- <sup>1</sup> Characterizing North Carolina's Deaf/Hard-of-Hearing Infants and Toddlers: Predictors of
- Vocabulary, Diagnosis, and Intervention
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7 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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In the United States, 1-2 children are born with hearing loss, per 1,000 births (CDC, 26 2018). This translates to 114,000 Deaf or Hard of Hearing (DHH) children born in the U.S. 27 per year (Martin, Hamilton, Osterman, & Driscoll, 2019). Of these 114,000, ~90\% will be 28 born to hearing parents (Mitchell & Karchmer, 2004), in a home where spoken language is likely the dominant communication method. Depending on the type and degree of hearing loss and whether the child uses amplification, spoken linguistic input will be partially or totally inaccessible. Some of these children will develop spoken language proficiency within the range of their hearing peers (Geers, Mitchell, Warner-Czyz, Wang, & Eisenberg, 2017; 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. 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 diagnosis and
- intervention initiatives.

### 54 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,

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

<sup>58</sup> 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

60 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.,

<sup>63</sup> 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

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

67 (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,

 $_{70}$  2016; Özçalışkan & Goldin-Meadow, 2010). This finding appears to be consistent across

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

<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.

<sup>72</sup> 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 (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 100 delay and disorder (Barre, Morgan, Doyle, & Anderson, 2011; Carter & Msall, 2017; Cusson, 101 2003; Rechia, Oliveira, Crestani, Biaggio, & de Souza, 2016; Van Noort-van Der Spek, 102 Franken, & Weisglas-Kuperus, 2012; Vohr, 2014). Unfortunately, research on language 103 development in premature DHH children is scant (Vohr, 2016), so it remains unclear how 104 hearing loss and prematurity may interact within spoken language skills. One study of 105 premature infants finds that auditory brainstem response during newborn hearing screening 106 predicts language performance on the PLS-4 at age 3 (Amin, Vogler-Elias, Orlando, & Wang, 107 2014), suggesting a link between prematurity, hearing loss, and language development in 108 early childhood, though further research is needed in this domain. In extremely premature 109 DHH children, incidence of additional disabilities may be as high as 73% (Robertson, 110 Howarth, Bork, & Dinu, 2009). Indeed, pre-term infants with comorbidities have been found 111 to be more likely to also have hearing loss than those without comorbidities (Schmidt et al., 112 2003), further complicating language development for this population. 113

Audiological Characteristics. Hearing loss varies in severity, ranging from slight 114 to profound (Clark, 1981). More severe hearing loss (less access to spoken language) 115 typically results in more difficulty with spoken language in infancy (Vohr et al., 2008), early 116 childhood (Ching et al., 2010, 2013; Sarant et al., 2008; Sininger, Grimes, & Christensen, 117 2010; Tomblin et al., 2015) and school-age children (Wake, Hughes, Poulakis, Collins, & 118 Rickards, 2004). Although profound hearing loss is associated with more pronounced spoken 119 language difficulty, even mild to moderate hearing loss is associated with elevated risk of 120 language disorders (Blair, Peterson, & Viehweg, 1985; Delage & Tuller, 2007). 121

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

environments (Ching, Van Wanrooy, & Dillon, 2007). The literature on hearing aids and 124 cochlear implants also points to benefits for bilateral auditory input (Lovett, Kitterick, 125 Hewitt, & Summerfield, 2010; Sarant, Harris, Bennet, & Bant, 2014; Smulders et al., 2016). 126 At school-age, 3-6% of children have unilateral hearing loss (Ross, Visser, Holstrum, Qin, & 127 Kenneson, 2010). Although children with unilateral hearing loss have one "good ear," even 128 mild unilateral hearing loss has been tied to higher risk of language delays and educational 129 challenges relative to hearing children (Kiese-Himmel, 2002; Lieu, 2004, 2013; Lieu, 130 Tye-Murray, & Fu, 2012; Vila & Lieu, 2015). Just as in the bilateral case, more severe 131 hearing loss leads to greater deficits in spoken language and educational outcomes for 132 children with unilateral hearing loss (Anne, Lieu, & Cohen, 2017; Lieu, 2013). 133

