1   Characterizing North Carolina's Deaf/Hard-of-Hearing Infants and Toddlers: Predictors of

2   Vocabulary, Diagnosis, and Intervention

3   Erin Campbell[1] & Elika Bergelson[1]

4   [1] Duke University

Characterizing North Carolina's Deaf/Hard-of-Hearing Infants and Toddlers: Predictors of
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Abstract

Purpose: The study sought to 1) characterize the demographic, audiological, and
intervention variability in the population of DHH children receiving state services for hearing
loss; 2) identify predictors of vocabulary delays; and 3) evaluate the success of early
identification and intervention efforts at a state level.

Method: One hundred Deaf/Hard-of-Hearing infants and toddlers (aged 4-36 months)
enrolled in early intervention completed the MacArthur-Bates Communicative Development
Inventory, and detailed information about their audiological and clinical history was
collected. We examined the influence of demographic, clinical, and audiological factors on
vocabulary outcomes and early intervention efforts.

Results: We found this sample showed spoken language vocabulary delays
(comprehension and production) relative to hearing peers, and room for improvement in rates
of early diagnosis and intervention. These delays in vocabulary and early support services
were predicted by an overlapping subset of hearing-, health-, and home-related variables.

Conclusions: In a diverse sample of Deaf/Hard-of-Hearing children receiving early
intervention, the variables which predict delays in vocabulary and early support services
reflected *both* dimensions that are immutable, and those that clinicians and caretakers can
potentially alter. We provide a discussion on the implications for clinical practice.

Introduction

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[26] In the United States, 1-2 children are born with hearing loss, per 1,000 births (CDC,

[27] 2018). This translates to 114,000 Deaf or Hard of Hearing (DHH) children born in the U.S.

[28] per year (Martin, Hamilton, Osterman, & Driscoll, 2019). Of these 114,000, ~90% will be

[29] born to hearing parents (Mitchell & Karchmer, 2004), in a home where spoken language is

[30] likely the dominant communication method. Depending on the type and degree of hearing

[31] loss and whether the child uses amplification, spoken linguistic input will be partially or

[32] totally inaccessible. Some of these children will develop spoken language proficiency within

[33] the range of their hearing peers (Geers, Mitchell, Warner-Czyz, Wang, & Eisenberg, 2017;

[34] Verhaert, Willems, Van Kerschaver, & Desloovere, 2008), but many will face persistent

[35] spoken language deficits (Eisenberg, 2007; Luckner & Cooke, 2010; Moeller, Tomblin,

[36] Yoshinaga-Itano, Connor, & Jerger, 2007; Sarchet et al., 2014), which may later affect

[37] reading ability (Kyle & Harris, 2010) and academic achievement (Karchmer & Mitchell,

[38] 2003; Qi & Mitchell, 2012).

[39] Despite many excellent studies examining language development in DHH children,

[40] there is still a gap in the literature describing and analyzing spoken language development

[41] across the full range of children receiving services for hearing loss, with many studies

[42] focusing in on specific subgroups (e.g. children under age X with Y level of hearing loss and

[43] Z amplification approach, e.g., Vohr et al., 2008; Yoshinaga-Itano, Sedey, Wiggin, & Mason,

[44] 2018). In what follows, we first summarize the previous literature on predictors of spoken

[45] language outcomes in DHH children. We then provide a brief overview of a common

[46] vocabulary measure used in the current study, the MacArthur-Bates Communicative

[47] Development Inventory (CDI). Finally, we turn to an empirical analysis of early vocabulary

[48] in a wide range of young children receiving state services in North Carolina. We have two

[49] broad goals. First, we aim to provide a comprehensive description of a heterogeneous group

[50] of young children who receive state services for hearing loss. Second, we aim to connect the

intervention approaches and child characteristics of this sample with children's spoken

vocabulary[1], with the broader goal of considering the success of early diagnosis and

intervention initiatives.

Predictors of Language Outcomes

Though the literature points towards spoken language delays and deficits for DHH

children, this is a highly variable population with highly variable outcomes (Pisoni,

Kronenberger, Harris, & Moberly, 2018). Previous research indicates that gender (Ching et

al., 2013; Kiese-Himmel & Ohlwein, 2002), additional disability (Ching et al., 2013; Verhaert

et al., 2008; Yoshinaga-Itano, Sedey, Wiggin, & Chung, 2017), degree and configuration of

hearing loss (Ching et al., 2013; de Diego-Lázaro, Restrepo, Sedey, & Yoshinaga-Itano, 2018;

Vohr et al., 2011; Yoshinaga-Itano et al., 2017), amplification (Walker et al., 2015),

communication (Geers et al., 2017), and early diagnosis/intervention (Yoshinaga-Itano et al.,

2017, 2018) predict language outcomes in DHH children. We first provide a brief literature

review on the effect of these predictors on language skills in DHH children.

Gender. For hearing children, the literature points to a female gender advantage in

early language acquisition. Girls speak their first word earlier (Macoby, 1966), have a larger

(Bornstein, Hahn, & Haynes, 2004; Fenson et al., 1994; Frank, Braginsky, Yurovsky, &

Marchman, 2017) and faster-growing vocabulary (Huttenlocher, Haight, Bryk, Seltzer, &

Lyons, 1991), and stronger grammatical and phonological skills (Lange, Euler, & Zaretsky,

2016; Özçalışkan & Goldin-Meadow, 2010). This finding appears to be consistent across

studies (Wallentin, 2009), various spoken languages (Frank, Braginsky, Marchman, &

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[1] Despite exciting, increasing, and converging evidence for benefits of early sign language exposure (e.g.,
Clark et al., 2016, Davidson et al., 2014; Hrastinski & Wilbur, 2016; Magnuson, 2000; Schick et al., 2007;
Spencer, 1993), the majority of DHH children will not be raised in a sign language environment. This is
particularly true for North Carolina, which does not have a large community of sign language users, relative
to states like Maryland or areas like Washington D.C. or Rochester, NY. For this reason, and because no
families in our sample used a full-fledged signed language, we focus on spoken language development.

72 Yurovsky, 2019), and gesture (Özçalışkan & Goldin-Meadow, 2010).

73 The DHH literature presents a more mixed (though rather understudied) picture. On

74 one hand, DHH girls, like hearing girls, have been found to have a larger spoken vocabulary

75 than DHH boys (Ching et al., 2013; Kiese-Himmel & Ohlwein, 2002). However, in contrast

76 to their hearing peers, DHH children do not seem to show a gender-based difference for some

77 aspects of syntactic development (Pahlavannezhad & Tayarani Niknezhad, 2014).

78 **Comorbidities.** Additional co-morbid disabilities occur frequently in the DHH

79 population, perhaps as much as three times more than in the hearing population (Pollack,

80 1997). Incidence estimates for co-occurring disabilities in DHH children range from 25-51%

81 (Bruce & Borders, 2015; Guardino, 2008; Holden-Pitt & Diaz, 1998; Luckner & Carter, 2001;

82 Picard, 2004; Schildroth & Hotto, 1996; Soukup & Feinstein, 2007), with approximately 8%

83 of DHH children living with 2 or more co-occurring disabilities (Schildroth & Hotto, 1996).

84 Some of these conditions, particularly those which carry risk of developmental delay

85 (e.g., Down syndrome), result in language delays independent of hearing loss (Chapman,

86 1997; Kristoffersen, 2008; Weismer, Lord, & Esler, 2010). These effects vary by the nature of

87 the specific disability (Cupples et al., 2014, 2018), with cognitive ability more predictive of

88 language outcomes than presence or absence of additional disability (Meinzen-Derr, Wiley,

89 Grether, & Choo, 2011; Sarant, Holt, Dowell, Richards, & Blamey, 2008). Disability and

90 hearing loss likely each contribute to a given child's spoken language development (Ching et

91 al., 2013; Rajput, Brown, & Bamiou, 2003; Van Nierop et al., 2016), with differential effects

92 of each (Vesseur et al., 2016). In some cases, additional disabilities appear to interact with

93 hearing loss to intensify developmental delays (Birman, Elliott, & Gibson, 2012; Pierson et

94 al., 2007).

95 Furthermore, incidence of hearing loss is higher among children born premature

96 (defined as < 37 weeks gestational age). Compared to an incidence of 0.2% in full-term

97 infants, incidence of hearing loss in extremely premature infants (defined as < 33 weeks

gestational age) ranges 2–11%, with increased prematurity associated with increased rates of hearing loss (Wroblewska-Seniuk, Greczka, Dabrowski, Szyfter-Harris, & Mazela, 2017).

Independently of hearing status, prematurity is linked to increased risk of language delay and disorder (Barre, Morgan, Doyle, & Anderson, 2011; Carter & Msall, 2017; Cusson, 2003; Rechia, Oliveira, Crestani, Biaggio, & de Souza, 2016; Van Noort-van Der Spek, Franken, & Weisglas-Kuperus, 2012; Vohr, 2014). Unfortunately, research on language development in premature DHH children is scant (Vohr, 2016), so it remains unclear how hearing loss and prematurity may interact within spoken language skills. One study of premature infants finds that auditory brainstem response during newborn hearing screening predicts language performance on the PLS-4 at age 3 (Amin, Vogler-Elias, Orlando, & Wang, 2014), suggesting a link between prematurity, hearing loss, and language development in early childhood, though further research is needed in this domain. In extremely premature DHH children, incidence of additional disabilities may be as high as 73% (Robertson, Howarth, Bork, & Dinu, 2009). Indeed, pre-term infants with comorbidities have been found to be more likely to also have hearing loss than those without comorbidities (Schmidt et al., 2003), further complicating language development for this population.

