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, 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 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 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 state 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, and 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 vocabulary, with the broader goal of considering the success of early diagnosis and intervention initiatives.

Predictors of Language Outcomes

Though the literature points towards spoken language delays and deficits for DHH children, this is a highly variable population with highly variable outcomes (Pisoni, Kronenberger, Harris, & Moberly, 2018). Previous research indicates that gender (Ching et al., 2013; Kiese-Himmel & Ohlwein, 2002), additional disability (Ching et al., 2013; Verhaert et al., 2008; Yoshinaga-Itano, Sedey, Wiggin, & Chung, 2017), degree and configuration of hearing loss (Ching et al., 2013; de Diego-Lázaro, Restrepo, Sedey, & Yoshinaga-Itano, 2018; Vohr et al., 2011; Yoshinaga-Itano et al., 2017), amplification (Walker et al., 2015), communication (Geers et al., 2017), and early diagnosis/intervention (Yoshinaga-Itano et al., 2017, 2018) predict language outcomes in DHH children. We first provide a brief literature review on the effect of these predictors on language skills in DHH children.

Gender. For hearing children, the literature points to a female gender advantage in early language acquisition. Girls speak their first word earlier (Macoby, 1966), have a larger (Bornstein, Hahn, & Haynes, 2004; Fenson et al., 1994; Frank, Braginsky, Yurovsky, & Marchman, 2017) and faster-growing vocabulary (Huttenlocher, Haight, Bryk, Seltzer, & Lyons, 1991), and stronger grammatical and phonological skills (Lange, Euler, & Zaretsky, 2016; Özçalışkan & Goldin-Meadow, 2010). This finding appears to be consistent across studies (Wallentin, 2009), various spoken languages (Frank, Braginsky, Marchman, & 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 delay and disorder (Barre, Morgan, Doyle, & Anderson, 2011; Carter & Msall, 2017; Cusson,

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

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

Hearing loss also varies in whether it affects one ear or both. Bilateral hearing assists speech perception, sound localization, and loudness perception in quiet and noisy environments (Ching, Van Wanrooy, & Dillon, 2007). The literature on hearing aids and cochlear implants also points to benefits for bilateral auditory input (Lovett, Kitterick, Hewitt, & Summerfield, 2010; Sarant, Harris, Bennet, & Bant, 2014; Smulders et al., 2016). At school-age, 3–6% of children have unilateral hearing loss (Ross, Visser, Holstrum, Qin, & Kenneson, 2010). Although children with unilateral hearing loss have one "good ear," even

mild unilateral hearing loss has been tied to higher risk of language delays and educational challenges relative to hearing children (Kiese-Himmel, 2002; Lieu, 2004, 2013; Lieu, Tye-Murray, & Fu, 2012; Vila & Lieu, 2015). Just as in the bilateral case, more severe hearing loss leads to greater deficits in spoken language and educational outcomes for 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 with children in HAs (Stiles, Bentler, & McGregor, 2012).

For both hearing aids and cochlear implants, earlier fit leads to better spoken language skills, if the amplification is effective. For hearing aids, some studies find that children with milder hearing loss who receive hearing aids earlier have better early language achievement than children who are fit with hearing aids later (Tomblin et al., 2015), but this finding does not hold for children with severe to profound hearing loss (Kiese-Himmel, 2002; Watkin et al., 2007) (for whom hearing aids are generally ineffective). Analogously, children who are eligible and receive cochlear implants earlier have better speech 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; 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 it often includes elements of sign, such as individual signs, is not a sign language, such as American Sign Language. Clinicians currently employ total communication as an alternative

or augmentative communication method for children with a wide range of disabilities (Branson & Demchak, 2009; Gibbs & Carswell, 1991; Mirenda, 2003).

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

1-3-6 Guidelines. Early identification (Apuzzo & Yoshinaga-Itano, 1995; Kennedy et al., 2006; Robinshaw, 1995; White & White, 1987; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998; Yoshinaga-Itano et al., 2018) and timely enrollment in early intervention programs (Ching et al., 2013; Holzinger, Fellinger, & Beitel, 2011; Vohr et al., 2008, 2011; Watkin et al., 2007) are associated with better language proficiency. Indeed, DHH children who receive prompt diagnosis and early access to services have been found to meet age-appropriate developmental outcomes, including language (Stika et al., 2015).

In line with these findings, the American Academy of Pediatricians (AAP) has set an initiative for Early Hearing Detection and Intervention (EHDI). Their EHDI guidelines recommend that DHH children are screened by 1 month old, diagnosed by 3 months old, and enter early intervention services by 6 months old. We refer to this guideline as 1-3-6. Meeting this standard appears to improve spoken language outcomes for children with HL (Yoshinaga-Itano et al., 2017, 2018) and the benefits appear consistent across a range of demographic characteristics.

At a federal level in the U.S., the Early Hearing Detection and Intervention Act of 2010 (Capps, 2009) was passed to develop state-wide systems for screening, evaluation,

diagnosis, and "appropriate education, audiological, medical interventions for children identified with hearing loss," but policies for early diagnosis and intervention vary by state. As of 2011, 36 states (including North Carolina, ("15A NCAC 21F .1201 - .1204," 2000)] mandate universal newborn hearing screening (National Conference of State Legislatures, 2011). All states have some form of early intervention programs that children with hearing loss can access (NAD, n.d.), but these also vary state-by-state. For instance, half of the states in the US do not consider mild hearing loss an eligibility criterion for early intervention (Holstrum, Gaffney, Gravel, Oyler, & Ross, 2008).

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 may be breaks in the chain from screening to diagnosis and from diagnosis to intervention, and the effect may be further delays in language development for children not meeting these guidelines.

Quantifying vocabulary growth in DHH children

The MacArthur Bates Communicative Development Inventory (CDI, Fenson et al., 1994) is a parent-report instrument that gathers information about children's vocabulary development. The Words and Gestures version of the form is normed for 8–18-month-olds. On Words and Gestures, parents indicate whether their child understands or produces each of the 398 vocabulary items, and answer questions about young children's early communicative milestones. The Words and Sentences version of the form is normed for 16-30-month-olds. On Words and Sentences, parents indicate whether their child produces each of the 680 vocabulary items, and answer some questions about grammatical development. The CDI has been normed on a large set of participants across many languages (Anderson & Reilly, 2002; Frank et al., 2017; Jackson-Maldonado et al., 2003).

The CDI has also been validated for DHH children with cochlear implants (Thal, Desjardin, & Eisenberg, 2007). More specifically, in this validation, researchers asked parents to complete the CDI, administered the Reynell Developmental Language Scales, and collected a spontaneous speech sample. All comparisons between the CDI and the other measures yielded significant correlations ranging from 0.58 to 0.93. Critically, the children in this study were above the normed age range for the CDI, and thus this validation helps to confirm that the CDI is a valid measurement tool for older DHH children. In further work, Castellanos, Pisoni, Kronenberger, and Beer (2016) finds that in children with CIs, number of words produced on the CDI predicts language, executive function, and academic skills up to 16 years later. Building on this work, several studies have used the CDI to measure vocabulary development in DHH children (Ching et al. (2013); Yoshinaga-Itano et al. (2017); Yoshinaga-Itano et al. (2018); de Diego-Lázaro et al. (2018); Vohr et al. (2008); Vohr et al. (2011); summarized in 1).