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 139 skills, if the amplification is effective. For hearing aids, some studies find that children with 140 milder hearing loss who receive hearing aids earlier have better early language achievement 141 than children who are fit with hearing aids later (Tomblin et al., 2015), but this finding does 142 not hold for children with severe-to-profound hearing loss (Kiese-Himmel, 2002; Watkin et 143 al., 2007) (for whom hearing aids are generally ineffective). Analogously, for children eligible for cochlear implants, those who receive cochlear implants earlier have better speech 145 perception and spoken language outcomes than those implanted later (Artières, Vieu, Mondain, Uziel, & Venail, 2009; Dettman, Pinder, Briggs, Dowell, & Leigh, 2007; Miyamoto, Hay-McCutcheon, Kirk, Houston, & Bergeson-Dana, 2008; Svirsky, Teoh, & Neuburger, 2004; 148 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 158 have better speech intelligibility (Dillon, Burkholder, Cleary, & Pisoni, 2004; Geers et al., 159 2017; Geers, Spehar, & Sedey, 2002; Hodges, Dolan Ash, Balkany, Schloffman, & Butts, 160 1999) and auditory perception (Geers et al., 2017; O'Donoghue, Nikolopoulos, & Archbold, 161 2000). That said, there is some debate as to whether an oral approach facilitates higher 162 spoken language performance, or whether children who demonstrate aptitude for spoken 163 language are steered towards the oral approach rather than total communication (Hall, 164 Levin, & Anderson, 2017). 165

1-3-6 Guidelines. Early identification (Apuzzo & Yoshinaga-Itano, 1995; Kennedy 166 et al., 2006; Robinshaw, 1995; White & White, 1987; Yoshinaga-Itano, Sedey, Coulter, & 167 Mehl, 1998; Yoshinaga-Itano et al., 2018) and timely enrollment in early intervention 168 programs (Ching, Dillon, Leigh, & Cupples, 2018; Ching et al., 2013; Holzinger, Fellinger, & 169 Beitel, 2011; Vohr et al., 2008, 2011; Watkin et al., 2007) are associated with better language 170 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 172 et al., 2015). In line with these findings, the American Academy of Pediatricians (AAP) has 173 set an initiative for Early Hearing Detection and Intervention (EHDI). Their EHDI 174 guidelines recommend that DHH children are screened by 1 month old, diagnosed by 3 175 months old, and enter early intervention services by 6 months old. We refer to this guideline 176

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 180 2010 (Capps, 2009) was passed to develop state-wide systems for screening, evaluation, 181 diagnosis, and "appropriate education, audiological, medical interventions for children 182 identified with hearing loss," but policies for early diagnosis and intervention vary by state. 183 As of 2011, 36 states (including North Carolina; "15A NCAC 21F .1201 - .1204," 2000) 184 mandate universal newborn hearing screening (UNHS; National Conference of State 185 Legislatures, 2011). All states have some form of early intervention programs that children 186 with hearing loss can access (NAD, n.d.), but the specifics vary state-by-state. For instance, 187 half of the states in the US do not consider mild hearing loss an eligibility criterion for early 188 intervention (Holstrum, Gaffney, Gravel, Oyler, & Ross, 2008); North Carolina does include 189 mild hearing loss in early intervention. 190

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 202 8–18-month-olds. On Words and Gestures, parents indicate whether their child understands 203 and/or produces each of the 398 vocabulary items, and answer questions about young 204 children's early communicative milestones. The Words and Sentences version of the form is 205 normed for 16–30-month-olds. On Words and Sentences, parents indicate whether their child 206 produces each of the 680 vocabulary items, and answer some questions about grammatical 207 development. The CDI has been normed on a large set of participants across many 208 languages (Anderson & Reilly, 2002; Frank et al., 2017; Jackson-Maldonado et al., 2003). 209

