Characterizing North Carolina's Deaf/Hard-of-Hearing Infants and Toddlers: Predictors of Word Learning, 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. In the following paragraphs, I will 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-occuring 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 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 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 (TC) 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 TC 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 TC (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 (CDI-WG) is normed for 8–18-month-olds. On CDI-WG, 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 (CDI-WS) is normed for 16-30-month-olds. On CDI-WS, 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 and third goal above, we did not have specific hypotheses and sought to provide descriptive information about 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.

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 WG or WS version of the CDI based on clinician judgement. Children who were too old for WG, but who were not producing many words at the time of assessment, were often given WG (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. Families in this sample either spoke English or Spanish. For one child, who was adopted from India at 28 months, we recorded the primary language as Hindi, athough 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 ≤ 3 mo. and an age of intervention ≤ 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.

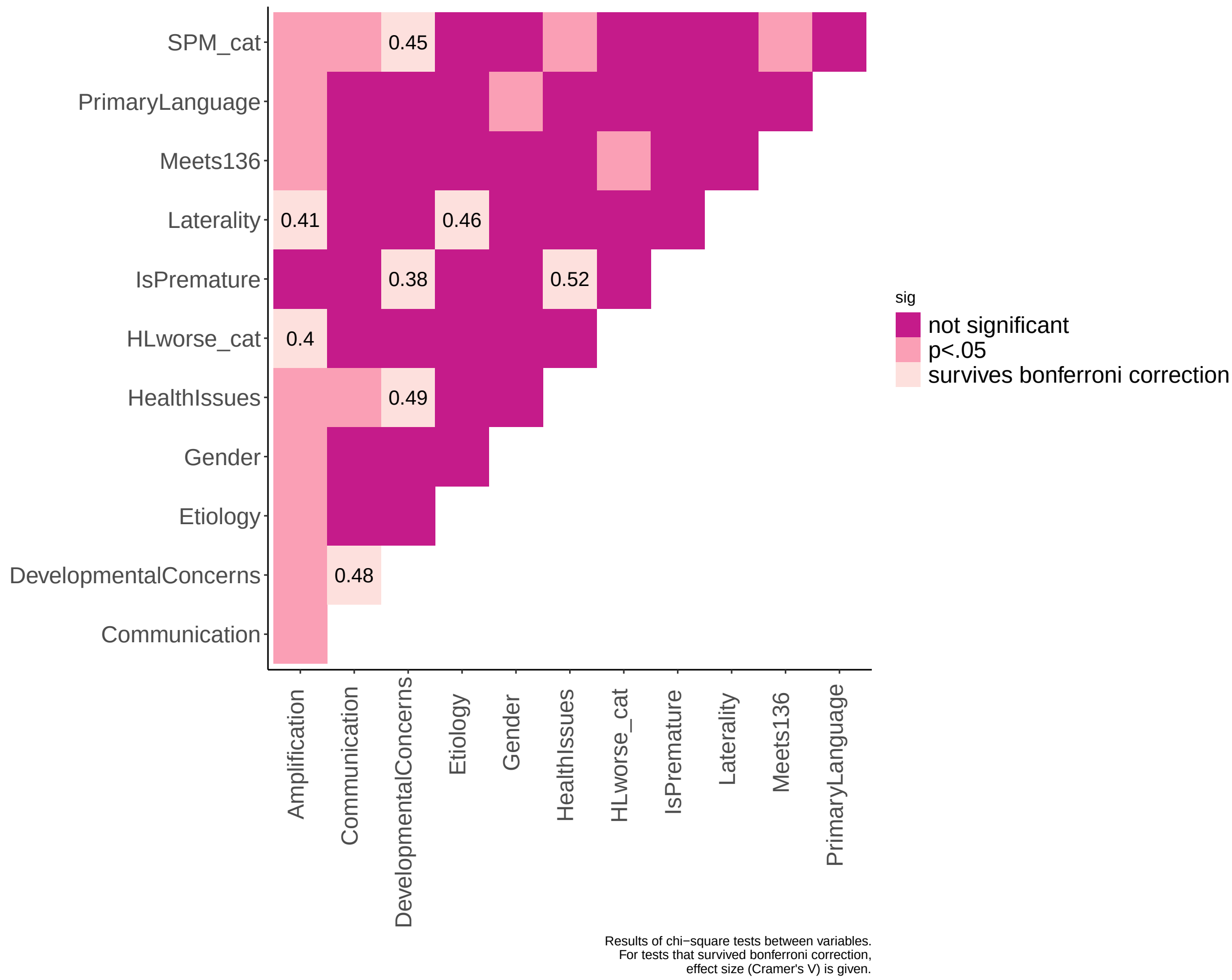
Results

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

Before we explore how these variables may be related to vocabulary, we would like to describe the variables' relationships to each other. Our goal in doing so is to demonstrate that many of these characteristics are not distributed randomly throughout the population. We approach this with bonferroni-corrected chi-square tests between each of our variables (gender, laterality, health issues, developmental delays, prematurity, language background, 1-3-6, degree of hearing loss (binned into mild, moderate, severe/profound), etiology, services received per month (binned into 0-3, 4-10, and >10), and amplification).

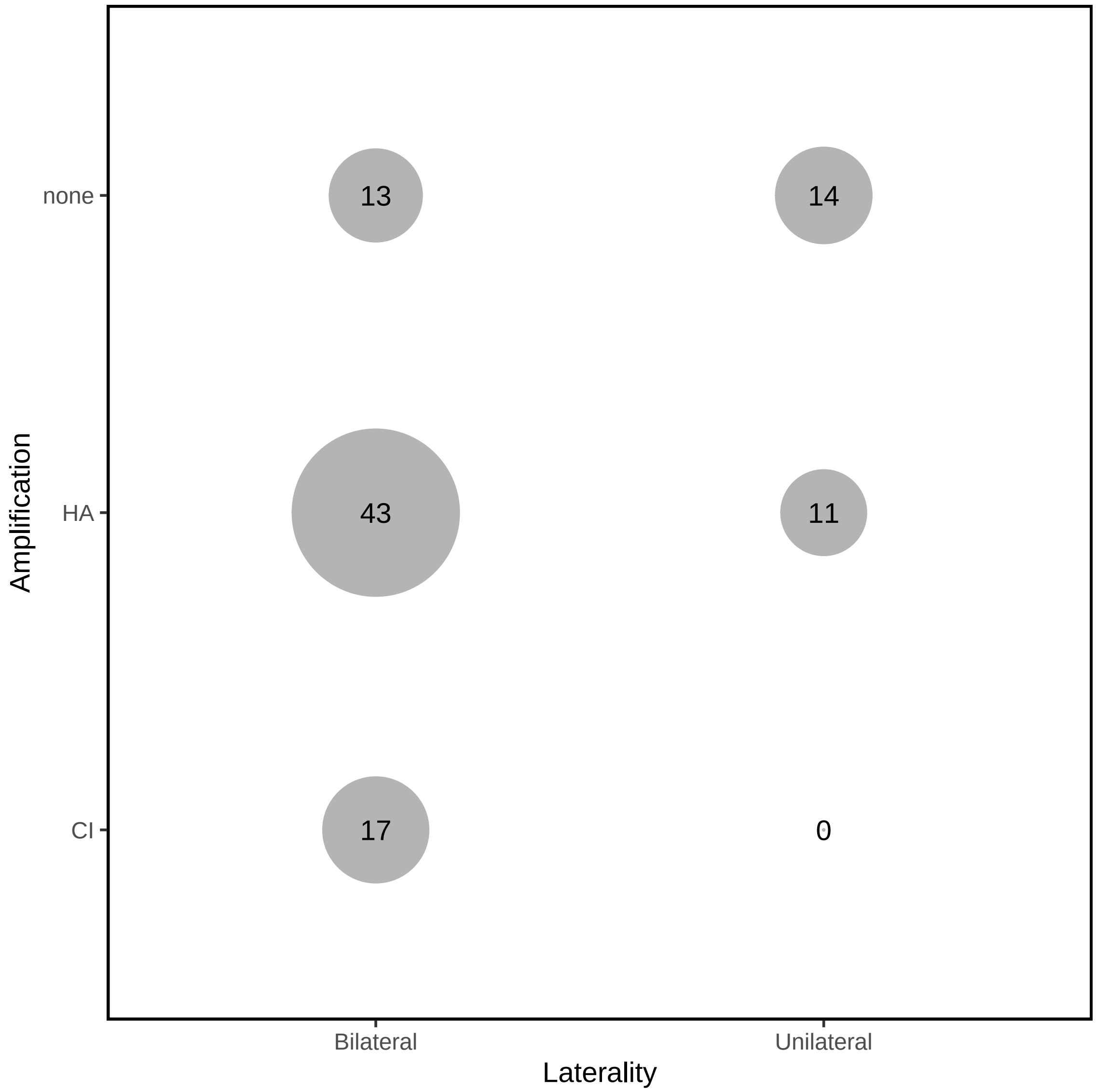
Bonferroni-corrected alpha for this set of analyses was $p < 0.0007$. Of the 66 combinations of variables, p < .05 for 22, and 8 survived bonferroni correction. We are only discussing the results of tests that survived bonferroni correction, but the full set of results can be found in figure **??**.

