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# Language Growth in Children with Mild to Severe Hearing Loss who Received Early Intervention by 3 Months or 6 Months of Age

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## Abstract

**Purpose:** To evaluate the impact of hearing screening, diagnosis, and early intervention (EI) by 3 months or 6 months of age on language growth trajectories for children with hearing loss (HL) relative to children with normal hearing (NH).

**Method:** We recruited 133 children with mild to severe HL through universal newborn hearing screening records and referrals from audiologists in the United States and 116 children with NH who served as a comparison group. Examiners administered a battery of developmentally appropriate language measures between 12 months and 8 years of age. We constructed latent growth curve models of global language, grammar, and vocabulary using Bayesian statistics.

**Results:** Children with HL demonstrated no significant differences in initial language skills compared to children with NH. Children in the 1-3-6 group also showed no difference in language growth compared to children with NH. The slope for the 1-2-3 group was significantly steeper than children with NH for global language and grammar.

**Conclusions:** This study documents the positive impact of EI on language outcomes in children with congenital HL. It is among the first to provide evidence to support the potential effects of very early intervention by 3 months of age.

**Keywords:** hearing loss; language development; EHDI

**Acronyms:** BEPTA = better ear pure tone average; CASL = Comprehensive Assessment of Spoken Language; CELF-4 = Clinical Evaluation of Language Fundamentals, 4th ed.; EHDI = Early Hearing Detection and Intervention; EI = early intervention; HA = hearing aid; HL = hearing loss; NH = normal hearing; NHS = Newborn Hearing Screening; OCHL = Outcomes of Children with Hearing Loss, PPVT-4 = Peabody Picture Vocabulary Test, 4th ed.; PTA = pure tone average; WASI-II = Wechsler Abbreviated Scale of Intelligence

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Hearing loss (HL) in childhood is a common condition, with a prevalence around 3 per 1000 births (Mehra et al., 2009). Children with HL are at risk for significant communication delays (Tomblin, Harrison et al., 2015). Reduced access to linguistic input limits children with HL from achieving their full developmental potential (Meinzen-Derr et al., 2018; Moeller & Tomblin, 2015). With the advent of universal newborn hearing screenings, a majority of children with HL in the United States are meeting one or more of the Early Hearing Detection and Intervention (EHDI) goals of screening by 1 month of age, HL confirmation by 3 months, and entry into early intervention by 6 months (Holte et al., 2012; Walker et al., 2014; Walker et al., 2017). Meeting these benchmarks has a positive impact on language, psychosocial, and academic outcomes for children with HL (Joint Committee on Infant Hearing [JCIH], 2019).

Because early intervention facilitates the acquisition of age-appropriate language skills (Moeller, 2000; Pimperton & Kennedy, 2012; Sininger et al., 2010; Watkin et al., 2007; Yoshinaga-Itano et al., 1998; Yoshinaga-Itano et al., 2017), researchers and clinicians have debated whether to pursue more aggressive milestones for EHDI services, referred to as the "1-2-3" benchmarks: screening by 1 month, confirmation by 2 months, entry into early intervention by 3 months. In the 2019 JCIH position statement, the committee recommended that U.S. states who currently are meeting 1-3-6 benchmarks should strive toward accomplishing 1-2-3 benchmarks. However, there is currently little direct evidence to suggest that reaching benchmarks earlier would result in further improvements in outcomes compared to the current 1-3-6 benchmarks. Documenting the potential effects of very early intervention has important public health significance and would provide empirical evidence to guide best-practice models for children with HL for physicians, audiologists, parents, and other stakeholders. It is critical to have these data before we can encourage states to devote the time and resources needed to implement an accelerated EHDI timeline.

Historically, prospective and longitudinal cohorts of children with HL who experienced early identification and intervention have not been available to study. Thus, the field of audiology has not had the opportunity to study the effects of accelerated EHDI timelines on language growth in children with HL compared to the traditional 1-3-6 recommendations. It is critical to address these questions about earlier intervention because evidence suggests that children with HL, including those with mild HL, remain at risk for language delays even in an era of newborn hearing screening (Walker et al., 2020; Yoshinaga-Itano et al., 2017).