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).

For the first goal, we had reason to expect that many of these variables would be related, due to known causal relations (e.g., cochlear implants recommended for severe hearing loss, but not mild hearing loss). We sought to provide descriptive documentation about the distrubution of demographic, audiological, and intervention characteristics in a diverse sample of DHH children receiving state services. For the second, we hypothesized

that male gender, more severe degree of hearing loss, bilateral hearing loss, no amplification use, prematurity, and presence of additional disabilities would predict larger spoken vocabulary delay. We did not have strong predictions regarding the effects of communication method or presence of other health issues (e.g., congenital heart malformation) on vocabulary. For the third goal, we hypothesized that children with less residual hearing (i.e., bilateral, more severe) and no co-occurring conditions would be earlier diagnosed and earlier to begin language services, and that earlier diagnosis would predict earlier intervention.

Methods

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

The clinical evaluations included demographic and audiological information, CDI vocabulary scores, and the results of any clinical assessments administered (e.g., PPVT), all 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 data from 100 children. Thus, the reported sample below consists of 100 children (56 male / 44 female) ages 4.20–36.17(M=21.21, SD=9.08). Race and SES information were not available. Families were administered either the Words and Gestures or Words and Sentences version of the CDI based on clinician judgement. Children who were too old for Words and

Gestures, but who were not producing many words at the time of assessment, were often given Words and Gestures (n=37). Families for whom Spanish was the primary language (n=14) completed the Spanish language version of the CDI (Jackson-Maldonado et al., 2003).

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 prematurity (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).

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). Using the ASHA hearing loss guidelines, each of these was coded with a dB HL value corresponding with the median dB HL for the level of hearing loss (e.g., moderate hearing loss was coded as 48 dB HL), and sloping hearing loss was coded as the average of the levels (e.g. mild to moderate was coded as 40.5 dB HL). Participants were also coded for unilateral or bilateral hearing loss; presence or absence of Auditory Neuropathy Spectrum Disorder; and etiology of hearing loss (sensorineural, conditive, or mixed). Amplification was recorded as the device the child used at the time of assessment: either hearing aid, cochlear implant, or none.

Communication method was recorded as spoken language, total communication, or cued speech. One participant had a parent fluent in sign language, but the reported communication method in the home was total communication. No child in our sample used American Sign Language or another signed language. The forms also listed the primary language spoken at home, which we binned into English-speaking and non-English-speaking. 85% of families spoke English, and 14% spoke Spanish. For one child, who was adopted from another country after her second birthday, we recorded the language background as

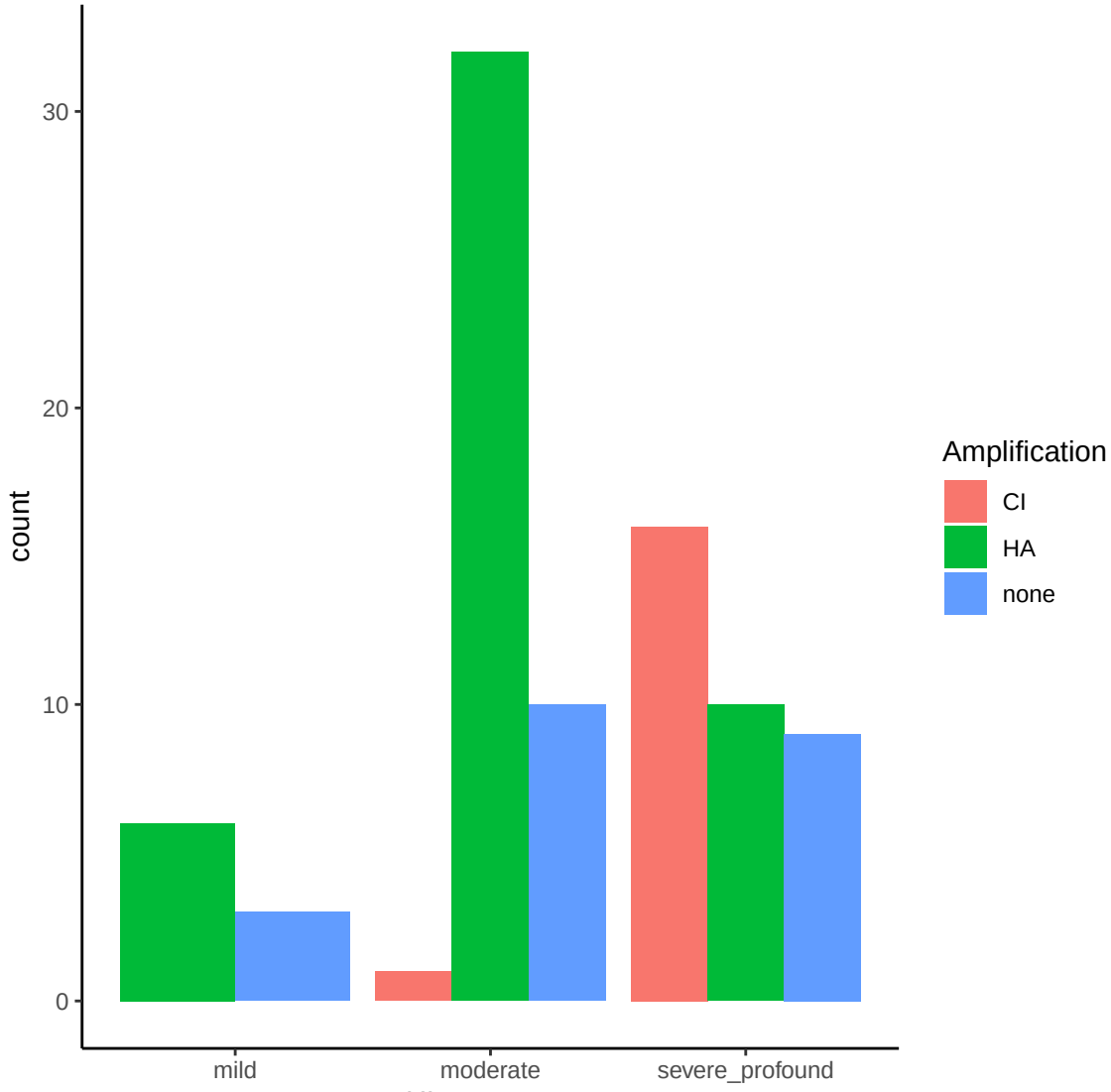
non-English-speaking, although the child's adoptive parents are English-speaking, because the child had lived most of her life in a non-English-speaking environment.

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 newborn hearing screening, 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. From the clinician report, we calculated the number of hours of early intervention services received per month (including service coordination, speech therapy, and occupational therapy, among others). Because of the relatively sparse data on screening age, if participants had an age at diagnosis $\leq$ 3 mo. and an age of intervention $\leq$ 6 mo., they were recorded as meeting 1-3-6. It is possible that a participant did not receive screening by 1 month, but did receive diagnosis by 3 months and services by 6 months. This special case would be coded as meeting 1-3-6 by our criteria.