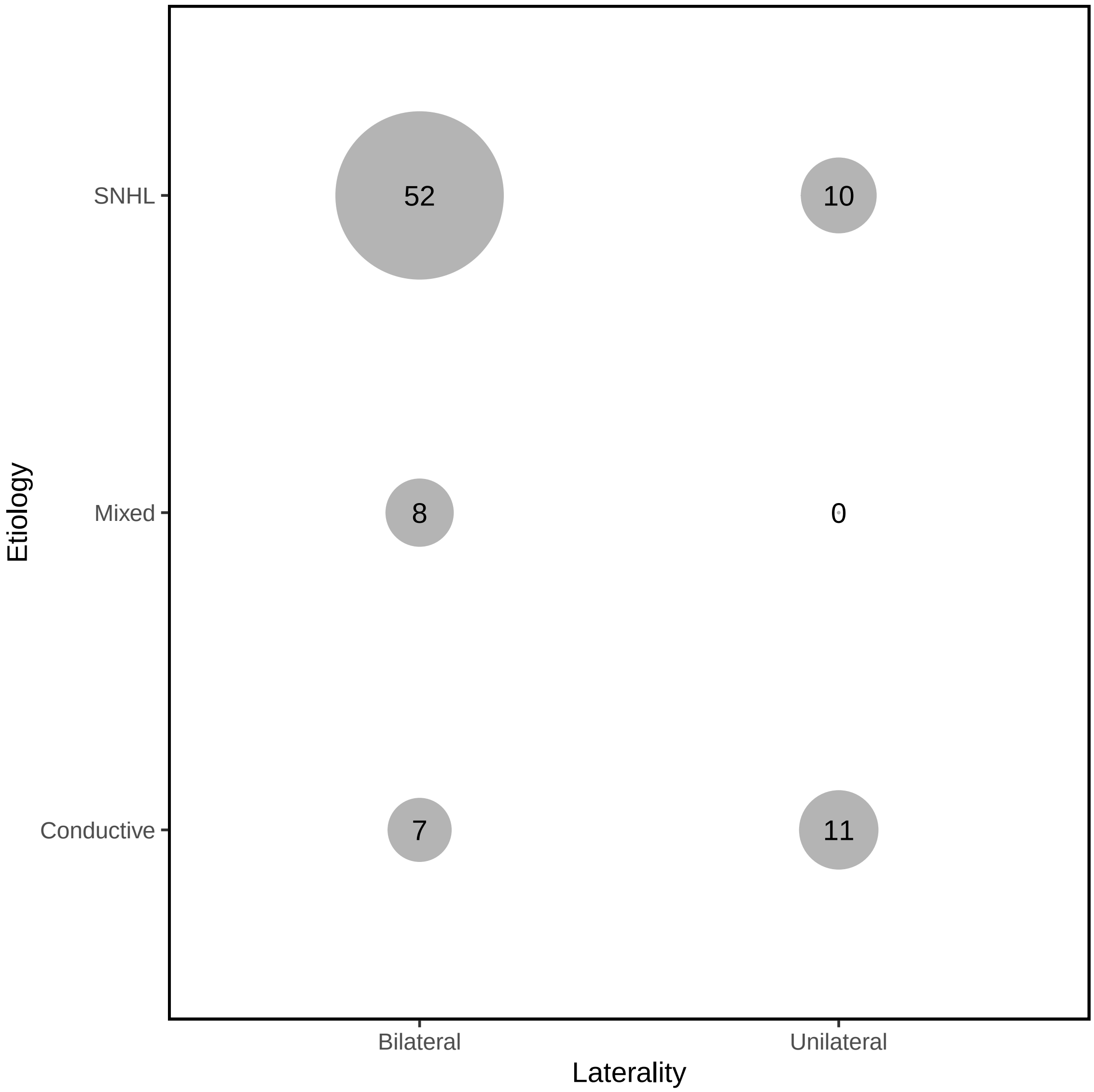


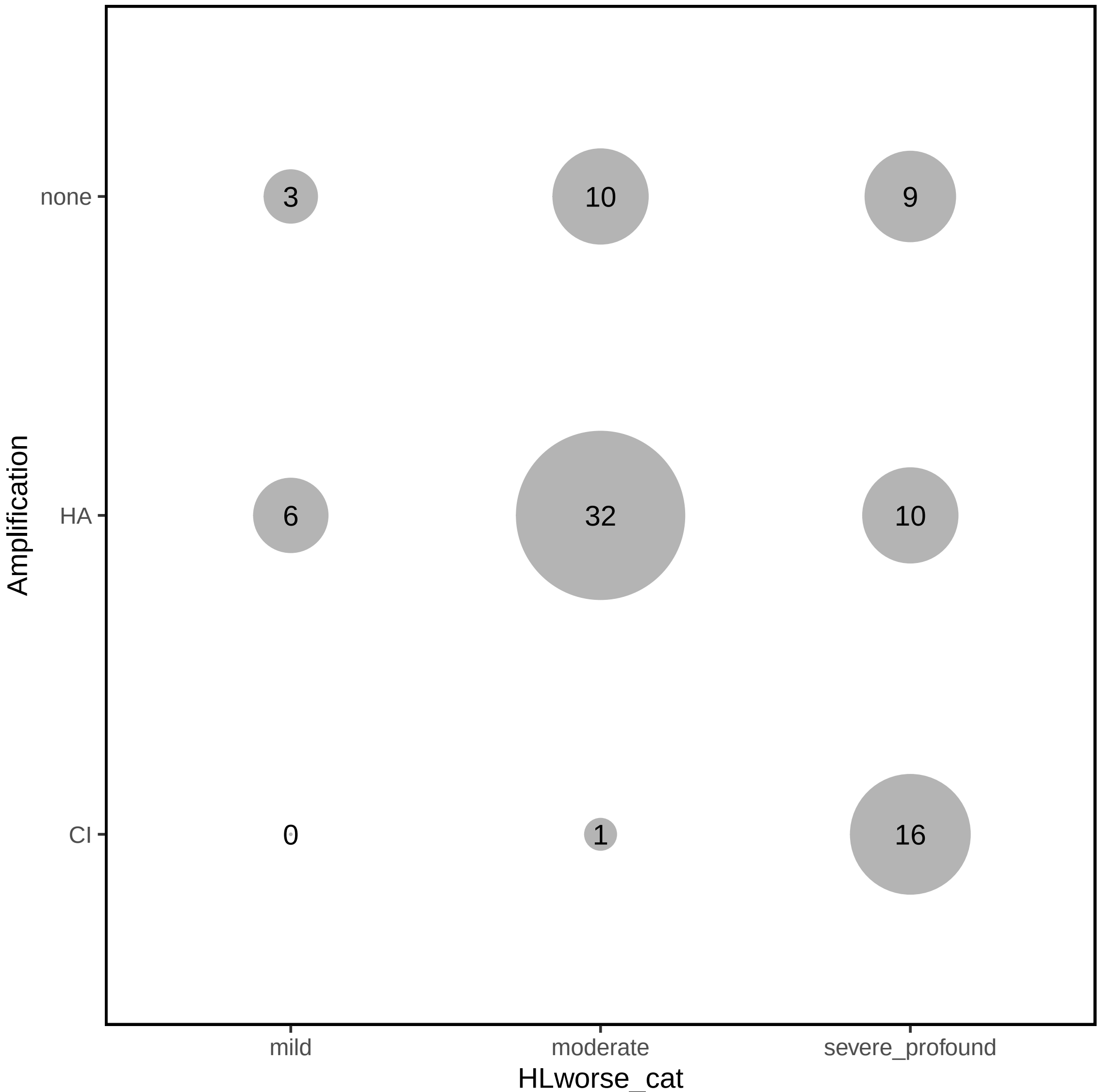
From this set of analyses, we found 8 relationships between variables that survived bonferroni correction. Health issues, developmental delays, and prematurity were highly interrelated in our sample, such that children born premature were more likely to also experience health issues (X^2 (1, N = 98) = 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

