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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:	<p>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.</p> <p>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.</p> <p>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.</p> <p>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.</p>	
Response to Reviewers:	<p>Dear Editor,</p> <p>Thank you very much for taking the time to consider our manuscript for publication at JSLHR and for allowing our resubmission. We were glad to hear that the reviewers thought that the study topic is “very interesting and relevant” (R2) and had “interesting findings” (R1), that the results section was “clearly written” (R1), and that our analyses were informative and appropriate. We are very grateful to you and the 2 reviewers for providing many helpful suggestions for reframing, shortening, and clarifying our manuscript.</p> <p>In our attached Revision Letter, we address your and each of the reviewer’s concerns point-by-point.</p> <p>We note that while we wholly agree with the majority of feedback, in a few cases, reviewers made suggestions not about the scientific content but about wording choices or what’s better handled in text vs. table. We largely took reviewers’ suggestions, but in</p>	

a few cases opted to retain the original wording or format; we are open to Editor feedback on these points.

Finally, to facilitate review and minimize portal download headaches, in addition to following the JSLHR guidelines for table and figure formats, we've added a supplementary pdf that includes all figures, tables, and supplementary tables in one place with their titles and captions.

We hope that you will find that the revised manuscript is clearer and more concise than the original. We thank you and the reviewers for your helpful and thoughtful comments.

Running head: DHH VOCABULARY, DIAGNOSIS, AND INTERVENTION

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

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Author note

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

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20 **Abstract**

21 **Purpose:** The study sought to 1) characterize the demographic, audiological, and
22 intervention variability in the population of DHH children receiving state services for hearing
23 loss; 2) identify predictors of vocabulary delays; and 3) evaluate factors influencing the success
24 and timing of early identification and intervention efforts at a state level.

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25 **Method:** One hundred Deaf/Hard-of-Hearing infants and toddlers (aged 4-36 months)
26 enrolled in early intervention completed the MacArthur-Bates Communicative Development
27 Inventory, and detailed information about their audiological and clinical history was collected.
28 We examined the influence of demographic, clinical, and audiological factors on vocabulary
29 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.

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

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 and whether there is any access to sign language, linguistic input will may be partially or totally inaccessible. Some 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), but 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 achievement¹ (Karchmer & Mitchell, 2003; Qi & Mitchell, 2012).

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¹ 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. Given this, we focus primarily on spoken language development.

52 ~~However, Though the literature points towards spoken language delays and deficits for~~
53 Deaf⁴ or Hard-of-Hearing ~~community(DHH) children, this~~ is a highly variable population with
54 highly variable ~~language~~ outcomes (Pisoni, Kronenberger, Harris, & Moberly, 2018).
55 ~~Previous~~For instance, previous research indicates that gender (Ching et al., 2013; Kiese-Himmel
56 & Ohlwein, 2002), additional disability (Ching et al., 2013; Verhaert et al., 2008; Yoshinaga-
57 Itano, Sedey, Wiggin, & Chung, 2017), degree and configuration of hearing loss (Ching et al.,
58 2013; de Diego-Lázaro, Restrepo, Sedey, & Yoshinaga-Itano, 2018; Vohr et al., 2011;
59 Yoshinaga-Itano et al., 2017), amplification (Walker et al., 2015), communication (Geers et al.,
60 2017), and early diagnosis/intervention (Yoshinaga-Itano et al., 2017; Yoshinaga-Itano, Sedey,
61 Wiggin, & Mason, 2018) ~~may~~ influence language outcomes in DHH children. A brief literature
62 review on the effect~~Although many~~ of these ~~predictors on~~variables reflect immutable
63 characteristics of the child, such as comorbid diagnoses or configuration of hearing loss, some
64 represent opportunities for clinicians and policy makers to intervene and potentially improve
65 language skills ~~in~~outcomes for DHH children ~~is provided in the Supplemental Materials (see~~
66 ~~table S1).~~

67 ~~Despite~~More specifically, early identification (Apuzzo & Yoshinaga-Itano, 1995;
68 Kennedy et al., 2006; Robinshaw, 1995; White & White, 1987; Yoshinaga-Itano, Sedey, Coulter,
69 & Mehl, 1998; Yoshinaga-Itano et al., 2018) and timely enrollment in early intervention
70 programs (Ching et al., 2013; Holzinger, Fellinger, & Beitel, 2011; Vohr et al., 2008, 2011;
71 Watkin et al., 2007) are associated with better language proficiency. Indeed, DHH children who
72 receive prompt diagnosis and early access to services have been found to meet age-appropriate
73 developmental outcomes, including language (Stika et al., 2015). In line with these findings, the
74 American Academy of Pediatricians (AAP) has set an initiative for Early Hearing Detection and

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

82 Notably, the variables linked to hearing loss mentioned above don't occur in a vacuum,
83 yet past work has largely attempted to measure their effects as if they were independent. For
84 instance, many excellent studies examining language focus on vocabulary development in DHH
85 children, there is still a gap in the literature describing and analyzing spoken language
86 development across the full range of children receiving services for hearing loss, with many
87 studies focusing in on specific subgroups (e.g. children under age X with Y level of hearing loss
88 and Z amplification approach, e.g., Vohr et al., Vohr et al., 2008; Yoshinaga-Itano et al., 2018).
89 In what follows, we 2008; Yoshinaga-Itano et al., 2018), which are not representative of the
90 broader population of DHH children. We take a different tack, asking instead how these factors
91 co-occur and interact in the context of the broad diversity of the DHH community, how they are
92 linked to early vocabulary, and how this connects with intervention and policy guidelines, within
93 a single state in the U.S.

94 **Goals, Predictions, and Key Contributions**

95 We present an empirical analysis of early vocabulary in a wide range of young DHH
96 children receiving state services in North Carolina. We have two broad goals. First, we aim to
97 provide a comprehensive description of a heterogeneous group of young children who receive

98 state services for hearing loss. Second, we aim to connect the intervention approaches and child
99 characteristics of this sample with children's spoken vocabulary², with the broader goal of
100 considering the success of early diagnosis and intervention initiatives.

101 In the present study, we analyze data from the MacArthur-Bates Communicative
102 Development Inventory (CDI; Fenson et al., 1994). This parent report instrument gathers
103 information about children's vocabulary development, and is commonly used in both research
104 and applied settings. The Words and Gestures version of the form is normed for 8–18-month-
105 olds. On Words and Gestures, parents indicate whether their child understands and/or produces
106 each of the 398 vocabulary items, and answer questions about young children's early
107 communicative milestones. The Words and Sentences version of the form is normed for 16–30-
108 month-olds. On Words and Sentences, parents indicate whether their child produces each of the
109 680 vocabulary items, and answer some questions about grammatical development. The CDI has
110 been normed on a large set of participants across many languages (Frank, Braginsky, Yurovsky,
111 & Marchman, 2017; Jackson-Maldonado et al., 2003).

112 The CDI has also been validated for DHH children with cochlear implants (Thal,
113 Desjardin, & Eisenberg, 2007). More specifically, in this validation, researchers asked parents to

²Despite exciting, increasing, and 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 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, and because no families in our sample used a full-fledged signed language, we focus on spoken language development.

114 complete the CDI, administered the Reynell Developmental Language Scales, and collected a
115 spontaneous speech sample. All comparisons between the CDI and the other measures yielded
116 significant correlations ranging from 0.58 to 0.93. Critically, the children in this study were
117 above the normed age range for the CDI, and thus this validation helps to confirm that the CDI is
118 a valid measurement tool for older DHH children. In further work, Castellanos, Pisoni,
119 Kronenberger, and Beer (2016) find that in children with CIs, number of words produced on the
120 CDI predicts language, executive function, and academic skills up to 16 years later. Building on
121 this work, several studies have used the CDI to measure vocabulary development in DHH
122 children (e.g., Yoshinaga-Itano et al., 2017, 2018; de Diego Lázaro et al., 2018; Vohr et al., 2008,
123 2011). We build on this literature in our analyses below.

124 **Goals and Predictions**

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

132 For the first goal, we expected that many of these variables would be relatedlinked, due to
133 known causal relations (e.g., cochlear implants recommended for severe hearing loss, but not
134 mild hearing loss). We soughtThis study contributes to provide descriptive documentation
135 aboutthe literature by quantifying the distribution and co-occurrence of demographic,
136 audiological, and intervention characteristics in a diverseour broad sample of DHH, which

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137 includes many children receiving state services often excluded from research. For the second
138 goal, we hypothesized that male (vs. female) gender, more severe degree of hearing loss, bilateral
139 (vs. unilateral) hearing loss, no amplification (vs. hearing aids and/or cochlear implants),
140 premature birth, meeting 1-3-6 guidelines, and presence of additional disabilities would predict
141 larger spoken vocabulary delay. We did not have strong predictions regarding the effects of
142 communication method or presence of other health issues (e.g., congenital heart
143 malformation). This study builds on prior work by taking a new modeling approach for
144 quantifying vocabulary-delay across these variables. For the third goal, based on the prior
145 literature summarized above, we hypothesized that children with less residual hearing (i.e.,
146 bilateral, more severe) and no co-occurring conditions would be earlier diagnosed and earlier to
147 begin language services, and that in turn earlier diagnosis would predict earlier intervention. This
148 study helps assess compliance with EHDI guidelines, and considers pathways for improvement.

149 Methods

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150 Clinical evaluations were obtained through an ongoing collaboration with the North
151 Carolina Early Language Sensory Support Program (ELSSP), an early intervention program
152 serving children with sensory impairments from birth to 36 months. ELSSP passed along sent
153 deidentified evaluations to our team after obtaining consent to do so from each family³. No

³ Because the data we received were already deidentified, this study was exempt from Duke University Institutional Review Board.

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154 ~~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.~~

156 ~~The clinical evaluations included demographic and audiological information, CDI vocabulary scores, and the results of any clinical assessments administered (e.g., PPVT), detailed further below. For some children, 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.~~

161 While this collaboration is ongoing, we opted to pause for this analysis upon receiving
162 data from 100 children. Thus, (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.

166 The clinical evaluations included demographic and audiological information and MacArthur Bates Communicative Development Inventory vocabulary scores (CDI, Fenson et al., 1994). reported sample 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.)

172 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 the Words and Sentences version (normed for 16–30-month-olds), parents indicate whether their child produces each of the

176 680 vocabulary items. Normative data for this instrument (Frank, Braginsky, Yurovsky, &
177 Marchman, 2017; Jackson-Maldonado et al., 2003) is available from WordBank, an open
178 database of CDI data. The CDI has also been validated for DHH children with cochlear implants
179 (Thal, Desjardin, & Eisenberg, 2007) in 32–66-month-olds. We build on prior literature using the
180 CDI to measure vocabulary in DHH children (e.g., Yoshinaga-Itano et al., 2017, 2018; de Diego-
181 Lázaro et al., 2018; Vohr et al., 2008, 2011) with a new analytic approach below ~~consists of~~

182 For this analysis, 100 children (56 male / 44 female) ages 4.~~20~~–~~36~~.~~2010~~–~~35.70~~ months

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183 (M=21.20, SD=9.10) contributed data. Race and socioeconomic information were not available.
184 Families were administered either the Words and Gestures or Words and Sentences version of the
185 CDI based on clinician judgment ~~of linguistic ability~~. Children who were too old for Words and
186 Gestures, but who were not producing many words at the time of assessment, were often given
187 Words and Gestures (n = 37). Families ~~for whom Spanish was the~~ whose primary language ~~was~~
188 ~~Spanish~~ (n = ~~44~~15) completed the Spanish language version of the CDI (Jackson-Maldonado et
189 al., 2003). Both spoken words and signs counted as word productions. See Table 1 for additional
190 CDI information for our sample. A summary of all the variables we examined is available in
191 Table 2, and more detailed ~~demographic and audiological~~ information can be found in the
192 Supplemental Materials, tables S2–S4Tables S1-S3.

193 Results

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194 ~~We split the~~The results ~~into three parts. In the first~~are organized mirroring the goals
195 ~~outlined above. First~~, we explore relationships among child demographic, audiological, and
196 clinical variables. ~~In the second~~Second, we use these variables to predict vocabulary
197 development. Finally, ~~in the third~~, we describe the implementation of the EHDI 1-3-6 guidelines

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198 and predictors of early diagnosis and intervention in this sample. All analyses were conducted in
199 R. All code is available on our OSF page (<https://osf.io/kfes3/>). (R Core Team, 2020) and all
200 code to generate this manuscript in Rstudio (RStudio Team, 2020) is available via OSF.

201 Relationships Among Demographic, Audiological, and Clinical Variables

202 Before we testtesting how these variables may be relatedrelate to vocabulary and clinical ← Formatted: Space Before: 10 pt
203 milestones, we describe their relationships to each other. As would be expected, many health, ← Formatted: Space Before: 9 pt, After: 12 pt
204 audiological, and clinical characteristics are not distributed randomly across this sample of
205 children. To quantify this statistically, we used Bonferroni-corrected chi-square tests between
206 each of our variables. Because the chi-square statistic assumes n > 5 is expected in the majority of
207 the cells for each test (preferably ≥ 80% McHugh, 2013), we excluded mixed hearing loss (n = 8)
208 and cued speech (n = 1) from this section of the analysis.⁴ Strictly speaking, some variables are
209 not expected to be randomly distributed relative to each other (e.g., premature birth and health
210 issues; degree and amplification), but quantifying the differences via chi-square using a
211 conservative significance threshold lets us highlight the strongest relationships within this
212 dataset.

213 Given that we ran 66 Chi square tests, Bonferroni corrected alpha for this set of analyses
214 was $p < 0.0007$. Of these the 66 combinations of variables, $p < .05$ for 2627, and 9 survived

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⁴ Because the chi-square statistic assumes $n > 5$ is expected in the majority of the cells for each test (preferably $\geq 80\%$ McHugh, 2013), we excluded mixed hearing loss ($n = 8$) and cued speech ($n = 1$) from this analysis.

215 Bonferroni correction-($p < 0.0007$). We ~~are only discussing~~^{limit} discussion to the latter below,
216 but depict the full set ~~of results can be found~~ in Figure 1.

217 As expected, ~~we found that~~ health issues, developmental delays, and premature birth were
218 highly interrelated in our sample, such that children born premature were more likely to also
219 experience health issues ($X^2 (1, N = 98) = 23.997 = 24.72$, $p < .0001$) and developmental delays
220 ($X^2 (1, N = 98) = 11.6397 = 12.84$, $p = .00060003$), and children with developmental delays
221 were more likely to also experience health issues ($X^2 (1, N = 98) = 20.8797 = 19.38$, $p < .0001$).
222 Children with developmental delays received more services per month than typically-developing
223 children ($X^2 (2, N = 95) = 22.1794 = 23.47$, $p < .0001$) and were more likely to use total
224 communication ($X^2 (2, N = 98) = 22.5497 = 24.52$, $p < .0001$). Likewise, children who used
225 total communication received more services per month than children using spoken language (X^2
226 ($4, N = 9594$) = 21.3505 , $p = .0003$).

227 We also confirmed expected relationships among many of the audiological characteristics.
228 There was a significant relationship between laterality and etiology ($X^2 (2, N = 88) = 18.2987 =$
229 17.98, $p = .0001$), such that children with conductive hearing loss were more likely to have
230 unilateral hearing loss, and children with sensorineural hearing loss were more likely to have a
231 bilateral loss⁵. The chi-square tests further showed that amplification was related to laterality (X^2
232 ($2, N = 9897$) = 16.432 , $p = .0003$) and degree of hearing loss ($X^2 (4, N = 8786) = 28.4518$, $p <$
233 ~~.0001~~^{in our sample.}). Specifically, children with bilateral hearing loss were more likely than

⁵ All children with mixed hearing loss ($n = 8$) had bilateral hearing loss.

