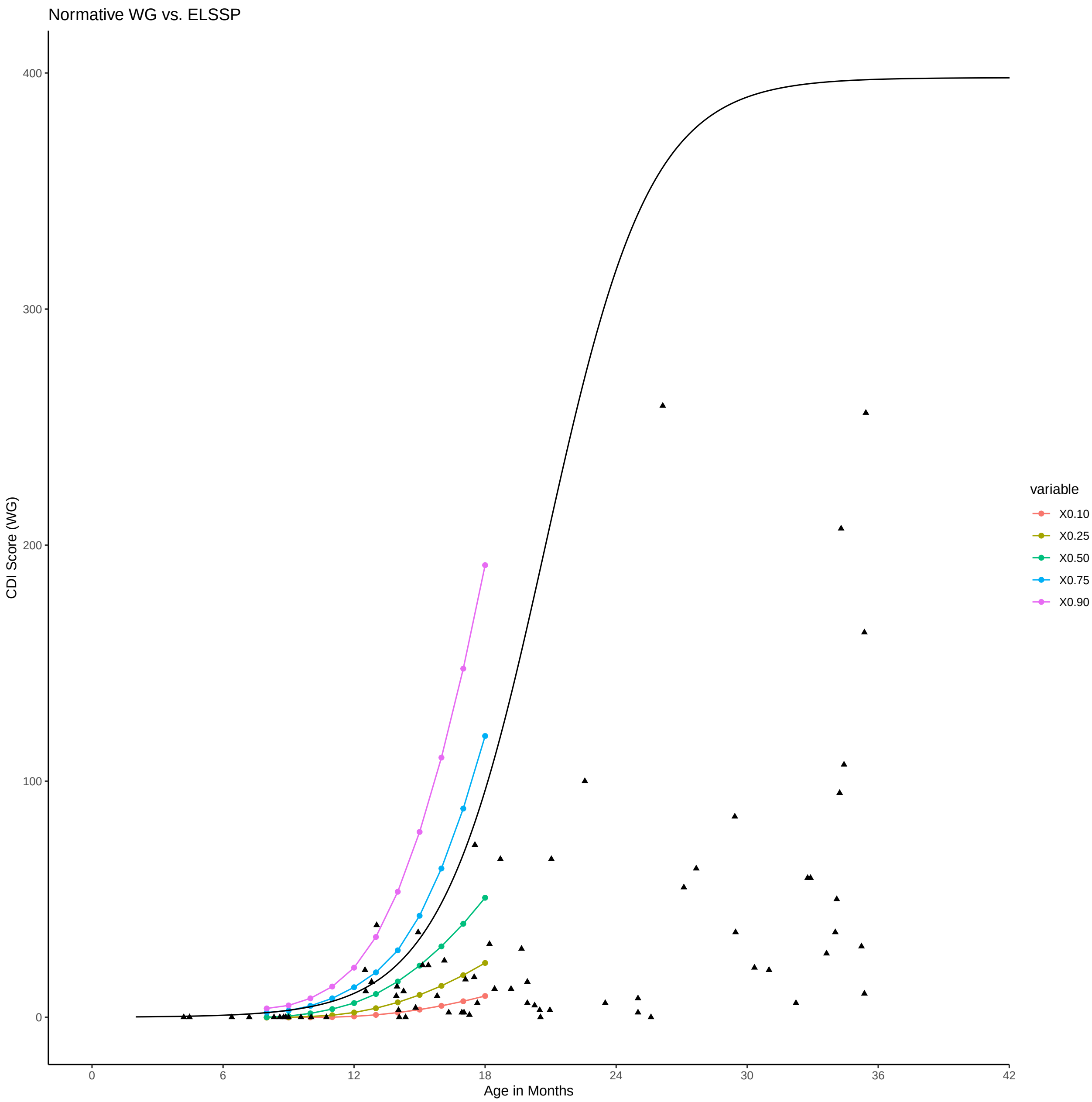
ELSSP


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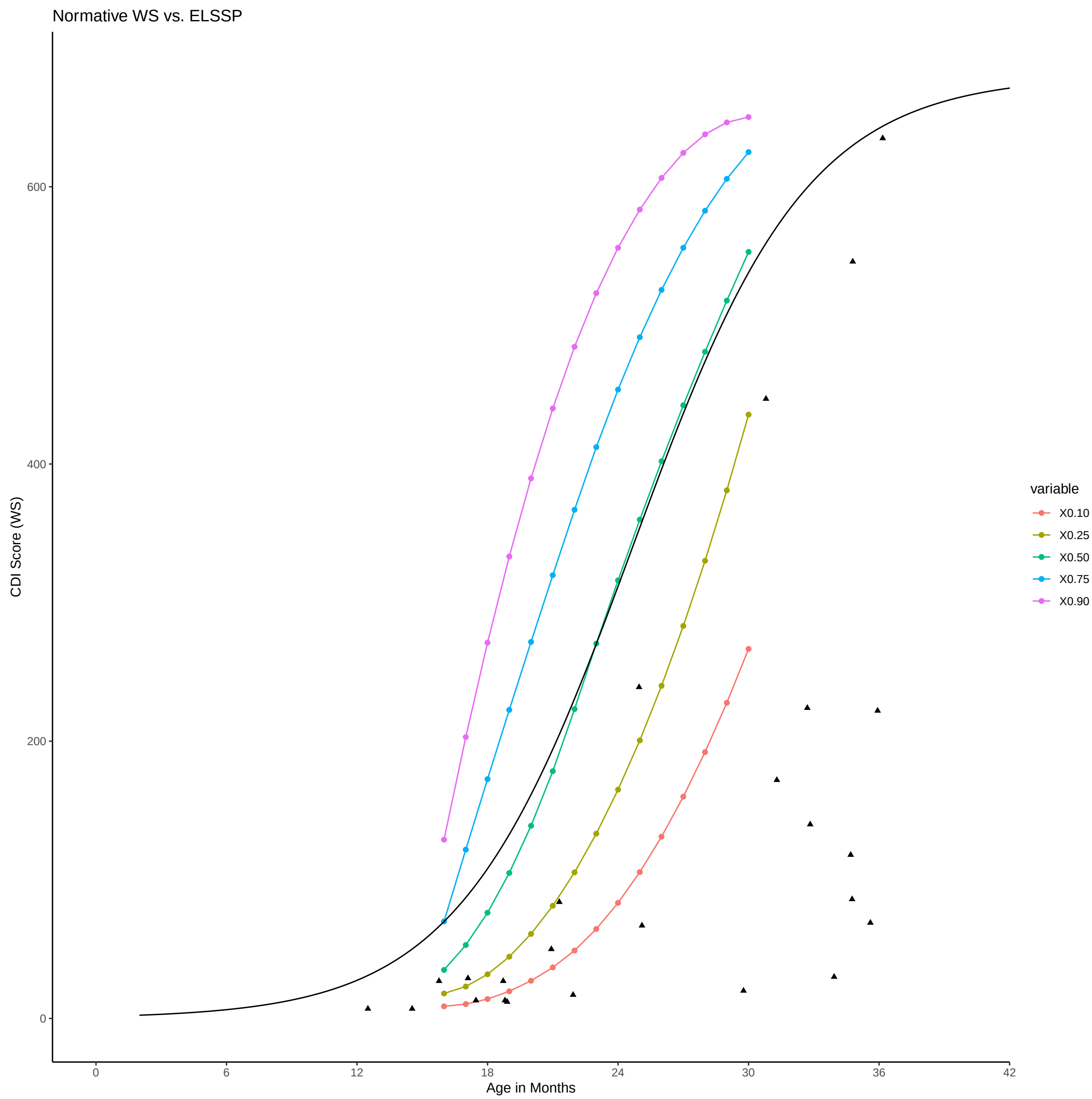