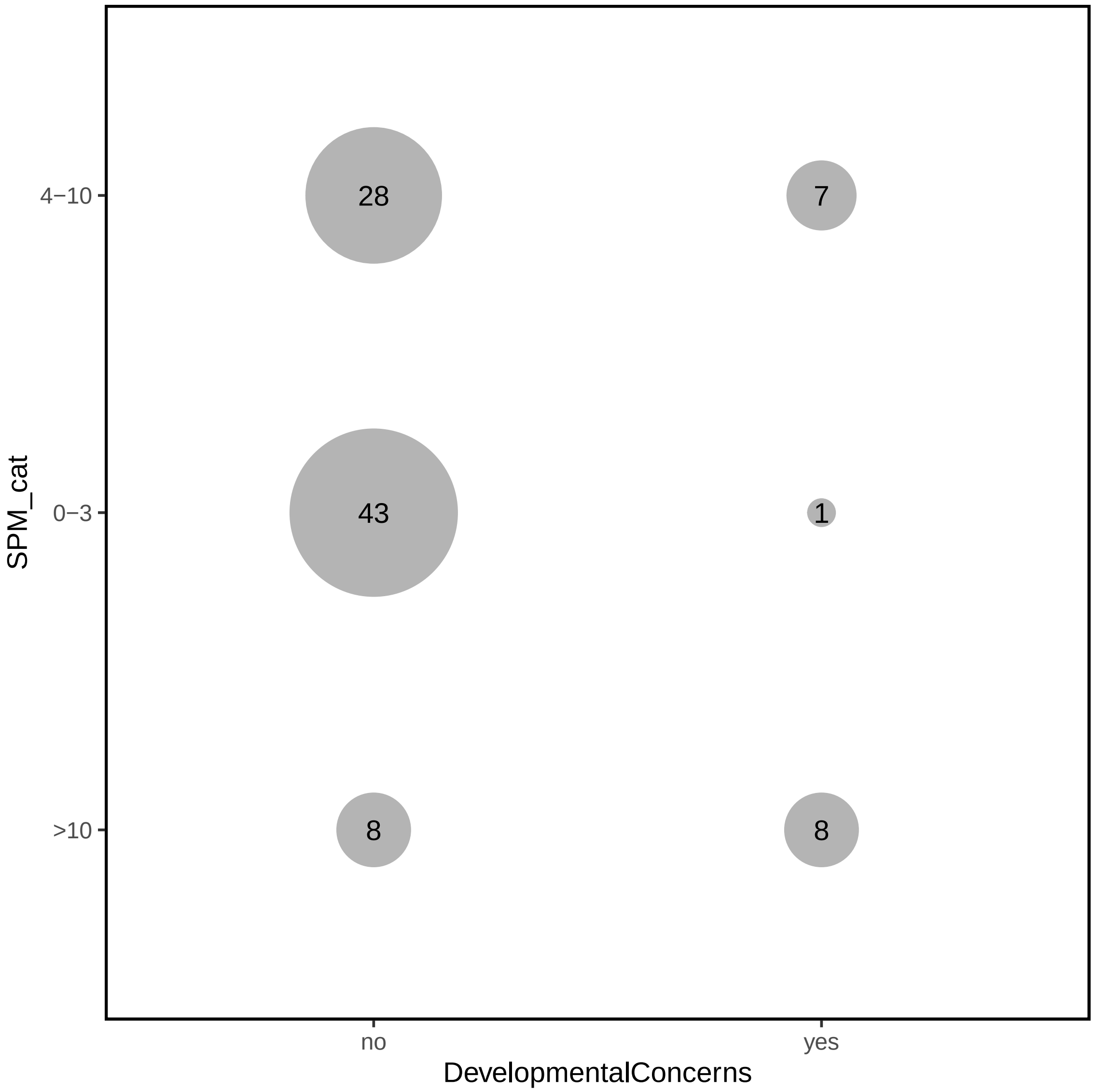
(X^2 (2, N = 95) = 19.48, p = 5.9e-05) and were more likely to use total communication (X^2 (2, N = 98) = 22.51, p = 1.3e-05). Likewise, children who used total communication received more services per month than children using spoken language (X^2 (4, N = 95) = 16.67, p = 0.0022).

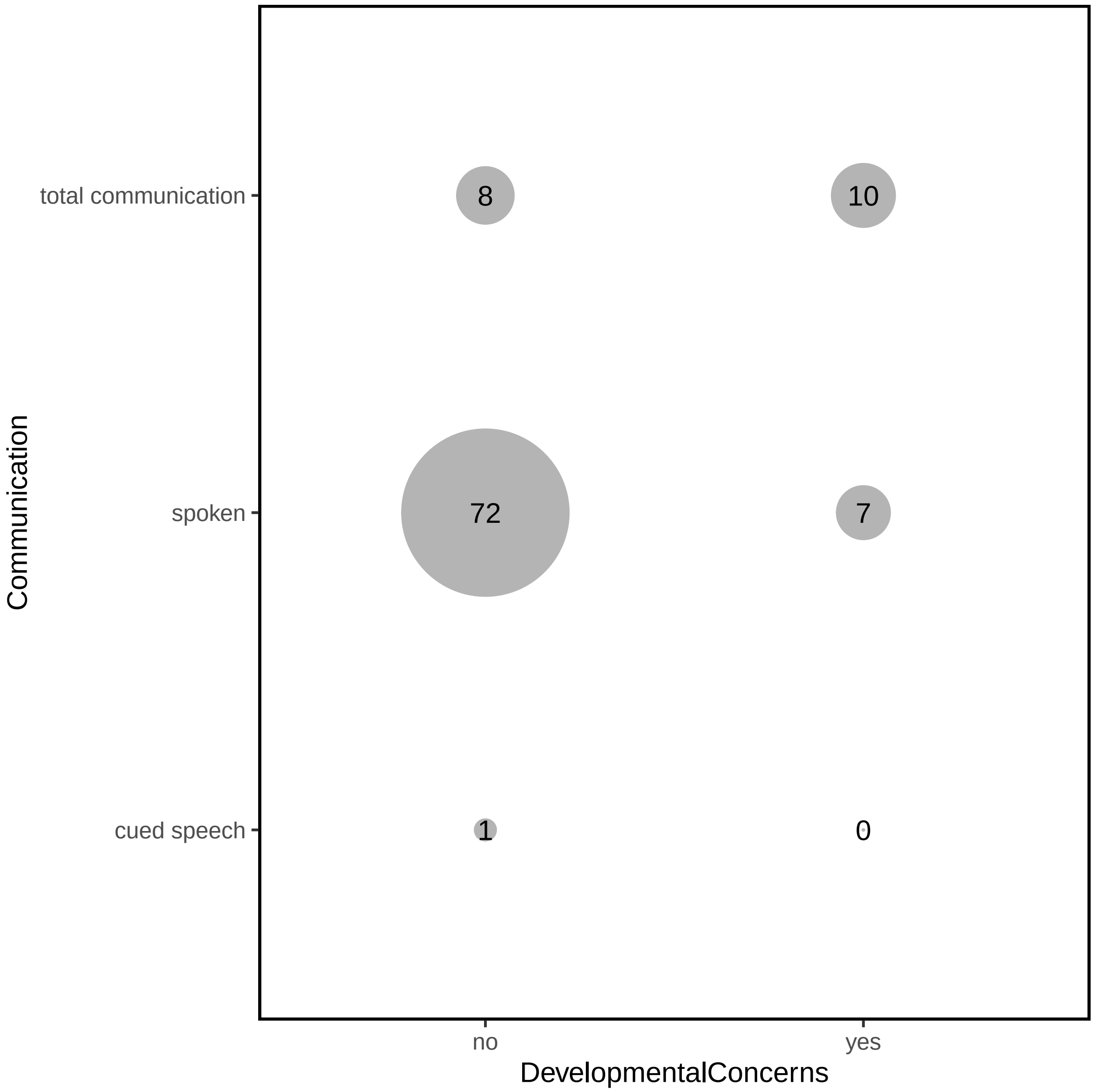
We also found relationships among many of the audiological characteristics. There was a significant relationship between laterality and etiology (X^2 (2, N = 88) = 18.29, p = 0.00011), such that children with conductive hearing loss were more likely to have unilateral hearing loss, children with sensorineural 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).