The primary goal of the current study is to estimate the language growth trajectories for children with mild to severe bilateral HL and to compare the growth rates for children who met the traditional 1-3-6 benchmarks versus the growth rates for children who met an accelerated 1-2-3 timeline with peers with normal hearing (NH). In these analyses, we examined growth of vocabulary (i.e., the content of language or word knowledge), grammar (i.e., the structural form of language or morphosyntactic knowledge), and global language (i.e., a combination of receptive and expressive language). A secondary goal was to examine the effects of HL severity and socio-economic status (SES) on growth trajectories for children in EI. We compared prospective longitudinal data in children who had cognitive skills within normal limits, no additional disabilities, and used spoken English. The current study rectifies some of the previous issues with investigations of treatment effects of EHDI. Specifically, we had a unique opportunity to examine prospective longitudinal data in children with HL or NH who have cognitive skills within or above normal limits, no major secondary disabilities, and were from monolingual English-speaking homes. Thus, the findings from this study provide us with an opportunity to address questions about timing of intervention without many of the additional confounds that are typical in this line of research (Ching et al., 2017; Ching et al., 2013).

In both aims, our interest is in language growth rates for children with HL from 6 months to 8 years of age. Due to this wide age range we needed to use developmentally appropriate measures; thus, different measurement instruments were used over time. Ideally, the measurement of growth employs a common scale across time that reflects a common trait. Our previous reports of language growth analysis used standard scores provided by norm-referenced tests to address the challenge of a common scale across time (Tomblin, Harrison, et al., 2015). Using standard scores placed all measures on the same scale but did not control for systematic differences in the norm-referenced groups. True changes in growth were confounded with changes in the ability levels of the norm-referenced groups. Further, the scaling of standard scores did not reflect the expected absolute gains in language ability over development, but instead indexed relative growth. Children with average growth were expected to have the same standard score across time. In this report, we have adopted a novel method for measuring language growth that draws on Bayesian methods to estimate a latent language ability based on the raw scores produced by our tests (Ward et al., 2020). Compared to traditional frequentist approaches, the Bayesian methods are valuable as they allow information to be borrowed across multiple tests of the same latent construct. The Bayesian model employed in the current study uses all available data to construct latent growth curves; thus, the statistical analysis did not require that children have the same measurements at every test visit.

## Method

### Participants

The Outcomes of Children with Hearing Loss (OCHL) study used an accelerated longitudinal design. We recruited children between 6 months and 7 years at enrollment into the study. They were followed beginning at time of enrollment biannually from 6 months to 2 years, and annually from 2 to 8 years (Holte et al., 2012; Tomblin, Walker, et al., 2015). Table 1 provides a summary of the participants' demographic and audiologic characteristics.

**Table 1**

*Demographic and Audiologic Characteristics for Children with Hearing Loss who met 1-2-3 Benchmarks or 1-3-6 Benchmarks and Children with Normal Hearing (CNH)*

