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- 1 Characterizing North Carolina's Deaf/Hard-of-Hearing Infants and Toddlers: Predictors of
- Vocabulary, Diagnosis, and Intervention
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Introduction

In the United States, 1-2 children are born with hearing loss, per 1,000 births (CDC, 8 2018). This translates to 114,000 Deaf or Hard of Hearing (DHH) children born in the U.S. per year (Martin, Hamilton, Osterman, & Driscoll, 2019). Of these 114,000, ~90\% will be 10 born to hearing parents (Mitchell & Karchmer, 2004), in a home where spoken language is 11 likely the dominant communication method. Depending on the type and degree of hearing 12 loss and whether the child uses amplification, spoken linguistic input will be partially or 13 totally inaccessible. Some of these children will develop spoken language proficiency within 14 the range of their hearing peers (Geers, Mitchell, Warner-Czyz, Wang, & Eisenberg, 2017; 15 Verhaert, Willems, Van Kerschaver, & Desloovere, 2008), but many will face persistent spoken language deficits (Eisenberg, 2007; Luckner & Cooke, 2010; Moeller, Tomblin, Yoshinaga-Itano, Connor, & Jerger, 2007; Sarchet et al., 2014), which may later affect reading ability (Kyle & Harris, 2010) and academic achievement (Karchmer & Mitchell, 2003; Qi & Mitchell, 2012).

Despite many excellent studies examining language development in DHH children,
there is still a gap in the literature describing and analyzing spoken language development
across the full range of children receiving services for hearing loss, with many studies
focusing in on specific subgroups (e.g. children under age X with Y level of hearing loss and
Z amplification approach, e.g. Vohr et al., 2008; Yoshinaga-Itano, Sedey, Wiggin, & Mason,
2018). In what follows, we first summarize the previous literature on predictors of spoken
language outcomes in DHH children. We then provide a brief overview of a common
vocabulary measure used in the current study, the MacArthur-Bates Communicative
Development Inventory (CDI). Finally, we turn to an empirical analysis of early vocabulary
in a wide range of young children receiving state services in North Carolina. We have two

broad goals in what follows. First, we aim to provide a comprehensive description of a

- heterogeneous group 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<sup>1</sup>, with the broader goal of considering the success of early
- 35 diagnosis and intervention initiatives.

## 36 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, &
- 51 Lyons, 1991), and stronger grammatical and phonological skills (Lange, Euler, & Zaretsky,

<sup>&</sup>lt;sup>1</sup> 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.

2016; Özçalışkan & Goldin-Meadow, 2010). This finding appears to be consistent across
 studies (Wallentin, 2009), various spoken languages (Frank, Braginsky, Marchman, &
 Yurovsky, 2019), and gesture (Özçalışkan & Goldin-Meadow, 2010).

The DHH literature presents a more mixed (though rather understudied) picture. On one hand, DHH girls, like hearing girls, have been found to have a larger spoken vocabulary than DHH boys (Ching et al., 2013; Kiese-Himmel & Ohlwein, 2002). However, in contrast to their hearing peers, DHH children do not seem to show a gender-based difference for some aspects of syntactic development (Pahlavannezhad & Tayarani Niknezhad, 2014).

Comorbidities. Additional co-morbid disabilities occur frequently in the DHH population, perhaps as much as three times more than in the hearing population (Pollack, 1997). Incidence estimates for co-occurring disabilities in DHH children range from 25-51% (Bruce & Borders, 2015; Guardino, 2008; Holden-Pitt & Diaz, 1998; Luckner & Carter, 2001; Picard, 2004; Schildroth & Hotto, 1996; Soukup & Feinstein, 2007), with approximately 8% of DHH children living with 2 or more co-occurring disabilities (Schildroth & Hotto, 1996).

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

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

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

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

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

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

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

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

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

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

al., 2007).

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

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(defined as < 37 weeks gestational age). Compared to an incidence of 0.2% in full-term 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 82 delay and disorder (Barre, Morgan, Doyle, & Anderson, 2011; Carter & Msall, 2017; Cusson, 83 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 (NBHS) 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 93 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 104 speech perception, sound localization, and loudness perception in quiet and noisy 105 environments (Ching, Van Wanrooy, & Dillon, 2007). The literature on hearing aids and 106 cochlear implants also points to benefits for bilateral auditory input (Lovett, Kitterick, 107 Hewitt, & Summerfield, 2010; Sarant, Harris, Bennet, & Bant, 2014; Smulders et al., 2016). 108 At school-age, 3-6% of children have unilateral hearing loss (Ross, Visser, Holstrum, Qin, & 109 Kenneson, 2010). Although children with unilateral hearing loss have one "good ear," even 110 mild unilateral hearing loss has been tied to higher risk of language delays and educational 111 challenges relative to hearing children (Kiese-Himmel, 2002; Lieu, 2004, 2013; Lieu, 112 Tye-Murray, & Fu, 2012; Vila & Lieu, 2015). Just as in the bilateral case, more severe 113 hearing loss leads to greater deficits in spoken language and educational outcomes for 114 children with unilateral hearing loss (Anne, Lieu, & Cohen, 2017; Lieu, 2013).

Many DHH children receive hearing aids (HAs) or cochlear implants (CIs) to boost access to the aural world. These devices have been associated with better speech perception and spoken language outcomes (Niparko et al., 2010; Walker et al., 2015; Waltzman et al., 1997). In turn, aided audibility predicts lexical abilities in children with HAs (Stiles, Bentler, & McGregor, 2012).

