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

Erin Campbell[1] & Elika Bergelson[1]

[1] Duke University

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Introduction

In the United States, 1-2 children are born with hearing loss, per 1,000 births (CDC, 2018). This translates to 114,000 Deaf or Hard of Hearing (DHH) children born in the U.S. per year (Martin, Hamilton, Osterman, & Driscoll, 2019). Of these 114,000, ~90% will be born to hearing parents (Mitchell & Karchmer, 2004), in a home where spoken language is likely the dominant communication method. Depending on the type and degree of hearing loss and whether the child uses amplification, spoken linguistic input will be partially or totally inaccessible. Some of these children will develop spoken language 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; Sarchet et al., 2014), which may later affect reading ability (Kyle & Harris, 2010) and academic achievement (Karchmer & Mitchell, 2003; Qi & Mitchell, 2012).

Despite many excellent studies examining language development in DHH children, there is still a gap in the literature describing and analyzing spoken language development across the full range of children receiving services for hearing loss, with many studies focusing in on specific subgroups (e.g. children under age X with Y level of hearing loss and Z amplification approach, e.g. Vohr et al., 2008; Yoshinaga-Itano, Sedey, Wiggin, & Mason, 2018). In what follows, we first summarize the previous literature on predictors of spoken language outcomes in DHH children. We then provide a brief overview of a common vocabulary measure used in the current study, the MacArthur-Bates Communicative Development Inventory (CDI). Finally, we turn to an empirical analysis of early vocabulary in a wide range of young children receiving state services in North Carolina. We have two broad goals in what follows. First, we aim to provide a comprehensive description of a heterogeneous group of young children who receive state services for hearing loss. Second, we aim to connect the intervention approaches and child characteristics of this sample with children's spoken vocabulary[1], with the broader goal of considering the success of early diagnosis and intervention initiatives.

Predictors of Language Outcomes

Though the literature points towards spoken language delays and deficits for DHH children, this is a highly variable population with highly variable outcomes (Pisoni, Kronenberger, Harris, & Moberly, 2018). Previous research indicates that gender (Ching et al., 2013; Kiese-Himmel & Ohlwein, 2002), additional disability (Ching et al., 2013; Verhaert et al., 2008; Yoshinaga-Itano, Sedey, Wiggin, & Chung, 2017), degree and configuration of hearing loss (Ching et al., 2013; de Diego-Lázaro, Restrepo, Sedey, & Yoshinaga-Itano, 2018; Vohr et al., 2011; Yoshinaga-Itano et al., 2017), amplification (Walker et al., 2015), communication (Geers et al., 2017), and early diagnosis/intervention (Yoshinaga-Itano et al., 2017, 2018) predict language outcomes in DHH children. We first provide a brief literature review on the effect of these predictors on language skills in DHH children.

Gender. For hearing children, the literature points to a female gender advantage in early language acquisition. Girls speak their first word earlier (Macoby, 1966), have a larger (Bornstein, Hahn, & Haynes, 2004; Fenson et al., 1994; Frank, Braginsky, Yurovsky, & Marchman, 2017) and faster-growing vocabulary (Huttenlocher, Haight, Bryk, Seltzer, & Lyons, 1991), and stronger grammatical and phonological skills (Lange, Euler, & Zaretsky,

[1] Despite 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.

2016; Özçalışkan & Goldin-Meadow, 2010). This finding appears to be consistent across studies (Wallentin, 2009), various spoken languages (Frank, Braginsky, Marchman, & Yurovsky, 2019), and gesture (Özçalışkan & Goldin-Meadow, 2010).

The DHH literature presents a more mixed (though rather understudied) picture. On one hand, DHH girls, like hearing girls, have been found to have a larger spoken vocabulary than DHH boys (Ching et al., 2013; Kiese-Himmel & Ohlwein, 2002). However, in contrast to their hearing peers, DHH children do not seem to show a gender-based difference for some aspects of syntactic development (Pahlavannezhad & Tayarani Niknezhad, 2014).

Comorbidities. Additional co-morbid disabilities occur frequently in the DHH population, perhaps as much as three times more than in the hearing population (Pollack, 1997). Incidence estimates for co-occurring disabilities in DHH children range from 25-51% (Bruce & Borders, 2015; Guardino, 2008; Holden-Pitt & Diaz, 1998; Luckner & Carter, 2001; Picard, 2004; Schildroth & Hotto, 1996; Soukup & Feinstein, 2007), with approximately 8% of DHH children living with 2 or more co-occurring disabilities (Schildroth & Hotto, 1996).

Some of these conditions, particularly those which carry risk of developmental delay (e.g., Down syndrome), result in language delays independent of hearing loss (Chapman, 1997; Kristoffersen, 2008; Weismer, Lord, & Esler, 2010). These effects vary by the nature of the specific disability (Cupples et al., 2014, 2018), with cognitive ability more predictive of language outcomes than presence or absence of additional disability (Meinzen-Derr, Wiley, Grether, & Choo, 2011; Sarant, Holt, Dowell, Richards, & Blamey, 2008). Disability and hearing loss likely each contribute to a given child's spoken language development (Ching et al., 2013; Rajput, Brown, & Bamiou, 2003; Van Nierop et al., 2016), with differential effects of each (Vesseur et al., 2016). In some cases, additional disabilities appear to interact with hearing loss to intensify developmental delays (Birman, Elliott, & Gibson, 2012; Pierson et al., 2007).

Furthermore, incidence of hearing loss is higher among children born premature (defined as < 37 weeks gestational age). Compared to an incidence of 0.2% in full-term infants, incidence of hearing loss in extremely premature infants (defined as < 33 weeks gestational age) ranges 2–11%, with increased prematurity associated with increased rates of hearing loss (Wroblewska-Seniuk, Greczka, Dabrowski, Szyfter-Harris, & Mazela, 2017).

Independently of hearing status, prematurity is linked to increased risk of language delay and disorder (Barre, Morgan, Doyle, & Anderson, 2011; Carter & Msall, 2017; Cusson, 2003; Rechia, Oliveira, Crestani, Biaggio, & de Souza, 2016; Van Noort-van Der Spek, Franken, & Weisglas-Kuperus, 2012; Vohr, 2014). Unfortunately, research on language development in premature DHH children is scant (Vohr, 2016), so it remains unclear how hearing loss and prematurity may interact within spoken language skills. One study of premature infants finds that auditory brainstem response during newborn hearing screening predicts language performance on the PLS-4 at age 3 (Amin, Vogler-Elias, Orlando, & Wang, 2014), suggesting a link between prematurity, hearing loss, and language development in early childhood, though further research is needed in this domain. In extremely premature DHH children, incidence of additional disabilities may be as high as 73% (Robertson, Howarth, Bork, & Dinu, 2009). Indeed, pre-term infants with comorbidities have been found to be more likely to also have hearing loss than those without comorbidities (Schmidt et al., 2003), further complicating language development for this population.

Audiological Characteristics. Hearing loss varies in severity, ranging from slight to profound (Clark, 1981). More severe hearing loss (less access to spoken language) typically results in more difficulty with spoken language in infancy (Vohr et al., 2008), early childhood (Ching et al., 2010, 2013; Sarant et al., 2008; Sininger, Grimes, & Christensen, 2010; Tomblin et al., 2015) and school-age children (Wake, Hughes, Poulakis, Collins, & Rickards, 2004). Although profound hearing loss is associated with more pronounced spoken language difficulty, even mild to moderate hearing loss is associated with elevated risk of language disorders (Blair, Peterson, & Viehweg, 1985; Delage & Tuller, 2007).