| | 1–2–3 (*n* = 60) | | 1–3–6 (*n* = 73) | | CNH (*n* = 116) |
|---|---|---|---|---|---|
| Characteristic | Percentage of Participants | | | | |
| Sex, male | 48% (29/60) | | 60% (44/73) | | 47% (54/116) |
| Race | | | | | |
| White | 87% (52/60) | | 79% (58/73) | | 79% (92/116) |
| Black | 3% (2/60) | | 8% (6/73) | | 6% (7/116) |
| Asian | 2% (1/60) | | 3% (2/73) | | 2% (2/116) |
| Other | 8% (5/60) | | 10% (7/73) | | 13% (15/116) |
| Receive private insurance[1] | 80% (48/60) | | 75% (55/73) | | 78% (87/112) |
| Full-term birth (> 36 weeks)[2] | 88% (52/59) | | 77% (56/73) | | 86% (90/105) |
| Maternal education level[3] | | | | | |
| High school or less | 10% (6/60) | | 17% (12/71) | | 19% (21/108) |
| Some college | 28% (17/60) | | 28% (20/71) | | 14% (15/108) |
| Bachelor's degree | 30% (18/60) | | 30% (21/71) | | 32% (35/108) |
| Post-graduate education | 32% (19/60) | | 25% (18/71) | | 34% (37/108) |
| | Mean | SD | Mean | SD | |
| Better-ear pure tone average (dB HL) | 49.80 | 13.20 | 50.06 | 12.97 | N/A |
| Age at first hearing evaluation (months) | 1.21 | 0.55 | 2.53 | 1.25 | N/A |
| Age at confirmation (months) | 1.56 | 0.74 | 3.21 | 1.42 | N/A |
| Age at hearing aid fitting (months) | 3.05 | 1.46 | 5.72 | 4.64 | N/A |
| Age at entry into early intervention (months) | 2.30 | 0.76 | 4.59 | 1.18 | N/A |

*Note.* HL = hearing loss.

[1]Insurance status not reported for 4 CNH.

[2]Pregnancy length not reported for 1 child in the 1-2-3 group and 11 CNH.

[3]Maternal education level not reported for 2 children in 1-3-6 group and 8 CNH.

The full cohort consisted of 317 children with HL. Inclusion criteria were (a) permanent, bilateral, better-ear 4-frequency pure-tone average (BEPTA) of 20 to 75 dB HL, (b) at least one primary caregiver who used spoken English, and (c) no known additional sensory or neurodevelopmental disorders. Children who used sign language as their primary communication mode were excluded because inclusion would require a different approach to language outcome measurement, making it difficult to compare groups.

Within the cohort, 78 passed their Newborn Hearing Screening (NHS) or did not receive an NHS; these children were excluded from these analyses due to the inability to document whether the HL was congenital. Two hundred thirty-nine failed their NHS: 25% (*n* = 60) met 1-2-3 benchmarks, 31% (*n* = 73) met 1-3-6 benchmarks, and 44% (*n* = 106) had a delay in diagnosis or EI. Data from the 106 children who were delayed in diagnosis or EI have been examined in previous analyses and are excluded from the current study (Tomblin, Harrison, et al., 2015). In the current study, we examined the 133 children enrolled in EI by 3 or 6 months, to directly test the effect of a 1-2-3 timeline relative to the current JCIH 1-3-6 recommendations. Inclusion in the 1-2-3 group was defined as NHS by 1 month, diagnostic testing by 2 months, and EI by 3 months. Children in the 1-3-6 group had NHS by 1 month, diagnostic testing between 3 and 5 months, and EI between 3 and 6 months. All children were fitted with hearing aids. Children in the 1-2-3 group were fitted at an average age of 3.05 months (*SD* = 1.46) and wore their hearing aids an average of 10.33 hours per day across visits (*SD* = 3.24). Children in the 1-3-6 group were fitted at an average age of 5.72 months (*SD* = 4.64) and wore their hearing aids an average of 10.59 hours per day across visits (*SD* = 3.03). There was no significant difference in hearing aid use time between the two groups ($p = .38$). Ten percent (*n* = 6/60) and 11% (*n* = 8/73) of the 1-2-3 and 1-3-6 groups, respectively, presented with a progressive HL (defined as more than a 10 dB HL increase in PTA between visits). Because the number of children who demonstrated progressive HL in either group was small, we did not control for progression of hearing loss in the statistical analyses.

One hundred sixteen children with NH, matched by age and maternal education level (as a proxy for SES) with the

children with HL, also participated in the OCHL study. The children with NH were included to provide a comparison group that was well matched with the children with HL in terms of home and family background. Children with NH used spoken English to communicate. Nonverbal cognition in both children with HL and children with NH was average to above-average, as measured by nonverbal subtests of the Wechsler Preschool and Primary Scale of Intelligence-III (Wechsler, 2002) at age 4 years or Wechsler Abbreviated Scale of Intelligence-2 (WASI-2; Wechsler & Hsiao-pin, 2011) at age 6 years.

