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Speech Sound Production in Two-Year-Olds who are Hard of Hearing

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Abstract

Purpose—The purpose of the study was to 1) compare the speech sound production abilities of 2-year-old children who are hard of hearing (HH) to children with normal hearing (NH), 2) identify sources of risk for individual children who are HH, and 3) determine whether speech sound production skills at age two were predictive of speech sound production skills at age three.

Method—Seventy children with bilateral, mild-to-severe hearing loss who use hearing aids and 37 age- and SES-matched children with NH participated. Children's speech sound production abilities were assessed at 2 and 3 years of age.

Results—At age two, the HH group demonstrated vowel production abilities on par with their NH peers, but weaker consonant production abilities. Within the HH group, better outcomes were associated with hearing aid fittings by 6 months of age, hearing loss of less than 45 dB HL, stronger vocabulary scores, and being female. Positive relationships existed between children's speech sound production abilities at 2 and 3 years of age.

Conclusions—Assessment of early speech sound production abilities in combination with demographic, audiologic, and linguistic variables may be useful in identifying HH children who are at risk of delays in speech sound production.

Perceptual access to linguistic input is critical for the development of phonological representations that underlie spoken word production (Kuhl, 2000), which becomes evident in studies of young children with hearing loss (von Hapsburg & Davis, 2006; Warner-Czyz, Davis, & MacNeilage, 2010). Hearing loss (HL) can prevent children from experiencing consistent and complete access to words in the ambient language, which may slow their development of phonological representations and/or their production accuracy (Tomblin, Oleson, Ambrose, Walker, & Moeller, 2014; von Hapsburg & Davis, 2006). Provision of early and consistent access to linguistic input is critical for promoting optimal outcomes for

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Conflicts of Interest
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these children and thus has become a fundamental best-practice goal in the management of infants with HL (Bagatto et al., 2011; Sininger, Grimes, & Christensen, 2010). Two relatively recent service innovations support this goal: early identification through universal newborn hearing screening and provision of hearing aids (HAs) and/or cochlear implants at much earlier ages than in the past (Halpin, Smith, Widen, & Chertoff, 2010; Sininger et al., 2010).

The current study examines speech production abilities in children with mild-to-severe HL who use HAs. It is necessary to examine outcomes for this group separately from those of children with profound HL who utilize cochlear implants, given differences in their auditory experiences. Contemporary studies have documented substantial progress in speech production for children with early receipt of cochlear implants, showing that speech delays in these children relative to hearing peers are much less pronounced than for previous generations of children with profound HL (Ertmer & Goffman, 2011; Ertmer, Kloiber, Jung, Kirleis, & Bradford, 2012; Warner-Czyz et al., 2010). However, studies exploring the outcomes of children who are hard of hearing (HH) are relatively rare (Eisenberg, 2007; Fitzpatrick, Crawford, Ni, & Durieux-Smith, 2011; Holte et al., 2012; Tomblin et al., 2014; von Hapsburg & Davis, 2006). As a result, there is little evidence-based research regarding early speech outcomes of children who are HH and factors that contribute to minimizing speech delays. The current study strives to address this research gap.

Speech Sound Development of Young Children with Normal Hearing

One goal of early intervention with children who are HH is to promote typical speech sound development. With this goal in mind, it is important to review developmental milestones in speech production for children with normal hearing (NH). In typical development, learning to produce words is influenced by multiple factors, including perceptual, cognitive, linguistic (e.g., semantic), and motor demands (Stoel-Gammon, 1998; von Hapsburg & Davis, 2006). Children with NH usually undergo rapid development of their speech sound systems during the months just prior to their second birthdays (Preisser, Hodson, & Paden, 1988). Stoel-Gammon (1991) described the consonant repertoire of an average 2-year-old as including voiced and voiceless labial, alveolar, and velar stop consonants along with labial and alveolar nasals, glides, and a few fricatives (typically /f/ and /s/). Furthermore, some consonant clusters are produced in the word initial and word final positions (Stoel-Gammon, 2011). Longitudinal studies of spontaneous word productions in typically-developing 24-month-olds revealed consonant inventories including /b, t, d, k, g, m, n, h, w, f, s/ in the word-initial position and /p, t, k, n, r, s/ in the word-final position (Stoel-Gammon, 1985, 1987). Dyson (1988) reported that by 3 years of age, children had typically expanded their consonant inventories to include palatal consonants /j, ʃ/, voiced fricatives /v, z/, and liquids /l, r/. The greatest changes over the third year of life were in the repertoire of consonants used in the word-final position.

McIntosh and Dodd (2008) reported that the 25 to 29 month old children in their study were producing consonants with 64% accuracy and vowels with 88% accuracy. Accuracy was higher for children approaching their third birthday, with 73% accuracy for consonant production and 95% accuracy for vowel production. This led the authors to conclude that vowel errors are relatively rare in comparison to consonant errors for typically-developing children in these age groups. Consonant errors generally followed typical developmental phonological patterns, the most frequent of which were cluster reduction, final consonant deletion, stopping, fronting, gliding of /r/, and deaffrication. Children who demonstrated high rates of atypical errors were likely to be diagnosed with a speech sound disorder at age 3.

In each of these reports of speech sound development, the authors have emphasized the fact that there is tremendous individual variability in early stages of speech production. This individual variability is interesting, in part, because research indicates that children's progress in developing a consonantal inventory and expanding its use in words can serve as a predictor of children's later expressive language outcomes. For example, Watt, Wetherby, and Shumway (2006) found that the consonant inventories produced by 160 children with NH late in the 2nd year of life contributed uniquely to their expressive language outcomes at 3 years of age. Additionally, Stoel-Gammon (1991) reported that the diversity of syllable and consonant types in the prelinguistic period were related to speech and language outcomes at 5 years of age in a group of children with NH. Taken together, these results suggest that changes in the consonantal inventory, particularly late in the 2nd year of life, may be associated with later expressive language outcomes. Thus, benchmarks in speech sound development can be used to monitor early spoken language progress in children with NH (Eilers & Oller, 1994; Oller & Eilers, 1988; Stoel-Gammon, 1991, 2011; Watt et al., 2006).

