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

2   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, & 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), with cognitive ability more

predictive of language outcomes than presence or absence of a specific 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 and hearing loss 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). That is, 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, and includes 398 vocabulary items that parents indicate whether their child understands or produces, along with 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, and includes 680 vocabulary items that parents indicate whether their child produces, along with 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).

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].

200 **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 two subgroups of DHH children

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

206 disabilities and children with unilateral hearing loss (e.g., Yoshinaga-Itano et al., 2018).

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

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

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

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

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

212 regarding communication method, language background, or presence of other health issues

213 (e.g., congenital heart malformation).

214   **Methods**

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

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

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

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

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

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

221 Carolina.

222   The clinical evaluations included demographic and audiological information, CDI

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

224 detailed further below. For some children (n=0), multiple evaluations were available from

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

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

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

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

229 44 female) ages 4.20–36.17(M=21.21, SD=9.08). Race and SES information were not

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

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

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

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

234 (Jackson-Maldonado et al., 2003).

235   Children in this sample were coded as yes/no for cognitive development concerns (e.g.,

236 Down syndrome, global developmental delays; Cornelia de Lange syndrome), yes/no for

237 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 48dB 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, even though the child's adoptive parents are English-speaking.

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 (CITE). 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 NC 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

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.
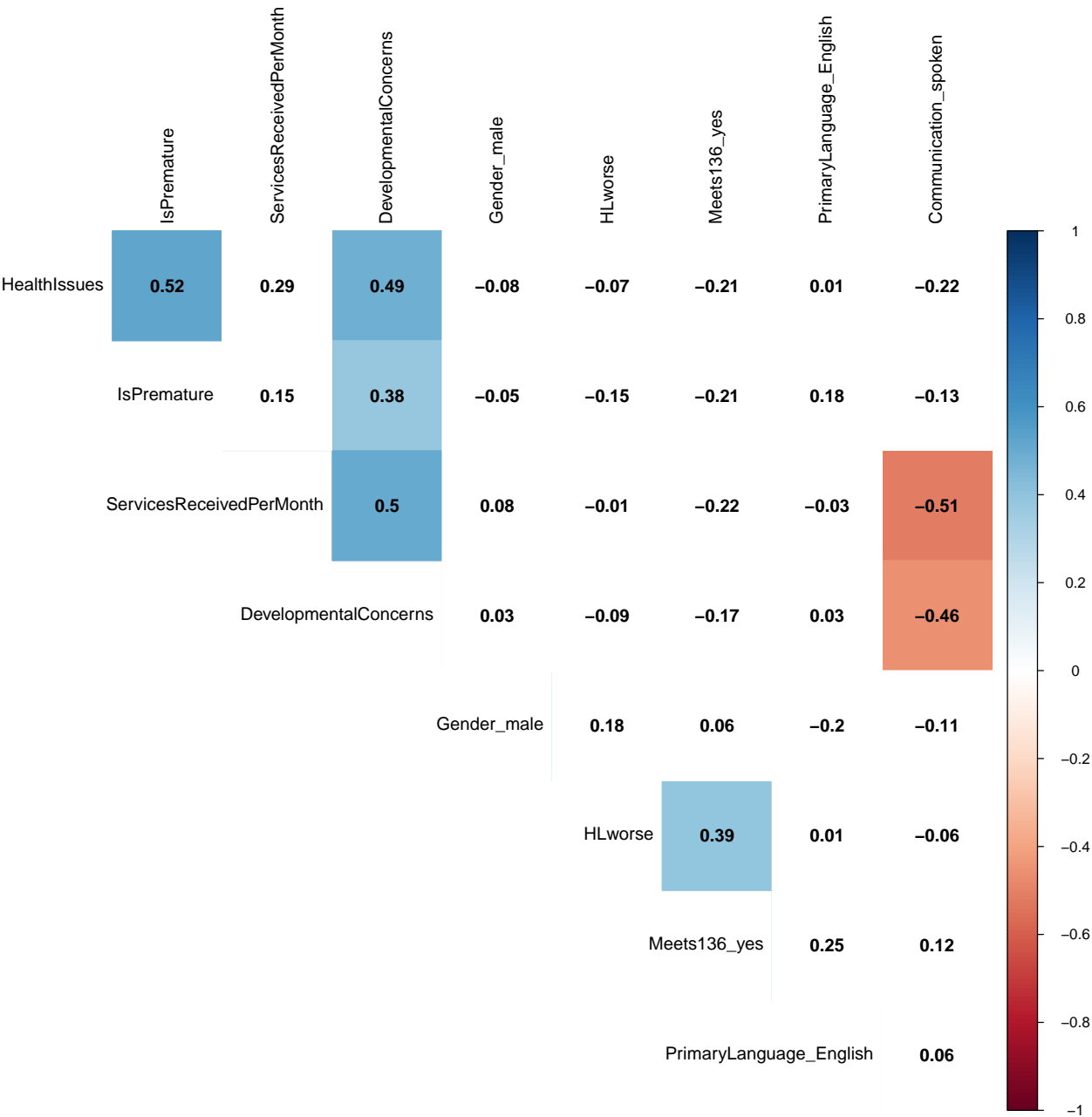
Results

All analyses were conducted in R. All code is available on Github. 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
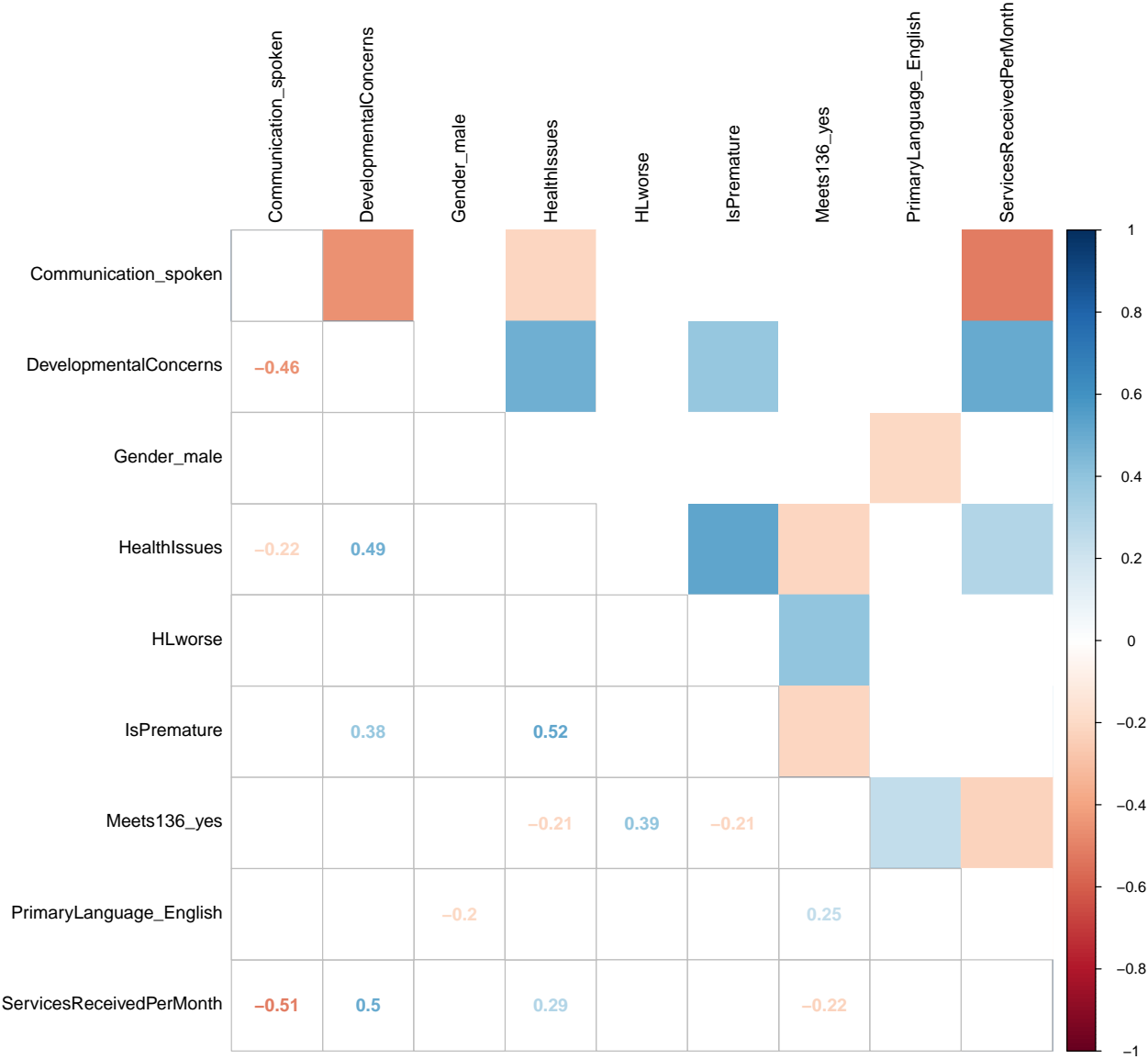
early diagnosis and intervention.

