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

2   Word Learning, Diagnosis, and Intervention

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

4   [1] Duke University

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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

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

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

164 mandate universal newborn hearing screening (National Conference of State Legislatures,

165 2011). All states have some form of early intervention programs that children with hearing

166 loss can access (NAD, n.d.), but these also vary state-by-state. For instance, half of the

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

168 intervention (Holstrum, Gaffney, Gravel, Oyler, & Ross, 2008).

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

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

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

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

173 that there may be breaks in the chain from screening to diagnosis and from diagnosis to

174 intervention, and the effect may be further delays in language development for children not

175 meeting these guidelines.

Quantifying vocabulary growth in DHH children

177 The MacArthur Bates Communicative Development Inventory (CDI, Fenson et al.,

178 1994) is a parent-report instrument that gathers information about children's vocabulary

179 development. The Words and Gestures version of the form (CDI-WG) is normed for

180 8–18-month-olds. On CDI-WG, parents indicate whether their child understands or produces

181 each of the 398 vocabulary items, and answer questions about young children's early

182 communicative milestones. The Words and Sentences version of the form (CDI-WS) is

183 normed for 16-30-month-olds. On CDI-WS, parents indicate whether their child produces

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

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

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

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

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

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

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

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

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

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

194 Castellanos, Pisoni, Kronenberger, and Beer (2016) finds that in children with CIs, number

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

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

197 vocabulary development in DHH children (Ching et al. (2013); Yoshinaga-Itano et al. (2017);

198 Yoshinaga-Itano et al. (2018); de Diego-Lázaro et al. (2018); Vohr et al. (2008); Vohr et al.

199 (2011); summarized in table XXX).

Goals and Predictions

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

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

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

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

205 traditionally excluded from studies of language development: children with additional

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

207 non-English-speaking households (e.g., Yoshinaga-Itano et al., 2018).

208   For the first and third goal above, we did not have specific hypotheses and sought to

209 provide descriptive information about a diverse sample of DHH children receiving state

210 services. For the second, we hypothesized that male gender, more severe degree of hearing

211 loss, bilateral hearing loss, no amplification use, prematurity, and presence of additional

212 disabilities would predict larger spoken vocabulary delay. We did not have strong predictions

²¹³ regarding the effects of communication method or presence of other health issues (e.g.,

²¹⁴ congenital heart malformation) on vocabulary.

₂₁₅   **Methods**

²¹⁶   Clinical evaluations were obtained through an ongoing collaboration with the North

²¹⁷ Carolina Early Language Sensory Support Program (ELSSP), an early intervention program

²¹⁸ serving children with sensory impairments from birth to 36 months. ELSSP passed along

²¹⁹ deidentified evaluations to our team after obtaining consent to do so from each family. No

²²⁰ eligibility criteria beyond hearing loss and receiving an ELSSP evaluation were imposed,

²²¹ given our goal of characterizing the full range of DHH children with hearing loss in North

²²² Carolina.

²²³   The clinical evaluations included demographic and audiological information, CDI

²²⁴ vocabulary scores, and the results of any clinical assessments administered (e.g., PPVT), all

²²⁵ detailed further below. For some children (n=47), multiple evaluations were available from

²²⁶ different timepoints. In these cases, only the first evaluation was considered for this study,

²²⁷ due to concerns regarding within-subjects variance for statistical analysis.

²²⁸   While this collaboration is ongoing, we opted to pause for this analysis upon receiving

²²⁹ data from 100 children. Thus, the reported sample below consists of 100 children (56 male /

²³⁰ 44 female) ages 4.20–36.17(M=21.21, SD=9.08). Race and SES information were not

²³¹ available. Families were administered either the WG or WS version of the CDI based on

²³² clinician judgement. Children who were too old for WG, but who were not producing many

²³³ words at the time of assessment, were often given WG (n=37). Families for whom Spanish

²³⁴ was the primary language (n=14) completed the Spanish language version of the CDI

²³⁵ (Jackson-Maldonado et al., 2003).

²³⁶   With regard to comorbid diagnoses, children in this sample were coded as yes/no for

²³⁷ cognitive development concerns (e.g., Down syndrome, global developmental delays; Cornelia

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

239 health issues (e.g., heart defects, kidney malformations, VACTERL association), and yes/no

240 for vision loss (not corrected to normal by surgery or glasses).

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

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

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

244 better and worse ear). Using the ASHA hearing loss guidelines, each of these was coded with

245 a dB HL value corresponding with the median dB HL for the level of hearing loss (e.g.,

246 moderate hearing loss was coded as 48 dB HL), and sloping hearing loss was coded as the

247 average of the levels (e.g. mild to moderate was coded as 40.5 dB HL). Participants were also

248 coded for unilateral or bilateral hearing loss; presence or absence of Auditory Neuropathy

249 Spectrum Disorder; and etiology of hearing loss (sensorineural, conditive, or mixed).

250 Amplification was recorded as the device the child used at the time of assessment: either

251 hearing aid, cochlear implant, or none.

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

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

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

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

256 language spoken at home. Families in this sample either spoke English or Spanish. For one

257 child, who was adopted from India at 28 months, we recorded the primary language as Hindi,

258 athough the child's adoptive parents are English-speaking, because the child had lived most

259 of her life in a non-English-speaking environment.

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

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

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

263 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

289 combinations of variables, p < .05 for 22, and 8 survived bonferroni correction. We are only

290 discussing the results of tests that survived bonferroni correction, but the full set of results

291 can be found in table XXX.

292

Results of chi−square tests between variables.
For tests that survived bonferroni correction,
effect size (Cramer's V) is given.
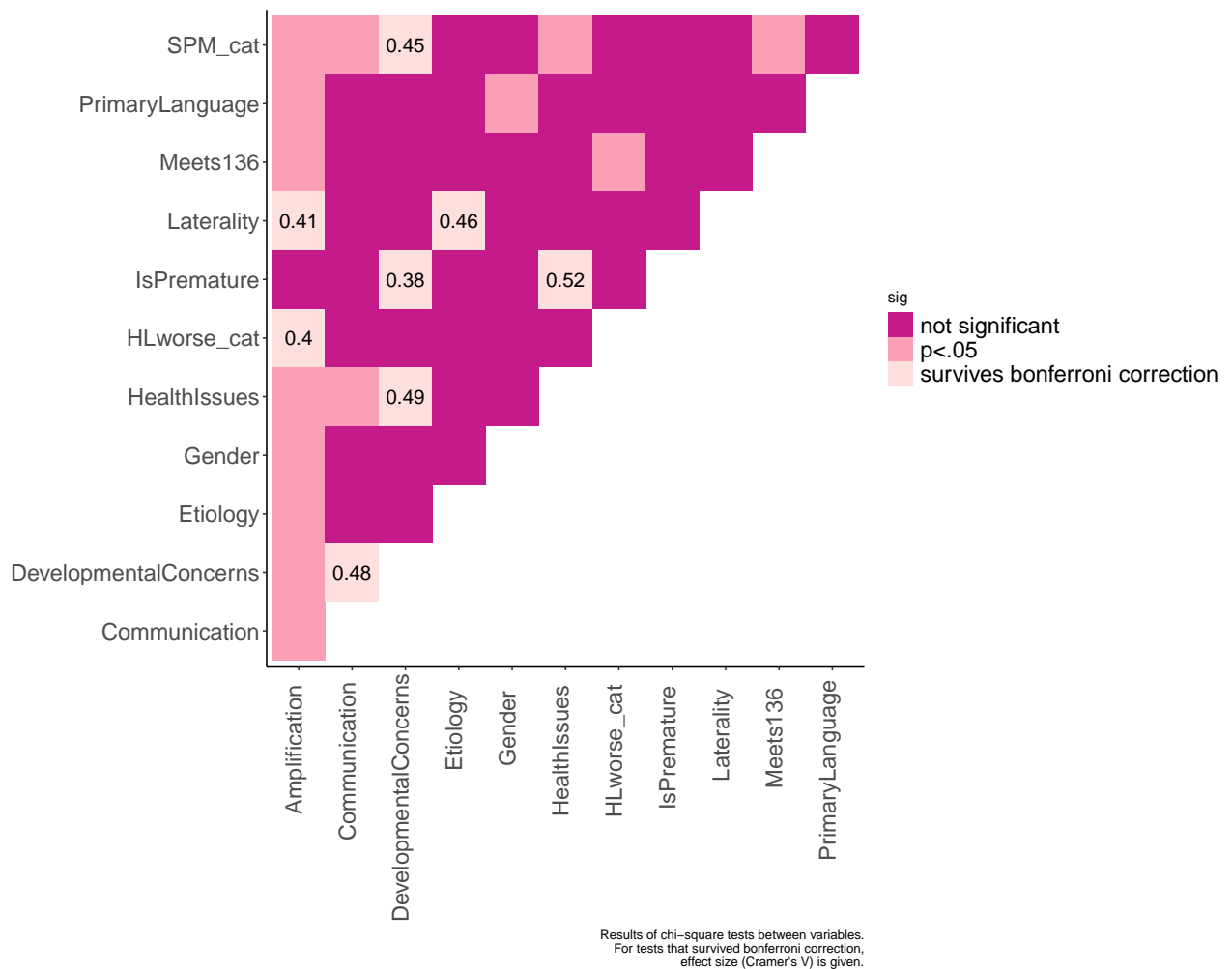
293   From this set of analyses, we found 8 relationships between variables that survived

294 bonferroni correction. Health issues, developmental delays, and prematurity were highly

295 interrelated in our sample, such that children born premature were more likely to also

