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# Characterizing North Carolina's Deaf/Hard-of-Hearing Infants and Toddlers: Predictors of Vocabulary, Diagnosis, and Intervention

--Manuscript Draft--

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| **Abstract:** | Purpose : The study sought 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 factors influencing the success and timing of early identification and intervention efforts at a state level.<br>Method : One hundred Deaf/Hard-of-Hearing infants and toddlers (aged 4-36 months) enrolled in early intervention completed the MacArthur-Bates Communicative Development Inventory, and detailed information about their audiological and clinical history was collected. We examined the influence of demographic, clinical, and audiological factors on vocabulary outcomes and early intervention efforts.<br>Results : We found this sample showed spoken language vocabulary delays (production) relative to hearing peers, and room for improvement in rates of early diagnosis and intervention. These delays in vocabulary and early support services were predicted by an overlapping subset of hearing-, health-, and home-related variables.<br>Conclusions : In a diverse sample of Deaf/Hard-of-Hearing children receiving early intervention, we identify variables which predict delays in vocabulary and early support services, which reflected both dimensions that are immutable, and those that clinicians and caretakers can potentially alter. We provide a discussion on the implications for clinical practice. |
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# Characterizing North Carolina's Deaf/Hard-of-Hearing Infants and Toddlers: Predictors of Vocabulary, Diagnosis, and Intervention

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# Characterizing North Carolina's Deaf/Hard-of-Hearing Infants and Toddlers: Predictors of Vocabulary, Diagnosis, and Intervention


## Abstract

**Purpose**: The study sought 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 factors influencing the success and timing of early identification and intervention efforts at a state level.

**Method**: One hundred Deaf/Hard-of-Hearing infants and toddlers (aged 4-36 months) enrolled in early intervention completed the MacArthur-Bates Communicative Development Inventory, and detailed information about their audiological and clinical history was collected. We examined the influence of demographic, clinical, and audiological factors on vocabulary outcomes and early intervention efforts.

**Results**: We found this sample showed spoken language vocabulary delays (~~comprehension and~~ production) relative to hearing peers, and room for improvement in rates of early diagnosis and intervention. These delays in vocabulary and early support services were predicted by an overlapping subset of hearing-, health-, and home-related variables.

**Conclusions**: In a diverse sample of Deaf/Hard-of-Hearing children receiving early intervention, we identify variables which predict delays in vocabulary and early support services, which reflected *both* dimensions that are immutable, and those that clinicians and caretakers can potentially alter. We provide a discussion on the implications for clinical practice.

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## Introduction

In the United States, 1-2 children are born with hearing loss, per 1,000 births (CDC, 2018), of which ~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, whether the child uses amplification, and whether there is any access to sign language, linguistic input may be partially or totally inaccessible. Despite growing, converging evidence for benefits of early sign language exposure (e.g., Clark et al., 2016, Davidson et al., 2014; Hrastinski & Wilbur, 2016; Magnuson, 2000; Schick et al., 2007; Spencer, 1993), the majority of U.S. DHH children (and particularly those in our North Carolina-based sample) are not raised in a sign language environment. While some of these children will develop spoken language proficiency within the range of their hearing peers (Geers, Mitchell, Warner-Czyz, Wang, & Eisenberg, 2017; Verhaert, Willems, Van Kerschaver, & Desloovere, 2008), many will face persistent ~~spoken~~ language deficits (Eisenberg, 2007; Luckner & Cooke, 2010; Moeller, Tomblin, Yoshinaga-Itano, Connor, & Jerger, 2007), which may later affect reading ability and academic achievement (Karchmer & Mitchell, 2003; Qi & Mitchell, 2012). Given this, we focus primarily on spoken language development.

Though the literature points towards spoken language delays and deficits for Deaf or Hard-of-Hearing (DHH) children, this is a highly variable population with highly variable language outcomes (Pisoni, Kronenberger, Harris, & Moberly, 2018). For instance, 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; Yoshinaga-Itano, Sedey, Wiggin, & Mason, 2018) influence language outcomes in DHH children. Although many of these variables reflect immutable characteristics of the child, such as comorbid diagnoses or configuration of hearing loss, some represent opportunities for clinicians and policy makers to intervene and potentially improve language outcomes for DHH children.

More specifically, 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). These EHDI guidelines recommend that DHH children are screened by 1 month old, diagnosed by 3 months, and enter early intervention services by 6 months. We refer to this guideline as 1-3-6. Meeting this standard appears to improve spoken language outcomes for children with hearing loss, and the benefits appear consistent across a range of demographic characteristics (Yoshinaga-Itano et al., 2017, 2018), so it remains an important research goal to ~~to~~ identify children at risk of receiving clinical support late, in order to help all children achieve prompt diagnosis and intervention.

Notably, the variables linked to hearing loss mentioned above don't occur in a vacuum, yet past work has largely attempted to measure their effects as if they were independent. For

instance, many studies focus on vocabulary development in 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 et al., 2018), which are not representative of the broader population of DHH children. We take a different tack, asking instead how these factors co-occur and interact in the context of the broad diversity of the DHH community, how they are linked to early vocabulary, and how this connects with intervention and policy guidelines, within a single state in the U.S.

**Goals, Predictions, and Key Contributions**

We present an empirical analysis of early vocabulary in a wide range of young DHH children receiving state services in North Carolina. This study aims to 1) characterize the demographic, audiological, and intervention variability in the population of DHH children receiving state services for hearing loss; 2) identify predictors of vocabulary delays; and 3) evaluate the success of early identification and intervention efforts at a state level. We include three subgroups of DHH children traditionally excluded from studies of language development: children with additional disabilities, children with unilateral hearing loss, and children from bilingual or non-English-speaking households (e.g., Yoshinaga-Itano et al., 2018).

For the first goal, we expected that many of these variables would be linked, due to known causal relations (e.g., cochlear implants recommended for severe hearing loss, but not mild hearing loss). ~~This study contributes to the literature by quantifying the distribution and co-occurrence of demographic, audiological, and intervention characteristics in our broad sample, which includes many children often excluded from research.~~ For the second goal, we hypothesized that male (vs. female) gender, more severe degree of hearing loss, bilateral (vs. unilateral) hearing loss, no amplification (vs. hearing aids and/or cochlear implants),

premature birth, not meeting 1-3-6 guidelines, and presence of additional disabilities would predict larger spoken vocabulary delay. This study builds on prior work (e.g., Ching et al., 2013; Lund, 2016; Yoshinaga-Itano et al., 2017) by taking a new modeling approach for quantifying *vocabulary delay* across these variables. For the third goal, we hypothesized that children with less residual hearing (i.e., bilateral, more severe) and no co-occurring conditions would be earlier diagnosed and earlier to begin language services, and that in turn earlier diagnosis would predict earlier intervention. This study helps assess compliance with EHDI guidelines, and considers pathways for improvement.

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## 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 sent deidentified evaluations to our team after obtaining consent to do so from each family[1]. While this collaboration is ongoing, we opted to pause for this analysis upon receiving data from 100 children (collected between 2010 and 2020, before the COVID-19 epidemic reached North Carolina in Spring 2020). Given our goal of characterizing the full range of DHH children with hearing loss in North Carolina, no eligibility criteria beyond hearing loss and receiving an ELSSP evaluation were imposed.

[1] Because the data we received were already deidentified, this study was exempt from Duke University Institutional Review Board.

The clinical evaluations included demographic and audiological information and MacArthur Bates Communicative Development Inventory vocabulary scores (CDI, Fenson et al., 1994). For some children, evaluations from multiple timepoints or other instruments were available (e.g. PPVT). We limit the scope of the present study to only the CDI (as this was available for all children), and only the first evaluation (due to concerns regarding within-subjects variance for statistical analysis~~.~~)).

The CDI is a parent-report instrument measuring children's vocabulary. On the Words and Gestures version of the form (normed for 8–18-month-olds), parents indicate whether their child understands and/or produces each of the 398 vocabulary items. ~~One~~On the Words and Sentences version (normed for 16–30-month-olds), parents indicate whether their child produces each of the 680 vocabulary items. Normative data for this instrument (Frank, Braginsky, Yurovsky, & Marchman, 2017; Jackson-Maldonado et al., 2003) is available from WordBank, an open database of CDI data. The CDI has also been validated for DHH children with cochlear implants (Thal, Desjardin, & Eisenberg, 2007) in 32–66-month-olds. We build on prior literature using the CDI to measure vocabulary in DHH children (e.g., Yoshinaga-Itano et al., 2017, 2018; de Diego-Lázaro et al., 2018; Vohr et al., 2008, 2011) with a new analytic approach below.