Hearing loss also varies in whether it affects one ear or both. Bilateral hearing assists speech perception, sound localization, and loudness perception in quiet and noisy environments (Ching, Van Wanrooy, & Dillon, 2007). The literature on hearing aids and cochlear implants also points to benefits for bilateral auditory input (Lovett, Kitterick, Hewitt, & Summerfield, 2010; Sarant, Harris, Bennet, & Bant, 2014; Smulders et al., 2016). At school-age, 3–6% of children have unilateral hearing loss (Ross, Visser, Holstrum, Qin, & Kenneson, 2010). Although children with unilateral hearing loss have one "good ear," even mild unilateral hearing loss has been tied to higher risk of language delays and educational challenges relative to hearing children (Kiese-Himmel, 2002; Lieu, 2004, 2013; Lieu, Tye-Murray, & Fu, 2012; Vila & Lieu, 2015). Just as in the bilateral case, more severe hearing loss leads to greater deficits in spoken language and educational outcomes for children with unilateral hearing loss (Anne, Lieu, & Cohen, 2017; Lieu, 2013).

Many DHH children receive hearing aids (HAs) or cochlear implants (CIs) to boost access to the aural world. These devices have been associated with better speech perception and spoken language outcomes (Niparko et al., 2010; Walker et al., 2015; Waltzman et al., 1997). In turn, aided audibility predicts lexical abilities in children with HAs (Stiles, Bentler, & McGregor, 2012).

For both hearing aids and cochlear implants, earlier fit leads to better spoken language skills, if the amplification is effective. For hearing aids, some studies find that children with milder hearing loss who receive hearing aids earlier have better early language achievement than children who are fit with hearing aids later (Tomblin et al., 2015), but this finding does not hold for children with severe-to-profound hearing loss (Kiese-Himmel, 2002; Watkin et al., 2007) (for whom hearing aids are generally ineffective). Analogously, children who are eligible and receive cochlear implants earlier have better speech perception and spoken language outcomes than those implanted later (Artières, Vieu, Mondain, Uziel, & Venail, 2009; Dettman, Pinder, Briggs, Dowell, & Leigh, 2007; Miyamoto, Hay-McCutcheon, Kirk, Houston, & Bergeson-Dana, 2008; Svirsky, Teoh, & Neuburger, 2004; Yoshinaga-Itano et al., 2018), with best outcomes for children receiving implants before their first birthday (Dettman et al., 2007).

Communication. Total Communication refers to communication that combines speech, gesture, and elements of sign, sometimes simultaneously. Total communication, while often including elements of sign such as individual signs, is not a full-fledged sign language like American Sign Language (Mueller, 2013; Scott & Henner, 2020). Clinicians currently employ total communication as an alternative or augmentative communication method for children with a wide range of disabilities (Branson & Demchak, 2009; Gibbs & Carswell, 1991; Mirenda, 2003).

Compared to total communication, DHH children using an exclusively oral approach have better speech intelligibility (Dillon, Burkholder, Cleary, & Pisoni, 2004; Geers et al., 2017; Geers, Spehar, & Sedey, 2002; Hodges, Dolan Ash, Balkany, Schloffman, & Butts, 1999) and auditory perception (Geers et al., 2017; O'Donoghue, Nikolopoulos, & Archbold, 2000). That said, there is some debate as to whether an oral approach facilitates higher spoken language performance, or whether children who demonstrate aptitude for spoken language are steered towards the oral approach rather than total communication (Hall, Levin, & Anderson, 2017).

1-3-6 Guidelines. Early identification (Apuzzo & Yoshinaga-Itano, 1995; Kennedy et al., 2006; Robinshaw, 1995; White & White, 1987; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998; Yoshinaga-Itano et al., 2018) and timely enrollment in early intervention programs (Ching, Dillon, Leigh, & Cupples, 2018; Ching et al., 2013; Holzinger, Fellinger, & Beitel, 2011; Vohr et al., 2008, 2011; Watkin et al., 2007) are associated with better language proficiency. Indeed, DHH children who receive prompt diagnosis and early access to services have been found to meet age-appropriate developmental outcomes, including language (Stika et al., 2015). In line with these findings, the American Academy of Pediatricians (AAP) has set an initiative for Early Hearing Detection and Intervention (EHDI). Their EHDI guidelines recommend that DHH children are screened by 1 month old, diagnosed by 3 months old, and enter early intervention services by 6 months old. We refer to this guideline as 1-3-6. Meeting this standard appears to improve spoken language outcomes for children with HL (Yoshinaga-Itano et al., 2017, 2018) and the benefits appear consistent across a range of demographic characteristics.

At a federal level in the U.S., the Early Hearing Detection and Intervention Act of 2010 (Capps, 2009) was passed to develop state-wide systems for screening, evaluation, diagnosis, and "appropriate education, audiological, medical interventions for children identified with hearing loss," but policies for early diagnosis and intervention vary by state. As of 2011, 36 states (including North Carolina; "15A NCAC 21F .1201 - .1204," 2000) mandate universal newborn hearing screening (UNHS; National Conference of State Legislatures, 2011). All states have some form of early intervention programs that children with hearing loss can access (NAD, n.d.), but the specifics vary state-by-state. For instance, half of the states in the US do not consider mild hearing loss an eligibility criterion for early intervention (Holstrum, Gaffney, Gravel, Oyler, & Ross, 2008); North Carolina does include mild hearing loss in early intervention.

In evaluating the success of this initiative, the AAP (EHDI, n.d.) finds that about 70% of US children who fail their newborn hearing screening test are diagnosed with hearing loss before 3 months old, and that 67% of those diagnosed (46% of those that fail newborn hearing screening) begin early intervention services by 6 months old. These findings suggest that there are breaks in the chain from screening to diagnosis and from diagnosis to intervention, with potential ramifications for the language development of children not meeting these guidelines. We return to this in the discussion.

Quantifying vocabulary growth in DHH children

In what follows, we analyze data from the MacArthur Bates Communicative Development Inventory (CDI, Fenson et al., 1994). This parent-report instrument gathers information about children's vocabulary development, and is commonly used in both research and applied settings. The Words and Gestures version of the form is normed for 8–18-month-olds. On Words and Gestures, parents indicate whether their child understands and/or produces each of the 398 vocabulary items, and answer questions about young children's early communicative milestones. The Words and Sentences version of the form is normed for 16–30-month-olds. On Words and Sentences, parents indicate whether their child produces each of the 680 vocabulary items, and answer some questions about grammatical development. The CDI has been normed on a large set of participants across many languages (Anderson & Reilly, 2002; Frank et al., 2017; Jackson-Maldonado et al., 2003).

The CDI has also been validated for DHH children with cochlear implants (Thal, Desjardin, & Eisenberg, 2007). More specifically, in this validation, researchers asked parents to complete the CDI, administered the Reynell Developmental Language Scales, and collected a spontaneous speech sample. All comparisons between the CDI and the other measures yielded significant correlations ranging from 0.58 to 0.93. Critically, the children in this study were above the normed age range for the CDI, and thus this validation helps to confirm that the CDI is a valid measurement tool for older DHH children. In further work, Castellanos, Pisoni, Kronenberger, and Beer (2016) find that in children with CIs, number of words produced on the CDI predicts language, executive function, and academic skills up to 16 years later. Building on this work, several studies have used the CDI to measure vocabulary development in DHH children (Yoshinaga-Itano et al. (2017); Yoshinaga-Itano et al. (2018); de Diego-Lázaro et al. (2018); Vohr et al. (2008); Vohr et al. (2011). We build on this literature in our analyses below.

