplification (Tomblin et al., this issue, pp. XXXX; Walker et al., this issue, pp. XXXX).

Quality of linguistic input—Interventions that seek to optimize the input, including maximizing audibility with HAs (McCreery et al. 2013), coaching families to provide high quality conversational interactions (DesJardin 2005; Stika et al. 2015), and providing high quality early intervention (Nittrouer & Burton 2001) would be expected to offer some protection against reduced access to input. In terms of qualitative features, parental responsiveness (Baumwell et al. 1997; Nittrouer 2010; Tamis-LeMonda et al. 2001) and use of language facilitation techniques (e.g., parallel talk, expansions, open-ended questions, and recasts) are expected to have buffering effects (Fey et al. 1999; Proctor-Williams et al. 2001; Szagun & Stumper 2012). Studies demonstrate that children with HL benefit from exposure to high quality, facilitative features of caregiver input in the home environment (Ambrose et al. 2013; Cruz et al. 2013; DesJardin & Eisenberg 2007; Szagun & Stumper 2012), with frequency of engagement in conversational interactions bearing special importance (Ambrose et al. 2013). Caregiver use of facilitative language features recently has been associated with better outcomes in children with CIs (DesJardin & Eisenberg 2007; Cruz et al. 2013; Szagun & Stumper 2012). Similar evidence has recently begun to emerge in relation to CHH (Vohr et al. 2010; Stika et al., 2015), but evidence is limited. Ambrose et al. (this volume, XXXX) further explores features of caregiver talk and its impact on child language development in CHH.

Educational interventions—Although our focus in the current set of studies is on the audiological intervention with HAs, the model includes a key role for educational interventions (explored in future reports from the OCHL team, Harrison et al. in review). Quality educational interventions would also be expected to offer protection, and this question was examined in school-age CHH by Nittrouer and Burton (2001). They proposed that HL could be modeled as a "deficit in language experience," not just a sensory deficit. To address this hypothesis, they asked whether school-aged CHH would exhibit problems in speech perception and language processing that were comparable to CNH who had histories of deficits in early language experience (e.g., children in low socioeconomic conditions,

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children with chronic otitis media, or both). Speech perception outcomes were reflective of the CHH's restricted access to acoustic information in the speech signal. However, two different groups of CHH (mainstreamed and non-mainstreamed) emerged from the data, and the respective groups relied on distinct perceptual strategies. Children in the mainstream group, particularly those who demonstrated perceptual strategies for speech that mirrored typically-developing children, also performed similarly to them on linguistic tasks. The authors speculated that early, specialized auditory/oral education received by the mainstreamed group allowed them to acquire necessary and sufficient experience to develop effective perceptual strategies for learning their native language. This study pointed to a role for high quality early intervention in shaping perceptual learning strategies underlying language development. Vohr and colleagues (2008) reported that entry to early intervention prior to 3 months of age was associated with better short- and long-term outcomes for children with HL.

The model in Figure 1 illustrates three predicted influences of interventions:

1. Moderating the relationship between HL and experience. The prediction is that provision of well-fit HAs, higher levels of audibility, and consistent HA use will lead to better access to linguistic input and higher levels of cumulative linguistic experience.
2. Contributing to the quality and quantity of experience by encouraging consistent HA use and coaching families to optimize language input and,
3. Contributing to the relationship between language experience and outcomes.

Bidirectional arrows reflect the expectation that audiological and educational interventions will jointly contribute to outcomes, as their effects are difficult to separate. For example, the success of the early language intervention efforts may be impacted by the degree that the HAs are fit appropriately. Consistent HA use may be fostered by joint problem solving of the audiologist and early intervention provider, and then implemented with the family by the early intervention professional. For example, a toddler's temperament can create challenges for parents working to maintain HA use (Moeller et al. 2009; Walker et al. 2013) but support from a knowledgeable early intervention specialist can foster increased HA use over time. Audiological interventions in tandem with educational interventions are also expected to influence the effectiveness of efforts to optimize audibility, promote consistent HA use, and encourage language-rich exposure.

Maternal education—It is widely recognized that factors related to socioeconomic status contribute to variation in children's developmental outcomes (Weisleder & Fernald 2013). The influence of socioeconomic status and/or maternal education on outcomes of CHH has been examined in several studies, but the findings are not uniform. Several find that maternal education level predicts language outcomes in children with HL (Fitzpatrick et al. 2007; Sarant et al. 2009; Fitzpatrick et al. 2011; Ching et al. 2013; Porter et al. 2014), but others do not (Pressman et al. 1999; Yoshinaga-Itano et al. 1998). Further study of the ways in which variations in maternal education level may impact the outcomes of CHH is needed so that interventions can be altered to address the needs of families with fewer resources (Holte et al. 2012; Sacks et al. 2013). Socioeconomic status is either controlled for or

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examined as a predictor in the OCHL study when exploring the effects of various moderators on outcomes.