For both hearing aids and cochlear implants, earlier fit leads to better spoken language 121 skills, if the amplification is effective. For hearing aids, some studies find that children with 122 milder hearing loss who receive hearing aids earlier have better early language achievement 123 than children who are fit with hearing aids later (Tomblin et al., 2015), but this finding does 124 not hold for children with severe-to-profound hearing loss (Kiese-Himmel, 2002; Watkin et 125 al., 2007) (for whom hearing aids are generally ineffective). Analogously, children who are 126 eligible and receive cochlear implants earlier have better speech perception and spoken 127 language outcomes than those implanted later (Artières, Vieu, Mondain, Uziel, & Venail, 128 2009; Dettman, Pinder, Briggs, Dowell, & Leigh, 2007; Miyamoto, Hay-McCutcheon, Kirk, 129

Houston, & Bergeson-Dana, 2008; Svirsky, Teoh, & Neuburger, 2004; 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 140 have better speech intelligibility (Dillon, Burkholder, Cleary, & Pisoni, 2004; Geers et al., 141 2017; Geers, Spehar, & Sedey, 2002; Hodges, Dolan Ash, Balkany, Schloffman, & Butts, 142 1999) and auditory perception (Geers et al., 2017; O'Donoghue, Nikolopoulos, & Archbold, 143 2000). That said, there is some debate as to whether an oral approach facilitates higher 144 spoken language performance, or whether children who demonstrate aptitude for spoken 145 language are steered towards the oral approach rather than total communication (Hall, 146 Levin, & Anderson, 2017). 147

1-3-6 Guidelines. Early identification (Apuzzo & Yoshinaga-Itano, 1995; Kennedy 148 et al., 2006; Robinshaw, 1995; White & White, 1987; Yoshinaga-Itano, Sedey, Coulter, & 149 Mehl, 1998; Yoshinaga-Itano et al., 2018) and timely enrollment in early intervention 150 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 153 have been found to meet age-appropriate developmental outcomes, including language (Stika 154 et al., 2015). In line with these findings, the American Academy of Pediatricians (AAP) has 155 set an initiative for Early Hearing Detection and Intervention (EHDI). Their EHDI 156

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 162 2010 (Capps, 2009) was passed to develop state-wide systems for screening, evaluation, 163 diagnosis, and "appropriate education, audiological, medical interventions for children 164 identified with hearing loss," but policies for early diagnosis and intervention vary by state. 165 As of 2011, 36 states (including North Carolina, ("15A NCAC 21F .1201 - .1204," 2000)] 166 mandate universal newborn hearing screening (UNHS; National Conference of State 167 Legislatures, 2011). All states have some form of early intervention programs that children 168 with hearing loss can access (NAD, n.d.), but the specifics vary state-by-state. For instance, 169 half of the states in the US do not consider mild hearing loss an eligibility criterion for early 170 intervention (Holstrum, Gaffney, Gravel, Oyler, & Ross, 2008). 171

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 182 research and applied settings. The Words and Gestures version of the form is normed for 183 8–18-month-olds. On Words and Gestures, parents indicate whether their child understands 184 and/or produces each of the 398 vocabulary items, and answer questions about young 185 children's early communicative milestones. The Words and Sentences version of the form is 186 normed for 16-30-month-olds. On Words and Sentences, parents indicate whether their child 187 produces each of the 680 vocabulary items, and answer some questions about grammatical 188 development. The CDI has been normed on a large set of participants across many 189 languages (Anderson & Reilly, 2002; Frank et al., 2017; Jackson-Maldonado et al., 2003). 190

The CDI has also been validated for DHH children with cochlear implants (Thal, 191 Desjardin, & Eisenberg, 2007). More specifically, in this validation, researchers asked parents 192 to complete the CDI, administered the Reynell Developmental Language Scales, and 193 collected a spontaneous speech sample. All comparisons between the CDI and the other 194 measures yielded significant correlations ranging from 0.58 to 0.93. Critically, the children in 195 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, 197 Castellanos, Pisoni, Kronenberger, and Beer (2016) find that in children with CIs, number of 198 words produced on the CDI predicts language, executive function, and academic skills up to 190 16 years later. Building on this work, several studies have used the CDI to measure 200 vocabulary development in DHH children (Yoshinaga-Itano et al. (2017); Yoshinaga-Itano et 201 al. (2018); de Diego-Lázaro et al. (2018); Vohr et al. (2008); Vohr et al. (2011); summarized 202 in Table 1). We build on this literature in our analyses below. 203

### 204 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 had reason to expect that many of these variables would be 212 related, due to known causal relations (e.g., cochlear implants recommended for severe 213 hearing loss, but not mild hearing loss). We sought to provide descriptive documentation 214 about the distrubution of demographic, audiological, and intervention characteristics in a 215 diverse sample of DHH children receiving state services. For the second, we hypothesized 216 that male gender, more severe degree of hearing loss, bilateral hearing loss, no amplification 217 use, premature birth, and presence of additional disabilities would predict larger spoken 218 vocabulary delay than female gender, less severe degree of hearing loss, unilateral hearing 219 loss, amplification use, full term birth, and lack of additional disabilities, respectively. We 220 did not have strong predictions regarding the effects of communication method or presence 221 of other health issues (e.g., congenital heart malformation) on vocabulary. For the third goal, based on the prior literatal summarized above, we hypothesized that children with less residual hearing (i.e., bilateral, more severe) and no co-occurring conditions would be earlier 224 diagnosed and earlier to begin language services, and that earlier diagnosis would predict 225 earlier intervention. 226

227 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

234 Carolina.

The clinical evaluations included demographic and audiological information, CDI vocabulary scores, and the results of any clinical assessments administered (e.g., PPVT), detailed further below. For some children (n=47), multiple evaluations were available from different timepoints. In these cases, only the first evaluation was considered for this study, due to concerns regarding within-subjects variance for statistical analysis.

While this collaboration is ongoing, we opted to pause for this analysis upon receiving 240 data from 100 children. Thus, the reported sample below consists of 100 children (56 male / 241 44 female) ages 4.20–36.17 months (M=21.21, SD=9.08). Race and socioeconomic 242 information were not available. Families were administered either the Words and Gestures or 243 Words and Sentences version of the CDI based on clinician judgement. Children who were 244 too old for Words and Gestures, but who were not producing many words at the time of 245 assessment, were often given Words and Gestures (n=37). Families for whom Spanish was 246 the primary language (n=14) completed the Spanish language version of the CDI 247 (Jackson-Maldonado et al., 2003). 248

With regard to comorbid diagnoses, children in this sample were coded as yes/no for cognitive development concerns (e.g., Down syndrome, global developmental delays; Cornelia de Lange syndrome), yes/no for premature birth (i.e., more than 3 weeks premature), yes/no for health issues (e.g., heart defects, kidney malformations, VACTERL association), and yes/no for vision loss (not corrected to normal by surgery or glasses); see Table 3.