Goals and Predictions

This study aims to 1) characterize the demographic, audiological, and intervention variability in the population of DHH children receiving state services for hearing loss; 2) identify predictors of vocabulary delays; and 3) evaluate the success of early identification and intervention efforts at a state level. We include 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; Nicholas & Geers, 2006).

For the first goal, we expected that many of these variables would be related, due to known causal relations (e.g., cochlear implants recommended for severe hearing loss, but not mild hearing loss). We sought to provide descriptive documentation about the distribution of demographic, audiological, and intervention characteristics in a diverse sample of DHH children receiving state services. For the second, 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, and presence of additional disabilities would predict larger spoken vocabulary delay. We did not have strong predictions regarding the effects of communication method or presence of other health issues (e.g., congenital heart malformation) on vocabulary. For the third goal, based on the prior literature summarized above, 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.

Methods

Clinical evaluations were obtained through an ongoing collaboration with the North Carolina Early Language Sensory Support Program (ELSSP), an early intervention program serving children with sensory impairments from birth to 36 months. ELSSP passed along deidentified evaluations to our team after obtaining consent to do so from each family. No eligibility criteria beyond hearing loss and receiving an ELSSP evaluation were imposed, given our goal of characterizing the full range of DHH children with hearing loss in North Carolina.

The clinical evaluations included demographic and audiological information, CDI vocabulary scores, and the results of any clinical assessments administered (e.g., PPVT), detailed further below. For some children (n=47), multiple evaluations were available from different timepoints. In these cases, only the first evaluation was considered for this study, due to concerns regarding within-subjects variance for statistical analysis.

While this collaboration is ongoing, we opted to pause for this analysis upon receiving data from 100 children. Thus, the reported sample below consists of 100 children (56 male / 44 female) ages 4.20–36.20 months (M=21.20, SD=9.10). Race and socioeconomic information were not available. Families were administered either the Words and Gestures or Words and Sentences version of the CDI based on clinician judgment. Children who were too old for Words and Gestures, but who were not producing many words at the time of assessment, were often given Words and Gestures (n=37). Families for whom Spanish was the primary language (n=14) completed the Spanish language version of the CDI (Jackson-Maldonado et al., 2003).

Table 1

CDI result summary. For each version of the CDI (WG = Words and Gestures; WS = Words and Sentences), the table shows the mean(SD) age, comprehension score, and production score of participants in our sample, along with the percent diagnosed with developmental delays.

CDI version	Mean Age (SD)	Mean Comprehension (SD)	Mean Production (SD)	% Developmental Delays
WG (n=74)	20 (8.8) months	105 (100) words	32 (53) words	18.9%
WS (n=24)	26 (7.8) months	NA	149 (180) words	4.2%

Table 2

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

Condition	Specific Condition	n
Premature		17
	Extremely Premature	11
	NICU stay	16
Health Issues		36
	Heart	9
	Lung	5
	Illness	15
	Feeding Issues	14
	Pregnancy/Birth Complications	11
	Musculoskeletal	9
	Cleft Lip/Palate	4
	Other	15
Developmental Concerns		17
	Down Syndrome	5
	Chromosomal Issues	2
	Neural Tube Defects	2
	Other	10
Vision Loss		5
	Retinopathy of Prematurity	1
	Nearsightedness	1
	Farsightedness	1
	Cortical Visual Impairment	1

With regard to comorbid diagnoses, children in this sample were coded as yes/no for cognitive development concerns (e.g., Down syndrome, global developmental delays; Cornelia de Lange syndrome), yes/no for premature birth (i.e., more than 3 weeks premature), yes/no for health issues (e.g., heart defects, kidney malformations, VACTERL association), and yes/no for vision loss (not corrected to normal by surgery or glasses); see Table 2.

Table 3

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

Laterality	Amplification	mean_HLbetter	mean_HLworse	mean_age_amplification	mean_age_implantation
Bilateral	CI	85.60	89.79	11.29	14.12
Bilateral	HA	47.02	55.57	8.28	NaN
Bilateral	none	49.67	53.65	NaN	NaN
Unilateral	HA	4.70	56.04	10.91	NaN
Unilateral	none	2.50	73.90	8.50	NaN

Degree of hearing loss was most often reported with a written description (e.g., "mild sloping to moderate" or "profound high frequency loss"). We created 3 variables: hearing loss in the better ear, hearing loss in the worse ear, and average hearing loss (average of better and worse ear). For the analyses below, we primarily use hearing loss in the worse ear to avoid any redundancies with laterality. Using the ASHA hearing loss guidelines, each of these hearing loss measures was coded with the decibels of hearing loss (dB HL) corresponding with the median dB HL for the level of hearing loss (e.g., moderate hearing loss was coded as 48 dB HL), and sloping hearing loss was coded as the average of the levels (e.g. mild to moderate was coded as 40.5 dB HL). Participants were also coded for unilateral or bilateral hearing loss; presence or absence of Auditory Neuropathy Spectrum Disorder; and etiology of hearing loss (sensorineural, conductive, or mixed). Amplification was recorded as the device the child used at the time of assessment: either hearing aid, cochlear implant, or none. See Table 3 for audiological characteristics of the sample.

Table 4

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

Communication	English	Hindi	Spanish	Total
cued speech	1	0	0	1
spoken	68	1	10	79
total communication	15	0	3	18

Communication method was recorded as spoken language, total communication, or cued speech. One participant had a parent fluent in sign language, but the reported communication method in the home was total communication. No child in our sample used American Sign Language or another signed language. The forms also listed the primary language spoken at home, which we binned into English-speaking and non-English-speaking. 85 out of the 100 had a primary language of English, while 14had Spanish. The remaining child was adopted from a non-English-speaking country after age 2 and had heard mostly non-English by the time of assessment; this child was coded as non-English-speaking. Language and communication information is summarized in Table 4.

Age at screening was measured as the child's age in months at their first hearing screening. Age at screening was available for 68 participants. All participants with a screening age available were screened at birth or while in the NICU. We presume that the vast majority of participants without age at screening received their screening as newborns, as North Carolina boasts a 98% newborn hearing screening rate (NCDHHS, 2013). Age at diagnosis was taken as the age in months when children received their first hearing loss diagnosis. All children were enrolled in birth-to-three early intervention services through ELSSP, and the date of enrollment was listed on the clinician evaluation. For determining whether participants met the 1-3-6 guidelines, given the very high rates of early screening reported in our sample and by the state, we imputed missing data by assuming that children met the “screening by 1 month” criterion if they met the “diagnoses by 3 months”. Finally, we also calculated the number of hours of early intervention services received per month (including service coordination, speech therapy, and occupational therapy, among others) based on the clinician report.

Table 5

Detailed information about the variables studied. For categorical variables, levels are described. For continuous variables, range, mean, and standard deviation are provided.