Audiological Characteristics. Hearing loss varies in severity, ranging from slight to profound (Clark, 1981). More severe hearing loss (less access to spoken language) typically results in more difficulty with spoken language in infancy (Vohr et al., 2008), early childhood (Ching et al., 2010, 2013; Sarant et al., 2008; Sininger, Grimes, & Christensen, 2010; Tomblin et al., 2015) and school-age children (Wake, Hughes, Poulakis, Collins, & Rickards, 2004). Although profound hearing loss is associated with more pronounced spoken language difficulty, even mild to moderate hearing loss is associated with elevated risk of language disorders (Blair, Peterson, & Viehweg, 1985; Delage & Tuller, 2007).

Hearing loss also varies in whether it affects one ear or both. Bilateral hearing assists speech perception, sound localization, and loudness perception in quiet and noisy

124 environments (Ching, Van Wanrooy, & Dillon, 2007). The literature on hearing aids and

125 cochlear implants also points to benefits for bilateral auditory input (Lovett, Kitterick,

126 Hewitt, & Summerfield, 2010; Sarant, Harris, Bennet, & Bant, 2014; Smulders et al., 2016).

127 At school-age, 3–6% of children have unilateral hearing loss (Ross, Visser, Holstrum, Qin, &

128 Kenneson, 2010). Although children with unilateral hearing loss have one "good ear," even

129 mild unilateral hearing loss has been tied to higher risk of language delays and educational

130 challenges relative to hearing children (Kiese-Himmel, 2002; Lieu, 2004, 2013; Lieu,

131 Tye-Murray, & Fu, 2012; Vila & Lieu, 2015). Just as in the bilateral case, more severe

132 hearing loss leads to greater deficits in spoken language and educational outcomes for

133 children with unilateral hearing loss (Anne, Lieu, & Cohen, 2017; Lieu, 2013).

134   Many DHH children receive hearing aids (HAs) or cochlear implants (CIs) to boost

135 access to the aural world. These devices have been associated with better speech perception

136 and spoken language outcomes (Niparko et al., 2010; Walker et al., 2015; Waltzman et al.,

137 1997). In turn, aided audibility predicts lexical abilities in children with HAs (Stiles, Bentler,

138 & McGregor, 2012).

139   For both hearing aids and cochlear implants, earlier fit leads to better spoken language

140 skills, if the amplification is effective. For hearing aids, some studies find that children with

141 milder hearing loss who receive hearing aids earlier have better early language achievement

142 than children who are fit with hearing aids later (Tomblin et al., 2015), but this finding does

143 not hold for children with severe-to-profound hearing loss (Kiese-Himmel, 2002; Watkin et

144 al., 2007) (for whom hearing aids are generally ineffective). Analogously, for children eligible

145 for cochlear implants, those who receive cochlear implants earlier have better speech

146 perception and spoken language outcomes than those implanted later (Artières, Vieu,

147 Mondain, Uziel, & Venail, 2009; Dettman, Pinder, Briggs, Dowell, & Leigh, 2007; Miyamoto,

148 Hay-McCutcheon, Kirk, Houston, & Bergeson-Dana, 2008; Svirsky, Teoh, & Neuburger, 2004;

149 Yoshinaga-Itano et al., 2018), with best outcomes for children receiving implants before their

first birthday (Dettman et al., 2007).

Communication.   Total Communication refers to communication that combines speech, gesture, and elements of sign, sometimes simultaneously. Total communication, while often including elements of sign such as individual signs, is not a full-fledged sign language like American Sign Language (Mueller, 2013; Scott & Henner, 2020). Clinicians currently employ total communication as an alternative or augmentative communication method for children with a wide range of disabilities (Branson & Demchak, 2009; Gibbs & Carswell, 1991; Mirenda, 2003).

Compared to total communication, DHH children using an exclusively oral approach have better speech intelligibility (Dillon, Burkholder, Cleary, & Pisoni, 2004; Geers et al., 2017; Geers, Spehar, & Sedey, 2002; Hodges, Dolan Ash, Balkany, Schloffman, & Butts, 1999) and auditory perception (Geers et al., 2017; O'Donoghue, Nikolopoulos, & Archbold, 2000). That said, there is some debate as to whether an oral approach facilitates higher spoken language performance, or whether children who demonstrate aptitude for spoken language are steered towards the oral approach rather than total communication (Hall, Levin, & Anderson, 2017).

1-3-6 Guidelines.   Early identification (Apuzzo & Yoshinaga-Itano, 1995; Kennedy et al., 2006; Robinshaw, 1995; White & White, 1987; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998; Yoshinaga-Itano et al., 2018) and timely enrollment in early intervention programs (Ching, Dillon, Leigh, & Cupples, 2018; Ching et al., 2013; Holzinger, Fellinger, & Beitel, 2011; Vohr et al., 2008, 2011; Watkin et al., 2007) are associated with better language proficiency. Indeed, DHH children who receive prompt diagnosis and early access to services have been found to meet age-appropriate developmental outcomes, including language (Stika et al., 2015). In line with these findings, the American Academy of Pediatricians (AAP) has set an initiative for Early Hearing Detection and Intervention (EHDI). Their EHDI guidelines recommend that DHH children are screened by 1 month old, diagnosed by 3 months old, and enter early intervention services by 6 months old. We refer to this guideline

as 1-3-6. Meeting this standard appears to improve spoken language outcomes for children

with HL (Yoshinaga-Itano et al., 2017, 2018) and the benefits appear consistent across a

range of demographic characteristics.

At a federal level in the U.S., the Early Hearing Detection and Intervention Act of

2010 (Capps, 2009) was passed to develop state-wide systems for screening, evaluation,

diagnosis, and "appropriate education, audiological, medical interventions for children

identified with hearing loss," but policies for early diagnosis and intervention vary by state.

As of 2011, 36 states (including North Carolina; "15A NCAC 21F .1201 - .1204," 2000)

mandate universal newborn hearing screening (UNHS; National Conference of State

Legislatures, 2011). All states have some form of early intervention programs that children

with hearing loss can access (NAD, n.d.), but the specifics vary state-by-state. For instance,

half of the states in the US do not consider mild hearing loss an eligibility criterion for early

intervention (Holstrum, Gaffney, Gravel, Oyler, & Ross, 2008); North Carolina does include

mild hearing loss in early intervention.

In evaluating the success of this initiative, the AAP (EHDI, n.d.) finds that about 70%

of US children who fail their newborn hearing screening test are diagnosed with hearing loss

before 3 months old, and that 67% of those diagnosed (46% of those that fail newborn

hearing screening) begin early intervention services by 6 months old. These findings suggest

that there are breaks in the chain from screening to diagnosis and from diagnosis to

intervention, with potential ramifications for the language development of children not

meeting these guidelines. We return to this in the discussion.

Quantifying vocabulary growth in DHH children

In what follows, we analyze data from the MacArthur Bates Communicative

Development Inventory (CDI, Fenson et al., 1994). This parent-report instrument gathers

information about children's vocabulary development, and is commonly used in both

research and applied settings. The Words and Gestures version of the form is normed for

8–18-month-olds. On Words and Gestures, parents indicate whether their child understands

and/or produces each of the 398 vocabulary items, and answer questions about young

children's early communicative milestones. The Words and Sentences version of the form is

normed for 16–30-month-olds. On Words and Sentences, parents indicate whether their child

produces each of the 680 vocabulary items, and answer some questions about grammatical

development. The CDI has been normed on a large set of participants across many

languages (Anderson & Reilly, 2002; Frank et al., 2017; Jackson-Maldonado et al., 2003).

The CDI has also been validated for DHH children with cochlear implants (Thal,

Desjardin, & Eisenberg, 2007). More specifically, in this validation, researchers asked parents

to complete the CDI, administered the Reynell Developmental Language Scales, and

collected a spontaneous speech sample. All comparisons between the CDI and the other

measures yielded significant correlations ranging from 0.58 to 0.93. Critically, the children in

this study were above the normed age range for the CDI, and thus this validation helps to

confirm that the CDI is a valid measurement tool for older DHH children. In further work,

Castellanos, Pisoni, Kronenberger, and Beer (2016) find that in children with CIs, number of

words produced on the CDI predicts language, executive function, and academic skills up to

16 years later. Building on this work, several studies have used the CDI to measure

vocabulary development in DHH children (Yoshinaga-Itano et al. (2017); Yoshinaga-Itano et

al. (2018); de Diego-Lázaro et al. (2018); Vohr et al. (2008); Vohr et al. (2011). We build on

this literature in our analyses below.