Speech Sound Production Skills and Influential Factors in Children who are HH

Studies of speech sound benchmarks and consonant production accuracy are particularly limited in regard to early-identified children who are HH at 2 and 3 years of age (Eisenberg, 2007). There is a pressing need for research to determine whether contemporary populations of children who are HH approximate typical benchmarks in speech sound development and to determine whether early speech sound production abilities may serve as a gauge for how children who are HH progress with amplification and other auditory interventions. Although one might expect that early identification and early provision of HA technology would provide children who are HH with optimum access to the speech spectrum and the fullest possible access to linguistic input, in reality HAs often fall short of this goal, especially for children with greater degrees of HL (McCreery, Bentler, & Roush, 2013). For example, restrictions in HA bandwidth limit the audibility of consonants with high frequency energy, particularly for female and child talkers (Stelmachowicz, Pittman, Hoover, & Lewis, 2001). Additionally, sound quality may be distorted as a result of the physiological effects of sensorineural HL. Environmental factors such as noise, reverberation, and distance from the talker also contribute to variable audibility of the input. Given these issues, it is not surprising that several studies of infants and toddlers with HL suggest that these children are at risk for early speech sound production delays (McGowan, Nittrouer, & Chenausky, 2008; Moeller, Hoover, Putman, Arbataitis, Bohnenkamp, Peterson, Lewis, et al., 2007;

Yoshinaga-Itano & Sedey, 2000), even when identification is early and HL is mild (von Hapsburg & Davis, 2006).

In one study of the speech sound development of infants and toddlers with HL, Moeller and colleagues employed a longitudinal design to compare prelinguistic and early lexical stages of children with HL to children with NH (Moeller, Hoover, Putman, Arbataitis, Bohnenkamp, Peterson, Lewis, et al., 2007; Moeller, Hoover, Putman, Arbataitis, Bohnenkamp, Peterson, Wood, et al., 2007). Nine of 12 children followed in the study wore HAs; the remaining three had cochlear implants. Results indicated that, on average, children with HL who were otherwise typically-developing were comparable to children with NH on measures of vowel inventory size and accuracy of vowel production in words. In contrast, however, delays in consonant and syllable structure development were noted for the children with HL. Specifically, fricative and affricate production showed atypically protracted development, whereas other consonant manners developed later but in parallel to children with NH. These findings match up with those of perceptual studies indicating that children with HL, especially those who are HH as opposed to deaf, demonstrate better perception and production of vowels as compared to consonants (Markides, 1970; Sininger et al., 2010).

In another study, Yoshinaga-Itano & Sedey (2000) found that the strongest predictors of speech production outcomes for deaf and HH children in the age range of 12 to 60 months were age, expressive language skills, and the degree of HL. The same research team also conducted another study with 19 children with mild to profound HL in which they explored whether prelinguistic and early speech sound behaviors at 16 to 23 months of age were contributors to speech intelligibility at 36 months of age (Obenchain, Menn, & Yoshinaga-Itano, 2000), a question particularly relevant to the goals of the current study. Early variables that were correlated with later speech skills included degree of HL, vocabulary size, use of meaningful gestures with vocalizations, phonetic inventory size, volubility, and production of syllables that contained one or more consonants. These findings provide support for examining whether, as for children with NH, benchmarks in speech sound development may be useful for monitoring early spoken language progress in children who are HH and identifying children who are HH who are at risk for persisting speech delays.

In addition to degree of HL, factors related to HA fitting may also be associated with speech production outcomes. Recently, Tomblin et al. (2014) examined factors influencing speech sound production scores at 3 and 5 years of age in 180 children who are HH. Results showed that children who are HH had poorer speech outcomes than children with NH, on average, and that the largest differences were seen for children with pure tone averages (PTAs) ≥ 45 dB HL. Furthermore, it was reported that aided audibility (i.e., the degree of access to the speech spectrum provided by HAs) had a beneficial effect on speech development in children who are HH. These results supported the proposal that a measure of aided hearing, the Speech Intelligibility Index, might be a more sensitive measure than PTA regarding how children access speech input for use in language learning through their HAs (Stiles, McGregor, & Bentler, 2012).

Sininger et al. (2010) examined the protective effects of early fitting of HAs. They followed 44 infants and toddlers with mild-to-profound HL longitudinally. Although they examined degree of HL as a predictor of later speech outcomes, they did not examine aided audibility due to the strong relationship between audibility and degree of HL. Results showed that age at HA fitting and degree of HL were significant predictors of speech outcomes, which were measured beginning at age three in this group. In contrast, in a large, epidemiological study of children with HL, aged 5 to 11 years, Kennedy et al. (2006) did not find age at confirmation of HL to be predictive of longer-term speech outcomes. These different conclusions may relate to methodological differences between the studies (e.g., speech was assessed by parent report rather than direct assessment in Kennedy et al.), amount of variance in ages of identification/HA fitting, and in the ages at which the children were assessed. Further research is needed to examine how perceptual abilities and audiological histories (audibility, duration of HA experience) are related to early speech production for children who are HH.

Research Questions

The following questions were addressed:

1. How do the speech sound production skills of children who are HH compare to age- and socioeconomic status (SES)-matched children with NH at 2 years of age?

   It was predicted that children who are HH, on average, would be delayed relative to children with NH in consonant production, but not vowel production at age two.

2. What factors explain variability in speech sound production outcomes of children who are HH?

   It was predicted that children with longer periods of HA use (earlier fitting) by age two would outperform children with less HA experience. It was also predicted that children with more than 45 dB HL would be outperformed by their peers with less HL and that better aided audibility would be associated with better speech production outcomes.

3. Are speech sound production skills at age two predictive of speech sound production skills at age three?

   It was predicted that speech sound production at age two will be positively related to speech sound production skills at age three for children who are HH. If this is the case, it will have clinical implications related to the benefits of measuring speech sound production at age 2.

Methods

Participants

Participants were seventy 2-year-olds (39 male, 31 female) with bilateral, mild-to-severe HL (HH group) who were age- and SES-matched to 37 (21 male, 16 female) children with NH (NH group). An additional 17 HH children (20%) and two NH children (5%) were recruited but either could not be administered the primary speech elicitation task because they were unable or unwilling to imitate or they imitated an insufficient number of words on the task. Independent samples *t*-tests and a Chi-square test indicated that there were no significant differences on maternal education levels, age at HA fit, or gender between HH children who contributed speech production data at 2-years and those who did not (all p values > .20). However, on average, the HH children who contributed data had significantly better average hearing thresholds (*M* = 49.35 dB HL, *SD* = 12.66) than HH children who did not contribute data (*M* = 57.60 dB HL, *SD* = 15.04), *t* = 2.42, *p* = .018, *d* = 1.18.