The CDI has also been validated for DHH children with cochlear implants (Thal, 210 Desjardin, & Eisenberg, 2007). More specifically, in this validation, researchers asked parents 211 to complete the CDI, administered the Reynell Developmental Language Scales, and 212 collected a spontaneous speech sample. All comparisons between the CDI and the other 213 measures yielded significant correlations ranging from 0.58 to 0.93. Critically, the children in 214 this study were above the normed age range for the CDI, and thus this validation helps to 215 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 217 words produced on the CDI predicts language, executive function, and academic skills up to 218 16 years later. Building on this work, several studies have used the CDI to measure 219 vocabulary development in DHH children (Yoshinaga-Itano et al. (2017); Yoshinaga-Itano et 220 al. (2018); de Diego-Lázaro et al. (2018); Vohr et al. (2008); Vohr et al. (2011). We build on 221 this literature in our analyses below. 222

### 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 231 known causal relations (e.g., cochlear implants recommended for severe hearing loss, but not 232 mild hearing loss). We sought to provide descriptive documentation about the distribution of 233 demographic, audiological, and intervention characteristics in a diverse sample of DHH 234 children receiving state services. For the second, we hypothesized that male (vs. female) 235 gender, more severe degree of hearing loss, bilateral (vs. unilateral) hearing loss, no 236 amplification (vs. hearing aids and/or cochlear implants), premature birth, and presence of 237 additional disabilities would predict larger spoken vocabulary delay. We did not have strong 238 predictions regarding the effects of communication method or presence of other health issues 239 (e.g., congenital heart malformation) on vocabulary. For the third goal, based on the prior 240 literature summarized above, we hypothesized that children with less residual hearing (i.e., 241 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 243 intervention.

245 Methods

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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<sup>2</sup>. 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

<sup>&</sup>lt;sup>2</sup> Because the data we received were already deidentified, this study was exempt from Duke University Institutional Review Board.

252 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, 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 258 data from 100 children. Thus, the reported sample below consists of 100 children (56 male / 259 44 female) ages 4.20-36.20 months (M=21.20, SD=9.10). Race and socioeconomic 260 information were not available. Families were administered either the Words and Gestures or 261 Words and Sentences version of the CDI based on clinician judgment. Children who were too 262 old for Words and Gestures, but who were not producing many words at the time of 263 assessment, were often given Words and Gestures (n = 37). Families for whom Spanish was 264 the primary language (n = 14) completed the Spanish language version of the CDI 265 (Jackson-Maldonado et al., 2003). See Table 1 for additional CDI information for our sample.

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 Supplemental Materials, Table S1).

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 277 hearing loss measures was coded with the decibels of hearing loss (dB HL) corresponding 278 with the median dB HL for the level of hearing loss (e.g., moderate hearing loss was coded as 279 48 dB HL), and sloping hearing loss was coded as the average of the levels (e.g. mild to 280 moderate was coded as 40.5 dB HL). Participants were also coded for unilateral or bilateral 281 hearing loss; presence or absence of Auditory Neuropathy Spectrum Disorder; and etiology of 282 hearing loss (sensorineural, conductive, or mixed). Amplification was recorded as the device 283 the child used at the time of assessment: either hearing aid, cochlear implant, or none. See 284 Supplemental Materials, Table S2 for audiological characteristics of the sample. 285

Communication method was recorded as spoken language, total communication, or 286 cued speech. One participant had a parent fluent in sign language, but the reported 287 communication method in the home was total communication. No child in our sample used 288 American Sign Language or another signed language. The forms also listed the primary 289 language spoken at home, which we binned into English-speaking and non-English-speaking. 290 85 out of the 100 had a primary language of English, while 14had Spanish. The remaining 291 child was adopted from a non-English-speaking country after age 2 and had heard mostly 292 non-English by the time of assessment; this child was coded as non-English-speaking. 293 Language and communication information is summarized in Supplemental Materials, Table 294 S3. 295

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,
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.

A summary of all the variables we examined is available in Table 2.

311 Results

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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.