Measurement of Outcomes

This brings us to the final aspect of the model presented in Figure 1: children's outcomes. In general, the OCHL study was designed to examine a comprehensive set of longitudinal child outcomes to serve as dependent variables in the analyses. The overall approach is described in the Methodological article in this issue (Tomblin et al., this issue, pp. XXXX). The primary focus of the current set of studies includes language outcomes during the preschool years along with outcomes related to speech perception and functional auditory skills. It is expected that early deficits in these areas may cascade to disrupt academic development in the school years, but that topic is currently being explored by the OCHL team and is beyond the scope of the present work. Above we considered factors that may explain individual differences in children's outcomes. One more issue that deserves attention based on the literature is whether CHH may be at differential risk for delays in selected domains of language development. This question was pursued in the OCHL study with measures of varying language domains to offer additional perspectives on the role of access to input in children's language learning.

Linguistic domains at differential risk in CHH—For CHH, it is possible that domains of language that depend on access to the phonetic structure of the input may be especially vulnerable to delays (see Nittrouer et al. 2013 for a discussion in children with CIs). A possible reason for this vulnerability is that HL has the effect of reducing opportunities for perceiving tokens (morphemes, syntactic patterns), particularly those that are perceptually subtle. The OCHL study explored the possibility that grammatical morphology may be especially challenging for CHH by comparing children's vocabulary and morphology outcomes. We reasoned that grammatical morphology is particularly reliant on access to phonetic structure and may be especially sensitive to the effects of reduced access to the input. On the other hand, lexical cues may occur with greater contextual and linguistic support.

The prediction that morphosyntax is vulnerable in CHH due to inconsistent access has some of its roots in the ***surface hypothesis***, originally proposed by Leonard (1989) to explain the grammatical morphology deficits of children with specific language impairment (SLI). Leonard found that grammatical morphemes with short duration and limited perceptual salience presented particular learning challenges for children with SLI. In English, morphemes are often represented by phonemes that are challenging to hear [s, z, t], that may occur in sentence positions that lead to reduced amplitude, or that vary in their frequency of occurrence in the input (Hsieh et al., 1999). In the case of CHH, risk appears to relate to the reduced perceptibility of morphemes in running speech, differential effects of input frequency, and the limited bandwidth of HAs, which further reduces perceptibility and production of English morphemes realized as -s or –z (Kortekaas & Stelmachowicz 2000; Stelmachowicz et al. 2002; Koehlinger et al. 2015).

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A small body of research has focused on the development of morphosyntax in CHH, which is predicted to be vulnerable in the context of HL. Norbury and colleagues (2001) examined the use of finite verb morphology in English in children with SLI compared to children with mild-moderate sensorineural HL. Results showed that CHH were comparable to age-matched hearing controls on accuracy of morphemes on verbs and they outperformed the children with SLI. These findings could be interpreted to refute auditory-based explanations of morphology deficits. However, the youngest CHH demonstrated deficits in morphosyntax, prompting Norbury et al. (2001) to conclude that degraded or disrupted auditory input during early development can lead to delays in linguistic skills, including morphosyntax. Hansson et al. (2007) also reported deficits in verb morphology in CHH that were believed to be associated with low-level perceptual deficits and weaknesses in phonological short-term memory. McGuckian and Henry (2007) found that CHH made a larger number of errors on morphemes that involved challenging-to-hear phonemes (–s or -z) and for those that occur less frequently in the input than other forms (i.e., -3s [walks, runs]), suggesting roles for both perceptibility and input frequency.

Further confirmation that CHH are at risk for delays in morphological development was reported by Koehlinger and colleagues (2013) as part of the OCHL study. Spontaneous production of verb morphology was examined in language samples of 3-and 6-year-old CHH compared to CNH. Results indicated that CHH lagged behind hearing peers in morphosyntax, even at the later age. Although some children performed at age-appropriate levels, more than half fell below the 25th percentile compared to CNH. Koehlinger and colleagues (2015) documented effects of both perceptibility and articulation skills on development of morphosyntax in young CHH. More perceptually-salient morphemes were produced more accurately than less salient forms by the CHH. Finally, Delage and Tuller (2007) documented evidence of *persistent* grammatical delays in French-speaking CHH. More than half of the adolescents they studied with mild and moderate HL demonstrated deficits in phonology and grammar. These results support our view that perceptibility and variations in access may have differential effects on the development of morphosyntax in CHH. Questions about differential vulnerability of morphology in CHH are further explored in Tomblin et al. (this issue, pp. XXX) because they offer an additional test of the effects of inconsistent access.