All of the children were participants in a longitudinal, multisite study on the outcomes of children with mild-to-severe HL (Outcomes of Children with Hearing Loss; OCHL). Children were recruited by research teams at the University of Iowa, Boys Town National Research Hospital, and the University of North Carolina-Chapel Hill and resided in nine U.S. states (Alabama, Georgia, Iowa, Kansas, Minnesota, Missouri, North Carolina, Nebraska, and Virginia). Participation criteria for the children who are HH included 1) permanent bilateral HL (sensorineural, mixed, or permanent conductive) with a better-ear three- or four-frequency pure-tone average (BEPTA) between 25 dB HL and 75 dB HL[1], 2) no significant cognitive, visual, or motor impairments, 3) spoken English as the primary communication mode, and 4) at least one primary caregiver using spoken English in the home. The children with NH met the same criteria, but all were confirmed to have hearing thresholds at or better than 20 dB HL. Table 1 summarizes key demographic variables for the two groups, including maternal education, which was used to represent SES and was coded as a continuous variable representing years of education. With the exception of one child in the NH group, parents reported race and ethnicity for their children. Fifty-eight children in the HH group were white, six were black, two were Asian/Pacific Islander, two were multi-racial, and two parents selected “other.” For the NH group, parent reported indicated that 32 children were white, one was black, two were multi-racial, and one parent selected “other.” With regard to ethnicity, one child in the HH group was reported to be Hispanic.

Procedures

As part of the OCHL protocol, children over the age of two and their families participated in an initial baseline visit, followed by visits once a year for up to 4 consecutive years. The current study involved data collected at the 2- and 3-year visits, which were conducted as close as possible to the children’s second and third birthdays.

Hearing assessments—An audiologist with pediatric experience and a test assistant completed all hearing evaluations. Air-conduction thresholds were measured at 500, 1000, 2000, and 4000 Hz using visual reinforcement or conditioned play audiometry procedures. BEPTA was calculated for subsequent analyses. Ear-specific thresholds were obtained using insert earphones, circumaural headphones, or the child’s own earmolds coupled to insert earphones. Audiologists obtained soundfield thresholds if the child would not tolerate the testing with earphones or headphones. If a full audiogram could not be completed, the audiologist obtained a copy of the child’s most recent unaided audiogram from their personal audiologist. Audiological results indicated that there were 42 children with bilateral

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[1] Two children in the HH group had PTAs better than 25 dB HL because the HL was primarily in the high frequency range; one child initially met the study criterion, but subsequently had HL greater than 75 dB HL related to progression of the loss.

sensorineural HL, three with Auditory Neuropathy Spectrum Disorder, and five with permanent conductive HL. For the remaining 20 children, bone-conduction testing could not be completed during the study visit. Therefore, the type of HL could not be definitively categorized.

Hearing aid verification measures—Real-ear to coupler difference was measured whenever possible; if children would not cooperate for real-ear measures, age-appropriate average real-ear measures were used. In order to estimate the proportion of the amplified speech spectrum that was audible to children who are HH when wearing their HAs, Audioscan Verifit software was used to calculate unaided and aided audibility measures (Bentler, Hu, & Cole, 2011) based on the Speech Intelligibility Index (SII; ANSI, 1997). The SII measure is reported on a scale from 0 to 1, with 0 representing completely inaudible and 1 representing completely audible. The better-ear aided SII (BESII) was used in subsequent analyses in the current study. All but two children were fit binaurally with behind-the-ear air-conduction HAs. One child was fit with a bone-anchored HA and the other was fit with a soft-band bone-conduction device.

Parents provided information about the age at HA fitting, frequency of service provision, and maternal education level via interviews and questionnaires at the time of testing. When information regarding HA fitting could not be obtained from parents, research assistants collected it via medical/educational chart reviews. Table 2 summarizes key audiological and intervention variables for the children with HL. The majority of these children (94.1%) received early intervention services; children received an average of 3.61 sessions per month, and the majority of sessions were home-based. For all children receiving early intervention services, surveys were sent to their primary service providers to query information related to service provision. Surveys were returned by one or more service providers for 43 children. The majority of children (67%) were reported to be receiving early intervention services from more than one type of provider. Seventy-nine percent of the sample received services from a teacher of the deaf, 56% received services from a speech-language pathologist, 40% received services from an early intervention specialist, 19% received services from an early childhood speech educator, and 7% received services from an auditory-verbal therapist.

Speech and language procedures—Speech language pathologists and/or experienced and trained examiners completed speech and language assessments for all children.

Open and Closed Set Test: At the 2-year test interval, examiners administered the *Open and Closed Set Test* (*O&C*; Ertmer, Miller, & Quesenberry, 2004), a measure that uses early-emerging vocabulary as stimuli to examine speech perception, word comprehension, and speech sound production abilities in very young children with bilateral HL. This measure was chosen to serve as the elicitation task for speech sound production abilities because it can be quickly administered to children as young as 2-years of age. The *O&C* consists of three lists of ten words that are found in the spoken vocabularies of 75% of typically-developing 2-year-olds (Dale & Fenson, 1996). The *O&C* is intended to be administered at 6-month intervals to monitor within-child progress after fitting of HAs or cochlear implants. The lists are balanced for the number of syllables in the stimulus words and the presence of consonant clusters and later-emerging consonants. For the purposes of the current study, only one of the *O&C* lists was administered to participants. To administer the *O&C*, each stimulus word was presented in both an open-set and closed-set task format. First, the open-set task was presented. In this task, the parent (or examiner) named the stimulus word and the child was asked to repeat it. The examiner broadly transcribed the child's imitated productions online. Then, the closed-set word identification task was completed, wherein the child identified the target word from a closed set of three pictures; nontarget items were from the same semantic class as the target (e.g., "elephant" and "bear" served as distractors for the target word "cow"). Children's results were included in the analysis if they attempted to imitate at least five of the target words.

Twenty percent of the 107 samples were audio and video recorded for reliability analysis from only one site (Boys Town National Research Hospital). Children wore a vest that had been adapted to hold a wireless lavaliere microphone (Shure Model LX1-V), positioned on the chest to maintain a consistent microphone-to-mouth distance of approximately 2 inches.

Scoring procedures for the *O&C* included deriving three subtest scores: 1) phonological accuracy, 2) word acceptability, and 3) word identification. Phonological accuracy represented the percent of phonemes (including both vowels and consonants) the child produced correctly. Word acceptability represented the percent of words in which the child's production included at least two accurately produced phonemes (vowels or consonants) and the correct number of syllables. For these two measures, errors in consonant voicing were ignored, as per the *O&C* protocol. This is accepted practice in studies of children this young, because of children's limited control of the voicing feature (Macken & Barton, 1980). For word identification, 2 points were awarded if the picture was correctly identified following a single exposure and 1 point was awarded if the picture was identified after the target word was repeated. These scores were then converted to percent correct out of 20 possible points.