Variable	Scale	Range
Age	Continuous	4-36 months (mean (SD): 21 (9))
Age at Amplification	Continuous	2-30 months (mean (SD): 9 (7))
Age at Diagnosis	Continuous	0-30 months (mean (SD): 5 (7))
Age at Implantation	Continuous	7-32 months (mean (SD): 14 (7))
Age at Intervention	Continuous	1-33 months (mean (SD): 11 (9))
Amplification	Categorical	Hearing Aid / Cochlear Implant / None
Communication	Categorical	Spoken / Total Communication / Cued Speech
Degree Hearing Loss (worse ear)	Continuous	18-100 dB HL (mean (SD): 64 (24))
Developmental Delay	Categorical	Yes / No
Gender	Categorical	Female / Male
Health Issues	Categorical	Yes / No
Language in Home	Categorical	English / Other
Laterality	Categorical	Unilateral / Bilateral
1-3-6	Categorical	Yes / No
Premature Birth	Categorical	Full-term / Premature
Services Per Month	Continuous	0-43 services per month (mean (SD): 6 (6))
Etiology	Categorical	Sensorineural / Conductive / Mixed
CDI - Words Produced	Continuous	0-635 words (mean (SD): 61 (111))

Results

We split the results into three parts. In the first, we explore relationships among child demographic, audiological, and clinical variables. In the second, we use these variables to predict vocabulary development. Finally, in the third, we describe the implementation of the EHDI 1-3-6 guidelines and predictors of early diagnosis and intervention in this sample. All analyses were conducted in R. All code is available on Github.

Relationships Among Demographic, Audiological, and Clinical Variables

Before we test how these variables may be related to vocabulary, we describe their relationships to each other. As would be expected, many health, audiological, and clinical characteristics are not distributed randomly across this sample of children. To quantify this statistically, we used Bonferroni-corrected chi-square tests between each of our variables (gender (male/female), laterality (bi-/uni-lateral hearing loss), health issues (yes/no), developmental delays (yes/no), premature birth (yes/no), language background (English/non-English), 1-3-6 (yes/no), degree of hearing loss (mild, moderate, severe/profound as defined above), etiology (sensorineural/conductive), services received per month (binned into 0-2, 3-6, and >7 - to create maximally evenly sized bins), communication (spoken/total communication) and amplification (hearing aids/cochlear implants/none)). 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 section of the analysis. Strictly speaking, some variables are not expected to be randomly distributed relative to each other (e.g., premature birth and health issues; degree and amplification), but quantifying the differences via chi-square using a conservative significance threshold lets us highlight the strongest relationships within this dataset.

Given that we ran 66 Chi-square tests, Bonferroni-corrected alpha for this set of analyses was $p < 0.0007$. Of these 66 combinations of variables, $p < .05$ for 26, and 9 survived Bonferroni correction. We are only discussing the latter below, but the full set of results can be found in Figure 1.

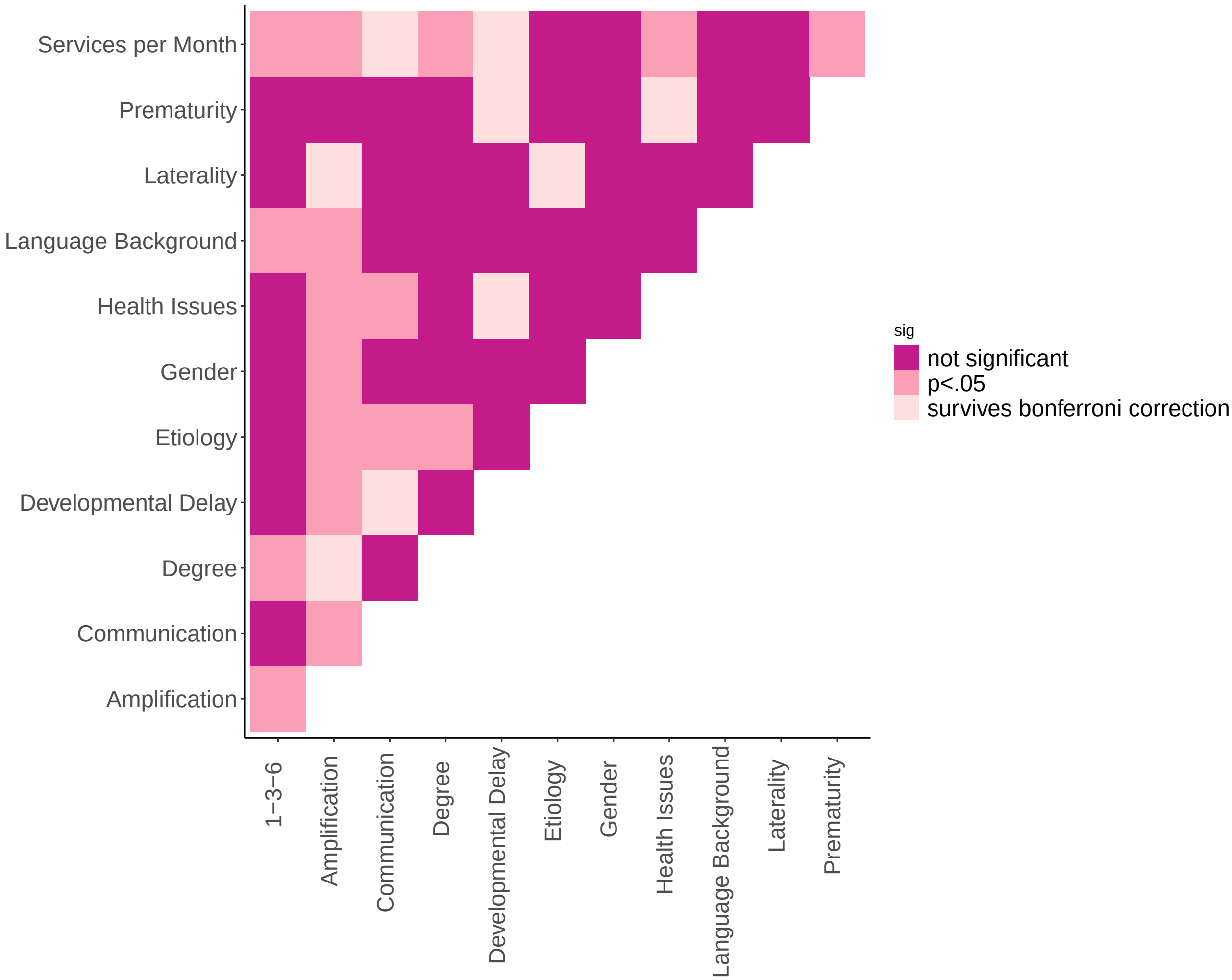

Figure 1. Results of chi-square tests between variables. X- and y-axes show the variables compared. Color of the square represents significance of the corresponding chi-square test. For tests that survived Bonferroni correction (p<.0007), effect size (Cramer's V) is given.

As expected, we found that health issues, developmental delays, and premature birth were highly interrelated in our sample, such that children born premature were more likely to also experience health issues (X^2 (1, N = 98) = 23.9, p < .0001) and developmental delays (X^2 (1, N = 98) = 11.63, p = .0006), and children with developmental delays were more likely to also experience health issues (X^2 (1, N = 98) = 20.87, p < .0001). Children with developmental delays received more services per month than typically-developing children (X^2 (2, N = 95) = 22.17, p < .0001) and were more likely to use total communication (X^2 (2, N = 98) = 22.51, p < .0001). Likewise, children who used total communication received more services per month than children using spoken language (X^2 (4, N = 95) = 21.35, p = .0003).