X0.25
X0.50
X0.75
X0.90


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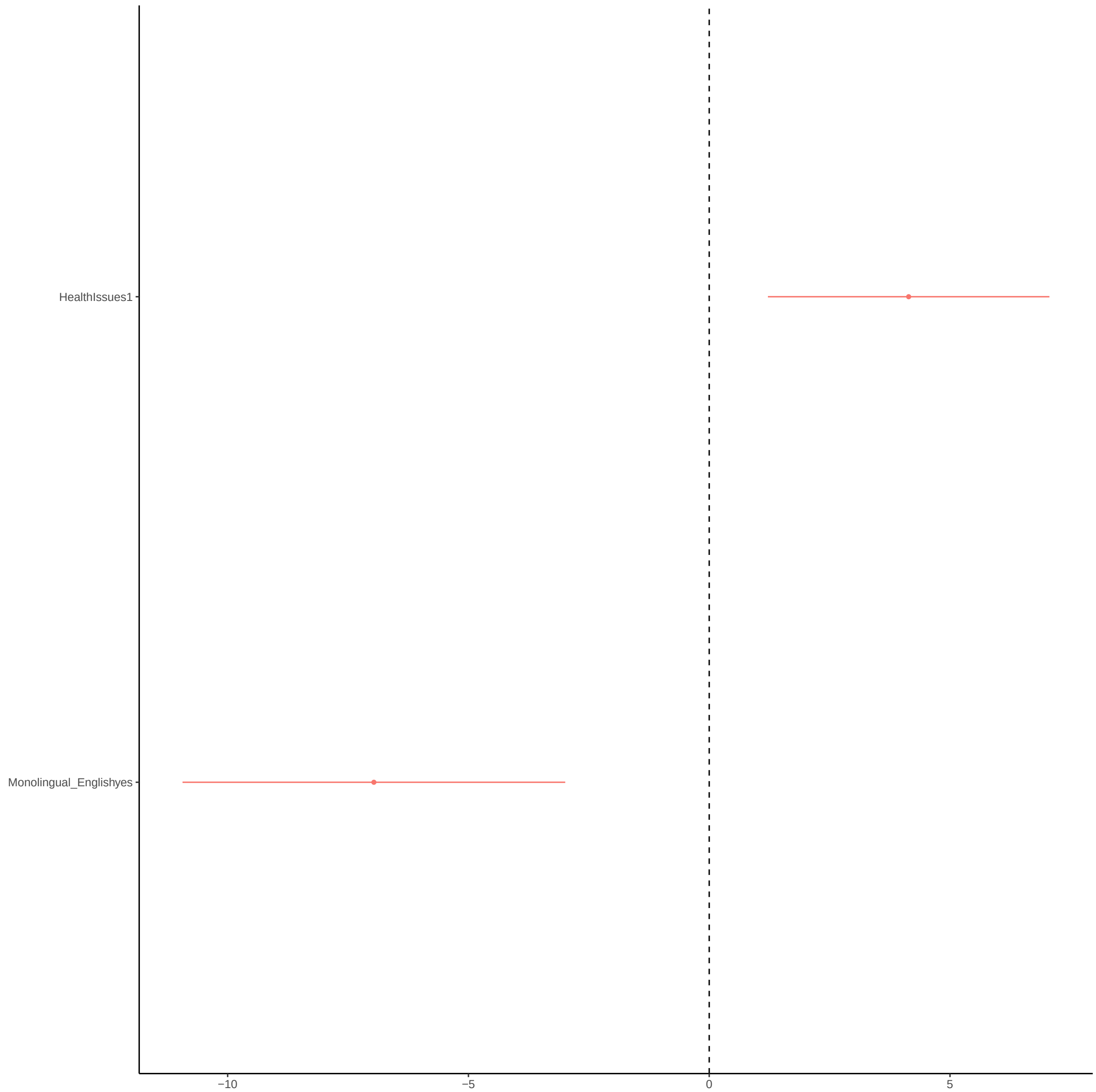
X0.25
X0.50
X0.75
X0.90