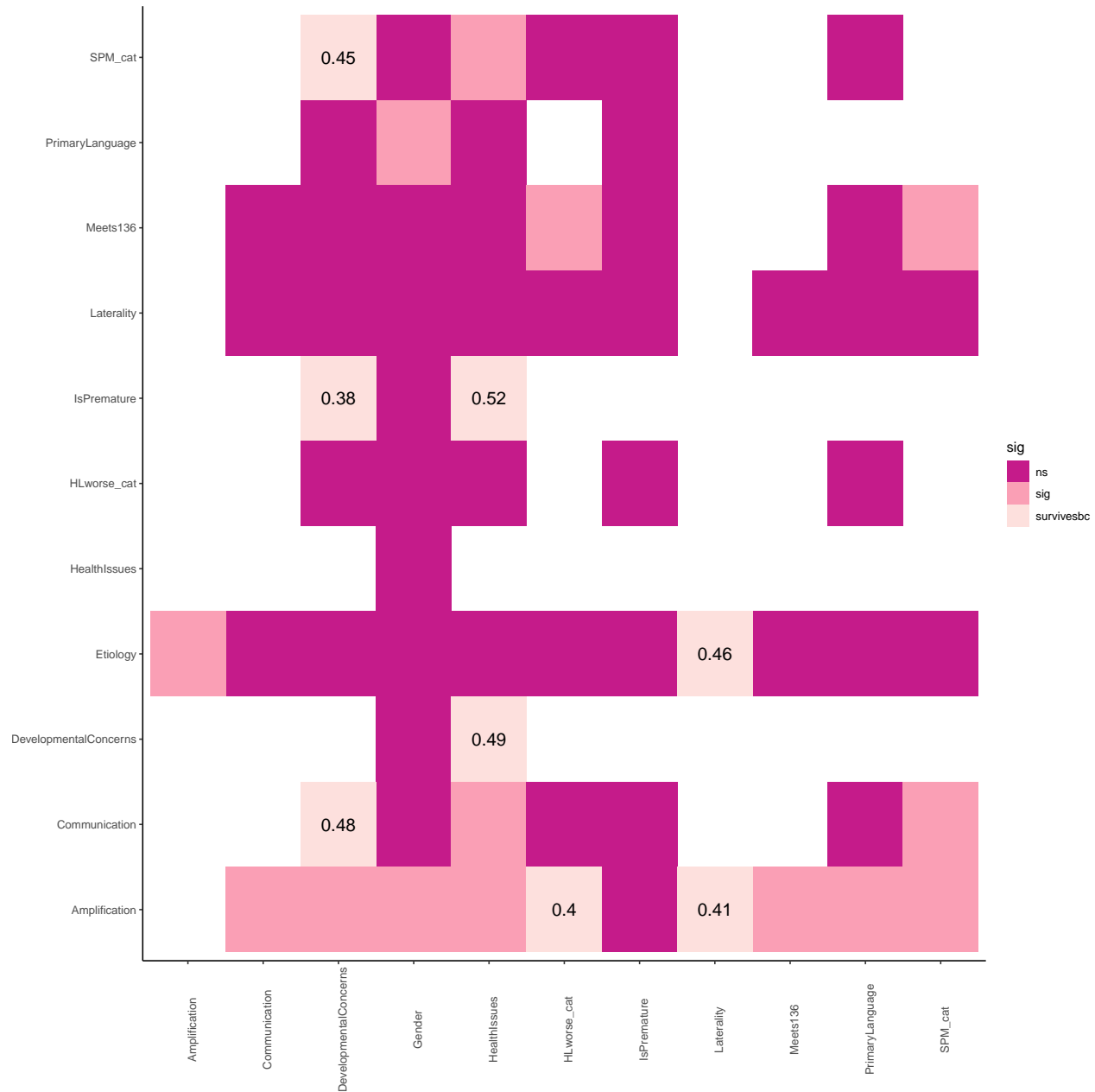**Part I: Interactions Among Variables**

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,

meets 1-3-6, amplification (binned into mild, moderate, severe/profound), etiology, services

received per month (binned into 0-3, 4-10, and >10), and degree of hearing loss).

Bonferroni-corrected alpha for this set of analyses was p < (.05/# predictors). Of the

XXX combinations of variables, p < .05 for XXX, and XXX survived bonferroni correction.

We are only discussing the results of tests that survived bonferroni correction, but the full
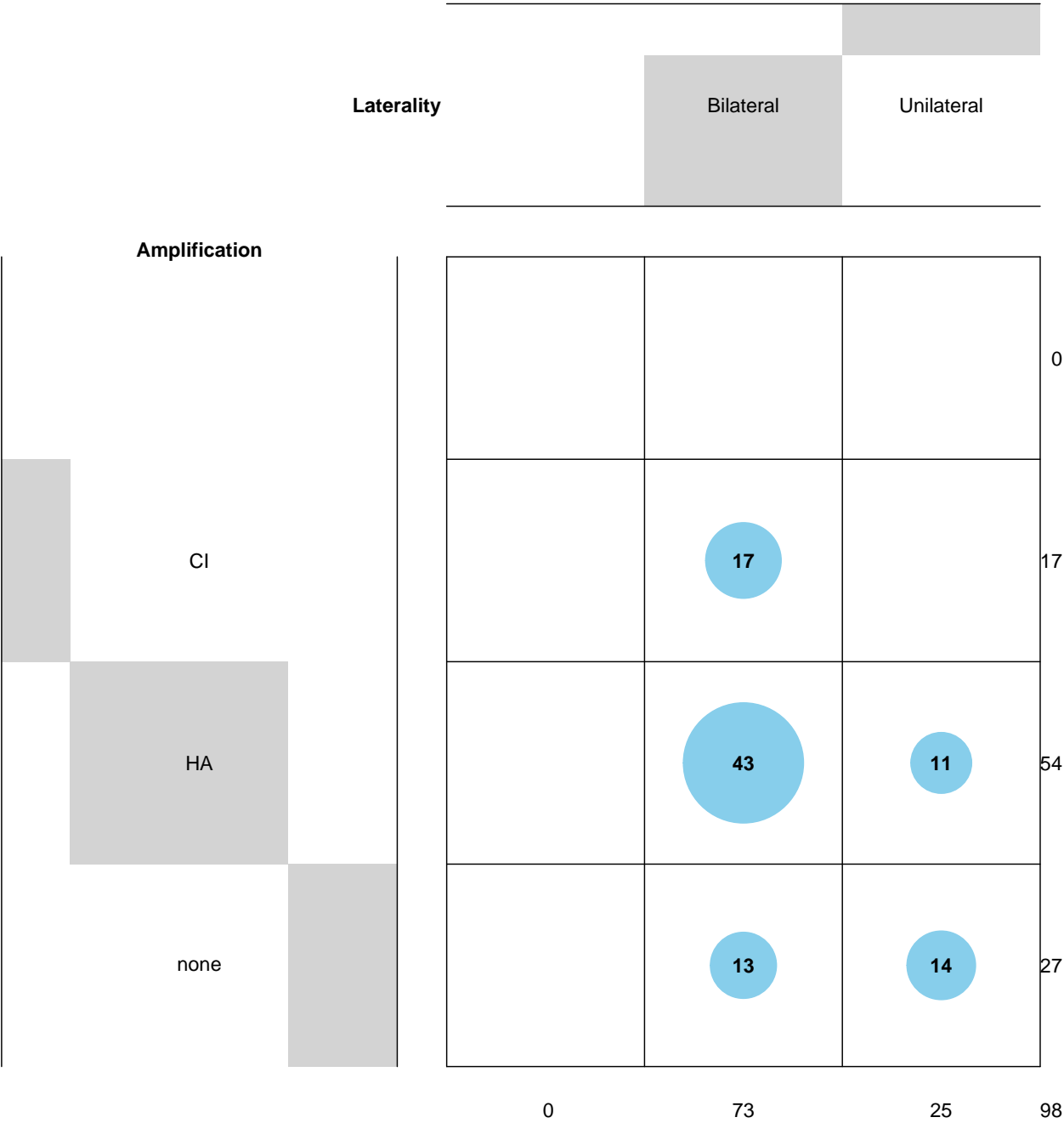
289 set of results can be found in table XXX.

	IsPremature	ServicesReceivedPerMonth	DevelopmentalConcerns	Gender_male	HLworse	Meets136_yes	PrimaryLanguage_English	Communication_spoken
HealthIssues	0.52	0.29	0.49	−0.08	−0.07	−0.21	0.01	−0.22
IsPremature		0.15	0.38	−0.05	−0.15	−0.21	0.18	−0.13
ServicesReceivedPerMonth			0.5	0.08	−0.01	−0.22	−0.03	−0.51
DevelopmentalConcerns				0.03	−0.09	−0.17	0.03	−0.46
Gender_male					0.18	0.06	−0.2	−0.11
HLworse						0.39	0.01	−0.06
Meets136_yes							0.25	0.12
PrimaryLanguage_English								0.06

290

From this set of analyses, we found XXX 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

299 developmental delays received more services per month than typically-developing children

300 (X^2 (2, N = 95) = 19.48, p = 5.9e-05) and were more likely to use total communication (X^2

301 (3, N = 98) = NaN, p = NA). Likewise, children who used total communication received

302 more services per month than children using spoken language (X^2 (6, N = 95) = NaN, p =

303 NA).

304   We also found relationships among many of the audiological characteristics. There was

305 a significant relationship between laterality and etiology (X^2 (6, N = 88) = NaN, p = NA),

306 such that children with conductive hearing loss were more likely to have unilateral hearing

307 loss, children with sensorineural hearing loss were more likely to have a bilateral loss, and all

308 children with mixed hearing loss (n = XXX) had bilateral hearing loss. Chi-square tests

309 showed that laterality (X^2 (6, N = 98) = NaN, p = NA) and degree of hearing loss (X^2 (6,

310 N = 87) = NaN, p = NA) were related to amplification in our sample. Children with

311 bilateral hearing loss were more likely than children with unilateral hearing loss to use a

312 hearing aid or cochlear implant; no child with unilateral hearing loss used a cochlear implant,

313 and many children with unilateral hearing loss used no amplification. Regarding degree,

314 children with severe-profound hearing loss were more likely to use a cochlear implant than