Amplification
none
HA
CI
13
14
43
11
17
0
Bilateral
Unilateral
Laterality

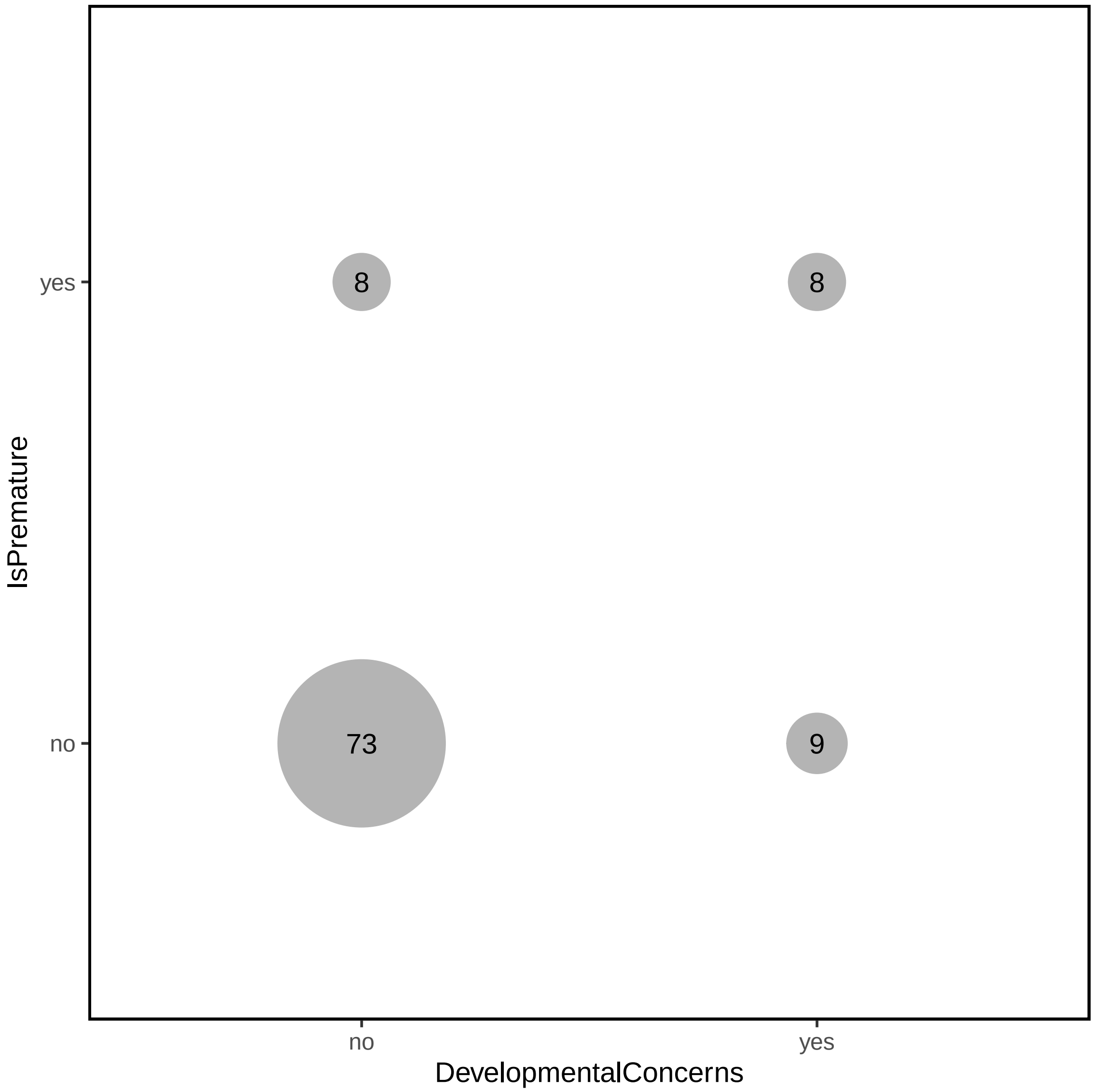

SNHL
Mixed
Conductive
Etiology
52
10
8
0
7
11
Bilateral
Unilateral
Laterality

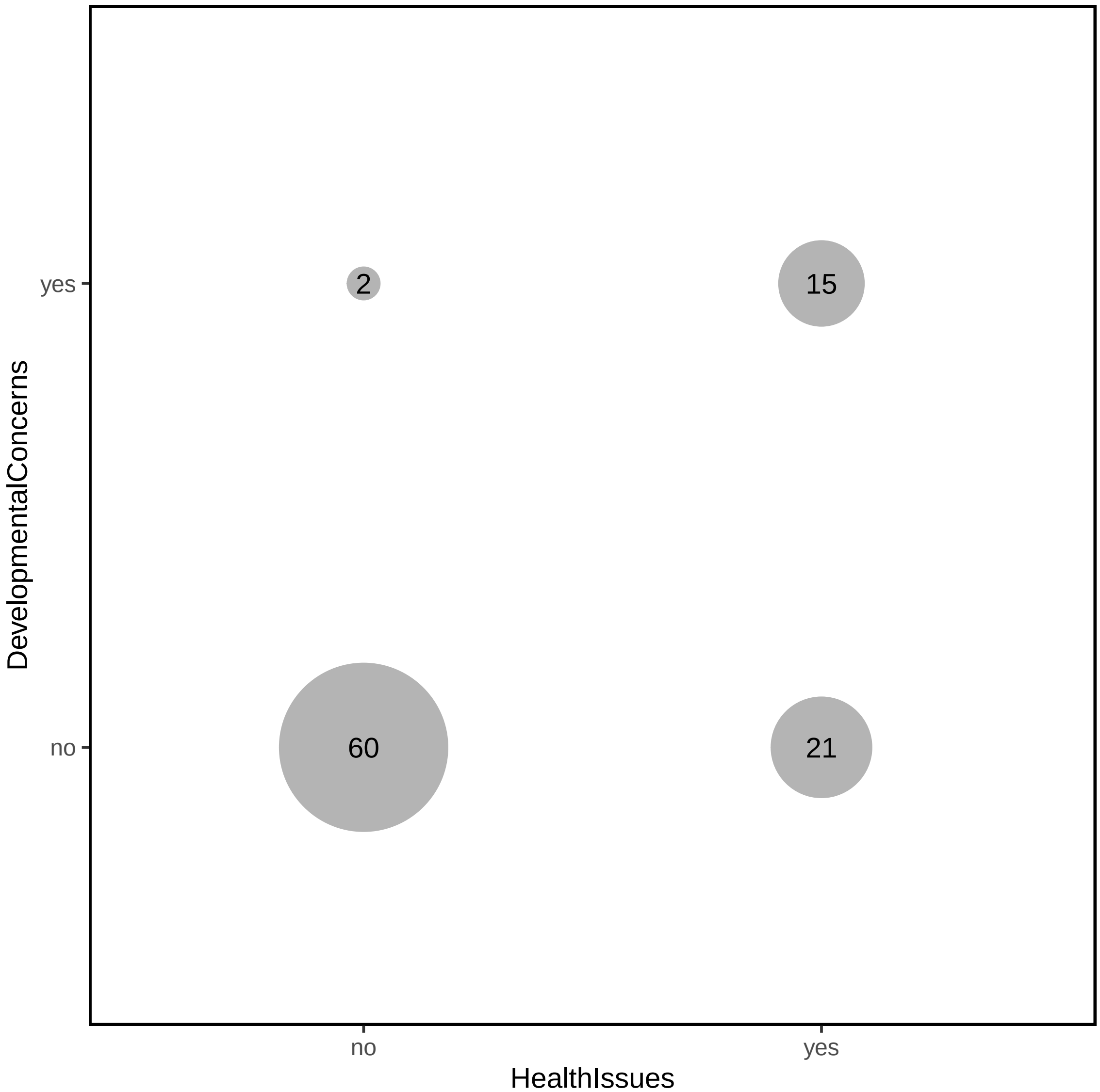

none
HA
CI
Amplification
3
10
9
6
32
10
0
1
16
mild
moderate
severe_profound
HLworse_cat

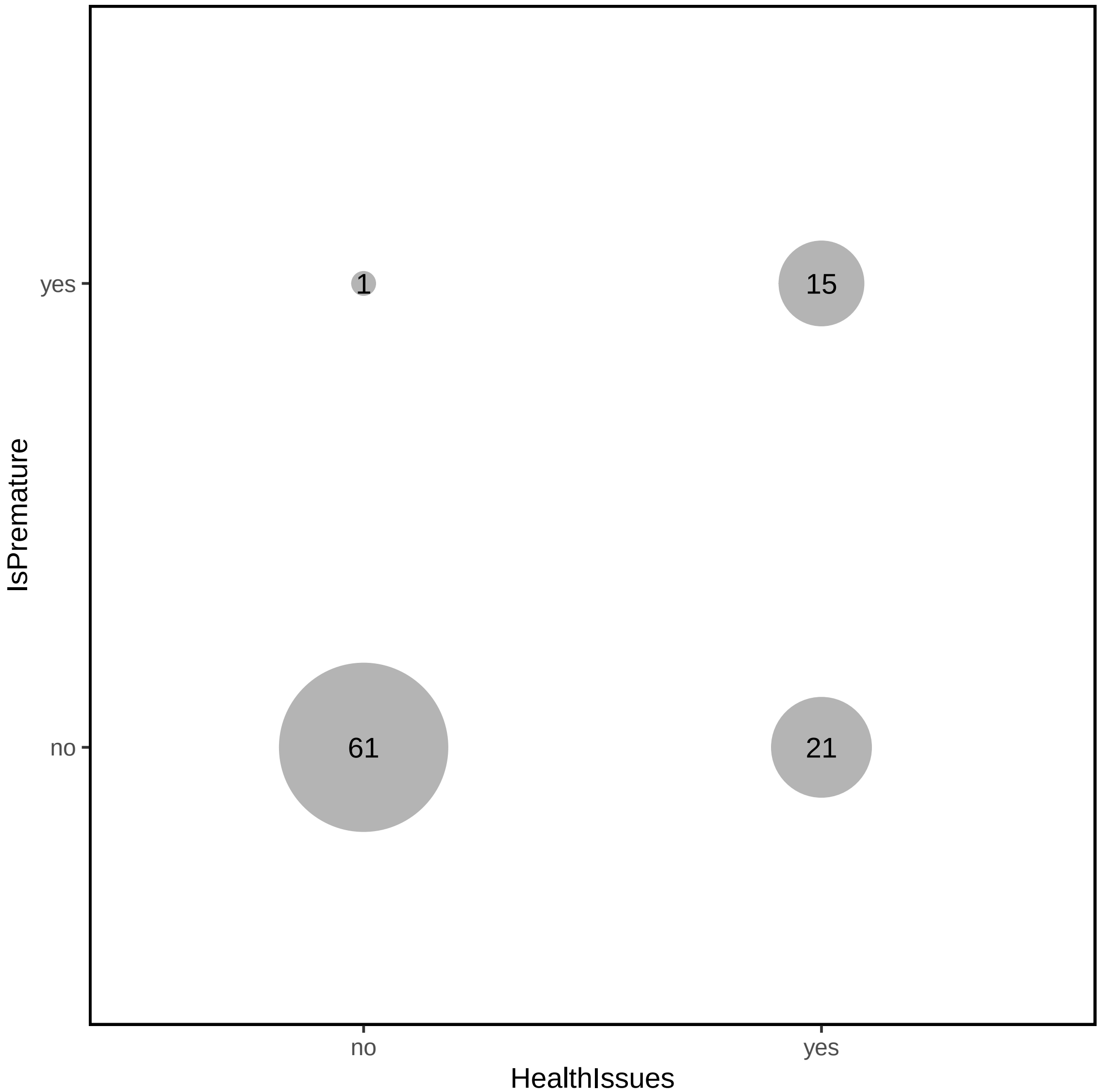

SPM_cat
4–10
0–3
>10
28
7
43
1
8
8
no
yes
DevelopmentalConcerns



total communication
spoken
cued speech
Communication
8
10
72
7
1
0
no
yes
DevelopmentalConcerns