Goals and Predictions

This study aims to 1) characterize the demographic, audiological, and intervention

variability in the population of DHH children receiving state services for hearing loss; 2)

identify predictors of vocabulary delays; and 3) evaluate the success of early identification

and intervention efforts at a state level. We include three subgroups of DHH children

traditionally excluded from studies of language development: children with additional

disabilities, children with unilateral hearing loss, and children from bilingual or

non-English-speaking households (e.g., Yoshinaga-Itano et al., 2018; Nicholas & Geers, 2006).

For the first goal, we expected that many of these variables would be related, due to
known causal relations (e.g., cochlear implants recommended for severe hearing loss, but not
mild hearing loss). We sought to provide descriptive documentation about the distribution of
demographic, audiological, and intervention characteristics in a diverse sample of DHH
children receiving state services. For the second, we hypothesized that male (vs. female)
gender, more severe degree of hearing loss, bilateral (vs. unilateral) hearing loss, no
amplification (vs. hearing aids and/or cochlear implants), premature birth, and presence of
additional disabilities would predict larger spoken vocabulary delay. We did not have strong
predictions regarding the effects of communication method or presence of other health issues
(e.g., congenital heart malformation) on vocabulary. For the third goal, based on the prior
literature summarized above, we hypothesized that children with less residual hearing (i.e.,
bilateral, more severe) and no co-occurring conditions would be earlier diagnosed and earlier
to begin language services, and that in turn earlier diagnosis would predict earlier
intervention.

Methods

Clinical evaluations were obtained through an ongoing collaboration with the North
Carolina Early Language Sensory Support Program (ELSSP), an early intervention program
serving children with sensory impairments from birth to 36 months. ELSSP passed along
deidentified evaluations to our team after obtaining consent to do so from each family. No
eligibility criteria beyond hearing loss and receiving an ELSSP evaluation were imposed,
given our goal of characterizing the full range of DHH children with hearing loss in North
Carolina.

253   The clinical evaluations included demographic and audiological information, CDI

254   vocabulary scores, and the results of any clinical assessments administered (e.g., PPVT),

255   detailed further below. For some children (n = 47), multiple evaluations were available from

256   different timepoints. In these cases, only the first evaluation was considered for this study,

257   due to concerns regarding within-subjects variance for statistical analysis.

258   While this collaboration is ongoing, we opted to pause for this analysis upon receiving

259   data from 100 children. Thus, the reported sample below consists of 100 children (56 male /

260   44 female) ages 4.20–36.20 months (M=21.20, SD=9.10). Race and socioeconomic

261   information were not available. Families were administered either the Words and Gestures or

262   Words and Sentences version of the CDI based on clinician judgment. Children who were too

263   old for Words and Gestures, but who were not producing many words at the time of

264   assessment, were often given Words and Gestures (n = 37). Families for whom Spanish was

265   the primary language (n = 14) completed the Spanish language version of the CDI

266   (Jackson-Maldonado et al., 2003). See Table 1 for additional CDI information for our sample.

267   With regard to comorbid diagnoses, children in this sample were coded as yes/no for

268   cognitive development concerns (e.g., Down syndrome, global developmental delays; Cornelia

269   de Lange syndrome), yes/no for premature birth (i.e., more than 3 weeks premature), yes/no

270   for health issues (e.g., heart defects, kidney malformations, VACTERL association), and

271   yes/no for vision loss (not corrected to normal by surgery or glasses); see Table 2.

272   Degree of hearing loss was most often reported with a written description (e.g., "mild

273   sloping to moderate" or "profound high frequency loss"). We created 3 variables: hearing

274   loss in the better ear, hearing loss in the worse ear, and average hearing loss (average of

275   better and worse ear). For the analyses below, we primarily use hearing loss in the worse ear

276   to avoid any redundancies with laterality. Using the ASHA hearing loss guidelines, each of

277   these hearing loss measures was coded with the decibels of hearing loss (dB HL)

278   corresponding with the median dB HL for the level of hearing loss (e.g., moderate hearing

loss was coded as 48 dB HL), and sloping hearing loss was coded as the average of the levels

(e.g. mild to moderate was coded as 40.5 dB HL). Participants were also coded for unilateral

or bilateral hearing loss; presence or absence of Auditory Neuropathy Spectrum Disorder;

and etiology of hearing loss (sensorineural, conductive, or mixed). Amplification was

recorded as the device the child used at the time of assessment: either hearing aid, cochlear

implant, or none. See Table 3 for audiological characteristics of the sample.

Communication method was recorded as spoken language, total communication, or

cued speech. One participant had a parent fluent in sign language, but the reported

communication method in the home was total communication. No child in our sample used

American Sign Language or another signed language. The forms also listed the primary

language spoken at home, which we binned into English-speaking and non-English-speaking.

85 out of the 100 had a primary language of English, while 14had Spanish. The remaining

child was adopted from a non-English-speaking country after age 2 and had heard mostly

non-English by the time of assessment; this child was coded as non-English-speaking.

Language and communication information is summarized in Table 4.

Age at screening was measured as the child's age in months at their first hearing

screening. Age at screening was available for 68 participants. All participants with a

screening age available were screened at birth or while in the NICU. We presume that the

vast majority of participants without age at screening received their screening as newborns,

as North Carolina boasts a 98% newborn hearing screening rate (NCDHHS, 2013). Age at

diagnosis was taken as the age in months when children received their first hearing loss

diagnosis. All children were enrolled in birth-to-three early intervention services through

ELSSP, and the date of enrollment was listed on the clinician evaluation. For determining

whether participants met the 1-3-6 guidelines, given the very high rates of early screening

reported in our sample and by the state, we imputed missing data by assuming that children

met the "screening by 1 month" criterion if they met the "diagnoses by 3 months". Finally,

305 we also calculated the number of hours of early intervention services received per month

306 (including service coordination, speech therapy, and occupational therapy, among others)

307 based on the clinician report.

308   A summary of all the variables we examined is available in Table 5.

Results

310   We split the results into three parts. In the first, we explore relationships among child

311 demographic, audiological, and clinical variables. In the second, we use these variables to

312 predict vocabulary development. Finally, in the third, we describe the implementation of the

313 EHDI 1-3-6 guidelines and predictors of early diagnosis and intervention in this sample. All

314 analyses were conducted in R. All code is available on Github.

Relationships Among Demographic, Audiological, and Clinical Variables

316   Before we test how these variables may be related to vocabulary, we describe their

317 relationships to each other. As would be expected, many health, audiological, and clinical

318 characteristics are not distributed randomly across this sample of children. To quantify this

319 statistically, we used Bonferroni-corrected chi-square tests between each of our variables[2].

320 Because the chi-square statistic assumes n > 5 is *expected* in the majority of the cells for

321 each test (preferably $\geq 80\%$ McHugh, 2013), we excluded mixed hearing loss (n = 8) and

322 cued speech (n = 1) from this section of the analysis. Strictly speaking, some variables are

323 not expected to be randomly distributed relative to each other (e.g., premature birth and

324 health issues; degree and amplification), but quantifying the differences via chi-square using

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[2] Variables for chi-square: gender (male/female), laterality (bi-/uni-lateral hearing loss), health issues (yes/no), developmental delays (yes/no), premature birth (yes/no), language background (English/non-English), 1-3-6 (yes/no), degree of hearing loss (mild, moderate, severe/profound as defined above), etiology (sensorineural/conductive), services received per month (binned into 0-2, 3-6, and >7 - to create maximally evenly sized bins), communication (spoken/total communication) and amplification (hearing aids/cochlear implants/none.

a conservative significance threshold lets us highlight the strongest relationships within this dataset.

Given that we ran 66 Chi-square tests, Bonferroni-corrected alpha for this set of analyses was p < 0.0007. Of these 66 combinations of variables, p < .05 for 26, and 9 survived Bonferroni correction. We are only discussing the latter below, but the full set of results can be found in Figure 1.

As expected, we found that health issues, developmental delays, and premature birth were highly interrelated in our sample, such that children born premature were more likely to also experience health issues (X^2 (1, N = 98) = 23.9, p < .0001) and developmental delays (X^2 (1, N = 98) = 11.63, p = .0006), and children with developmental delays were more likely to also experience health issues (X^2 (1, N = 98) = 20.87, p < .0001). Children with developmental delays received more services per month than typically-developing children (X^2 (2, N = 95) = 22.17, p < .0001) and were more likely to use total communication (X^2 (2, N = 98) = 22.51, p < .0001). Likewise, children who used total communication received more services per month than children using spoken language (X^2 (4, N = 95) = 21.35, p = .0003).

We also confirmed expected relationships among many of the audiological characteristics. There was a significant relationship between laterality and etiology (X^2 (2, N = 88) = 18.29, p = .0001), such that children with conductive hearing loss were more likely to have unilateral hearing loss, and children with sensorineural hearing loss were more likely to have a bilateral loss[3]. The chi-square tests further showed that amplification was related to laterality (X^2 (2, N = 98) = 16.43, p = .0003) and degree of hearing loss (X^2 (4, N = 87) = 28.45, p < .0001) in our sample. Specifically, children with bilateral hearing loss were more likely than children with unilateral hearing loss to use a hearing aid or cochlear

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[3] All children with mixed hearing loss (n = 8) had bilateral hearing loss.

implant; no child with unilateral hearing loss used a cochlear implant, and many children

with unilateral hearing loss used no amplification. Regarding degree, children with severe to

profound hearing loss were more likely to use a cochlear implant than children with less

severe hearing loss (i.e., mild or moderate).