Degree of hearing loss was most often reported with a written description (e.g., "mild sloping to moderate" or "profound high frequency loss"). We created 3 variables: hearing loss in the better ear, hearing loss in the worse ear, and average hearing loss (average of better and worse ear). For the analyses below, we primarily use hearing loss in the worse ear to avoid any redundancies with laterality. Using the ASHA hearing loss guidelines, each of

these hearing loss measures was coded with the decibels of hearing loss (dB HL) 259 corresponding with the median dB HL for the level of hearing loss (e.g., moderate hearing 260 loss was coded as 48 dB HL), and sloping hearing loss was coded as the average of the levels 261 (e.g. mild to moderate was coded as 40.5 dB HL). Participants were also coded for unilateral 262 or bilateral hearing loss; presence or absence of Auditory Neuropathy Spectrum Disorder; 263 and etiology of hearing loss (sensorineural, conductive, or mixed). Amplification was 264 recorded as the device the child used at the time of assessment: either hearing aid, cochlear 265 implant, or none. See Table 4 for audiological characteristics of the sample. 266

Communication method was recorded as spoken language, total communication, or 267 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 269 American Sign Language or another signed language. The forms also listed the primary 270 language spoken at home, which we binned into English-speaking and non-English-speaking. 271 85% of families spoke English, and 14% spoke Spanish. For one child, who was adopted from 272 a non-English-speaking country after their second birthday, we recorded the language 273 background as non-English-speaking, although the child's adoptive parents are 274 English-speaking, because the child had lived most of her life in a non-English-speaking 275 environment. Language and communication information is summarized in Table 5. 276

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% NBHS 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" and "service enrollment by 6 months" criteria; see Table 6. Finally, we also calculated the number of hours of early intervention services received per month (including service coordination, speech therapy, and occupational therapy, among others) based on the clinician report.

291 Results

We split the results into three parts. In the first, we explore relationships among child demographic, audiological, and clinical variables. In the second, we use these variables to predict vocabulary development. Finally, in the third, we describe the implementation of the EHDI 1-3-6 guidelines and predictors of early diagnosis and intervention in this sample. All analyses were conducted in R. All code is available on Github.

# Part I: Interactions Among Variables

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Before we test how these variables may be related to vocabulary, we describe their 298 relationships to each other. As would be expected, many health, audiological, and clinical 290 characteristics are not distributed randomly across this sample of children. To quantify this 300 statistically, we used Bonferroni-corrected chi-square tests between each of our variables 301 (gender (male/female), laterality (bi-/uni-lateral hearing loss), health issues (yes/no), 302 developmental delays (yes/no), premature birth (yes/no), language background 303 (English/non-English), 1-3-6 (yes/no), degree of hearing loss (mild, moderate, 304 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)). Because the chi-square statistic assumes n > 5 is expected in the majority of the cells for 308 each test (preferably  $\geq 80\%$  McHugh, 2013), we excluded mixed hearing loss (n=8) and cued 309 speech (n=1) from this section of the analysis. Strictly speaking, some of these variables are 310

not expected to be randomly distributed relative to each other (e.g., premature birth and health issues; degree and amplification), but quantifying the differences via chi-square using 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 319 were highly interrelated in our sample, such that children born premature were more likely 320 to also experience health issues  $(X^2 (1, N = 98) = 23.9, p < .0001)$  and developmental 321 delays  $(X^2 (1, N = 98) = 11.63, p = .0006)$ , and children with developmental delays were 322 more likely to also experience health issues ( $X^2$  (1, N = 98) = 20.87, p < .0001). Children 323 with developmental delays received more services per month than typically-developing 324 children  $(X^2 (2, N = 95) = 22.17, p < .0001)$  and were more likely to use total 325 communication  $(X^2 (2, N = 98) = 22.51, p < .0001)$ . Likewise, children who used total 326 communication received more services per month than children using spoken language ( $X^2$ 327 (4, N = 95) = 21.35, p = .0003).328

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<sup>2</sup>. Chi-square tests showed that laterality ( $X^2$  (2, N = 98) = 16.43, p = .0003) and degree of hearing loss ( $X^2$  (4, N = 87) = 28.45, p < .0001) were related to

<sup>&</sup>lt;sup>2</sup> All children with mixed hearing loss (n=8) had bilateral hearing loss.

amplification in our sample. Children with bilateral hearing loss were more likely than
children with unilateral hearing loss to use a hearing aid or cochlear 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).

# Part II: Influence on vocabulary

We next turn to the relationship between each of these variables and children's productive vocabulary, as measured by the CDI. Figures 2 & 3 show the vocabulary scores of children in our samples relative to norms for hearing children for English and Spanish respectively. 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 <sup>3</sup> (Frank et al., 2017) to create binary logistic growth curves separately for the Words

 $<sup>^{3} \</sup> n(WG-English) = 526932, \ n(WG-Spanish) = 399000, \ n(WS-English) = 1164417, \ n(WS-Spanish) = 815724817, \ n(WS-Spanish) = 1164417, \ n(WS-Spanish$ 

and Gestures and Words and Sentences versions of the CDI for American English and 359 Mexican Spanish. For each child, we took the number of words they produced divided by the 360 number of words on the instrument, to give us the proportion of words produced. We used 361 this proportion in an inverse prediction from the binary logistic regression curves to generate 362 a predicted age. That is, for each possible CDI score, the growth curve provided the age that 363 the score would be achieved for the 50th percentile trajectory. Finally, we subtracted the 364 predicted age from each child's chronological age to calculate their vocabulary delay. 365 However, for children producing 0 words, this approach was not appropriate due to the long 366 tails on the growth curves. Thus, for this subset of children, we took the x-intercept from 367 Wordbank (8 months for English, and 9 months for Spanish), and subtracted that value from 368 the child's chronological age to get their vocabulary delay. 369

To look at the relationship between our predictor variables and CDI scores, we next conducted multiple linear regression, using vocabulary delay as our outcome variable. <sup>4</sup>

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

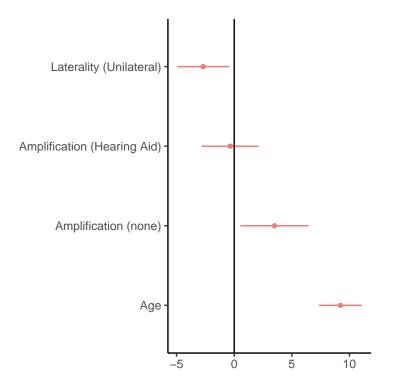
Developmental Delay + Health Issues + Premature Birth + Laterality + Degree +

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

Background.