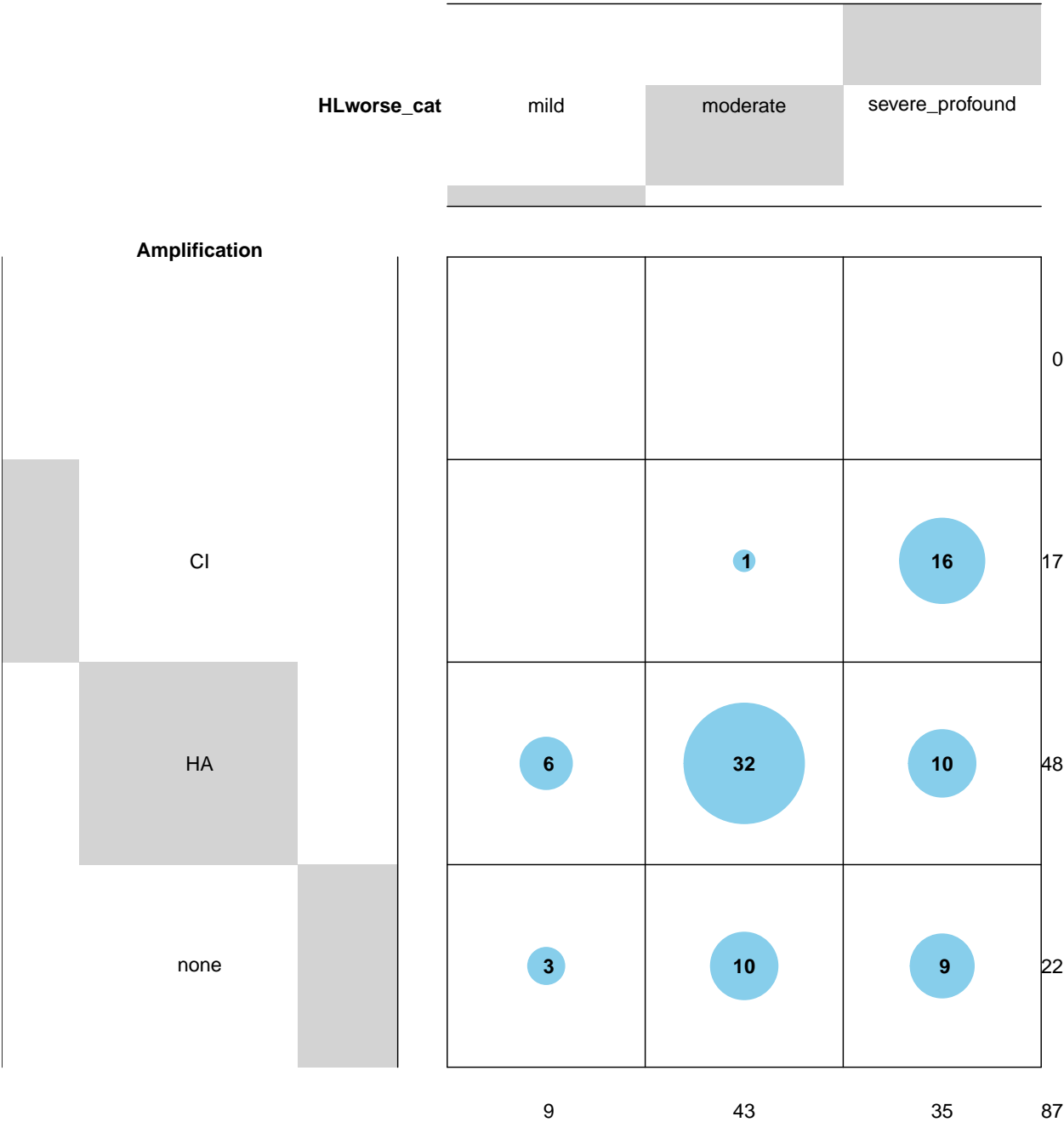315 children with less severe hearing loss (i.e., mild or moderate).
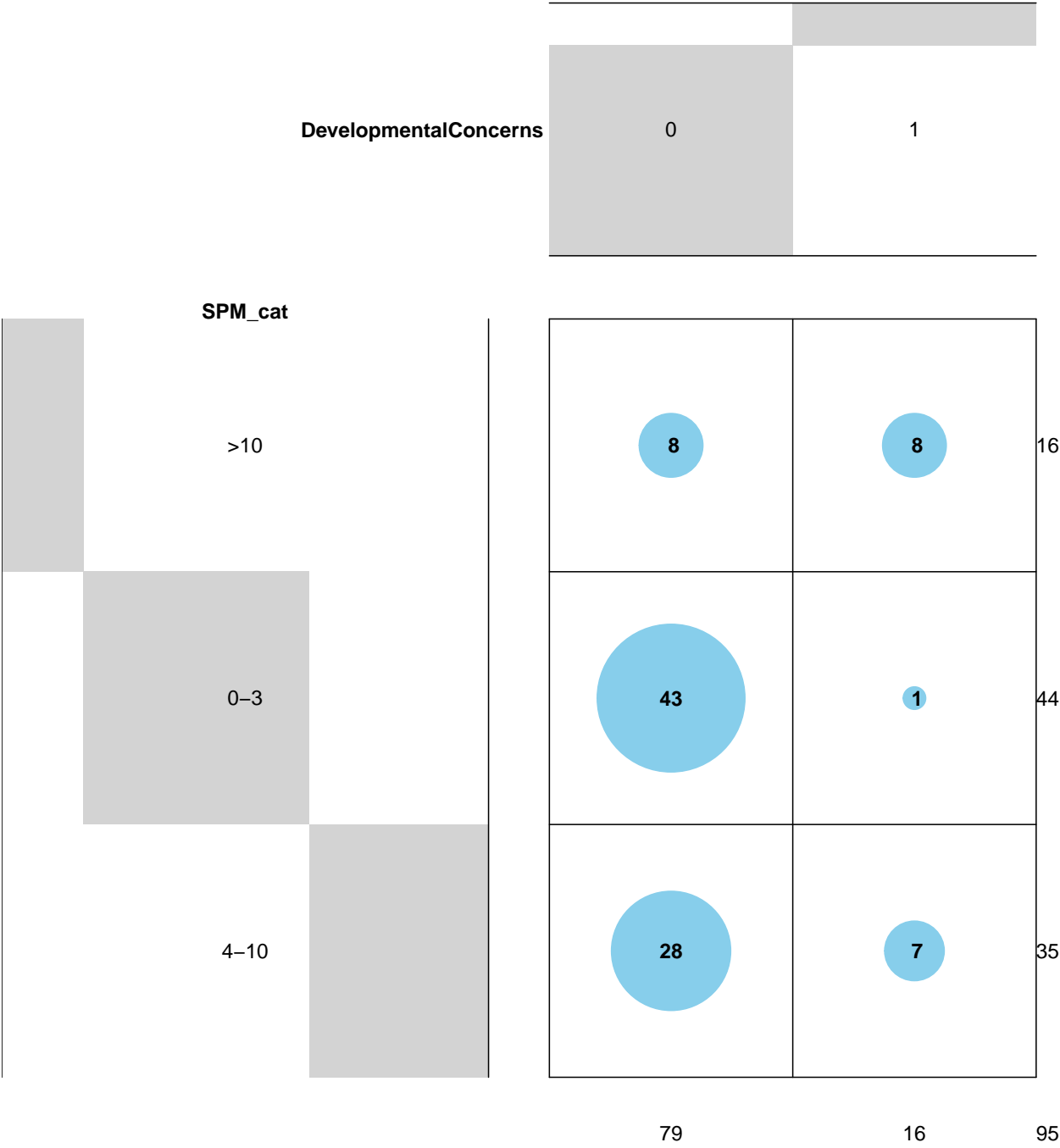
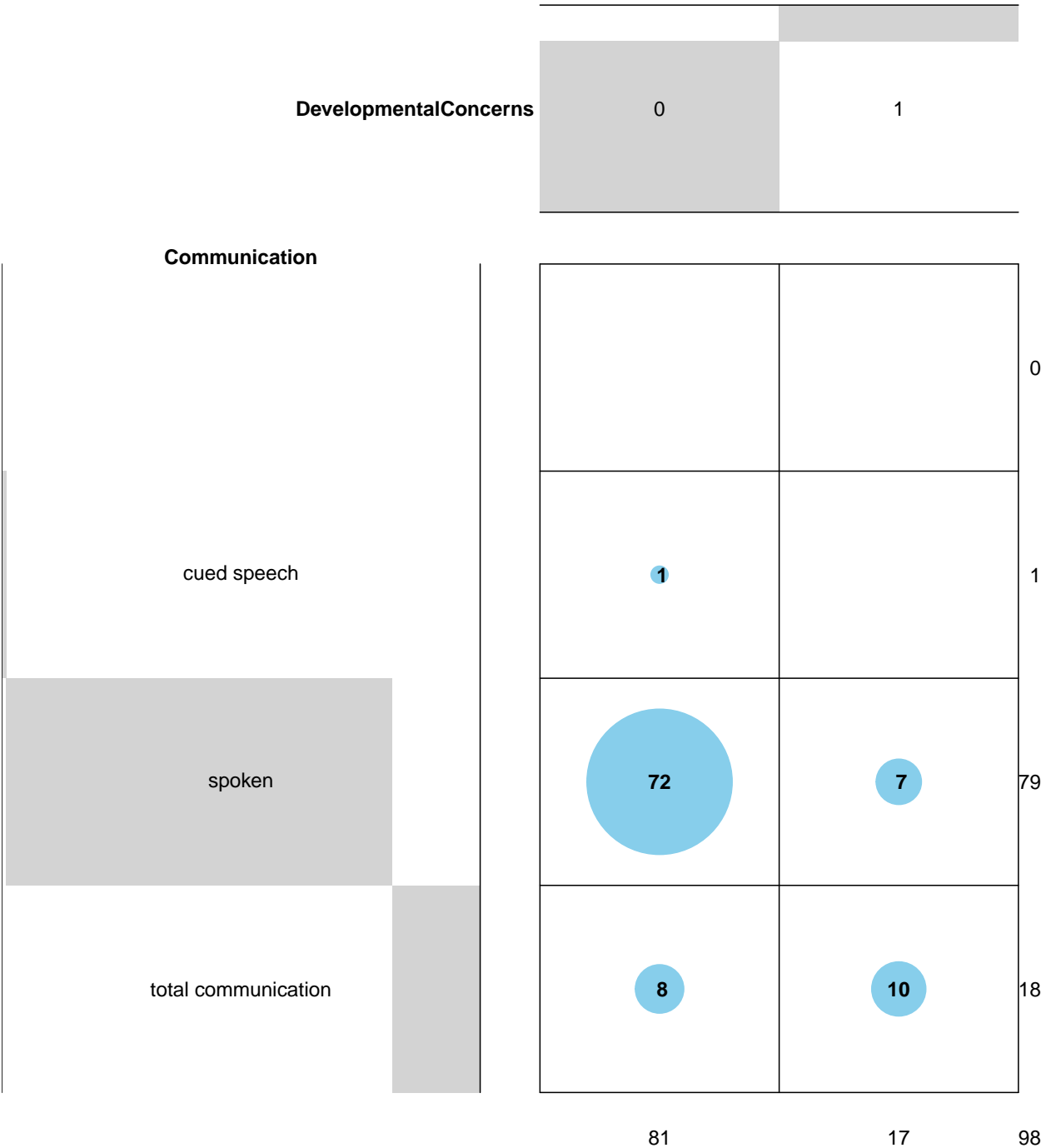## Laterality by Amplification