Taken together, the results in this set of analyses serve to highlight the notable

interconnectedness among early health and development on the one hand (i.e. health issues,

prematurity, and developmental delays), and audiological characteristics (i.e. links among

laterality, etiology, amplification, and degree of hearing loss) on the other.

Predictors of Vocabulary Delay

We next turn to the relationship between each of these variables and children's

productive vocabulary, as measured by the CDI. Figure 2 shows the vocabulary scores of

children in our samples relative to norms for hearing children for each CDI form.

Descriptively, we found widespread vocabulary delays on both Words and Gestures and

Words and Sentences, with the majority of DHH children testing around or below the 25th

percentile for hearing children (based on WordBank norms; Frank et al., 2017).

As noted above, the CDI is composed of two instruments, which differ in number of

questions (i.e. the maximum vocabulary score is 398 on Words and Gestures and 680 on

Words and Sentences; 428 and 680 respectively for Spanish language CDI). To take this into

account, rather than using the raw number of words produced as our outcome variable, we

use WordBank norms to establish the difference (in months) between the child's

chronological age and their predicted age based on their vocabulary, derived from the

WordBank norms (Frank et al., 2017). We call this derived variable *vocabulary delay.*

More specifically, to compute a child's predicted age from their vocabulary score, we

used the 50th percentile for productive vocabulary from Wordbank data typically-developing

infants (Frank et al., 2017) to create binary logistic growth curves separately for the "Words

374 and Gestures" (WG) and "Words and Sentences" (WS) versions of the CDI for American

375 English and Mexican Spanish[4]. For each child, we took the number of words they produced

376 divided by the number of words on the instrument, to give us the proportion of words

377 produced. We used this proportion in an inverse prediction from the binary logistic

378 regression curves to generate a predicted age. That is, for each possible CDI score, the

379 growth curve provided the age that the score would be achieved for the 50th percentile

380 trajectory. Finally, we subtracted the predicted age from each child's chronological age to

381 calculate their vocabulary delay. However, for children producing 0 words, this approach was

382 not appropriate due to the long tails on the growth curves. Thus, for this subset of children,

383 we took the x-intercept from Wordbank (8 months for English, and 9 months for Spanish),

384 and subtracted that value from the child's chronological age to get their vocabulary delay.

385   To look at the relationship between our predictor variables and CDI scores, we next

386 conducted multiple linear regression, using vocabulary delay as our outcome variable. [5]

387   Our full regression model included all variables: Vocabulary Delay ~ Gender +

388 Developmental Delay + Health Issues + Premature Birth + Laterality + Degree +

389 Amplification + Communication + Meets 1-3-6 + Services Received Per Month + Language

390 Background.

391   This model accounted for significant variance in vocabulary delay (adjusted-$R^2 = 0.59$,

392 $p < .001$). We next performed stepwise model comparison using stepAIC (MASS) to pare

393 down the model. This process selects only the predictors which incrementally improved

394 model fit, measured by Akaike's Information Criterion (AIC), which considers goodness of fit

395 and model complexity (penalizing models with many predictors). We started model selection

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[4] Number of hearing children in normative sample for each growth curve: WG-English=1071,

WG-Spanish=760; WS-English=1461, WS-Spanish=1092

[5] We excluded the adopted child from this section of the analysis due to concerns about comparing her score

to the American English CDI norms.

396 with the full model, as described above. We then filtered out data from children for whom

397 Meets 1-3-6 (n = 5) or Degree (n = 13) was unknown, as this stepwise AIC approach does

398 not permit missing values across predictors. Since this initial filtered analysis found that

399 Degree and 1-3-6 did not improve model fit, we manually removed the Degree and 1-3-6

400 terms from the model selection so that the 15 participants with missing cases for these

401 variables could be retained.[6]

402   Based on this iterative process, we arrived at the following final model: Vocabulary

403 Delay ~ Age + Laterality + Amplification. This model accounted for significant variance in

404 children's vocabulary delay to a nearly identical degree as the full model (adjusted-R^2 =

405 0.58, p = < .001, see Table 6 & Figure 5.A). We found significant main effects for Age,

406 Amplification, and Laterality, such that older age, no amplification, and bilateral hearing loss

407 predicted greater vocabulary delays. Compared to children with no amplification, children

408 with cochlear implants had a 3.50 months smaller spoken vocabulary delay (p = .021), and

409 similarly children with hearing aids had a 3.84 months smaller delay (p = .001). Children

410 with unilateral hearing loss had a 2.70 months smaller delay (p = .020) than children with

411 bilateral hearing loss. With regard to Age, for each month older, the model predicted a 0.55

412 months *larger* vocabulary delay (p < .001).

413   Given our results above revealing relationships among several of these variables (e.g.,

414 laterality and amplification), we tested for collinearity concerns by computing the model's

415 VIF (variance inflation factor). This revealed low levels of collinearity among predictors in

416 our final model (all VIF < 1.20; James, Witten, Hastie, & Tibshirani, 2013). In sum, the

417 analyses in this section revealed that over half of the variance in DHH children's vocabulary

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[6] For transparency, we note that the model fitted with only complete cases of Degree did include a

non-significant main effect of Developmental Delay. However, ANOVA revealed that including a

Developmental Delay term did not significantly improve model fit when including the 15 participants without

Degree information.

scores was explained by their age, whether their receive amplification, and whether their

hearing loss was unilateral or bilateral.

Success in Meeting 1-3-6 Guidelines

Perhaps of greatest importance to clinicians and policymakers is the implementation

and effect of existing policies. Although whether a child met 1-3-6 guidelines was not

included in our final model predicting vocabulary delay through our model selection process,

its demonstrated importance for language outcomes (e.g., Yoshinaga-Itano et al., 2018)

merits further discussion. To this end, we looked at the ages at which children received

diagnosis and intervention, and how this mapped onto the 1-3-6 guidelines. In this section,

we provide a brief description of the implementation of 1-3-6 in our sample, examine its

effect on vocabulary delay, and describe the results of exploratory linear regression models

for age at diagnosis and age at intervention.

Overall, 37% of our sample met 1-3-6 guidelines for early diagnosis and intervention.

Among the children for whom screening information was available (n = 68), 100% were

screened at birth or during NICU stay. 69% of children received diagnosis by 3 months of

age, and 39% began early intervention by 6 months of age. Among children with

comorbidities, 21.05% met 1-3-6 guidelines, compared to 47.37% of children without

comorbidities. Figure 3 shows the age at first diagnosis, intervention, amplification, and

implantation for each child in our sample.

We first tested the link between 1-3-6 and vocabulary directly in an exploratory

analysis. An independent samples t-test showed that children who did not meet 1-3-6

guidelines had significantly larger vocabulary delays than children who met 1-3-6 guidelines

($t(68.78)$=2.62, p=0.01; see Figure 4). On average, the group that did not meet 1-3-6

guidelines was 3.62 months more delayed with regard to vocabulary (relative to the same

50th percentile benchmark from hearing children in Wordbank described above).

⁴⁴³   To better understand implementation of 1-3-6 guidelines, we next zoomed in on

⁴⁴⁴ diagnosis and intervention. We conducted two linear regressions, one for age at diagnosis and

⁴⁴⁵ one for age at intervention, considering only the predictors that would have been available or

⁴⁴⁶ relevant at each of these stages (as detailed below). Model selection followed the same

⁴⁴⁷ stepwise AIC-based process as described in the preceding section.

⁴⁴⁸   For age at diagnosis, we included the set of child-specific factors that would be relevant

⁴⁴⁹ *before* diagnosis of hearing loss (e.g., we excluded amplification type because a child would

⁴⁵⁰ not receive a hearing aid or cochlear implant prior to being diagnosed with hearing loss.) We

⁴⁵¹ began with: gender, degree, developmental delay, health issues, prematurity, laterality,

⁴⁵² language background, and etiology.

⁴⁵³   The best fit model was: Age at Diagnosis ~ Health Issues + Language Background +

⁴⁵⁴ Laterality, with significant main effects of Health Issues and Language Background (see

⁴⁵⁵ Table 7 & Figure 5.B). This model accounted for 16.41% of the variance in age at diagnosis

⁴⁵⁶ (p = .001). Average age at diagnosis was 4.65(7.19) months. Relative to English-speaking

⁴⁵⁷ families, children from Spanish-speaking families were diagnosed 6.47 months later (p =

⁴⁵⁸ .001). Children with health issues were diagnosed 3.70 months later than children without

⁴⁵⁹ health issues (p = .01).