This model accounted for significant variance in vocabulary delay (adjusted- $R^2 = 0.59$ , p < .001; see Figure 4). We next performed stepwise model comparison using stepAIC (MASS) to pare down the model. This process selected only the predictors which incrementally improved model fit, measured by Akaike's Information Criterion (AIC), which considers goodness of fit and model complexity (penalizing models with many predictors).

<sup>&</sup>lt;sup>4</sup> We excluded the adopted child from this section of the analysis due to concerns about comparing her score to the American English CDI norms.



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Based on this iterative process, we removed several predictors from the model, leaving 382 the following final model: Vocabulary Delay  $\sim$  Age + Laterality + Amplification. This 383 model accounted for significant variance in children's vocabulary delay to a nearly identical 384 degree as the full model (adjusted- $R^2 = 0.58$ , p = < .001). We found significant main effects 385 for Age (ß = 0.55, p < .01), Amplification (Hearing Aid: (ß = -0.34, p = .79); No 386 amplification: ( $\beta = 3.49$ , p = .02)), and Laterality ( $\beta = -2.69$ , p = .02), such that older age, 387 no amplification, and bilateral hearing loss predicted greater vocabulary delays. Given our 388 results in Part I revealing relationships exist among several of these variables (e.g., laterality 389 and amplification), we tested for collinearity concerns by computing the model's VIF (variance inflation factor). This revealed low levels of collinearity among predictors in our final model (all VIF < 1.5; see Table ??; James, Witten, Hastie, & Tibshirani, 2013). In 392 sum, the analyses in this section revealed that over half of the variance in DHH's children's 393 vocabulary scores was explained by their age, whether their receive amplification, and 394 whether their hearing loss was unilateral or bilateral. 395

### Part III: 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 (see Table 2). 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 5 shows the age at first diagnosis, intervention, amplification, and implantation for each child in our sample.

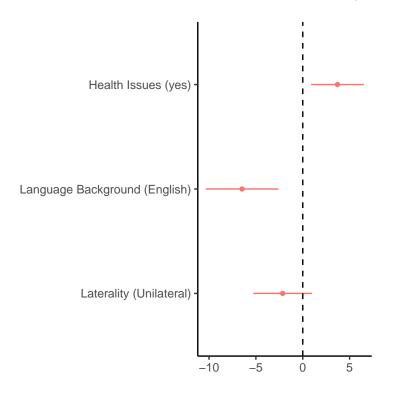
We first tested the link between 1-3-6 and vocabulary directly in an exploratory analysis. An independent samples t-test showed that children who did not meet 1-3-6 guidelines had significantly larger vocabulary delays than children who met 1-3-6 guidelines (t(68.78)=2.62, p=0.01; see Figure 6).

To better understand implementation of 1-3-6 guidelines, we next zoomed in on diagnosis and intervention. We conducted two linear regressions, one for age at diagnosis and one for age at intervention, considering only the predictors that would have been available or relevant at each of these stages (as detailed below). Model selection followed the same

stepwise AIC-based process as Part II.

For age at diagnosis, we included the set of child-specific factors that would be relevant before diagnosis of hearing loss (e.g., we excluded amplification type because a child would not receive a hearing aid or cochlear implant prior to being diagnosed with hearing loss.) We began with: gender, degree, developmental delay, health issues, prematurity, laterality, language background, and etiology.

The best fit model was: Age at Diagnosis  $\sim$  Health Issues + Language Background + Laterality, with significant main effects of Health Issues and Language Background. This model accounted for 16.41% of the variance in age at diagnosis (p = .001). Average age at diagnosis for English-speaking children with bilateral hearing loss and no health issues was 2.91 months. Relative to English-speaking families, children from Spanish-speaking families were diagnosed 6.47 months later (p = .001). Children with health issues were diagnosed 3.70 months later than children without health issues (p = .01).

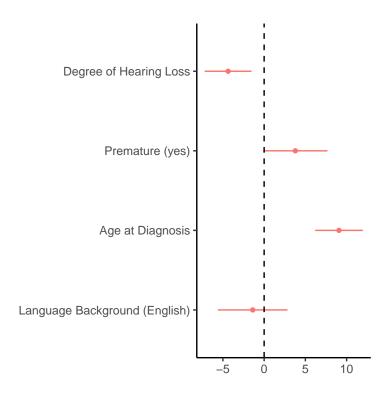


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We repeated this model selection process for age at intervention. In addition to the

variables used to fit the intervention model, we included age at diagnosis. The best fit model was: Age at Intervention ~ Premature Birth + Degree + Age at Diagnosis + Language Background ( $R^2$ =0.43 , p < .001), with significant main effects of degree and age at diagnosis. Less severe hearing loss ( $\beta$  = -0.09, p < .01) and later diagnosis ( $\beta$  = 0.65, p < .01), and coming from a non-English-speaking household ( $\beta$  = -1.38, p = .52) predicted later intervention. Prematurity and language background were not significant predictors on their own, but their inclusion improved model fit.



444 Discussion

443

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. This analysis was exploratory and descriptive, and the results should be interpreted accordingly.