We also confirmed expected relationships among many of the audiological characteristics. There was a significant relationship between laterality and etiology (X^2 (2, N = 88) = 18.29, p = .0001), such that children with conductive hearing loss were more likely to have unilateral hearing loss, and children with sensorineural hearing loss were more likely to have a bilateral loss[2]. The chi-square tests further showed that amplification was related to laterality (X^2 (2, N = 98) = 16.43, p = .0003) and degree of hearing loss (X^2 (4, N = 87) = 28.45, p < .0001) in our sample. Specifically, children with bilateral hearing loss were more likely than children with unilateral hearing loss to use a hearing aid or cochlear implant; no child with unilateral hearing loss used a cochlear implant, and many children with unilateral hearing loss used no amplification. Regarding degree, children with severe to profound hearing loss were more likely to use a cochlear implant than children with less severe hearing loss (i.e., mild or moderate).

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

Predictors of Vocabulary Delay

We next turn to the relationship between each of these variables and children's productive vocabulary, as measured by the CDI. Figures 2 & **??** show the vocabulary scores

[2] All children with mixed hearing loss (n=8) had bilateral hearing loss.

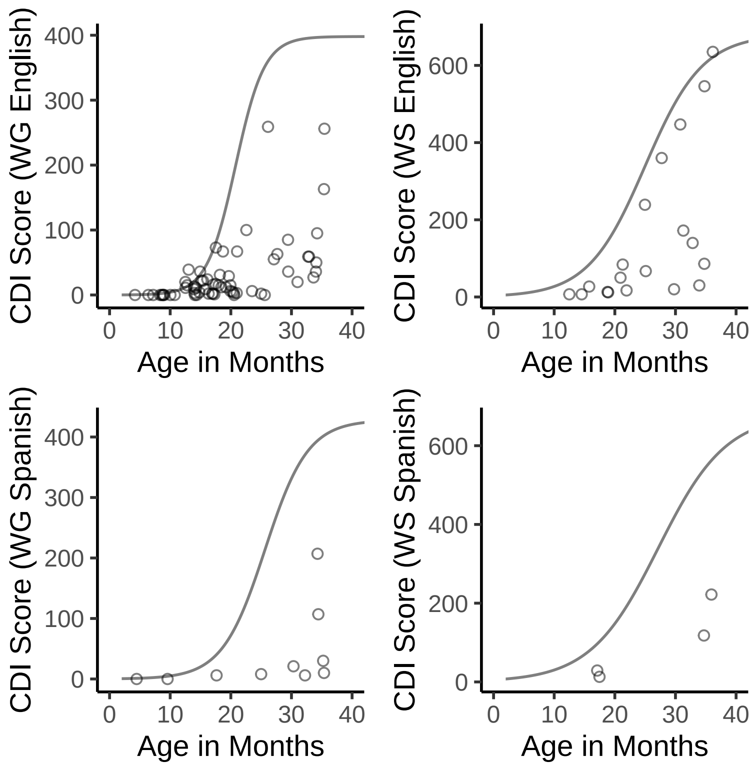

Figure 2. Lines show the growth curves created from Wordbank 50th percentile data. Left panels show Words & Gestures; right panels show Words & Sentences. Top row is American English data; bottom row is Mexican Spanish data. Dots represent vocabulary scores of individual DHH children in the sample.

of children in our samples relative to norms for hearing children for English and Spanish respectively. Descriptively, we found widespread vocabulary delays on both Words and Gestures and Words and Sentences, with the majority of DHH children testing around or below the 25th percentile for hearing children (based on WordBank norms; Frank et al., 2017).

As noted above, the CDI is composed of two instruments, which differ in number of questions (i.e. the maximum vocabulary score is 398 on Words and Gestures and 680 on Words and Sentences; 428 and 680 respectively for Spanish language CDI). To take this into account, rather than using the raw number of words produced as our outcome variable, we use WordBank norms to establish the difference (in months) between the child's chronological age and their predicted age based on their vocabulary, derived from the WordBank norms (Frank et al., 2017). We call this derived variable *vocabulary delay*.

More specifically, to compute a child's predicted age from their vocabulary score, we used the 50th percentile for productive vocabulary from Wordbank data typically-developing infants (Frank et al., 2017) to create binary logistic growth curves separately for the "Words and Gestures" (WG) and "Words and Sentences" (WS) versions of the CDI for American English and Mexican Spanish[3]. For each child, we took the number of words they produced divided by the number of words on the instrument, to give us the proportion of words produced. We used this proportion in an inverse prediction from the binary logistic regression curves to generate a predicted age. That is, for each possible CDI score, the growth curve provided the age that the score would be achieved for the 50th percentile trajectory. Finally, we subtracted the predicted age from each child's chronological age to calculate their vocabulary delay. However, for children producing 0 words, this approach was not appropriate due to the long tails on the growth curves. Thus, for this subset of children, we took the x-intercept from Wordbank (8 months for English, and 9 months for Spanish), and subtracted that value from the child's chronological age to get their vocabulary delay.

To look at the relationship between our predictor variables and CDI scores, we next conducted multiple linear regression, using vocabulary delay as our outcome variable. [4]

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

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

[4] We excluded the adopted child from this section of the analysis due to concerns about comparing her score to the American English CDI norms.

Background.

This model accounted for significant variance in vocabulary delay (adjusted-$R^2 = 0.59$, $p < .001$; see Table **??**). We next performed stepwise model comparison using stepAIC (MASS) to pare down the model. This process selects only the predictors which incrementally improved model fit, measured by Akaike's Information Criterion (AIC), which considers goodness of fit and model complexity (penalizing models with many predictors). We started model selection with the full model, as described above. We then filtered out data from children for whom Meets 1-3-6 (n=5) or Degree (n = 13) was unknown, as this stepwise AIC approach does not permit missing values across predictors. Since this initial filtered analysis found that Degree and 1-3-6 did not improve model fit, we manually removed the Degree and 1-3-6 terms from the model selection so that the 15 participants with missing cases for these variables could be retained.[5]

Table 6

Unstandardized beta weights (months of vocabulary delay) for the model of vocabulary delay selected by AIC: Vocabulary Delay Age + Laterality + Amplification.

term	estimate	std.error	statistic	p.value
(Intercept)	-0.54	1.71	-0.31	0.76
Laterality (Unilateral)	-2.70	1.14	-2.37	0.02
Amplification (Cochlear Implant)	-3.50	1.49	-2.36	0.02
Amplification (Hearing Aid)	-3.84	1.13	-3.38	0.00
Age	0.55	0.06	9.83	0.00

Based on this iterative process, we arrived at the following final model: Vocabulary

[5] 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 15 participants without Degree information.

Delay ~ Age + Laterality + Amplification. This model accounted for significant variance in children's vocabulary delay to a nearly identical degree as the full model (adjusted-R^2 = 0.58, $p = < .001$, see Table 6). We found significant main effects for Age, Amplification, and Laterality, such that older age, no amplification, and bilateral hearing loss predicted greater vocabulary delays. Compared to children with no amplification, children with cochlear implants had a 3.50 months smaller spoken vocabulary delay (p = .021), and similarly children with hearing aids had a 3.84 months smaller delay (p = .001). Children with unilateral hearing loss had a 2.70 months smaller delay (p = .020) than children with bilateral hearing loss. With regard to Age, for each month older, the model predicted a 0.55 months *larger* vocabulary delay (p < .001).