296 experience health issues (X^2 (1, N = 98) = 23.9, p = 1e-06) and developmental delays (X^2

297 (1, N = 98) = 11.63, p = 0.00065), and children with developmental delays were more likely

298 to also experience health issues (X^2 (1, N = 98) = 20.87, p = 4.9e-06). Children with

299 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
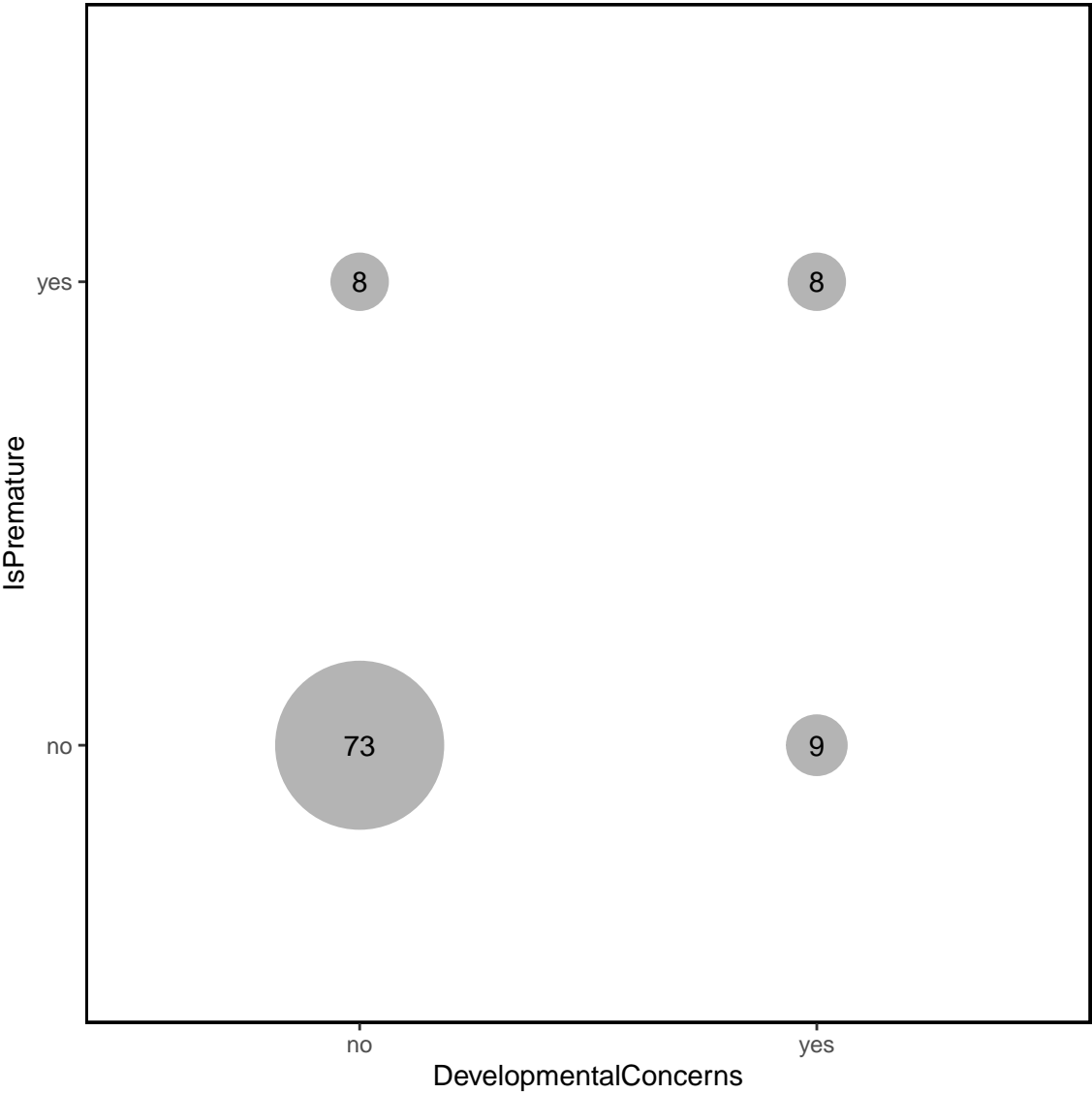
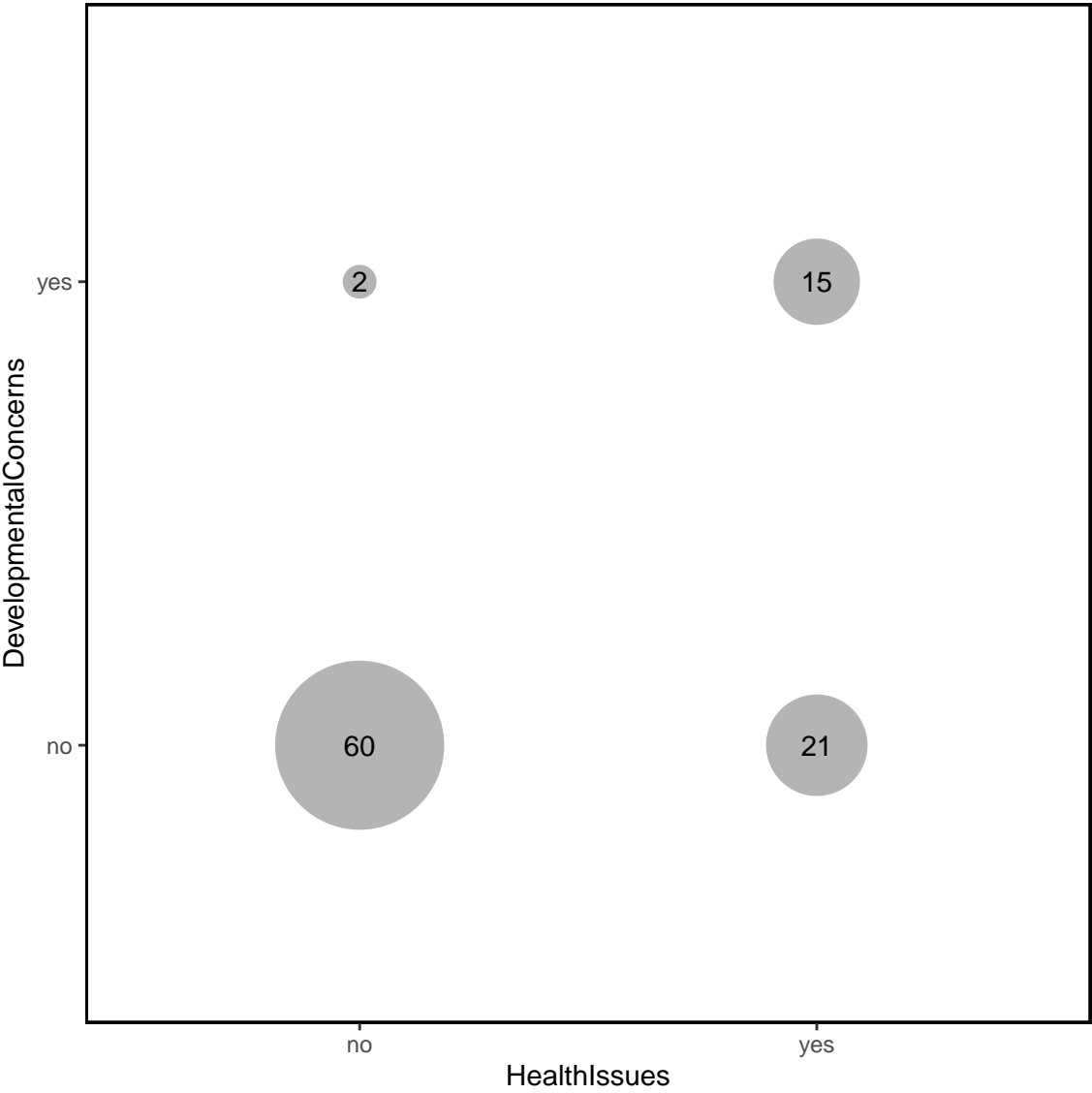children with less severe hearing loss (i.e., mild or moderate).