For this analysis, 100 children (56 male / 44 female) ages 4.10–35.70 months (M=21.20, SD=9.10) contributed data. Race and socioeconomic information were not available. Families were administered either the Words and Gestures or Words and Sentences version of the CDI based on clinician judgment of linguistic ability. Children who were too old for Words and Gestures, but who were not producing many words at the time of assessment, were often given Words and Gestures (n = 37). Families whose primary language was Spanish (n = 15) completed the Spanish language version of the CDI (Jackson-Maldonado et al., 2003). Both spoken words

and signs counted as word productions. ~~See Table 1 for additional CDI information for our sample.~~ A summary of all the variables we examined is available in Table 1~~2~~, and more detailed information can be found in the Supplemental Materials, Tables S1-S3.

## Results

The results are organized mirroring the goals outlined above. First, we explore relationships among child demographic, audiological, and clinical variables. Second, we use these variables to predict vocabulary development. Finally, we describe the implementation of the EHDI 1-3-6 guidelines and predictors of early diagnosis and intervention in this sample. All analyses were conducted in R (R Core Team, 2020) and all code to generate this manuscript in Rstudio (RStudio Team, 2020) is available via OSF.

### Relationships Among Demographic, Audiological, and Clinical Variables

Before testing how these variables relate to vocabulary and clinical milestones, we describe their relationships to each other. To quantify this statistically, we used Bonferroni-corrected chi-square tests between each of our variables. Because the chi-square statistic assumes n>5 is *expected* in the majority of the cells for each test (preferably ≥80%, McHugh, 2013), we excluded mixed hearing loss (n = 8) and cued speech (n = 1) from this analysis. Strictly speaking, some variables are not expected to be randomly distributed relative to each other (e.g., premature birth and health issues; degree and amplification), but quantifying the differences via chi-square using a conservative significance threshold lets us highlight the strongest relationships within this dataset.

Of the 66 combinations of variables, $p < .05$ for 26, and 9 survived Bonferroni correction ($p < 0.0007$). We limit discussion to the latter below, but depict the full set in Figure 1.

As expected, health issues, developmental delays, and premature birth 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 < .0001) and developmental delays ($X^2$ (1, N = 98) = 13.06, p = .0003), and children with developmental delays were more likely to also experience health issues ($X^2$ (1, N = 98) = 18.67, p < .0001). Children with developmental delays received more services per month than typically-developing children ($X^2$ (2, N = 95) = 23.99, p < .0001) and were more likely to use total communication ($X^2$ (2, N = 98) = 24.88, p < .0001). Likewise, children who used total communication received more services per month than children using spoken language ($X^2$ (4, N = 95) = 21.53, p = .0002).

We also confirmed expected relationships among many of the audiological characteristics. There was a significant relationship between laterality and etiology ($X^2$ (2, N = 89) = 18.72, p = .0001), such that children with conductive hearing loss were more likely to have unilateral hearing loss, and children with sensorineural hearing loss were more likely to have a bilateral loss. All children with mixed hearing loss (n = 8), though excluded from statistical analysis due to low N, had bilateral hearing loss. The chi-square tests further showed that amplification was related to laterality ($X^2$ (2, N = 98) = 17.55, p = .0002) and degree of hearing loss ($X^2$ (4, N = 88) = 28.76, p < .0001). Specifically, 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 of hearing loss, children with severe-to-profound

hearing loss were more likely to use a cochlear implant than children with mild or moderate hearing loss.

Taken together, the results in this set of analyses highlight the notable interconnectedness among early health and development (i.e. health issues, prematurity, and developmental delays), and audiological characteristics (i.e. links among laterality, etiology, amplification, and degree of hearing loss).

**Predictors of Vocabulary Delay**

We next turn to the relationship between these variables and children's productive vocabulary, as measured by the CDI. Figure 2 shows the vocabulary scores of children in our ~~samples~~sample relative to norms for hearing children for each CDI form. Descriptively, we found widespread vocabulary delays, with the majority of DHH children testing around or below the 25th percentile for hearing children (based on WordBank norms; Frank et al., 2017).

As noted above, the two CDI forms differ in how many vocabulary items they contain. To take this into account, we establish the difference (in months) between the child's chronological age and their predicted age based on their productive vocabulary, derived from the WordBank norms (Frank et al., 2017), rather than using the raw vocabulary scores. We call this derived variable *vocabulary delay*.

More specifically, to compute a child's predicted age from their vocabulary score, we used the 50th percentile for productive vocabulary from WordBank data for typically-developing

infants (Frank et al., 2017) to create binary logistic growth curves separately for the "Words and Gestures" (WG) and "Words and Sentences" (WS) versions of the CDI for American English and Mexican Spanish[2]. For each child, we took the number of words they produced (spoken and/or signed, though the latter was only provided for children using Total Communication (n = 18) as all others were reported to exclusively use spoken language). We then divided this production score by the number of words on the instrument, to give us the proportion of words produced. We used this proportion in an inverse prediction from the binary logistic regression curves to generate a predicted age. That is, for each possible CDI score, the growth curve provided the age that the score would be achieved for the 50th percentile trajectory. Finally, we subtracted the predicted age from each child's chronological age to calculate their vocabulary delay. However, for children producing 0 words, this approach was not appropriate due to the long tails on the growth curves. Thus, for this subset of children, we took the x-intercept from Wordbank (8 months for English, and 9 months for Spanish), and subtracted that value from the child's chronological age to get their vocabulary delay.

To look at the relationship between our predictor variables and CDI scores, we next conducted multiple linear regression, using vocabulary delay as our outcome variable. Children who were too young for the CDI version they were administered (n = 7) were excluded from this portion of the analysis, as was the adopted child due to concerns about comparing their score to the American English CDI norms.

[2] Number of hearing children in normative sample for each growth curve: WG-English=1071, WG-Spanish=760; WS-English=1461, WS-Spanish=1092

Our full regression model included all variables: Vocabulary Delay ~ Gender + Developmental Delay + Health Issues + Premature Birth + Laterality + Degree + Amplification + Communication + Meets 1-3-6 + Services Received Per Month + Language Background.

This model accounted for significant variance in vocabulary delay (adjusted-$R^2 = 0.59$, $p < .001$). We next performed stepwise model comparison using stepAIC (MASS) to pare down the model. This process selects only the predictors which incrementally improved model fit, measured by Akaike's Information Criterion (AIC). We started model selection with the full model, as described above. We then filtered out data from children for whom Meets 1-3-6 (n = 5) or Degree (n = 12) was unknown, as this stepwise AIC approach does not permit missing values across predictors. Since this initial filtered analysis found that Degree and 1-3-6 did not improve model fit, we manually removed the Degree and 1-3-6 terms from the model selection so that the 14 participants with missing cases for these variables could be retained[3].

Based on this iterative process, we arrived at the following final model: Vocabulary Delay ~ Age + Laterality + Amplification. No other variables from the full model above significantly improved model fit, and are thus not discussed further. Our final model accounted for significant

[3] 3 participants had missing values for both 1-3-6 and Degree. For transparency, we note that the model fitted with only complete cases of Degree did include a non-significant main effect of Developmental Delay. However, ANOVA revealed that including a Developmental Delay term did not significantly improve model fit when including the 14 participants without Degree information.

variance in children's vocabulary delay to a nearly identical degree as the full model (adjusted-$R^2$ = 0.58, $p$ = < .001, see Table S4 & Figure 5.A). We found significant main effects for Age, Laterality, and Amplification, such that older age, bilateral hearing loss, and no amplification predicted greater vocabulary delays. Compared to children with no amplification, children with cochlear implants had a 3.58 months smaller spoken vocabulary delay ($p$ = .019), and similarly children with hearing aids had a 3.89 months smaller delay ($p$ = .001). Children with unilateral hearing loss had a 3.03 months smaller delay ($p$ = .009) than children with bilateral hearing loss. For Age, the model predicted a 0.55 months *larger* vocabulary delay ($p$ < .001) for each additional month of age.