#### 450 How are child-level variables intertwined?

In our sample, we found significant overlap among demographic, audiological, and 451 clinical variables. Prematurity, health issues, and developmental delay frequently co-occurred, 452 such that there was a moderate relationship between each of these variables (Cramer's V = 453 0.38 - 0.52, p < .0007). Children with one of these conditions (prematurity, developmental 454 delay, health issues) were more likely to have any other condition. This is not surprising. 455 Many conditions that cause developmental delays have a high incidence of health issues (e.g., 456 heart problems in Down Syndrome; vomiting and seizures with hydrocephalus), and it is well 457 documented that there is a higher incidence of developmental delay in preterm infants 458 (Aarnoudse-Moens, Weisglas-Kuperus, van Goudoever, & Oosterlaan, 2009; Pierrat et al., 2017). Children born premature, especially those born extremely premature, are at increased 460 risk for a number of health issues at birth (Costeloe et al., 2012; Robertson et al., 2009; York & DeVoe, 2002) and throughout the lifespan (Luu, Katz, Leeson, Thébaud, & Nuyt, 2016).

Each of these conditions may affect language and development in different ways. The 463 literature points to increased risk of language delay for children with developmental delays 464 (Chapman, 1997; Kristoffersen, 2008; Weismer et al., 2010) and children born premature 465 (Foster-Cohen, Friesen, Champion, & Woodward, 2010), with differential effects based on the 466 nature of the child's diagnosis (Cupples et al., 2014, 2018). Together, these risks may 467 interact and multiply. In our sample, we also had a large range of health conditions (76 468 unique conditions in our sample of 100 children; see 3 and Appendix XXX for more detailed 460 information about comorbidities), and it appears probable that those conditions would vary 470 in whether and how they influence vocabulary growth.

We found that children with developmental delays (e.g., Down syndrome) were much more likely to use a total communication approach than typically-developing DHH children. Assignment to "spoken language" and "total communication" groups was not randomly distributed, with use of total communication appearing to follow 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
should temper the interpretation of correlational studies finding links between total
communication and language delays (e.g., Geers et al., 2017).

Additionally, in our sample, children with developmental delays were considerably more likely to receive >10 services per month, perhaps in response to increased need (or increased perceived need). The services per month variable also includes occupational therapy, physical therapy, which typically-developing DHH children may be unlikely to receive. Likewise, children who used total communication were more likely to receive frequent services.

We also found relationships among many of our audiological variables. In particular,
etiology and laterality were related, such that conductive hearing loss was more likely
unilateral, and sensorineural hearing loss was more likely bilateral. There were only seven
cases of mixed hearing loss, and all were bilateral. One possible explanation is that certain
underlying causes of conductive hearing loss (e.g., aural atresia, impacted cerumen, trauma
to the tympanic membrane) may be more likely to affect one ear than two.

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 in speech perception and quality of life for amplification for unilateral hearing loss (Briggs, Davidson, & Lieu, 2011; Hassepass et al., 2013; Priwin, Jönsson, Hultcrantz, & Granström, 2007; Winiger, Alexander, & Diefendorf, 2016) and spoken language vocabulary and grammar for mild hearing loss (Walker et al., 2015).

These results are not necessarily surprising, given causal links among some of the variables (e.g., increased health issues in children born premature). Nevertheless, it 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?

# 507 Predicting vocabulary outcomes

88.89% of DHH children in our sample fell below the 50th percentile for spoken vocabulary. 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). To have such a strong of majority DHH children 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).

In contrast to our predictions, the best model predicting vocabulary delay had just a 515 few variables: age, amplification, and laterality. Notably, we did not simply find that DHH 516 children were learning words at the same rate (albeit delayed) as hearing children, which 517 would have led to a constain delay across developmental time. Instead, we see that the 518 spoken vocabulary delay widens with age, indicating that the rate of spoken vocabulary 519 acquisition is slower for DHH children. The result is a population increasingly behind on 520 spoken language milestones, and given that none of the children here use sign language, on 521 language development more broadly. 522

An alternative explanation is that the age effect is related to age at intervention. We
analyzed the first evaluation available from each participant, which was often (but not
always) their first evaluation with the early intervention program. Perhaps children who

were older at first evaluation were more likely to have a wider delay. Exploring the longitudinal administrations would help us clarify whether age effect reflects an artifact of our sampling or an increasing delay with age.

In our sample, children without amplification were 2.15 months more delayed than 529 children with a cochlear implant. Vocabulary delay was not significantly different between 530 children with hearing aids and children with cochlear implants. This increased delay for 531 children without amplification could reflect both increased spoken language audibility and/or 532 a difference in demographic characteristics of amplification candidates / non-candidates. Of 533 the typically-developing children in our sample, 20.99% received cochlear implants, 55.56% of 534 children received hearing aids, and 23.46% received no amplification. Meanwhile, for children 535 with developmental concerns, 0\% received cochlear implants, 52.94\% received hearing aids, 536 and 47.06% received no amplification (p<.05, but does not survive Bonferroni correction; see 537 Figure 1). 538

In our model, children with bilateral hearing loss had an 3.70 months greater delay 539 than children with unilateral hearing loss. While few studies directly assess language 540 development differences between unilateral and bilateral hearing loss, we know that both bilateral and unilateral hearing loss have detrimental effects on spoken language development 542 (Kiese-Himmel, 2002; Lieu, 2004, 2013; Lieu et al., 2012; Vila & Lieu, 2015). Our model 543 results suggest that children with bilateral hearing loss are at a quantifiable disadvantage over children with unilateral hearing loss. This falls in line with research that finds better spoken language outcomes for individuals with more residual hearing (Anne et al., 2017; Lieu, 2013; Tomblin et al., 2015; Vohr et al., 2008). However even children with unilateral hearing loss, who have one ear with normal hearing, experience notable delays both in the literature (Kiese-Himmel, 2002; Lieu, 2004, 2013; Lieu et al., 2012; Vila & Lieu, 2015) and in 549 our sample (Mean delay  $\{\text{unilateral}\} = 7.18$ ). 550

### Predicting early diagnosis and intervention

Our findings from North Carolina parallel AAP's national findings: approximately 70% of children in our sample were diagnosed by 3 months, and only about 40% began services by 6 months. Only 36% of children met the EHDI guidelines, despite ample evidence suggesting early diagnosis and intervention improve language outcomes (Apuzzo & Yoshinaga-Itano, 1995; Ching et al., 2013; Holzinger et al., 2011; Kennedy et al., 2006; Robinshaw, 1995; Vohr et al., 2008, 2011; Watkin et al., 2007; White & White, 1987; Yoshinaga-Itano et al., 1998, 2018). Children in our sample who met 1-3-6 guidelines were 3.62 months less delayed in spoken vocabulary than children who were late to receive diagnosis and/or services (p=0.01).