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234 children with unilateral hearing loss to use a hearing aid or cochlear implant; no child with
235 unilateral hearing loss used a cochlear implant, and many children with unilateral hearing loss
236 used no amplification. Regarding degree, children with severe-to-profound hearing loss were
237 more likely to use a cochlear implant than children with less severe hearing loss (i.e., mild or
238 moderate),hearing loss.

239 Taken together, the results in this set of analyses serve to highlight the notable
240 interconnectedness among early health and development on the one hand (i.e. health issues,
241 prematurity, and developmental delays), and audiological characteristics (i.e. links among
242 laterality, etiology, amplification, and degree of hearing loss)on the other).

243 Predictors of Vocabulary Delay

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

250 As noted above, the two CDI is composed of two instruments, which differ in
251 number of questions (i.e. the maximum how many vocabulary score is 398 on Words and
252 Gestures and 680 on Words and Sentences; 428 and 680 respectively for Spanish language
253 CDI).items they contain. To take this into account, rather than using the raw number of words
254 produced as our outcome variable, we use WordBank norms to establish the difference (in
255 months) between the child's chronological age and their predicted age based on their productive

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256 vocabulary, derived from the WordBank norms (Frank et al., 2017), rather than using the raw
257 vocabulary scores. We call this derived variable *vocabulary delay*.

258 More specifically, to compute a child's predicted age from their vocabulary score, we used
259 the 50th percentile for productive vocabulary from WordbankWordBank data for typically-
260 developing infants (Frank et al., 2017) to create binary logistic growth curves separately for the
261 "Words and Gestures" (WG) and "Words and Sentences" (WS) versions of the CDI for American
262 English and Mexican Spanish⁶. For each child, we took the number of words they produced
263 divided(spoken and/or signed, though the latter was only provided for children using Total
264 Communication (n = 18) as all others were reported to exclusively use spoken language). We
265 then divided this production score by the number of words on the instrument, to give us the
266 proportion of words produced. We used this proportion in an inverse prediction from the binary
267 logistic regression curves to generate a predicted age. That is, for each possible CDI score, the
268 growth curve provided the age that the score would be achieved for the 50th percentile trajectory.
269 Finally, we subtracted the predicted age from each child's chronological age to calculate their
270 vocabulary delay. However, for children producing 0 words, this approach was not appropriate
271 due to the long tails on the growth curves. Thus, for this subset of children, we took the x-
272 intercept from Wordbank (8 months for English, and 9 months for Spanish), and subtracted that
273 value from the child's chronological age to get their vocabulary delay.

⁶ Number of hearing children in normative sample for each growth curve: WG-English=1071,

WG-Spanish=760; WS-English=1461, WS-Spanish=1092

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274 To look at the relationship between our predictor variables and CDI scores, we next
275 conducted multiple linear regression, using vocabulary delay as our outcome variable.⁷

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

279 This model accounted for significant variance in vocabulary delay (adjusted-R² = 0.5958,
280 $p < .001$). We next performed stepwise model comparison using stepAIC (MASS) to pare down
281 the model. This process selects only the predictors which incrementally improved model fit,
282 measured by Akaike's Information Criterion (AIC), which considers goodness of fit and model
283 complexity (penalizing models with many predictors). We started model selection with the full
284 model, as described above. We then filtered out data from children for whom Meets 1-3-6 (n =
285 56) or Degree (n = 4314) was unknown, as this stepwise AIC approach does not permit missing
286 values across predictors. Since this initial filtered analysis found that Degree and 1-3-6 did not
287 improve model fit, we manually removed the Degree and 1-3-6 terms from the model selection so
288 that the 4516 participants with missing cases for these variables could be retained.⁸

⁷ Children who were too young for the CDI version they were administered (n = 9) were excluded from this analysis. Additionally, we excluded, as was the adopted child due to concerns about comparing her/his score to the American English CDI norms.

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⁸ 4 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

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289 Based on this iterative process, we arrived at the following final model: Vocabulary Delay
290 ~ Age + Laterality + Amplification. ~~This~~~~No other variables from the full model above~~
291 ~~significantly improved model fit, and are thus not discussed further. Our final model accounted~~
292 for significant variance in children's vocabulary delay to a nearly identical degree as the full
293 model (adjusted-R² = 0.58, $p < .001$, see Table ~~SSS4~~ & Figure 5.A). We found significant main
294 effects for Age, ~~Laterality, and Amplification, and Laterality,~~ such that older age, ~~no~~
295 ~~amplification, and~~ bilateral hearing loss, ~~and no amplification~~ predicted greater vocabulary
296 delays. Compared to children with no amplification, children with cochlear implants had a 3.~~5049~~
297 months smaller spoken vocabulary delay ($p = .021022$), and similarly children with hearing aids
298 had a 3.~~8477~~ months smaller delay ($p = .001$). Children with unilateral hearing loss had a 2.~~7073~~
299 months smaller delay ($p = .020$) than children with bilateral hearing loss. ~~With regard to Age, for~~
300 ~~each month olderFor Age~~, the model predicted a 0.55 months *larger* vocabulary delay ($p <$
301 ~~.001~~~~) for each additional month of age.~~

302 Given our ~~first set of~~ results ~~above revealing~~~~regarding~~ relationships among several of
303 these variables (e.g., laterality and amplification), we tested for collinearity ~~econcerns~~ by
304 computing the model's VIF (variance inflation factor). This revealed low levels of collinearity
305 among predictors in our final model (all VIF < 1.20; James, Witten, Hastie, & Tibshirani, 2013).
306 In sum, the analyses in this section revealed that over half of the variance in DHH children's

Developmental Delay. However, ANOVA revealed that including a Developmental Delay term did not significantly improve model fit when including the ~~4516~~ participants without Degree information.

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307 vocabulary scores was explained by their age, whether they receive amplification, and whether
308 their hearing loss was unilateral or bilateral.

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

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

314 ~~To this end, we looked at the ages at which children received diagnosis and intervention, and how~~
315 ~~this mapped onto the 1-3-6 guidelines. In this section~~~~To this end,~~ we provide a brief description
316 of the implementation of 1-3-6 in our sample, examine its effect on vocabulary delay, and
317 describe the results of exploratory linear regression models for age at diagnosis and age at
318 intervention.

319 Overall, 37% of our sample met 1-3-6 guidelines for early diagnosis and intervention.
320 ~~Among~~~~Breaking this down further, among~~ the children for whom screening information was
321 available ($n = 6867$), 100% were screened at birth or during NICU stay. ~~69~~In our sample, 70% of
322 children received diagnosis by 3 months of age, and ~~3940~~40 began early intervention by 6 months
323 of age. ~~Among children with comorbidities, 21.05% met 1-3-6 guidelines, compared to 47.37%~~
324 ~~of children without comorbidities. (see~~ Figure 3 ~~shows the age at first diagnosis, intervention,~~
325 ~~amplification, and implantation for each child in our sample.)~~

326 We first tested the link between 1-3-6 and vocabulary directly ~~in an exploratory analysis~~.
327 An independent samples t-test showed that children who did not meet 1-3-6 guidelines had
328 significantly larger vocabulary delays than children who met 1-3-6 guidelines ($t(68.7869.27) =$

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329 2.6268, $p = 0.01$; see Figure 4). On average, the group that did not meet 1-3-6 guidelines was
330 3.6271 months more delayed with regard to vocabulary (relative to the same 50th percentile
331 benchmark ~~from hearing children in Wordbank~~ described above).

332 To better understand implementation of 1-3-6 guidelines, we next ~~zoomed in on turned~~
333 ~~our focus to factors influencing the timing of~~ diagnosis and intervention. We conducted two
334 linear regressions, one for ~~predicting~~ age at diagnosis and one for age at intervention, ~~considering~~
335 ~~only the predictors that would have been available or relevant at each of these stages (as detailed~~
336 ~~below)~~. Model selection followed the same stepwise AIC-based process as described in the
337 preceding section.

338 For age at diagnosis, we included the set of child-specific factors that would be relevant
339 before diagnosis of hearing loss (e.g., we excluded amplification type because ~~a child~~
340 ~~would children did~~ not receive ~~a hearing aid or cochlear implant amplification~~ prior to ~~being~~
341 ~~diagnosed with~~ hearing loss ~~diagnosis~~.) We began with: gender, degree, developmental delay,
342 health issues, prematurity, laterality, language background, and etiology.

343 The best ~~fitting~~ model was: Age at Diagnosis ~ Health Issues + Language Background +
344 Laterality, with significant main effects of Health Issues and Language Background (see Table
345 S6S5 & Figure 5.B). This model accounted for 16.4411% of the variance in age at diagnosis ($p =$
346 .001). Average age at diagnosis was 4.6558(7.4920) months. Relative to English-speaking
347 families, children from Spanish-speaking families were diagnosed 6.4751 months later ($p = .001$).
348 Children with health issues were diagnosed 3.7057 months later than children without health
349 issues ($p = .01$).

350 We repeated this model selection process for age at intervention. In addition to the
351 variables used to fit the intervention model, we included age at diagnosis. The best fit model was:
352 Age at Intervention ~ Premature Birth + Degree + Age at Diagnosis + Language Background
353 ($R^2=0.43$, $p < .001$; See Table S7S6 & Figure 5.C), with significant main effects of degree and
354 age at diagnosis. Prematurity ($\beta = 3.7879$, $p = .06$) and language background ($\beta = -1.3839$, $p =$
355 $.52$) were not significant predictors on their own, but their inclusion improved model fit. Average
356 age at intervention was $11.4205(8.5455)$ months. More severe hearing loss predicted earlier
357 intervention, such that for every additional 10 dB HL, predicted age at intervention was 1 month
358 earlier ($p < .01$). With regard to age at diagnosis, for every month diagnosis was delayed,
359 intervention was delayed by 2.80 weeks ($p < .01$). Taken together, these analyses reveal that
360 beyond aspects of the child's hearing status, other variables to children's audiological
361 characteristics, comorbid diagnoses, and language background contribute to delays in both
362 diagnoses and intervention. We return to this point in the discussion.

Discussion

363
364 In this study, we examined the demographic, audiological, and clinical characteristics of
365 100 young DHH children in North Carolina. We documented the distribution of these
366 characteristics and explored the relationships between these variables, vocabulary, diagnosis, and
367 intervention. In prior work with tightly controlled samples, the variables studied here have been
368 shown to be relevant for language development, but their effects have rarely been examined in
369 the full heterogeneity they naturally occur within. We took this big-tent approach by including
370 any children receiving services for hearing loss.

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371 Returning to our original three questions, we asked first: how are child-level variables
372 intertwined? We found significant structure across many of the variables, suggesting that in a
373 real-world sample of children with hearing loss, many factors are intrinsically not dissociable.
374 This was particularly true for many of the auditory characteristics and comorbid diagnoses. To
375 our knowledge, this paper provides the first population-based documentation of this distribution.
376 We next asked whether these characteristics can predict vocabulary outcomes for DHH children.
377 We found that a model including only children's age, laterality of hearing loss, and amplification
378 type best accounted for the variability in ~~spoken~~-vocabulary outcomes. Finally, we asked: how
379 successful ~~were~~ the 1-3-6 guidelines were for early detection and intervention, both in terms of
380 improving child outcomes and ensuring timely diagnosis and intervention ~~for all children with~~
381 ~~hearing loss?~~ Here, we found that children who met 1-3-6 guidelines indeed had a smaller
382 vocabulary delay than those who didn't. However, only 37% of children met these guidelines.
383 Our results highlight family- and health-related variables (~~e.g. language background, health~~
384 ~~issues~~) that accounted for significant variability in when children received diagnosis and/or
385 intervention.

386 ~~To us, We believe~~ the inherent complexity in these results is an important piece of
387 understanding ~~spoken language~~vocabulary outcomes ~~for children with hearing loss~~ within the
388 diverse population of ~~Deaf/Hard of Hearing~~DHH children. We next highlight some implications
389 of this study for future research and clinical practice.

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

391 In our sample, we found significant overlap among demographic, audiological, and
392 clinical variables. ~~Prematurity~~To highlight a few of these findings, prematurity, health issues, and

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393 developmental delay frequently co-occurred, such that children with one of these factors were
394 more likely to have the others.~~This is not surprising. Many conditions that cause developmental~~
395 ~~delays have a high incidence of health issues (e.g., heart problems in Down Syndrome; vomiting~~
396 ~~and seizures, consistent with hydrocephalus), and it is well documented that there is a higher~~
397 ~~incidence of developmental delay and health issues in preterm infants prior research~~ (Luu, Katz,
398 Leeson, Thébaud, & Nuyt, 2016; Pierrat et al., 2017). ~~In our sample, we also had a large~~
399 ~~range Given that the constellation of health comorbid conditions is so varied~~ (76 unique conditions
400 in our sample of 100 children; see Table S1 ~~in Supplemental Materials for more detailed~~
401 ~~information about comorbidities). Some studies to date have examined the outcomes of DHH~~
402 ~~children with certain conditions (e.g., Clibbens, 2001; Szymanski, Brice, Lam, & Hotto, 2012).~~
403 ~~But given that the constellation of comorbid conditions is so varied.~~) an important direction for
404 future research is whether cognitive and social abilities, as well as family's treatment resources,
405 are predictive of language outcomes across conditions.

406 We also found that children with developmental delays (e.g., Down syndrome) were much
407 more likely to use a total communication approach than DHH children without developmental
408 delays (i.e., total communication used by ~~58.82~~^{62.50}% of DHH children with developmental
409 delay vs. 9.88% of those without). That is, ~~communication modality was not distributed randomly~~
410 ~~throughout our sample, with~~ use of total communication ~~linked to~~ was more likely for children
411 already at greater risk for verbal delays. ~~Such a pattern Quantifying this confound~~ is ~~in line with~~
412 ~~clinical use an important contribution of manual communication approaches for young children~~
413 ~~with disabilities (e.g., Branson & Demehak, 2009). This result tempers this work, as it calls for~~
414 ~~tempering~~ the interpretation of correlational studies finding links between total communication
415 and language delays (e.g., Geers et al., 2017).

416 Our audiological variables too were not randomly distributed relative to each other. To
417 highlight one such result, amplification devices were more common for children with less hearing
418 (i.e., children with bilateral hearing loss and children with moderate to profound hearing loss).
419 This may be due to the assumption that a hearing aid or cochlear implant will not benefit children
420 with minimal hearing loss, although several studies have found benefits for amplification for mild
421 or unilateral hearing loss (Briggs, Davidson, & Lieu, 2011; Hassepass et al., 2013; Walker et al.,
422 2015).

423 The relationships we found among variables were more confirmatory than surprising,
424 particularly those reflecting known causal links (e.g., increased health issues in children
425 born premature). Nevertheless, they should caution us to think critically about how we construct
426 samples for controlled lab experiments. During study design: how likely is it If a researcher
427 desires to collect a desired sample of (e.g.) 32 typically-developing pediatric cochlear implant
428 users with bilateral, severe-to-profound hearing loss, how representative would the results be,
429 given that such a subsample may only represent roughly 14% of the DHH population, as it does
430 here? During interpretation of the results: how might the findings generalize to the rest of the
431 DHH population given the constraints of the study at hand? Such considerations are important for
432 properly representing, understanding, and supporting DHH children and their families. This
433 becomes doubly important in the context of interpreting language outcomes like vocabulary.