Transcriptions of all word productions were entered into Computerized Profiling Software (Long, Fey, & Channel, 2006) to facilitate further speech sound analyses through the Profile of Phonology (PROPH). Computer-based analyses were used to derive specific measures: 1) Percentage of Vowels Correct-Revised (PVC-R; Shriberg, Austin, Lewis, McSweeny, & Wilson, 1997), 2) Percentage of Consonants Correct-Revised (PCC-R; Shriberg, 1993; Shriberg et al., 1997), 3) consonant accuracy by developmental sound class, 4) consonant accuracy by place of articulation, and 5) frequency of phonological pattern use. Target productions are listed in Appendix A, along with additional information regarding the number of opportunities for observing the features of interest in this study. PVC-R is a measure of the intended vowels and diphthongs produced correctly with deletions and substitutions counted as incorrect, but clinical distortions counted as correct. The *O&C* word list provided children with 14 opportunities for vowel and diphthong production, including two unique rhotic vowels, three unique diphthongs, and six unique monophthongs. PCC-R is calculated in the same way as PVC-R, with all deletions and substitutions of consonants counted as incorrect, but clinical distortions counted as correct. The *O&C* word list samples 10 unique consonants, some of which are sampled multiple times, providing 22 opportunities for consonant production.

Consonant accuracy was also calculated for developmental sound class and place of articulation. Shriberg (1993) classified consonants into three developmental sound classes: Early-8 (/m, b, j, n, w, d, p, h/), Middle-8 (/t, ŋ, k, g, f, v, ʧ, dʒ/), and Late-8 (/ʃ, θ, s, z, ð, l, r, ʒ/). On the *O&C* word list, five of the sampled consonants were in the Early class (/m, b, n, d, p/), two were in the Middle class (/t, k/), and three were in the Late class (/ʃ, z, l/). For consonant accuracy by place of articulation, only those categories that were sufficiently sampled to allow for meaningful analysis were analyzed: bilabial, alveolar, and velar. Information regarding opportunities for production of consonants within each developmental sound class and place of articulation is contained in Appendix A.

PROPH was also used to identify occurrences of three phonological patterns (velar fronting, final consonant deletion, and cluster reduction) to determine the frequency with which these patterns occurred for each group. Scores were represented as the percentage of times these patterns were used when an opportunity existed. Velar fronting, final consonant deletion, and cluster reduction were singled out for analysis because they are common in young children's speech samples, and there were a sufficient number of possible opportunities in the *O&C* words to yield meaningful analyses (four, five, and three, respectively).

Vocabulary: Information regarding children's expressive vocabularies was collected by having parents complete the Words and Sentences version of the *MacArthur-Bates Communication Development Inventory* (MBCDI; Fenson et al., 2007). Scores for the Words Produced section were converted to percentiles, based on the normative data from the *MBCDI*. Table 1 includes results for 56 HH children and 30 NH children, because parents of 14 HH children and seven NH children did not return the forms. There were no significant differences on maternal education levels or any of the audiological variables between children who contributed *MBCDI* data and those who did not (all p values > .05).

Later speech outcomes: At 3 years of age, children's speech sound production skills were assessed via the *Goldman-Fristoe Test of Articulation-2* (*GFTA-2*; Goldman & Fristoe, 2000), which examines consonant production accuracy in single word productions. For a variety of reasons, data were unavailable for 14 HH children (progressive HL, child unable to do the task, attrition), and eight NH children (attrition). There were no significant differences on maternal education levels or any of the audiological variables between children who contributed data at both sessions and those who only contributed data at the 2-year visit (all p values > .05).

Transcription reliability: Transcription reliability was assessed through re-transcription of 20% of the *O&C* samples for both the children who are HH and children with NH. A trained listener, who was not involved with the original transcription and was blinded to the children's hearing status, independently transcribed samples from the video-audio recordings. For vowel reliability, intrajudge and interjudge agreement was calculated for correct vs. incorrect production. Interjudge reliability ranged from 71% to 100%, with an average of 90.8%. Intrajudge reliability ranged from 71% to 100%, with an average of 85.9%. For consonant transcription, point-to-point percentage agreement was assessed for consonant transcription. Interjudge reliability ranged from 77 to 100%, with an average of 86.2%. Intrajudge reliability also was calculated for consonant transcription by requiring two of the original testers to independently re-transcribe test words from the recordings. Intrajudge agreements ranged from 75 to 100%, with an average of 92.0%. Transcription from video was, at times, challenging due to the young ages of the children, whose movements sometimes obscured their faces. Thus, transcriptions based on the face-to-face interactions were used in the analysis.

Results

Group Differences

To answer the first research question, which asked whether children who are HH demonstrate speech sound production skills that are similar to children with NH, a series of independent sample *t*-tests was conducted, with the alpha value adjusted to .01 to correct for multiple comparisons. Statistical results for the *O&C* are shown in Table 3. Results revealed that scores for the children with NH were significantly higher than those of the children who are HH for all three subtests of this measure. Effect sizes (Cohen's *d*) were medium, based on Cohen's guidelines (1988). From this point forward the research questions were addressed using the transcriptions and PROPH analyses (PCC-R and PVC-R) rather than the clinical scores from the *O&C* measure, as the PROPH analyses allow for more detailed measures of speech sound production accuracy and are more directly comparable to findings from other studies.

PVC-R and PCC-R scores were derived for imitative productions of the *O&C* words from PROPH in order to further compare the groups on early speech sound production skills. In addition to total PCC-R, PCC-R was also calculated for developmental sound class (Early, Middle, Late) and place of articulation (bilabial, alveolar, and velar). Results are summarized in Table 3. Results indicated that there was no difference between the groups on PVC-R (p = .156). Differences were observed for consonant production: the children with NH outperformed the children who are HH on total PCC-R (p = .001). Overall, results for the analysis of PCC-R by developmental sound class indicated that both groups showed a predictable pattern related to developmental sound class, with stronger performance, on average, on consonants in the Early class compared to consonants in the Middle and Late classes. However, the children with NH scored at significantly higher levels on Early and Middle class consonants than children who are HH (p = .009 and $p < .001$, respectively). The Late class of consonants was challenging for both groups (p = .057), as expected at this young age. In general, the results suggest a delayed but parallel pattern of consonant production for the children who are HH compared to the NH group. Effect sizes were moderate to large (see Table 3).

In the next analysis, PCC-R scores were compared for accuracy of consonants produced at the bilabial, alveolar, and velar places of articulation. Results showed that the groups did not differ significantly on bilabial consonants (p = .026). However, the children who are HH were significantly less accurate than children with NH on both alveolar (p = .006) and velar consonants (p = .001), and medium effects were observed (see Table 3). In the final analysis of consonant accuracy, use of three phonological error patterns was examined. Results indicated that the children with NH and the children who are HH demonstrated evidence of velar fronting (14% and 20% of opportunities, respectively) and cluster reduction (49% and

54% of opportunities, respectively) to similar degrees (p values > .01). However, the children who are HH were significantly more likely to delete final consonants than the children with NH ($M_{NH} = 10.27\%$, $SD_{NH} = 22.42$; $M_{HH} = 34.98\%$, $SD_{HH} = 31.94$, $t = -4.66$, $p < .001$, $d = 0.89$).