Additionally, in support of this policy, Ching and colleagues (Ching et al., 2013) find a 560 pronounced effect of early diagnosis. In this study, which was conducted in Australia, 561 researchers compared groups of children in regions that had implemented universal newborn 562 hearing screenings to children in regions that had not yet implemented UNHS. Children were 563 otherwise similar in diagnostic, demographic, and intervention characteristics. The UNHS 564 group had higher global language scores. By contrast, in our sample, by dint of accepting all 565 children receiving early intervention services in one state, we were able to document 566 naturally occurring variance in who received on-time diagnosis and intervention. We found 567 that variance in age at diagnosis and intervention were explained by children's demographic 568 and audiological characteristics. We hypothesized that children with less residual hearing 560 (i.e., bilateral, more severe) and no co-occurring conditions would be earlier diagnosed and 570 earlier to begin language services, and that earlier diagnosis would predict earlier 571 intervention. ### Diagnosis

In the case of diagnosis, presence of health issues, primary language in the home, and laterality of hearing loss accounted for 16.41% of variance in age at diagnosis. Having diagnosed health issues and non-English language background predicted later diagnosis; laterality was not significant.

Children with health issues were diagnosed 3.70 months later than infants without 577 health issues. One possible explanation is that the health issues caused acquired hearing loss 578 that wouldn't be detected by the NBHS, thus delaying identification of hearing loss. Of the 579 76 unique health issues experienced by children in the sample, only 9 conditions might cause 580 acquired hearing loss (i.e., meningitis, sepsis, jaundice, seizures, hydrocephalus, MRSA, 581 anemia, frequent fevers, cytomegalovirus), affecting 16 out of 36 children with health issues 582 in our sample. Another possible explanation is that the health issues required more pressing 583 medical attention than the possible hearing loss, and that families and medical providers had 584 to prioritize treatment for the health issue (e.g., surgery for congenital heart defect) over 585 diagnostic audiology services. 586

Infants from Spanish-speaking families were diagnosed 3.78 months later than infants 587 from English-speaking families. This may be due to cultural differences in attitudes towards deafness (Caballero, Muñoz, Schultz, Graham, & Meibos, 2018; Rodriguez & Allen, 2020; 589 Steinberg, Dávila, Collazo, Loew, & Fischgrund, 1997, p. @steinberg2003) or it may result 590 from a lack of linguistically accessible and culturally appropriate audiology services. Only 591 5.6% of American audiologists identify as a bilingual service provider (ASHA, 2019), and 592 services from a monolingual provider may be insufficient. Caballero et al. (2017) found that 593 Hispanic-American parents of DHH children want more concrete resources, comprehensive 594 information, and emotional support from their audiologist. In a nationwide survey, the 595 majority of audiologists reported using ad-hoc interpreters (e.g., client's family) to overcome 596 language barriers. Survey respondents reported that language barriers presented a major 597 challenge in working with Spanish-speaking families, specifically in obtaining the child's case 598 history and providing recommendations for follow-up services (Abreu, Adriatico, & DePierro, 2011). 600

Intervention. As expected, more severe hearing loss predicted earlier intervention, such that for every additional 10 dB HL, predicted age at diagnosis was 4.02 weeks 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 604 hearing loss. This could be related to diagnosis; certain methods of hearing screening (i.e., 605 Auditory Brainstem Responses) can miss milder hearing loss (Johnson et al., 2005), and 606 parents may not suspect hearing loss in children who respond to loud noises. Alternatively, 607 parents and clinicians may adopt a wait-and-see approach to intervention for children with 608 some residual hearing. Nevertheless, mild-to-moderate hearing loss is associated with 600 language delays and academic challenges (Blair et al., 1985; Delage & Tuller, 2007), which 610 early intervention may offset. 611

Age at start of services was also associated with age at diagnosis: for every month
diagnosis was delayed, intervention was delayed by 2.84 weeks. Early diagnosis puts children
in the pipeline towards intervention earlier. 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.

This sample is composed of children receiving birth-to-3 services. An estimated 67% of 618 children with hearing loss enroll in early intervention services (CDC, 2018). While this 619 represents a tremendous step forward in prompt early intervention services, early 620 intervention may not be early enough. Less than 39% of our sample of children in early 621 intervention meet the 6-month EHDI benchmark. Furthermore, an unknown fraction of the 622 DHH population in North Carolina aren't included in this analysis because they have not 623 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 627 by the EHDI-recommended 6 months may be closer to 26%. These children may not receive 628 clinical support until school-age or later. 629

### 630 Educational and Clinical Implications

Despite high rates of NBHS 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 642 (relative to hearing peers), and studies of spoken vocabulary development in older DHH 643 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 645 (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. Prior to intervention or amplification, provision of accessible language input (i.e., sign language) 651 may mitigate negative effects of auditory deprivation on language skills (Davidson, 652 Lillo-Martin, & Pichler, 2014; Hassanzadeh, 2012; Spellun & Kushalnagar, 2018). 653

Indeed, while we recognize that learning sign language may pose a challenge for some 654 families for myriad reasons, we nevertheless feel it is worth underscoring as an important 655 language support for DHH children and their families. We encourage sign language at least 656 prior to mastery of spoken language, and when possible for the family, we encourage its 657 continued use as a language resource. While some children in our sample exhibited 658 vocabulary development at or even slightly above the 50th percentile for hearing peers 659 (n=10), the vast majority of children fell below this threshold (n=80). Even in the **best** case 660 scenario for spoken language development (i.e., unilateral hearing loss, uses amplification, 661 etc.), children in our sample did not excel with spoken language. Offering from-diagnosis 662 access to structured linguistic input through sign language may help overcome this deficit. 663

In recommending sign language, we follow the rationale set forth by Hall, Hall, and 664 Caselli (2019), summarized here: Spoken language outcomes for DHH children are variable 665 and unpredictable (Ganek, McConkey Robbins, & Niparko, 2012; Szagun & Schramm, 2016), 666 and even in optimal situations, many DHH children do not achieve age-appropriate spoken 667 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 669 development (Amraei, Amirsalari, & Ajalloueyan, 2017; Dammeyer, 2010; Desselle, 1994; 670 Hall et al., 2017; Hrastinski & Wilbur, 2016; Kushalnagar et al., 2011; Moeller & Schick, 671 n.d.; 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 675 early access to a natural sign language offers children the best chance of achieving language 676 mastery, and use of sign language does not preclude learning spoken language. 677

## Limitations and Opportunities for Future Work

678

These analyses were exploratory, and 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. Additionally, many children in our sample have
multiple evaluations, so we have an opportunity to see whether these results hold up
longitudinally.