HealthIssues1
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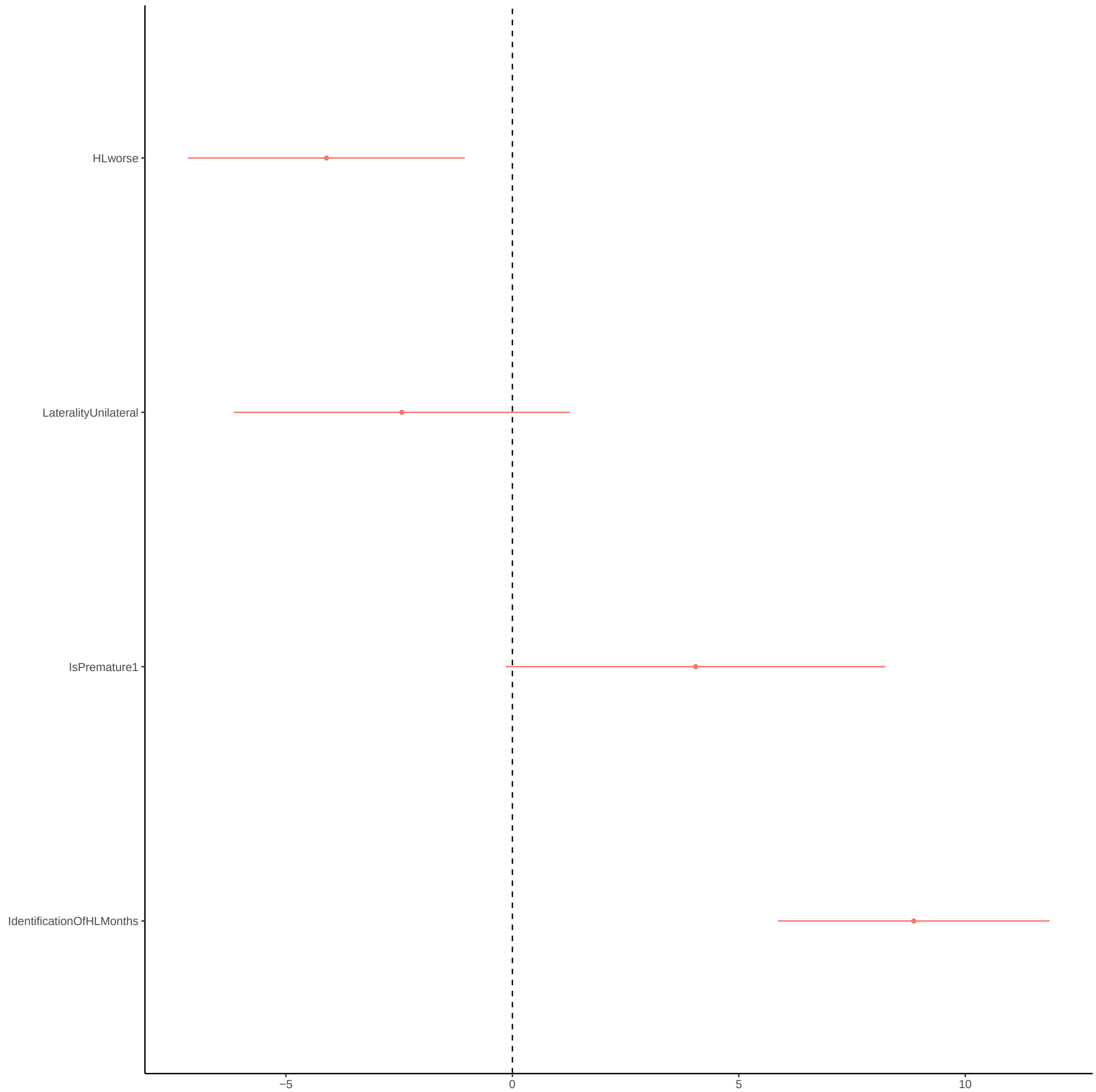
−5
0
5