IsPremature
yes
no
8
8
73
9
no
yes
DevelopmentalConcerns


DevelopmentalConcerns
yes
no
2
15
60
21
no
yes
HealthIssues

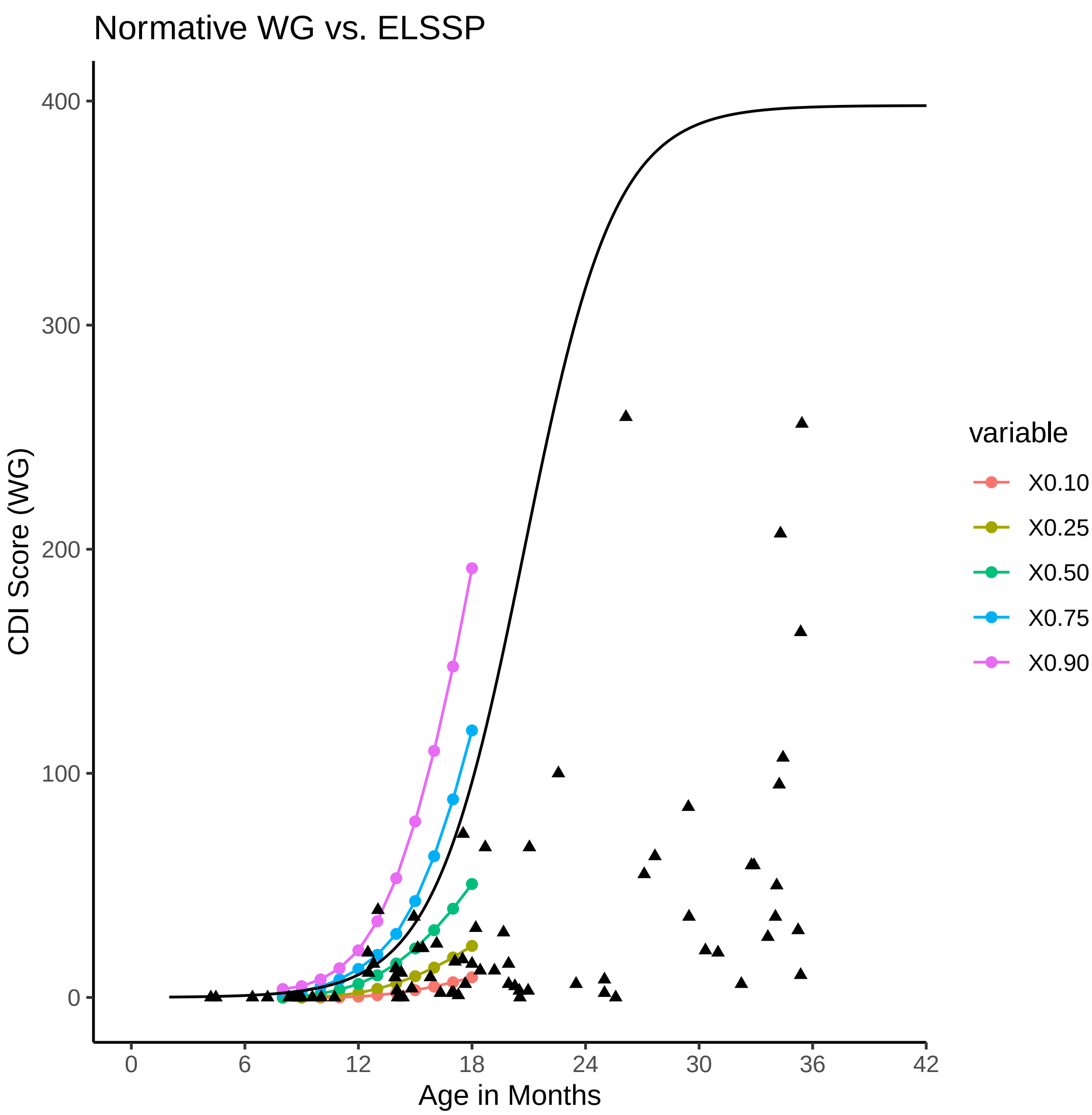


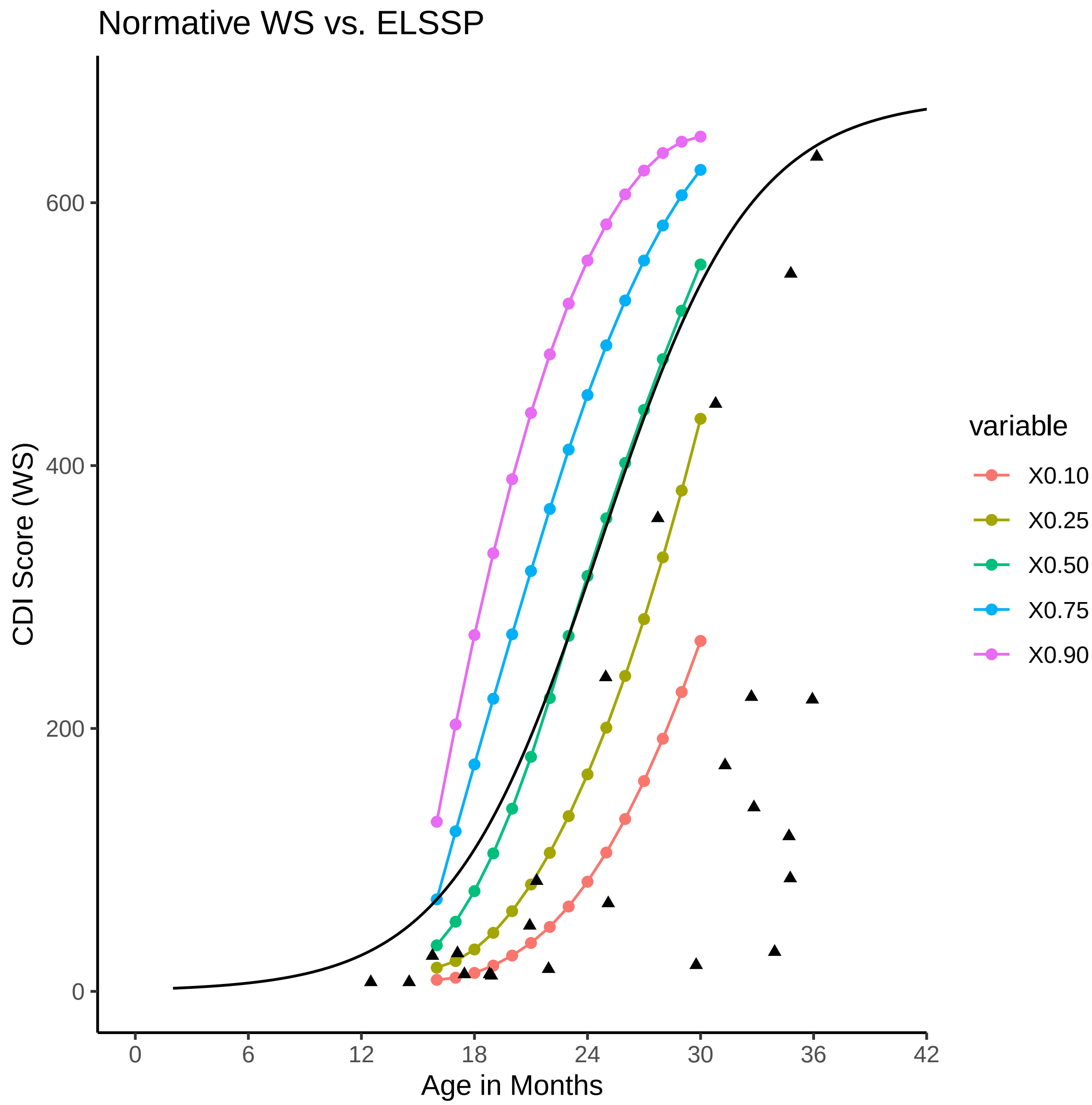
Part II: Influence on vocabulary

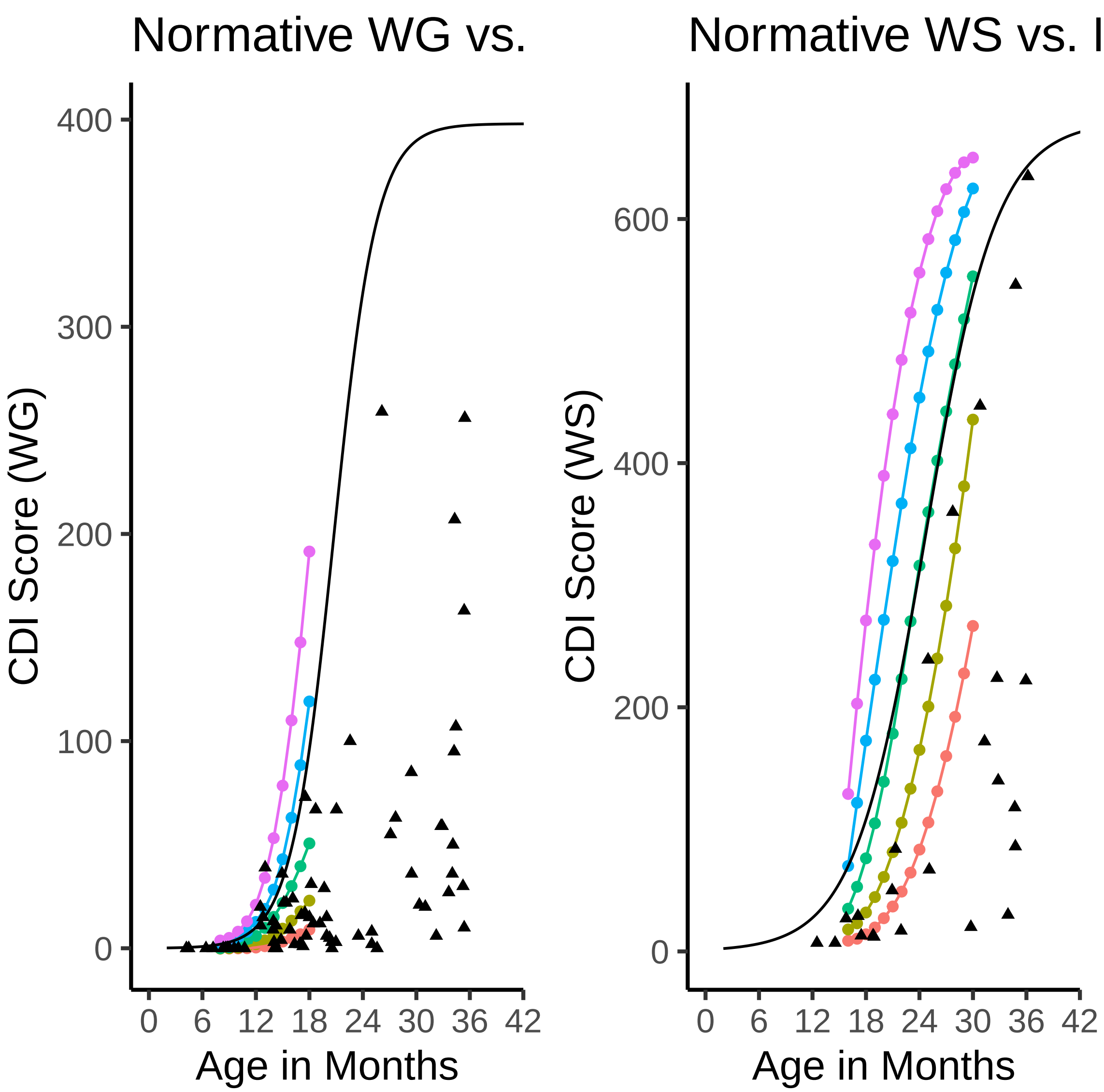
We next turn to the relationship between each of these variables and children's productive vocabulary, measured on the CDI. Figure **??** shows the vocabulary scores of children in our samples relative to norms for hearing children. Descriptively, we found widespread vocabulary delays on both Words and Gestures and Words and Sentences, with the majority of DHH children testing around or below the 25th percentile for hearing children.

The CDI is composed of two instruments, which are typically given to different age groups, but in this sample, are administered based on clinician assessment of the child's language ability. The two instruments differ in number of questions; the max score is 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 (8,300 typically-developing infants; Frank et al. (2017)) to create a binary logistic growth curve. The