Results

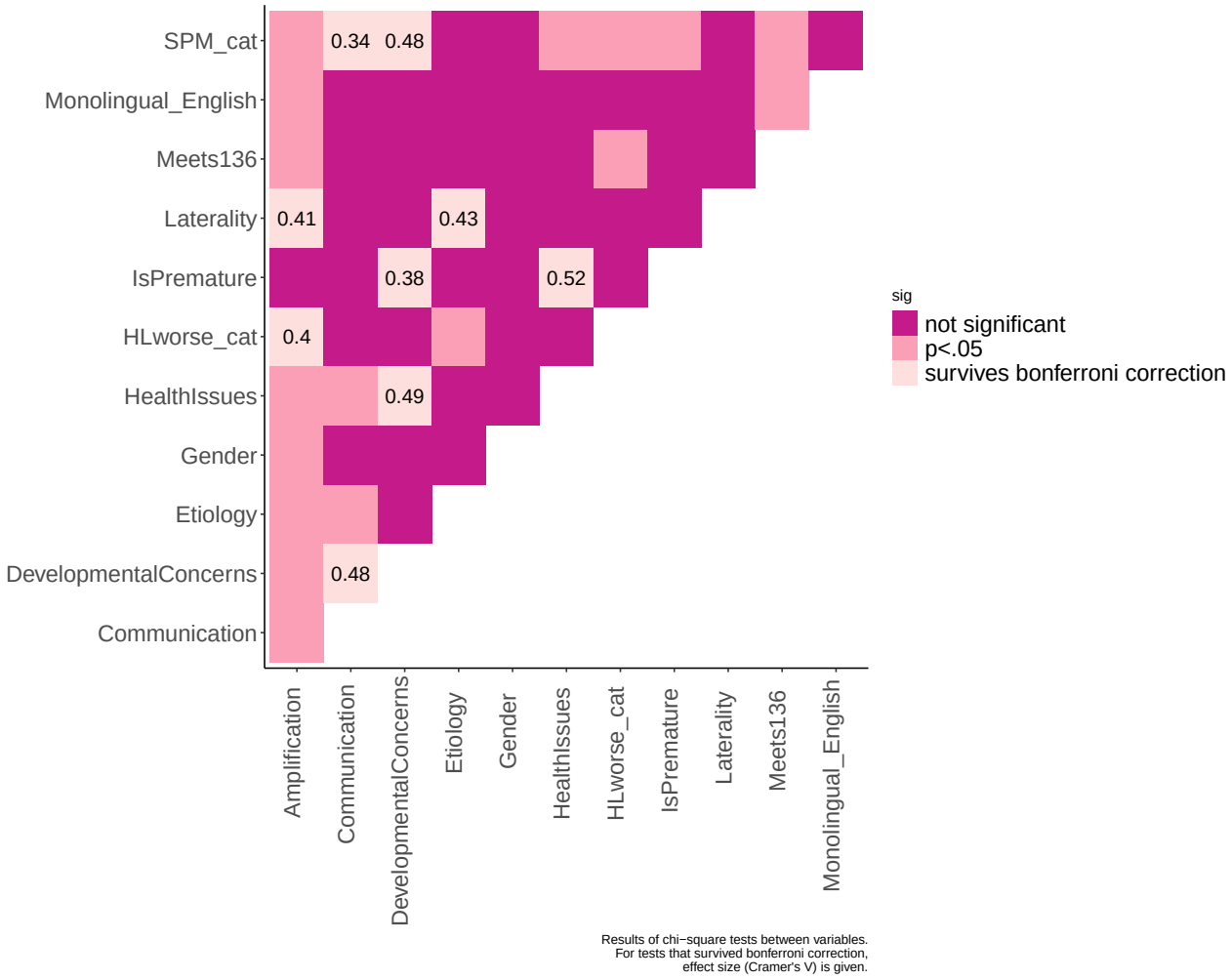
In the first section, we explore relationships among child demographic, audiological, and clinical variables. In the second section, we examine the influence of these factors on vocabulary development. In the third section, we describe the implementation of the EHDI 1-3-6 guidelines and predictors of early diagnosis and intervention. All analyses were conducted in R. All code is available on Github.

Part I: Interactions Among Variables



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 (gender (male/female), laterality (bi-/uni-lateral hearing loss), health issues (yes/no), developmental delays (yes/no), prematurity (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)). Because the chi-square statistic assumes $n > 5$ is *expected* in the majority of the cells for 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 of these variables are not expected to be randomly distributed relative to each other (e.g., prematurity 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 **??**.



We found that health issues, developmental delays, and prematurity were highly interrelated in our sample, such that children born premature were more likely to also experience health issues (X^2 (1, N = 98) = 23.9, p = 1e-06) and developmental delays (X^2 (1, N = 98) = 11.63, p = 0.00065), and children with developmental delays were more likely to also experience health issues (X^2 (1, N = 98) = 20.87, p = 4.9e-06). Children with developmental delays received more services per month than typically-developing children (X^2 (2, N = 95) = 22.17, p = 1.5e-05) and were more likely to use total communication (X^2 (2, N = 98) = 22.51, p = 1.3e-05). Likewise, children who used total communication received more services per month than children using spoken language (X^2 (4, N = 95) = 21.35, p = 0.00027).

We also found relationships among many of the audiological characteristics. There was a significant relationship between laterality and etiology (X^2 (2, N = 88) = 18.29, p = 0.00011), such that children with conductive hearing loss were more likely to have unilateral hearing loss, children with sensorineural hearing loss were more likely to have a bilateral loss, and all children with mixed hearing loss (n=8) had bilateral hearing loss. Chi-square tests showed that laterality (X^2 (2, N = 98) = 16.43, p = 0.00027) and degree of hearing loss (X^2 (4, N = 87) = 28.45, p = 1e-05) were related to 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-profound hearing loss were more likely to use a cochlear implant than children with less severe hearing loss (i.e., mild or moderate).