Inconsistent and/or distorted access to input might also be expected to impact word learning and the size of the lexicon. Fernald and colleagues demonstrated that lower levels of language experience were associated with reduced language processing efficiency and poorer vocabulary development (Fernald et al. 2012; Weisleder & Fernald 2013). If HL impacts language experience in a way that affects language processing, vocabulary deficits would be expected. However, results reported in the literature are also mixed in regard to vocabulary outcomes (see Moeller et al. 2007 for a review). Some recent studies confirm that CHH, on average, are delayed in lexical development compared to CNH (Blamey et al., 2001; Pittman et al., 2005), and that CHH require more exposures to a word than CNH to add it to their lexicon (Pittman et al. 2005; Lederberg & Spencer, 2009; Stelmachowicz et al. 2004). However, Stiles and colleagues (2012) found differences between CNH and CHH in word and nonword repetition and receptive vocabulary, but groups did not differ on word

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learning tasks. These same authors (2013) found that school-aged CHH, like CNH, relied on wordlikeness (i.e., phonotactic probabilities) of English in the service of word learning, but subtle differences between groups were observed. These studies suggest effects of HL on subtle aspects of lexical development, but it is unclear if the extent of impact is comparable to that experienced in morphology. Therefore, we further explored the possible differential effects of HL on grammatical morphology by making a comparison to children's vocabulary outcomes as a first step in looking at this question. If grammatical morphology is particularly challenging for CHH, this finding may have implications for intervention.

Summary

In summary, multiple factors can be expected to impact a child's access to linguistic input and, in turn, moderate the influences of the child's HL on outcomes. The articles in this volume explore the inconsistent access hypothesis by first examining three key factors that are predicted to explain individual differences through their effects on cumulative auditory-linguistic experience. The factors predicted to be influential include aided audibility, duration and consistency of HA use, and characteristics of the child's language environment. Next, we test for the influence of multiple factors, including those proposed, on children's auditory and language outcomes from both cross-sectional and longitudinal viewpoints. We conclude with a summary of key findings and implications for research and service delivery (Moeller & Tomblin, this issue, pp. XXXX).

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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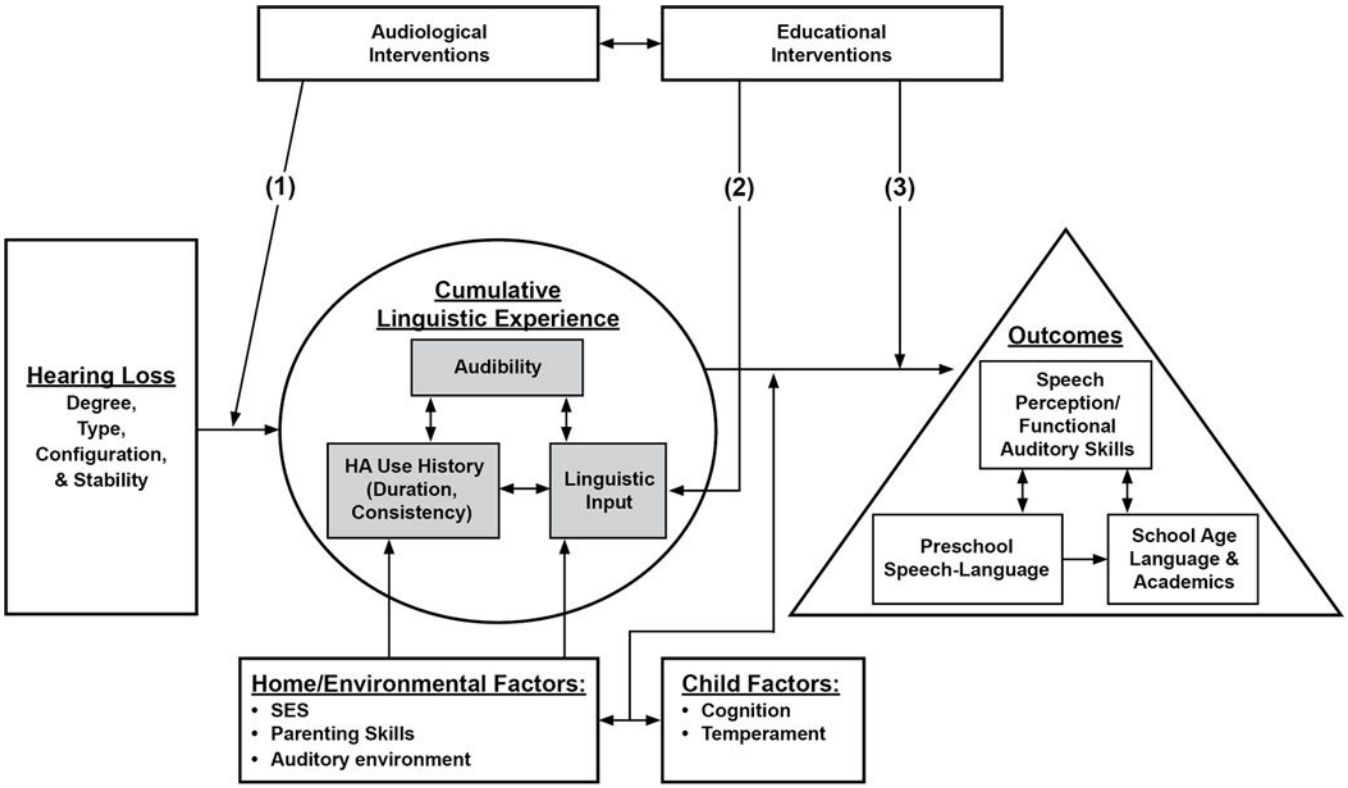

Figure 1.

Model of hypothesized factors that may influence the relationship between childhood hearing loss and developmental outcomes.