Laterality by Etiology

Etiology	Laterality	Bilateral	Unilateral	
				0
Conductive		7	11	18
Mixed		8		8
SNHL		52	10	62
		0 67	21	88
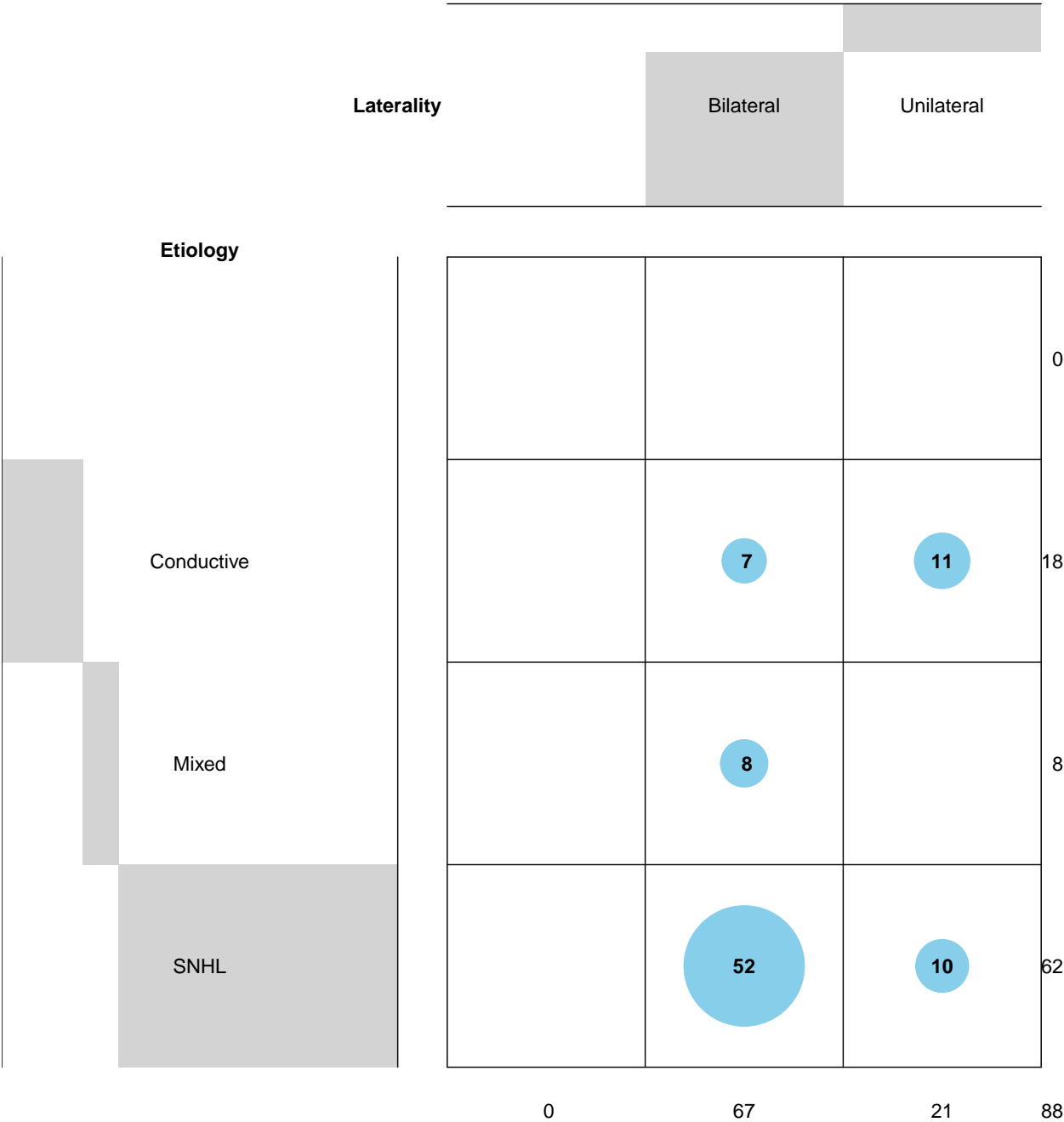
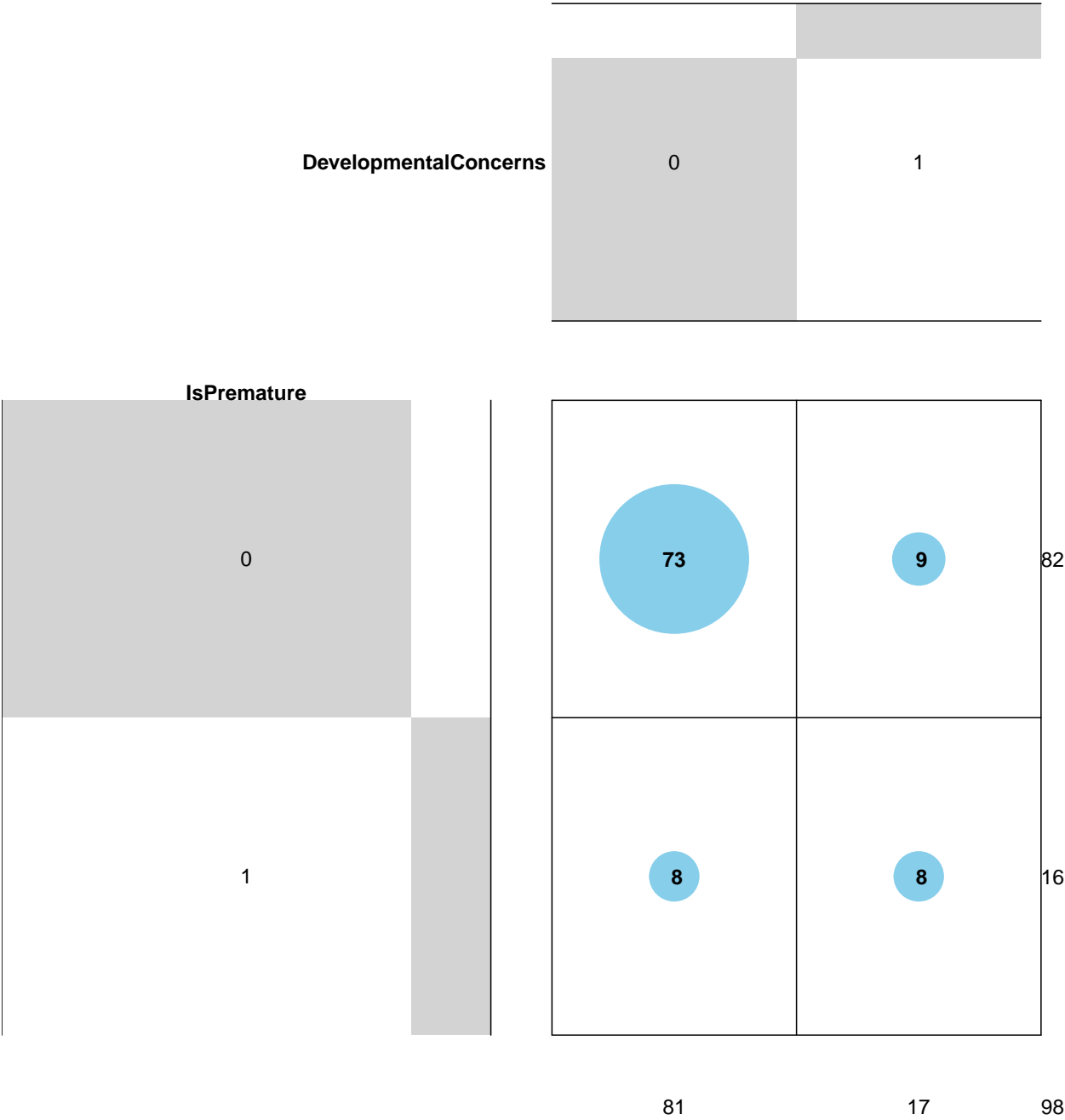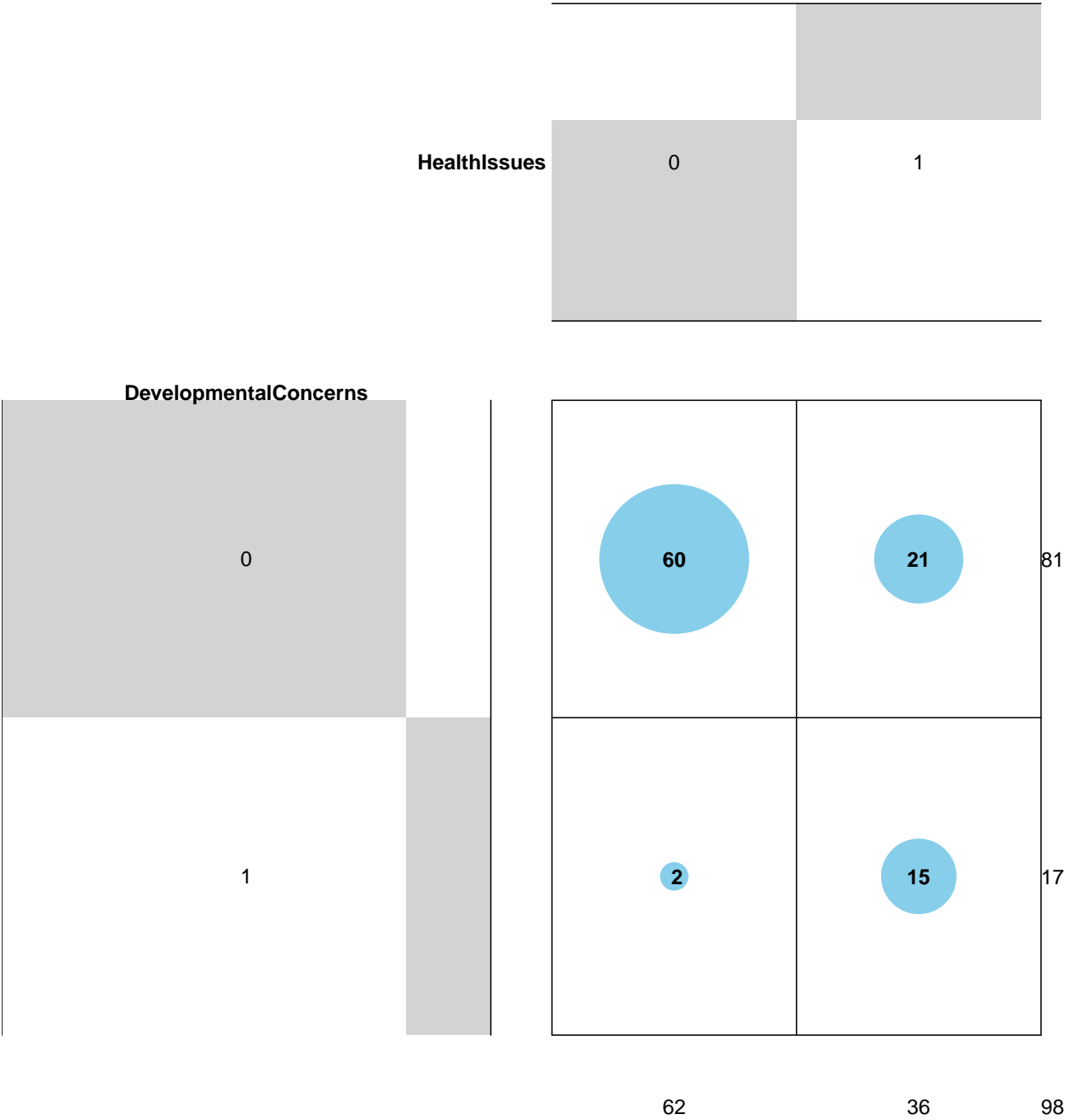**HLworse_cat by Amplification**