⁴⁶⁰   We repeated this model selection process for age at intervention. In addition to the

⁴⁶¹ variables used to fit the intervention model, we included age at diagnosis. The best fit model

⁴⁶² was: Age at Intervention ~ Premature Birth + Degree + Age at Diagnosis + Language

⁴⁶³ Background (R^2=0.43 , p < .001; See Table 8 & Figure 5.C), with significant main effects of

⁴⁶⁴ degree and age at diagnosis. Prematurity (ß = 3.78, p = .06) and language background (ß =

⁴⁶⁵ -1.38, p = .52) were not significant predictors on their own, but their inclusion improved

⁴⁶⁶ model fit. Average age at intervention was 11.12(8.54) months. More severe hearing loss

⁴⁶⁷ predicted earlier intervention, such that for every additional 10 dB HL, predicted age at

⁴⁶⁸ intervention was 1 month earlier (p < .01). With regard to age at diagnosis, for every month

diagnosis was delayed, intervention was delayed by 2.80 weeks (p < .01). Taken together these analyses reveal that beyond aspects of the child's hearing status, other variables too contribute to delays in both diagnoses and intervention. We return to this point in the discussion.

Discussion

In this study, we examined the demographic, audiological, and clinical characteristics of 100 young DHH children in North Carolina. We documented the distribution of these characteristics and explored the relationships between these variables, vocabulary, diagnosis, and intervention. In prior work with tightly controlled samples, the variables studied here have been shown to be relevant for language development, but their effects have rarely examined in the full heterogeneity they naturally occur within. We took this big-tent approach by including any children receiving services for hearing loss.

Returning to our original three questions, we asked first: how are child-level variables intertwined? We found significant structure across many of the variables, suggesting that in a real-world sample of children with hearing loss, many factors are intrinsically not dissociable. This was particularly true for many of the auditory characteristics and comorbid diagnoses. To our knowledge, this paper provides the first population-based documentation of this distribution. We next asked whether these characteristics can predict vocabulary outcomes for DHH children. We found that a model including only children's age, laterality of hearing loss, and amplification type best accounted for the variability in spoken vocabulary outcomes. Finally, we asked: how successful were the 1-3-6 guidelines for early detection and intervention, both in terms of improving child outcomes and ensuring timely diagnosis and intervention for all children with hearing loss? Here, we found that children who met 1-3-6 guidelines indeed had a smaller vocabulary delay than those who didn't. However, only 37% of children met these guidelines. Our results highlight family- and health-related variables (e.g. language background, health issues) that accounted for

significant variability in when children received diagnosis and/or intervention.

To us, the inherent complexity in these results is an important piece of understanding spoken language outcomes for children with hearing loss within the diverse population of Deaf/Hard-of-Hearing children. We next highlight some implications of this study for future research and clinical practice.

How are child-level variables intertwined?

In our sample, we found significant overlap among demographic, audiological, and clinical variables. Prematurity, health issues, and developmental delay frequently co-occurred, such that children with one of these factors were more likely to have the others. This is not surprising. Many conditions that cause developmental delays have a high incidence of health issues (e.g., heart problems in Down Syndrome; vomiting and seizures with hydrocephalus), and it is well documented that there is a higher incidence of developmental delay and health issues in preterm infants (Aarnoudse-Moens, Weisglas-Kuperus, van Goudoever, & Oosterlaan, 2009; Costeloe et al., 2012; Luu, Katz, Leeson, Thébaud, & Nuyt, 2016; Pierrat et al., 2017; Robertson et al., 2009; York & DeVoe, 2002). In our sample, we also had a large range of health conditions (76 unique conditions in our sample of 100 children; see Table 2 and elssp_comorbidities.csv on OSF for more detailed information about comorbidities). Some studies to date have examined the outcomes of DHH children with certain conditions (e.g., Clibbens, 2001; Szymanski, Brice, Lam, & Hotto, 2012). But given that the constellation of comorbid conditions is so varied, an important direction for future research is whether cognitive and social abilities, as well as family's treatment resources, are predictive of language outcomes across conditions.

We also found that children with developmental delays (e.g., Down syndrome) were much more likely to use a total communication approach than DHH children without developmental delays (i.e., total communication used by 58.82% of DHH children with

developmental delay vs. 9.88% of those without). That is, communication modality was not distributed randomly throughout our sample, with use of total communication linked to children already at greater risk for verbal delays. Such a pattern is in line with clinical use of manual communication approaches for young children with disabilities (e.g., Branson & Demchak, 2009). This result tempers the interpretation of correlational studies finding links between total communication and language delays (e.g., Geers et al., 2017).

Our audiological variables too were not randomly distributed relative to each other. To highlight one such result, amplification devices were more common for children with less hearing (i.e., children with bilateral hearing loss and children with moderate to profound hearing loss). This may be due to the assumption that a hearing aid or cochlear implant will not benefit children with minimal hearing loss (Updike, 1994), although several studies have found benefits for amplification for mild or unilateral hearing loss (Briggs, Davidson, & Lieu, 2011; Hassepass et al., 2013; Priwin, Jönsson, Hultcrantz, & Granström, 2007; Walker et al., 2015; Winiger, Alexander, & Diefendorf, 2016).

The relationships we found among variables were more confirmatory than surprising, particular those reflecting known causal links (e.g., increased health issues in children born premature). Nevertheless, they should caution us to think critically about how we construct samples for controlled lab experiments. During study design: how likely is it to collect a desired sample of (e.g.) 32 typically-developing pediatric cochlear implant users with bilateral, severe-to-profound hearing loss, given that such a subsample may only represent roughly 14% of the DHH population, as it does here? During interpretation of the results: how might the findings generalize to the rest of the DHH population given the constraints of the study at hand? Such considerations are important for properly representing, understanding, and supporting DHH children and their families. This becomes doubly important in the context of interpreting language outcomes like vocabulary.

Predicting vocabulary outcomes

In our sample, 88.89% of DHH children fell below the 50th percentile for spoken vocabulary. Moreover, of the 11.11% who were at or above the 50th percentile, 55.56% were 8-to-9-month olds who were not yet producing any words (as expected at this age). Finding that nearly 90% of DHH children are below the 50th percentile for vocabulary development indicates that this group is not yet well-equipped to acquire spoken language. This disadvantage can have lasting consequences in the lives of DHH children (Karchmer & Mitchell, 2003; Kyle & Harris, 2010; Qi & Mitchell, 2012), highlighting the importance of understanding what factors contribute to it.

We predicted that male gender, more severe hearing loss, bilateral hearing loss, no amplification, premature birth, and presence of additional disabilities would be associated with larger spoken vocabulary delay. In contrast to our predictions, the best model predicting vocabulary delay had just a few variables: age, amplification, and laterality. Notably, we did not simply find that DHH children were learning words at the same rate (albeit delayed) as hearing children, which would have led to a constant delay across developmental time. Instead, we see that the spoken vocabulary delay widens with age, indicating that the *rate* of spoken vocabulary acquisition is slower for DHH children. The result is a population increasingly behind on spoken language milestones. Given that none of the children here use sign language (which can ensure earlier language access) this vocabulary delay is likely to have knock-on effects for language development more broadly as well. This in turn has policy implications that are critical to consider.

Predicting early diagnosis and intervention

Our exploration of the implementation of 1-3-6 guidelines revealed that only 36.84% of children met the EHDI guidance for diagnosis by 3 months and intervention by 6 months, despite ample evidence suggesting early diagnosis and intervention improve language outcomes (Apuzzo & Yoshinaga-Itano, 1995; Ching et al., 2013; Holzinger et al., 2011;

571 Kennedy et al., 2006; Robinshaw, 1995; Vohr et al., 2008, 2011; Watkin et al., 2007; White &

572 White, 1987; Yoshinaga-Itano et al., 1998, 2018). Children in our sample who met 1-3-6

573 guidelines were 3.62 months *less* delayed in spoken vocabulary than children who were late

574 to receive diagnosis and/or services. With these demonstrable benefits in mind, our sample,

575 by dint of accepting all children receiving early intervention services in one state, was able to

576 explore naturally occurring variance in *who* received on-time diagnosis and intervention.

577 **Diagnosis.** Having health issues or a non-English language background predicted

578 later diagnosis. Children with health issues were diagnosed 3.70 months later than infants

579 without health issues. One possible explanation is that the health issues caused acquired

580 hearing loss that wouldn't be detected by the newborn hearing screening, thus delaying

581 identification of hearing loss. In our sample, 16 of the 36 children with health issues had

582 conditions that might cause acquired hearing loss (i.e., meningitis, sepsis, jaundice, seizures,

583 hydrocephalus, MRSA, anemia, frequent fevers, cytomegalovirus). While acquired hearing

584 loss may be one driver of delayed diagnosis for children with health issues, this accounts for

585 only a fraction of the subpopulation with health issues. Another possible explanation is that

586 the health issues required more pressing medical attention than the possible hearing loss. For

587 instance, families and medical providers are likely to prioritize treatment for certain health

588 issues (e.g., surgery for congenital heart defect) over diagnostic audiology services.

589 Nevertheless, it is possible that in some cases, clinician awareness of the increased delays in

590 language related to health issues more broadly may facilitate improvements in timely

591 diagnosis.