Factors Influencing Speech Sound Production Accuracy

The next research question explored which factors contributed to variability in the speech sound production outcomes of children who are HH. Given the strong correlation between PVC-R and PCC-R for this group ($r = .629$, $p < .001$), and the lack of theoretical arguments indicating that these two variables would be affected differently by audiologic or demographic variables, PCC-R served as the dependent variable in analyses for this question. As a first step toward addressing this question, a two-way ANOVA that examined the effect of age at HA fitting and degree of HL on PCC-R was conducted. Participants were divided into groups according to age at HA fitting (no HA [i.e., children with NH], ≤ 6 months, > 6 months) and degree of HL (< 20 dB HL [i.e., children with NH], 20–45 dB HL, > 45 dB HL). There was a significant overall effect of age at HA fitting on PCC-R scores, $F(1, 106) = 8.08$, $p = .005$, $\eta_p^2 = .073$. A follow-up Tukey test showed that children who were fitted with HAs by 6 months were not different from the NH group on average ($p = .139$). However, the children fit after 6 months were significantly different than the NH group ($p < .001$) as well as the group fit by 6 months ($p = .005$), with the group fit after 6 months having lower scores, on average. There was also a significant overall effect of degree of HL on PCC-R scores, $F(1, 106) = 5.24$, $p = .024$, $\eta_p^2 = .049$. A follow-up Tukey test revealed that children with 20–45 dB HL were not significantly different from the NH group ($p = .113$). However, the children with > 45 dB HL were significantly different than both the NH group ($p < .001$) and the 20–45 dB HL group ($p = .024$), with the > 45 dB HL group performing worse than either of the other two groups. No interaction was observed ($p = .518$), suggesting that age at HA fitting had similar effects at each level of HL. These effects are shown in Figure 1 for PCC-R.

Multiple linear regression was then utilized to further explore which factors explained variance in PCC-R. Only children who are HH with data for the variables maternal education, BESII, *MBCDI*, and PCC-R ($n = 54$) were included in these analyses. Because BEPTA and BESII are strongly correlated ($r = -0.80$, $p < .001$), only BESII was entered in the model, given that this measure reflects the child's aided audibility and may be more sensitive than BEPTA to how children access speech input through their HAs. Furthermore, neither age of identification of HL or age at HA fitting were entered in the regression because limited variability and skewed distributions make these variables hard to analyze as continuous predictors in the regression model. In the resulting analysis, there was no evidence of multiple linear regression assumptions being violated and no evidence of multicollinearity. With maternal education, sex, BESII, and *MBCDI* included in the model, there were two significant predictors of PCC-R: sex ($\beta = .231$, $t = 2.09$, $p = .042$) and *MBCDI* ($\beta = .597$, $t = 5.36$, $p < .001$). BESII and maternal education were not significant in the model (both $p > .10$). This model accounted for 45.2% of the variance in PCC-R, which was significant ($p < .001$).

Relationships with Speech Sound Production Skills at Age 3

The final objective was to determine if speech scores for the children who are HH at age two predicted their speech sound production scores at age three. Speech sound production abilities at age 3 were represented by children's standard scores on the *GFTA*. Standard scores for both groups are displayed in Table 1, with results of an independent samples *t*-test indicating that the average standard score for the HH group was significantly lower than that of the NH group. Thirty nine percent (22/56) of the children who are HH obtained a standard score at or below 85 on the *GFTA*; only 1 child with NH scored at this level. Pearson correlation coefficients were calculated to explore the relationships of PVC-R and PCC-R at age two with *GFTA* at age 3. PVC-R and *GFTA* scores were moderately correlated for both groups (HH: $r = 0.497$, $p < .001$; NH: $r = 0.443$, $p = .016$). Relationships were stronger between PCC-R and *GFTA* scores, with large correlations for both groups (HH: $r = 0.730$, $p < .001$; NH: $r = 0.542$, $p = .002$). The correlations of PVC-R and PCC-R at age 2 with *GFTA* standard scores at age 3 are illustrated in Figure 2 for the HH group.

Given that PCC-R was more strongly correlated with *GFTA* scores than was PVC-R, PCC-R was used in the remaining analyses for this question. First, to further understand contributory factors, a multiple linear regression analysis was conducted, which allowed for controlling the variables sex, *MBCDI*, maternal education, and BESII. Using only the children who are HH and only those with data for sex, *MBCDI*, maternal education, BESII, PCC-R, and *GFTA*, the relevance of using PCC-R scores to predict *GFTA* scores (measured at age three) was assessed. There was no evidence of multiple linear assumptions being violated. Calculation of variance inflation factors and condition indices indicated that multicollinearity was not a problem (all VIFs < 2.5). Interestingly, PCC-R was a significant predictor of *GFTA*, even after controlling for sex, maternal education, *MBCDI*, and BESII. With all the predictors on the model, only PCC-R contributed significant unique variance to *GFTA* scores ($\beta = .503$, $t = 3.25$, $p = .002$). Fifty eight percent of the variance in *GFTA* was explained by the predictor variables in this regression model.

To assess the predictive ability of PCC-R for determining whether children who are HH would have delayed speech sound production abilities at age 3, PCC-R was entered into a logistic regression, with *GFTA* category (Low [< 85], Average [> 85]) as the dependent variable for the HH group only ($n = 22$ and 44, respectively). PCC-R was a significant contributor ($\beta = -.084$, SE = .023, $p < .001$, Log Likelihood Ratio = 53.23) and the model was statistically significant ($\chi^2 = 21.81$, $p < .001$). The predictive accuracy of the model can be assessed from the area under the receiver operator curve (ROC). The area under the curve ranges from 0.5 for a model representing random classification to 1.0 for a perfect model. The area under the curve in this model was 0.85 which means that if we randomly select one individual from the Low group and one individual from the Average group, then 85% of the time the person from the Low group will have a lower PCC-R score (i.e., higher predicted probability of being in the Low group). We used the ROC curve and Youden index (Youden, 1950) to determine the optimal cutoff points for sensitivity and specificity. Sensitivity for this model was 81.8 (Confidence Interval = [61, 93]) and specificity was 79.4 (Confidence Interval = [63, 90]) corresponding to a 40% probability rule. The model predicted that children with PCC-R scores below 57.4 would be in the Low group and children with PCC-R scores at or above 57.4 would be in the Average group. The odds ratio was 1.09 (95% Confidence Interval = [1.04, 1.14]), thus with every one percent increase in PCC-R, the odds of being in the Normal group are 9% higher than the odds of being in the Low group.