434 Predicting vocabulary outcomes

435 In our sample, 88.89% of DHH children fell below the 50th percentile for spoken
436 vocabulary. Moreover, of the 11.11% who were at or above the 50th percentile, 55.56% were 8-
437 to 9-month-olds who were not yet producing any words (as expected at this age). Finding that
438 nearly 90% of DHH children are below the 50th percentile for vocabulary development indicates

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439 ~~that this group is not yet well equipped to acquire spoken language vocabulary, indicating that a~~
440 ~~large majority of this sample is behind a normative sample of their hearing peers in word~~
441 ~~learning.~~ This disadvantage can have lasting consequences in the lives of DHH children
442 (Karchmer & Mitchell, 2003; Qi & Mitchell, 2012), highlighting the importance of understanding
443 what factors contribute to it.

444 ~~We predicted that male gender, more severe hearing loss, bilateral hearing loss, no~~
445 ~~amplification, premature birth, and presence of additional disabilities would be associated with~~
446 ~~larger spoken vocabulary delay.~~ In contrast to our predictions, the best model predicting
447 vocabulary delay had just a few variables: age, amplification, and laterality. ~~Notably, we did not~~
448 ~~simply find that DHH children were learning words at the same rate (albeit delayed) as hearing~~
449 ~~children, which would have led to a constant delay across developmental time. Instead~~
450 ~~Notably,~~ we see that the spoken vocabulary delay widens with age, indicating that the *rate* of spoken
451 vocabulary acquisition is slower for DHH children. ~~The result is a population increasingly behind~~
452 ~~on spoken language milestones.~~ Given that none of the children here use sign language (which
453 can ensure earlier language access), this vocabulary delay is likely to have knock-on effects for
454 language development more broadly ~~as well. This in turn has policy, alongside~~ implications ~~that~~
455 ~~are critical to consider for public policy.~~

456 Predicting early diagnosis and intervention

457 Our exploration of the implementation of 1-3-6 guidelines revealed that only ~~36.84~~~~37.23%~~ ←
458 of children met the EHDI guidance for diagnosis by 3 months and intervention by 6 months,
459 ~~despite ample evidence suggesting early diagnosis and intervention improve language outcomes.~~
460 Our results were consistent with prior work (e.g., Yoshinaga-Itano, Sedey, Coulter, & Mehl, et

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461 al., 1998 @ ching2013). Children in our sample; Ching et al., 2013), finding that children who
462 met ~~1-3-6~~the guidelines were 3.~~62~~71 months less delayed in spoken vocabulary than children
463 who were late to receive diagnosis and/or services. With these demonstrable benefits in mind, our
464 sample, byBy dint of accepting all children receiving early intervention services in one state, was
465 able to explore naturally occurring variance in our dataset let us delve deeper into who received
466 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.~~70~~57 months later than infants without health issues. One possible explanation is that the For a small fraction of cases, this may have been because health issues caused acquired hearing loss that wouldn't be detected by the newborn hearing screening, thus, delaying its identification of hearing loss. In our sample, 16 of the 36 children with health issues had conditions that might cause acquired hearing loss (i.e., meningitis, sepsis, jaundice, seizures, hydrocephalus, MRSA, anemia, frequent fevers, cytomegalovirus). While acquired hearing loss may be one driver of delayed diagnosis for children with health issues, this accounts for only a fraction of the subpopulation with health issues. Another possible explanation is that the health issues required more pressing medical attention than the possible hearing loss. For instance,⁹. Of course, some situations may require families and medical providers are likely to prioritize treatment for certain health issues (e.g.,

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⁹ 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).

479 surgery for congenital heart defect) over diagnostic audiology services. ~~Nevertheless, it is~~
480 ~~possible~~ That said, our results raise the possibility that ~~in some cases,~~ clinician awareness of ~~the~~
481 increased delays in language ~~related~~linked to ~~the prevalence of~~ health issues ~~more broadly~~ may
482 facilitate improvements in timely diagnosis.

483 Language background too predicted age at diagnosis. ~~Infants, such that infants~~ from
484 Spanish-speaking families were diagnosed 3.~~7879~~ months later than infants from English-
485 speaking families. This may be due to cultural differences in attitudes towards deafness
486 (Caballero, Muñoz, Schultz, Graham, & Meibos, 2018; Rodriguez & Allen, 2020; Steinberg,
487 Bain, Li, Delgado, & Ruperto, 2003) or ~~it may result from~~ a lack of linguistically accessible and
488 culturally appropriate audiology services. Only 5.6% of American audiologists identify as
489 bilingual service providers (ASHA, 2019), and services from a monolingual provider may be
490 insufficient. ~~To this point, Caballero et al. (2017) found that Hispanic American parents of DHH~~
491 ~~children wish for more concrete resources, comprehensive information, and emotional support~~
492 ~~from their audiologist. In a nationwide survey of audiologists, the majority of audiologists~~
493 ~~reported that language barriers presented a major challenge in working with Spanish speaking~~
494 ~~families, specifically, particularly~~ in obtaining the child's case history and providing
495 recommendations for follow-up services (Abreu, Adriatico, & DePierro, 2011).

Intervention.

As expected, more severe hearing loss predicted earlier intervention;

497 ~~such that for every additional 10 dB HL, predicted age at intervention was 0.93 month earlier.~~
498 This ~~converges with findings by Harrison, Roush, and Wallace (2003) in which severe~~ may be
499 ~~due to profound hearing loss was diagnosed 2-5 months earlier than mild-to-moderate hearing~~
500 ~~loss. Parents~~ parents and clinicians ~~may adopt~~adopting a wait-and-see approach to intervention
501 for children with some residual hearing. ~~Nevertheless,~~ despite associations between mild-to-

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502 moderate hearing loss ~~is associated with, and~~ language delays and academic challenges (Blair,
503 Peterson, & Viehweg, 1985; Delage & Tuller, 2007),~~which early~~. Early intervention may help
504 offset these associations.

505 Age at start of services was also associated with age at diagnosis: for ~~everyeach~~ month
506 diagnosis was delayed, intervention was delayed by 2.80 weeks. Ching et al. (2013) found that
507 age at intervention predicted better outcomes for DHH children, above and beyond age at
508 diagnosis. Of course, these two variables are related, ~~such that we cannot hope to~~
509 ~~achieveunderscoring the importance of~~ early ~~intervention goals without ensuring children receive~~
510 ~~timely~~ diagnosis. ~~Early diagnosis puts for putting~~ children in the pipeline towards ~~intervention~~
511 earlier intervention.

512 Finally, it's important to note that this sample is composed of children receiving birth-to-3
513 services. ~~An estimated 67% of children with hearing loss enroll in early intervention services~~
514 ~~(CDC, 2018)~~. While this represents a tremendous step forward in prompt early intervention
515 ~~services relative to just a few decades ago, early intervention may not be early enough. Less than~~
516 ~~39% of our sample of children in early intervention meet the 6-month EHDI benchmark.~~
517 ~~Furthermore, an unknown fraction of the DHH population in North Carolina aren't included in~~
518 ~~this analysis because they have not been enrolled in services by 36 months. The AAP estimates~~
519 ~~that almost 36% of infants who do not pass a newborn hearing screening are lost to follow-up.~~
520 ~~Assuming that the population of children in early intervention only represents two thirds of the~~
521 ~~population with hearing loss, Less than 40% of our sample of children in early intervention meet~~
522 ~~the 6-month EHDI benchmark. Given that only about 67% of children with hearing loss enroll in~~
523 ~~early intervention services (CDC, 2018)~~, our data suggest that the actual proportion of DHH
524 children who receive intervention by the EHDI-recommended 6 months may be closer to 26%.

525 These children may not receive clinical support until school-age or later, exacerbating concerns
526 for language development, which lays an important foundation for literacy and academic success
527 (Hemphill & Tivnan, 2008; Stæhr, 2008).

528 **Educational and Clinical Implications**

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

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

540 Additionally, the vast majority of children in our sample experienced vocabulary delays
541 (relative to hearing peers), and studies of spoken vocabulary development in older DHH children
542 suggest that they may not catch up (Lund, 2016). This should set clinicians and educators on high
543 alert, ~~due to the demonstrated importance of vocabulary skills in literacy (Stæhr, 2008) and in education more broadly (e.g., Young, 2005; Monroe & Orme, 2002)~~. As early intervention
544 predicts vocabulary outcomes in study after study (including this present study and e.g., Vohr et
545 al., 2008; Ching, Dillon, Leigh, & Cupples, 2018), ensuring intervention by 6 months for all
546

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547 DHH children may be one way to address spoken vocabulary deficits. Another ~~solution: even~~
548 ~~prior to intervention or amplification, option may be the~~ provision of structured, accessible
549 language input (i.e., sign language) ~~may mitigate even prior to intervention or amplification,~~
550 ~~potentially mitigating~~ negative effects of auditory deprivation on language skills (Davidson,
551 Lillo-Martin, & Pichler, 2014; Hassanzadeh, 2012). ~~Indeed, while we recognize that While~~
552 learning sign language may pose a challenge for some families for myriad reasons, ~~and (as noted~~
553 ~~above, underscored by its absence as a communication modality within~~ our sample ~~did not use~~
554 ~~sign language.)~~ we nevertheless ~~feel it is worth underseoring highlight its potential~~ as an
555 important language support for DHH children and their families.

556 In recommending sign language, we endorse the rationale set forth by Hall, Hall, and
557 Caselli (2019). Summarizing their view, they note that spoken language outcomes for DHH
558 children are variable and unpredictable (Ganek, McConkey Robbins, & Niparko, 2012; Szagun &
559 Schramm, 2016), and even in optimal situations, many DHH children do not achieve age-
560 appropriate spoken language outcomes (e.g., Geers et al., 2017). Failing to achieve language
561 proficiency (in any language) confers higher risk of disrupted cognitive, academic, and
562 socioemotional development (Amraei, Amirsalari, & Ajalloueyan, 2017; Dammeyer, 2010;
563 Desselle, 1994; Hall, Levin, & Anderson, 2017; Hrastinski & Wilbur, 2016; Kushalnagar et al.,
564 2011; Schick, De Villiers, De Villiers, & Hoffmeister, 2007). The available data do not suggest
565 that sign language harms spoken language development (Davidson et al., 2014; Park et al., 2013),
566 and in fact, some studies suggest that sign language *benefits* spoken language development (e.g.,
567 Hassanzadeh, 2012). Providing early access to a natural sign language offers children another
568 path to language mastery, and use of sign language does not preclude learning spoken language.

569 Thus, we encourage sign language use *at least* prior to mastery of spoken language, and when
570 possible for the family, we encourage its continued use as a language resource.

571 Limitations and Opportunities for Future Work

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572 This study represents an important first step in quantifying variability in demographic
573 characteristics, language outcomes, and 1-3-6 attainment. At the same time, it is exploratory, has
574 limited geographic scope, and analyzed data from a (deliberately) high-variability sample. We
575 see these limitations as opportunities for future investigation into the complex factors influencing
576 DHH children's outcomes.

577 Given our exploratory analyses, there were many possible analytic routes. That said, our
578 results largely converge with or replicate key aspects of past studies (e.g., Ching et al., 2013) and
579 received wisdom among clinicians. In the interest of transparency, these data and all code
580 generating our results are available on our OSF page (<https://osf.io/kfes3/>), and we We encourage
581 those interested readers to explore further analyses- using the data and/or code provided on our
582 OSF page.

583 This sample is composed only of children in North Carolina, and, While certain factors
584 vary by country and by state (e.g., diagnosis and early intervention practices; NAD, n.d.).
585 However, based on other demographic research (Blackorby & Knokey, 2006; Institute, 2014),,
586 our sample largely resembles the national DHH population in terms of degree of hearing loss,
587 percentage of children with additional disabilities, cochlear implant and hearing aid use, language
588 background, and gender. We would exercise caution (Blackorby & Knokey, 2006; Gallaudet
589 Research Institute, 2014). It did diverge from the national sample in applying these results to
590 communication modality: our sample had no signers while 20% of DHH children have sign as

591 their primary modality (Gallaudet Research Institute, 2014). A similar naturalistic study in
592 regions where sign language access for DHH children is more common (e.g. Washington D.C.) ~~A~~
593 ~~similar naturalistic study in those regions could help illuminate~~~~would be a welcome addition to~~
594 ~~the present work, in illuminating~~ the effects of different clinical and demographic factors in a
595 signing population. One further limitation to our analyses and to assessing representativeness of
596 ~~the sample is that race and socioeconomic status information was not available.~~

597 Finally, the considerable variability in the sample did not allow us to easily isolate effects
598 of different factors. ~~However, as discussed above, this (e.g., degree vs. amplification). This~~
599 reflects real-world variability ~~that is often does not make sense to isolate. Instead, this~~
600 ~~limitation and~~ would be best addressed by larger sample sizes. As researchers continue to study
601 influences on vocabulary in DHH children, a meta-analytic approach too may be able to better
602 estimate ~~effects and~~ effect sizes within the varied outcomes of this heterogeneous population.

Conclusion

603 The present study explored interrelations among demographic and audiological
604 characteristics, vocabulary outcomes, and clinical milestones within a diverse sample of 100
605 DHH children enrolled in early intervention services, in North Carolina. Our population-based
606 description underscores heavily interlocking demographic, audiological, and clinical
607 characteristics (e.g. communication approach and presence of developmental delays). Our models
608 highlight the outsized roles of age, amplification, and laterality relative to other predictors,
609 together accounting for over half of variance in productive vocabulary. We also explicitly
610 examined the roles of prompt achievement of early intervention milestones on vocabulary. We
611 found that overall, this sample showed spoken language-vocabulary delays relative to hearing

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613 peers ~~on average~~, and room for improvement in rates of early diagnosis and intervention.

614 Critically, we also found that the variables predicting these delays in both vocabulary and early

615 support services reflected *both* dimensions that are immutable, and those that clinicians and

616 caretakers can potentially alter in particular. This in turn highlights potential paths forward in

617 ensuring that regardless of hearing status, we are able to provide language access and early

618 childhood support to help children attain their potential.

619 Acknowledgement

620 Thank you to the Early Language Sensory Support Program for generously sharing their
621 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. ~~Blacks dot~~ 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. ~~Blacks dot~~ 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.

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

900 **Table 2:** Detailed information about the variables studied. For categorical variables, levels
901 are described. and Ns provided. For continuous variables, range, mean, and standard deviation
902 are provided.

903 **Supplemental Materials S1:** A summary of previous research on predictors of
904 vocabulary in DHH children. + equals bigger vocab, - equals smaller vocab.

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905 **Supplemental Materials S2:** Additional Diagnoses (n = 39): Ns of participants in our
906 sample diagnosed with other conditions. N.B.: Ns do not sum to total because many participants
907 had multiple diagnoses.

908 **Supplemental Materials S3S2:** Audiological Characteristics of the Sample: First two
909 columns describe laterality and amplification type (cochlear implant (CI), hearing aid (HA), or
910 none). Mean decibels of hearing loss (HL) in better ear, worse ear, and the mean age (in months)
911 of amplification, and cochlear implantation (when applicable) for each laterality and
912 amplification combination.

913 **Supplemental Materials S4S3:** Language and Communication Characteristics of the
914 Sample: Ns of participants by language background and communication method.

915 **Supplemental Materials S5S4:** Unstandardized beta weights (months of vocabulary

916 delay) for the model of vocabulary delay selected by AIC.