Given our first set of results regarding relationships among several of these variables (e.g., laterality and amplification), we tested for collinearity by computing the model's VIF (variance inflation factor). This revealed low levels of collinearity among predictors in our final model (all VIF < 1.23; James, Witten, Hastie, & Tibshirani, 2013). In sum, the analyses in this section revealed that over half of the variance in DHH children's vocabulary scores was explained by their age, whether they receive amplification, and whether their hearing loss was unilateral or bilateral.

**Success in Meeting 1-3-6 Guidelines**

Perhaps of greatest importance to clinicians and policymakers is the implementation and effect of existing policies. Although ~~whether a child met~~ 1-3-6 ~~guidelines~~status was not included in our final model predicting vocabulary delay through our model selection process, its demonstrated importance for language outcomes (e.g., Yoshinaga-Itano et al., 2018) merits further discussion. To this end, we provide a brief description of the implementation of 1-3-6 in

our sample, examine its effect on vocabulary delay, and describe the results of exploratory linear regression models for age at diagnosis and age at intervention.

Overall, 36% of our sample met 1-3-6 guidelines for early diagnosis and intervention. Breaking this down further, among the children for whom screening information was available (n = 68), 100% were screened at birth or during NICU stay. In our sample, 69% of children received diagnosis by 3 months of age, and 38% began early intervention by 6 months of age (see Figure 3).

We first tested the link between 1-3-6 and vocabulary directly. An independent samples t-test showed that children who did not meet 1-3-6 guidelines had significantly larger vocabulary delays than children who met 1-3-6 guidelines ($t(66.29) = 2.66$, $p = 0.01$; see Figure 4). On average, the group that did not meet 1-3-6 guidelines was 3.71 months more delayed with regard to vocabulary (relative to the same 50th percentile benchmark described above).

To better understand implementation of 1-3-6 guidelines, we next turned our focus to factors influencing the timing of diagnosis and intervention. We conducted two linear regressions, one for predicting age at diagnosis and one for age at intervention. Model selection followed the same stepwise AIC-based process as described in the preceding section.

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 children did not receive amplification prior to hearing loss diagnosis.) We began with: gender, degree, developmental delay, health issues, prematurity, laterality, language background, and etiology.

The best fitting model was: Age at Diagnosis ~ Health Issues + Language Background + Laterality, with significant main effects of Health Issues and Language Background (see Table S5 & Figure 5.B). This model accounted for 15.34% of the variance in age at diagnosis ($p$ = .002). Average age at diagnosis was 4.60(7.19) months. Relative to English-speaking families, children from Spanish-speaking families were diagnosed 6.18 months later ($p$ = .002). Children with health issues were diagnosed 3.65 months later than children without health issues ($p$ = .01). One possibility for this last predictor is that the health issues caused hearing loss *later* in infancy; in our sample, 16 of the 36 children with health issues reported conditions that can in some cases cause acquired hearing loss (i.e., meningitis, sepsis, jaundice, seizures, hydrocephalus, MRSA, anemia, frequent fevers, cytomegalovirus).

We repeated this model selection process for age at intervention. In addition to the variables used to fit the ~~intervention~~diagnosis model, we included age at diagnosis. The best fit model was: Age at Intervention ~ Premature Birth + Degree + Age at Diagnosis + Language Background ($R^2$=0.43 , $p$ < .001; See Table S6 & Figure 5.C), with significant main effects of degree and age at diagnosis. Prematurity (ß = 3.79, p = .06) and language background (ß = -1.37, p = .51) were not significant predictors on their own, but their inclusion improved model fit. Average age at intervention was 11.29(8.63) months. More severe hearing loss predicted earlier intervention, such that for every additional 10 dB HL, predicted age at intervention was 1 month earlier ($p$ < .01). With regard to age at diagnosis, for every month diagnosis was delayed, intervention was delayed by 2.80 weeks ($p$ < .01). Taken together, these analyses reveal that children's audiological characteristics, comorbid diagnoses, and language background contribute to delays in both diagnoses and intervention. We return to this point in the discussion.

## Discussion

In this study, we examined the demographic, audiological, and clinical characteristics of 100 young DHH children in North Carolina. We documented the distribution of these characteristics and explored the relationships between these variables, vocabulary, diagnosis, and intervention. In prior work with tightly controlled samples, the variables studied here have been shown to be relevant for language development, but their effects have rarely been examined in the full heterogeneity they naturally occur within. We took this big-tent approach by including any children receiving services for hearing loss.

Returning to our original three questions, we asked first: how are child-level variables intertwined? We found significant structure across many of the variables, suggesting that in a real-world sample of children with hearing loss, many factors are intrinsically not dissociable. This was particularly true for many of the auditory characteristics and comorbid diagnoses. To our knowledge, this paper provides the first population-based documentation of this distribution. We next asked whether these characteristics can predict vocabulary outcomes for DHH children. We found that a model including only children's age, laterality of hearing loss, and amplification type best accounted for the variability in vocabulary outcomes. Finally, we asked how successful the 1-3-6 guidelines were for early detection and intervention, both in terms of improving child outcomes and ensuring timely diagnosis and intervention. Here, we found that children who met 1-3-6 guidelines indeed had a smaller vocabulary delay than those who didn't. However, only 36% of children met these guidelines. Our results highlight family- and health-related variables that accounted for significant variability in when children received diagnosis and/or intervention.

We believe the inherent complexity in these results is an important piece of understanding vocabulary outcomes within the diverse population of DHH children. We next highlight some implications of this study for future research and clinical practice.

**How are child-level variables intertwined?**

This study contributes to the literature by quantifying the distribution and co-occurrence of demographic, audiological, and intervention characteristics in our broad sample, which includes many children often excluded from research. In our sample, we found significant overlap among demographic, audiological, and clinical variables. To highlight a few of these findings, prematurity, health issues, and developmental delay frequently co-occurred, such that children with one of these factors were more likely to have the others, consistent with prior research (Luu, Katz, Leeson, Thébaud, & Nuyt, 2016; Pierrat et al., 2017). Given that the constellation of comorbid conditions is so varied (76 unique conditions in our sample of 100 children; see Table S1), an important direction for future research is whether cognitive and social abilities, as well as ~~family's~~families' treatment resources, are predictive of language outcomes across conditions.

We also found that children with developmental delays (e.g., Down syndrome) were much more likely to use a total communication approach than DHH children without developmental delays (i.e., total communication used by 62.50% of DHH children with developmental delay vs. 9.76% of those without). That is, use of total communication was more likely for children already at greater risk for verbal delays. Quantifying this confound is an important contribution of this work, as it calls for tempering the interpretation of correlational studies finding links between total communication and language delays (e.g., Geers et al., 2017).

The relationships we found among variables were more confirmatory than surprising, particularly those reflecting known causal links (e.g., increased health issues in children born premature). Nevertheless, they should caution us to think critically about how we construct samples for controlled lab experiments. ~~If a researcher desires to collect~~For example, if an eyetracking experiment has a sample of ~~(e.g.,)~~ typically-developing pediatric cochlear implant users with bilateral, severe-to-profound hearing loss, ~~how representative would the results be, given that~~ such a subsample may only represent roughly 14% of the DHH population~~, as it does here?~~. Such considerations are important for properly representing and supporting DHH children and their families. This becomes doubly important in the context of interpreting language outcomes like vocabulary.

**Predicting vocabulary outcomes**

In our sample, 87.78% of DHH children fell below the 50th percentile for vocabulary, indicating that a large majority of this sample is behind a normative sample of their hearing peers in word learning. This disadvantage can have lasting consequences in the lives of DHH children (Karchmer & Mitchell, 2003; Qi & Mitchell, 2012), highlighting the importance of understanding what factors contribute to it.