# Relationships Among Demographic, Audiological, and Clinical Variables

Before we test how these variables may be related to vocabulary, we describe their relationships to each other. As would be expected, many health, audiological, and clinical characteristics are not distributed randomly across this sample of children. To quantify this statistically, we used Bonferroni-corrected chi-square tests between each of our variables<sup>3</sup>. Because the chi-square statistic assumes n > 5 is *expected* in the majority of the cells for

<sup>&</sup>lt;sup>3</sup> 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.

each test (preferably  $\geq 80\%$  McHugh, 2013), we excluded mixed hearing loss (n = 8) and cued speech (n = 1) from this section of the analysis. Strictly speaking, some variables are 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 ??.

As expected, we found that health issues, developmental delays, and premature birth 333 were highly interrelated in our sample, such that children born premature were more likely 334 to also experience health issues  $(X^2 (1, N = 98) = 23.9, p < .0001)$  and developmental 335 delays  $(X^2 (1, N = 98) = 11.63, p = .0006)$ , and children with developmental delays were 336 more likely to also experience health issues  $(X^2 (1, N = 98) = 20.87, p < .0001)$ . Children 337 with developmental delays received more services per month than typically-developing 338 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<sup>4</sup>. The chi-square tests further showed that amplification was related 347 to laterality  $(X^2 (2, N = 98) = 16.43, p = .0003)$  and degree of hearing loss  $(X^2 (4, N = 87))$ 348 = 28.45, p < .0001) in our sample. Specifically, children with bilateral hearing loss were 349 more likely than children with unilateral hearing loss to use a hearing aid or cochlear 350 implant; no child with unilateral hearing loss used a cochlear implant, and many children 351 with unilateral hearing loss used no amplification. Regarding degree, children with severe to 352 profound hearing loss were more likely to use a cochlear implant than children with less 353 severe hearing loss (i.e., mild or moderate). 354

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.

## 359 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 ?? 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

 $<sup>^4</sup>$  All children with mixed hearing loss (n = 8) had bilateral hearing loss.

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 373 used the 50th percentile for productive vocabulary from Wordbank data typically-developing 374 infants (Frank et al., 2017) to create binary logistic growth curves separately for the "Words 375 and Gestures" (WG) and "Words and Sentences" (WS) versions of the CDI for American 376 English and Mexican Spanish<sup>5</sup>. For each child, we took the number of words they produced 377 divided by the number of words on the instrument, to give us the proportion of words 378 produced. We used this proportion in an inverse prediction from the binary logistic regression curves to generate a predicted age. That is, for each possible CDI score, the growth curve provided the age that the score would be achieved for the 50th percentile trajectory. Finally, we subtracted the predicted age from each child's chronological age to calculate their vocabulary delay. However, for children producing 0 words, this approach was 383 not appropriate due to the long tails on the growth curves. Thus, for this subset of children, 384 we took the x-intercept from Wordbank (8 months for English, and 9 months for Spanish), 385 and subtracted that value from the child's chronological age to get their vocabulary delay. 386

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>6</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.

<sup>&</sup>lt;sup>5</sup> Number of hearing children in normative sample for each growth curve: WG-English=1071, WG-Spanish=760; WS-English=1461, WS-Spanish=1092

<sup>&</sup>lt;sup>6</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.

This model accounted for significant variance in vocabulary delay (adjusted- $R^2 = 0.59$ , 393 p < .001). We next performed stepwise model comparison using stepAIC (MASS) to pare 394 down the model. This process selects only the predictors which incrementally improved 395 model fit, measured by Akaike's Information Criterion (AIC), which considers goodness of fit 396 and model complexity (penalizing models with many predictors). We started model selection 397 with the full model, as described above. We then filtered out data from children for whom 398 Meets 1-3-6 (n = 5) or Degree (n = 13) was unknown, as this stepwise AIC approach does 399 not permit missing values across predictors. Since this initial filtered analysis found that 400 Degree and 1-3-6 did not improve model fit, we manually removed the Degree and 1-3-6 401 terms from the model selection so that the 15 participants with missing cases for these 402 variables could be retained.<sup>7</sup> 403