917 **Supplemental Materials S5:** Unstandardized beta coefficients (months) for the model of

918 age at diagnosis selected by AIC.

919 **Supplemental Materials S6:** Unstandardized beta coefficients (months) for the model of

920 age at diagnosisintervention selected by AIC.

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921 **Supplemental Materials S7:** Unstandardized beta coefficients (months)PDF containing

922 all figures and tables with their captions in situ (for the model of age at intervention selected by

923 AIC-reader/reviewer ease).

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

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Author note

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

18 **Abstract**

19 **Purpose:** The study sought to 1) characterize the demographic, audiological, and
20 intervention variability in the population of DHH children receiving state services for hearing
21 loss; 2) identify predictors of vocabulary delays; and 3) evaluate factors influencing the success
22 and timing of early identification and intervention efforts at a state level.

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

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

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

36

Introduction

37 In the United States, 1-2 children are born with hearing loss, per 1,000 births (CDC,
38 2018), of which ~90% will be born to hearing parents (Mitchell & Karchmer, 2004), in a home
39 where spoken language is likely the dominant communication method. Depending on the type
40 and degree of hearing loss, whether the child uses amplification, and whether there is any access
41 to sign language, linguistic input may be partially or totally inaccessible. While some of these
42 children will develop spoken language proficiency within the range of their hearing peers (Geers,
43 Mitchell, Warner-Czyz, Wang, & Eisenberg, 2017; Verhaert, Willems, Van Kerschaver, &
44 Desloovere, 2008), many will face persistent spoken language deficits (Eisenberg, 2007; Luckner
45 & Cooke, 2010; Moeller, Tomblin, Yoshinaga-Itano, Connor, & Jerger, 2007), which may later
46 affect reading ability and academic achievement¹ (Karchmer & Mitchell, 2003; Qi & Mitchell,
47 2012).

48 Though the literature points towards spoken language delays and deficits for Deaf or
49 Hard-of-Hearing (DHH) children, this is a highly variable population with highly variable
50 language outcomes (Pisoni, Kronenberger, Harris, & Moberly, 2018). For instance, previous

¹ 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. Given this, we focus primarily on spoken language development.

51 research indicates that gender (Ching et al., 2013; Kiese-Himmel & Ohlwein, 2002), additional
52 disability (Ching et al., 2013; Verhaert et al., 2008; Yoshinaga-Itano, Sedey, Wiggin, & Chung,
53 2017), degree and configuration of hearing loss (Ching et al., 2013; de Diego-Lázaro, Restrepo,
54 Sedey, & Yoshinaga-Itano, 2018; Vohr et al., 2011; Yoshinaga-Itano et al., 2017), amplification
55 (Walker et al., 2015), communication (Geers et al., 2017), and early diagnosis/intervention
56 (Yoshinaga-Itano et al., 2017; Yoshinaga-Itano, Sedey, Wiggin, & Mason, 2018) influence
57 language outcomes in DHH children. Although many of these variables reflect immutable
58 characteristics of the child, such as comorbid diagnoses or configuration of hearing loss, some
59 represent opportunities for clinicians and policy makers to intervene and potentially improve
60 language outcomes for DHH children.

61 More specifically, early identification (Apuzzo & Yoshinaga-Itano, 1995; Kennedy et al.,
62 2006; Robinshaw, 1995; White & White, 1987; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998;
63 Yoshinaga-Itano et al., 2018) and timely enrollment in early intervention programs (Ching et al.,
64 2013; Holzinger, Fellinger, & Beitel, 2011; Vohr et al., 2008, 2011; Watkin et al., 2007) are
65 associated with better language proficiency. Indeed, DHH children who receive prompt diagnosis
66 and early access to services have been found to meet age-appropriate developmental outcomes,
67 including language (Stika et al., 2015). In line with these findings, the American Academy of
68 Pediatricians (AAP) has set an initiative for Early Hearing Detection and Intervention (EHDI).
69 These EHDI guidelines recommend that DHH children are screened by 1 month old, diagnosed
70 by 3 months, and enter early intervention services by 6 months. We refer to this guideline as 1-3-
71 6. Meeting this standard appears to improve spoken language outcomes for children with hearing
72 loss and the benefits appear consistent across a range of demographic characteristics (Yoshinaga-
73 Itano et al., 2017, 2018), so it remains an important research goal to identify children at risk of

74 receiving clinical support late, in order to help all children achieve prompt diagnosis and
75 intervention.

76 Notably, the variables linked to hearing loss mentioned above don't occur in a vacuum,
77 yet past work has largely attempted to measure their effects as if they were independent. For
78 instance, many studies focus on vocabulary development in specific subgroups (e.g. children
79 under age X with Y level of hearing loss and Z amplification approach, e.g., Vohr et al., 2008;
80 Yoshinaga-Itano et al., 2018), which are not representative of the broader population of DHH
81 children. We take a different tack, asking instead how these factors co-occur and interact in the
82 context of the broad diversity of the DHH community, how they are linked to early vocabulary,
83 and how this connects with intervention and policy guidelines, within a single state in the U.S.

84 **Goals, Predictions, and Key Contributions**

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

93 For the first goal, we expected that many of these variables would be linked, due to known
94 causal relations (e.g., cochlear implants recommended for severe hearing loss, but not mild
95 hearing loss). This study contributes to the literature by quantifying the distribution and co-

96 occurrence of demographic, audiological, and intervention characteristics in our broad sample,
97 which includes many children often excluded from research. For the second goal, we
98 hypothesized that male (vs. female) gender, more severe degree of hearing loss, bilateral
99 (vs. unilateral) hearing loss, no amplification (vs. hearing aids and/or cochlear implants),
100 premature birth, meeting 1-3-6 guidelines, and presence of additional disabilities would predict
101 larger spoken vocabulary delay. This study builds on prior work by taking a new modeling
102 approach for quantifying vocabulary delay across these variables. For the third goal, we
103 hypothesized that children with less residual hearing (i.e., bilateral, more severe) and no co-
104 occurring conditions would be earlier diagnosed and earlier to begin language services, and that
105 in turn earlier diagnosis would predict earlier intervention. This study helps assess compliance
106 with EHDI guidelines, and considers pathways for improvement.

107

Methods

108 Clinical evaluations were obtained through an ongoing collaboration with the North
109 Carolina Early Language Sensory Support Program (ELSSP), an early intervention program
110 serving children with sensory impairments from birth to 36 months. ELSSP sent deidentified
111 evaluations to our team after obtaining consent to do so from each family². While this
112 collaboration is ongoing, we opted to pause for this analysis upon receiving data from 100
113 children (collected between 2010 and 2020, before the COVID-19 epidemic reached North

² Because the data we received were already deidentified, this study was exempt from Duke University Institutional Review Board.

114 Carolina in Spring 2020). Given our goal of characterizing the full range of DHH children with
115 hearing loss in North Carolina, no eligibility criteria beyond hearing loss and receiving an ELSSP
116 evaluation were imposed.

117 The clinical evaluations included demographic and audiological information and
118 MacArthur Bates Communicative Development Inventory vocabulary scores (CDI, Fenson et al.,
119 1994). For some children, evaluations from multiple timepoints or other instruments were
120 available (e.g. PPVT). We limit the scope of the present study to only the CDI (as this was
121 available for all children), and only the first evaluation (due to concerns regarding within-subjects
122 variance for statistical analysis.)

123 The CDI is a parent-report instrument measuring children's vocabulary. On the Words and
124 Gestures version of the form (normed for 8–18-month-olds), parents indicate whether their child
125 understands and/or produces each of the 398 vocabulary items. One the Words and Sentences
126 version (normed for 16–30-month-olds), parents indicate whether their child produces each of the
127 680 vocabulary items. Normative data for this instrument (Frank, Braginsky, Yurovsky, &
128 Marchman, 2017; Jackson-Maldonado et al., 2003) is available from WordBank, an open
129 database of CDI data. The CDI has also been validated for DHH children with cochlear implants
130 (Thal, Desjardin, & Eisenberg, 2007) in 32–66-month-olds. We build on prior literature using the
131 CDI to measure vocabulary in DHH children (e.g., Yoshinaga-Itano et al., 2017, 2018; de Diego-
132 Lázaro et al., 2018; Vohr et al., 2008, 2011) with a new analytic approach below.

133 For this analysis, 100 children (56 male / 44 female) ages 4.10–35.70 months ($M=21.20$,
134 $SD=9.10$) contributed data. Race and socioeconomic information were not available. Families
135 were administered either the Words and Gestures or Words and Sentences version of the CDI

136 based on clinician judgment of linguistic ability. Children who were too old for Words and
137 Gestures, but who were not producing many words at the time of assessment, were often given
138 Words and Gestures ($n = 37$). Families whose primary language was Spanish ($n = 15$) completed
139 the Spanish language version of the CDI (Jackson-Maldonado et al., 2003). Both spoken words
140 and signs counted as word productions. See Table 1 for additional CDI information for our
141 sample. A summary of all the variables we examined is available in Table 2, and more detailed
142 information can be found in the Supplemental Materials, Tables S1-S3.

143 **Results**

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

150 **Relationships Among Demographic, Audiological, and Clinical Variables**

151 Before testing how these variables relate to vocabulary and clinical milestones, we
152 describe their relationships to each other. To quantify this statistically, we used Bonferroni-

153 corrected chi-square tests between each of our variables³. Strictly speaking, some variables are
154 not expected to be randomly distributed relative to each other (e.g., premature birth and health
155 issues; degree and amplification), but quantifying the differences via chi-square using a
156 conservative significance threshold lets us highlight the strongest relationships within this
157 dataset.

158 Of the 66 combinations of variables, $p < .05$ for 27, and 9 survived Bonferroni correction
159 ($p < 0.0007$). We limit discussion to the latter below, but depict the full set in Figure 1.

160 As expected, health issues, developmental delays, and premature birth were highly
161 interrelated in our sample, such that children born premature were more likely to also experience
162 health issues ($X^2 (1, N = 97) = 24.72, p < .0001$) and developmental delays ($X^2 (1, N = 97) =$
163 12.84, $p = .0003$), and children with developmental delays were more likely to also experience
164 health issues ($X^2 (1, N = 97) = 19.38, p < .0001$). Children with developmental delays received
165 more services per month than typically-developing children ($X^2 (2, N = 94) = 23.47, p < .0001$)
166 and were more likely to use total communication ($X^2 (2, N = 97) = 24.52, p < .0001$). Likewise,
167 children who used total communication received more services per month than children using
168 spoken language ($X^2 (4, N = 94) = 21.05, p = .0003$).

³ Because the chi-square statistic assumes $n > 5$ is *expected* in the majority of the cells for each test (preferably $\geq 80\%$ McHugh, 2013), we excluded mixed hearing loss ($n = 8$) and cued speech ($n = 1$) from this analysis.

169 We also confirmed expected relationships among many of the audiological characteristics.
170 There was a significant relationship between laterality and etiology (X^2 (2, N = 87) = 17.98, p =
171 .0001), such that children with conductive hearing loss were more likely to have unilateral
172 hearing loss, and children with sensorineural hearing loss were more likely to have a bilateral
173 loss⁴. The chi-square tests further showed that amplification was related to laterality (X^2 (2, N =
174 97) = 16.2, p = .0003) and degree of hearing loss (X^2 (4, N = 86) = 28.18, p < .0001).
175 Specifically, children with bilateral hearing loss were more likely than children with unilateral
176 hearing loss to use a hearing aid or cochlear implant; no child with unilateral hearing loss used a
177 cochlear implant, and many children with unilateral hearing loss used no amplification.
178 Regarding degree, children with severe-to-profound hearing loss were more likely to use a
179 cochlear implant than children with mild or moderate hearing loss.

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

184 **Predictors of Vocabulary Delay**

185 We next turn to the relationship between these variables and children's productive
186 vocabulary, as measured by the CDI. Figure 2 shows the vocabulary scores of children in our
187 samples relative to norms for hearing children for each CDI form. Descriptively, we found

⁴ All children with mixed hearing loss (n = 8) had bilateral hearing loss.

188 widespread vocabulary delays, with the majority of DHH children testing around or below the
189 25th percentile for hearing children (based on WordBank norms; Frank et al., 2017).

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

195 More specifically, to compute a child's predicted age from their vocabulary score, we used
196 the 50th percentile for productive vocabulary from WordBank data for typically-developing
197 infants (Frank et al., 2017) to create binary logistic growth curves separately for the "Words and
198 Gestures" (WG) and "Words and Sentences" (WS) versions of the CDI for American English and
199 Mexican Spanish⁵. For each child, we took the number of words they produced (spoken and/or
200 signed, though the latter was only provided for children using Total Communication ($n = 18$) as
201 all others were reported to exclusively use spoken language). We then divided this production
202 score by the number of words on the instrument, to give us the proportion of words produced. We
203 used this proportion in an inverse prediction from the binary logistic regression curves to
204 generate a predicted age. That is, for each possible CDI score, the growth curve provided the age
205 that the score would be achieved for the 50th percentile trajectory. Finally, we subtracted the

⁵ Number of hearing children in normative sample for each growth curve: WG-English=1071,
WG-Spanish=760; WS-English=1461, WS-Spanish=1092

206 predicted age from each child's chronological age to calculate their vocabulary delay. However,
207 for children producing 0 words, this approach was not appropriate due to the long tails on the
208 growth curves. Thus, for this subset of children, we took the x-intercept from Wordbank (8
209 months for English, and 9 months for Spanish), and subtracted that value from the child's
210 chronological age to get their vocabulary delay.

211 To look at the relationship between our predictor variables and CDI scores, we next
212 conducted multiple linear regression, using vocabulary delay as our outcome variable⁶.

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

216 This model accounted for significant variance in vocabulary delay (adjusted- $R^2 = 0.58$, $p < .001$). We next performed stepwise model comparison using stepAIC (MASS) to pare down the
217 model. This process selects only the predictors which incrementally improved model fit,
218 measured by Akaike's Information Criterion (AIC). We started model selection with the full
219 model, as described above. We then filtered out data from children for whom Meets 1-3-6 ($n = 6$)
220 or Degree ($n = 14$) was unknown, as this stepwise AIC approach does not permit missing values
222 across predictors. Since this initial filtered analysis found that Degree and 1-3-6 did not improve

⁶ Children who were too young for the CDI version they were administered ($n = 9$) were excluded from this analysis, as was the adopted child due to concerns about comparing their score to the American English CDI norms.

223 model fit, we manually removed the Degree and 1-3-6 terms from the model selection so that the
224 16 participants with missing cases for these variables could be retained.⁷

225 Based on this iterative process, we arrived at the following final model: Vocabulary Delay
226 ~ Age + Laterality + Amplification. No other variables from the full model above significantly
227 improved model fit, and are thus not discussed further. Our final model accounted for significant
228 variance in children's vocabulary delay to a nearly identical degree as the full model (adjusted-R²
229 = 0.58, $p = < .001$, see Table S4 & Figure 5.A). We found significant main effects for Age,
230 Laterality, and Amplification, such that older age, bilateral hearing loss, and no amplification
231 predicted greater vocabulary delays. Compared to children with no amplification, children with
232 cochlear implants had a 3.49 months smaller spoken vocabulary delay ($p = .022$), and similarly
233 children with hearing aids had a 3.77 months smaller delay ($p = .001$). Children with unilateral
234 hearing loss had a 2.73 months smaller delay ($p = .020$) than children with bilateral hearing loss.
235 For Age, the model predicted a 0.55 months *larger* vocabulary delay ($p < .001$) for each
236 additional month of age.