In contrast to our predictions, the best model predicting vocabulary delay had just a few variables: age, amplification, and laterality. We did not find that gender, developmental delay, health issues, premature birth, degree of hearing loss, communication modality, 1-3-6 status, number of services per month, or language background significantly improved model fit. Notably, we see that the spoken vocabulary delay widens with age, indicating that the *rate* of spoken vocabulary acquisition is slower for DHH children. Given that none of the children here

use sign language (which can ensure earlier language access), this vocabulary delay is likely to have knock-on effects for language development more broadly, alongside implications for public policy.

## Predicting early diagnosis and intervention

Our exploration of the implementation of 1-3-6 guidelines revealed that only 35.79% of children met the EHDI guidance for diagnosis by 3 months and intervention by 6 months. Our results were consistent with prior work (e.g., Yoshinaga-Itano et al., 1998; Ching et al., 2013), finding that children who met the guidelines were 3.71 months *less* delayed in spoken vocabulary than children who were late to receive diagnosis and/or services. By dint of accepting all children receiving early intervention services in one state, our dataset let us delve deeper into *who* received on-time diagnosis and intervention.

**Diagnosis.** Having health issues or a non-English language background predicted later diagnosis. Children with health issues were diagnosed 3.65 months later than infants without health issues. For a small fraction of cases, this may have been because health issues caused acquired hearing loss, delaying its identification[4]. Of course, some situations may require families and medical providers to prioritize treatment for certain health issues (e.g., surgery for congenital heart defect) over diagnostic audiology services. That said, our results raise the possibility that

[4] In our sample, 16 of the 35 children with health issues had conditions that might cause acquired hearing loss (i.e., meningitis, sepsis, jaundice, seizures, hydrocephalus, MRSA, anemia, frequent fevers, cytomegalovirus).

clinician awareness of increased delays in language linked to the prevalence of health issues may facilitate improvements in timely diagnosis.

Language background too predicted age at diagnosis, such that infants from Spanish-speaking families were diagnosed 3.79 months later than infants from English-speaking families. This may be due to cultural differences in attitudes towards deafness (Caballero, Muñoz, Schultz, Graham, & Meibos, 2018; Rodriguez & Allen, 2020; Steinberg, Bain, Li, Delgado, & Ruperto, 2003) or a lack of linguistically accessible and culturally appropriate audiology services. Only 5.6% of American audiologists identify as bilingual service providers (ASHA, 2019), and services from a monolingual provider may be insufficient, particularly in obtaining the child's case history and providing recommendations for follow-up services (Abreu, Adriatico, & DePierro, 2011).

**Intervention.** As expected, more severe hearing loss predicted earlier intervention. This may be due to parents and clinicians adopting a wait-and-see approach to intervention for children with some residual hearing, despite associations between mild-to-moderate hearing loss, and language delays and academic challenges (Blair, Peterson, & Viehweg, 1985; Delage & Tuller, 2007). Early intervention may help offset these associations.

Age at start of services was also associated with age at diagnosis: for each month diagnosis was delayed, intervention was delayed by 2.80 weeks. Ching et al. (2013) found that age at intervention predicted better outcomes for DHH children, above and beyond age at diagnosis. Of course, these two variables are related, underscoring the importance of early diagnosis for putting children in the pipeline towards earlier intervention.

Finally, it's important to note that this sample is composed of children receiving birth-to-3 services. Less than 38% of our sample of children in early intervention meet the 6-month EHDI benchmark. Given that only about 67% of children with hearing loss enroll in early intervention services (CDC, 2018), our data suggest that the actual proportion of DHH children who receive intervention by the EHDI-recommended 6 months may be closer to 25%. These children may not receive clinical support until school-age or later, exacerbating concerns for language development, which lays an important foundation for literacy and academic success (Hemphill & Tivnan, 2008; Stæhr, 2008).

**Educational and Clinical Implications**

Despite high rates of newborn hearing screening in North Carolina, and even relatively high rates of diagnosis by 3 months (66/100 children in our sample), most children did not meet the 1-3-6 guidelines. Based on our analyses, we have the following recommendations for increasing attainment of 1-3-6 guidelines:

1. Frequent hearing screenings for children receiving medical or therapeutic care for health issues.
2. Service coordination for families balancing multiple co-occurring conditions.
3. Expansion of bilingual clinicians both in-person and for teletherapy ~~clinicians~~ to provide therapy and service coordination to non-English-speaking families.
4. Provision and encouragement of early intervention services for children with mild to moderate hearing loss.

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Additionally, the vast majority of children in our sample experienced vocabulary delays (relative to hearing peers), and studies of spoken vocabulary development in older DHH children suggest that they may not catch up (Lund, 2016). This should set clinicians and educators on high alert. As early intervention predicts vocabulary outcomes ~~in study after study~~across multiple studies (including this present study and e.g., Vohr et al., 2008; Ching, Dillon, Leigh, & Cupples, 2018), ensuring intervention by 6 months for all DHH children may be one way to address spoken vocabulary deficits. Another option may be the provision of structured, accessible language input (i.e., sign language) even prior to intervention or amplification, potentially mitigating negative effects of auditory deprivation on language skills (Davidson, Lillo-Martin, & Pichler, 2014; Hassanzadeh, 2012). While learning sign language may pose a challenge for some families for myriad reasons (as underscored by its absence as a communication modality within our sample), we nevertheless highlight its potential as an important language support for DHH children and their families.

## Limitations and Opportunities for Future Work

This study represents an important first step in quantifying variability in demographic characteristics, language outcomes, and 1-3-6 attainment. At the same time, it is exploratory, has limited geographic scope, and analyzed data from a (deliberately) high-variability sample.

Given our exploratory analyses, there were many possible analytic routes. We encourage interested readers to explore further analyses using the data and/or code provided on our OSF page.

This sample is composed only of children in North Carolina. While certain factors vary by country and by state (e.g., diagnosis and early intervention practices; NAD, n.d.), our sample

largely resembles the national DHH population in terms of degree of hearing loss, percentage of children with additional disabilities, cochlear implant and hearing aid use, language background, and gender (Blackorby & Knokey, 2006; Gallaudet Research Institute, 2014). It did diverge from the national sample in communication modality: our sample had no signers while 20% of DHH children have sign as their primary modality (Gallaudet Research Institute, 2014). A similar naturalistic study in regions where sign language access for DHH children is more common (e.g. Washington D.C.) would be a welcome addition to the present work, in illuminating the effects of different clinical and demographic factors in a signing population. One further limitation to our analyses and to assessing representativeness of the sample is that race and socioeconomic status information was not available.

Finally, the considerable variability in the sample did not allow us to easily isolate effects of different factors (e.g., degree vs. amplification). This reflects real-world variability and would be best addressed by larger sample sizes. As researchers continue to study influences on vocabulary in DHH children, a meta-analytic approach too may be able to better estimate effect sizes within the varied outcomes of this heterogeneous population.

## Conclusion

The present study explored interrelations among demographic and audiological characteristics, vocabulary outcomes, and clinical milestones within a diverse sample of 100 DHH children enrolled in early intervention services in North Carolina. Our population-based description underscores heavily interlocking demographic, audiological, and clinical characteristics (e.g. communication approach and presence of developmental delays). Our models highlight the outsized roles of age, amplification, and laterality relative to other predictors,

together accounting for over half of variance in productive vocabulary. We also explicitly examined the roles of prompt achievement of early intervention milestones on vocabulary. We found that overall, this sample showed vocabulary delays relative to hearing peers, and room for improvement in rates of early diagnosis and intervention in particular. This in turn highlights potential paths forward in ensuring that regardless of hearing status, we are able to provide language access and early childhood support to help children attain their potential.

**Acknowledgement**

Thank you to the Early Language Sensory Support Program for generously sharing their vocabulary assessments. We also thank Stephan Meylan for lending growth curve knowledge.

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## Captions

**Figure 1**: Results of chi-square tests between variables. X- and y-axes show the variables compared. Color of the square represents significance of the corresponding chi-square test. For tests that survived Bonferroni correction ($p < .0007$), effect size (Cramer's V) is given. (For the chi-square test, services received per month was binned into 0-2, 3-6, and > 7 services/month to create maximally evenly sized bins.)

**Figure 2**: Lines show the growth curves created from Wordbank 50th percentile data. Left panels show Words & Gestures; right panels show Words & Sentences. Top row is American English data; bottom row is Mexican Spanish data. Dots represent vocabulary scores of individual DHH children in the sample.