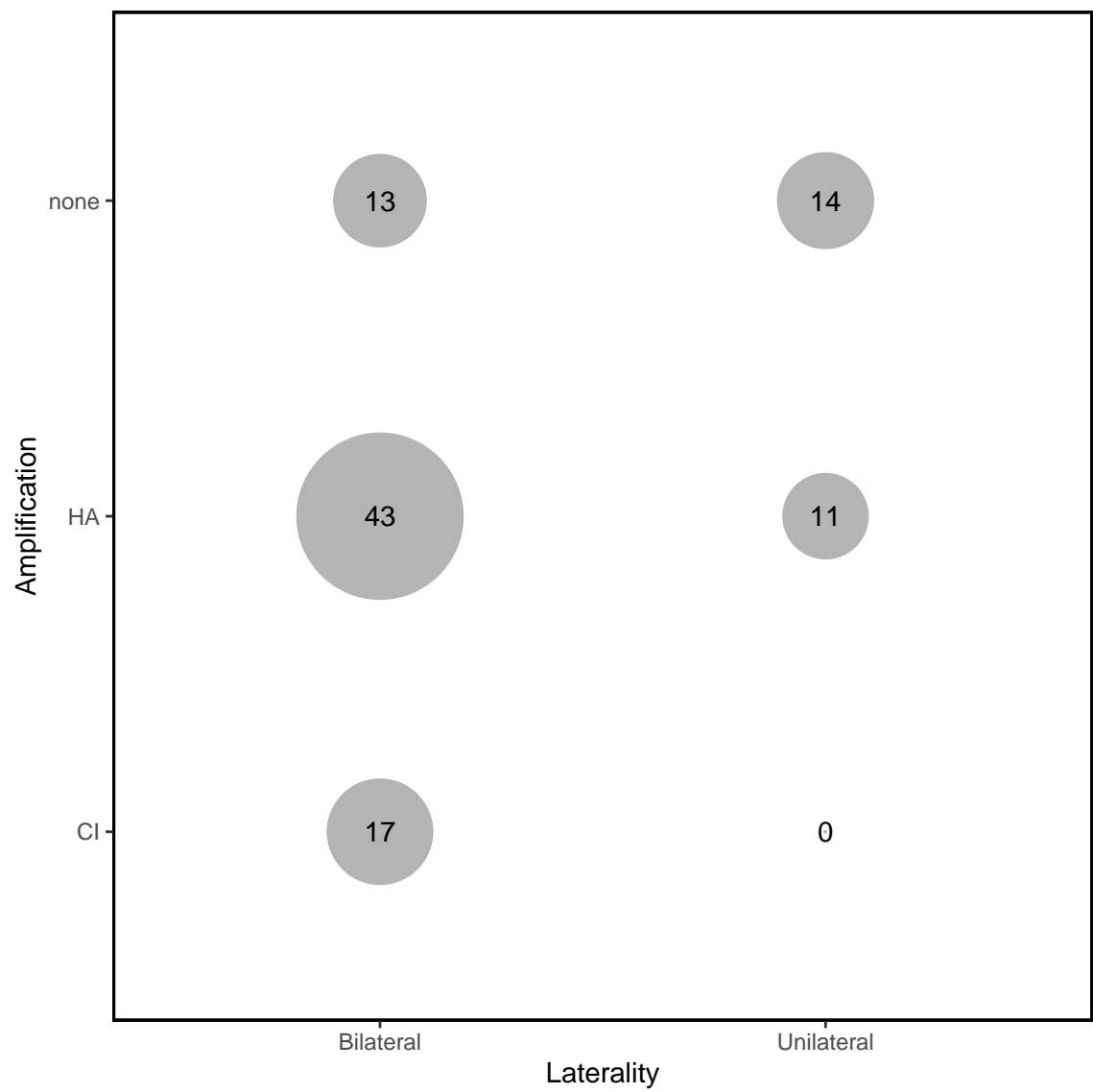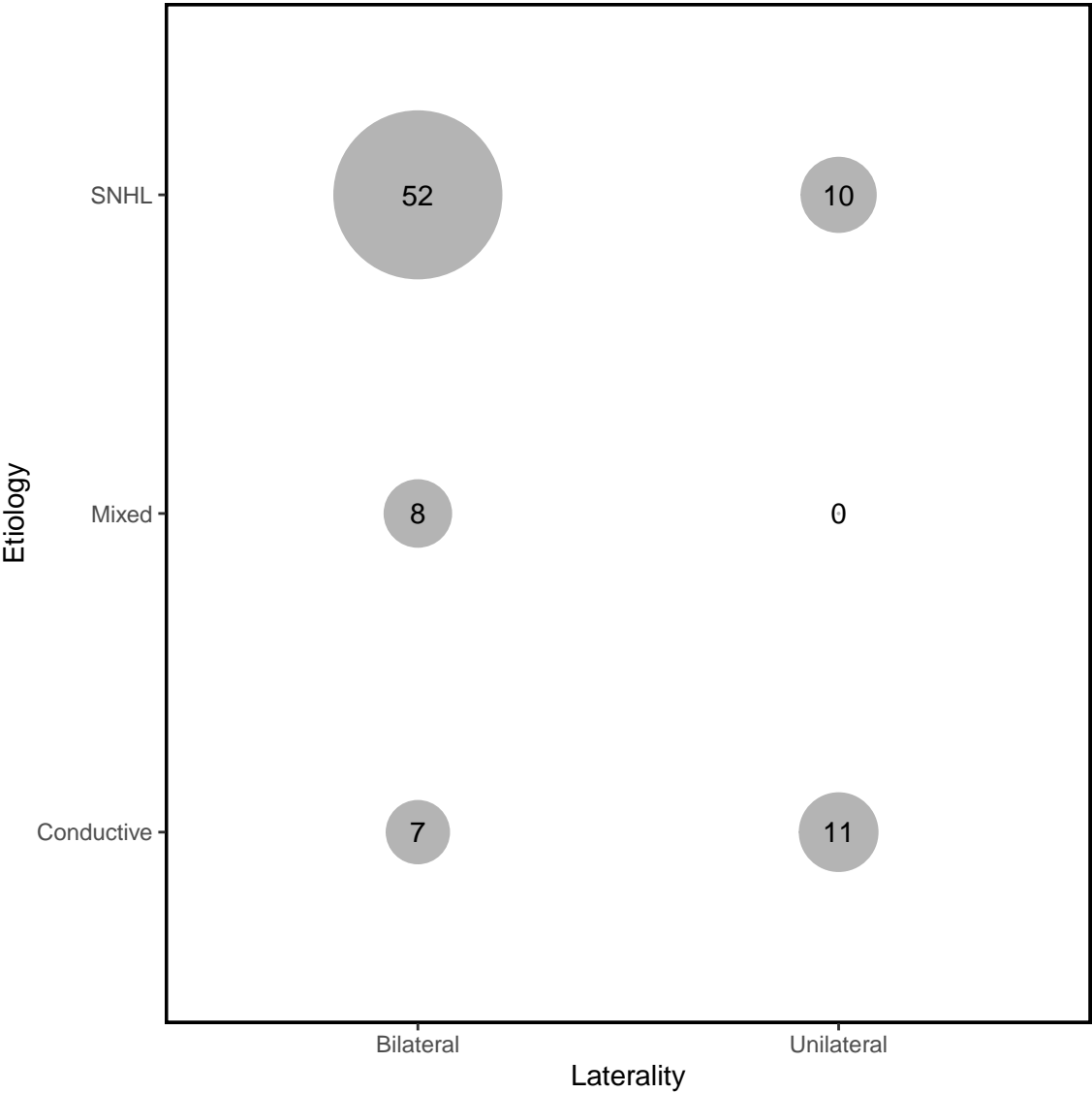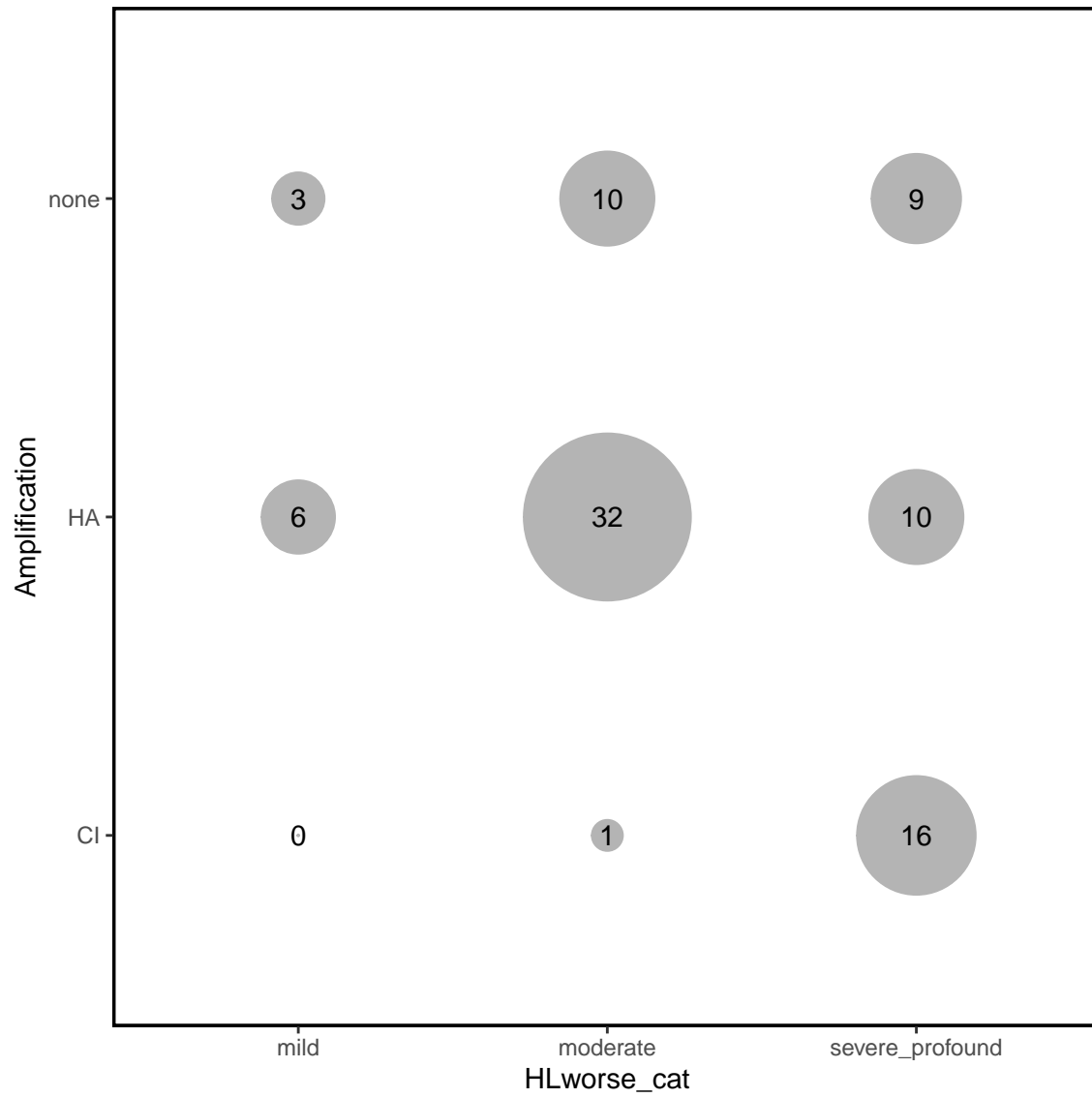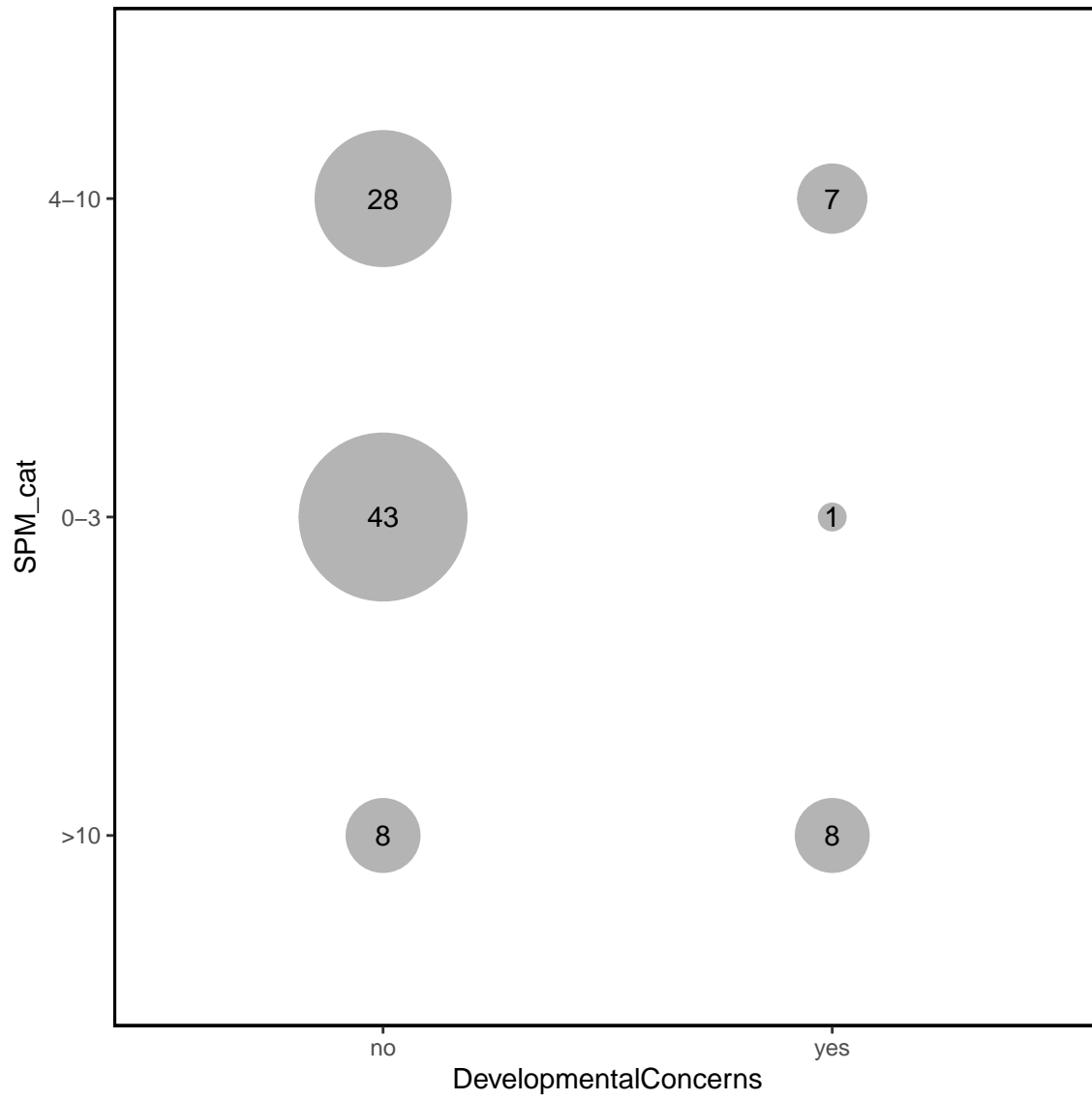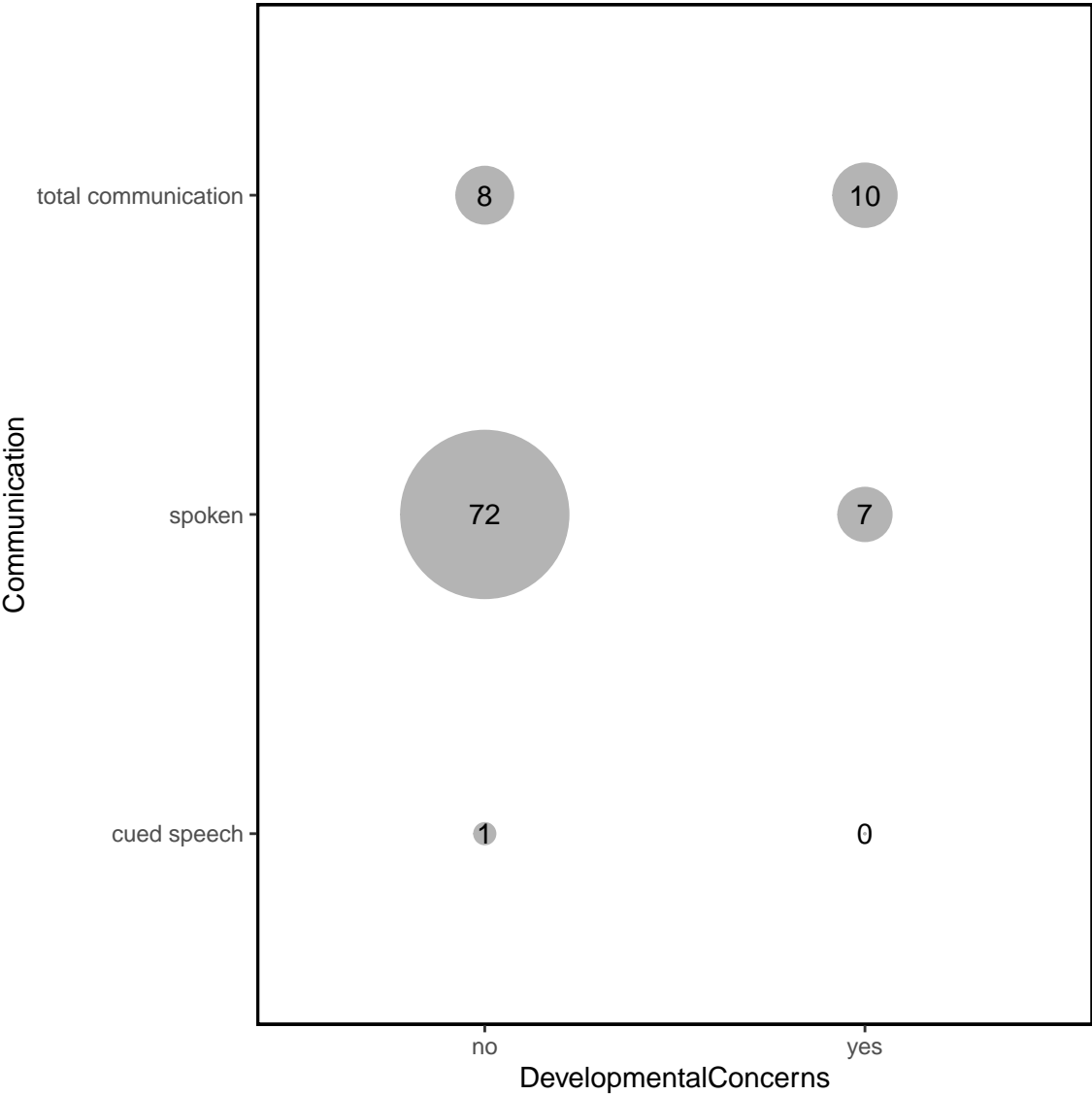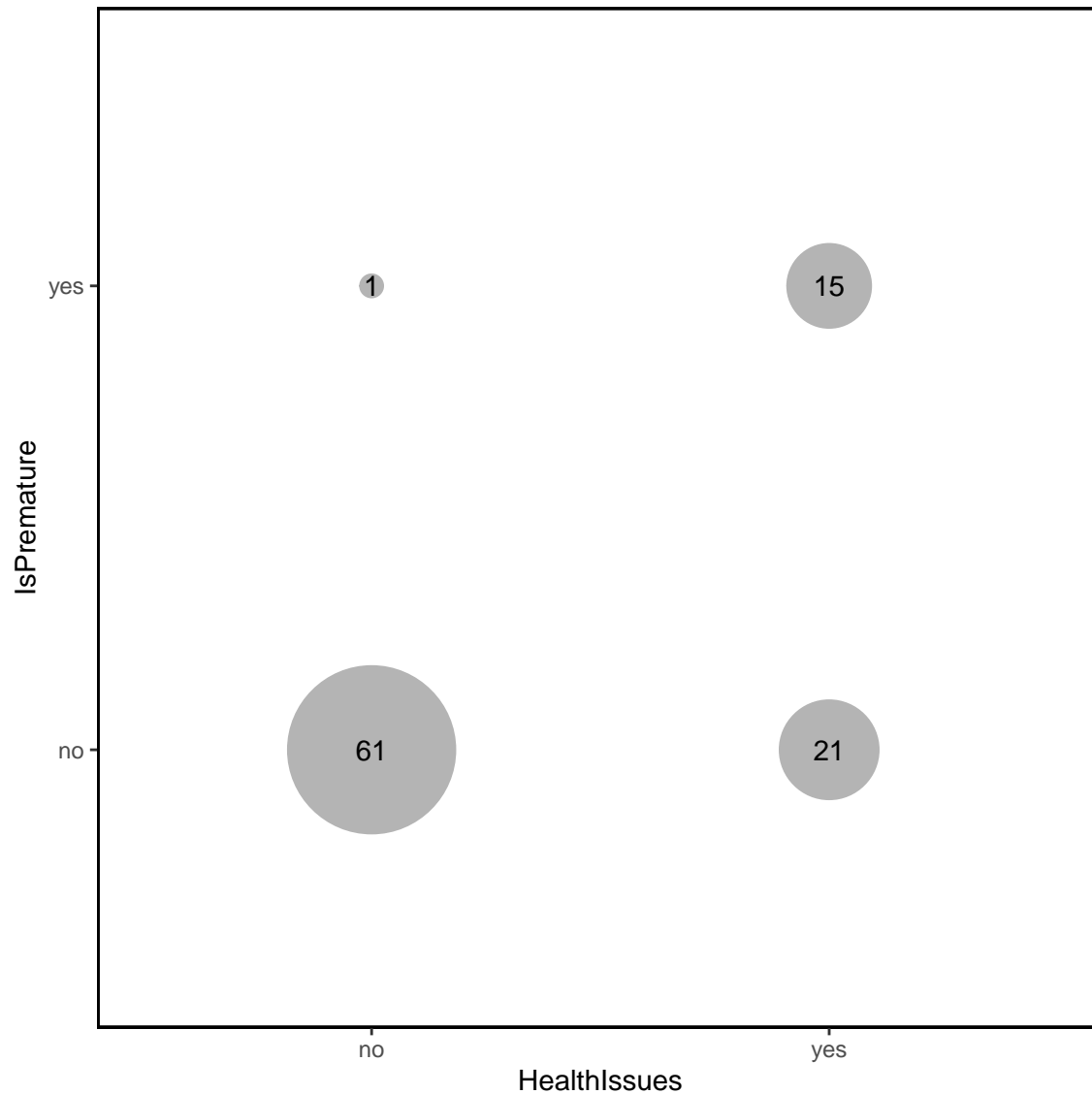
Part II: Influence on vocabulary