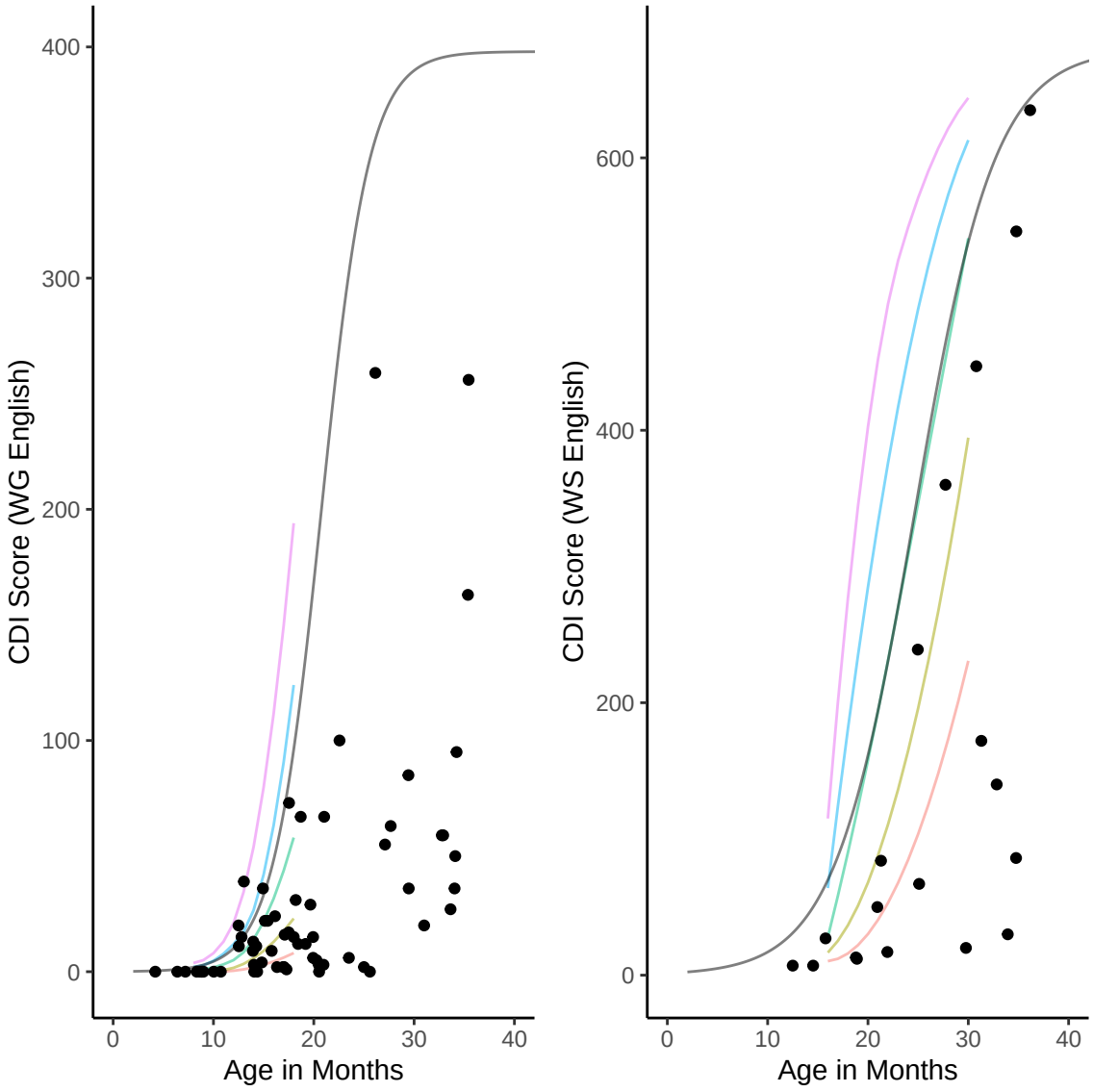
Part II: Influence on vocabulary

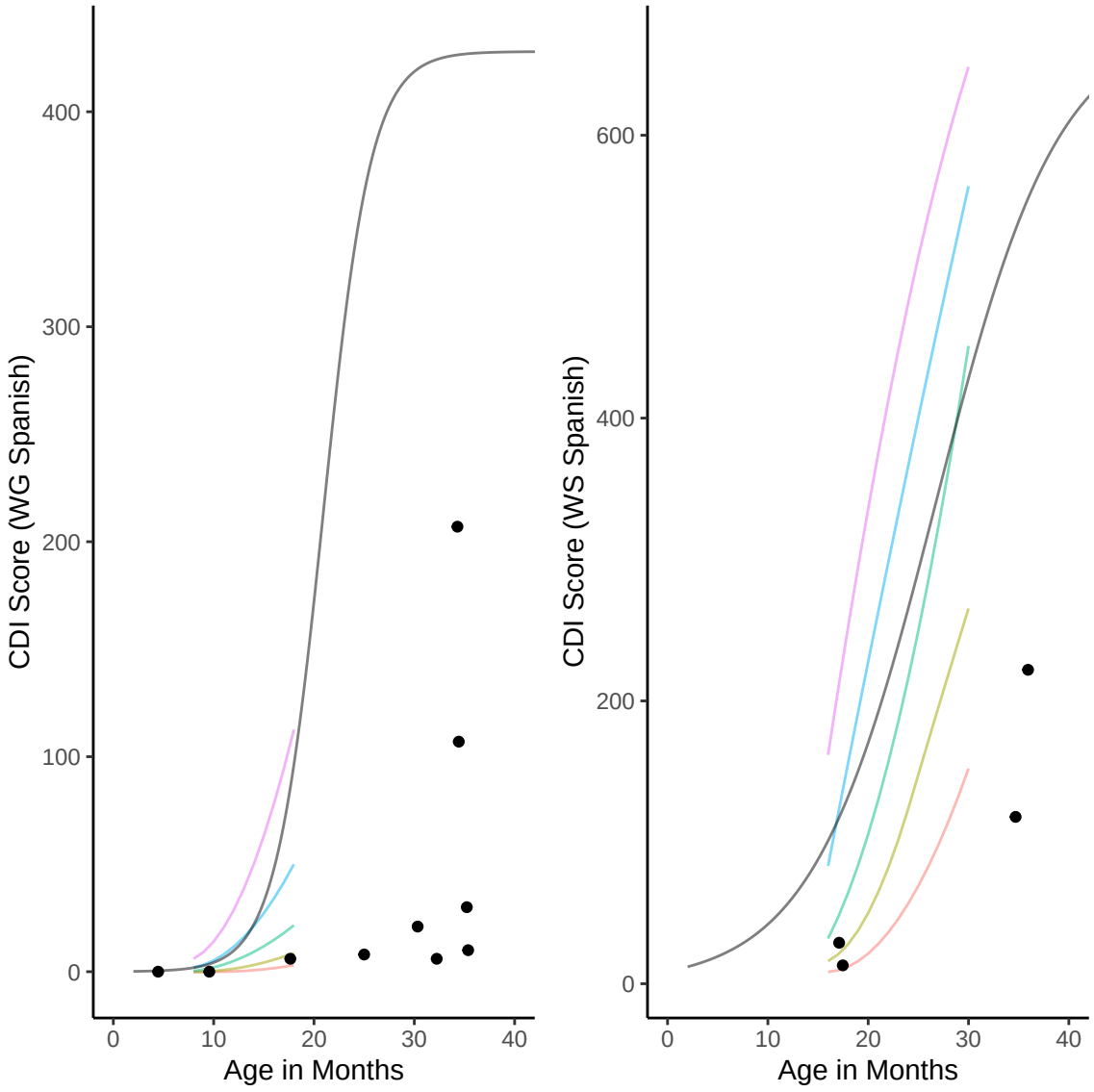
We next turn to the relationship between each of these variables and children's productive vocabulary, measured on the CDI. Figure **??** shows the vocabulary scores of children in our samples relative to norms for hearing children. Descriptively, we found widespread vocabulary delays on both Words and 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)).

The CDI is composed of two instruments, which differ in number of questions(the max score of 398 on Words and Gestures and 680 on Words and Sentences). For this reason, instead of using the raw number of words produced as our outcome variable, we use the difference (in months) between the child's chronological age and their predicted age for their vocabulary – we call this derived variable **vocabulary delay**.