growth curve modelled the 50th percentile language trajectories for WG-CDI and WS-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: predicted_age = (log(proportion / (1-proportion)) - b0)/ b1 (cite). We subtracted the predicted age from the chronological age to get the vocabulary delay variable.


Normative WG vs. ELSSP
CDI Score (WG)
400
300
200
100
0
0
6
12
18
24
30
36
42
Age in Months
variable
X0.10
X0.25
X0.50
X0.75
X0.90


Normative WS vs. ELSSP
CDI Score (WS)
0
200
400
600
Age in Months
0
6
12
18
24
30
36
42
variable
X0.10
X0.25
X0.50
X0.75
X0.90

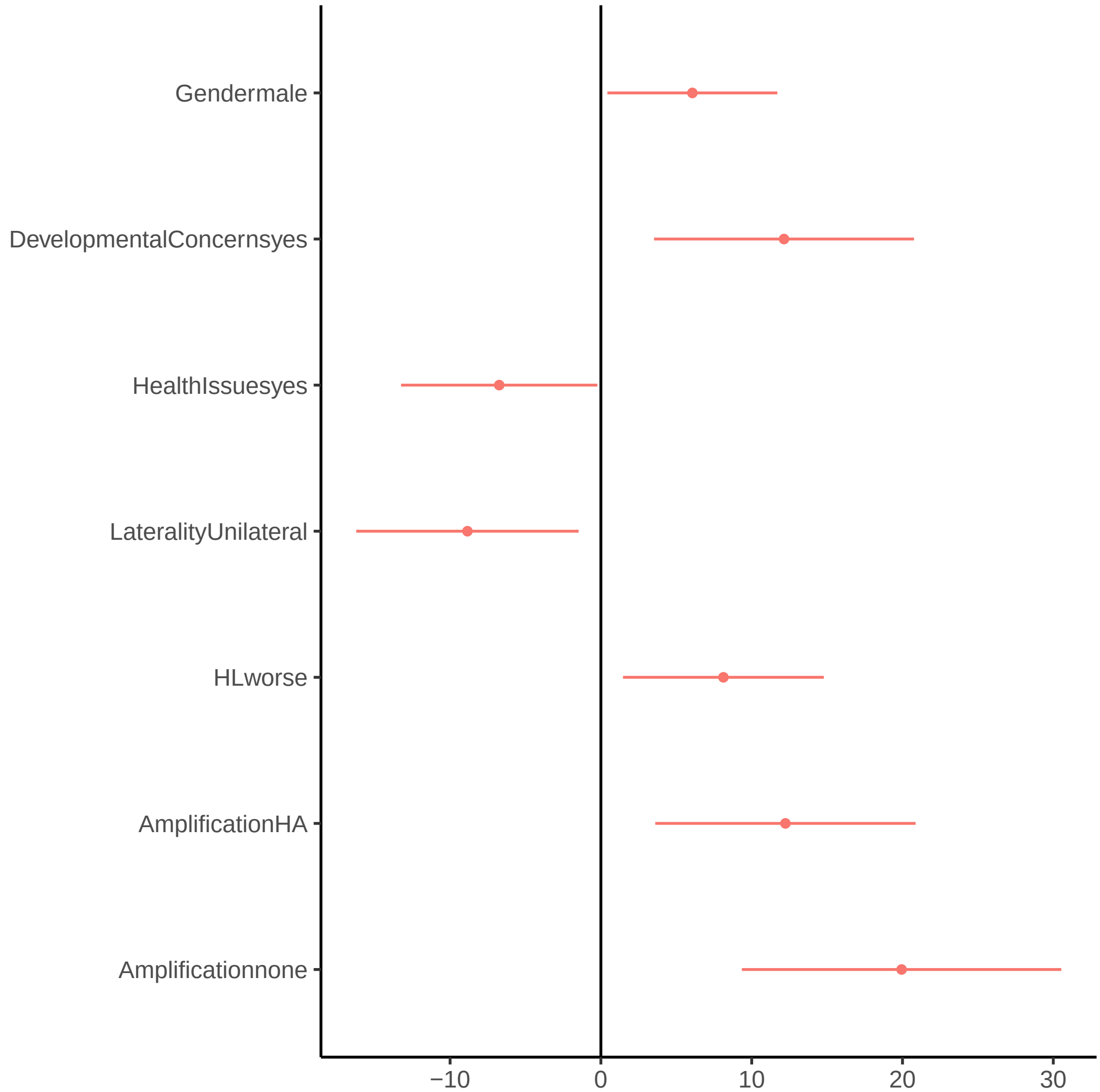


To look at the relationship between our predictor variables and vocabulary delay, we conducted multiple linear regression, using vocabulary delay as the outcome variable. We also present bonferroni-corrected zero-order tests of each predictor on vocabulary delay. We exclude children from non-English-speaking families from this section of the analysis because our growth curves are based on the English language CDI.

Our full regression model included all of our 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.