We next turn to the relationship between each of these variables and children's productive vocabulary, measured on the CDI. Figure XXX 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

Normative WS vs. ELSSP

Normative WG vs.   Normative WS vs. I
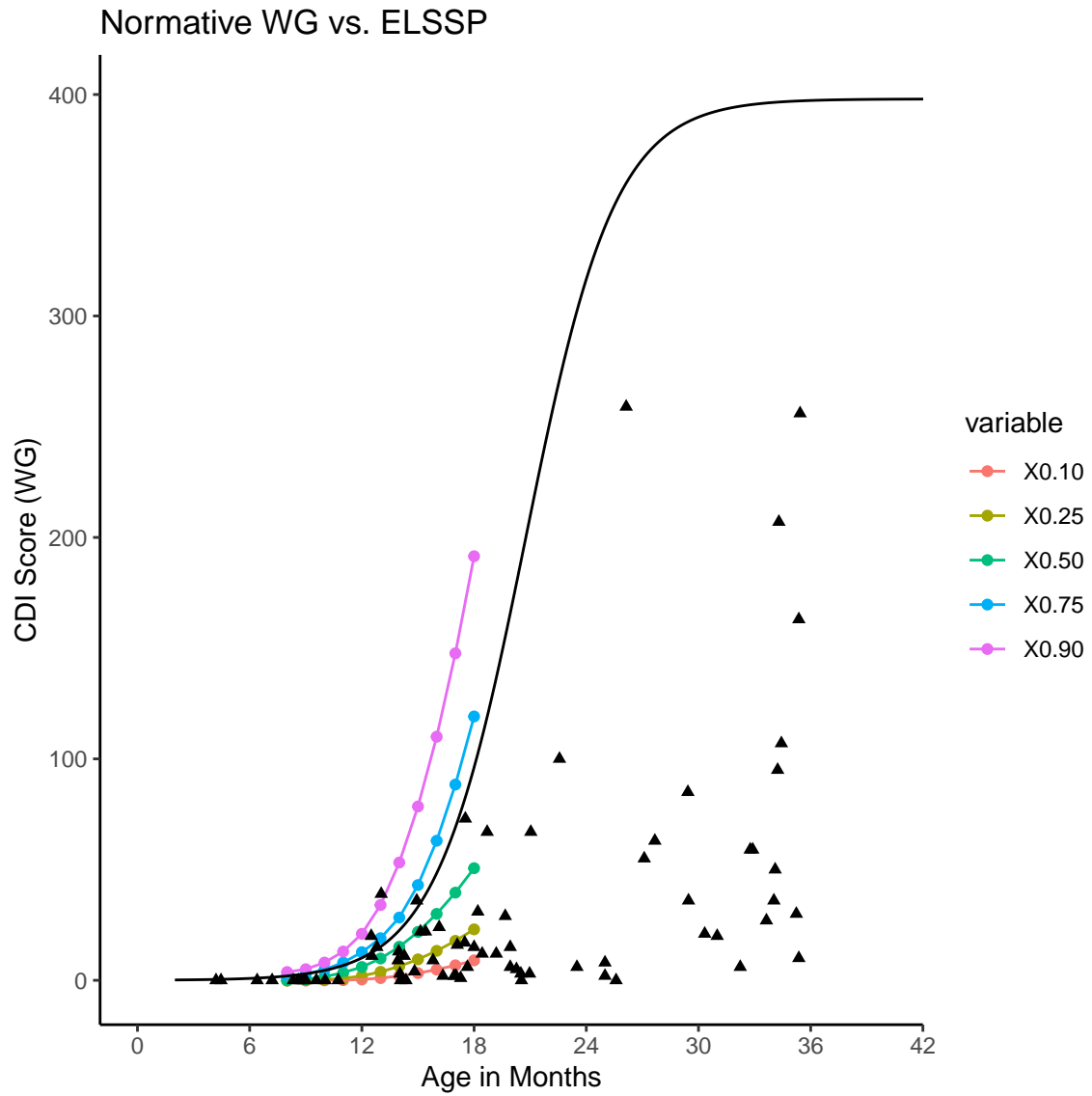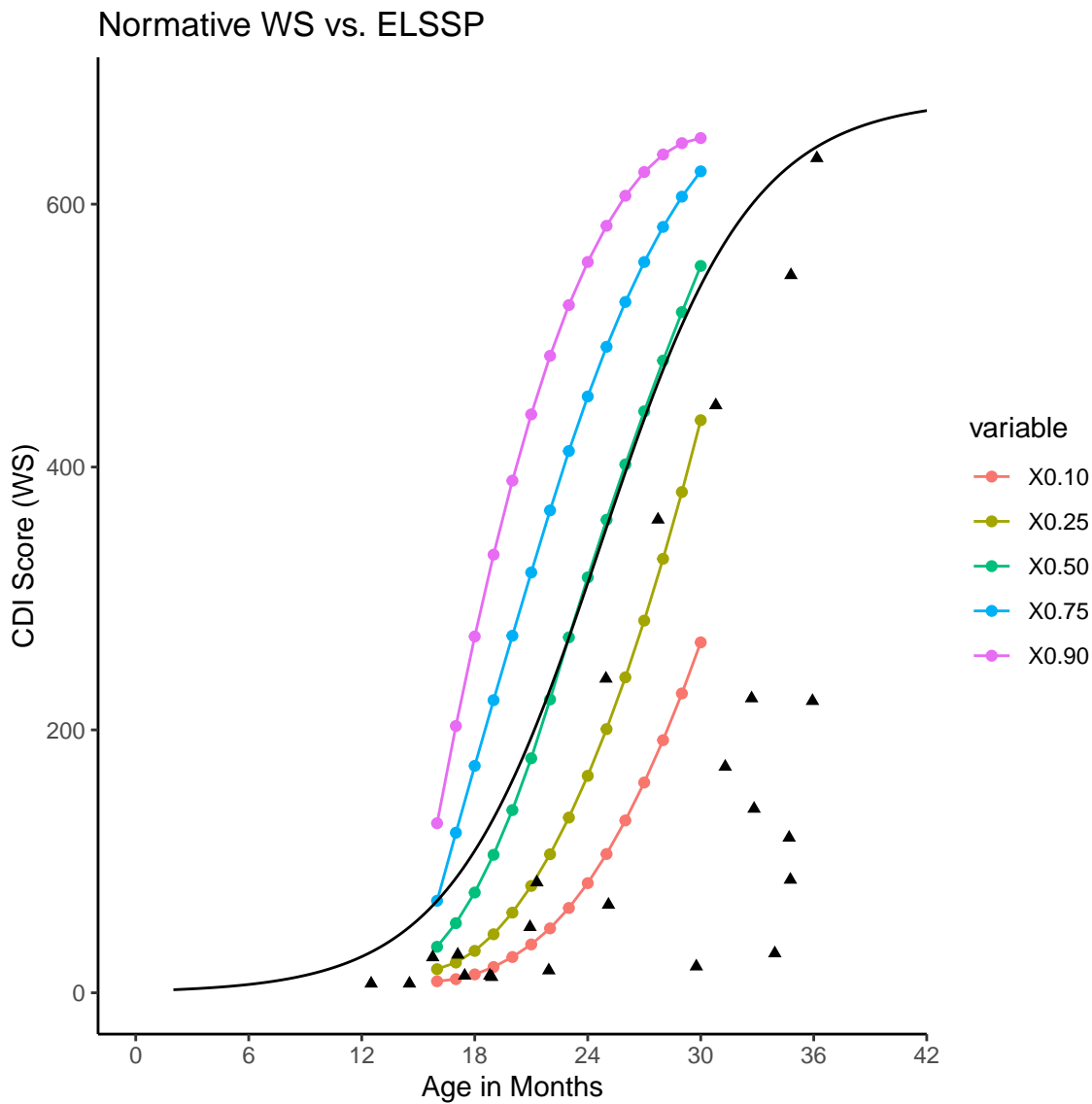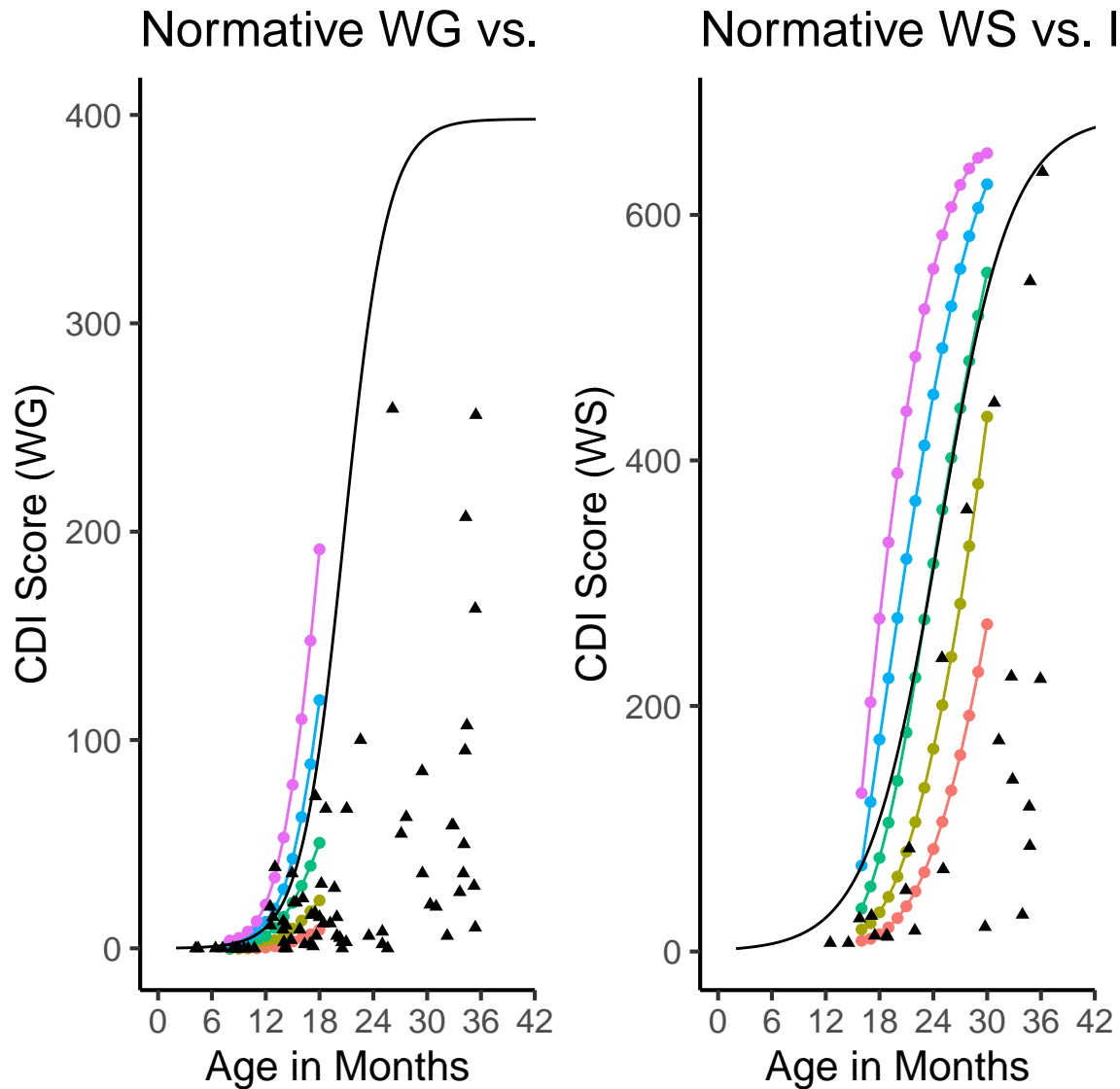
To look at the relationship between our predictor variables and vocabulary delay, we conducted multiple linear regression, using vocabulary delay as the outcome variable. We also present bonferroni-corrected zero-order tests of each predictor on vocabulary delay. We exclude children from non-English-speaking 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 +