⁷ 4 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 16 participants without Degree information.

237 Given our first set of results regarding relationships among several of these variables (e.g.,
238 laterality and amplification), we tested for collinearity by computing the model's VIF (variance
239 inflation factor). This revealed low levels of collinearity among predictors in our final model (all
240 $VIF < 1.20$; James, Witten, Hastie, & Tibshirani, 2013). In sum, the analyses in this section
241 revealed that over half of the variance in DHH children's vocabulary scores was explained by
242 their age, whether they receive amplification, and whether their hearing loss was unilateral or
243 bilateral.

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

245 Perhaps of greatest importance to clinicians and policymakers is the implementation and
246 effect of existing policies. Although whether a child met 1-3-6 guidelines was not included in our
247 final model predicting vocabulary delay through our model selection process, its demonstrated
248 importance for language outcomes (e.g., Yoshinaga-Itano et al., 2018) merits further discussion.
249 To this end, we provide a brief description of the implementation of 1-3-6 in our sample, examine
250 its effect on vocabulary delay, and describe the results of exploratory linear regression models for
251 age at diagnosis and age at intervention.

252 Overall, 37% of our sample met 1-3-6 guidelines for early diagnosis and intervention.
253 Breaking this down further, among the children for whom screening information was available (n
254 = 67), 100% were screened at birth or during NICU stay. In our sample, 70% of children received
255 diagnosis by 3 months of age, and 40% began early intervention by 6 months of age (see Figure
256 3).

257 We first tested the link between 1-3-6 and vocabulary directly. An independent samples t-
258 test showed that children who did not meet 1-3-6 guidelines had significantly larger vocabulary

259 delays than children who met 1-3-6 guidelines ($t(69.27) = 2.68, p = 0.01$; see Figure 4). On
260 average, the group that did not meet 1-3-6 guidelines was 3.71 months more delayed with regard
261 to vocabulary (relative to the same 50th percentile benchmark described above).

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

266 For age at diagnosis, we included the set of child-specific factors that would be relevant
267 *before* diagnosis of hearing loss (e.g., we excluded amplification type because children did not
268 receive amplification prior to hearing loss diagnosis.) We began with: gender, degree,
269 developmental delay, health issues, prematurity, laterality, language background, and etiology.

270 The best fitting model was: Age at Diagnosis ~ Health Issues + Language Background +
271 Laterality, with significant main effects of Health Issues and Language Background (see Table
272 S5 & Figure 5.B). This model accounted for 16.11% of the variance in age at diagnosis ($p =$
273 .001). Average age at diagnosis was 4.58(7.20) months. Relative to English-speaking families,
274 children from Spanish-speaking families were diagnosed 6.51 months later ($p = .001$). Children
275 with health issues were diagnosed 3.57 months later than children without health issues ($p = .01$).

276 We repeated this model selection process for age at intervention. In addition to the
277 variables used to fit the intervention model, we included age at diagnosis. The best fit model was:
278 Age at Intervention ~ Premature Birth + Degree + Age at Diagnosis + Language Background
279 ($R^2=0.43, p < .001$; See Table S6 & Figure 5.C), with significant main effects of degree and age
280 at diagnosis. Prematurity ($\beta = 3.79, p = .06$) and language background ($\beta = -1.39, p = .52$) were

not significant predictors on their own, but their inclusion improved model fit. Average age at intervention was 11.05(8.55) 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.

296 Returning to our original three questions, we asked first: how are child-level variables
297 intertwined? We found significant structure across many of the variables, suggesting that in a
298 real-world sample of children with hearing loss, many factors are intrinsically not dissociable.
299 This was particularly true for many of the auditory characteristics and comorbid diagnoses. To
300 our knowledge, this paper provides the first population-based documentation of this distribution.
301 We next asked whether these characteristics can predict vocabulary outcomes for DHH children.
302 We found that a model including only children's age, laterality of hearing loss, and amplification

303 type best accounted for the variability in vocabulary outcomes. Finally, we asked how successful
304 the 1-3-6 guidelines were for early detection and intervention, both in terms of improving child
305 outcomes and ensuring timely diagnosis and intervention. Here, we found that children who met
306 1-3-6 guidelines indeed had a smaller vocabulary delay than those who didn't. However, only
307 37% of children met these guidelines. Our results highlight family- and health-related variables
308 that accounted for significant variability in when children received diagnosis and/or intervention.

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

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

313 In our sample, we found significant overlap among demographic, audiological, and
314 clinical variables. To highlight a few of these findings, prematurity, health issues, and
315 developmental delay frequently co-occurred, such that children with one of these factors were
316 more likely to have the others, consistent with prior research (Luu, Katz, Leeson, Thébaud, &
317 Nuyt, 2016; Pierrat et al., 2017). Given that the constellation of comorbid conditions is so varied
318 (76 unique conditions in our sample of 100 children; see Table S1), an important direction for
319 future research is whether cognitive and social abilities, as well as family's treatment resources,
320 are predictive of language outcomes across conditions.

321 We also found that children with developmental delays (e.g., Down syndrome) were much
322 more likely to use a total communication approach than DHH children without developmental
323 delays (i.e., total communication used by 62.50% of DHH children with developmental delay
324 vs. 9.88% of those without). That is, use of total communication was more likely for children

325 already at greater risk for verbal delays. Quantifying this confound is an important contribution of
326 this work, as it calls for tempering the interpretation of correlational studies finding links between
327 total communication and language delays (e.g., Geers et al., 2017).

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

337 **Predicting vocabulary outcomes**

338 In our sample, 88.89% of DHH children fell below the 50th percentile for vocabulary,
339 indicating that a large majority of this sample is behind a normative sample of their hearing peers
340 in word learning. This disadvantage can have lasting consequences in the lives of DHH children
341 (Karchmer & Mitchell, 2003; Qi & Mitchell, 2012), highlighting the importance of understanding
342 what factors contribute to it.

343 In contrast to our predictions, the best model predicting vocabulary delay had just a few
344 variables: age, amplification, and laterality. Notably, we see that the spoken vocabulary delay
345 widens with age, indicating that the *rate* of spoken vocabulary acquisition is slower for DHH
346 children. Given that none of the children here use sign language (which can ensure earlier

347 language access), this vocabulary delay is likely to have knock-on effects for language
348 development more broadly, alongside implications for public policy.

349 **Predicting early diagnosis and intervention**

350 Our exploration of the implementation of 1-3-6 guidelines revealed that only 37.23% of
351 children met the EHDI guidance for diagnosis by 3 months and intervention by 6 months. Our
352 results were consistent with prior work (e.g., Yoshinaga-Itano et al., 1998; Ching et al., 2013),
353 finding that children who met the guidelines were 3.71 months *less* delayed in spoken vocabulary
354 than children who were late to receive diagnosis and/or services. By dint of accepting all children
355 receiving early intervention services in one state, our dataset let us delve deeper into *who*
356 received on-time diagnosis and intervention.

Diagnosis. Having health issues or a non-English language background predicted

358 later diagnosis. Children with health issues were diagnosed 3.57 months later than infants without
359 health issues. For a small fraction of cases, this may have been because health issues caused
360 acquired hearing loss, delaying its identification⁸. Of course, some situations may require families
361 and medical providers to prioritize treatment for certain health issues (e.g., surgery for congenital
362 heart defect) over diagnostic audiology services. That said, our results raise the possibility that

⁸ 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).

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

365 Language background too predicted age at diagnosis, such that infants from Spanish-
366 speaking families were diagnosed 3.79 months later than infants from English-speaking families.
367 This may be due to cultural differences in attitudes towards deafness (Caballero, Muñoz, Schultz,
368 Graham, & Meibos, 2018; Rodriguez & Allen, 2020; Steinberg, Bain, Li, Delgado, & Ruperto,
369 2003) or a lack of linguistically accessible and culturally appropriate audiology services. Only
370 5.6% of American audiologists identify as bilingual service providers (ASHA, 2019), and
371 services from a monolingual provider may be insufficient, particularly in obtaining the child's
372 case history and providing recommendations for follow-up services (Abreu, Adriatico, &
373 DePierro, 2011).

Intervention. As expected, more severe hearing loss predicted earlier intervention.

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

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

384 Finally, it's important to note that this sample is composed of children receiving birth-to-3
385 services. Less than 40% of our sample of children in early intervention meet the 6-month EHDI
386 benchmark. Given that only about 67% of children with hearing loss enroll in early intervention
387 services (CDC, 2018), our data suggest that the actual proportion of DHH children who receive
388 intervention by the EHDI-recommended 6 months may be closer to 26%. These children may not
389 receive clinical support until school-age or later, exacerbating concerns for language
390 development, which lays an important foundation for literacy and academic success (Hemphill &
391 Tivnan, 2008; Stæhr, 2008).

392 **Educational and Clinical Implications**

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

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

404 Additionally, the vast majority of children in our sample experienced vocabulary delays
405 (relative to hearing peers), and studies of spoken vocabulary development in older DHH children

406 suggest that they may not catch up (Lund, 2016). This should set clinicians and educators on high
407 alert. As early intervention predicts vocabulary outcomes in study after study (including this
408 present study and e.g., Vohr et al., 2008; Ching, Dillon, Leigh, & Cupples, 2018), ensuring
409 intervention by 6 months for all DHH children may be one way to address spoken vocabulary
410 deficits. Another option may be the provision of structured, accessible language input (i.e., sign
411 language) even prior to intervention or amplification, potentially mitigating negative effects of
412 auditory deprivation on language skills (Davidson, Lillo-Martin, & Pichler, 2014; Hassanzadeh,
413 2012). While learning sign language may pose a challenge for some families for myriad reasons
414 (as underscored by its absence as a communication modality within our sample), we nevertheless
415 highlight its potential as an important language support for DHH children and their families.

416 **Limitations and Opportunities for Future Work**

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

420 Given our exploratory analyses, there were many possible analytic routes. We encourage
421 interested readers to explore further analyses using the data and/or code provided on our [OSF](#)
422 [page](#).

423 This sample is composed only of children in North Carolina. While certain factors vary by
424 country and by state (e.g., diagnosis and early intervention practices; NAD, n.d.), our sample
425 largely resembles the national DHH population in terms of degree of hearing loss, percentage of
426 children with additional disabilities, cochlear implant and hearing aid use, language background,
427 and gender (Blackorby & Knokey, 2006; Gallaudet Research Institute, 2014). It did diverge from

428 the national sample in communication modality: our sample had no signers while 20% of DHH
429 children have sign as their primary modality (Gallaudet Research Institute, 2014). A similar
430 naturalistic study in regions where sign language access for DHH children is more common
431 (e.g. Washington D.C.) would be a welcome addition to the present work, in illuminating the
432 effects of different clinical and demographic factors in a signing population. One further
433 limitation to our analyses and to assessing representativeness of the sample is that race and
434 socioeconomic status information was not available.

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

440 **Conclusion**

441 The present study explored interrelations among demographic and audiological
442 characteristics, vocabulary outcomes, and clinical milestones within a diverse sample of 100
443 DHH children enrolled in early intervention services in North Carolina. Our population-based
444 description underscores heavily interlocking demographic, audiological, and clinical
445 characteristics (e.g. communication approach and presence of developmental delays). Our models
446 highlight the outsized roles of age, amplification, and laterality relative to other predictors,
447 together accounting for over half of variance in productive vocabulary. We also explicitly
448 examined the roles of prompt achievement of early intervention milestones on vocabulary. We
449 found that overall, this sample showed vocabulary delays relative to hearing peers, and room for

450 improvement in rates of early diagnosis and intervention in particular. This in turn highlights
451 potential paths forward in ensuring that regardless of hearing status, we are able to provide
452 language access and early childhood support to help children attain their potential.

453 **Acknowledgement**

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

456

457

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- 632

633

Captions

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

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

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

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

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

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

660 **Table 2:** Detailed information about the variables studied. For categorical variables, levels
661 are described and Ns provided. For continuous variables, range, mean, and standard deviation are
662 provided.

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

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

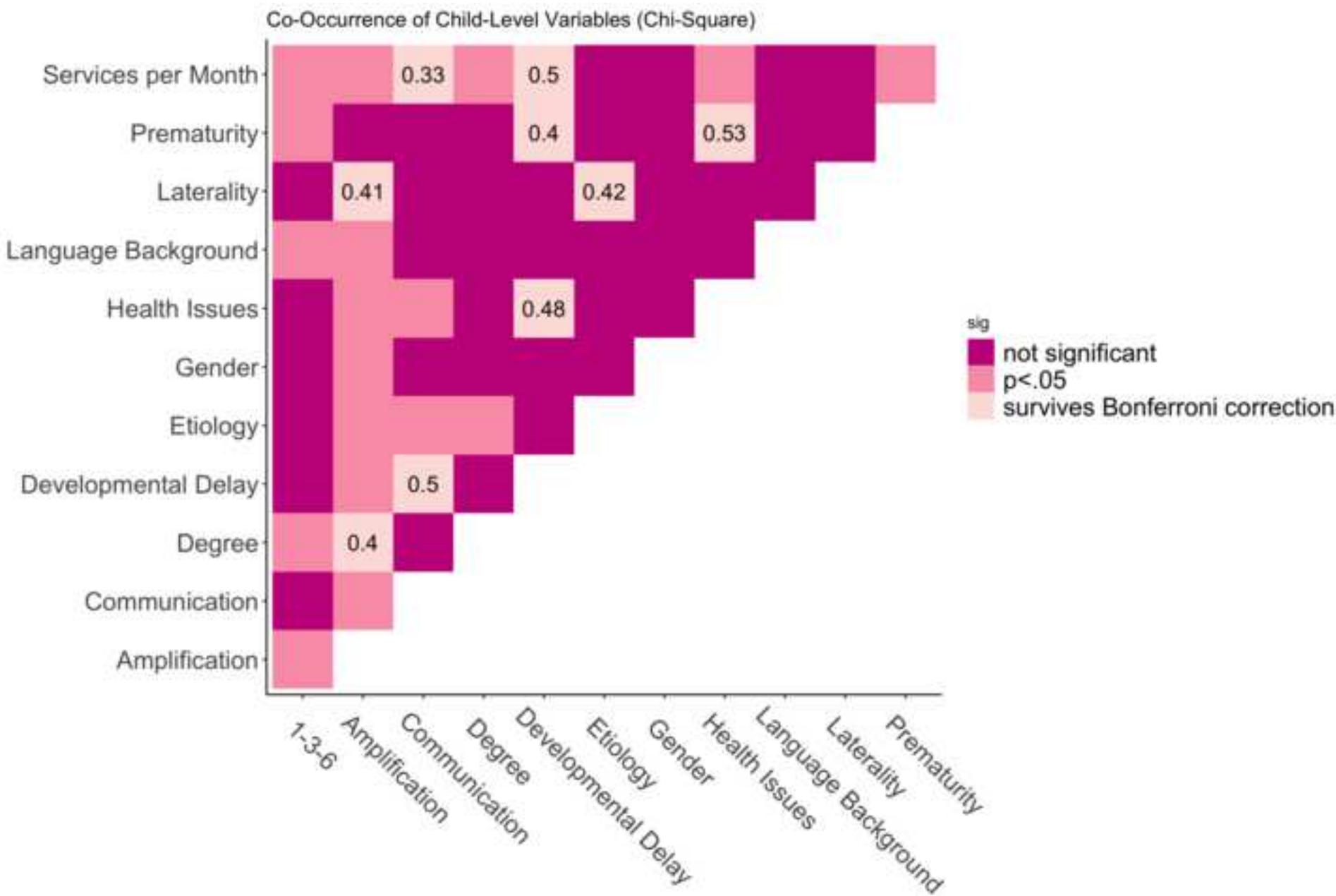
671 **Supplemental Materials S3:** Language and Communication Characteristics of the
672 Sample: Ns of participants by language background and communication method.