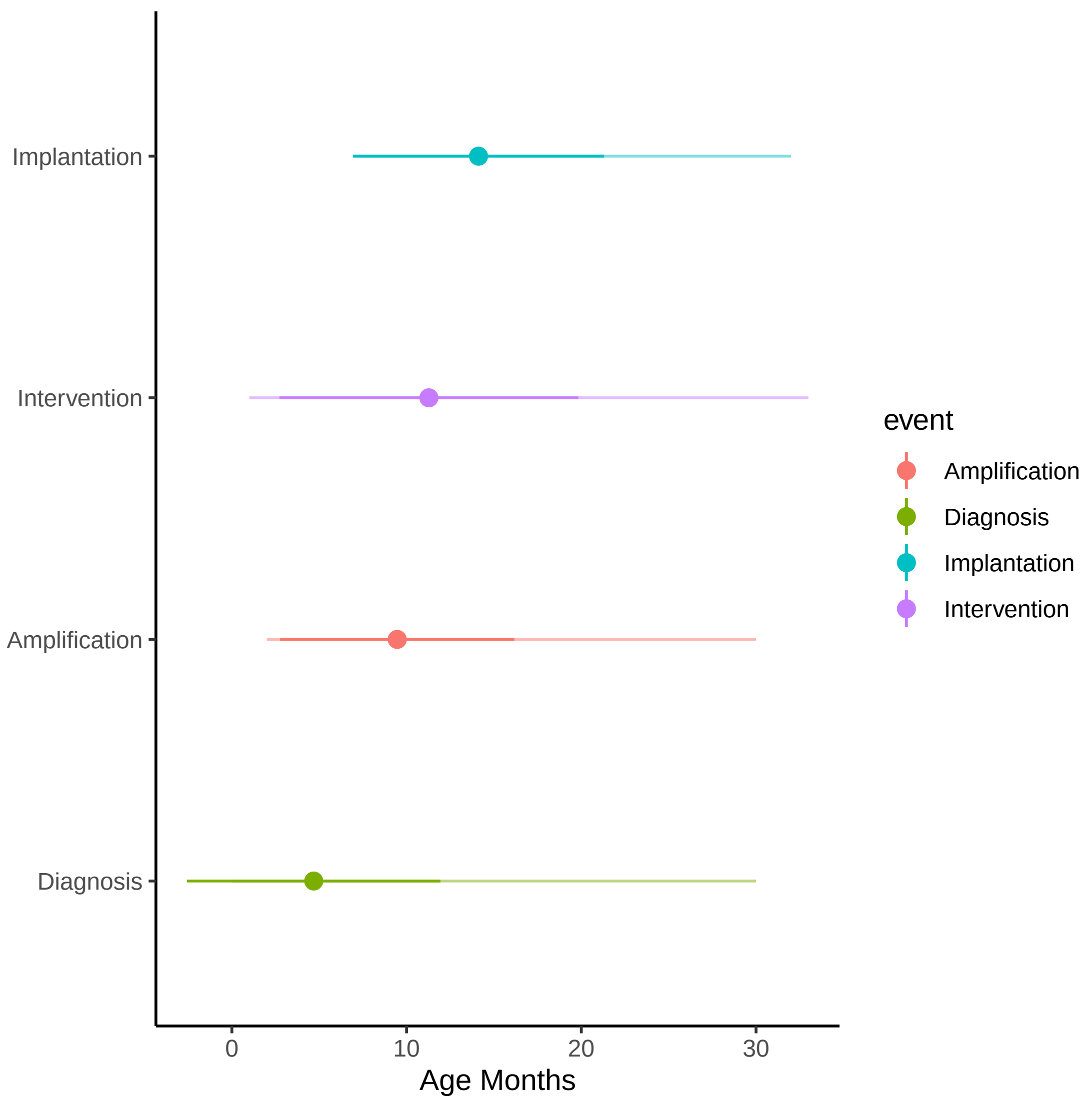
Our final model included: Vocabulary Delay ~ Gender + Developmental Delay + Health Issues + Laterality + Degree + Amplification. This model accounted for significant variance in children's vocabulary delay (adjusted-R2 = 0.29, p =). There were significant main effects of gender, developmental delay, laterality, degree, health issues, and amplification. In this model, being male (ß = 6.07, p = 0.035), having a developmental delay (ß = 12.14, p = 0.0065), bilateral hearing loss (ß = -8.85, p = 0.019), and more severe hearing loss predicted a larger delay (ß = 0.17, p = 0.018). Having a cochlear implant (ß = 12.24, p = 0.0061)or hearing aid (ß = 19.94, p = 0.00036) predicted a smaller delay, relative to no amplification. Presence of health issues indicated a smaller vocabulary delay (ß = -6.74, p = 0.043). 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).



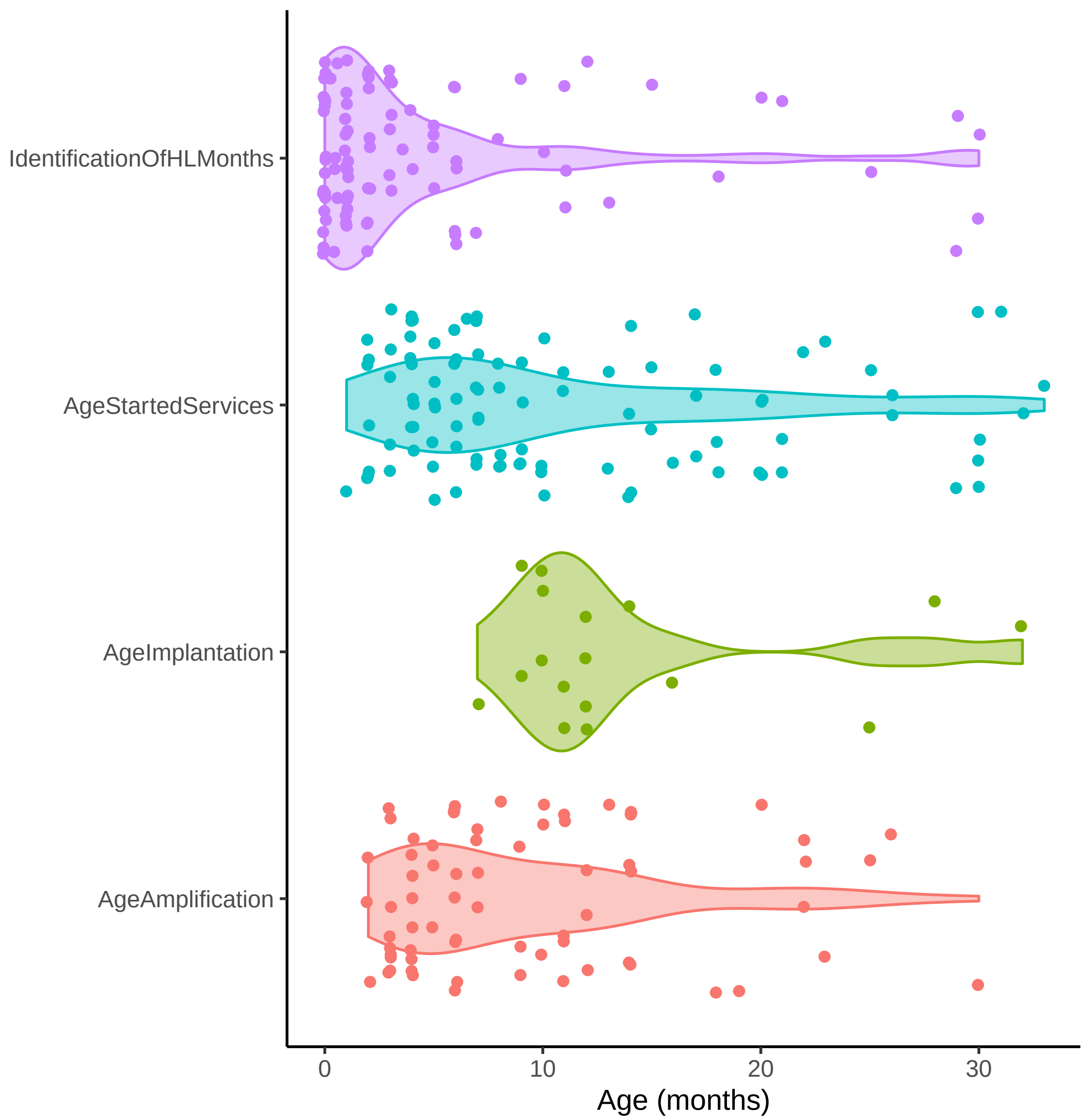
Part III: Meets136 success

Lastly, we looked at the ages at which children received diagnosis and intervention, and how this mapped onto the 1-3-6 guidelines. Perhaps of greatest importance to clinicians and policymakers is the implementation and effect of existing policies. 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.