This sample is comprised only of children in North Carolina, and certain factors, such 686 as diagnosis and early intervention practices, vary by country and by state (NAD, n.d.). 687 However, based on Gallaudet's 2014 Annual Survey of Deaf and Hard-of-Hearing Children 688 and the SEELS report of elementary and middle school aged DHH children (Blackorby & 689 Knokey, 2006; Institute, 2014), our sample largely resembles the national DHH population in 690 terms of degree of hearing loss, percentage of children with additional disabilities, cochlear 691 implant and hearing aid use, and gender. Descriptively, compared to the Gallaudet sample, 692 more children in our sample spoke Spanish, and many fewer children in our sample used sign 693 language. We would exercise caution in applying these results to regions where sign language 694 access for DHH children is more common (e.g. Washington D.C.; Rochester, New York.) A 695 similar naturalistic study in those regions could help illuminate the effects of different clinical 696 and demographic factors in a signing population 697

There were several variables reported to influence outcomes for DHH children that we did not have access to (e.g., socioeconomic status, maternal education, parental hearing status). Measuring these variables may have provided additional predictive power.

Furthermore, the considerable variability in the sample did not allow us to easily isolate effects of different characteristics. However, this variability is real-world variability, and as we demonstrated earlier, many of these variables co-occurred such that it may not make

sense to isolate. Larger sample sizes, which are often difficult to achieve in research with

DHH children, would help to tease apart different effects. As researchers continue to study

influences on vocabulary in DHH children, a meta-analytic approach may be able to better

estimate effects and effect sizes within the varied outcomes of this diverse population.

708 Conclusion

Using a diverse sample of 100 children enrolled in early intervention, we provide a 709 description of children's demographic and audiological characteristics, vocabulary outcomes, 710 and clinical milestones. Many variables in this sample co-occurred disproportionately (e.g., 711 amplification and degree of hearing loss; communication modality and developmental delay); 712 this paper provides the first population-based documentation of this distribution. The vast 713 majority of vocabulary scores in our sample were well below established norms for hearing 714 infants. Significant predictors of vocabulary delays were: age, amplification, and laterality. 715 Only 37% of children met 1-3-6 guidelines for early detection and intervention. English 716 language background and having no co-occurring health issues significantly predicted earlier 717 diagnosis. More severe hearing loss and earlier diagnosis significantly predicted earlier 718 intervention.

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 $\label{thm:continuous} \begin{tabular}{ll} Table 1 \\ Summary of findings of CDI studies in DHH children \\ \end{tabular}$ 

Study	Population	Gender	1-3-6	Laterality	Degree	Amplification	Communication	Comorbidities
Yoshinaga-Itano et al., 2017	$8\mbox{-}39$ month children with bilateral hearing loss	No effect	1-3-6 +	Did not study	More severe -	Did not study	Did not study	Comorbidities -
Yoshinaga-Itano et al., 2018	Children with cochlear implants	Did not study	1-3-6 +	Did not study	Did not study	Earlier CI activation +	Did not study	Did not study
De Diego-Lazaro et al., 2018	Spanish speaking children with bilateral hearing loss $$	No effect	Earlier intervention +	Did not study	Milder +	More functional hearing $+$	Did not study	Did not study
Vohr et al., 2011	18-24 month olds with hearing loss	Did not study	Earlier intervention +	Did not study	Milder +	Did not study	Did not study	NICU stay -; Comorbidities -

a + equals bigger vocab, - equals smaller vocab

Table 2

CDI details

CDI version	Average Age (SD)	Average Comprehension (SD)	Average Production (SD)	% Developmental Delays
WG (n=74)	20.05 (8.82) months	105 (99.7) words	32 (53.4) words	18.92%
WS (n=24)	26.03 (7.78) months	NA	149 (180.1) words	4.17%

Table 3  $Additional\ Diagnoses\ (n=39)$ 

		I
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

 $\label{eq:audiological} Table \ 4$   $\mbox{\it Audiological Characteristics of the Sample}$ 

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

 $\label{eq:communication} \begin{tabular}{ll} Table 5 \\ Language and communication characteristics of the sample \\ \end{tabular}$ 

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

Table 6

Meets 1-3-6 table

Diagnosis by 3 months	69.47%
Average Age Diagnosis (SD)	4.65 (7.19) months
Intervention by 6 months	39.18%
Average Age Intervention (SD)	11.12 (8.54) months
Meets 1-3-6	36.84%

Table 7
Variables table

Variable	Scale	Range
Age	Continuous	4.2-36 months (mean (SD): 21 (9.1))
Age at Amplification	Continuous	2-30 months (mean (SD): 9 (6.7))
Age at Diagnosis	Continuous	0-30 months (mean (SD): 5 (7.2))
Age at Implantation	Continuous	7-32 months (mean (SD): 14 (7.2))
Age at Intervention	Continuous	1-33 months (mean (SD): 11 (8.5))
Amplification	Categorical	Hearing Aid / Cochlear Implant / None
Communication	Categorical	Spoken / Total Communication / Cued Speech
Degree Hearing Loss (worse ear)	Continuous	17.75-100 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 (6.4))
Etiology	Categorical	Sensorineural / Conductive / Mixed
CDI - Words Produced	Continuous	0-635 words (mean (SD): 61 (111.2))

term	estimate	std.error	statistic	p.value
(Intercept)	-4.1011902	1.5880766	-2.5824889	0.0115637
LateralityUnilateral	-2.6914102	1.1359432	-2.3693176	0.0201422
AmplificationHA	-0.3407579	1.2490380	-0.2728163	0.7856720
Amplificationnone	3.4923095	1.4891773	2.3451266	0.0214079
Age	0.5522518	0.0559563	9.8693442	0.0000000

term	estimate	std.error	statistic	p.value
(Intercept)	9.384015	1.967599	4.769272	0.0000069
HealthIssuesyes	3.703441	1.418520	2.610778	0.0105472
Monolingual_Englishyes	-6.469065	1.957318	-3.305066	0.0013545
LateralityUnilateral	-2.148902	1.575573	-1.363886	0.1759312