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

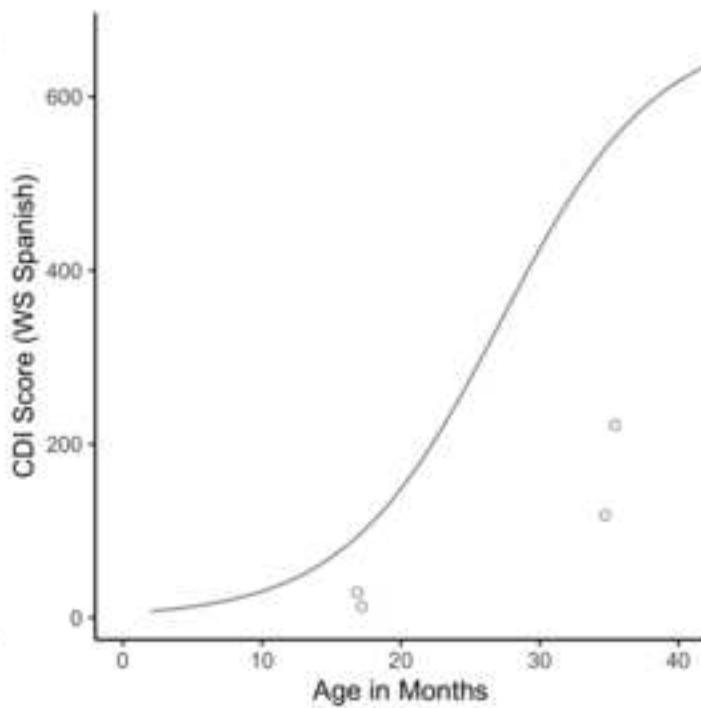
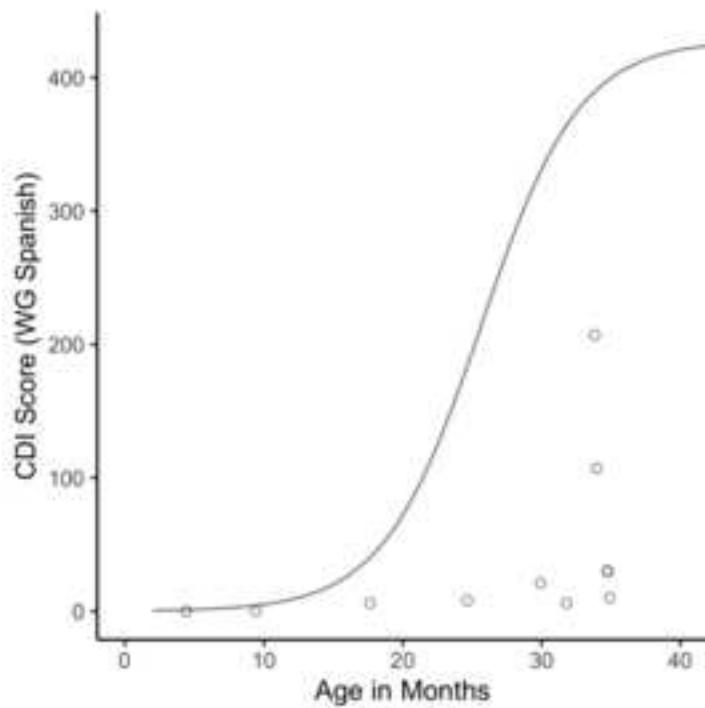
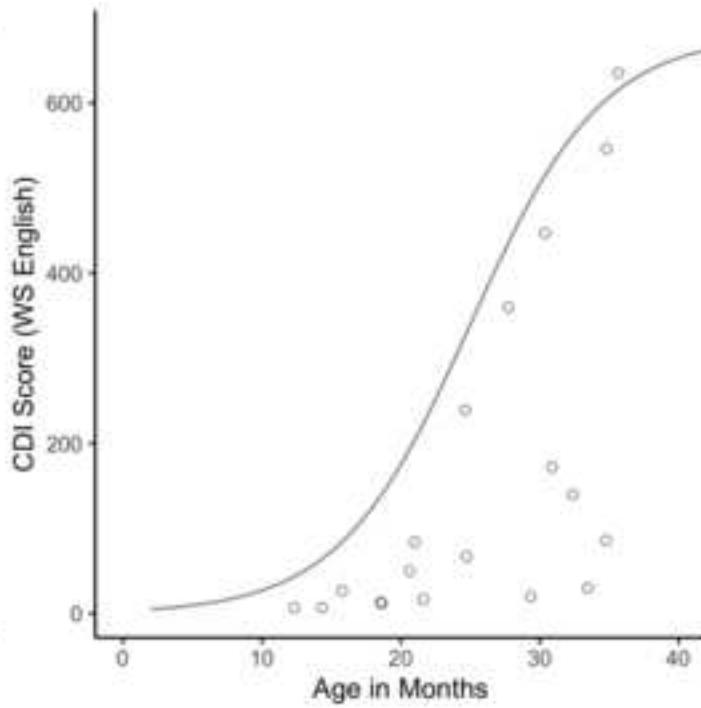
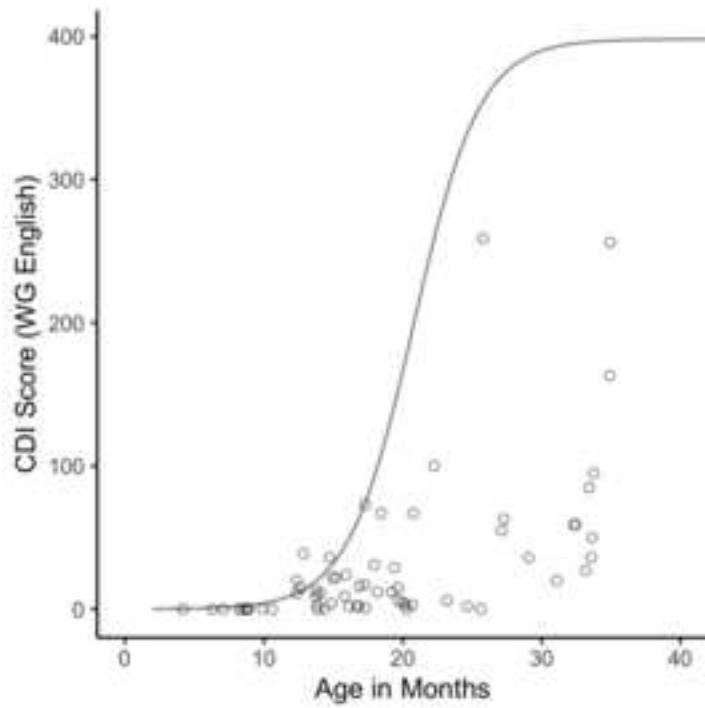
675 **Supplemental Materials S5:** Unstandardized beta coefficients (months) for the model of
676 age at diagnosis selected by AIC.

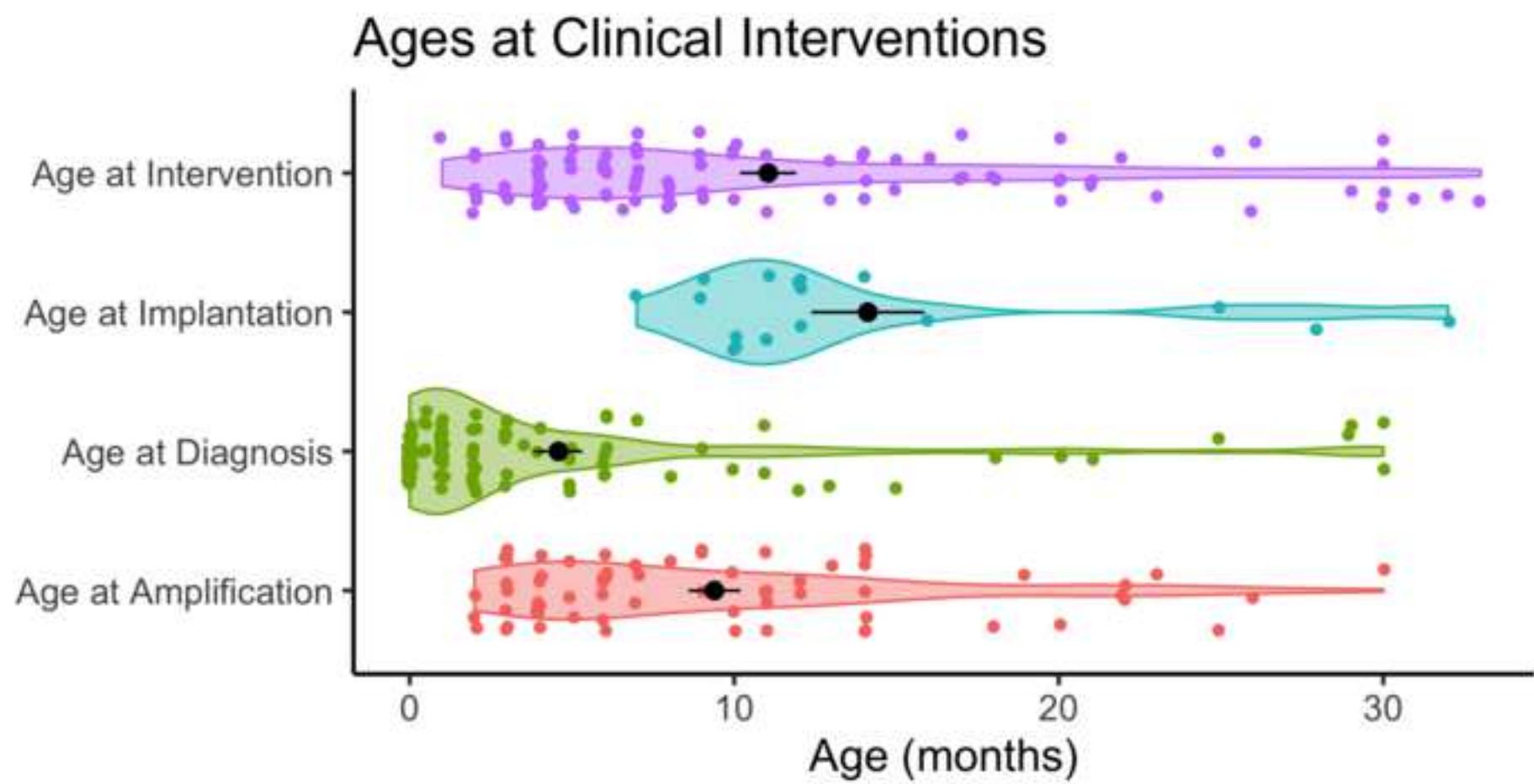
677 **Supplemental Materials S6:** Unstandardized beta coefficients (months) for the model of
678 age at intervention selected by AIC.

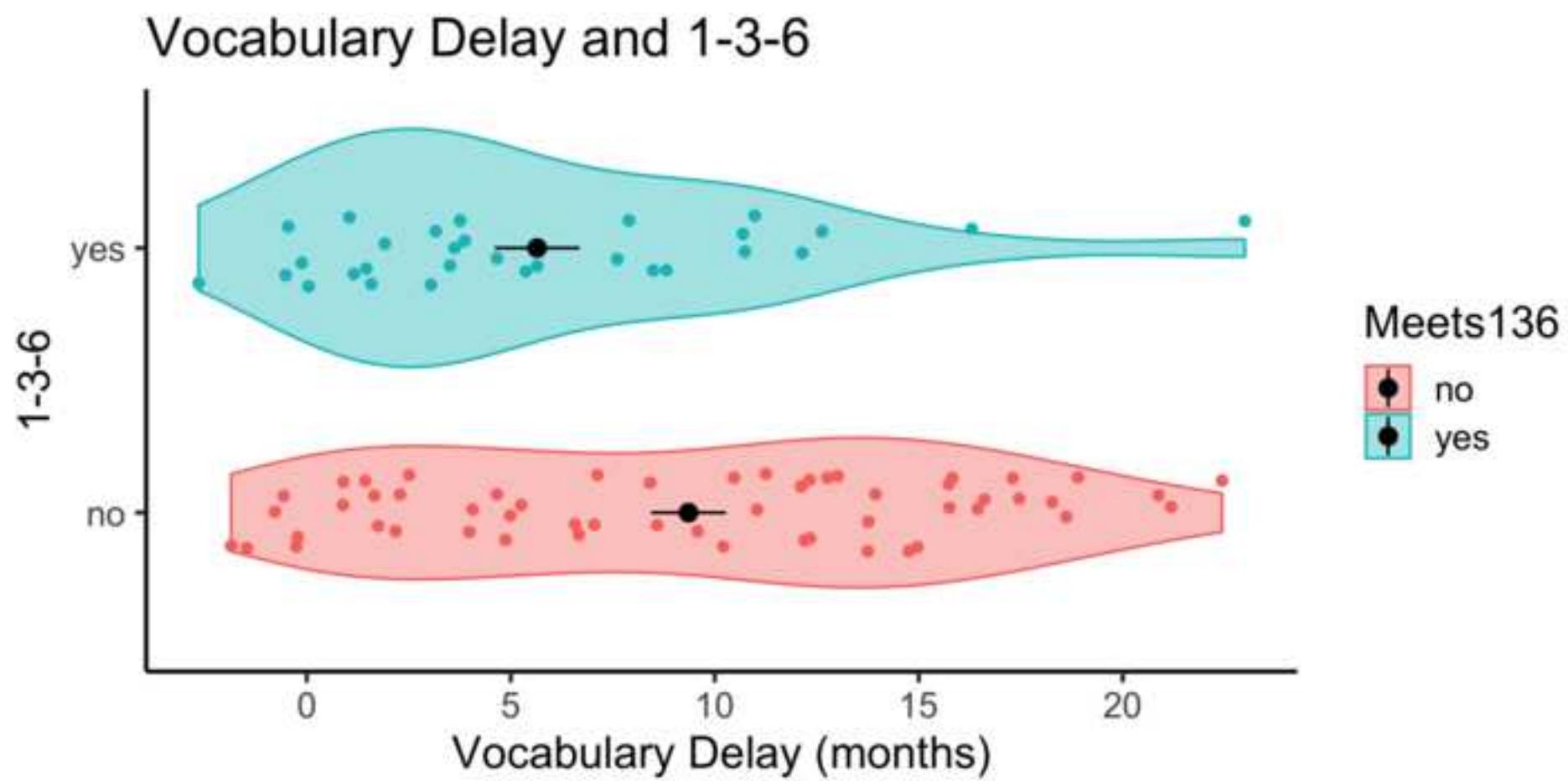
679 **Supplemental Materials S7:** PDF containing all figures and tables with their captions in
680 situ (for reader/reviewer ease).