DevelopmentalConcerns by SPM_cat

DevelopmentalConcerns by Communication

DevelopmentalConcerns by IsPremature

HealthIssues by DevelopmentalConcerns

HealthIssues

| | 0 | 1 |

DevelopmentalConcerns

| DevelopmentalConcerns	0	1
	60	21	81
	2	15	17
	62	36	98

HealthIssues by IsPremature
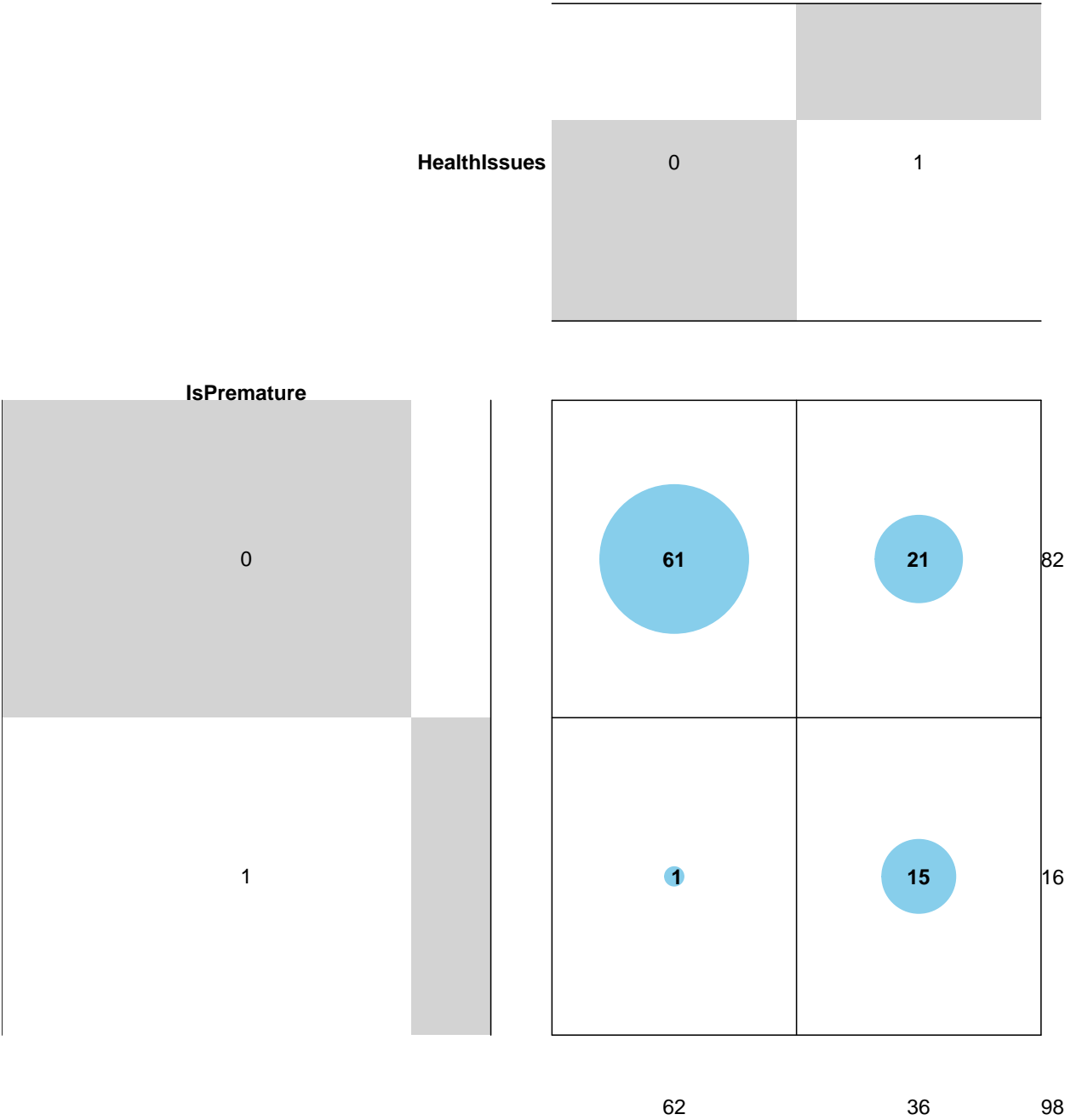
Part II: Influence on vocabulary

We next turn to the relationship between each of these variables and children's

productive vocabulary, measured on the CDI. 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 XXX on Words and Gestures and XXX 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 (X number typically-developing infants; (CITE)) 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. ELSSP