592 Language background too predicted age at diagnosis. Infants from Spanish-speaking

593 families were diagnosed 3.78 months later than infants from English-speaking families. This

594 may be due to cultural differences in attitudes towards deafness (Caballero, Muñoz, Schultz,

595 Graham, & Meibos, 2018; Rodriguez & Allen, 2020; Steinberg, Bain, Li, Delgado, & Ruperto,

596 2003; Steinberg, Dávila, Collazo, Loew, & Fischgrund, 1997) or it may result from a lack of

597 linguistically accessible and culturally appropriate audiology services. Only 5.6% of

American audiologists identify as bilingual service providers (ASHA, 2019), and services from a monolingual provider may be insufficient. To this point, Caballero et al. (2017) found that Hispanic-American parents of DHH children wish for more concrete resources, comprehensive information, and emotional support from their audiologist. In a nationwide survey of audiologists, the majority of audiologists reported that language barriers presented a major challenge in working with Spanish-speaking families, specifically in obtaining the child's case history and providing recommendations for follow-up services(Abreu, Adriatico, & DePierro, 2011).

Intervention. As expected, more severe hearing loss predicted earlier intervention, such that for every additional 10 dB HL, predicted age at intervention was 0.93 month earlier. This converges with findings by Harrison, Roush, and Wallace (2003) in which severe-to-profound hearing loss was diagnosed 2-5 months earlier than mild-to-moderate hearing loss. Parents and clinicians may adopt a wait-and-see approach to intervention for children with some residual hearing. Nevertheless, mild-to-moderate hearing loss is associated with language delays and academic challenges (Blair et al., 1985; Delage & Tuller, 2007), which early intervention may offset.

Age at start of services was also associated with age at diagnosis: for every month diagnosis was delayed, intervention was delayed by 2.80 weeks. Ching et al. (2013) found that age at intervention predicted better outcomes for DHH children, above and beyond age at diagnosis. Of course, these two variables are related, such that we cannot hope to achieve early intervention goals without ensuring children receive timely diagnosis. Early diagnosis puts children in the pipeline towards intervention earlier.

Finally, it's important to note that this sample is composed of children receiving birth-to-3 services. An estimated 67% of children with hearing loss enroll in early intervention services (CDC, 2018). While this represents a tremendous step forward in prompt early intervention services relative to just a few decades ago, early intervention may

not be early enough. Less than 39% of our sample of children in early intervention meet the 6-month EHDI benchmark. Furthermore, an unknown fraction of the DHH population in North Carolina aren't included in this analysis because they have not been enrolled in services by 36 months. The AAP estimates that almost 36% of infants who do not pass a newborn hearing screening are lost to follow-up. Assuming that the population of children in early intervention only represents two thirds of the population with hearing loss, our data suggest that the actual proportion of DHH children who receive intervention by the EHDI-recommended 6 months may be closer to 26%. These children may not receive clinical support until school-age or later, exacerbating concerns for language development, which lays an important foundation for literacy and academic success (Biemiller, 2003; Hemphill & Tivnan, 2008; Monroe & Orme, 2002; Stæhr, 2008; Young, 2005).

Educational and Clinical Implications

Despite high rates of newborn hearing screening in North Carolina, and even relatively high rates of diagnosis by 3 months (66/100 children in our sample), most children in our sample did not meet the 1-3-6 guidelines. Based on our analyses, we have the following recommendations for increasing attainment of 1-3-6 guidelines:

1. Frequent hearing screenings for children receiving medical or therapeutic care for health issues.

2. Service coordination for families balancing multiple co-occurring conditions.

3. Expansion of bilingual clinicians both in-person and teletherapy clinicians to provide therapy and service coordination to non-English-speaking families.

4. Provision and encouragement of early intervention services for children with mild to moderate hearing loss.

Additionally, the vast majority of children in our sample experienced vocabulary delays

(relative to hearing peers), and studies of spoken vocabulary development in older DHH children suggest that they may not catch up (Lund, 2016). This should set clinicians and educators on high alert, due to the demonstrated importance of vocabulary skills in literacy (Biemiller, 2003; Hemphill & Tivnan, 2008; Stæhr, 2008) and in education more broadly (e.g., Young, 2005; Monroe & Orme, 2002). As early intervention predicts vocabulary outcomes in study after study (including this present study and e.g., Vohr et al., 2008, 2011; Ching et al., 2018, 2013; Holzinger et al., 2011; Watkin et al., 2007), ensuring intervention by 6 months for all DHH children may be one way to address spoken vocabulary deficits. Another solution: even prior to intervention or amplification, provision of structured, accessible language input (i.e., sign language) may mitigate negative effects of auditory deprivation on language skills (Davidson, Lillo-Martin, & Pichler, 2014; Hassanzadeh, 2012; Spellun & Kushalnagar, 2018). Indeed, while we recognize that learning sign language may pose a challenge for some families for myriad reasons, and as noted above, our sample did not use sign language, we nevertheless feel it is worth underscoring as an important language support for DHH children and their families.

In recommending sign language, we endorse the rationale set forth by Hall, Hall, and Caselli (2019). Summarizing their view, they note that spoken language outcomes for DHH children are variable and unpredictable (Ganek, McConkey Robbins, & Niparko, 2012; Szagun & Schramm, 2016), and even in optimal situations, many DHH children do not achieve age-appropriate spoken language outcomes (e.g., Geers et al., 2017). Failing to achieve language proficiency (in any language) confers higher risk of disrupted cognitive, academic, and socioemotional development (Amraei, Amirsalari, & Ajalloueyan, 2017; Dammeyer, 2010; Desselle, 1994; Hall et al., 2017; Hrastinski & Wilbur, 2016; Kushalnagar et al., 2011; Moeller & Schick, 2006; Preisler, Tvingstedt, & Ahlström, 2002; Schick, De Villiers, De Villiers, & Hoffmeister, 2007). The available data do not suggest that sign language harms spoken language development (Davidson et al., 2014; Park et al., 2013), and in fact, some studies suggest that sign language *benefits* spoken language development (e.g.,

Hassanzadeh, 2012). Providing early access to a natural sign language offers children another path to language mastery, and use of sign language does not preclude learning spoken language. Thus, we encourage sign language use *at least* prior to mastery of spoken language, and when possible for the family, we encourage its continued use as a language resource.

Limitations and Opportunities for Future Work

This study represents an important first step in quantifying variability in demographic characteristics, language outcomes, and 1-3-6 attainment. At the same time, it is exploratory, has limited geographic scope, and analyzed data from a (deliberately) high-variability sample. We see these limitations as opportunities for future investigation into the complex factors influencing DHH children's outcomes.

Given our exploratory analyses, there were many possible analytic routes. That said, our results largely converge with or replicate key aspects of past studies (e.g., Ching et al., 2013) and received wisdom among clinicians. In the interest of transparency, these data and all code generating our results are available on our OSF page (XXX) and we encourage those interested to explore further analyses.

This sample is composed only of children in North Carolina, and certain factors vary by country and by state (e.g., diagnosis and early intervention practices; NAD, n.d.). However, based on other demographic research (Blackorby & Knokey, 2006; Institute, 2014), our sample largely resembles the national DHH population in terms of degree of hearing loss, percentage of children with additional disabilities, cochlear implant and hearing aid use, language background, and gender. We would exercise caution in applying these results to regions where sign language access for DHH children is more common (e.g. Washington D.C.) A similar naturalistic study in those regions could help illuminate the effects of different clinical and demographic factors in a signing population.

Finally, the considerable variability in the sample did not allow us to easily isolate

effects of different factors. However, as discussed above, this reflects real-world variability

that is often does not make sense to isolate. Instead, this limitation would be best addressed

by larger sample sizes. As researchers continue to study influences on vocabulary in DHH

children, a meta-analytic approach too may be able to better estimate effects and effect sizes

within the varied outcomes of this heterogeneous population.

Conclusion

The present study explored demographic and audiological characteristics, vocabulary

outcomes, and clinical milestones within a diverse sample of 100 DHH children enrolled in

early intervention services. We found that overall, this sample showed spoken language

vocabulary delays relative to hearing peers on average and room for improvement in rates of

early diagnosis and intervention. Critically, we also found that the variables predicting these

delays in both vocabulary and early support services reflected *both* dimensions that are

immutable, and those that clinicians and caretakers can potentially alter. This in turn

highlights potential paths forward in ensuring that regardless of hearing status, we are able to

provide language access and early childhood support to help children attain their potential.

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Table 1

For each version of the CDI (WG = Words and Gestures; WS = Words and Sentences), the table shows the mean(SD) age, comprehension score, and production score of participants in our sample, along with the percent diagnosed with developmental delays.

CDI version	Mean Age (SD)	Mean Comprehension (SD)	Mean Production (SD)	% Developmental Delays
WG (n = 74)	20 (8.8) months	105 (100) words	32 (53) words	18.9%
WS (n = 24)	26 (7.8) months	NA	149 (180) words	4.2%

Table 2

Additional Diagnoses (n = 39): Ns of participants in our sample diagnosed with other conditions. N.B.: Ns do not sum to total because many participants had multiple diagnoses.