358 ServicesReceivedPerMonth. We performed stepwise model comparison using stepAIC

359 (MASS) to pare down the model. This process selected only the predictors which

360 incrementally improved model fit, measured by Akaike's Information Criterion (AIC), which

361 considers goodness of fit and model complexity (penalizing models with many predictors).

362 Based on this iterative process, we removed Prematurity, Communication, Meets 1-3-6, and

363 ServicesReceivedPerMonth from the model.

364   Our final model included: Vocabulary Delay ~ Gender + Developmental Delay +

365 Health Issues + Laterality + Degree + Amplification (R2=0.35 , p=0.00). In this model,

366 being male, having a developmental delay, bilateral hearing loss, and more severe hearing

367 loss predicted a larger delay. Presence of developmental delay predicted larger vocabulary

368 delay. Having a cochlear implant or hearing aid predicted a smaller delay, relative to no

369 amplification. Presence of health issues indicated a smaller vocabulary delay. This model

370 accounted for roughly 0.35% of the variance in children's vocabulary delay. Although we

371 showed in Part I that relationships exist among several of these variables (e.g., degree and

372 amplification), a vif test on our model revealed that each predictor was responsible for a

373 unique share of the variance.
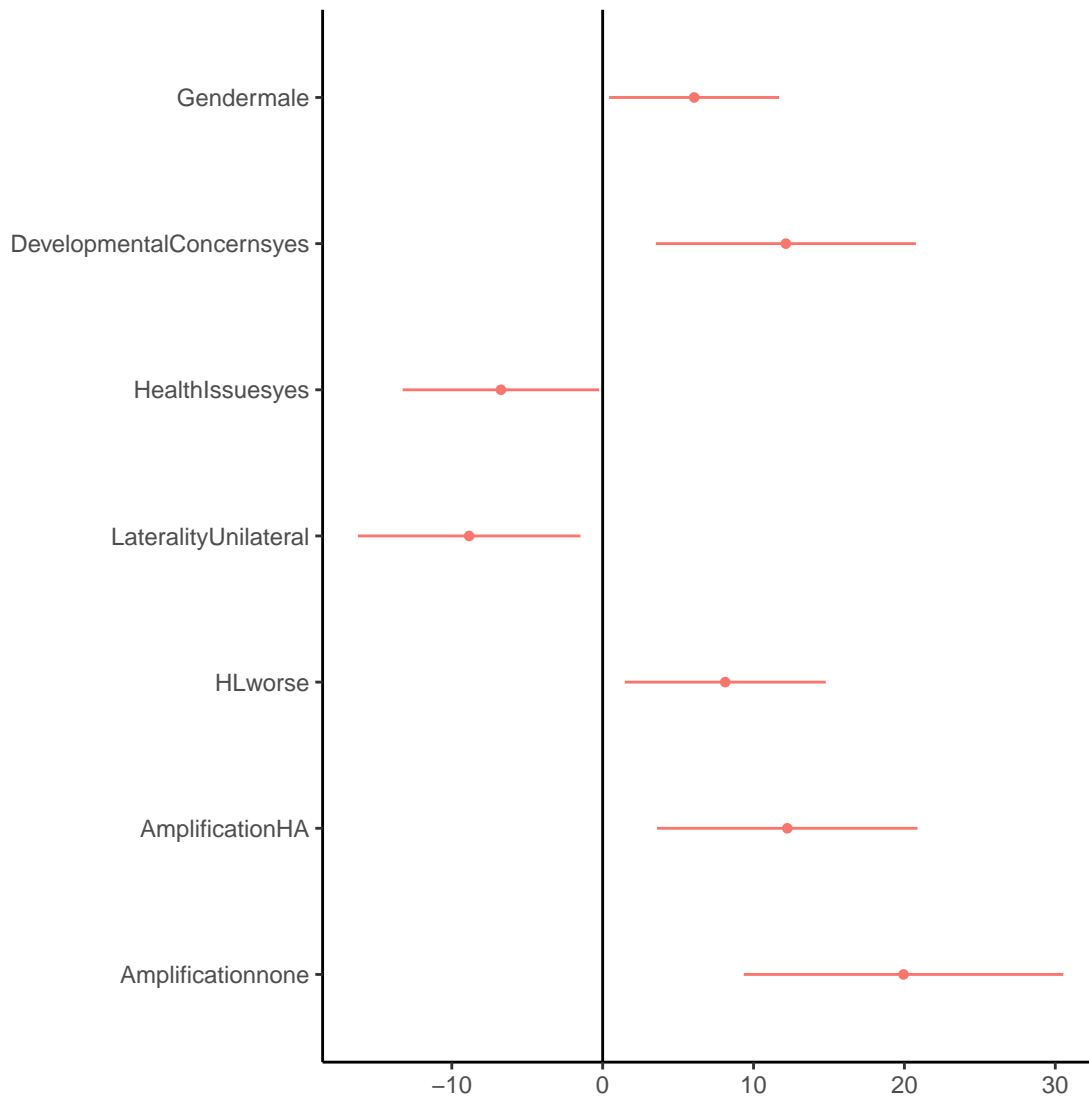
Part III: Meets136 success

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.