Normative WS vs. ELSSP
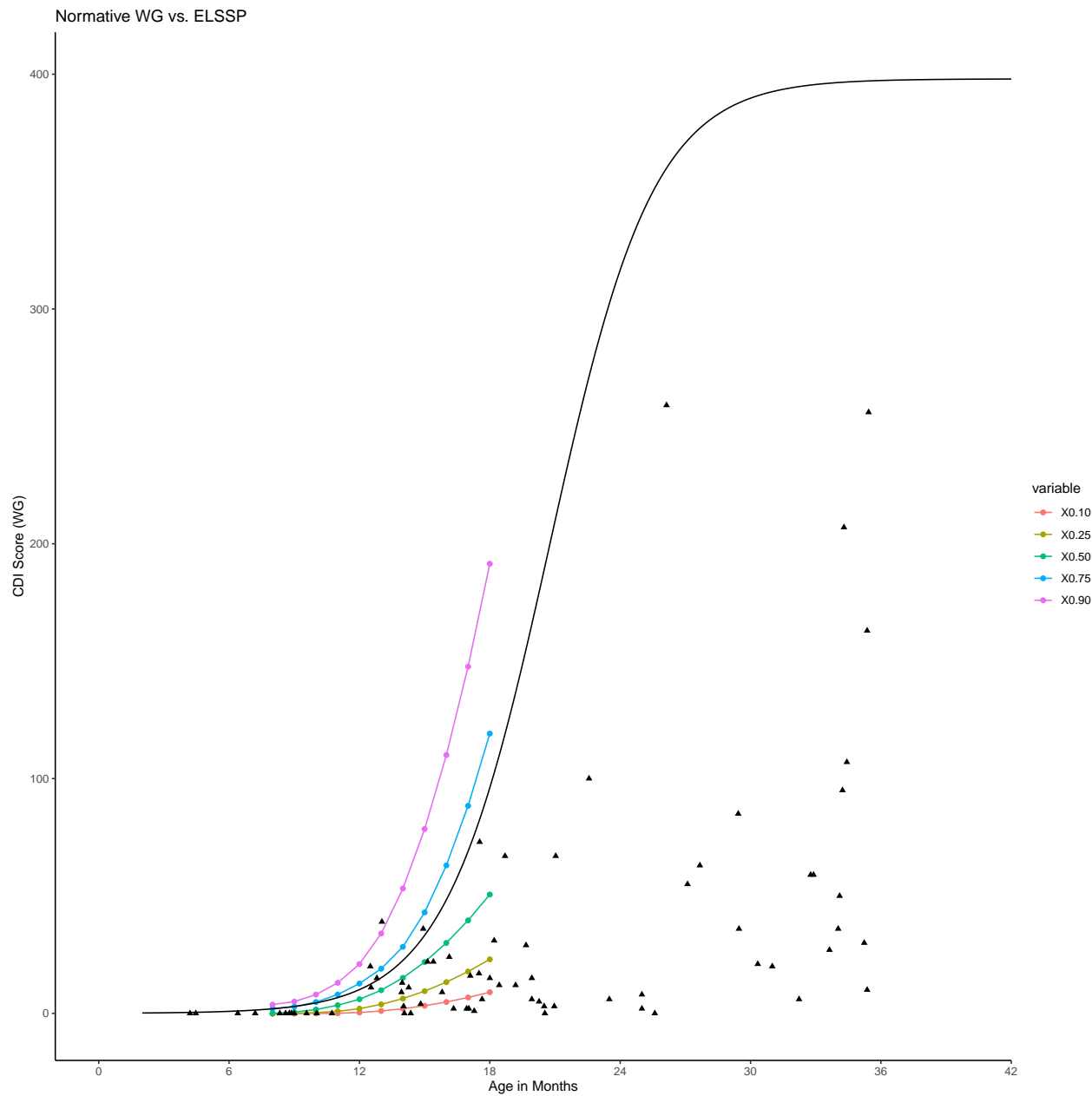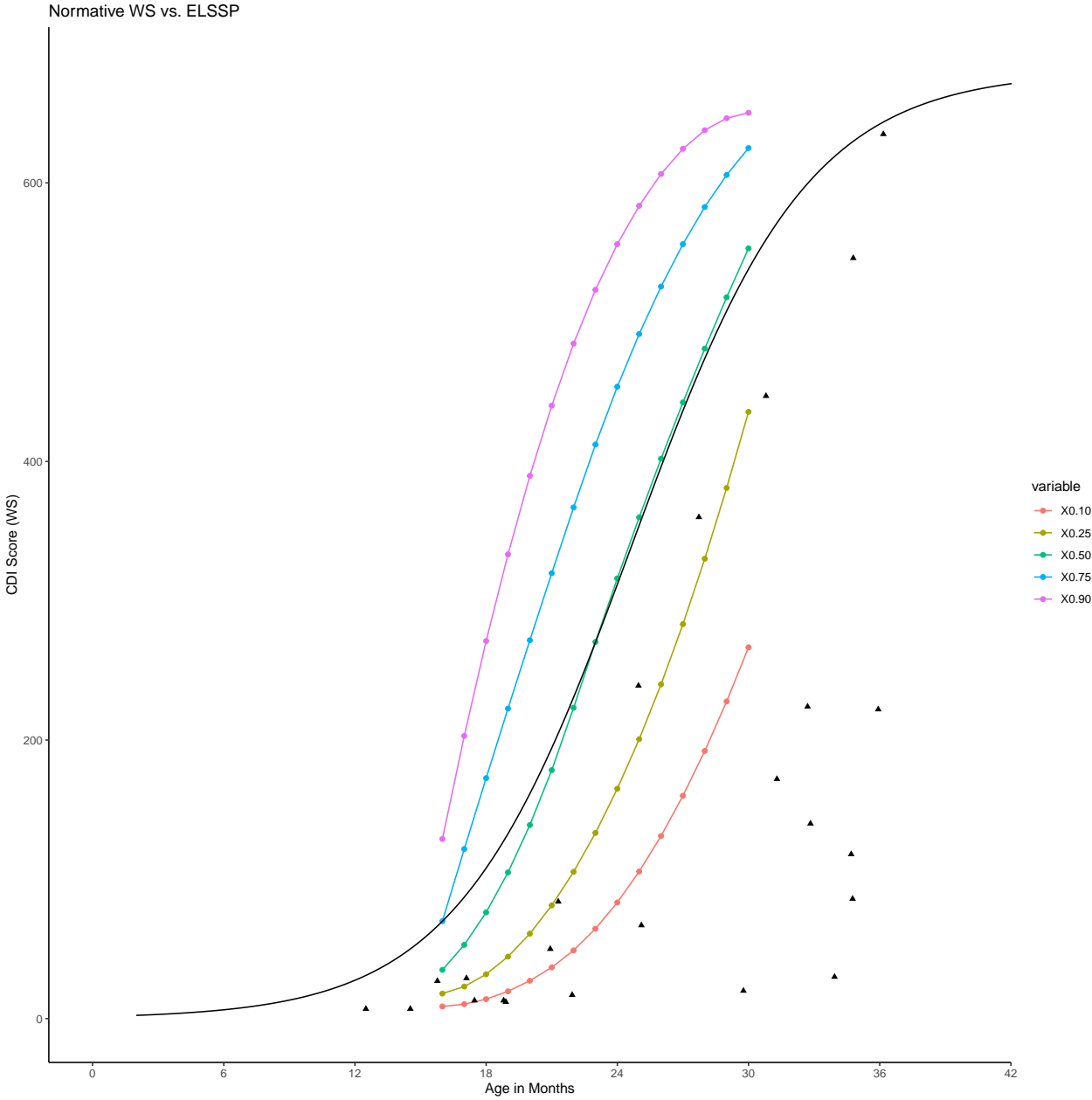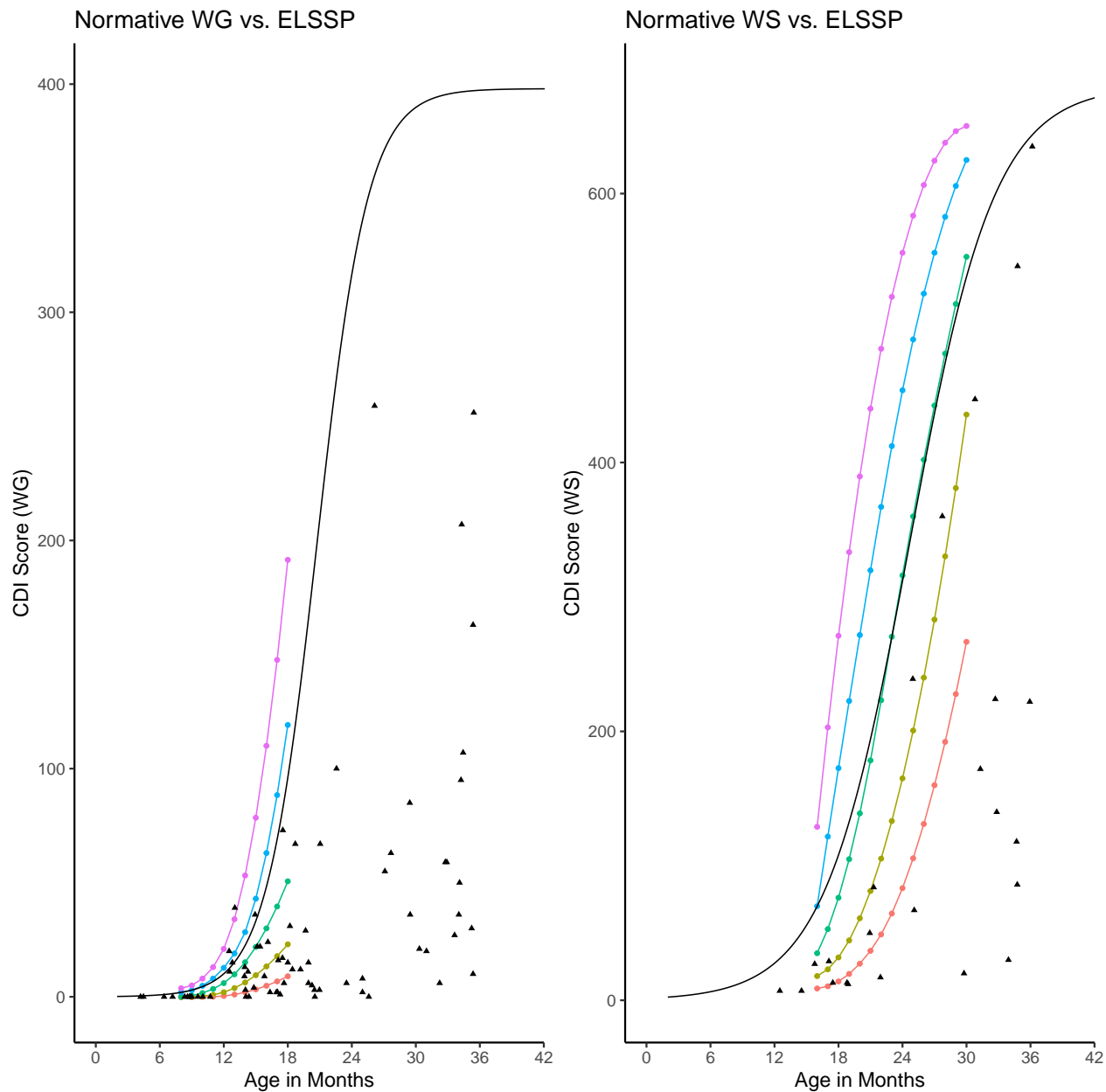
To look at the relationship between our predictor variables and vocabulary delay, we
created a linear regression model using stepAIC. We also present bonferroni-corrected
zero-order tests of each predictor on vocabulary delay. We exclude children from
non-English-speaking families from this section of the analysis because our growth curves are
based on the English language CDI.

Our full regression model included all of our variables (STATS): Vocabulary Delay ~

Gender + Developmental Delay + Health Issues + Prematurity + Laterality + Degree +
Amplification + Communication + Meets 1-3-6 + ServicesReceivedPerMonth. We performed
stepwise model comparison using stepAIC (cite 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.

```
FALSE Start:  AIC=373.89
FALSE diff_age_from_expected ~ Gender + DevelopmentalConcerns + HealthIssues +
FALSE     IsPremature + Laterality + HLworse + Amplification + Communication +
FALSE     Meets136
FALSE
FALSE                         Df Sum of Sq    RSS    AIC
FALSE - Meets136              2      8.01 8154.4 369.97
FALSE - Communication        2     50.79 8197.1 370.35
FALSE - IsPremature          1    104.43 8250.8 372.84
FALSE <none>                            8146.4 373.89
FALSE - Gender               1    462.99 8609.3 375.98
FALSE - Laterality           1    593.94 8740.3 377.10
FALSE - HealthIssues         1    652.02 8798.4 377.59
FALSE - HLworse              1    660.37 8806.7 377.66
FALSE - DevelopmentalConcerns 1    706.09 8852.4 378.04
FALSE - Amplification        2   1320.27 9466.6 381.01
FALSE
FALSE Step:  AIC=369.97
FALSE diff_age_from_expected ~ Gender + DevelopmentalConcerns + HealthIssues +
FALSE     IsPremature + Laterality + HLworse + Amplification + Communication
```

```
FALSE

FALSE                          Df Sum of Sq    RSS    AIC

FALSE - Communication          2     50.85  8205.2 366.43

FALSE - IsPremature            1    102.98  8257.3 368.89

FALSE <none>                              8154.4 369.97

FALSE - Gender                 1    463.89  8618.3 372.06

FALSE - Laterality             1    617.44  8771.8 373.37

FALSE - HealthIssues           1    668.30  8822.7 373.80

FALSE - DevelopmentalConcerns  1    707.17  8861.5 374.12

FALSE - HLworse                1    719.53  8873.9 374.22

FALSE - Amplification          2   1360.12  9514.5 377.38

FALSE

FALSE Step:  AIC=366.43

FALSE diff_age_from_expected ~ Gender + DevelopmentalConcerns + HealthIssues +

FALSE      IsPremature + Laterality + HLworse + Amplification

FALSE

FALSE                          Df Sum of Sq    RSS    AIC

FALSE - IsPremature            1    103.99  8309.2 365.36

FALSE <none>                              8205.2 366.43

FALSE - Gender                 1    547.26  8752.5 369.20

FALSE - HealthIssues           1    642.12  8847.3 370.00

FALSE - Laterality             1    676.82  8882.0 370.29

FALSE - HLworse                1    773.97  8979.2 371.10

FALSE - DevelopmentalConcerns  1    779.33  8984.5 371.14

FALSE - Amplification          2   1758.61  9963.8 376.80

FALSE

FALSE Step:  AIC=365.36
```

```
408  FALSE diff_age_from_expected ~ Gender + DevelopmentalConcerns + HealthIssues +

409  FALSE      Laterality + HLworse + Amplification

410  FALSE

411  FALSE                              Df Sum of Sq      RSS      AIC

412  FALSE <none>                                      8309.2 365.36

413  FALSE - HealthIssues            1    538.16   8847.4 368.00

414  FALSE - Gender                  1    581.61   8890.8 368.36

415  FALSE - Laterality              1    723.01   9032.2 369.53

416  FALSE - HLworse                 1    746.60   9055.8 369.73

417  FALSE - DevelopmentalConcerns   1    996.31   9305.5 371.74

418  FALSE - Amplification           2   1783.90  10093.1 375.75


419  FALSE

420  FALSE Call:

421  FALSE lm(formula = diff_age_from_expected ~ Gender + DevelopmentalConcerns +

422  FALSE      HealthIssues + Laterality + HLworse + Amplification, data = (full_elssp %>%

423  FALSE      filter(PrimaryLanguage == "English")))

424  FALSE

425  FALSE Coefficients:

426  FALSE           (Intercept)                Gendermale  DevelopmentalConcerns

427  FALSE               -9.6942                    6.0664                12.1442

428  FALSE          HealthIssues  LateralityUnilateral                 HLworse

429  FALSE               -6.7421                   -8.8503                 0.1731

430  FALSE         AmplificationHA       Amplificationnone

431  FALSE               12.2372                   19.9404
```