To predict age from vocabulary score, we used the 50th percentile for productive

vocabulary from Wordbank data from 8586 typically-developing infants (Frank et al., 2017) to create binary logistic growth curves. The growth curves modeled the 50th percentile language trajectories for Words and Gestures and Words and Sentences for the American English and Mexican Spanish forms of the CDI. For each child, we took the number of words they produced divided by the number of words on the instrument, to give us the proportion of words produced. We used the proportion of words in an inverse prediction from the binary logistic regression curves to generate a predicted age; such for each possible CDI score, the growth curve provided the age that score would be achieved for the 50th percentile trajectory. We subtracted the predicted age from each child's chronological age to get the vocabulary delay variable.


CDI Score (WG English)
400
300
200
100
0
0
10
20
30
40
Age in Months
CDI Score (WS English)
600
400
200
0
0
10
20
30
40
Age in Months

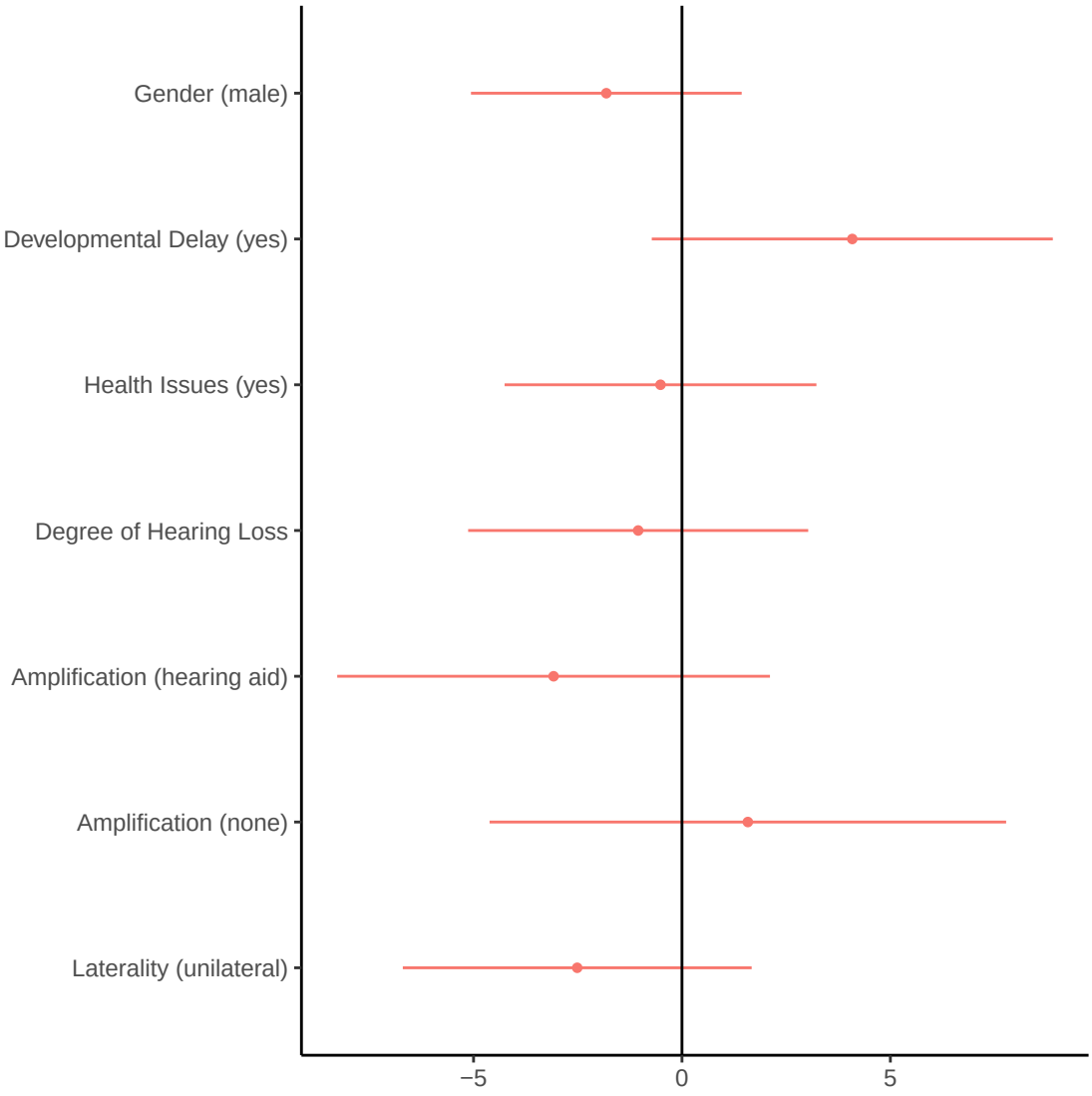


To look at the relationship between our predictor variables and vocabulary delay, we conducted multiple linear regression, using vocabulary delay as the outcome variable. We excluded the Hindi-speaking child from this section of the analysis due to concerns about comparing her score to the American English CDI norms.

Our full regression model included all variables except Language Background: Vocabulary Delay ~ Gender + Developmental Delay + Health Issues + Prematurity + Laterality + Degree + Amplification + Communication + Meets 1-3-6 + ServicesReceivedPerMonth. We 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). Based on this iterative process, we removed Prematurity, Communication, Meets 1-3-6, and ServicesReceivedPerMonth from the model.