Given our results above revealing relationships among several of these variables (e.g., laterality and amplification), we tested for collinearity concerns by computing the model's VIF (variance inflation factor). This revealed low levels of collinearity among predictors in our final model (all VIF < 1.20; see Table **??**; James, Witten, Hastie, & Tibshirani, 2013). In sum, the analyses in this section revealed that over half of the variance in DHH children's vocabulary scores was explained by their age, whether their receive amplification, and whether their hearing loss was unilateral or bilateral.

Success in Meeting 1-3-6 Guidelines

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

Overall, 37% of our sample met 1-3-6 guidelines for early diagnosis and intervention (see Table **??**). Among the children for whom screening information was available (n=68), 100% were screened at birth or during NICU stay. 69% of children received diagnosis by 3 months of age, and 39% began early intervention by 6 months of age. Among children with comorbidities, 21.05% met 1-3-6 guidelines, compared to 47.37% of children without comorbidities. Figure 3 shows the age at first diagnosis, intervention, amplification, and implantation for each child in our sample.

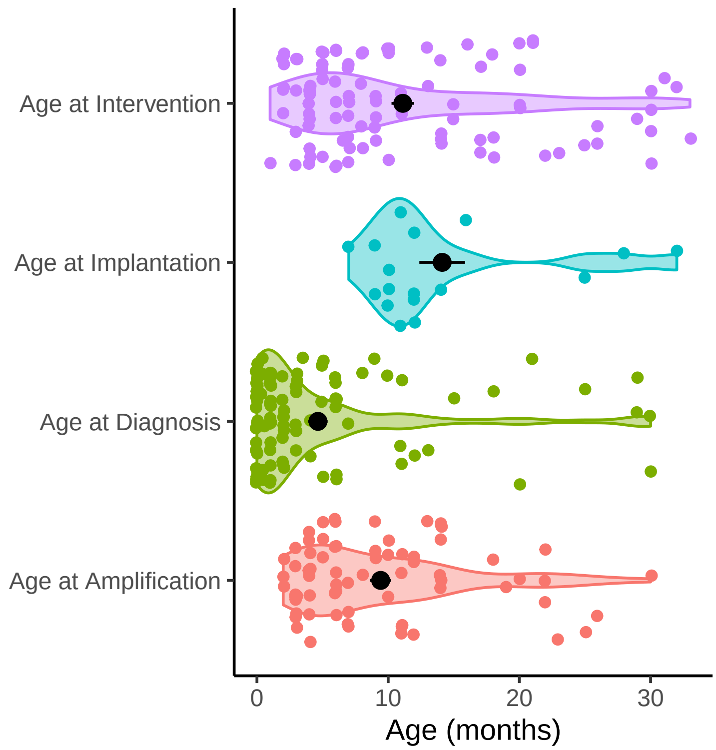

Figure 3. Age at diagnosis, intervention, amplification, and cochlear implantation across participants. Each dot represents the age that one child received the clinical service; violin width reflects data distribution. Blacks dot and whiskers show means and standard deviations. Not all children received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

We first tested the link between 1-3-6 and vocabulary directly in an exploratory analysis. An independent samples t-test showed that children who did not meet 1-3-6

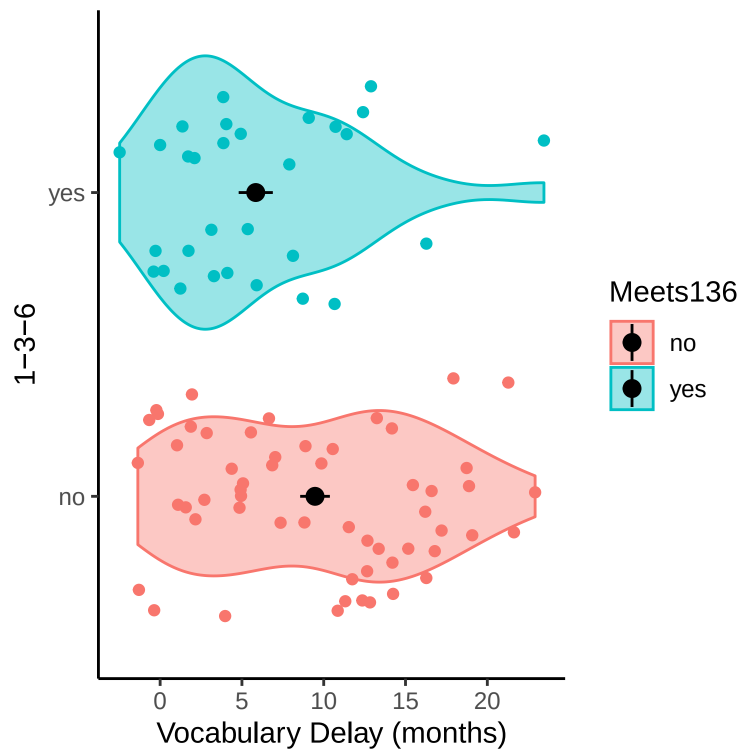

Figure 4. Estimated vocabulary delay for children who meet 1-3-6 guidelines for diagnosis/intervention (top) and children who do not (bottom). Each dot represents one child in the sample; violin width reflect data distribution. Blacks dot and whiskers show means and standard error.

guidelines had significantly larger vocabulary delays than children who met 1-3-6 guidelines (t(68.78)=2.62, p=0.01; see Figure 4). On average, the group that did not meet 1-3-6 guidelines was 3.62 months more delayed with regard to vocabulary (relative to the same 50th percentile benchmark from hearing children in Wordbank described above).

To better understand implementation of 1-3-6 guidelines, we next zoomed in on diagnosis and intervention. We conducted two linear regressions, one for age at diagnosis and one for age at intervention, considering only the predictors that would have been available or relevant at each of these stages (as detailed below). Model selection followed the same stepwise AIC-based process as described in the preceding section.

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

Table 7

Unstandardized beta coefficients (months) for the model of age at diagnosis selected by AIC: Age at Diagnosis   Health Issues + Language Background + Laterality.

term	estimate	std.error	statistic	p.value
(Intercept)	9.38	1.97	4.77	0.00
Health Issues (yes)	3.70	1.42	2.61	0.01
Language Background (English)	-6.47	1.96	-3.31	0.00
Laterality (Unilateral)	-2.15	1.58	-1.36	0.18

The best fit model was: Age at Diagnosis ~ Health Issues + Language Background + Laterality, with significant main effects of Health Issues and Language Background (see Table 7). This model accounted for 16.41% of the variance in age at diagnosis ($p = .001$). Average age at diagnosis was 4.65 months. Relative to English-speaking families, children from Spanish-speaking families were diagnosed 6.47 months later ($p = .001$). Children with health issues were diagnosed 3.70 months later than children without health issues ($p = .01$).