**Figure 3**: Age at diagnosis, intervention, amplification, and cochlear implantation across participants. Each dot represents the age that one child received the clinical service; violin width reflects data distribution. Black dots and whiskers show means and standard errors. Not all children received amplification (hearing aids) or implantation (cochlear implants).

**Figure 4**: Estimated vocabulary delay for children who meet 1-3-6 guidelines for diagnosis/intervention (top) and children who do not (bottom). Each dot represents one child in the sample; violin width reflect data distribution. Black dots and whiskers show means and standard error.

**Figure 5**: Unstandardized coefficients (measured in months) with 95% confidence intervals for the models selected by AIC for: (A) vocabulary delay, (B) age at diagnosis, (C) age at intervention.

**Table 1**: ~~CDI Ages, Vocabulary Scores and Rate of Developmental Delay: For each version of the CDI (WG = Words and Gestures; WS = Words and Sentences), the table shows the mean(SD) age, comprehension score (spoken + signed), and production score (spoken + signed, where relevant) of participants in our sample, along with the percent diagnosed with developmental delays. (N.B. signs were only reported for the 18 children using total communication as the rest reported solely spoken language as the communication modality).~~

~~**Table 2**~~Variables List: Detailed information about the variables studied. *Note*. For categorical variables, levels are described ~~and Ns provided.~~. Some participants had missing information for some variables, thus totals may not sum to 100. For continuous variables, range, mean, and standard deviation are provided. For CDI, participants were *either* administered Words and Gestures *or* Words and Sentences.

**Supplemental Materials S1**: Additional Diagnoses (n = 39): Ns of participants in our sample diagnosed with other conditions. N.B.: Ns do not sum to total because many participants had multiple diagnoses.

**Supplemental Materials S2**: Audiological Characteristics of the Sample: First two columns describe laterality and amplification type (cochlear implant (CI), hearing aid (HA), or none). Mean decibels of hearing loss (HL) in better ear, worse ear, and the mean age (in months) of amplification, and cochlear implantation (when applicable) for each laterality and amplification combination.

**Supplemental Materials S3**: Language and Communication Characteristics of the Sample: Ns of participants by language background and communication method.

**Supplemental Materials S4**: Unstandardized beta weights (months of vocabulary delay) for the model of vocabulary delay selected by AIC.

**Supplemental Materials S5**: Unstandardized beta coefficients (months) for the model of age at diagnosis selected by AIC.

**Supplemental Materials S6**: Unstandardized beta coefficients (months) for the model of age at intervention selected by AIC.

~~**Supplemental Materials S7**: PDF containing all figures and tables with their captions in situ (for reader/reviewer ease).~~