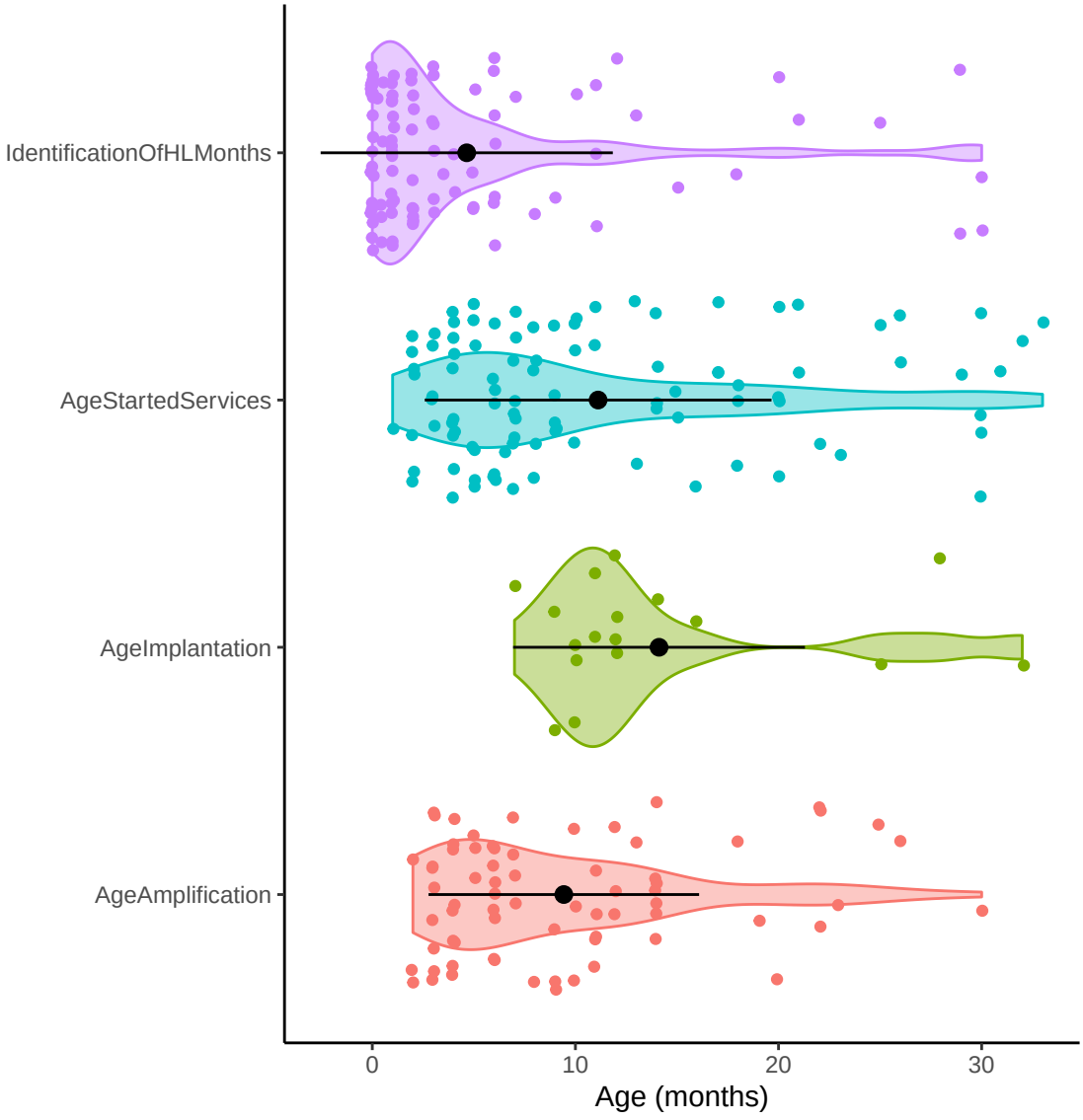
Our final model included: Vocabulary Delay ~ Gender + Developmental Delay + Health Issues + Laterality + Degree + Amplification. This model accounted for significant variance in children's vocabulary delay (adjusted-$R^2 = 0.07$, $p =$). There were significant main effects of gender, developmental delay, laterality, degree, health issues, and amplification. In this model, being male (ß = -1.81, p = 0.27), having a developmental delay (ß = 4.09, p = 0.095), bilateral hearing loss (ß = -2.51, p = 0.23), and more severe hearing loss predicted a larger delay (ß = -0.02, p = 0.61). Having a cochlear implant (ß = -3.08, p = 0.24)or hearing aid (ß = 1.58, p = 0.61) predicted a smaller delay, relative to no amplification. Presence of health issues indicated a smaller vocabulary delay (ß = -0.51, p = 0.78). Although we showed in Part I that relationships exist among several of these variables (e.g., degree and amplification), a vif test on our model revealed that each predictor was responsible for a unique share of the variance (all GVIF < 3; see table **??**; James, Witten, Hastie, and Tibshirani (2013)).



Part III: Meets136 success

Perhaps of greatest importance to clinicians and policymakers is the implementation and effect of existing policies. Lastly, we looked at the ages at which children received diagnosis and intervention, and how this mapped onto the 1-3-6 guidelines. Although we did not find 1-3-6 guidelines to significantly predict vocabulary delay in our sample, we wanted to examine the reach of 1-3-6 and discuss factors that may influence whether a given child receives early diagnosis and intervention. In this section, we provide a brief description of the implementation of 1-3-6 in our sample and describe the results of linear regression models for age at diagnosis and age at intervention.

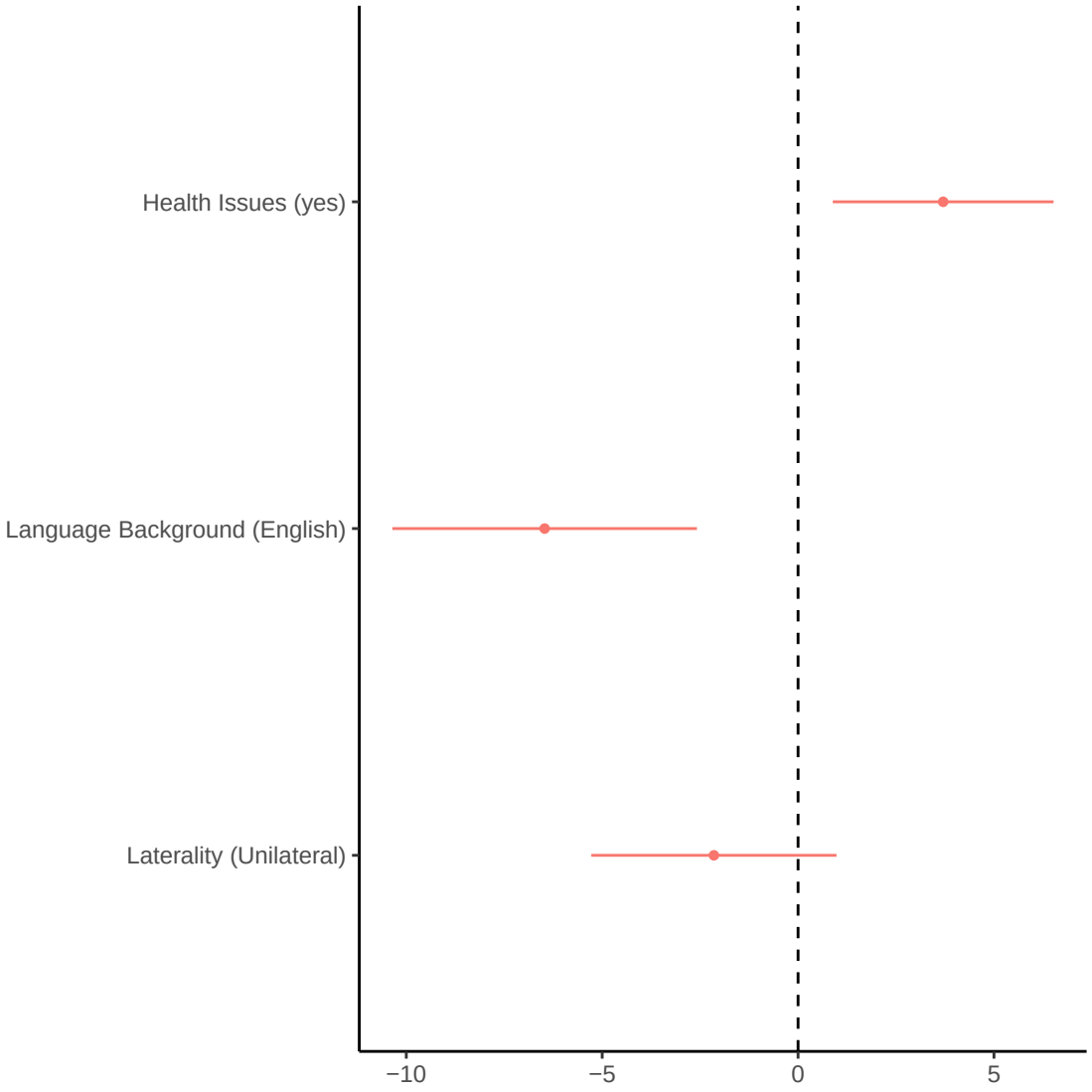
Overall, 37% of our sample met 1-3-6 guidelines for early diagnosis and intervention (see 2). Among the children for which screening information was available (n=68), 100% were screened at birth or during NICU stay. 69% of children received diagnosis by 3 months of age, and 39% began early intervention 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.