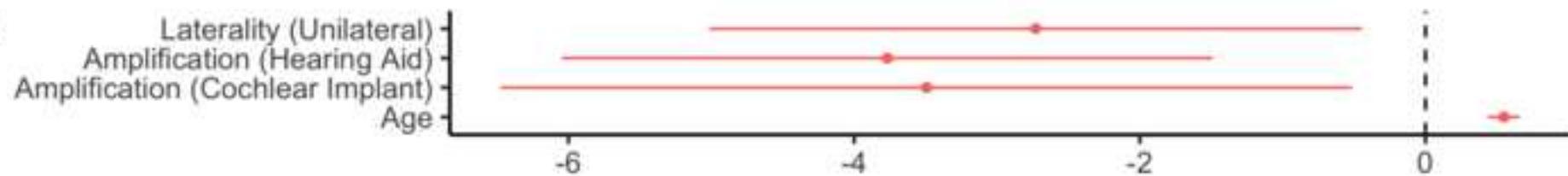
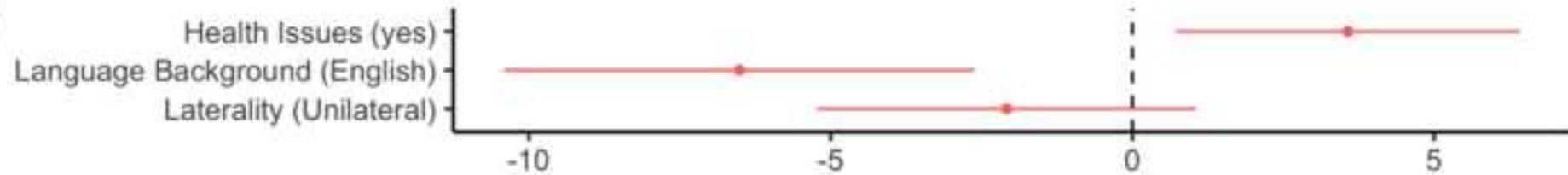
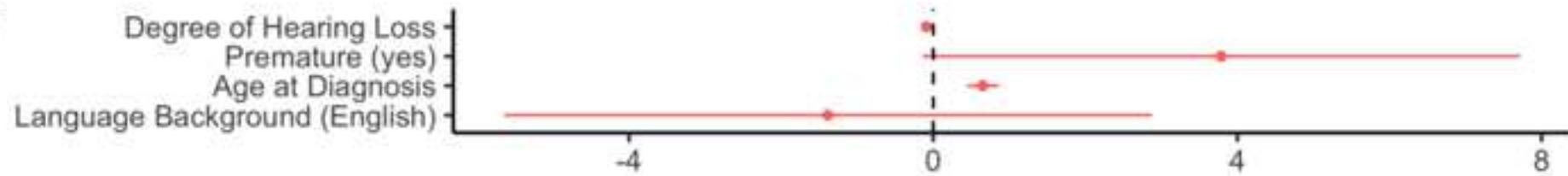
Vocabulary Growth Curves by Instrument







Beta Weights for Vocabulary, Diagnosis, and Intervention Models

A**B****C**

CDI version	Mean Age (SD	Mean Compre	Mean Word P %	Developmental Delays
WG (n = 74)	20.1 (8.9)	mor 107 (99)	word 32 (53)	words 17.6%
WS (n = 24)	25.8 (7.7)	mor NA	149 (180)	wor 4.2%

Variable	Range
Age	4-36 months (mean (SD): 21 (9))
Age at Amplifi	2-30 months (mean (SD): 9 (7))
Age at Diagno	0-30 months (mean (SD): 5 (7))
Age at Implan	7-32 months (mean (SD): 14 (7))
Age at Interve	1-33 months (mean (SD): 11 (9))
Amplification	Hearing Aid (53) / Cochlear Implant (17) / None (27)
Communicativ	Spoken (78) / Total Communication (18) / Cued Speech (1)
Degree Hearir	18-100 dB HL (mean (SD): 65 (23))
Development:	Yes (16) / No (81)
Gender	Female (44) / Male (56)
Health Issues	Yes (35) / No (62)
Language in H	English (84) / Other (16)
Laterality	Unilateral (25) / Bilateral (72)
1-3-6	Yes (35) / No (59)
Premature Bir	Full-term (16) / Premature (81)
Services Per M	0-43 services per month (mean (SD): 6 (6))
Etiology	Sensorineural (61) / Conductive (18) / Mixed (8)
CDI - Words P	0-635 words (mean (SD): 61 (111))

Dear Editor,

Thank you very much for taking the time to consider our manuscript for publication at JSLHR and for allowing our resubmission. We were glad to hear that the reviewers thought that the study topic is “very interesting and relevant” (R2) and had “interesting findings” (R1), that the results section was “clearly written” (R1), and that our analyses were informative and appropriate. We are very grateful to you and the 2 reviewers for providing many helpful suggestions for reframing, shortening, and clarifying our manuscript.

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*.

We note that while we wholly agree with the majority of feedback, in a few cases, reviewers made suggestions not about the scientific content but about wording choices or what’s better handled in text vs. table. We largely took reviewers’ suggestions, but in a few cases opted to retain the original wording or format; we are open to Editor feedback on these points.

Finally, to facilitate review and minimize portal download headaches, in addition to following the JSLHR guidelines for table and figure formats, we’ve added a supplementary pdf that includes all figures, tables, and supplementary tables in one place with their titles and captions.

We hope that you will find that the revised manuscript is clearer and more concise than the original. We thank you and the reviewers for your helpful and thoughtful comments.

Editor Comments:

Two expert reviewers and I have read your manuscript. We share several major concerns about the work in its current state.

- 1. First, there are concerns about what is added to the literature. A more complete justification and adjustment of how space is used in the introduction may help alleviate this concern.**

RESPONSE: We have significantly overhauled the introduction in light of this feedback; see replies 5 and 10 below.

- 2. Second, there are major concerns about the length. I think one possible solution is to move some of the detail of the results to supplemental material, and eliminate much of the irrelevant information, largely in the discussion and several figures.**

Some relevant info is presently located in supplemental material that instead is vital to readers' understanding. Reorganizing the manuscript extensively is necessary for readability.

RESPONSE: As detailed below, we have made cuts and thoroughly reorganized the manuscript, particularly in the discussion section, and in the section regarding child-level variables. We have retained the Figures but have clarified their significance in our streamlining of the results. We have not added further content to the supplementals as it became clear that we'd inadvertently left content out of our initial submission (S1-3, which were formerly labelled S2-4) and did not want to extend the supplemental materials further (to this end we did take R1's suggestion to drop Table S1). We hope to have struck a better balance of detail and brevity.

- 3. Third, and perhaps most importantly, the focus of the article, in my opinion, doesn't quite highlight the interesting findings. Both reviewers question the contribution to the literature, and I think that shifting the focus to some novel or unexpected findings would really enhance the paper. In particular, I suggest that a focus on the 1-3-6 piece could address this concern. The reviewers both provide important critiques and suggestions as well.**

RESPONSE: We agree that we had insufficiently highlighted the interesting findings, and that the 1-3-6 component is an important part of our results. Along with addressing the reviewers' comments below, we have added a paragraph in the introduction to promote the contribution of the section regarding 1-3-6, which we have included in the response to comment 8.

Reviewer #1 Comments:

- 4. Your study had interesting findings related to the language outcomes of children who are Deaf and Hard of Hearing for children within your state. This is a fantastic sample size that can nicely capture outcomes within your state.**

More studies that include a diverse sample of children who are DHOH are needed. This study is a piece in describing outcomes for children with multiple needs and are bilingual. It also demonstrates the importance of early identification and early intervention.

RESPONSE: We thank R1 for appreciating the size and diversity of our sample, and our results highlighting the importance of early identification and intervention for DHH children and their families.

5. The literature review did not set up the study well. In other words, after reading this section, I was not convinced that this study would contribute something new to the dearth of information related to DHOH outcomes.

RESPONSE: We have extensively reorganized the literature review to clarify that a central contribution of our study is to: identify predictors for delays in vocabulary, diagnosis, and intervention, within the context of the considerable diversity of the DHH population. This led to big changes through the introduction, as exemplified by this new text on lines 86-93: *"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)."*

6. The authors should consider using this section to discuss the process of early intervention referral and services in North Carolina as well as the demographics of communication modality choice that may be unique to the state. Your footnote on page four hints at this conversation.

RESPONSE: We now reiterate the potentially state-specific modality choice in the discussion on lines 424-433:

"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. "

RESPONSE: Given the editor's concerns about the article length, we have not added further NC-specific referral or demographic information, but are happy to add this in if R1 or the editor would like.

7. It would be beneficial to have a discussion related to how the sample collected represents children who are DHOH overall (e.g. 2nd language learners, LSL vs TC). I think this discussion should not be focused on your sample specifically but the trends in the country related to communication modalities and demongraphics

RESPONSE: We have added information about demographic trends in the discussion on lines 424-430:

"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)."

8. You focused on 1-3-6 guidelines in your analysis. This is significant when considering child outcomes. A brief summary of the guidelines and previous research would better support why you examined this variable.

RESPONSE: We have added a brief summary of the 1-3-6 guidelines and previous research (now on lines 62-76):

"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 identify children at risk of receiving clinical support late, in order to help all children achieve prompt diagnosis and intervention."

9. I understand that manuscript space is limited. My suggestion would be to remove the discussion of the CDI. This would be more appropriate, in a more summarized form, in the methods section. The purpose of the paper was not to analyze the CDI and its validity, but to describe a certain population and their outcomes. If the paper was about the CDI, then it would be appropriate to have this discussion in the literature review.

RESPONSE: We thank R1 for this helpful suggestion. We have shortened the explanation of the CDI and moved it to Methods:

"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. 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.”

10. Focus on WHY the paper was written and how it contributes to the limited information related to the outcomes of children who are DHOH

RESPONSE: We removed portions of the introduction that were not relevant to our “why”, and added a more explicit justification of the study on lines 77-84:

“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”

We also now clarify the specific contribution of each of our 3 goals (beginning line 94):
“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, meeting 1-3-6 guidelines, and presence of additional disabilities would predict larger spoken vocabulary delay. This study builds on prior work 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.”

11. In your 3rd hypothesis, you mentioned “based on the prior literature summarized above” . There was very little summary of these variables and how they are related to child outcomes. Consider either adding to the literature review discussion centered around the impact of these variables or rewording your hypothesis statement

RESPONSE: We have reworded our hypothesis statement as follows:

“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.”

12. How many families in ELSSP did not agree to participate? Report the acceptance percentage

RESPONSE: Unfortunately, our partner organization did not track this information systematically, although our liaison reports that they're not aware of families who did not consent to participate when asked by their individual Teachers of the Deaf, who then passed the deidentified data along to our liaison. We have not added this to the text at present, but would be glad to add a footnote to this effect if the Editor or reviewer feels this information is useful to the reader given that we are unable to precisely quantify it.

13. Was the CDI just spoken language or were signs also accounted for?

RESPONSE: Clinicians also reported the number of signs reported though this only applied to the 18 children who used total communication (none exclusively signed and the rest used only the spoken modality). We have added this information to Table 1, and on lines 140-141, we added text to clarify:

"Both spoken words and signs counted as word productions."

14. If early intervention is birth to 36 months, how were some children older than 36 months? (line 131)

RESPONSE: We thank the reviewer for catching this! We discovered a small error in how the Age column was rounding days in a month. We have adjusted the data preparation script to address the problem, which resulted in a mean difference of 0.18 months between the ages reported in the original manuscript and the revised manuscript; no children were >36mo.

15. Table 1 does not add significant information. This can be added to the text and not documented in a chart.

RESPONSE: We respectfully disagree that putting numeric information regarding the number of words children understand and produce, and the percent of children with developmental delays for each instrument is easier to understand in text form than in table, and have retained Table 1.

16. Table 2- is not necessary to report the type of variable (continuous vs categorical). The ranges can be reported within the text of the methods section.

RESPONSE: We removed the "type of variable" column per R1's suggestion, but opted to keep the ranges in the table, rather than in text, for readability.

17. Supplementary material 1- this study was not a metanalysis, this chart does not seem necessary

RESPONSE: We have removed Table S1 from the supplemental materials.

18. I do not have access to S2-4, and cannot comment on them

RESPONSE: We are terribly sorry for this error on our part in the upload portal. We have confirmed that all supplementary tables are included for the revised resubmission.

19. Clearly written results section. I appreciated the organization and the reminder of the purpose at the start of each section.

RESPONSE: We thank R1 for their kind words regarding the Result section's clarity.

20. Line 197- A better explanation of what WordBank norms are is warranted. It is difficult to make comment/ draw conclusions on how the results, using this value, that were derived without further explanation. Was the following paragraph (starting at line 199) an explanation of this? If so, this is unclear.

RESPONSE: We have added a brief explanation of the Wordbank norms to the Methods on lines 128-130:

"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."

21. In the predictors of vocabulary section- when talking about vocabulary delays, was this just spoken language? Should be very clear that a child can have vocabulary, but may not be in spoken language.

RESPONSE: We fully agree. Our calculation of vocabulary delay includes both spoken and signs in the vocabulary production score from the CDI. But to be clear, given the rarity of non-spoken language only participants, very few signs are included in the group-level vocabulary means. We have clarified the text throughout to reflect this, particularly in the Table 1 caption (new text bolded):

*"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)."*

22. There were several variables put into the regression model and discussed in previous sections that are not again mentioned. For example, were there any significant findings for gender and how it relates to a vocabulary delay?

RESPONSE: We thank R1 for pointing out this oversight. We have added text on line 227-228 to clarify:

"No other variables from the full model above significantly improved model fit, and are thus not discussed further."

To address the reviewer's specific question, gender did not improve model fit for predicting vocabulary delay. Given that discussing these variables further was not statistically justified, we do not mention them further in the manuscript, but if R1 had any specific requests or suggestions regarding gender, we'd be glad to add information to the Supplemental Materials.

23. When considering vocabulary delay, why was communication modality not considered? This would make a significant impact on spoken language / overall language use.

RESPONSE: We're not sure we understand the Reviewer's concern here. Communication modality was included in our models of vocabulary delay, but did not emerge as one of the variables in the best-fitting models (similar to gender in the preceding comment).

Perhaps Reviewer 1 is asking whether the vocabulary delay model results differ by communication modality? Currently, the model uses total vocabulary production (spoken + sign) as the outcome variable. But to look into R1's question (if we've understood it correctly), we reran our models using only spoken words (versus spoken+sign) as the DV. In this model, total communication does become a significant predictor of vocabulary delay; its use is associated with an 8 month larger delay than using spoken language.

But given that (a) total communication was not randomly assigned to children (and as our results show, more common for those with developmental delays; see comment 46) and (b) that only "counting" spoken vocabulary is not representative of children's full communication skills, we opted to use spoken+sign to calculate and predict vocabulary delay (but clarify that this is what we're doing as noted in comments 13 and 21).

24. Several great clinical points were made in this section. You nicely summed up your findings and made them relevant clinically and for a bigger picture (legislative changes that may need to be made)

RESPONSE: We're glad R1 felt that this part of the paper was clear and relevant to the bigger picture.

25. While I personally agree with the discussion related to using sign language as a foundation for language prior to spoken language, it seems to go beyond the scope

of the current paper to make these claims. I think it is a point worth mentioning, but maybe more in passing, rather than such a full discussion

RESPONSE: We have shortened this section as follows:

"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."

26. As clearly said a few times in the child-level variables sections (results and discussion), the relationships were not surprising or contributory to the findings of this study. It seemed like this section could be significantly reduced.

RESPONSE: We have significantly shortened the sections on child-level variables, both the results and especially our discussion of them, which, as R1 observes, was somewhat redundant. We hope R1 now finds the space allotted to them more appropriate.

Could the manuscript benefit from the addition of supplemental material?

Reviewer 1: No:

Is additional information regarding the research methodology needed to replicate the study?