Based on this iterative process, we arrived at the following final model: Vocabulary 404 Delay ~ Age + Laterality + Amplification. This model accounted for significant variance in 405 children's vocabulary delay to a nearly identical degree as the full model (adjusted- $\mathbf{R}^2$ 406 0.58, p = < .001, see Table ?? & Figure ??.A). We found significant main effects for Age, 407 Amplification, and Laterality, such that older age, no amplification, and bilateral hearing loss 408 predicted greater vocabulary delays. Compared to children with no amplification, children 409 with cochlear implants had a 3.50 months smaller spoken vocabulary delay (p = .021), and similarly children with hearing aids had a 3.84 months smaller delay (p = .001). Children 411 with unilateral hearing loss had a 2.70 months smaller delay (p = .020) than children with 412 bilateral hearing loss. With regard to Age, for each month older, the model predicted a 0.55 413 months larger vocabulary delay (p < .001). 414

<sup>&</sup>lt;sup>7</sup> 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.

Given our results above revealing relationships among several of these variables (e.g., laterality 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 < 1.20; James, Witten, Hastie, & Tibshirani, 2013). In sum, the analyses in this section revealed that over half of the variance in DHH children's vocabulary 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

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Perhaps of greatest importance to clinicians and policymakers is the implementation 423 and effect of existing policies. Although whether a child met 1-3-6 guidelines was not 424 included in our final model predicting vocabulary delay through our model selection process, 425 its demonstrated importance for language outcomes (e.g., Yoshinaga-Itano et al., 2018) 426 merits further discussion. To this end, we looked at the ages at which children received 427 diagnosis and intervention, and how this mapped onto the 1-3-6 guidelines. In this section, 428 we provide a brief description of the implementation of 1-3-6 in our sample, examine its 429 effect on vocabulary delay, and describe the results of exploratory linear regression models 430 for age at diagnosis and age at intervention. 431

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 ?? 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 ??). 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).

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 described in the preceding section.

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 ~ Health Issues + Language Background +

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

Table ?? & Figure ??.B). This model accounted for 16.41% of the variance in age at

diagnosis (p = .001). Average age at diagnosis was 4.65(7.19) 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).

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<sup>2</sup>=0.43, p < .001; See Table ?? & Figure ??.C), with significant main effects 465 of degree and age at diagnosis. Prematurity ( $\beta = 3.78$ , p = .06) and language background ( $\beta$ 466 = -1.38, p = .52) were not significant predictors on their own, but their inclusion improved 467 model fit. Average age at intervention was 11.12(8.54) months. More severe hearing loss 468 predicted earlier intervention, such that for every additional 10 dB HL, predicted age at 460 intervention was 1 month earlier (p < .01). With regard to age at diagnosis, for every month 470 diagnosis was delayed, intervention was delayed by 2.80 weeks (p < .01). Taken together 471 these analyses reveal that beyond aspects of the child's hearing status, other variables too 472 contribute to delays in both diagnoses and intervention. We return to this point in the 473 discussion. 474

475 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 503 clinical variables. Prematurity, health issues, and developmental delay frequently 504 co-occurred, such that children with one of these factors were more likely to have the others. 505 This is not surprising. Many conditions that cause developmental delays have a high 506 incidence of health issues (e.g., heart problems in Down Syndrome; vomiting and seizures 507 with hydrocephalus), and it is well documented that there is a higher incidence of 508 developmental delay and health issues in preterm infants (Aarnoudse-Moens, 509 Weisglas-Kuperus, van Goudoever, & Oosterlaan, 2009; Costeloe et al., 2012; Luu, Katz, 510 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?? and elssp comorbidities.csv on OSF for more 513 detailed information about comorbidities). Some studies to date have examined the 514 outcomes of DHH children with certain conditions (e.g., Clibbens, 2001; Szymanski, Brice, 515 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 519 much more likely to use a total communication approach than DHH children without 520 developmental delays (i.e., total communication used by 58.82% of DHH children with 521 developmental delay vs. 9.88% of those without). That is, communication modality was not 522 distributed randomly throughout our sample, with use of total communication linked to 523 children already at greater risk for verbal delays. Such a pattern is in line with clinical use of 524 manual communication approaches for young children with disabilities (e.g., Branson & 525 Demchak, 2009). This result tempers the interpretation of correlational studies finding links 526 between total communication and language delays (e.g., Geers et al., 2017). 527

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.