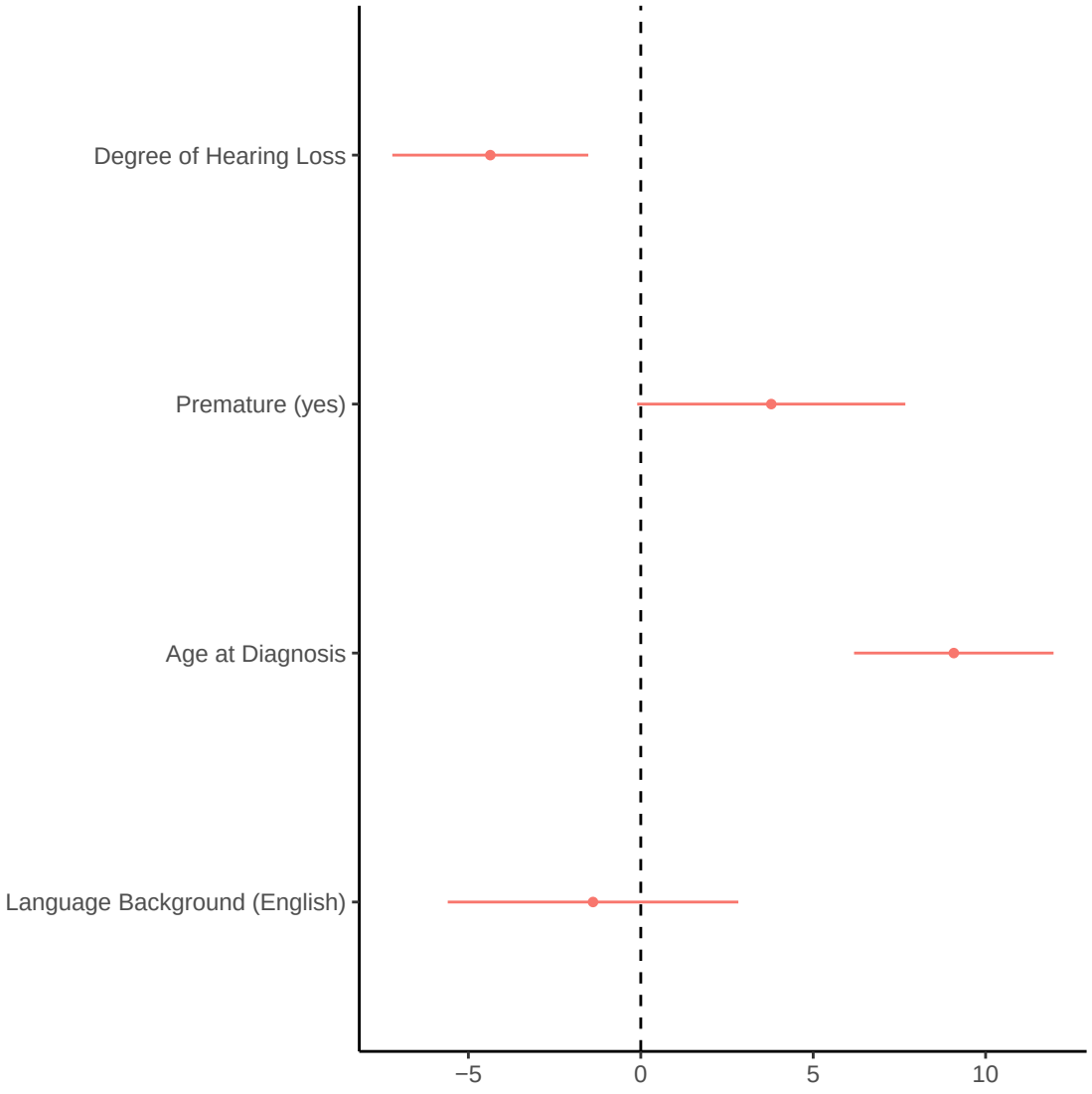
To better understand implementation of 1-3-6 guidelines, we zoomed in on diagnosis

and intervention. We created two linear regression models, one for age at diagnosis and one for age at intervention. For each model, we started with the set of predictors that would have been present prior to or during diagnosis or intervention, respectively. We then pared down each model using stepwise regression by AIC (MASS package), using the process described above in Part II of Results.

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. Under the best fit model (R^2=0.16 , p=0.00), children with no additional health issues (ß = 3.7, p = 0.011), children from English-speaking households (ß = -6.47, p = 0.0014), and children with unilateral hearing loss (ß = -2.15, p = 0.18) predicted earlier diagnosis. This model accounted for roughly 16.41% of the variance in age at diagnosis.



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 Intervention ~ prematurity + degree + age at diagnosis + language background (R^2=0.43 , p=0.00). Prematurity (ß = 3.78, p = 0.056), less severe hearing loss (ß = -0.09, p = 0.003), later diagnosis (ß = 0.65, p = 1.9e-08), and coming from a non-English-speaking household (ß = -1.38, p = 0.52) predicted later intervention and accounted for roughly 43.41% of the variance in age at intervention.



Discussion

(This isn't ready yet)

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.

How are child-level variables intertwined?

In our sample, we found significant overlap among demographic, audiological, and clinical variables. Prematurity, health issues, and developmental delay frequently co-occurred, such that there was a moderate relationship between each of these variables (cramer's V = 0.38 - 0.52, $p < .0007$). Children with one of these conditions (prematurity, developmental delay, health issues) were more likely to have any other condition. This is not surprising. Many conditions that cause developmental delays have a high incidence of health issues (e.g., heart problems in Down Syndrome; vomiting and seizures with hydrocephalus), and it is well documented that there is a higher incidence of developmental delay in preterm infants (**???**; Pierrat et al., 2017). Children born premature, especially those born extremely premature, are at increased 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 literature points to increased risk of language delay for children with developmental delays (Chapman, 1997; Kristoffersen, 2008; Weismer et al., 2010) and children born premature (CITE), with differential effects based on the nature of the developmental delay (Cupples et al., 2014, 2018) or the gestation duration (CITE). Together, these risks may interact and multiply. In our sample, we also had a large range of health conditions (76 unique conditions in our sample of 100 children; see 3 and Appendix XXX for more detailed information about comorbidities), and it appears probable that those conditions would vary in whether and how they influence vocabulary growth. Unfortunately, we lack sufficient Ns to measure the unique effect of each condition in children with hearing loss. 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. Additionally, in our sample, children with developmental delays were

considerably more likely to receive >10 services per month, perhaps accounting for 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 (**???**; **???**; **???**; **???**) and spoken language vocabulary and grammar for mild hearing loss (Walker et al., 2015). *Add paragraph here about how while this overlap is not necessarily surprising, it should tell us to be careful. how replicable are controlled lab samples when X% of children with X have Y*

skipping discussion part II while we sort out growth curves

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).