Figure 1



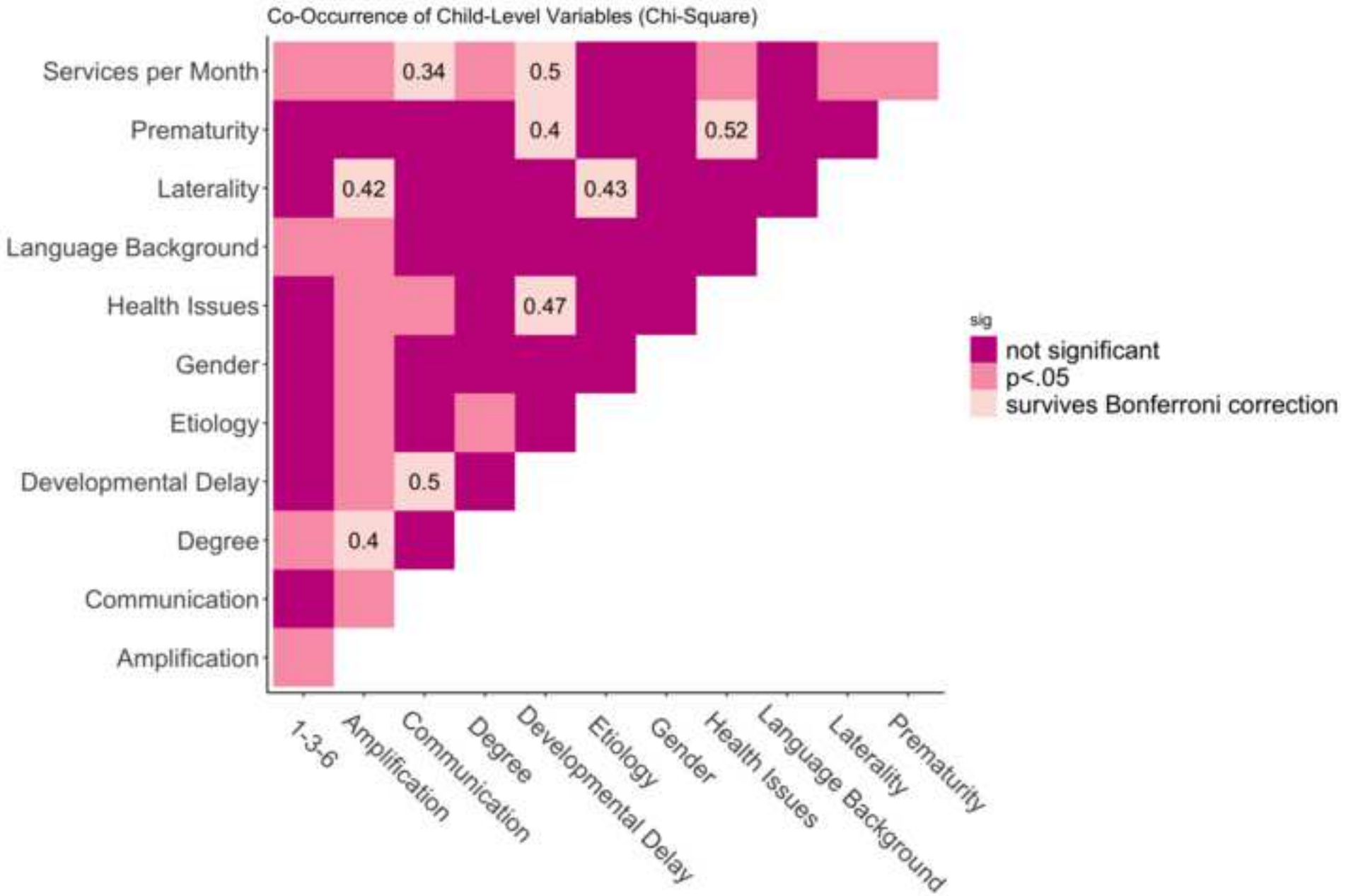

Figure 2



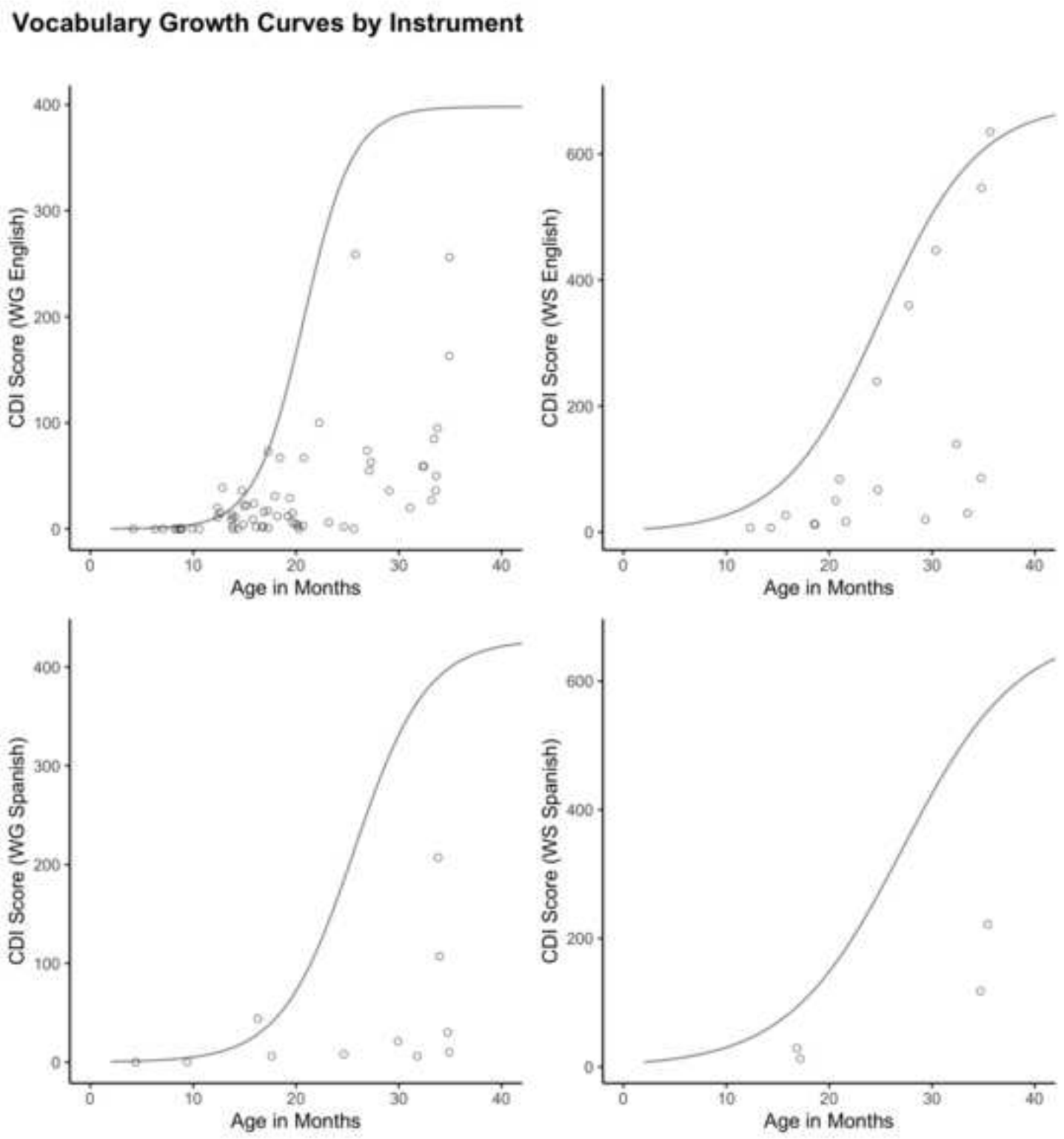

Figure 3



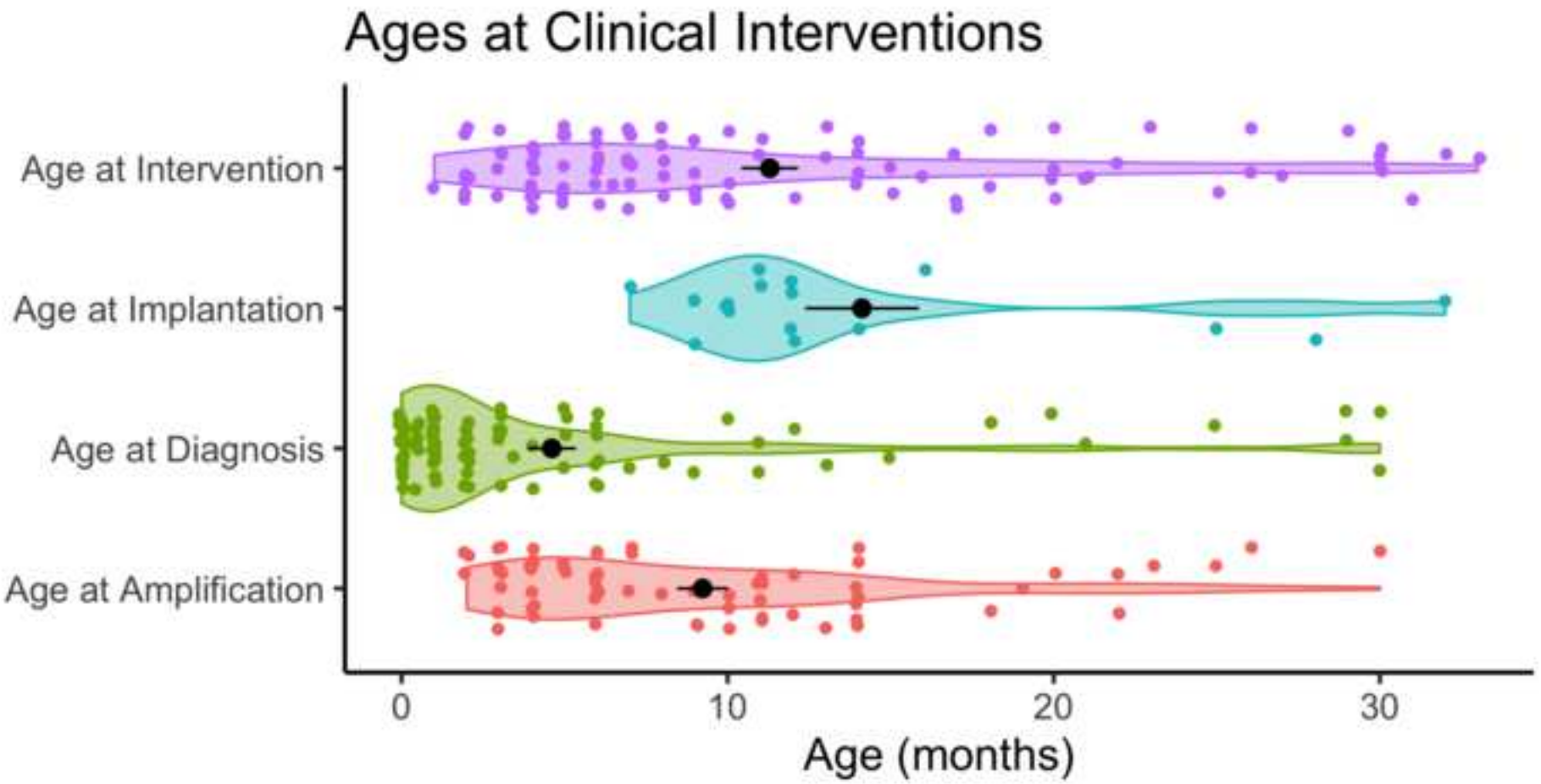

Figure 4



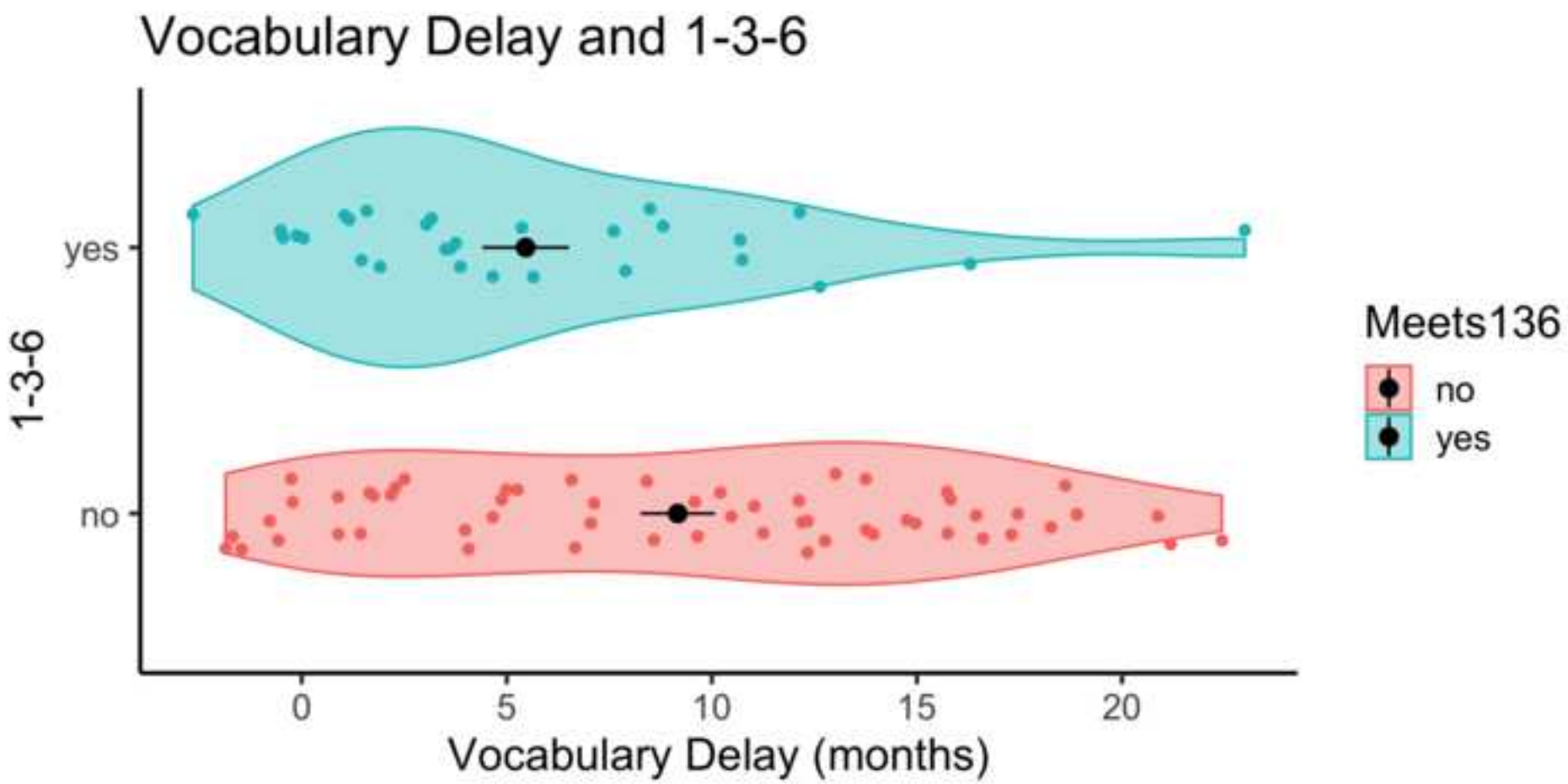

Figure 5



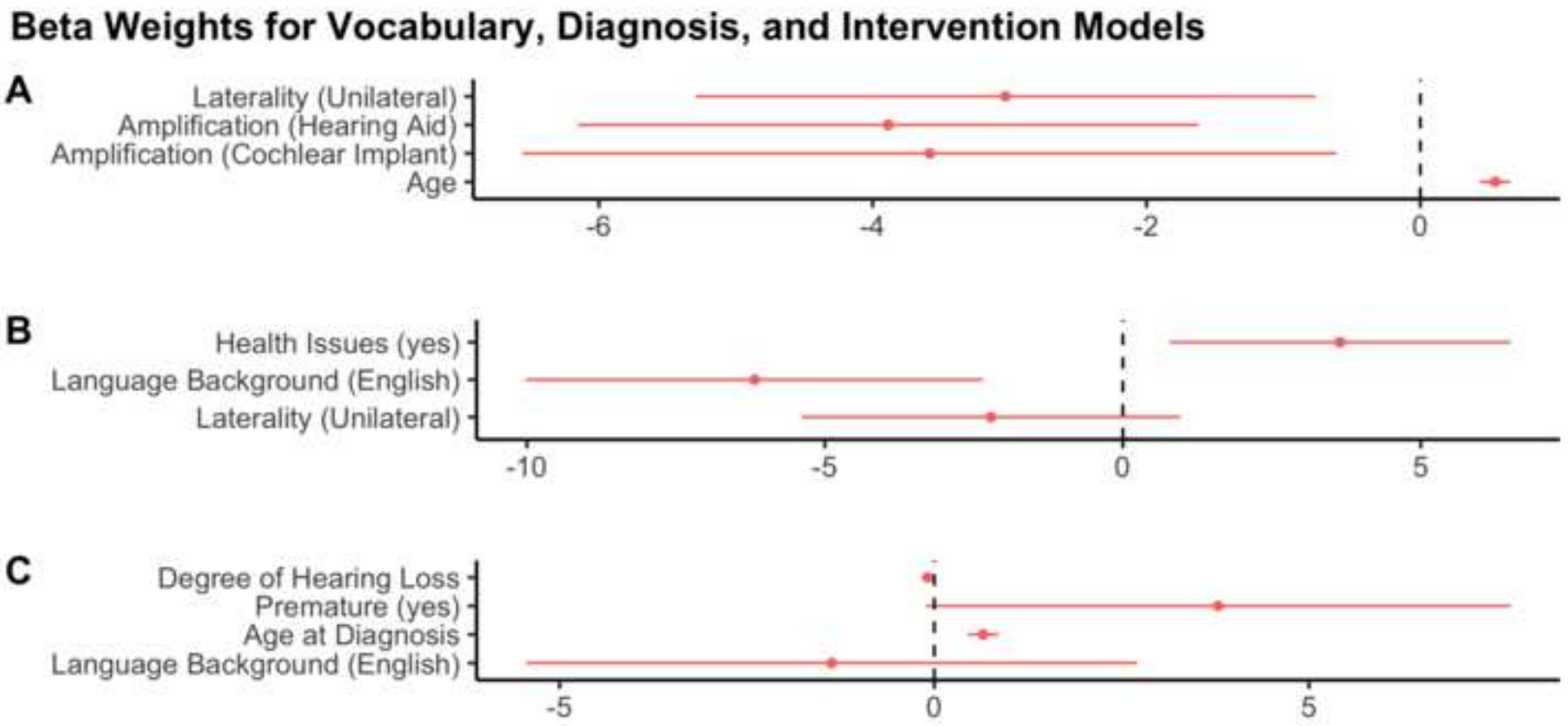

Table 1: *Variables List: Detailed information about the variables studied.*

| Variable | Range |
|---|---|
| Age | 4-36 months (mean (SD): 21 (9)) |
| Age at Amplification | 2-30 months (mean (SD): 9 (7)) |
| Age at Diagnosis | 0-30 months (mean (SD): 5 (7)) |
| Age at Implantation | 7-32 months (mean (SD): 14 (7)) |
| Age at Intervention | 1-33 months (mean (SD): 11 (9)) |
| Amplification | Hearing Aid (53) / Cochlear Implant (17) / None (28) |
| Communication | Spoken (79) / Total Communication (18) / Cued Speech (1) |
| Degree Hearing Loss (worse ear) | 18-100 dB HL (mean (SD): 64 (23)) |
| Developmental Delay | Yes (16) / No (82) |
| Gender | Female (43) / Male (57) |
| Health Issues | Yes (36) / No (62) |
| Language in Home | English (84) / Other (16) |
| Laterality | Unilateral (26) / Bilateral (72) |
| 1-3-6 | Yes (34) / No (61) |
| Premature Birth | Full-term (16) / Premature (82) |
| Services Per Month | 0-43 services per month (mean (SD): 5 (6)) |
| Etiology | Sensorineural (62) / Conductive (19) / Mixed (8) |
| Words and Gestures CDI - Words Produced | 0-259 words (mean (SD): 33 (53)) |
| Words and Sentences CDI - Words Produced | 7-635 words (mean (SD): 148 (184)) |