## 47 Predicting vocabulary outcomes

In our sample, 88.89% of DHH children fell below the 50th percentile for spoken 548 vocabulary. Moreover, of the 11.11% who were at or above the 50th percentile, 55.56% were 540 8-to-9-month olds who were not yet producing any words (as expected at this age). Finding 550 that nearly 90% of DHH children are below the 50th percentile for vocabulary development 551 indicates that this group is not yet well-equipped to acquire spoken language. This 552 disadvantage can have lasting consequences in the lives of DHH children (Karchmer & 553 Mitchell, 2003; Kyle & Harris, 2010; Qi & Mitchell, 2012), highlighting the importance of 554 understanding what factors contribute to it. 555

We predicted that male gender, more severe hearing loss, bilateral hearing loss, no 556 amplification, premature birth, and presence of additional disabilities would be associated 557 with larger spoken vocabulary delay. In contrast to our predictions, the best model 558 predicting vocabulary delay had just a few variables: age, amplification, and laterality. 559 Notably, we did not simply find that DHH children were learning words at the same rate 560 (albeit delayed) as hearing children, which would have led to a constant delay across 561 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 565 vocabulary delay is likely to have knock-on effects for language development more broadly as 566 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 569 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; 572 Kennedy et al., 2006; Robinshaw, 1995; Vohr et al., 2008, 2011; Watkin et al., 2007; White & 573 White, 1987; Yoshinaga-Itano et al., 1998, 2018). Children in our sample who met 1-3-6 574 guidelines were 3.62 months less delayed in spoken vocabulary than children who were late 575 to receive diagnosis and/or services. With these demonstrable benefits in mind, our sample, 576 by dint of accepting all children receiving early intervention services in one state, was able to 577 explore naturally occurring variance in who received on-time diagnosis and intervention. 578

Having health issues or a non-English language background predicted 579 later diagnosis. Children with health issues were diagnosed 3.70 months later than infants 580 without health issues. One possible explanation is that the health issues caused acquired 581 hearing loss that wouldn't be detected by the newborn hearing screening, thus delaying 582 identification of hearing loss. In our sample, 16 of the 36 children with health issues had 583 conditions that might cause acquired hearing loss (i.e., meningitis, sepsis, jaundice, seizures, 584 hydrocephalus, MRSA, anemia, frequent fevers, cytomegalovirus). While acquired hearing 585 loss may be one driver of delayed diagnosis for children with health issues, this accounts for 586 only a fraction of the subpopulation with health issues. Another possible explanation is that 587 the health issues required more pressing medical attention than the possible hearing loss. For instance, families and medical providers are likely to prioritize treatment for certain health issues (e.g., surgery for congenital heart defect) over diagnostic audiology services. Nevertheless, it is possible that in some cases, clinician awareness of the increased delays in 591 language related to health issues more broadly may facilitate improvements in timely 592 diagnosis. 593

Language background too predicted age at diagnosis. Infants from Spanish-speaking 594 families were diagnosed 3.78 months later than infants from English-speaking families. This 595 may be due to cultural differences in attitudes towards deafness (Caballero, Muñoz, Schultz, 596 Graham, & Meibos, 2018; Rodriguez & Allen, 2020; Steinberg, Bain, Li, Delgado, & Ruperto, 597 2003; Steinberg, Dávila, Collazo, Loew, & Fischgrund, 1997) or it may result from a lack of 598 linguistically accessible and culturally appropriate audiology services. Only 5.6% of 590 American audiologists identify as bilingual service providers (ASHA, 2019), and services 600 from a monolingual provider may be insufficient. To this point, Caballero et al. (2017) found 601 that Hispanic-American parents of DHH children wish for more concrete resources, 602 comprehensive information, and emotional support from their audiologist. In a nationwide 603 survey of audiologists, the majority of audiologists reported that language barriers presented 604 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). 607