### Procedures

All study procedures were approved by Institutional Review Boards at the University of Iowa, Boys Town National Research Hospital, and the University of North Carolina.

#### *Hearing and Language Measures*

Parents completed an intake questionnaire that documented age at NHS, HL confirmation, hearing aid (HA) fitting, and EI. Clinically certified audiologists who were experienced in working with children completed hearing assessments at each visit, including air-conduction and bone-conduction thresholds at 500, 1000, 2000, and 4000 Hz at a minimum. The BEPTA at these four frequencies was calculated for subsequent analyses.

The language test battery consisted of a combination of parent-report measures and standardized, norm-referenced tests. The language examiners included audiologists, clinically certified speech-language pathologists, or licensed teachers. The measures varied depending on the chronological age of the children. The Vineland Adaptive Behavior Scales-2 Parent/Caregiver Form (Sparrow et al., 2005) was administered between 6 and 48 months of age. For the current analysis we only included the Vineland Receptive and Expressive language subscales. At ages 3, 4, 6, and 8 years we administered the Comprehensive Assessment of Spoken Language (CASL; Carrow-Woolfolk, 1999). Scores on the Basic Concepts subtest (vocabulary) and the Grammar Construction (grammar) are included in the current analysis. The Clinical Evaluation of Language Fundamentals-4 (CELF-4; Semel et al., 2004) Word Structure subtest, which assesses grammar, was administered at 5 and 7 years of age. The Peabody Picture Vocabulary Test-4 (PPVT-4; Dunn & Dunn, 2007) was administered at 5 and 7 years of age, and the WASI-2 Vocabulary subtest (Wechsler & Hsiao-pin, 2011) was administered at 7 and 8 years of age. Both tests measure vocabulary knowledge. Table 2 displays the constructs, test names, descriptions, types of scores, and the ages at which the language assessments were administered.

**Table 2**

*Constructs, Test Names, Descriptions, Types of Scores, and Visit Administered for Language Measures*

| Construct/test name | Description | Score | Visit Administered | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | 12m | 18m | 2yr | 3yr | 4yr | 5yr | 6yr | 7yr | 8yr |
| **Global Language** | | | | | | | | | | | |
| VABS-2 Expressive Language and Receptive Language | Parent-report checklist | Raw score | X | X | X | X | X | | | | |
| **Grammar** | | | | | | | | | | | |
| CASL Syntax Construction | Expressive morphosyntax; cloze procedure with picture support | Raw score | | | | X | X | | X | | X |
| CELF-4 Word Structure | Expressive morphosyntax; cloze procedure with picture support | Raw score | | | | | | X | | X | |
| **Vocabulary** | | | | | | | | | | | |
| CASL Basic Concepts | Lexical/semantic knowledge; picture pointing task with four-item closed set | Raw score | | | | X | X | | | | |
| PPVT-4 | Receptive vocabulary; picture pointing task with four-item closed set | Raw score | | | | | | X | | X | |
| WASI-2 Vocabulary | Expressive vocabulary; definitions | Raw score | | | | | | | | X | X |

*Note.* VABS-2 = Vineland Adaptive Behavior Scales; CASL = Comprehensive Assessment of Spoken Language; CELF-4 = Clinical Evaluation of Language Fundamentals; PPVT-4 = Peabody Picture Vocabulary Test; WASI-2 = Wechsler Abbreviated Scale of Intelligence.

Children were followed for an average of 1.8 years, and throughout follow-up were measured between two and 13 times across all instruments, with 50% of the children having between 8 and 10 measurements. Most children (53.6%) were measured on four instruments and 21.8% were measured on all seven instruments.