HLworse
LateralityUnilateral
IsPremature1
IdentificationOfHLMonths
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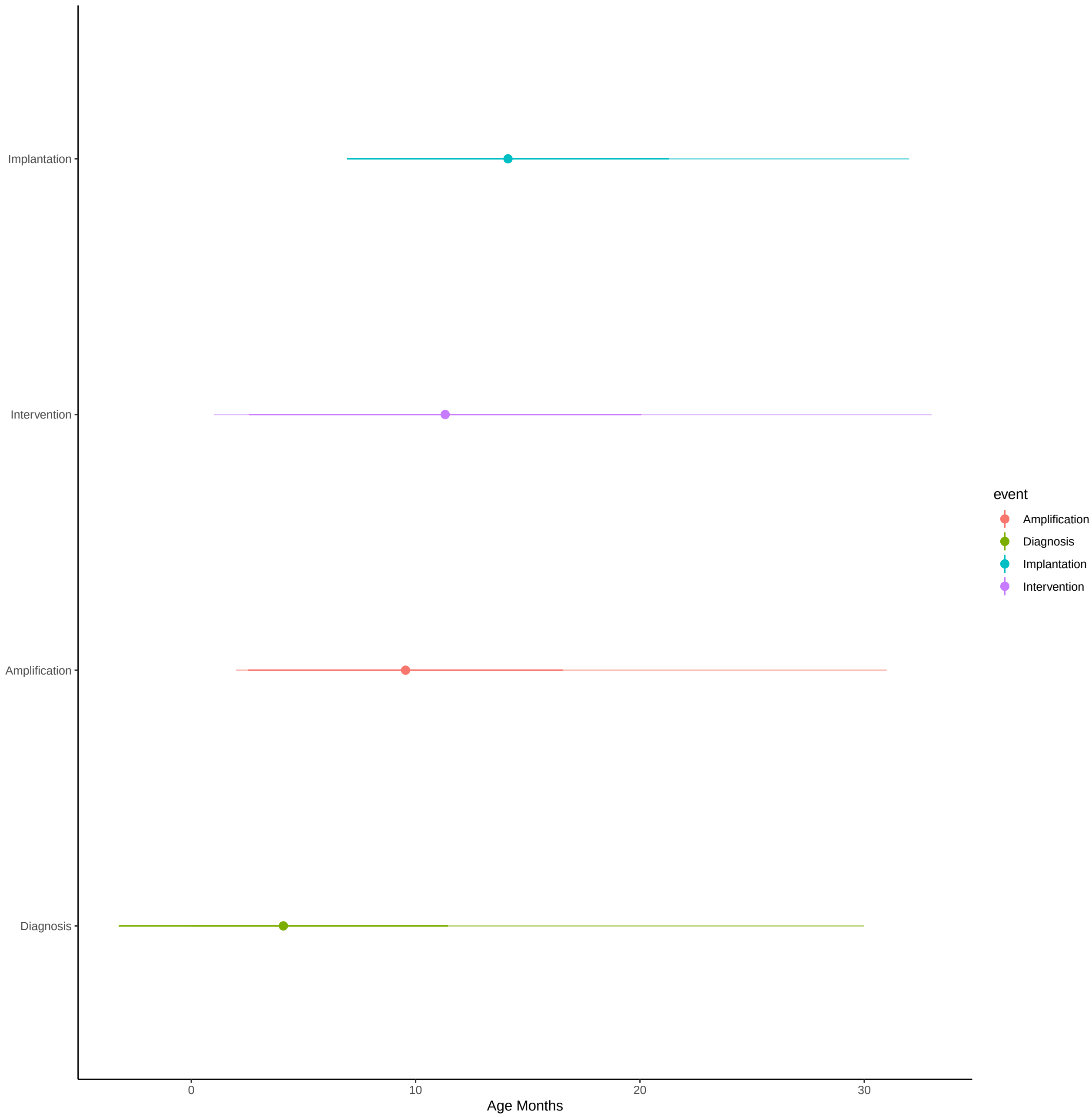


Introduction

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 et al. [2019]). Of these 114,000, ~90% will be born to hearing parents (Mitchell and Karchmer [2004/00/00]), 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 (Verhaert et al. [2008]; Geers et al. [2017]), but many will face persistent spoken language deficits (**?**; Moeller et al. [2007]; Luckner and Cooke [2010]; Sarchet et al. [2014]), which may later affect reading ability (**?**) and academic achievement (Qi and Mitchell [2012]; Karchmer and Mitchell [2003]).

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. Yoshinaga-Itano et al. [2018]; Vohr et al. [2008]). 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 et al. [2018]). Previous research indicates that gender (Kiese-Himmel and Ohlwein [2002]; Ching et al. [2013]), additional disability (Ching et al. [2013]; Yoshinaga-Itano et al. [2017]; Verhaert et al. [2008]), degree and configuration of hearing loss (Ching et al. [2013]; de Diego-Lázaro et al. [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]; Yoshinaga-Itano et al. [2018]) predict language outcomes 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 et al. [2004]; Fenson et al. [1994]; Frank et al. [2017]) and faster-growing vocabulary (**?**), and stronger grammatical and phonological skills (Lange et al. [2016]; Özçalışkan and Goldin-Meadow [2010]). This finding appears to be consistent across studies (Wallentin [2009]), various spoken languages (Frank et al. [2019]), and gesture (Özçalışkan and 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 (Kiese-Himmel and Ohlwein [2002]; Ching et al. [2013]). However, in contrast to their hearing peers, DHH children do not seem to show a gender-based difference for some aspects of syntactic development (**?**).

####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% (**?**; Guardino [2008]; Luckner and Carter [2001]; Picard [2004]; Schildroth and Hotto [1996]; Soukup and Feinstein [2007]; **?**; **?**), with approximately 8% children living with 2 or more co-occurring disabilities (Schildroth and 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]; **?**; Weismer et al. [2010]), with cognitive ability more predictive of language outcomes than presence or absence of a specific disability (Meinzen-Derr et al. [2011]; Sarant et al. [2008]). Disability and hearing loss likely each contribute to a given child's language development (Rajput et al. [2003], Van Nierop et al. [2016]; Ching et al. [2013]), 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 et al. [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 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 (**?**).

Independently of hearing status, prematurity is linked to increased risk of language delay and disorder (Rechia et al. [2016]; Van Noort-van Der Spek et al. [2012]; Carter and Msall [2017]; Vohr [2014]; Cusson [2003]; Barre et al. [2011]). 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 et al. [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 et al. [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]; Ching et al. [2013]; Tomblin et al. [2015]; Sarant et al. [2008]; **?**) and school-age (Wake et al. [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 (Delage and Tuller [2007]; Blair et al. [1985/00/00]). 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 et al. [2007]). The literature on hearing aids and cochlear implants points to benefits for bilateral auditory input (Sarant et al. [2014]; Lovett et al. [2010]; Smulders et al. [2016]). At school-age, 3–6% of children have unilateral hearing loss (Ross et al. [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 (Vila and Lieu [2015]; Lieu [2013]; Lieu [2004]; Lieu et al. [2012], Kiese-Himmel [2002]). 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 (Lieu [2013];Anne et al. [2017]). 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 (Walker et al. [2015]; Waltzman et al. [1997]; Niparko et al. [2010]). In turn, aided audibility predicts lexical abilities with children in HAs (Stiles et al. [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 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 (Dettman et al. [2007]; Miyamoto et al. [2008]; Svirsky et al. [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 (but not a full sign language, such as American Sign Language), sometimes simultaneously. Clinicians currently employ TC as an alternative or augmentative communication method for children with a wide range of disabilities (Branson and Demchak [2009]; Gibbs and Carswell [1991]; Mirenda [2003]).