Our final model included: Vocabulary Delay ~ Gender + Developmental Delay + Health Issues + Laterality + Degree + Amplification ($R^2 = 0.35$, p=0.00). In this model,
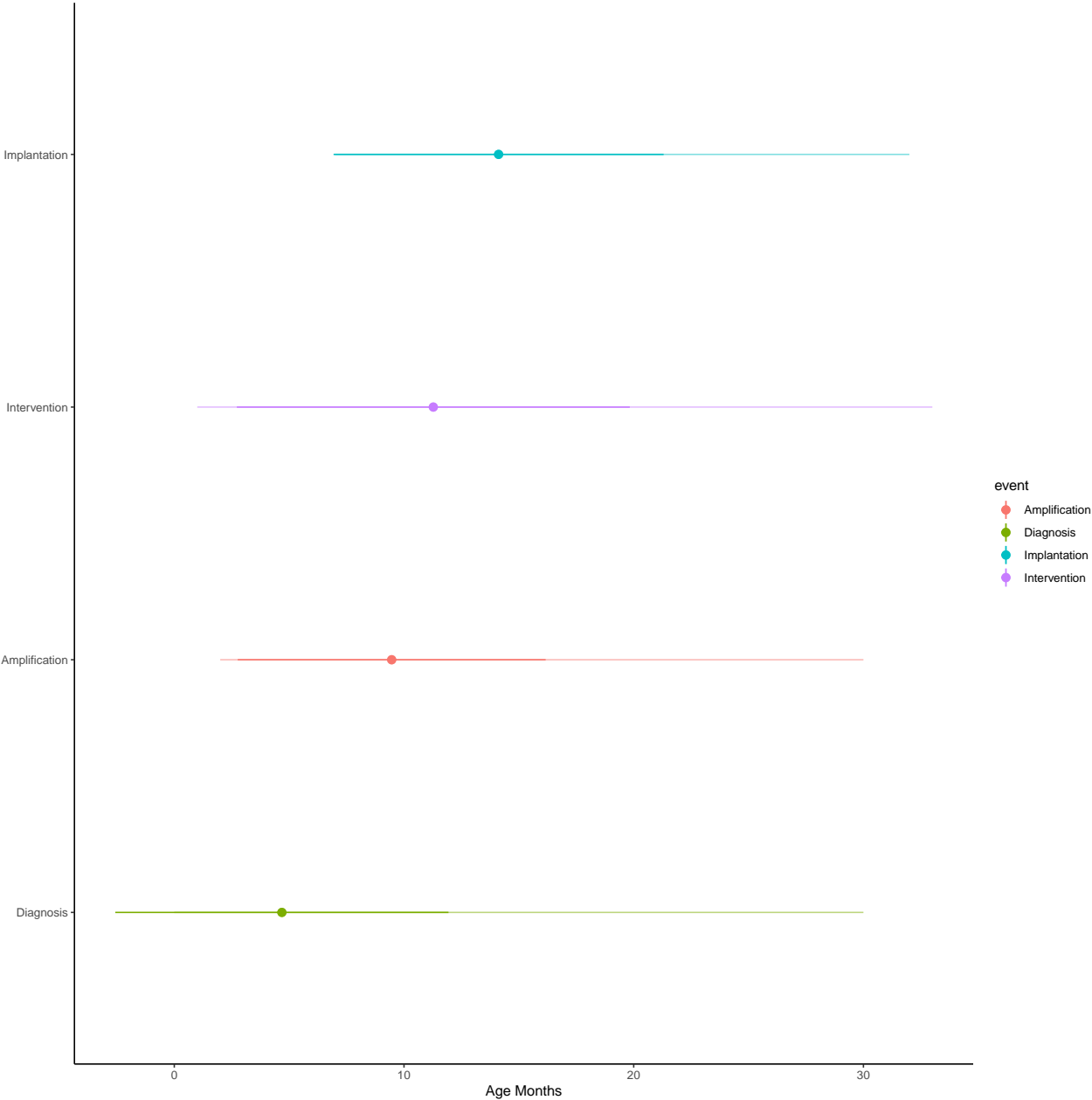
being male, having a developmental delay, bilateral hearing loss, and more severe hearing

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

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

amplification. Presence of health issues trended towards smaller vocabulary delay, but this

predictor was not significant. This model accounted for roughly 0% of the variance in

children's vocabulary delay.
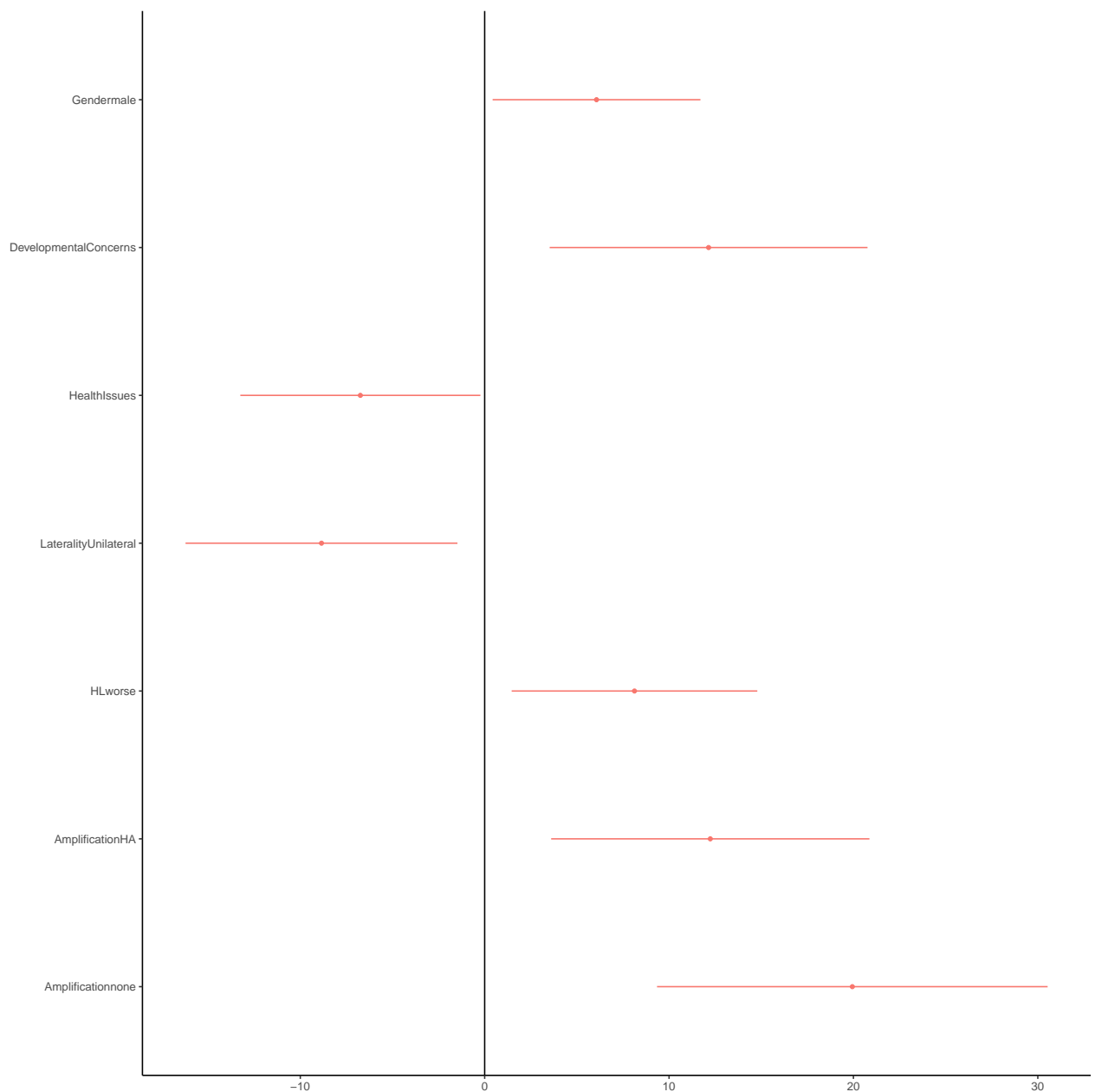
441 **Part III: Meets136 success**

442   Lastly, we looked at the ages at which children received diagnosis and intervention, and

443 how this mapped onto the 1-3-6 guidelines. Overall, 61% of our sample met 1-3-6 guidelines

444 for early diagnosis and intervention.