Discussion

The first goal of this study was to determine if the presence of mild-to-severe HL places children at risk for delays in the development of speech sound production skills. A positive finding was that the HH group performed similarly to the children with NH at age two on the measure of vowel production accuracy. However, consistent with the previously outlined predictions, the children who are HH were significantly less accurate, on average, than age- and SES-matched children with NH in imitative production of consonants in early-developing words. Consonant production may have been more sensitive to group differences than vowel production because vowels were generally produced with greater accuracy than consonants by the HH children in this study. This fits with the findings of previous work indicating that children with NH master vowel production earlier in development than consonant production and that children with HL more accurately perceive and produce vowels as compared to consonants (Ertmer & Goffman, 2011; Markides, 1970; McIntosh & Dodd, 2008; Moeller, Hoover, Putman, Arbataitis, Bohnenkamp, Peterson, Lewis, et al., 2007; Sininger et al., 2010; Warner-Czyz et al., 2010). This finding also makes sense in light of the fact that vowels are more sonorous, and thus perceived as louder, than consonants. Overall, the results underscore the need to understand factors that contribute to individual differences so that children at most risk may be identified early.

Consonant Production: Developmental Profiles

The children who are HH were less accurate in consonant production than the children with NH; however, they followed a typical developmental pattern. In general, these findings support earlier claims of delayed but parallel development of consonant production in children with HL compared to children with NH (Moeller, Hoover, Putman, Arbataitis, Bohnenkamp, Peterson, Lewis, et al., 2007). However, this earlier work also revealed that a small group of children with HL demonstrated especially protracted development of fricative production compared to age-matched children with NH. It was not possible to address this question in the current study because the fricative class was not comprehensively sampled (2 types, 3 tokens) on the *O&*C word list.

Accuracy was also measured by place of articulation for bilabial, alveolar, and velar consonants. Bilabial accuracy did not differ by hearing status, which may be, in part, a result of ceiling effects for this place of articulation. Although no between-group differences were observed for bilabials, the children with NH did outperform the children who are HH on production of alveolar and velar consonants. The differences in accuracy for bilabial versus alveolar and velar sounds may be related to the more limited visual cues that accompany the latter two places, making them more challenging than visually-salient bilabials for children who are HH (Stoel-Gammon, 1988; von Hapsburg & Davis, 2006). However, alternatively, differences may be attributed to sampling features of the *O&C* word list. As seen in

Appendix A, bilabials were not sampled in the postvocalic position, but 40% of alveolar consonants and 20% of velar consonants were sampled in the postvocalic position. Thus, it is possible that findings for alveolar and velar production were affected by the tendency of children who are HH to delete final consonants.

Final consonant deletion is a common phonological pattern in typically-developing 2-year-olds (Stoel-Gammon, 1991; Vihman, 1996). However, children who are HH in the current study were three times more likely than children with NH to delete final consonants when imitating the test words. It is possible that this finding reflects immature syllable structure development in this group of children. Although consonant-vowel-consonant syllable shapes are commonly produced by 2-year-olds (Stoel-Gammon, 2011), children with NH and delayed speech display a greater proportion of open syllables (vowel only or consonant-vowel) than typical age-mates (Paul & Jennings, 1992; Rescorla & Ratner, 1996). Thus, the results of the current study may indicate that the children who are HH simply resemble younger children with typical development. However, given the much higher occurrence of final consonant deletion in children who are HH, the potential role of audibility should be considered. It is possible that saliency of final consonants in conversational speech is reduced in noise, during periods without HA use, or as a result of limited HA bandwidth (Stelmachowicz, Pittman, Hoover, Lewis, & Moeller, 2004). Perceptual limitations such as these could influence the development of syllable closure. This hypothesis gains some support from the relative performances of the groups across the three phonological patterns examined. Children who are HH were no more likely than children with NH to exhibit velar fronting or cluster reduction, but were far more likely to exhibit final consonant deletion. Thus, it appears that syllable closure was relatively more challenging for the children who are HH than for the children with NH.

Factors Contributing to Speech Sound Production Outcomes

A second goal of the study was to identify factors that explain variability in speech sound production accuracy in the children who are HH. PCC-R was utilized in analyses for this research question. It is clear from Figure 1 that early-identified children with hearing losses of 20–45 dB HL were most likely to achieve typical performance. On the other hand, performance decrements were observed for children fitted with amplification later than six months of age in either of the HL categories, suggesting that even children with mild HL benefitted from earlier access to amplification. The results suggest that auditory experience with amplification plays a role in consonant development for children who are HH, thus supporting the practice of providing early access to amplification for young children who are HH.

In contrast to the previously outlined predictions, a unique contribution of BESII to PCC-R at two years of age was not found. There are several possible reasons that this relationship was not observed, including the non-normal distribution of the BESII scores. Another possibility is that the effects of aided audibility may not have been apparent at this early age, given the relatively limited experience children had with their HAs (average of 18 months). This latter possibility is supported by Tomblin et al.’s (2014) finding that the effects of audibility increase as children’s duration of HA use increases. In addition, although it has been suggested that the aided SII may be a better predictor of outcomes than BEPTA, limitations of the SII measure have been described. The SII is an idealized representation of audibility/access and reflects only performance in quiet settings. McCreery and Stelmachowicz (2011) found that the SII tends to overestimate speech understanding in children, particularly in noise (i.e. real world listening environments). Further research is needed to explore the ways in which aided audibility may interact with other variables (e.g., consistency and duration of HA use) to influence developmental outcomes. These questions are being pursued by the OCHL team.

Two non-audiological factors were found to contribute significant variance in PCC-R scores: sex and vocabulary scores. Among the children who are HH, girls were more likely to have higher PCC-R scores at 2 years of age. Although gender accounts for only small amounts (1–2%) of variance in early language development (Fenson et al., 1994), a meta-analysis showed that 10–15% of variance in speech production was accounted for by gender (Hyde & Linn, 1988). The finding that larger vocabularies were associated with stronger PCC-R scores finds support in the literature on children with HL. Obenchain et al. (2000) found that a larger lexical inventory in the second half of the 2nd year of life was a predictor of better speech outcomes at 36 months of age in children with HL. Studies of typically-developing children suggest strong bidirectional effects of phonology and lexical development in early stages. Stoel-Gammon (1998) originally proposed that at the onset of meaningful speech, the speech sound production abilities of the child play a key role in determining which words are likely to enter the lexicon. It was further documented experimentally that children are more likely to attempt to say words that contain consonants already within their speech sound inventories (Schwartz & Leonard, 1982). However, as children's vocabulary grows, the lexicon in turn prompts speech sound growth, as the child stretches to attempt to say new words (Stoel-Gammon, 2011). In the current study, the strong association between lexical development and PCC-R is logical based on these bidirectional effects of phonology and lexicon at this stage of development.