In support of this policy, Ching and colleagues (Ching et al., 2013) find a pronounced effect of early diagnosis. In this study, which was conducted in Australia, researchers compared groups of children in regions that had implemented universal newborn hearing screenings to children in regions that had not yet implemented UNHS. Children were otherwise similar in diagnostic, demographic, and intervention characteristics. The UNHS group had higher global language scores. By contrast, in our sample, by dint of accepting all children receiving early intervention services in one state, we were able to document naturally occurring variance in who received on-time diagnosis and intervention. We found that some of the variance in age at diagnosis and intervention could be explained by children's demographic and audiological characteristics.

Age at Diagnosis. In the case of diagnosis, presence of health issues, primary language in the home, and laterality of hearing loss accounted for 0.16% 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 (ß = 3.7, p = 0.011) months later than infants without health issues. One possible explanation is that the health issues caused acquired hearing loss that wouldn't be detected by the NBHS, thus delaying identification of hearing loss. Of the 76 unique health issues experienced by children in the sample, only 9 conditions might cause acquired hearing loss (i.e., meningitis, sepsis, jaundice, seizures, hydrocephalus, MRSA, anemia, frequent fevers, cytomegalovirus, affecting 16 out of 36 children with health issues in our sample. Another possible explanation is that the health issues required more pressing medical attention than the possible hearing loss, and that families and medical providers had to prioritize treatment for the health issue (e.g., surgery for congenital heart defect) over diagnostic audiology services.

Infants from Spanish-speaking families were diagnosed 3.78 months later than infants

from English-speaking families. This may be due to cultural differences in attitudes towards deafness (**???**; **???**, @steinberg2003; **???**) or it may result from a lack of linguistically accessible and culturally appropriate audiology services (CITE). Only 5.6% of American audiologists identify as a bilingual service provider (ASHA, 2019), and services from a monolingual provider may be insufficient. Caballero et al. (2017) found that Hispanic-American parents of DHH children want more concrete resources, comprehensive information, and emotional support from their audiologist. The majority of audiologists in a survey responded that they use ad-hoc interpreters (e.g., client's family) to overcome language barriers. Survey respondents reported that language barriers presented a major challenge in working with Spanish-speaking families, specifically in obtaining the child's case history and providing recommendations for follow-up services (**???**). Compounding this issue, Hispanic-Americans infants may be more likely to than non-Hispanic-American infants to experience hearing loss (Mehra, Eavey, & Keamy, 2009).

Intervention. Our age at intervention model accounted for 0.43% of the variance with main effects of degree of hearing loss, prematurity, language background, and age at diagnosis. Prematurity and language background were not significant main effects in our model, but remained in the model because their inclusion improved model fit.

More severe hearing loss predicted earlier diagnosis, such that for every additional 10 dB HL, predicted age at diagnosis was 4.02 weeks earlier. This parallels a 2003 finding by Harrison, Roush, and Wallace (2003) in which severe-to-profound hearing loss diagnosed 5 months later than mild-to-moderate hearing loss (for children with a known cause of hearing loss; 2 months later for hearing loss with unknown cause).

One reliable predictor of age at start of services was 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.

Unsurprisingly however, these two variables are highly associated, 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 children with hearing loss enroll in early intervention services [(**???**) https://www.cdc.gov/ncbddd/hearingloss/2016-data/01-data-summary.html). While this represents a tremendous step forward in prompt early intervention services, early intervention may not be early enough. Less than 39% of our sample of children in early intervention meet the 6-month EHDI benchmark. Furthermore, critically, there exists a significant chunk of the population who aren't included in this analysis and for whom we don't have any data because they have not been enrolled in services by 36 months. The AAP estimates that almost 36% of infants who do not pass a newborn hearing screening are lost to follow-up. Assuming that the population of children in early intervention only represents two thirds of the population with hearing loss, our data suggest that the actual proportion of DHH children who receive intervention by the EHDI-recommended 6 months may be closer to 26. These children may not receive clinical support until school-age or later.

We acknowledge that for both age at diagnosis and age at intervention, our models accounted for only a modest portion of the variance (R^2s=0.16&0.43, respectively), and it is likely that other factors that we did not measure (e.g., SES or geographic proximity to service providers) also play a role in when an individual receives diagnosis or therapeutic services. With that being said, based on the results of these two models, 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.

Limitations

These analyses were very much exploratory, and there were many possible analytic routes. In the interest of transparency, these data are available on our OSF page (XXX), and all of the code used to generate the statistics and figures from this article are available on Github (XXX). With that being said, our results largely confirmed past studies (e.g., XXX) or clinical intuitions. 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, so we cannot say with any certainty how these results would hold up in other geographic regions, particularly with regard to diagnosis and intervention policies which vary by country and by state (NAD, n.d.). Based on Gallaudet's 2014 Annual Survey of Deaf and Hard-of-Hearing Children and the SEELS report of elementary and middle school aged DHH children (Blackorby & Knokey, 2006; Institute, 2014), our sample largely represents the national DHH population in XXX .

There were several variables reported to influence outcomes for DHH children that we did not have access to (e.g., SES, aided hearing, maternal education, parental hearing status). Measuring these variables may have provided additional predictive power. For more information on these variables, see XXX, XXX, XXX, XXX.

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 Ns, which are often difficult to achieve in research with DHH children (XXX?), 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 this diverse population and varied outcomes.

Conclusion

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

Acknowledgments

Footnotes: Despite exciting, increasing, and converging evidence for benefits of early sign language exposure (e.g., Schick, De Villiers, De Villiers, & Hoffmeister, 2007; Clark et al., 2016; Davidson, Lillo-Martin, & Pichler, 2014; Hrastinski & Wilbur, 2016; Magnuson, 2000; 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 child in our sample used a legitimate signed language, we focus on spoken language development. *cite gallaudet national survey to show that our sample had many fewer signers than US as a whole*

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Table 1

Summary of findings of CDI studies in DHH children

Study	Population	Gender	1-3-6	Laterality	Degree	Amplification	Communication	Comorbidities
Yoshinaga-Itano et al., 2017	8-39 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)

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

Table 4

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

Table 5

Language and communication characteristics of the sample

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
Meets 1-3-6	Categorical	Yes / No
Prematurity	Categorical	Full-term / Premature
Services Received Per Month	Continuous	0-43 services per month (mean (SD): 6 (6.4))
Type of Hearing Loss	Categorical	Sensorineural / Conductive / Mixed
CDI - Words Produced	Continuous	0-635 words (mean (SD): 61 (111.2))

term	estimate	std.error	statistic	p.value
(Intercept)	10.9704460	4.3239663	2.5371257	0.0137944
Gendermale	-1.8147633	1.6234338	-1.1178548	0.2680850
DevelopmentalConcernsyes	4.0861442	2.4050613	1.6989771	0.0945035
HealthIssuesyes	-0.5149686	1.8700530	-0.2753765	0.7839732
LateralityUnilateral	-2.5129659	2.0921095	-1.2011637	0.2344080
HLworse	-0.0222659	0.0432189	-0.5151885	0.6083143
AmplificationHA	-3.0798163	2.5950698	-1.1867952	0.2399863
Amplificationnone	1.5804807	3.0978278	0.5101900	0.6117902

	GVIF	Df
Gender	1.185960	1
DevelopmentalConcerns	1.513090	1
HealthIssues	1.463388	1
Laterality	1.144936	1
HLworse	1.843961	1
Amplification	2.561074	2

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