Reviewer 1: No:

27. Use people first language - children who are DHOH

RESPONSE: Our understanding from practitioners, clinicians, Teachers of the Deaf, and families with children with hearing loss is that the Deaf/Hard-of-Hearing community often uses identity-first language, and indeed identity-first language is common in literature about children and individuals in this community (e.g., Terhune-Cotter, Conway, and Dye, 2021; Carrigan & Coppola, 2017). We also believe our usage complies with JSLHR's author guidelines, which specify that "ASHA adheres to the style guide of the American Psychological Association (APA) in using person-first or identity-first language to describe attributes and diagnoses of individuals or groups of people." We thus opt to retain identify-first language in this manuscript.

Writing Suggestions:

RESPONSE: We thank Reviewer 1 for taking the time to comment on our word choice and writing style. We have retained some of our original wording in instances where it seemed like the stylistic critique might be idiosyncratic to Reviewer 1.

28. Line 52- Dont start a sentence with the word "however".

RESPONSE: We reworded this sentence:

"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)."

29. Line 52- Dont use the word "excellent" this term is a vague descriptor and the author shouldnt put judgement on the quality of studies without discussing evidence (which I am sure they didnt want to do in this case).

RESPONSE: We reworded this section which now reads:

"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."

30. The footnote on page 4 was an odd placement. Should be included in the methods section.

RESPONSE: We agree that it was oddly placed, but respectfully disagree with the suggestion to move it to the methods. To us, it seems critical to let the reader know early on what the scope of the work is (and why our references to prior work focus primarily on spoken language). That said we have substantially shortened this footnote to make it more appropriate to its placement in the introduction (where it now appears at the end of the first paragraph):

"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. Given this, we focus primarily on spoken language development."

31. Line 120- "passed along" is informal language, consider revising

RESPONSE: We changed "passed along" to "sent":

"ELSSP sent deidentified evaluations to our team after obtaining consent to do so from each family"

32. Line 122- switch the clause order. Start with "Given our goal..." Some people call this "upside down subordination"

RESPONSE: The sentence now reads:

"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."

33. Line 135 "for whom Spanish" is awkward wording. Consider "Families, whose primary language was Spanish,

RESPONSE: We implemented reviewer's wording suggestion here:

"Families whose primary language was Spanish (n = 15) completed the Spanish language version of the CDI (Jackson-Maldonado et al., 2003)."

34. Footnote 5- there is a typo

RESPONSE: Fixed.

35. Line 257- dont start a sentence with a number, use the word form

RESPONSE: We reworded this sentence so that it no longer begins with a number.

"In our sample, 70% of children received diagnosis by 3 months of age, and 40% began early intervention by 6 months of age (see Figure 3)."

36. Line 267 - "next zoomed in on" is informal language, consider revising

RESPONSE: We revised this sentence:

"To better understand implementation of 1-3-6 guidelines, we next turned our focus to factors influencing the timing of diagnosis and intervention."

37. Line 301- don't end a sentence with "within", its a preposition

RESPONSE: We respectfully disagree with R1's suggestion, and have retained the original wording::

"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 examined in the full heterogeneity they naturally occur within."

38. Line 347- awkward sentence. I would reword to say "amplification devices were commonly used for children with more significant hearing loss"

RESPONSE: Done.

39. Line 388 this is an odd citation. Was this intentional?

RESPONSE: No, this was due to a formatting error. We have addressed the formatting of this citation, and thank R1 for noticing.

40. Line 398- was the use of "dint" purposeful? Odd wording in this sentence

RESPONSE: Yes, this was the intended wording. We've reworded this sentence, but retain that word:

"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."

41. Line 510- other factors... such as? (give examples)

RESPONSE: We now provide examples of relevant factors:

"Finally, the considerable variability in the sample did not allow us to easily isolate effects of different factors (e.g., degree vs. amplification)."

42. Line 293- awkward phrasing : Taken together these analyses reveal that beyond aspects of the 293 child's hearing status, other variables too contribute to delays in both diagnoses and intervention. 294 We return to this point in the discussion.

RESPONSE: We have changed wording to:

"Taken together, these analyses reveal that children's audiological characteristics, comorbid diagnoses, and language background contribute to delays in both diagnoses and intervention."

43. Line 441- need a citation for AAP estimate statement

RESPONSE: In shortening the discussion, this statement was removed.

44. The figures and tables were not in APA format. For example, all were missing a title, making it difficult to decipher the purpose of the document. Figures often have notes to explain the information.

RESPONSE: We thank R1 for noting this oversight; while captions were at the end of the document per JSHLR instructions, titles were accidentally omitted. We have reformatted all figures and tables in accordance with APA guidelines.

45. There were several figures and tables that seem to be unnecessary. For example, I am not understanding the value of figure 4. It seems this information could be reported within the body of the text. Maybe further explanation is warranted if the authors feel it is valuable.

RESPONSE: As noted in our agreement with the Editor above, the implications for 1-3-6 guidelines are an important contribution of our work. To this end, Figure 4 provides a critical

visualization of our results. To R1's point, we have included an explanation in the caption to make the result clear:

"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."

And we also underscore the importance of this contribution within our results in lines 352-357:

"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."

46. Can information within the supplemental material be combined into 1 chart? This would make the information easier to access.

RESPONSE: Unfortunately, combining this information into one table rendered it harder to understand in our view. While the tables were split into separate Excel files per JSLHR's guidelines, to R1's point, we have added a supplementary document that includes all tables and their captions in one place as a pdf with the table contents pasted in, simply for easier reviewer viewing.

47. I am not able to view S2-4 and cannot make comment on them.

RESPONSE: We are terribly sorry for this error on our part in the upload portal. We have confirmed that all supplementary tables are included for the revised resubmission.

Reviewer 2

48. The study had three parts. First, there was a characterization of the variability in the audiological, demographic and intervention factors within a sample of 100 DHH infants and toddlers, and the inter-relatedness of these factors. Second, the authors calculated vocabulary delays and investigated predictors of the vocabulary delays. Third, the authors investigated predictors of variation in age of diagnosis and age of intervention.

A major strength of this study was that the data and code are available on the OFS ensuring transparency. The statistics and violin plots were very clear and informative.

The calculation of the vocabulary delay scores was a strength of the study and it was well explained.

A strength of the discussion was the frank and open discussion about the complexity of the results in the sense that the factors are all interrelated. Many authors try to simplify the story but the current authors should be applauded for not doing this.

RESPONSE: We thank R2 for their kind words regarding our data transparency, analyses, figures, and discussion of a complex set of results.

49. The paragraph at the top of page 17 makes a very important point about total communication and language delays. Could more be made of this?

RESPONSE: We have elaborated on this point on line 322-328:

"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. 9.88% 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)."

50. The study addresses important questions about predictors of language delay in DHH children in a population-based sample. However, was the sample size sufficient enough to be able to address the important questions appropriately?

RESPONSE: Certainly more data might help us better answer some of these questions, however we believe that our sample of 100 DHH children from a single state is sufficient for our models and analysis. We address this sample size question further in our response to comment 55.

51. The rationale for the study and the knowledge gap that it was filling was not very clear. This left me with the feeling that we already knew the results which makes it difficult assess the importance of the study.

RESPONSE: We agree with R2 that our study motivation was not sufficiently explained. We have added text to the introduction and discussion to clarify our rationale and novel findings.

In particular, we've added the following text to crystalize our contribution in the introduction before laying out our goals (lines 77-84):

"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."

We also now clarify the specific contribution of each of our 3 goals (beginning line 94):

"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, meeting 1-3-6 guidelines, and presence of additional disabilities would predict larger spoken vocabulary delay. This study builds on prior work 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."

52. The rationale for the study was not clear. I was left with the feeling that we knew the findings already. However, I am not sure that is correct but because of the way the introduction was written, there was little detail about the background. The manuscript stated that the literature review had been moved to supplemental materials. I searched but I was unable to locate these materials. Regardless of that, without including sufficient background information in the introduction, the rationale for the study is not clear. What is the knowledge gap that the study is trying to fill? This made it difficult to see how the results added to what we already know from the literature, as described in the paragraph at the bottom of page 3. Is the rationale that these factors haven't been investigated in one sample before? Why was a fuller overview of the literature included in the supplemental materials? Was it because of the length of the manuscript? If so, there was a lot of focus on the CDI in the introduction and that space could have been spent on the background overview.

RESPONSE: We thank R2 for articulating sections where we were insufficiently clear in our rationale. We have done a major overhaul of the introduction, highlighting that the factors in our models have not been studied together in one sample before, as R2 suspected. We removed extraneous information about the CDI from the introduction and moved it to Methods (or when not critical, removed it altogether).

In addition to the changes regarding rationale noted in the preceding reply (51), we have also extensively reorganized the literature review to clarify that a central contribution of our study is to: identify predictors for delays in vocabulary, diagnosis, and intervention, within

the context of the considerable diversity of the DHH population. This led to many changes through the introduction, as exemplified by this new text on lines 86-93:

"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)."

53. There was missing information about the participants. How many had comorbid factors?

RESPONSE: Seventeen participants had a developmental delay, sixteen participants were born premature, and thirty-six participants had a health issue. There was a lot of overlap across these three categories, so thirty-nine participants total had any of these comorbidities. We have added this information to Table 2.

54. Table 2 contains a list of information about the background/audiological factors. The range and mean/SD were included for the continuous factors. But the categorical categories had the category labels only and no frequency information. This information is crucial to be able to understand the characteristics of the sample.

RESPONSE: We have added this information to Table 2.

55. The sample is representative on the one hand because it is a population based study – but on the other hand, it has a very small sample size for a population-based study. So what does this add to our knowledge over what we know already from larger scale cohort population studies? What was the rationale for stopping at 100? Was a power analysis conducted to explain the stopping point? If the data were coming from an ongoing collaboration, why not continue to collect more data which would allow more factors to be investigated? Is 100 sufficient to say it is a representative sample? Was it stratified across factors before stopping at 100?

RESPONSE: We were sorry to see that R2 disagreed with R1's comment that "This is a fantastic sample size that can nicely capture outcomes within your state." Given that part of our goal was to characterize a generally under-described 'take all comers' type of sample in which the degree of interaction across variables was challenging to quantify in advance, we did not conduct an *a priori* power analysis, or do a preregistration (which is our preferred approach).

Given that we aimed to collect all eligible infants receiving services through the Early Language Sensory Support Program, we were not able to stratify across factors. Our sample size was determined pragmatically: we established our partnership with the program in 2018, and were fortunate to receive data from 2010–2020. In stopping at 100, we were trying to balance collecting a sufficient sample size with disseminating the results in a timely manner. Unfortunately, continuing to collect more data is not a viable approach, as this sample is entirely pre-COVID-19; adding newer data would introduce potential effects of COVID-19, which is outside of the scope for the present study.

We do note that a sample size of 100 is, generally speaking, large for a sample of CDI data from DHH children (e.g. Thal et al., 2007; Stika et al., 2015; Castellanos et al., 2016 all have Ns from 24-32). The more population-oriented studies R2 alludes to generally span a far wider geographic region, adding further factors for consideration, as state laws regarding services and support vary widely (e.g. Yoshinaga-Itano et al 2017 have a sample of >400 infants, but span 12 states). Taken together, our sample of 100 DHH children from a single state is dense relative to prior work, and sufficient for our modeling and analysis approach.

We now clarify that all data in the current analysis were collected pre-COVID:

"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)."

56. Data exclusions – 5 children who had missing data for 1-3-6 and 13 children had missing data for degree of deafness were excluded. Why were 13 excluded rather than 18? Line 227

RESPONSE: To answer the reviewer's question, several participants had missing data for both 1-3-6 and degree. We have added this information to the footnote on line 224 to clarify.

57. JSLHR has an international audience, please explain 1-3-6.

RESPONSE: We have added a brief explanation of 1-3-6 on lines 68-76:

"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 identify children at risk of receiving clinical support late, in order to help all children achieve prompt diagnosis and intervention."

58. Not having race or SES information is a limitation - is it possible that the identified predictors might have been acting as a proxy for race or SES?

RESPONSE: We agree that this is a limitation and have acknowledged this on lines 433-435:

"One further limitation to our analyses and to assessing representativeness of the sample is that race and socioeconomic status information was not available."

We don't feel like the present data provide appropriate grounds to speculate as to whether other factors may have acted as a proxy for race or SES without evidence but agree these are important variables that we wish we had direct access to for analysis.

59. The results section was well explained but it was very lengthy.

RESPONSE: We have made cuts wherever possible in the Results section..

60. The models were detailed and appropriate and the process was described clearly.

RESPONSE: We thank R2 for this feedback and are glad the models were clear.

61. Why were the tables in Excel sheets that had to be downloaded rather than being in APA format?

RESPONSE: Our apologies for the formatting difficulties: excel sheets are one of the options JSLHR provides for sharing tables; we too prefer these to be in situ and next to their corresponding title and caption! We have reformatted the figures and tables in accordance with APA guidelines. To R2's point, we have added a supplementary document that includes all tables and their captions in one place as a pdf with the table contents pasted in, simply for easier reviewer viewing.

62. My main concern with the paper in its current format is that after reading it several times, I am not entirely sure what the paper is telling me in terms of findings, but I think that might be because of the lack of a clear rationale in the introduction.

RESPONSE: We have reframed the introduction to better set the stage for our paper's contributions, which are exploring predictors of vocabulary delay and timing of diagnosis and intervention in a diverse sample of DHH children receiving state services (please see reply 51 and 52 where new text is pasted regarding rationale).

We have also reworded portions of the Discussion and Conclusions to highlight the findings, most germanely lines 432-453:

"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."

63. The discussion section was very lengthy and difficult to follow in parts because it goes off topic in several points and does not stick to discussing the results of the study. I read the paper and the discussion several times to try to make sense of the implications of the main findings.

RESPONSE: We have removed or reduced several extraneous sections and hope that the reviewers find the implications clearer. We've also reworded our summary of the main findings in Conclusions (as pasted in our reply to the preceding comment, 62).

64. The third paragraph on page 17 was about how researchers could construct samples – the argument didn't seem to follow on logically. If the target subpopulation represents 14% of a population, why would trying to recruit a sample of 32 be difficult? I didn't follow this argument as many studies achieve these sample sizes.

RESPONSE: We agree with R2 that this was confusingly put in our initial submission. We have reworded this section as follows:

"If a researcher desires to collect 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."

65. On Page 18 – third paragraph was about spoken vocab delays increasing with age – this is not a novel finding to warrant so much focus.

RESPONSE: We have shortened this section as follows:

"In contrast to our predictions, the best model predicting vocabulary delay had just a few variables: age, amplification, and laterality. Notably, we see that the spoken vocabulary delay widens with age, indicating that the rate of spoken vocabulary acquisition is slower for DHH children."

66. I agree that the authors should mention the advantages of sign language but including a whole page dedicated to it when it is not mentioned in the study doesn't seem in keeping with the rest of the study.

RESPONSE: We have significantly shortened this section:

"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."

Could the manuscript benefit from the addition of supplemental material?

Reviewer 2: No:

Is additional information regarding the research methodology needed to replicate the study?

Reviewer 2: Yes: See comments under section 4.

67. The topic is very interesting and relevant. I think it could have potential but there are some significant weaknesses in the current format. The main issues were that the manuscript is simply too long in its current format with too much relevant information contained in the supplemental materials (which were not even all accessible).

RESPONSE: We're glad R2 felt the work has potential. We hope the cuts we made and the reshuffling of material from the supplementals into the main manuscript has allowed the key points and contribution to read more clearly. We thank R2 for their helpful feedback in improving the work.



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