*Note.* For categorical variables, levels are described. Some participants had missing information for some variables, thus totals may not sum to 100. For continuous variables, range, mean, and standard deviation are provided. For CDI, participants were *either* administered Words and Gestures *or* Words and Sentences.

Dear Editor,

Thank you again for your thoughtful consideration of our manuscript for publication at JSLHR. We were delighted to read that you and the reviewers agreed that “the manuscript is much improved.”

In what follows, we address your and each of the reviewer’s concerns point-by-point, adding contiguous numbers across comments for expositional ease. Editor/Reviewer feedback is **bolded**, and our responses are plain text, with quoted text from the manuscript *italicized*.

In nearly all cases, we implemented the requested change, excepting reviewer comments 12 and 22, which we explain below.

We hope that you will find that the revised manuscript even clearer than the last, and we welcome any further feedback.

Kind regards,
Erin Campbell & Elika Bergelson

**Editor Comments:**

1. **Thank you for your attention to the previous comments. The reviewers and I agree that your manuscript is much improved. A few issues remain to address. Please see the reviewer comments below and address each concern. In addition, please ensure that all your table and figure captions adhere to APA format.**

   Thank you for your kind words, and for you and the reviewers’ role in improving the manuscript. We have addressed all reviewer comments as described below, and reformatted the tables to adhere to APA format.