Condition	Specific Condition	n
Premature		17
	Extremely Premature	11
	NICU stay	16
Health Issues		36
	Heart	9
	Lung	5
	Illness	15
	Feeding Issues	14
	Pregnancy/Birth Complications	11
	Musculoskeletal	9
	Cleft Lip/Palate	4
	Other	15
Developmental Concerns		17
	Down Syndrome	5
	Chromosomal Issues	2
	Neural Tube Defects	2
	Other	10
Vision Loss		5
	Retinopathy of Prematurity	1
	Nearsightedness	1
	Farsightedness	1
	Cortical Visual Impairment	1

Table 3

Audiological Characteristics of the Sample: First two columns describe laterality and amplification type (cochlear implant (CI), hearing aid (HA), or none). Mean decibels of hearing loss (HL) in better ear, worse ear, and the mean age (in months) of amplification, and cochlear implantation (when applicable) for each laterality and amplification combination.

Laterality	Amplification	mean_HLbetter	mean_HLworse	mean_age_amplification	mean_age_implantation
Bilateral	CI	85.60	89.79	11.29	14.12
Bilateral	HA	47.02	55.57	8.28	NaN
Bilateral	none	49.67	53.65	NaN	NaN
Unilateral	HA	4.70	56.04	10.91	NaN
Unilateral	none	2.50	73.90	8.50	NaN

Table 4

Language and Communication Characteristics of the Sample: Ns of participants by language background and communication method.

Communication	English	Hindi	Spanish	Total
cued speech	1	0	0	1
spoken	68	1	10	79
total communication	15	0	3	18

Table 5

Detailed information about the variables studied. For categorical variables, levels are described. For continuous variables, range, mean, and standard deviation are provided.

Variable	Scale	Range
Age	Continuous	4-36 months (mean (SD): 21 (9))
Age at Amplification	Continuous	2-30 months (mean (SD): 9 (7))
Age at Diagnosis	Continuous	0-30 months (mean (SD): 5 (7))
Age at Implantation	Continuous	7-32 months (mean (SD): 14 (7))
Age at Intervention	Continuous	1-33 months (mean (SD): 11 (9))
Amplification	Categorical	Hearing Aid / Cochlear Implant / None
Communication	Categorical	Spoken / Total Communication / Cued Speech
Degree Hearing Loss (worse ear)	Continuous	18-100 dB HL (mean (SD): 64 (24))
Developmental Delay	Categorical	Yes / No
Gender	Categorical	Female / Male
Health Issues	Categorical	Yes / No
Language in Home	Categorical	English / Other
Laterality	Categorical	Unilateral / Bilateral
1-3-6	Categorical	Yes / No
Premature Birth	Categorical	Full-term / Premature
Services Per Month	Continuous	0-43 services per month (mean (SD): 6 (6))
Etiology	Categorical	Sensorineural / Conductive / Mixed
CDI - Words Produced	Continuous	0-635 words (mean (SD): 61 (111))

Table 6

Unstandardized beta weights (months of vocabulary delay) for the model of vocabulary delay selected by AIC.

term	estimate	std.error	statistic	p.value
(Intercept)	-0.54	1.71	-0.31	0.76
Laterality (Unilateral)	-2.70	1.14	-2.37	0.02
Amplification (Cochlear Implant)	-3.50	1.49	-2.36	0.02
Amplification (Hearing Aid)	-3.84	1.13	-3.38	0.00
Age	0.55	0.06	9.83	0.00

Table 7

Unstandardized beta coefficients (months) for the model of age at diagnosis selected by AIC.

term	estimate	std.error	statistic	p.value
(Intercept)	9.38	1.97	4.77	0.00
Health Issues (yes)	3.70	1.42	2.61	0.01
Language Background (English)	-6.47	1.96	-3.31	0.00
Laterality (Unilateral)	-2.15	1.58	-1.36	0.18

Table 8

Unstandardized beta coefficients (months) for the model of age at intervention selected by AIC.

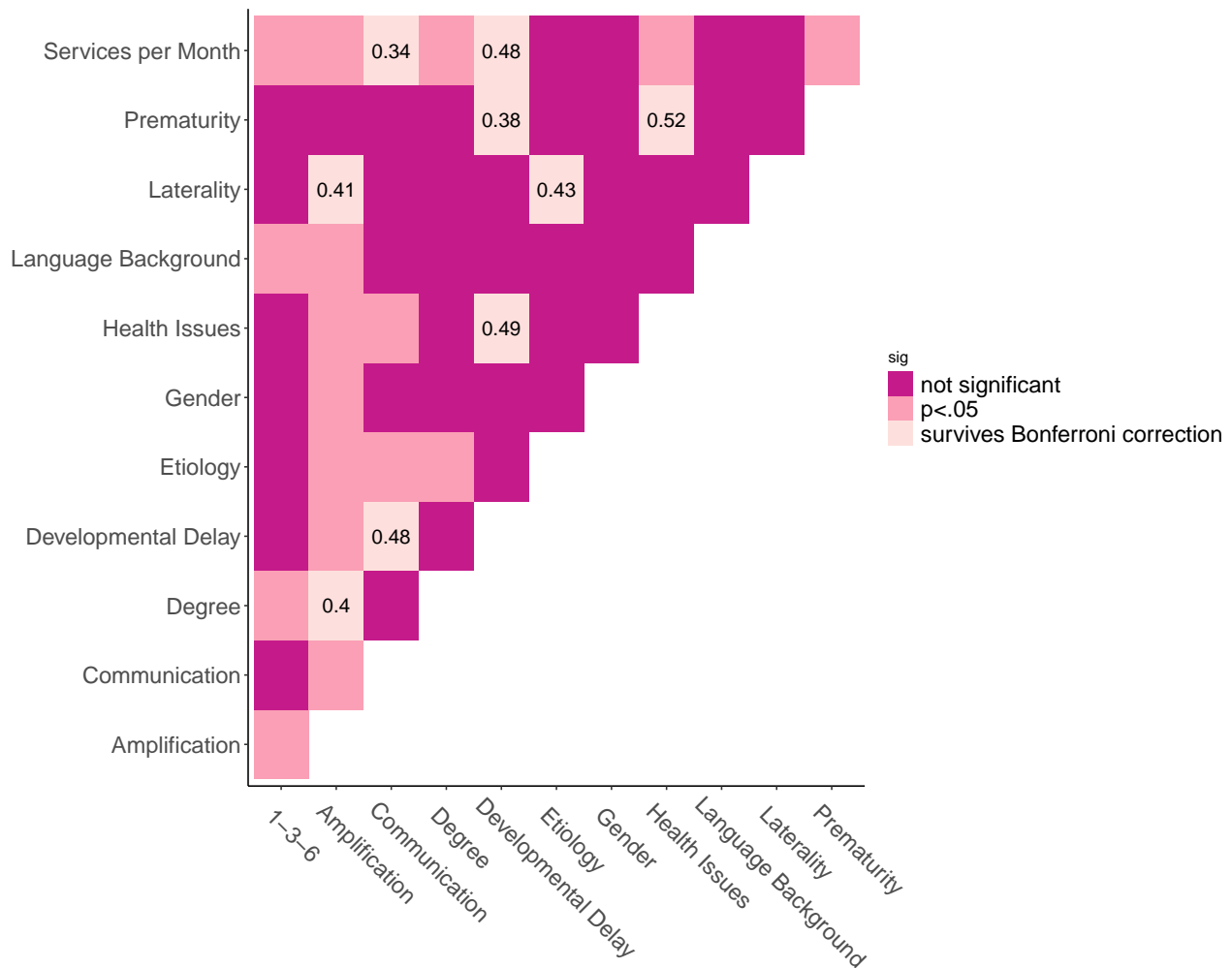| term	estimate	std.error	statistic	p.value
(Intercept)	14.65	2.87	5.10	0.00
Degree of Hearing Loss	-0.09	0.03	-3.06	0.00
Premature (yes)	3.78	1.95	1.94	0.06
Age at Diagnosis	0.65	0.10	6.24	0.00
Language Background (English)	-1.38	2.12	-0.65	0.52