Compared to total communication, DHH children using an exclusively oral approach have better speech intelligibility (Geers et al. [2017]; Dillon et al. [2004]; Geers et al. [2002]; Hodges et al. [1999]) and auditory perception (Geers et al. [2017]; **?**). 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 et al. [2017]).

####1-3-6 Guidelines Early identification (Yoshinaga-Itano et al. [2018]; Yoshinaga-Itano et al. [1998]; White and White [1987]; Robinshaw [1995]; Apuzzo and Yoshinaga-Itano [1995]; Kennedy et al. [2006]) and timely enrollment in early intervention programs (Vohr et al. [2008], Watkin et al. [2007]; Ching et al. [2013]; Holzinger et al. [2011]; Vohr et al. [2011]) 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]; Yoshinaga-Itano et al. [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, 200 [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), 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 et al. [2008]).

In evaluating the success of this initiative, the AAP (EHDI) 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 and Reilly [2002]; Frank et al. [2017]; Jackson-Maldonado et al. [2003]).

The CDI has also been validated for DHH children with cochlear implants (Thal et al. [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 et al. [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]).

##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 two subgroups of DHH children traditionally excluded from studies of language development: children with additional disabilities and children with unilateral hearing loss (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 communication method, language background, or presence of other health issues (e.g., congenital heart malformation).

##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=0), 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 98 children (55 male/43 female) ages 4.2–36.1666667(M=21.5992189, SD=9.007651). Race and SES information was 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 version of the CDI (Jackson-Maldonado et al. [2003]).

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 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; sensorineural, conditive, or mixed hearing loss. 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 sign language. Participants were also coded as monolingual or multilingual based on whether families reported using more than one language at home. Total communication was not counted as multilingualism.

Age at screening was measured as the child's age in months at their first hearing screening. Age at screening was only available for (XXX) participants. If participants received their newborn hearing screening, age at screening was recorded as 0 (months). 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 <= 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 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.

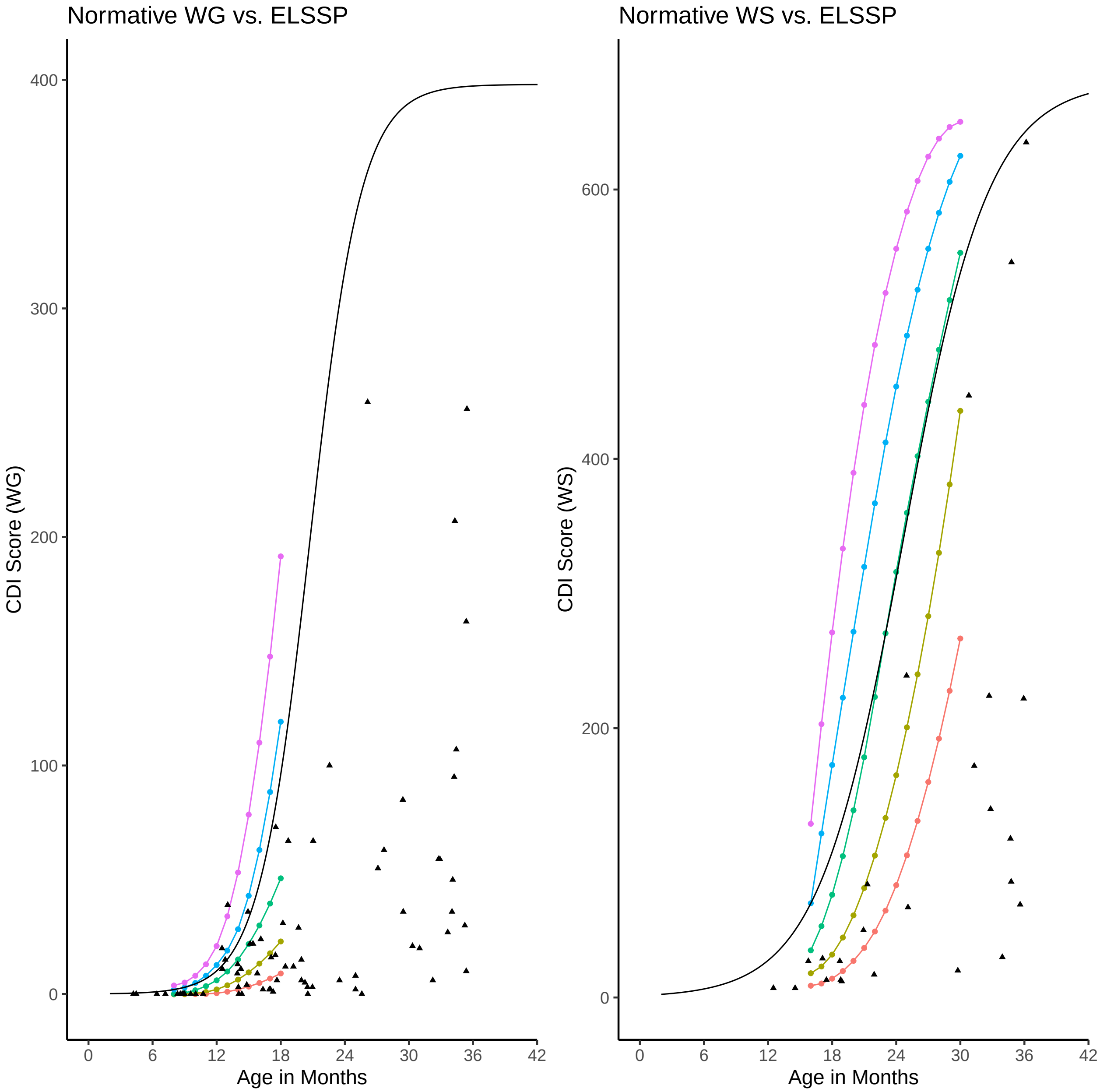
Part I: Structure of different variables with chi-square tests, correlation matrices, etc.