**Intervention.** As expected, more severe hearing loss predicted earlier intervention, 608 such that for every additional 10 dB HL, predicted age at intervention was 0.93 month 600 earlier. This converges with findings by Harrison, Roush, and Wallace (2003) in which 610 severe-to-profound hearing loss was diagnosed 2-5 months earlier than mild-to-moderate 611 hearing loss. Parents and clinicians may adopt a wait-and-see approach to intervention for 612 children with some residual hearing. Nevertheless, mild-to-moderate hearing loss is 613 associated with language delays and academic challenges (Blair et al., 1985; Delage & Tuller. 614 2007), which early intervention may offset. 615

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 622 birth-to-3 services. An estimated 67% of children with hearing loss enroll in early 623 intervention services (CDC, 2018). While this represents a tremendous step forward in 624 prompt early intervention services relative to just a few decades ago, early intervention may 625 not be early enough. Less than 39% of our sample of children in early intervention meet the 626 6-month EHDI benchmark. Furthermore, an unknown fraction of the DHH population in 627 North Carolina aren't included in this analysis because they have not been enrolled in 628 services by 36 months. The AAP estimates that almost 36% of infants who do not pass a 629 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 632 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 634 an important foundation for literacy and academic success (Biemiller, 2003; Hemphill & 635 Tivnan, 2008; Monroe & Orme, 2002; Stæhr, 2008; Young, 2005). 636

### 637 Educational and Clinical Implications

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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 649 (relative to hearing peers), and studies of spoken vocabulary development in older DHH 650 children suggest that they may not catch up (Lund, 2016). This should set clinicians and 651 educators on high alert, due to the demonstrated importance of vocabulary skills in literacy 652 (Biemiller, 2003; Hemphill & Tivnan, 2008; Stæhr, 2008) and in education more broadly 653 (e.g., Young, 2005; Monroe & Orme, 2002). As early intervention predicts vocabulary 654 outcomes in study after study (including this present study and e.g., Vohr et al., 2008, 2011; 655 Ching et al., 2018, 2013; Holzinger et al., 2011; Watkin et al., 2007), ensuring intervention by 656 6 months for all DHH children may be one way to address spoken vocabulary deficits. 657 Another solution: even prior to intervention or amplification, provision of structured, 658 accessible language input (i.e., sign language) may mitigate negative effects of auditory 659 deprivation on language skills (Davidson, Lillo-Martin, & Pichler, 2014; Hassanzadeh, 2012; 660 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,

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academic, and socioemotional development (Amraei, Amirsalari, & Ajalloueyan, 2017; 671 Dammeyer, 2010; Desselle, 1994; Hall et al., 2017; Hrastinski & Wilbur, 2016; Kushalnagar 672 et al., 2011; Moeller & Schick, 2006; Preisler, Tvingstedt, & Ahlström, 2002; Schick, De 673 Villiers, De Villiers, & Hoffmeister, 2007). The available data do not suggest that sign 674 language harms spoken language development (Davidson et al., 2014; Park et al., 2013), and 675 in fact, some studies suggest that sign language benefits spoken language development (e.g., 676 Hassanzadeh, 2012). Providing early access to a natural sign language offers children another 677 path to language mastery, and use of sign language does not preclude learning spoken 678 language. Thus, we encourage sign language use at least prior to mastery of spoken language, 679 and when possible for the family, we encourage its continued use as a language resource. 680

## 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 (https://osf.io/kfcs3/) 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.

707 Conclusion

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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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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.

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.")

- Table 2: Detailed information about the variables studied. For categorical variables, levels are described. For continuous variables, range, mean, and standard deviation are provided.
- Table 3: Unstandardized beta weights (months of vocabulary delay) for the model of vocabulary delay selected by AIC.
- Table 4: Unstandardized beta coefficients (months) for the model of age at diagnosis selected by AIC.
- Table 5: Unstandardized beta coefficients (months) for the model of age at intervention selected by AIC.
- Supplemental Materials S1: Additional Diagnoses (n = {n\_condition(anycomorbid)}):

  Ns of participants in our sample diagnosed with other conditions. N.B.: Ns do not sum to

  total because many participants had multiple diagnoses.
- Supplemental Materials S2: 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.
- Supplemental Materials S3: Language and Communication Characteristics of the Sample: Ns of participants by language background and communication method.