Figure 1. Results of chi-square tests between variables. X- and y-axes show the variables compared. Color of the square represents significance of the corresponding chi-square test. For tests that survived Bonferroni correction (p<.0007), effect size (Cramer's V) is given.
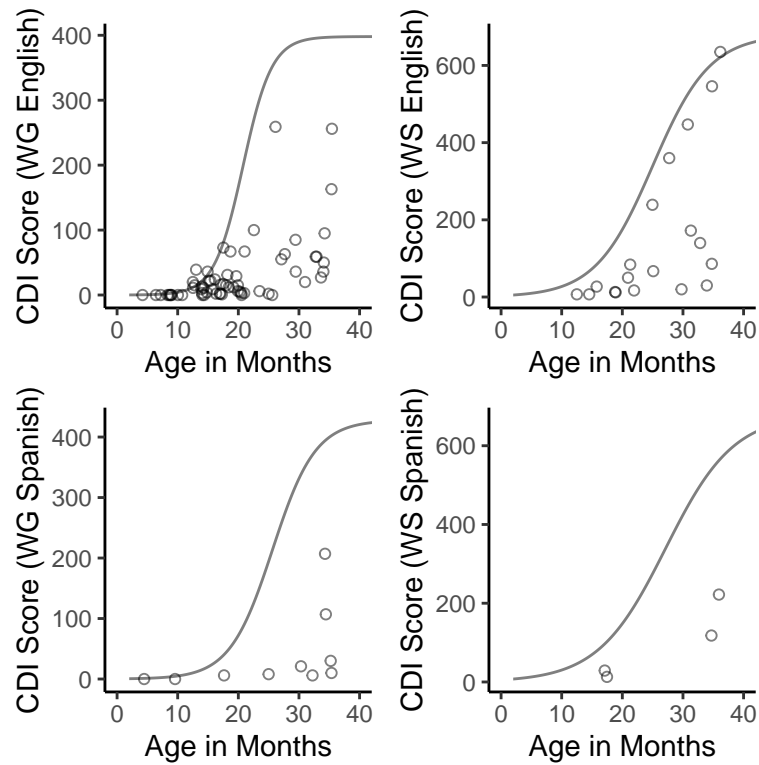
Figure 2. Lines show the growth curves created from Wordbank 50th percentile data. Left panels show Words & Gestures; right panels show Words & Sentences. Top row is American English data; bottom row is Mexican Spanish data. Dots represent vocabulary scores of individual DHH children in the sample.
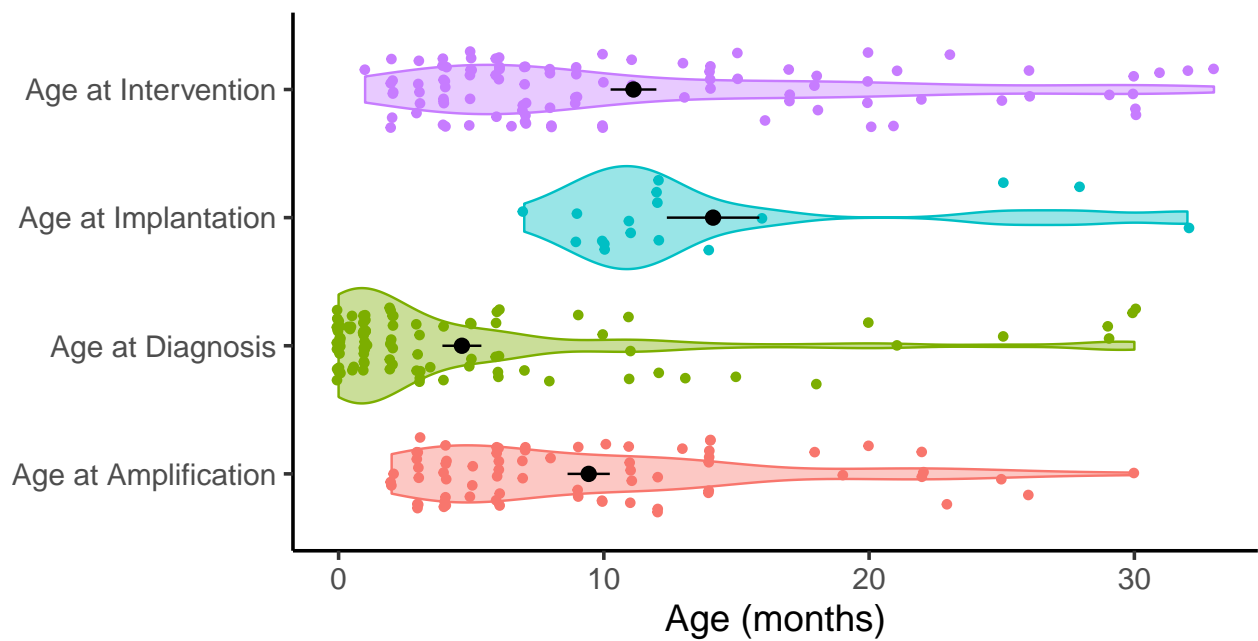
Figure 3. Age at diagnosis, intervention, amplification, and cochlear implantation across participants. Each dot represents the age that one child received the clinical service; violin width reflects data distribution. Blacks dot and whiskers show means and standard errors. Not all children received amplification (hearing aids) or implantation (cochlear implants)
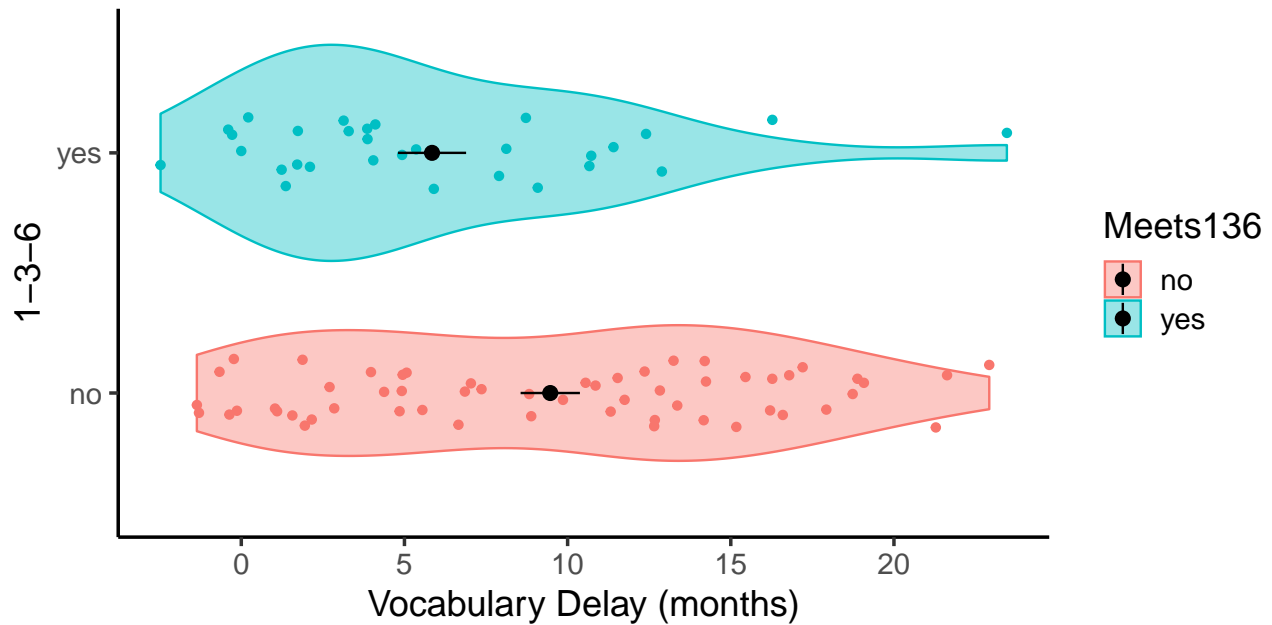
Figure 4. Estimated vocabulary delay for children who meet 1-3-6 guidelines for diagnosis/intervention (top) and children who do not (bottom). Each dot represents one child in the sample; violin width reflect data distribution. Blacks dot and whiskers show means and standard error.
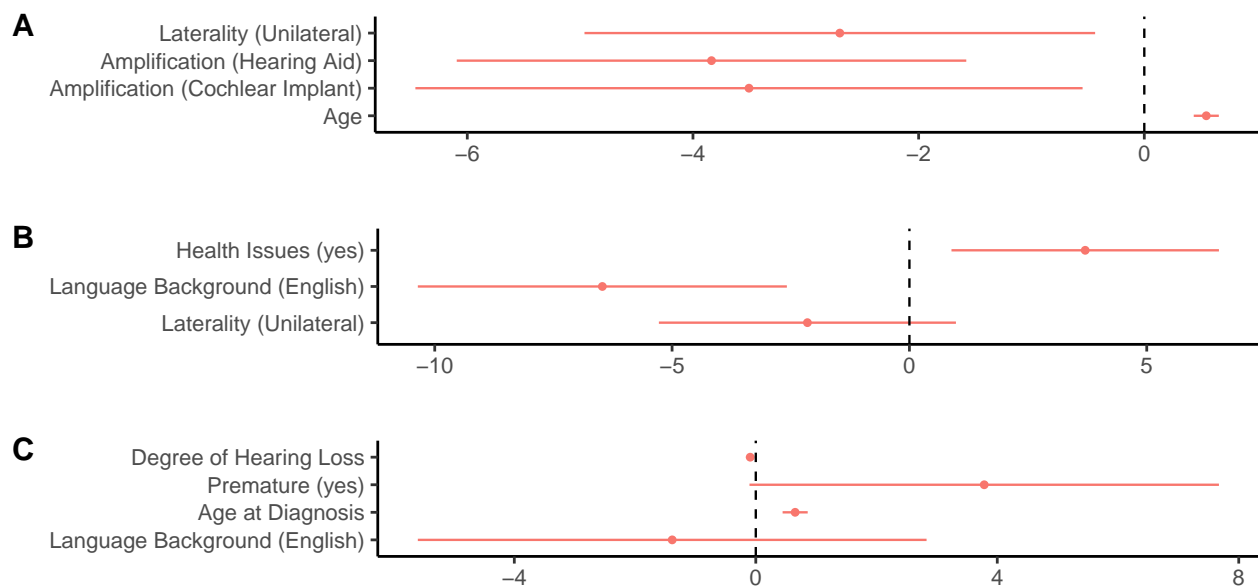

Figure 5. Unstandardized coefficients (measured in months) with 95% confidence intervals for the models selected by AIC for: (A) vocabulary delay, (B) age at diagnosis, (C) age at intervention.