Shapiro–Wilk tests revealed that all of our continuous measures (i.e. degree of hearing loss, services received per month, vocabulary delay) significantly differed from a normal distribution (ps $<.05$), so we used nonparametric tests to explore relationships among our variables. For categorical-categorical relationships, we used chi square tests; for continuous-categorical tests, we used mann-whitney U tests (2 levels for categorical variable) or kruskal-wallis tests (>2 levels for categorical variable; for continuous-continuous relationships, we used Of the fifty-five combinations of variables, $p < .05$ for sixteen, and seven survived bonferroni correction ($p < 9.1 \times 10^{-4}$). The full set of comparisons is shown in figure XXX.

From this analysis, we found that children born premature were more likely to also have health issues (X2 (1, N = 63, 18, 1, 14) = 25.6888889, $p = 4.0112993 \times 10^{-7}$.) Children with conductive hearing loss were more likely to have unilateral hearing loss (X2 (2, N = 7, 8, 50, 10, 0, 11) = 14.8356311, $p = 6.0045939 \times 10^{-4}$). Children with unilateral hearing loss were unlikely to receive a cochlear implant and more likely to use no amplification (X2 (2, N = 17, 42, 12, 0, 11, 14) = 17.191416, $p = 1.8489767 \times 10^{-4}$). Children with more severe hearing loss were more likely to use a cochlear implant than children with milder hearing loss (H(2)=23.8032827, $p=6.7792686 \times 10^{-6}$). Children with developmental delays received more services per month than typically developing DHH children (H()=134.5, $p=4.9880474 \times 10^{-6}$)and were more likely to use total communication (X2 (2, N = 1, 0, 73, 6, 8, 8)) = 19.3795263, p = 19.3795263). Children who used total communication received more services per month than children using spoken language (H(1)=15.6025986, $p=7.8147116 \times 10^{-5}$).

Part II: Influence on vocabulary

We first constructed a binary logistic growth curve for vocabulary from the 50th percentile data for typically developing children from Wordbank. With this function, each participant's CDI score yielded a predicted age from the normative data. For each child, we subtracted this predicted age (given the score) from the child's actual age to give us a measure of delay in months. 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.