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

385   (see Table 6). Among the children for which screening information was available (n = 68),

386   100% were screened at birth or during NICU stay. 69% of children received diagnosis by 3

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

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

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

390   implantation for each child in our sample.
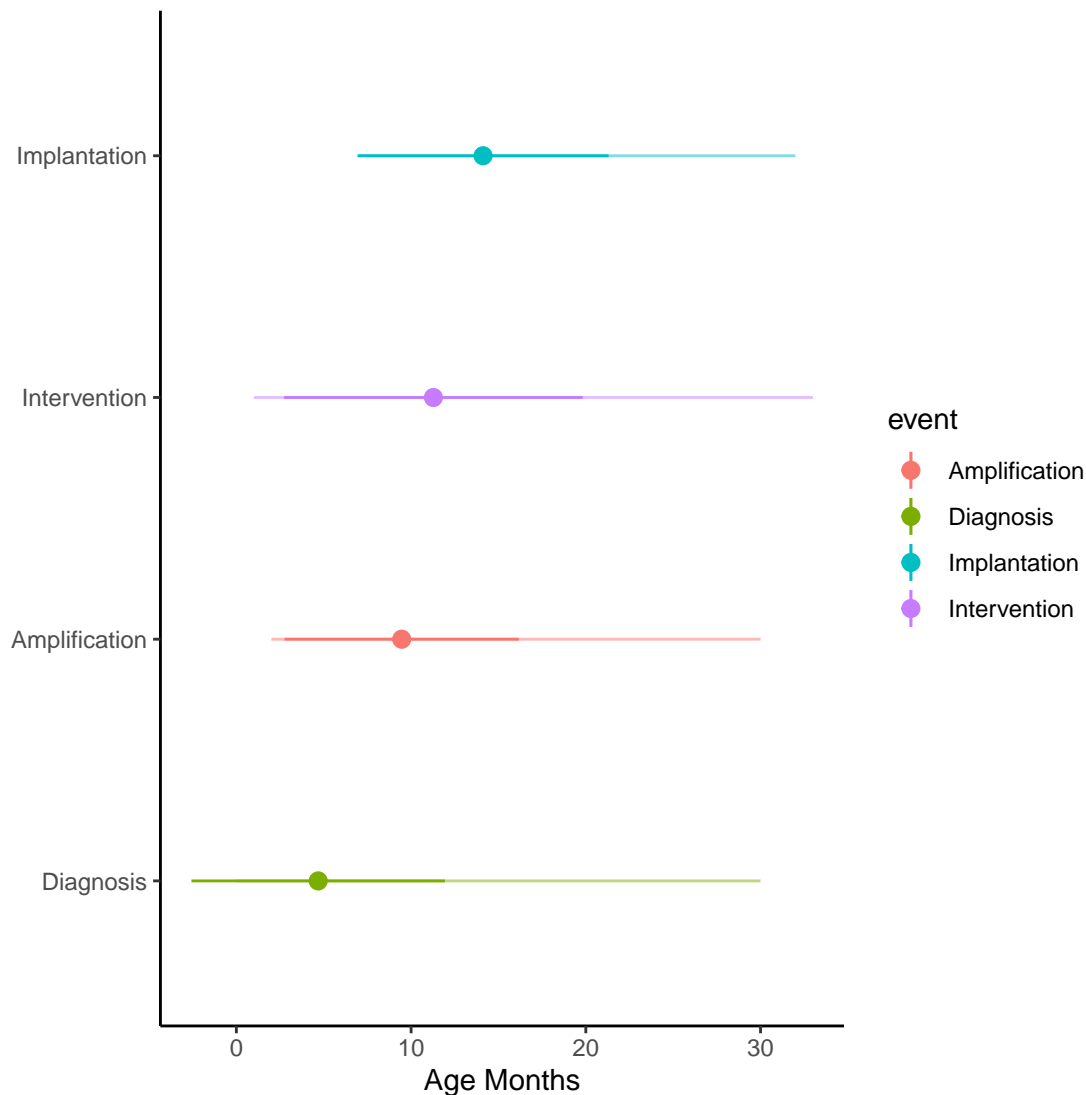
391   ...diagnosis, etc. Stronger lines represent standard deviation. Softer lines show range.

392
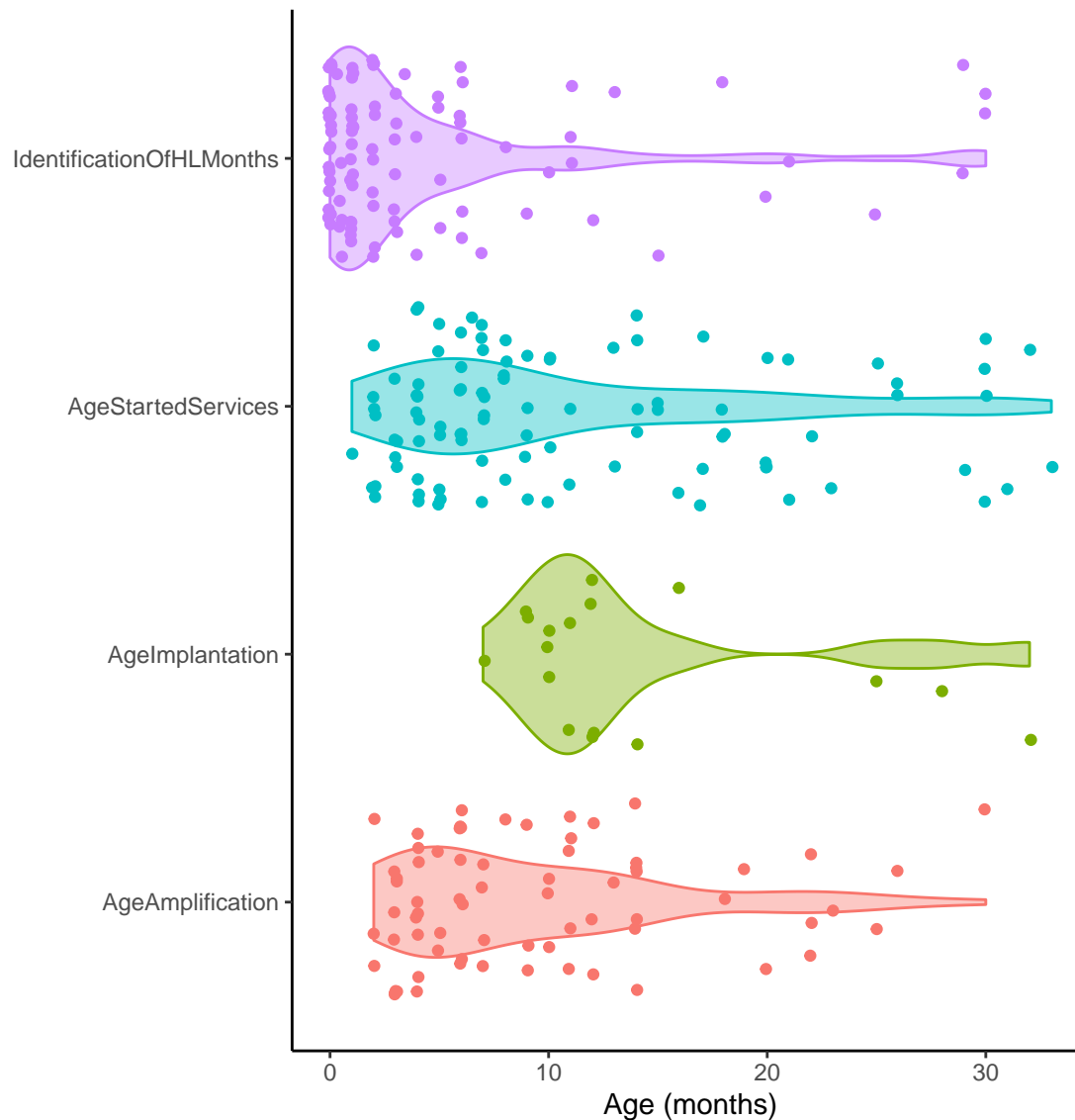
We created two linear regression models, one for age at diagnosis and one for age at
intervention. For each model, we started with the set of predictors that would have been
present prior to or during diagnosis / intervention. We then paired down using stepwise
regression by AIC (MASS package), using 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,

401 language background, and etiology.

402   Model selection removed gender, degree, developmental delay, prematurity, laterality,
403 and etiology as predictors. Under the best fit model (R2=0.16 , p=0.00), children with no
404 additional health issues (ß = 3.7, p = 0.011), children from English-speaking households (ß
405 = -6.47, p = 0.0014), and children with unilateral hearing loss (ß = -2.15, p = 0.18) were
406 earlier diagnosed. Presence of health issues indicated a 3.70 months later diagnosis, and
407 coming from a non-English-speaking household resulted in a -6.47 months later diagnosis.
408 This model accounted for roughly 0.16% of the variance in age at diagnosis.