We repeated this model selection process for age at intervention. In addition to the variables used to fit the intervention model, we included age at diagnosis. The best fit model was: Age at Intervention ~ Premature Birth + Degree + Age at Diagnosis + Language Background (R^2=0.43 , $p < .001$; see Table 8, with significant main effects of degree and age at diagnosis. Prematurity (ß = 3.78, $p = .06$) and language background (ß = -1.38, $p = .52$) were not significant predictors on their own, but their inclusion improved model fit. Average age at intervention was 11.12 months. More severe hearing loss predicted earlier intervention,

Table 8

Unstandardized beta coefficients (months) for the model of age at intervention selected by AIC: Age at Intervention   Degree + Prematurity + Language Background + Age at Diagnosis.

term	estimate	std.error	statistic	p.value
(Intercept)	14.65	2.87	5.10	0.00
Degree of Hearing Loss	-0.09	0.03	-3.06	0.00
Premature (yes)	3.78	1.95	1.94	0.06
Age at Diagnosis	0.65	0.10	6.24	0.00
Language Background (English)	-1.38	2.12	-0.65	0.52

such that for every additional 10 dB HL, predicted age at intervention was 4.02 weeks earlier ($p < .01$). With regard to age at diagnosis, for every month diagnosis was delayed, intervention was delayed by 2.84 weeks ($p < .01$). Taken together these analyses reveal that beyond aspects of the child's hearing status, other variables too 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 examined in the full heterogeneity they naturally occur within. We took this big-tent approach by including any children receiving services for hearing loss.

Returning to our original three questions, we asked first: how are child-level variables intertwined? We found significant structure across many of the variables, suggesting that in

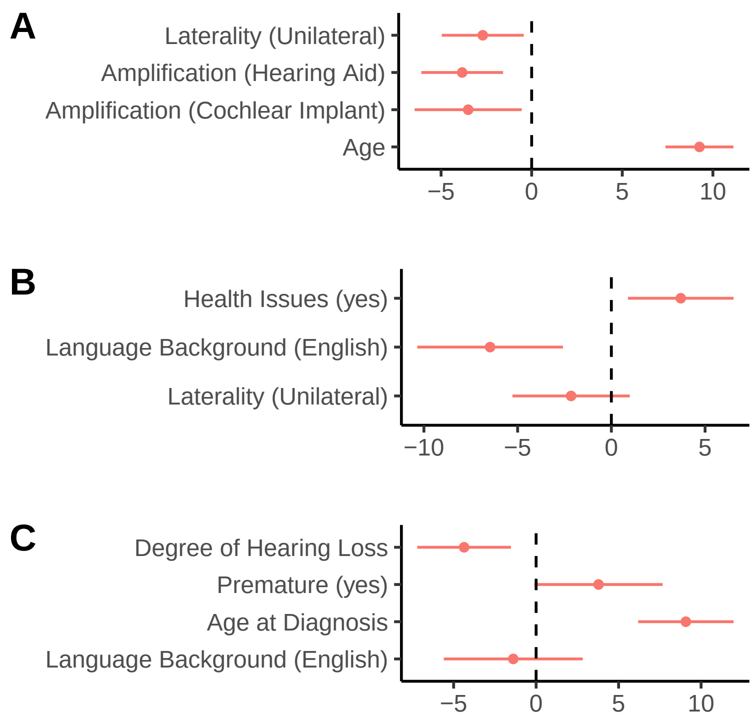

Figure 5. Unstandardized beta weights (months) for the models of (A) vocabulary delay, (B) age at diagnosis, (C) age, selected by AIC. A: ; B: Age at Diagnosis ~ Health Issues + Language Background + Laterality; C: Age at Intervention ~ Degree + Prematurity + Language Background + Age at Diagnosis.

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

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

How are child-level variables intertwined?

In our sample, we found significant overlap among demographic, audiological, and clinical variables. Prematurity, health issues, and developmental delay frequently co-occurred, such that children with one of these factors were more likely to have the others. This is not surprising. Many conditions that cause developmental delays have a high incidence of health issues (e.g., heart problems in Down Syndrome; vomiting and seizures with hydrocephalus), and it is well documented that there is a higher incidence of developmental delay and health issues in preterm infants (Aarnoudse-Moens, Weisglas-Kuperus, van Goudoever, & Oosterlaan, 2009; Costeloe et al., 2012; Luu, Katz, Leeson, Thébaud, & Nuyt, 2016; Pierrat et al., 2017; Robertson et al., 2009; York & DeVoe, 2002). In our sample, we also had a large range of health conditions (76 unique conditions in our sample of 100 children; see Table 2 and Appendix XXX for more detailed information about comorbidities). Some studies to date have examined the outcomes of DHH children with certain conditions (e.g., Clibbens, 2001; Szymanski, Brice, Lam, & Hotto, 2012). But given that the constellation of comorbid conditions is so varied, an important direction for future research is whether cognitive and social abilities, as well as family's treatment resources, are predictive of language outcomes across conditions.

We also found that children with developmental delays (e.g., Down syndrome) were much more likely to use a total communication approach than DHH children without developmental delays (i.e., total communication used by 58.82% of DHH children with developmental delay vs. 9.88% of those without). That is, communication modality was not distributed randomly throughout our sample, with use of total communication linked to children already at greater risk for verbal delays. Such a pattern is in line with clinical use of manual communication approaches for young children with disabilities (e.g., Branson & Demchak, 2009). This result tempers the interpretation of correlational studies finding links between total communication and language delays (e.g., Geers et al., 2017).

Our audiological variables too were not randomly distributed relative to each other. To highlight one such result, amplification devices were more common for children with less hearing (i.e., children with bilateral hearing loss and children with moderate to profound hearing loss). This may be due to the assumption that a hearing aid or cochlear implant will not benefit children with minimal hearing loss (Updike, 1994), although several studies have found benefits for amplification for mild or unilateral hearing loss (Briggs, Davidson, & Lieu, 2011; Hassepass et al., 2013; Priwin, Jönsson, Hultcrantz, & Granström, 2007; Walker et al., 2015; Winiger, Alexander, & Diefendorf, 2016).

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

Predicting vocabulary outcomes

In our sample, 88.89% of DHH children fell below the 50th percentile for spoken vocabulary. Moreover, of the 11.11% who were at or above the 50th percentile, 55.56% were 8-to-9-month olds who were not yet producing any words (as expected at this age). Finding that nearly 90% of DHH children are below the 50th percentile for vocabulary development indicates that this group is not yet well-equipped to acquire spoken language. This disadvantage can have lasting consequences in the lives of DHH children (Karchmer & Mitchell, 2003; Kyle & Harris, 2010; Qi & Mitchell, 2012), highlighting the importance of understanding what factors contribute to it.

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

Predicting early diagnosis and intervention

Our exploration of the implementation of 1-3-6 guidelines revealed that only 36.84% of children met the EHDI guidance for diagnosis by 3 months and intervention by 6 months, despite ample evidence suggesting early diagnosis and intervention improve language outcomes (Apuzzo & Yoshinaga-Itano, 1995; Ching et al., 2013; Holzinger et al., 2011;

Kennedy et al., 2006; Robinshaw, 1995; Vohr et al., 2008, 2011; Watkin et al., 2007; White & White, 1987; Yoshinaga-Itano et al., 1998, 2018). Children in our sample who met 1-3-6 guidelines were 3.62 months *less* delayed in spoken vocabulary than children who were late to receive diagnosis and/or services. With these demonstrable benefits in mind, our sample, by dint of accepting all children receiving early intervention services in one state, was able to explore naturally occurring variance in *who* received on-time diagnosis and intervention.

Diagnosis. Having health issues or a non-English language background predicted later diagnosis. Children with health issues were diagnosed 3.70 months later than infants without health issues. One possible explanation is that the health issues caused acquired hearing loss that wouldn't be detected by the newborn hearing screening, thus delaying 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, families and medical providers are likely to prioritize treatment for certain health issues (e.g., surgery for congenital heart defect) over diagnostic audiology services. Nevertheless, it is possible that in some cases, clinician awareness of the increased delays in language related to health issues more broadly may facilitate improvements in timely diagnosis.