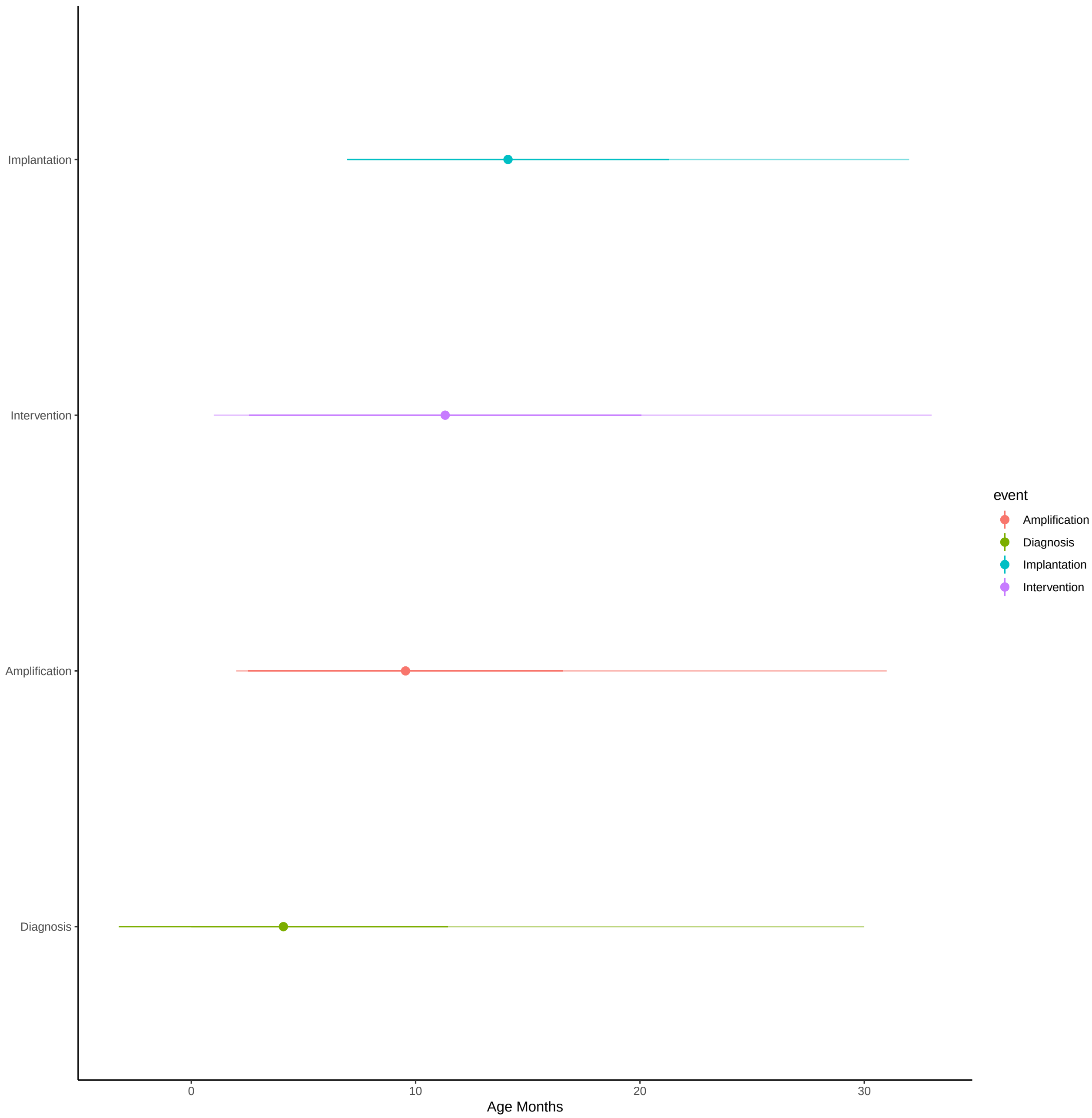


We next explored the effect of the different audiological, demographic, and intervention characteristics on vocabulary delay. Vocabulary delay did not meet the assumption of normality, so we used non-parametric tests for the following set of analyses. Mann-Whitney-Wilcoxen tests were conducted to examine the effects of gender, laterality, developmental delay, health issues, prematurity, meeting 1-3-6 guidelines, and communication on vocabulary delay. We used kruskal-wallis tests for amplification and etiology, and Kendall's rank correlations for degree of hearing loss (worse ear) and services received per month. These results are exploratory and descriptive, and their interpretation should be tempered accordingly.

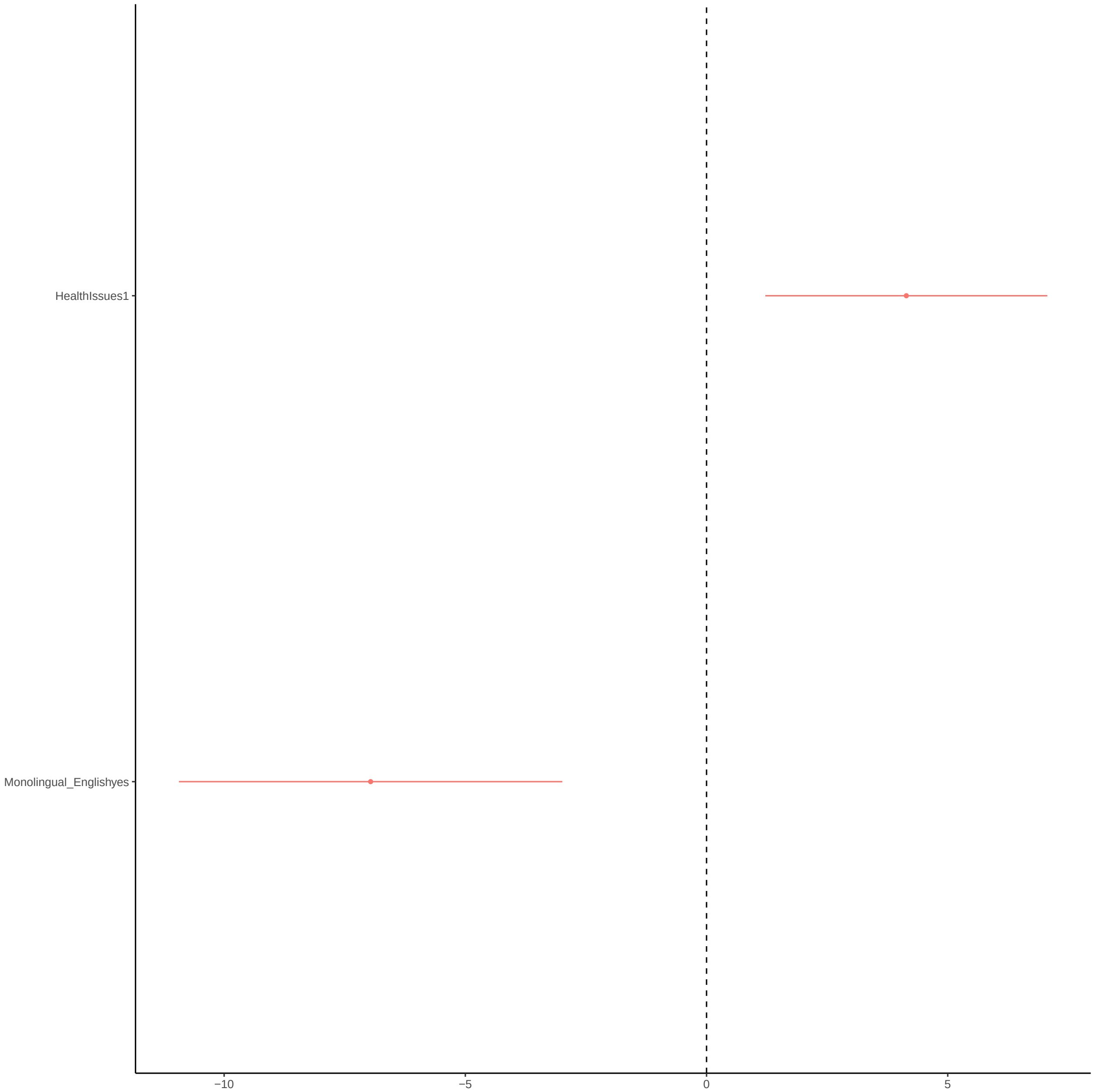
Boys were significantly more delayed than girls on Words and Sentences but not Words and Gestures. Children with developmental delays had larger vocabulary delays than children without developmental delays on Words and Gestures. Because only one child with a developmental delay took the Words and Sentences form, we did not perform the analysis for Words and Sentences. Premature children and children with health issues had smaller vocabularies than typically developing children on Words and Gestures but not Words and Sentences. Children who met 1-3-6 guidelines had larger vocabulary than children who did not on Words and Gestures but not Words and Sentences. On Words and Gestures but not Words and Sentences, receiving more early intervention services was correlated with lower vocabulary. We did not observe an effect of laterality, communication, degree, or etiology on vocabulary delay on either form of the CDI. For communication, we omitted cued speech from the analysis because only one child in our sample used this method of communication (shown on graph anyway for the curious). A kruskal-wallis test showed a significant effect of amplification on vocabulary delay on Words and Gestures, such that children with no amplification were more delayed than children without amplification.