Age at Diagnosis ~ Health Issues + Language Background

409
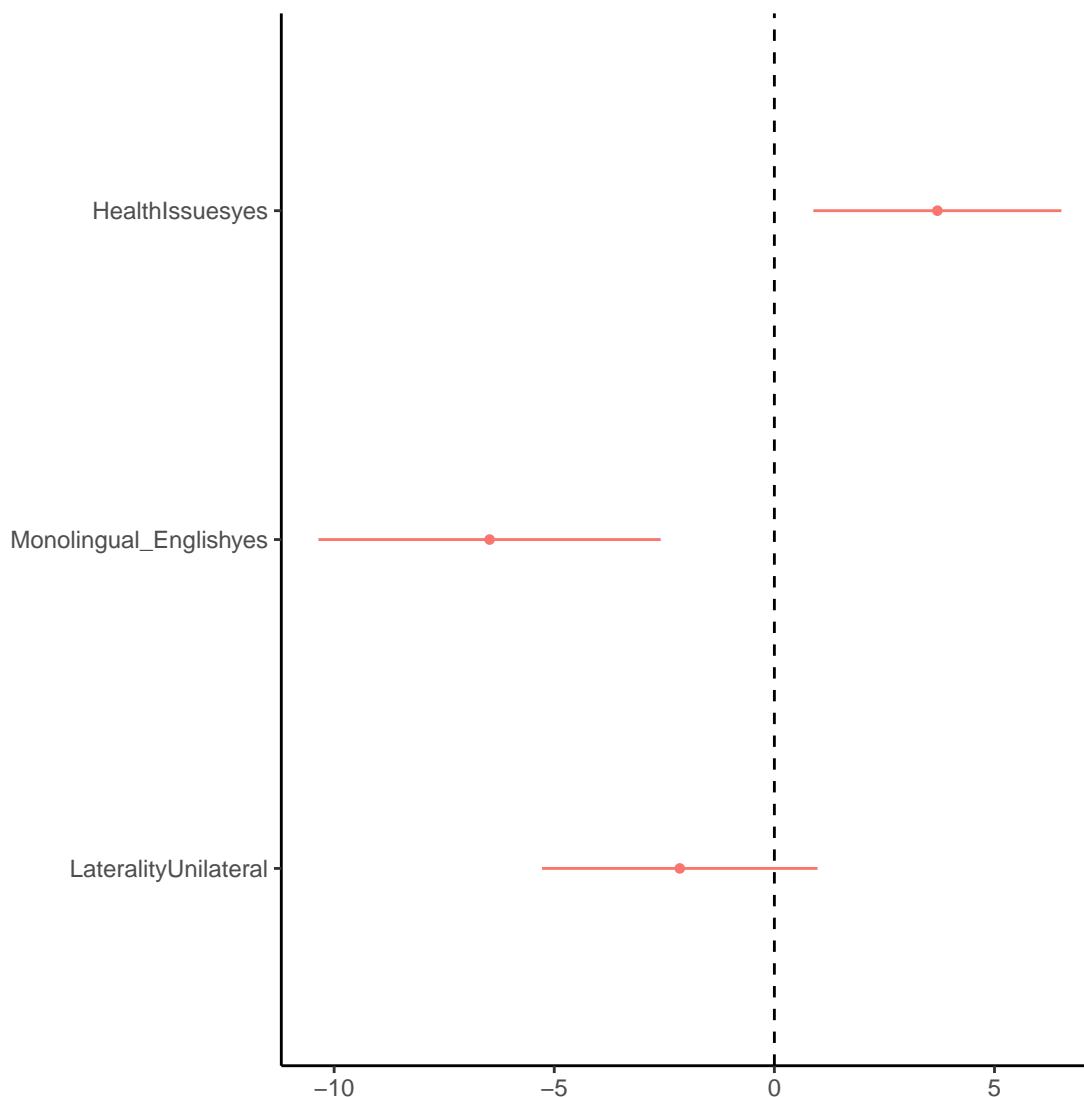
410   We repeated this model selection process for age at intervention. Our starting model

411   included the variables potentially relevant to intervention: Age intervention ~ gender +

412   degree (worse ear) + developmental delay + health issues + prematurity + laterality +

413   language background + etiology + age diagnosis Model selection by AIC removed Gender,

414   Developmental Delay, Health Issues, Prematurity, Language Background, and Etiology. In

415   the best fit model (R2=0.45 , p=0.00), prematurity (ß = 4.7, p = 0.02), less severe hearing

416   loss (ß = -0.09, p = 0.0042), later diagnosis (ß = 0.66, p = 1.1e-08), and coming from a

417   non-English-speaking household (ß = -1.38, p = 0.51) predicted later intervention and

418   accounted for roughly 0.45% of the variance in age at intervention. Infants born premature

419   were diagnosed 4.70 months later than full-term infants. A 1 decibel increase in hearing loss

420   results in 2.68 days earlier intervention. Each one month delay in diagnosis results in a 1.38

421   month delay in intervention.

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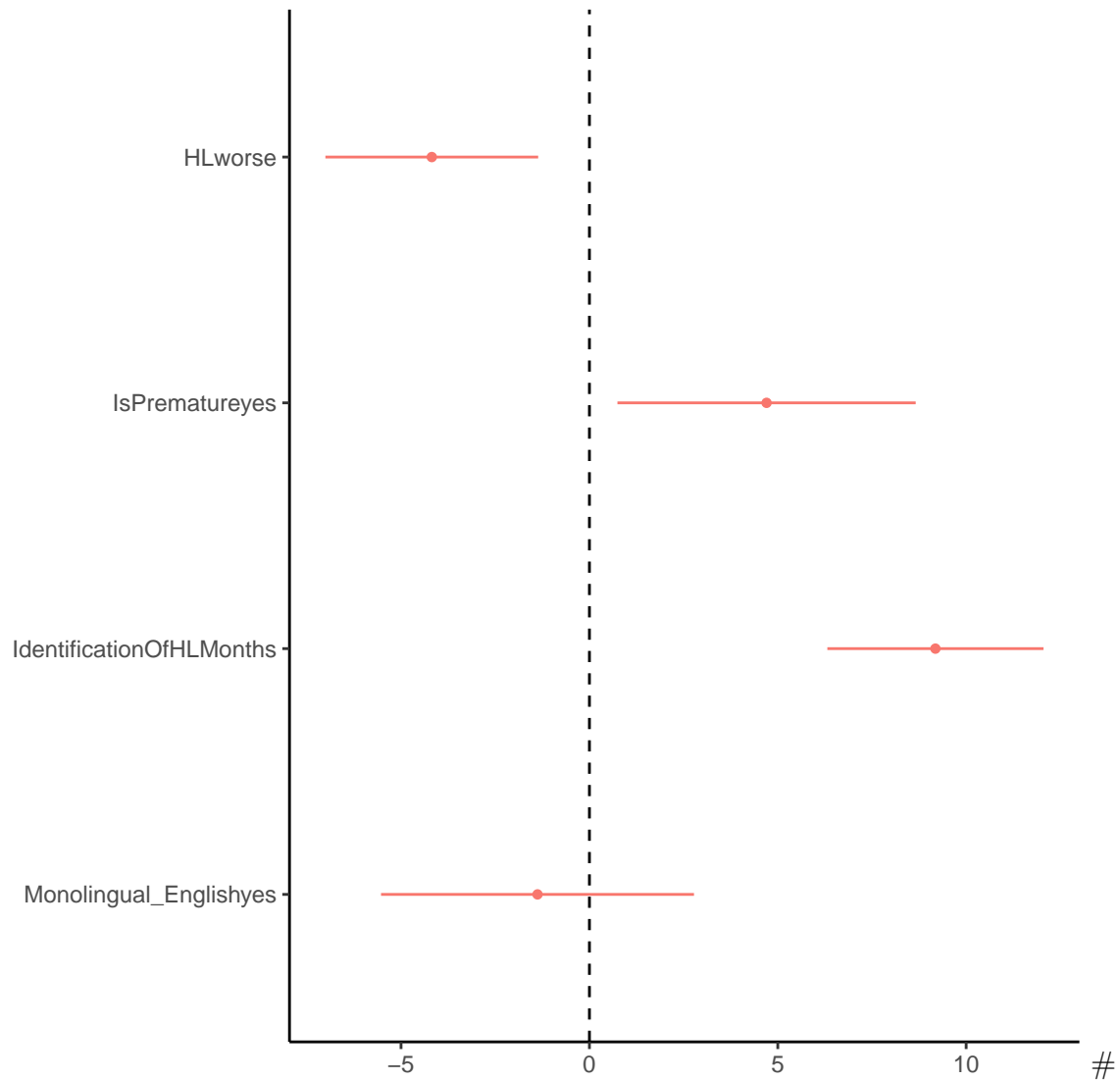
423 Discussion

Conclusion

425   Footnotes: Despite exciting, increasing, and converging evidence for benefits of early

426 sign language exposure (e.g., Schick, De Villiers, De Villiers, & Hoffmeister, 2007; Clark et

427 al., 2016; Davidson, Lillo-Martin, & Pichler, 2014; Hrastinski & Wilbur, 2016; Magnuson,

428 2000; Spencer, 1993), the majority of DHH children will not be raised in a sign language

429 environment. This is particularly true for North Carolina, which does not have a large

430 community of sign language users, relative to states like Maryland or areas like Washington

431 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