Language background too predicted age at diagnosis. Infants from Spanish-speaking families were diagnosed 3.78 months later than infants from English-speaking families. This may be due to cultural differences in attitudes towards deafness (Caballero, Muñoz, Schultz, Graham, & Meibos, 2018; Rodriguez & Allen, 2020; Steinberg, Bain, Li, Delgado, & Ruperto, 2003; Steinberg, Dávila, Collazo, Loew, & Fischgrund, 1997) or it may result from a lack of linguistically accessible and culturally appropriate audiology services. Only 5.6% of

American audiologists identify as bilingual service providers (ASHA, 2019), and services from a monolingual provider may be insufficient. To this point, Caballero et al. (2017) found that Hispanic-American parents of DHH children wish for more concrete resources, comprehensive information, and emotional support from their audiologist. In a nationwide survey of audiologists, the majority of audiologists reported that language barriers presented a major challenge in working with Spanish-speaking families, specifically in obtaining the child's case history and providing recommendations for follow-up services(Abreu, Adriatico, & DePierro, 2011).

Intervention. As expected, more severe hearing loss predicted earlier intervention, such that for every additional 10 dB HL, predicted age at intervention was 0.93 month earlier. This converges with findings by Harrison, Roush, and Wallace (2003) in which severe-to-profound hearing loss was diagnosed 2-5 months earlier than mild-to-moderate hearing loss. Parents and clinicians may adopt a wait-and-see approach to intervention for children with some residual hearing. Nevertheless, mild-to-moderate hearing loss is associated with language delays and academic challenges (Blair et al., 1985; Delage & Tuller, 2007), which early intervention may offset.

Age at start of services was also associated with age at diagnosis: for every month diagnosis was delayed, intervention was delayed by 2.84 weeks. Ching et al. (2013) found that age at intervention predicted better outcomes for DHH children, above and beyond age at diagnosis. Of course, these two variables are related, such that we cannot hope to achieve early intervention goals without ensuring children receive timely diagnosis. Early diagnosis puts children in the pipeline towards intervention earlier.

Finally, it's important to note that this sample is composed of children receiving birth-to-3 services. An estimated 67% of children with hearing loss enroll in early intervention services (CDC, 2018). While this represents a tremendous step forward in prompt early intervention services relative to just a few decades ago, early intervention may not be early enough. Less than 39% of our sample of children in early intervention meet the 6-month EHDI benchmark. Furthermore, an unknown fraction of the DHH population in North Carolina aren't included in this analysis because they have not been enrolled in services by 36 months. The AAP estimates that almost 36% of infants who do not pass a newborn hearing screening are lost to follow-up. Assuming that the population of children in early intervention only represents two thirds of the population with hearing loss, our data suggest that the actual proportion of DHH children who receive intervention by the EHDI-recommended 6 months may be closer to 26%. These children may not receive clinical support until school-age or later, exacerbating concerns for language development, which lays an important foundation for literacy and academic success (Biemiller, 2003; Hemphill & Tivnan, 2008; Monroe & Orme, 2002; Stæhr, 2008; Young, 2005).

Educational and Clinical Implications

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

1. Frequent hearing screenings for children receiving medical or therapeutic care for health issues.

2. Service coordination for families balancing multiple co-occurring conditions.

3. Expansion of bilingual clinicians both in-person and teletherapy clinicians to provide therapy and service coordination to non-English-speaking families.

4. Provision and encouragement of early intervention services for children with mild to moderate hearing loss.

Additionally, the vast majority of children in our sample experienced vocabulary delays (relative to hearing peers), and studies of spoken vocabulary development in older DHH children suggest that they may not catch up (Lund, 2016). This should set clinicians and educators on high alert, due to the demonstrated importance of vocabulary skills in literacy (Biemiller, 2003; Hemphill & Tivnan, 2008; Stæhr, 2008) and in education more broadly (e.g., Young, 2005; Monroe & Orme, 2002). As early intervention predicts vocabulary outcomes in study after study (including this present study and e.g., Vohr et al., 2008, 2011; Ching et al., 2018, 2013; Holzinger et al., 2011; Watkin et al., 2007), ensuring intervention by 6 months for all DHH children may be one way to address spoken vocabulary deficits. Another solution: even prior to intervention or amplification, provision of structured, accessible language input (i.e., sign language) may mitigate negative effects of auditory deprivation on language skills (Davidson, Lillo-Martin, & Pichler, 2014; Hassanzadeh, 2012; Spellun & Kushalnagar, 2018). Indeed, while we recognize that learning sign language may pose a challenge for some families for myriad reasons, and as noted above, our sample did not use sign language, we nevertheless feel it is worth underscoring as an important language support for DHH children and their families.

In recommending sign language, we endorse the rationale set forth by Hall, Hall, and Caselli (2019). Summarizing their view, they note that spoken language outcomes for DHH children are variable and unpredictable (Ganek, McConkey Robbins, & Niparko, 2012; Szagun & Schramm, 2016), and even in optimal situations, many DHH children do not achieve age-appropriate spoken language outcomes (e.g., Geers et al., 2017). Failing to achieve language proficiency (in any language) confers higher risk of disrupted cognitive, academic, and socioemotional development (Amraei, Amirsalari, & Ajalloueyan, 2017; Dammeyer, 2010; Desselle, 1994; Hall et al., 2017; Hrastinski & Wilbur, 2016; Kushalnagar et al., 2011; Moeller & Schick, 2006; Preisler, Tvingstedt, & Ahlström, 2002; Schick, De Villiers, De Villiers, & Hoffmeister, 2007). The available data do not suggest that sign language harms spoken language development (Davidson et al., 2014; Park et al., 2013), and in fact, some studies suggest that sign language *benefits* spoken language development (e.g., Hassanzadeh, 2012). Providing early access to a natural sign language offers children another path to language mastery, and use of sign language does not preclude learning spoken language. Thus, we encourage sign language use *at least* prior to mastery of spoken language, and when possible for the family, we encourage its continued use as a language resource.

Limitations and Opportunities for Future Work

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

Given our exploratory analyses, there were many possible analytic routes. That said, our results largely converge with or replicate key aspects of past studies (e.g., Ching et al., 2013) and received wisdom among clinicians. In the interest of transparency, these data and all code generating our results are available on our OSF page (XXX) and we encourage those interested to explore further analyses.

This sample is composed only of children in North Carolina, and certain factors vary by country and by state (e.g., diagnosis and early intervention practices; NAD, n.d.). However, based on other demographic research (Blackorby & Knokey, 2006; Institute, 2014), 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. We would exercise caution in applying these results to regions where sign language access for DHH children is more common (e.g. Washington D.C.; Rochester, New York.) A similar naturalistic study in those regions could help illuminate the effects of different clinical and demographic factors in a signing population.

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

Conclusion

The present study explored demographic and audiological characteristics, vocabulary outcomes, and clinical milestones within a diverse sample of 100 DHH children enrolled in early intervention services. We found that overall, this sample showed spoken language vocabulary delays relative to hearing peers on average and room for improvement in rates of early diagnosis and intervention. Critically, we also found that the variables predicting these delays in both vocabulary and early support services reflected *both* dimensions that are immutable, and those that clinicians and caretakers can potentially alter. 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. # References

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