Figure XXX – Dots represent average age of diagnosis, etc. Stronger lines represent standard deviation. Softer lines show range.

445

446   We created linear regression models for age at diagnosis and age at intervention.

447 Models were paired down using stepwise regression by AIC using the stepAIC function (cite

448 MASS package). For age at diagnosis, we included the set of child-specific factors that would

449 be relevant before diagnosis of hearing loss. We began with:

$$Age at Diagnosis \sim Gender + Degree of Hearing Loss(worse ear) + Developmental Delay + Health Issues + F$$

450 Age diagnosis ~ gender + laterality + degree (worse ear) + developmental delay + health

451 issues + prematurity + laterality + language background + etiology The best fit model

452 (R2=0.25 , p=0.00) included health issues (ß = 3.99, p = 0.0039) and language background

453 (ß = 27.69, p = 3.8e-05).

$$Age\ at\ Diagnosis \sim Health\ Issues + Language\ Background$$
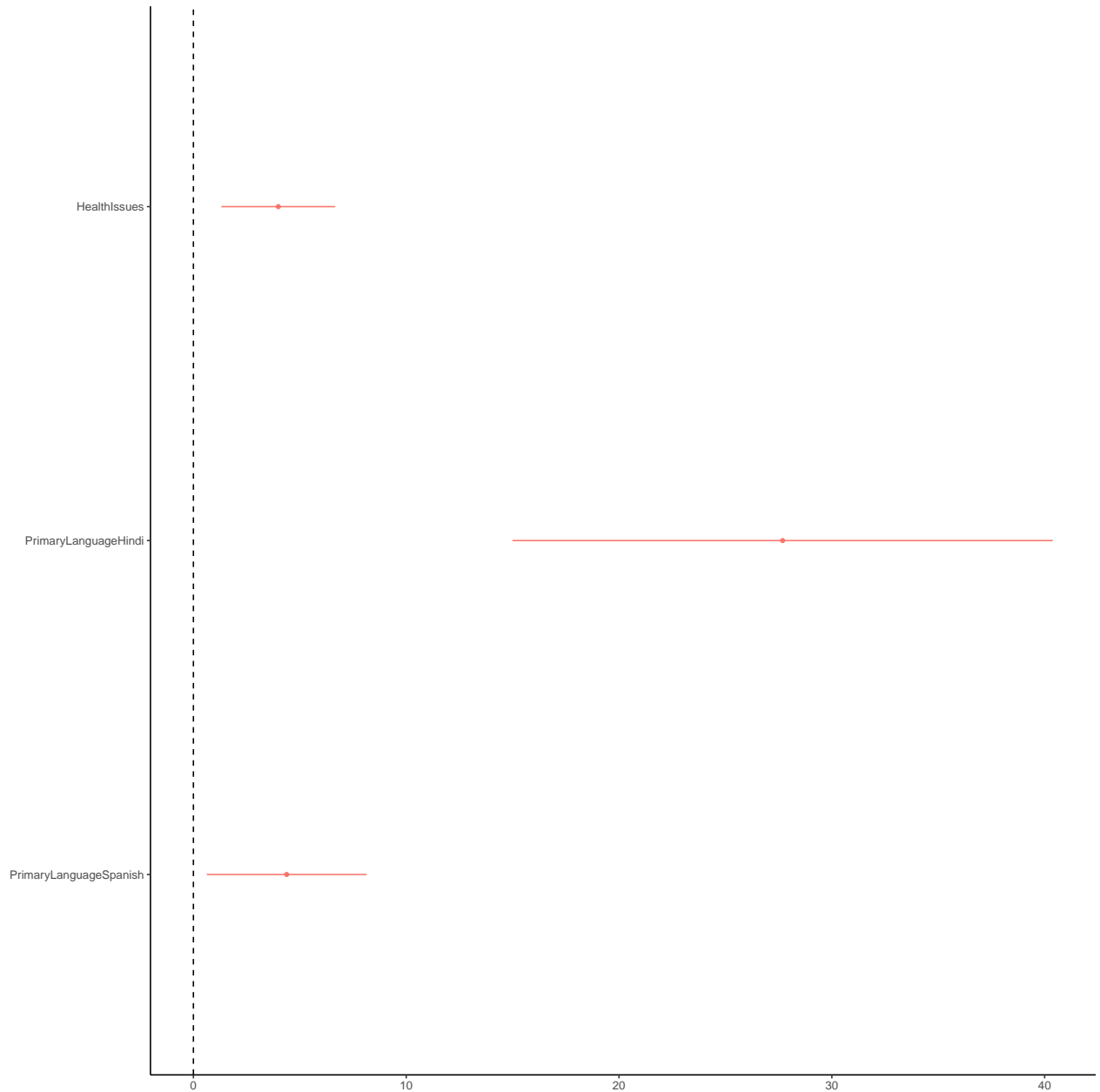
455 For age at intervention, we first included the variables potentially relevant prior to

456 intervention: Age intervention ~ gender + degree (worse ear) + developmental delay +

457 health issues + prematurity + laterality + language background + etiology + age diagnosis

$$AgeatIntervention \sim Gender + Degree of Hearing Loss(worse ear) + Developmental Delay + Health Issues$$

458 The best fit model (R2=0.45 , p=0.00) included prematurity (ß = 4.46, p = 0.025), degree of

459   hearing loss (ß = -0.09, p = 0.0038), and age at diagnosis (ß = 0.67, p = 1.9e-09).

$$AgeatIntervention \sim DegreeofHearingLoss(worseear) + Prematurity + AgeatDiagnosis$$
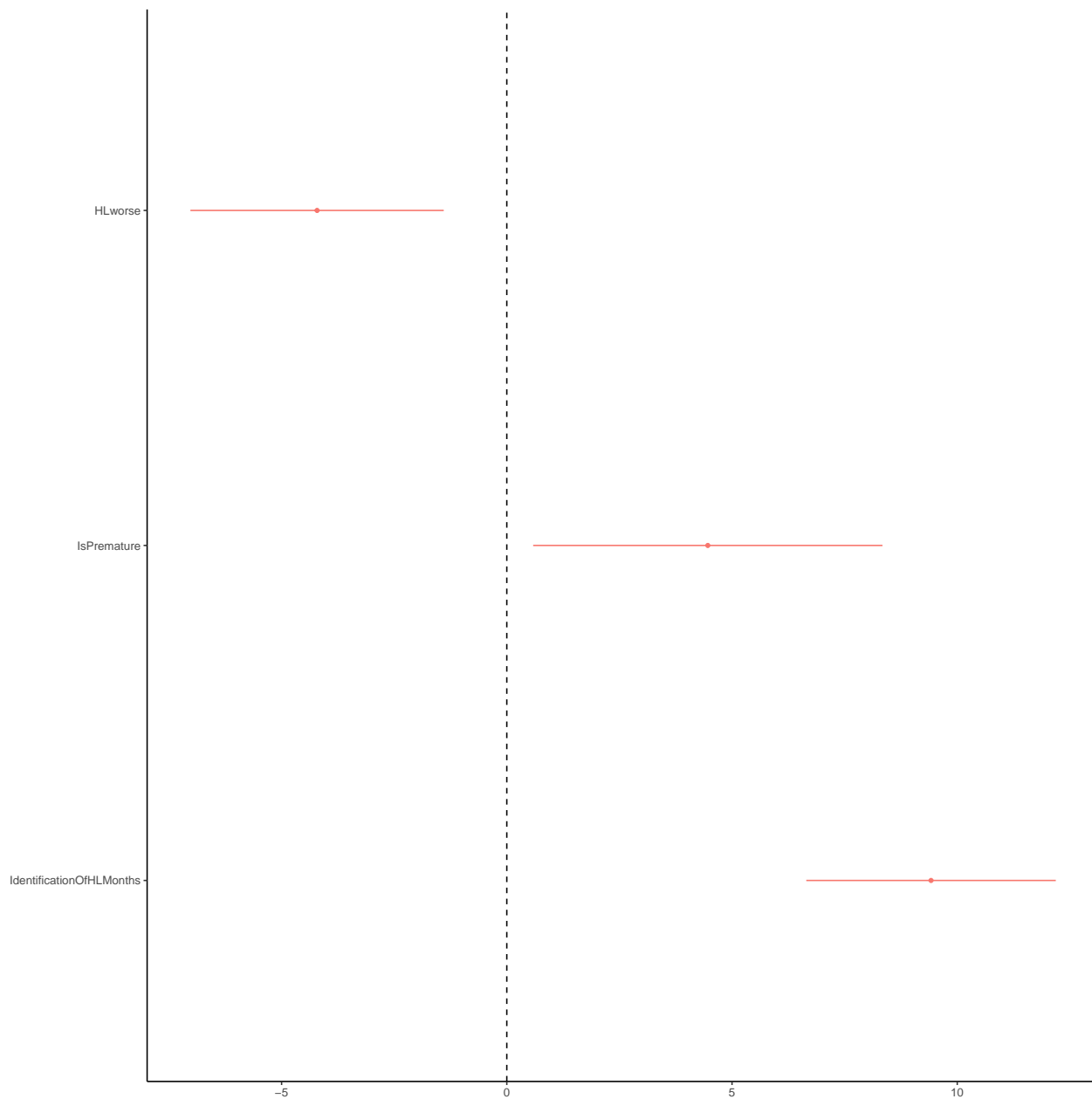
460

461   # Discussion

462   **Conclusion**

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

464   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

Table 8

Delay table

Variable	mean delays	Method
Gender	Boy: 16.3; Girl: 10.3	wilcox
Laterality	Unilateral: 11.1; Bilateral: 14.4	wilcox
Amplification	CI: 10; HA: 12.2, none: 18.6	kruskall
Health Issues	Yes: 12.9; No: 13.9	wilcox
Developmental Delay	Yes: 21.4; No: 12.1	wilcox
Prematurity	Premature: 17.2; Full-term: 12.9	wilcox
1-3-6 Guidelines	Meets: 12.3; Does not meet: 14.2	wilcox
Communication	Spoken Language: 12.5; Total Communication: 19.3	wilcox
Etiology	SNHL: 13.2; Mixed: 17.4, Conductive: 13.3	kruskall
Degree	More severe: 14.5; Less severe: 13.3	wilcox
Services Received Per Month	More services: 16.5; Less services: 11.9	wilcox