Speech Sound Production at Ages Two and Three

Longitudinal methods supported the exploration of the third question: are speech sound production scores at age two predictive of speech sound production abilities at age three? Both PVC-R and PCC-R scores at age two were positively correlated with *GFTA* scores at age three for the 56 children who are HH providing scores at both ages. This suggests that children who demonstrated the strongest speech sound production abilities at age two were likely to continue to demonstrate relatively strong speech production abilities at age three. PCC-R scores were more strongly associated with *GFTA* scores than were PVC-R scores, which may be partially attributable to the fact that *GFTA* scores only represent children's consonant production abilities. PCC-R explained unique and significant variance in *GFTA* scores, after controlling for other primary variables. *MBCDI* scores did not explain unique variance in *GFTA* scores that was not already explained by PCC-R. This does not suggest that *MBCDI* is unimportant, but rather that it shares variance with PCC-R.

It is concerning that 39% of the children who are HH demonstrated scores in the below average range on the *GFTA* at age 3, given that normative data would only predict that 16%

of children who are typically-developing would achieve scores at this level. Indeed, only 7% of the NH group in this study demonstrated such low scores, thus indicating that the children who are HH continue to demonstrate delays in speech sound development at 3 years of age. PCC-R scores at age two were 82% accurate in identifying children who were below average on the *GFTA* at age three, with each one percent increase in PCC-R resulting in a 9% increase in odds that a child would fall into the average range on the *GFTA* at age three. These findings imply that the PCC-R score based on imitation of *O&C* words holds promise as an index that can identify early risk for phonological delays in children who are HH. Of the scores that were predicted incorrectly, the measure was more likely to over-identify risk rather than miss potential risk. False positives predictions occurred at a rate of 25%; seven children had low scores at age two, but performed within the average range by 3 years of age. This is a lower percentage of spontaneous resolution of delay than observed in research on late talkers, where nearly 50% of early delays were observed to resolve (Rescorla, Roberts, & Dahlsgaard, 1997; Rescorla & Schwartz, 1990; Weismer, Murray-Branch, & Miller, 1994). It calls into question a practice of “waiting to see” if children who are HH will be “late bloomers.” A more conservative approach is to support the child’s phonological development proactively if risks are identified at age two. In the current study, false negative predictions occurred at a rate of 13%; relatively few children performing well at age two showed delays at age three.

The findings of this study indicate that administration of an imitative speech sound production task with as few as ten words may be useful in identifying 2-year-old children who are at risk for delays in speech sound production skills. The skills should be considered in conjunction with children’s development in related skill areas, given that early spoken word development is influenced by a variety of factors including children’s perceptual, cognitive, linguistic (e.g., semantic), and motor abilities (Stoel-Gammon, 1998; von Hapsburg & Davis, 2006). Indeed, in this study, children’s vocabulary abilities contributed unique variance to their early speech sound production scores.

Limitations and Future Directions

Several limitations need to be kept in mind when interpreting these results. First, 20% of the children who are HH and 5% of the children with NH either could not be administered the task or they did not contribute data because they imitated an insufficient number of words on the task. Additionally, children who were unable to contribute data were likely to have poorer BEPTAs than children who contributed data. Thus, the results presented in this study may be an overestimation of HH children’s speech production abilities.

Additionally, there were a number of limitations of the elicitation task utilized for speech sound production at 2-years in this study. First, the task required imitative productions, rather than spontaneous word attempts. Reliance on imitation could overestimate the stability of a child’s consonant production. Although imitation would not be expected to fully represent spontaneous speech or consonant production stability, this paradigm has been used by others to measure perception and production outcomes of young children with HL (Boothroyd, Eisenberg, & Martinez, 2010; Ertmer & Goffman, 2011). Another concern is that the elicitation task only contained ten words and these words over-represented earlyappearing phonological forms and had limited sampling of certain phonemes in various positions of words and limited opportunities for production of some error patterns. The limited word list also prevented analysis of syllable and word complexity in children's productions. Despite these limitations, however, the average PCC-R score of 73.8% in the current study for children with NH was quite similar to previous reports that utilized spontaneous speech samples for 2-year-olds and reported PCC-R scores around 70% (Paul & Jennings, 1992; Stoel-Gammon, 1987; Watson & Scukanec, 1997). Additionally, even though the task was not a comprehensive look at early speech production skills, it is striking that consonant production on the imitative measure was sensitive to between group differences and held up fairly well as a predictor of *GFTA* outcomes at age three. Nonetheless, future research should utilize a more comprehensive elicitation task that allows for assessment of syllable and word complexity and that samples a wider variety of speech sounds in the full array of word positions. Specifically, given the prediction that HA bandwidth limitations may reduce access to fricatives and affricates (Stelmachowicz et al., 2001), future investigations should ensure that these forms are better sampled, particularly /s/ and /f/, which are observed in the inventories of typically-developing 2-year-olds (Stoel-Gammon, 2011). This work should also explore the impact of perceptual limitations on syllable structure development and consonant development order and accuracy in children who are HH, including those with milder degrees of loss (von Hapsburg & Davis, 2006).

Future work should also examine additional factors that may have an impact on speech sound development, including variables related to children's intervention services. Although examination of intervention services was beyond the scope of the current manuscript, this work is currently being conducted with the OCHL cohort and will be reported in future manuscripts. Future work from the OCHL project will further examine speech sound production outcomes, including dimensions of phonology beyond consonant production, such as speech intelligibility.

Summary

This study examined the speech sound production skills of children who are HH as compared to those of children with NH. Results indicated that the children who are HH generally demonstrated delayed but parallel development of consonant production skills as compared to children with NH. No differences were identified between groups for vowel production accuracy. Among the children who are HH, those who received their HA by 6 months of age and/or had better pure tone thresholds tended to demonstrate better speech sound production accuracy than children who had later HA fittings or poorer hearing. Better speech production skills were also associated with being female and having stronger vocabulary scores. Speech sound production abilities at age two were positively correlated with speech sound production abilities at age three. It is concerning that most of the HH children identified with delays at age two did not resolve them by age three. These children may benefit from additional focus on listening and speaking within their early intervention programs.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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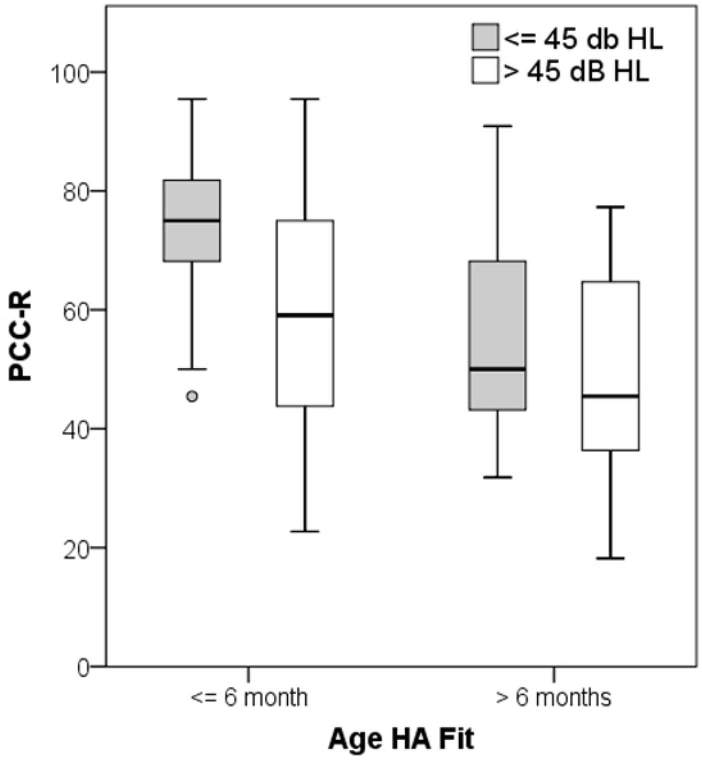