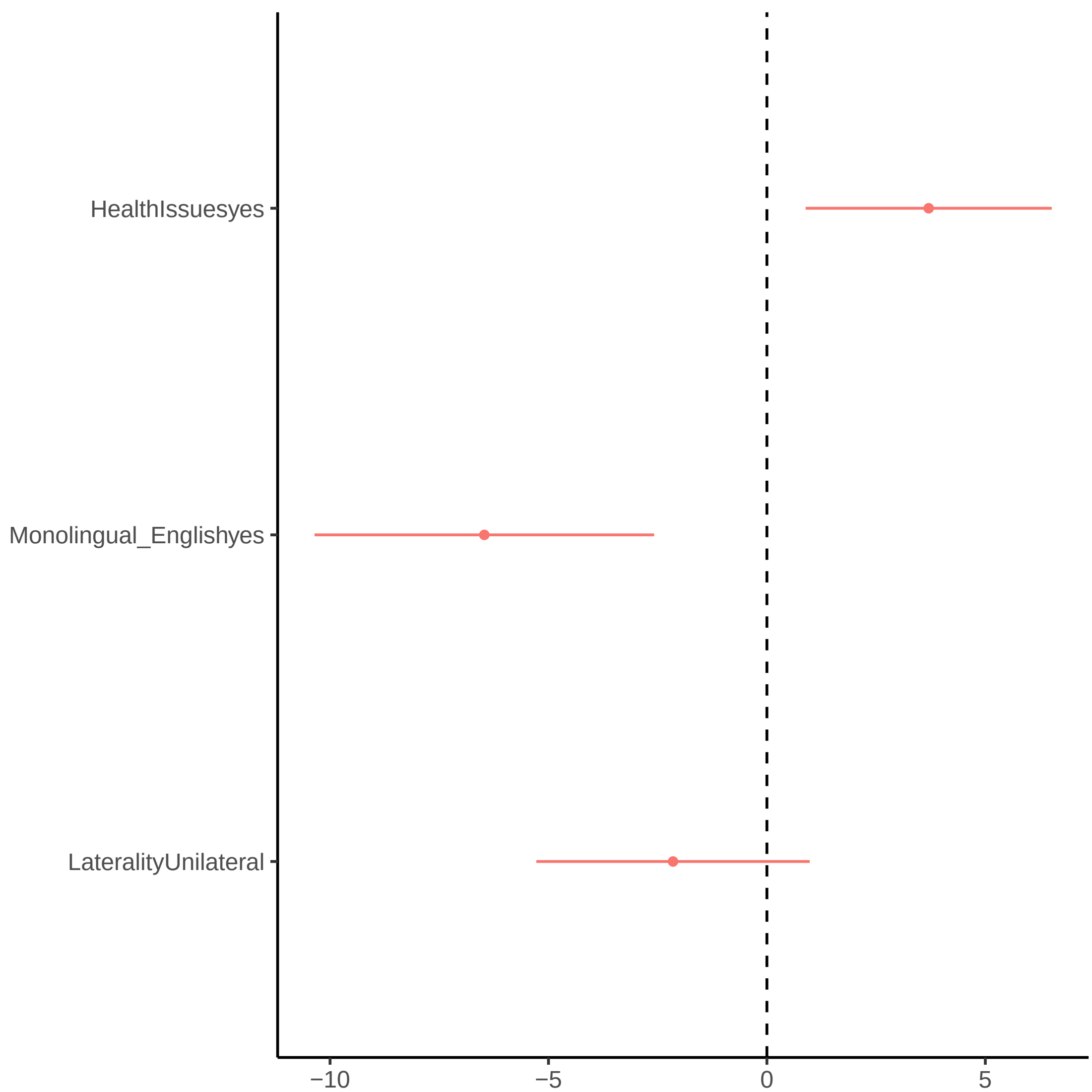
iagnosis, etc. Stronger lines represent standard deviation. Softer lines show range.



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 / intervention. We then paired down 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.

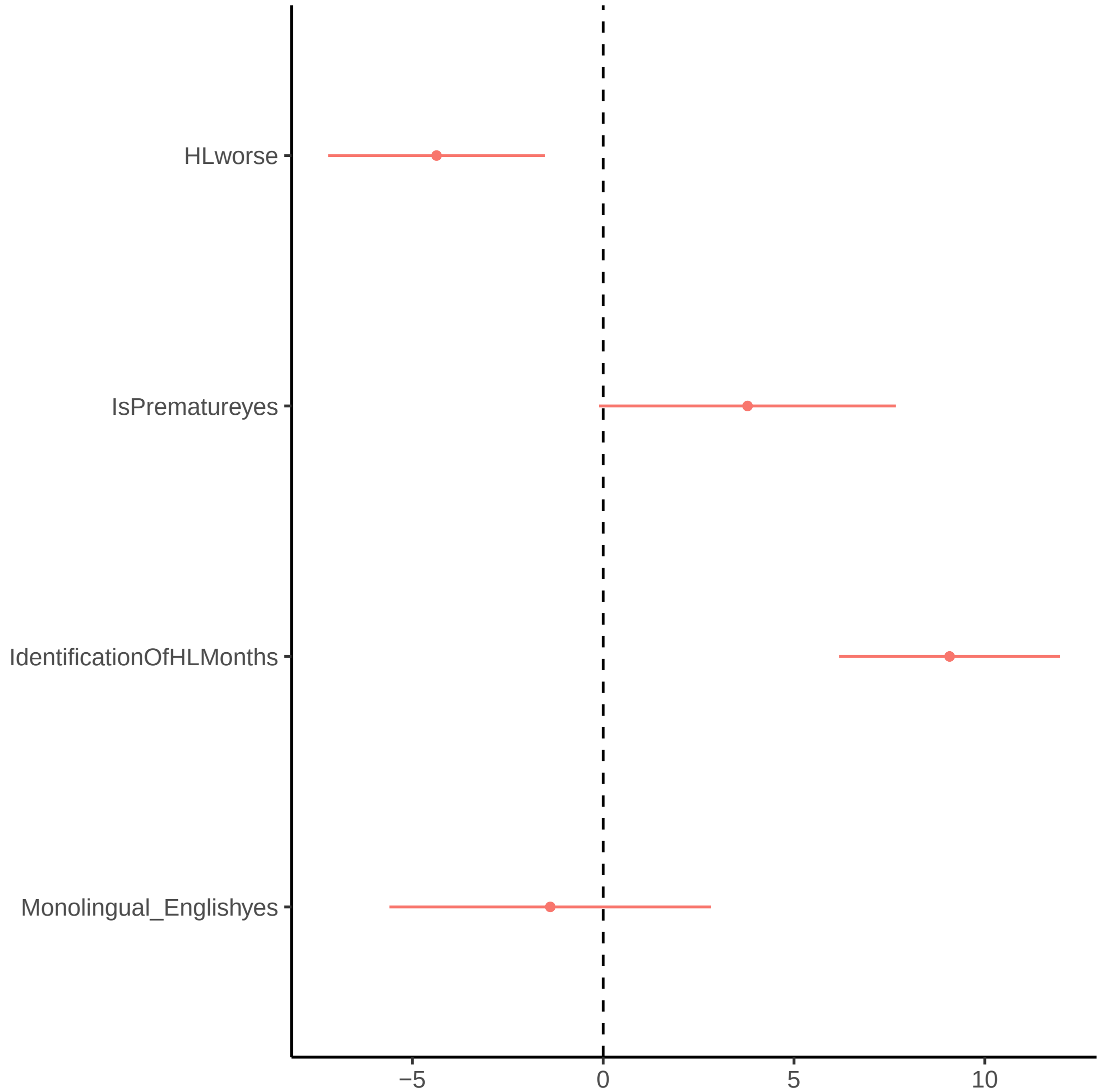
Model selection removed gender, degree, developmental delay, prematurity, laterality, and etiology as predictors. Under the best fit model (R2=0.16 , p=0.00), children with no additional health issues (ß = 3.7, p = 0.011), children from English-speaking households (ß = -6.47, p = 0.0014), and children with unilateral hearing loss (ß = -2.15, p = 0.18) were earlier diagnosed. Presence of health issues indicated a 3.70 months later diagnosis, and coming from a non-English-speaking household resulted in a -6.47 months later diagnosis. This model accounted for roughly 0.16% of the variance in age at diagnosis.

$$Age\ at\ Diagnosis \sim Health\ Issues + Language\ Background$$



We repeated this model selection process for age at intervention. Our starting model included the variables potentially relevant to intervention: Age intervention ~ gender + degree (worse ear) + developmental delay + health issues + prematurity + laterality + language background + etiology + age diagnosis Model selection by AIC removed Gender, Developmental Delay, Health Issues, Prematurity, Language Background, and Etiology. In the best fit model (R2=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 0.43% of the variance in age at intervention. Infants born premature were diagnosed 3.78 months later than full-term infants. A 1 decibel increase in hearing loss results in 2.81 days earlier intervention. Each one month delay in diagnosis results in a 1.38 month delay in intervention.

$$AgeatIntervention \sim DegreeofHearingLoss(worseear) + Prematurity + AgeatDiagnosis$$



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Discussion *(This isn't ready yet)* 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 (CITE) and throughout the lifespan (CITE). 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 (CITE) and children born premature(CITE), with differential effects based on the nature of the developmental delay (CITE) 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), 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. 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 random, 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 (Hassepass et al., 2013; priwin et al., 2007; briggs et al., 2011; dwyer et al., 2014) and spoken language vocabulary and grammar for mild hearing loss (walker et al., 2015).

Conclusion

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, we focus on spoken language development.

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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
Ching et al., 2013	3 year old children receiving services in Australia	Female +	Did not study	Did not study	More severe -	No effect	No effect	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
Age at Amplification	Continuous	2-30 months
Age at Diagnosis	Continuous	0-30 months
Age at Implantation	Continuous	7-32 months
Age at Intervention	Continuous	1-33 months
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
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
Type of Hearing Loss	Categorical	Sensorineural / Conductive / Mixed
CDI - Words Produced	Continuous	0-635 words