**Statistical Analysis**

We constructed latent growth curve models of three constructs: global language (Vineland Receptive and Expressive subscales), grammar (CASL Syntax Construction, CELF-4 Word Structure), and vocabulary (CASL Basic Concepts, PPVT-4, WASI-2 Vocabulary) in the Bayesian paradigm. For this analysis, the Bayesian paradigm is preferred because it provides an intuitive framework for estimation, dividing a complex process into a series of smaller, well-defined components using conditional probability (Oleson et al., 2019). Compared to Frequentist implementation, the Bayesian framework is advantageous as it allows information to be borrowed across multiple tests of the same latent construct. Children did not always have measurements on every test or multiple measurements on a test over time, and the Bayesian model leveraged all available data to construct latent growth curves.

Bayesian hierarchical models can be broken down into three components. The data model describes the distribution of the observed data, the process model describes the scientific process for the parameters of the data's distribution, and the parameter model sets the remaining prior distributions in the hierarchical model (Cressie & Wikle, 2015). These three components are combined using Bayes' rule to give the posterior distribution, which defines the probability distribution of the parameter values conditioned on any prior scientific evidence in the parameter model and the observed data in the data model. Another advantage of the Bayesian paradigm is in the natural and intuitive interpretation given by the posterior distribution.

For the data model of each of the three constructs, we assume that the individual score on each associated test over time is approximately normally distributed with its own mean and variance parameters. The process model defines the mean for each score over time using a linear equation which allows for group- and subject-specific intercepts and slopes. Time was scaled so that the intercept occurred at 6 months for interpretability. Information across multiple tests in each construct was combined by including effects for each measure in the intercept and slope and constraining the effects to sum to zero for identifiability. The subject-specific effects account for the within-subject correlation and borrow strength across each language measure. Each model controlled for HL severity and maternal education level. These two factors were included because they could influence the timing of hearing aid fitting and we wanted to control these effects. We treated HL severity as a continuous measure; children with NH were given a HL severity value of zero, corresponding to no HL. We treated maternal education level as an ordinal variable with four levels: (a) high school or less, (b) some college, (c) bachelor's degree, or (d) post-graduate education. Severity of HL and maternal education were included as covariates in the intercept terms, meaning the effects of these variables were held constant over time.

The final stage of the hierarchical model is to assign prior distributions for all the remaining parameters. We used vague but proper priors. The test effects were given Normal priors with mean 0 and a large, uninformative standard deviation of 10,000. The group effects were also given Normal priors with mean 0 and a large uninformative standard deviation of 1,000. The random subject effects were given a multivariate normal distribution with mean 0 and variance-covariance matrix which followed an inverse Wishart distribution. All remaining variance parameters were given non-informative inverse gamma (0.1, 0.1) priors. The vague priors reflect the lack of preexisting information on the parameters and ensures data driven final outcomes.

To compare growth curves for each language construct, we analyzed the posterior distributions of the differences in the intercept and slope between each of the three groups: children with NH, children with HL who met the 1-3-6 benchmark, and children with HL who met the 1-2-3 benchmark. The posterior mean of each difference provides an estimate of the average difference in the intercept or slope, given the observed data. We used Bayesian credible intervals (CI) to test for significant differences: A 95% CI defines the region where the true difference in intercepts or slopes lies with 95% probability, given the data. A 95% CI for the difference in intercepts or slopes that does not include zero indicates a statistically significant difference in that parameter between the groups at the 5% level. Analyses were performed with R version 4.0.2 and OpenBUGS version 3.2.3 (Lunn et al., 2000). For each model, three chains were run for 30,000 iterations after a burn-in period, with convergence indicated by all parameters achieving a Gelman-Rubin statistic less than 1.1 (Gelman & Rubin, 1992).

## Results

Table 3 summarizes the posterior distribution of the group differences in the intercept and slope and covariate effects for each language construct model (global language, grammar, and vocabulary). The posterior means of the group differences in the intercepts and slopes represent the average difference in the latent score at six months and the average difference in yearly growth of the latent score, respectively. Significant credible intervals are shaded.