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. 36.5591398% of our sample met 1-3-6 guidelines for early diagnosis and intervention. Of children with comorbidities (developmental concerns, prematurity, health issues), only `perc_136_DA`% met 1-3-6 guidelines, compared to `perc_136_TD`% of typically developing children. See table XXX for complete breakdown.

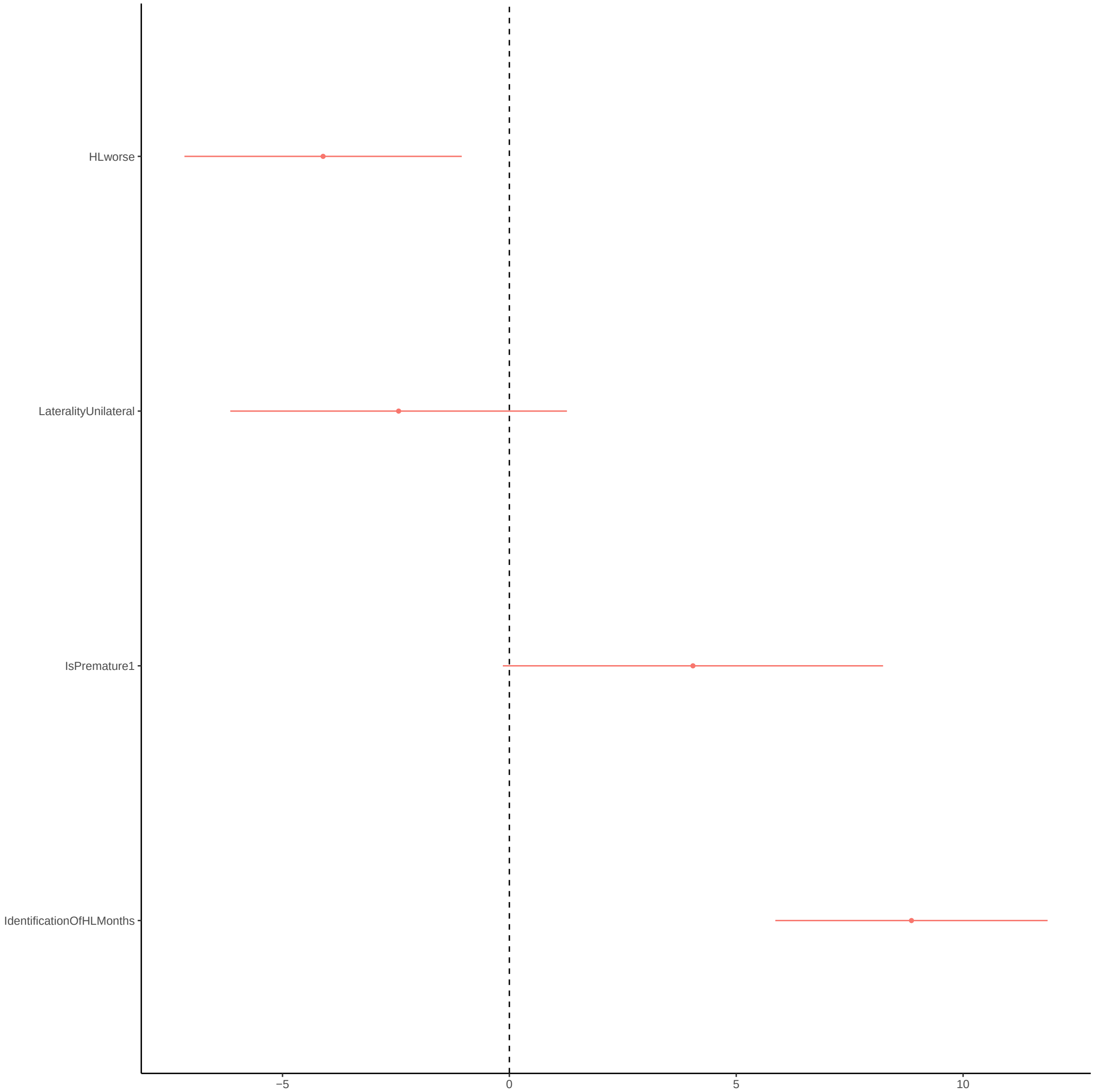


We created linear regression models for age at diagnosis and age at intervention. Models were paired down using stepwise regression by AIC using the stepAIC function (cite MASS package). For age at diagnosis, we included the set of child-specific factors that would be relevant before diagnosis of hearing loss. We began with: Age diagnosis ~ gender + laterality + degree (worse ear) + developmental delay + health issues + prematurity + laterality + language background + etiology The best fit model (R2=0.1887708 , p=8.1556828×10^{-5})included health issues (beta weight = 4.1415066, p = 0.0059947) and language background (beta weight = -6.9632948, p = 7.7174296×10^{-4}). Age diagnosis ~ health issues + language background



For age at intervention, we first included the variables potentially relevant prior to intervention: Age intervention ~ gender + degree (worse ear) + developmental delay + health issues + prematurity + laterality + language background + etiology + age diagnosis The best fit model (R2=0.4069216 , p=1.8631573×10^{-8}) included prematurity (beta weight = 4.0461265, p = 0.0582177), laterality (beta weight = -2.4409748, p = 0.1941197), degree of hearing loss (beta weight = 4.1415066, p = 0.009108), and age at diagnosis (beta weight = 0.6246784, p = 9.3856616×10^{-8}). Age intervention ~ laterality + degree (worse ear) + prematurity +

laterality + age diagnosis



Footnotes: Despite exciting, increasing, and converging evidence for benefits of early sign language exposure (e.g., Schick et al. [2007]; Spencer [1993]; **?**; Magnuson [2000]; Clark et al. [2016]; Hrastinski and Wilbur [2016]), 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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