**Reviewer #1 Comments:**

2. **This study demonstrates the highly eclectic nature of children who are Deaf or hard of hearing. It also shows that what is happening within a state may not be similar to nationwide trends (across all variables). With this in mind, the states should investigate their own population of children, just as the authors have done, and make recommendations based on their unique population residing there. The authors had a well thought out clinical implications section that can help to steer state-wide recommendations. The authors made fantastic revisions to the manuscript. The purpose was clear and the implications were easily linked. This paper makes a strong case for changes that may need to happen at the state-level. Rationale was clearly written and mapped to the methods and results . I saw a clear link to the 1-3-6 guidelines and the need to identify the population within the**

**author's state. The authors made notable edits to the literature review. It is now a clear, streamlined argument for the research study. Support for each variable was explored and extraneous information was removed.**

Thank you! We agree that implementing the reviewer feedback vastly improved the manuscript's clarity.

3. **Page 3- I appreciated the information in subpoint 1 (related to why the study focused on spoken language) was included. Maybe this sentence would be better in the main body versus a subpoint? It seems like an important justification for why the study focused on one modality over another, and should not get lost in the subpoints. It seems like this would fit into several places such as : after line 89 " we include 3 subgroups of DHH children tradtionally excluded..." , the methods section within the last paragraph (pg 11) or page 10 after line 165 " no eligibility criteria beyond hearing loss and 165 receiving an ELSSP evaluation were imposed".**

   RESPONSE: Thank you for these suggestions! We have moved subpoint 1 to the main text on line 42.

4. **Line 101- the authors refer to the study building off prior work, should have a citation identifying similar studies .**

   RESPONSE: We agree. We've added citations and this sentence now reads (line 103):

   *This study builds on prior work (e.g., Ching et al., 2013; Lund, 2016; Yoshinaga-Itano et al., 2017) by taking a new modeling approach for quantifying vocabulary delay across these variables.*

5. **The subpoint 3 (page 9) needs to be relabeled as subpoint "2".**

   RESPONSE: After moving several of the subpoints to the main text, we renumbered all of the subpoints and believe they are now consistent.

6. **Moving the discussion of the CDI from the introduction to the methods section was a great choice. It justified why the CDI was used.**

   RESPONSE: Thank you, it was an excellent reviewer suggestion!

7. **Much of the information included in table 2 is unnecessary, as this was included in the main body of the text. Maybe Table 1 and Table 2 could be combined as very little information is in table 1?**

   RESPONSE: We took Reviewer 1's suggestion to combine Tables 1&2.

8. **Table 1 and 2 are not in APA format. I am not sure if this is an issue of conversion to a PDF.**

   RESPONSE: Sorry about this. We have reformatted all tables to adhere to APA formatting guidelines.

**Just a few minor suggestions for [the Results] section:**

9. **Pg 12- subpoint 4 should be relabeled as subpoint “3”.**

   RESPONSE: All footnote numbering has been fixed.

10. **Subpoint 4 and 5 are important information related to the characteristics of the children within the study. They seem lost as subpoints. The authors should consider having them as their own paragraph within the results section.**

    RESPONSE: Done. This information is now in the main text directly, lines 146 and 168, respectively.

11. **Subpoint 7 also seems to be important information that should be included in the main body of the text.**

    RESPONSE: We moved footnote 7 to the main text on line 209.

12. **The font in line 306 looked different than the rest of the manuscript. This may just be due to changing to a pdf?**

    RESPONSE: Thank you for pointing out this formatting error! Unfortunately this doesn’t seem to be the case on our end, so we’re unsure how to troubleshoot it at this stage. We’ve double checked that all the font is the same throughout the document, and hopefully it renders properly!

13. **Line 178 in the line "regarding degree"...add "of loss"**

    RESPONSE: We added “of hearing loss” on line 174.

14. **Line 246- the wording "although whether" is awkward, consider revising.**

    RESPONSE: We reworded this sentence as follows (line 246):

    “Although 1-3-6 status was not included…”

15. **In this revision, the discussion section was clearer and more succinct, highlighting the authors main points.**

    RESPONSE: Thank you!

16. **Variables that the authors found were not be predictive should be specifically mentioned again.**

    RESPONSE: We have added this on line 351:

    *“We did not find that gender, developmental delay, health issues, premature birth, degree of hearing loss, communication modality, 1-3-6 status, number of services per month, or language background significantly improved model fit.“*

17. **Remove subpoint 9 from the discussion section. This information, if its not already, should be in the results section.**

    RESPONSE: We moved this information to line 276:

    *"One possibility for this last predictor is that the health issues caused hearing loss later in infancy; in our sample, 16 of the 35 children with health issues reported conditions that can in some cases cause acquired hearing loss (i.e., meningitis, sepsis, jaundice, seizures, hydrocephalus, MRSA, anemia, frequent fevers, cytomegalovirus)."*

18. **Line 407- the wording "in study after study" is informal. Consider revising**

    RESPONSE: We reworded this sentence as follows (line 417):

    "As early intervention predicts vocabulary outcomes across multiple studies…"

**Reviewer #2 Comments:**

19. **The authors have addressed all the issues and concerns that I raised in my initial review - thank you. I think the new manuscript is vastly improved and I would commend the authors for turning it around so well. The revised version continues to highlight the complexity of the results. Importantly, the manuscript now provides the rationale and background to the complexity in the introduction. The major concern over the previous version of the manuscript was that the novelty of the results and the contribution to the field was not clear. In my opinion, this has now been addressed. The rationale for the study is now clearly set out in the introduction. The discussion is still lengthy but it is more focused on the important findings and implications. I have four minor comments.**

    Thank you very much for your feedback on this and the previous version of the manuscript. We address your comments below.

20. **Line 134 - this sentence is written more akin to usual statements about the contribution of the study found in discussion sections rather than introduction sections.**

    RESPONSE: We agree with the reviewer's stylistic assessment, and we have moved this sentence to the discussion section on line 317.

21. **Line 140 - I was puzzled why the authors hypothesised that meeting the 1-3-6 guidelines would predict a larger spoken vocabulary delay. Isn't the aim of the guidelines to enable early access to support/intervention which should result in lower spoken vocabulary delays? Or is this prediction due to the inter-related nature of meeting the 1-3-6 guidelines being related to more severe levels of deafness which might result in greater spoken vocabulary delays? Upon reading the rest of the paper, I wonder if this is a typo?**

RESPONSE: Yes, thank you for catching this typo! We have fixed this on line 102.

22. **Line 161 - The authors provided an explanation of why the sample size was stopped at 100. The records were provided covering a 10 year period from 2010 to 2020. This explanation made me wonder whether this would have an effect upon generalisability of the results. Was this explored? Would we not expect that outcomes would have improved over time since 2010 once the 1-3-6 guidelines were implemented? This is a pertinent question considering the extent of the vocabulary delays reported for this group.**

    RESPONSE: Thank you for raising these questions! The EHDI 1-3-6 guidelines were implemented in the early 2000's (CDC, 2003; White, 2003; White, Forsman, Eichwald,& Munoz, 2010), and nationwide, rates of timely screening, diagnosis, and intervention have indeed increased in the time since (Subbiah, Mason, Gaffney, & Grosse, 2018). Within our North Carolina sample however, correlations between birth year and our outcomes were not found: we find no correlation with birth year and vocabulary delay (R=-0.15, p=.147), age at diagnosis (R=-0.02, p=0.88), or age at intervention (R=0.08, p=0.417). Given that our manuscript already runs on the long side, we have not included this in the manuscript at this point, but would be happy to do so if the editor or reviewers think it would be useful for the reader.

23. **Line 428 -this is a really important point the authors made; however I had to read this several times to understand what the representative refers to. Please clarify.**

    We have reworded this point as follows (line 338):

    *"For example, if an eyetracking experiment has a sample of typically-developing pediatric cochlear implant users with bilateral, severe-to-profound hearing loss, such a subsample may only represent roughly 14% of the DHH population."*

Table S1

Table S2

Table S3


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Supplemental Material
TableS3.docx

Table S4

Table S5

Table S6