	VIF	Df
HealthIssues	1.002092	1
Monolingual_English	1.025896	1
Laterality	1.027814	1

term	estimate	std.error	statistic	p.value
(Intercept)	14.6545372	2.8717392	5.1030181	0.0000022
HLworse	-0.0925203	0.0302741	-3.0560849	0.0030365
IsPrematureyes	3.7839323	1.9540853	1.9364212	0.0563036
IdentificationOfHLMonths	0.6520471	0.1044276	6.2440093	0.0000000
Monolingual_Englishyes	-1.3846263	2.1177275	-0.6538265	0.5150755

	VIF	Df
HLworse	1.030540	1
IsPremature	1.064463	1
IdentificationOfHLMonths	1.068221	1
Monolingual_English	1.101377	1

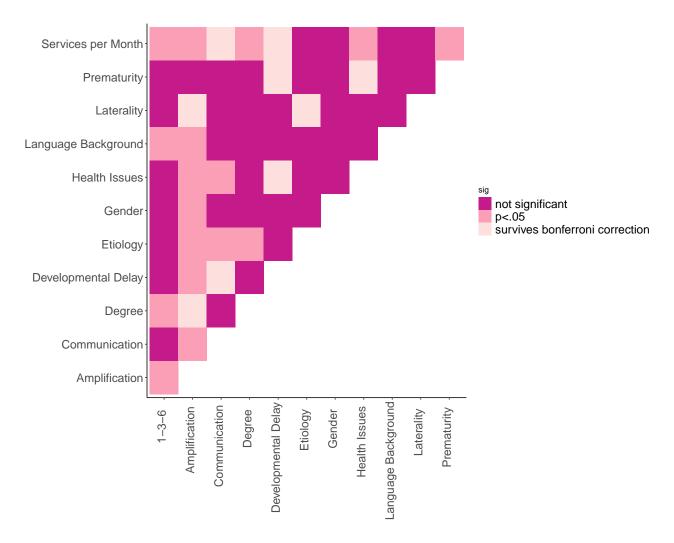


Figure 1. Results of chi-square tests between variables. For tests that survived Bonferroni correction, effect size (Cramer's V) is given.

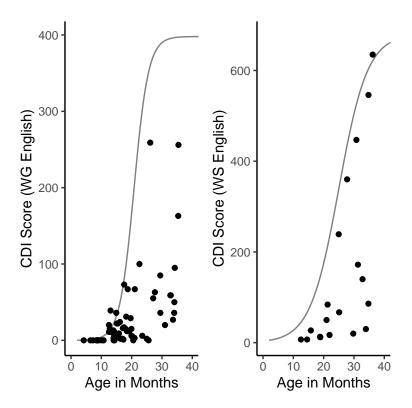


Figure 2. Growth curve from Wordbank American English 50th percentile data. Black triangles show vocabulary scores of individual DHH children.

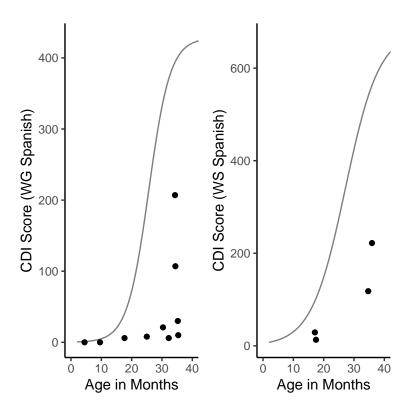


Figure 3. Growth curve from Wordbank Spanish (Mexican) 50th percentile data. Black triangles show vocabulary scores of individual DHH children.

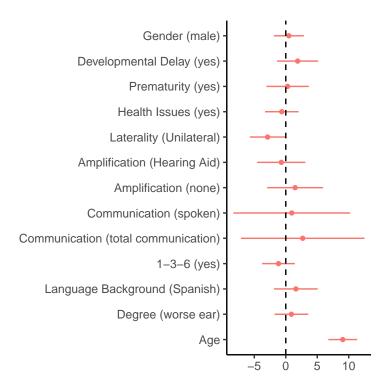


Figure 4. Coefficients for the full model.

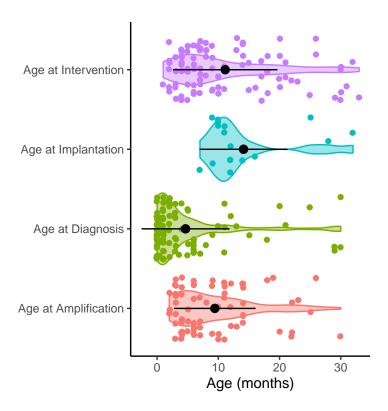


Figure 5. Timeline for diagnosis/intervention/etc.

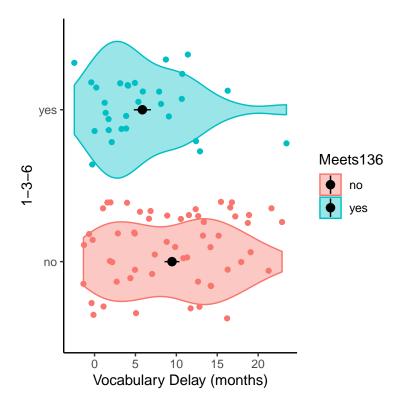


Figure 6. Months delay for children who meet / don't meet 1-3-6 guidelines.