Figure 1.

Boxplots displaying medians and quartiles for PCC-R scores for children who are hard of hearing with BEPTAs ≤ 45 dB HL or > 45 dB HL, plotted as a function of category of age at hearing aid fitting (≤ 6 months, > 6 months).

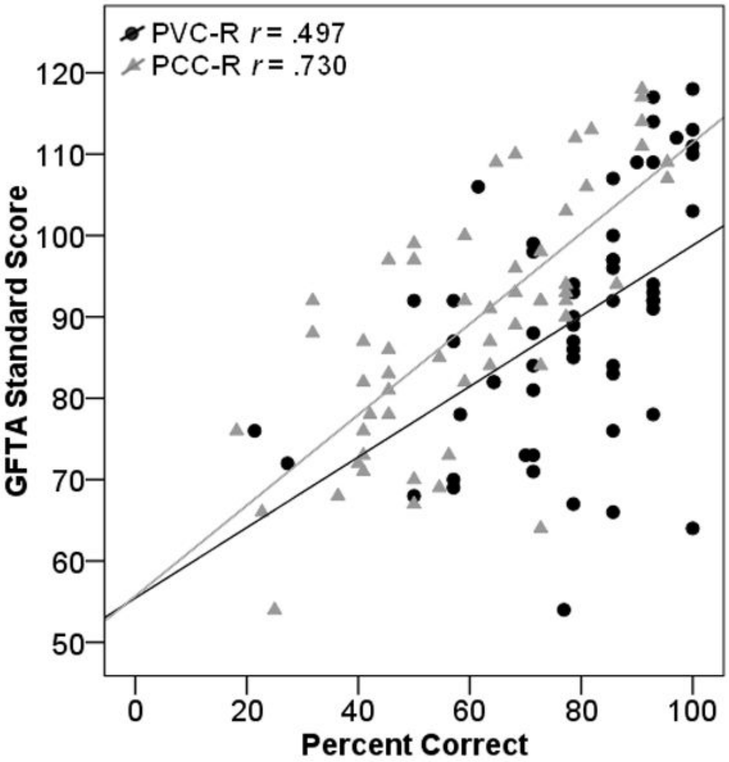

Figure 2.

Scatterplot of the relationships of Percent Vowels Correct – Revised (PVC-R) and Percent Consonants Correct – Revised (PCC-R) scores at age two and the *Goldman-Fristoe Test of Articulation-2* standard scores at age three for individual HH children.

Table 1

Demographic and outcome measures for the children with NH and children who are HH groups.

	Normal Hearing		Hard of Hearing		Comparisons Between Groups		
Measure	***n***	***M* (*SD*)**	***n***	***M* (*SD*)**	***t***	***p***	***d***
Demographics							
Maternal education (years)	37	16.08 (2.86)	70	15.13 (2.17)	1.93	.057	0.37
Age at 2-year test (months)	37	26.32 (2.81)	70	25.57 (2.41)	1.38	.171	0.29
Age at 3-year test (months)	29	36.48 (1.57)	56	36.50 (1.73)	−0.45	.964	−0.01
Outcome measures							
MBCDI-WS: WP (%ile)	30	53.13 (27.69)	56	37.13 (25.50)	2.69	.009	0.60
GFTA-2 (standard score)	29	104.31 (10.28)	56	89.36 (15.10)	4.78	<.001	1.16

Note. MBCDI-WS: WP = Words Produced section of the Words and Sentences version of *MacArthur-Bates Communicative Development Inventory* (administered at age two); *GFTA*-2 = Goldman-Fristoe Test of Articulation-2 (administered at age 3); *d* = Cohen's *d*, a measure of effect size.

Table 2

Audiological and intervention variables for children with hearing loss.

Measure	*n*	*M*	*SD*	range
Age at hearing aid fit (months)	70	6.89	4.98	2–22
BEPTA (db HL)	70	49.35	12.66	16 – 83
BESII	67	0.73	0.14	0.28–0.96
Service quantity (visits/month)	66	3.61	2.89	0–12

Note. BEPTA = Better-Ear Pure Tone Average; BESII = Better-Ear Aided Speech Intelligibility Index.

Table 3

Open and & Closed Set

test scores for the three subtests, Percent Vowels Correct –Revised scores, and Percent Consonant Correct - Revised scores (total, by developmental sound class, and by place of articulation) for the 37 children with children with NH and the 70 children who are HH.

	Normal Hearing	**Hard of Hearing**	**Comparisons Between Groups**	
Measure	***M* (*SD*)**	***M* (*SD*)**	***t***	***d***
O&C Subtest Scores				
Phonological accuracy	78.98 (15.96)	68.02 (17.06)	3.23*	0.66
Word acceptability	87.60 (14.37)	77.04 (22.90)	2.92*	0.55
Word identification	85.81 (14.65)	70.57 (29.93)	3.53*	0.65
PVC-R	82.12 (14.57)	77.26 (17.74)	1.43	0.30
PCC-R Total	73.79 (19.63)	60.01 (19.99)	3.41*	0.70
PCC-R for Developmental Class				
Early	88.91 (16.65)	79.35 (18.36)	2.64*	0.54
Middle	71.62 (33.62)	45.40 (34.52)	3.77*	0.77
Late	48.20 (28.54)	36.71 (29.74)	1.93	0.39
PCC-R for Place of Articulation				
Bilablial	92.92 (13.96)	84.19 (21.26)	2.25	0.49
Alveolar	74.41 (26.37)	58.98 (27.66)	2.79*	0.57
Velar	67.12 (36.54)	42.37 (36.75)	3.32*	0.68

Note. O&C = Open and Closed Set Test; PVC-R = Percent Vowels Correct – Revised; PCC-R = Percent Consonants Correct - Revised.

* $p < .01$