**Children who Met 1-3-6 Benchmarks Compared to Children with Normal Hearing**

Figure 1A displays growth trajectories of the 1-3-6 group compared to children with NH for each language construct. Across all three language constructs, there was no difference in intercepts or slopes between the 1-3-6 and NH groups. These results indicate that children with HL that met 1-3-6 benchmarks and children with NH had similar starting points and growth trajectories in global language, grammar, and vocabulary.

**Table 3**

*Posterior Mean, Standard Deviation, and 95% Credible Intervals (CI) for Each Parameter of Interest*

| | | Global Language | | Grammar | | Vocabulary | |
|---|---|---|---|---|---|---|---|
| | | Group | *n* | Group | *n* | Group | *n* |
| | | NH | 82 | NH | 105 | NH | 105 |
| | | 1–3–6 | 53 | 1–3–6 | 61 | 1–3–6 | 61 |
| | | 1–2–3 | 51 | 1–2–3 | 44 | 1–2–3 | 44 |
| | | Mean (SD) | 95% CI | Mean (SD) | 95% CI | Mean (SD) | 95% CI |
| 1-3-6 vs. NH | Intercept (6 months) | -2.43 (2.09) | (-6.50, 1.68) | -2.05 (2.30) | (-6.41, 2.38) | -2.88 (3.02) | (-8.69, 3.82) |
| | Slope | 0.67 (0.63) | (-0.57, 1.87) | 0.32 (0.44) | (-0.54, 1.20) | 0.52 (0.58) | (-0.62, 1.63) |
| 1-2-3 vs. NH | Intercept (6 months) | -3.58 (2.08) | (-7.74, 0.51) | -3.54 (2.37) | (-8.25, 1.06) | -2.49 (3.19) | (-8.50, 3.82) |
| | Slope | 1.64 (0.64) | (0.38, 2.88) | 1.01 (0.48) | (0.07, 1.97) | 0.77 (0.60) | (-0.42, 1.97) |
| 1-3-6 vs. 1-2-3 | Intercept (6 months) | 1.14 (1.48) | (-1.76, 4.05) | 1.49 (1.92) | (-2.41, 5.10) | -0.39 (2.64) | (-5.57, 4.79) |
| | Slope | -0.97 (0.65) | (-2.26, 0.31) | -0.69 (0.54) | (-1.76, 0.38) | -0.26 (0.66) | (-1.55, 1.04) |
| Severity of Hearing Loss | | -0.07 (0.03) | (-0.12, -0.01) | -0.04 (0.03) | (-0.10, 0.02) | -0.06 (0.04) | (-0.14, 0.02) |
| Maternal Education | | 1.31 (0.36) | (0.59, 2.00) | 2.28 (0.38) | (1.54, 3.01) | 2.86 (0.45) | (2.02, 3.77) |

*Note.* A 95% Credible Interval (CI) for the difference in intercepts or slopes that does not include zero indicates a statistically significant difference in that parameter between the groups at the 5% level. Shaded CIs indicate significant differences. NH = normal hearing.

### Children who Met 1-2-3 Benchmarks Compared to Children with Normal Hearing

Figure 1B displays growth trajectories of the 1-2-3 group compared to children with NH. The intercept for the 1-2-3 group was not significantly different than the intercept for children with NH for any language construct. The slope for the 1-2-3 group was significantly higher than the slope for the NH group in the global language and grammar models, indicating children who met 1-2-3 benchmarks had steeper growth in global language and grammar than their hearing peers. Children in the 1-2-3 group did not significantly differ from children with NH in vocabulary growth.

### Children who Met 1-2-3 Benchmarks Compared to Children who met 1-3-6 Benchmarks

Figure 1C displays the growth trajectories of the 1-2-3 versus 1-3-6 group. There was no difference in intercepts or slopes between the groups on any language construct. These results indicate that the two groups of children with HL had similar starting points in global language, grammar, and vocabulary. They also indicate that the two groups demonstrated similar language growth trajectories in these three constructs.

### Outcomes by HL Severity and Maternal Education Level

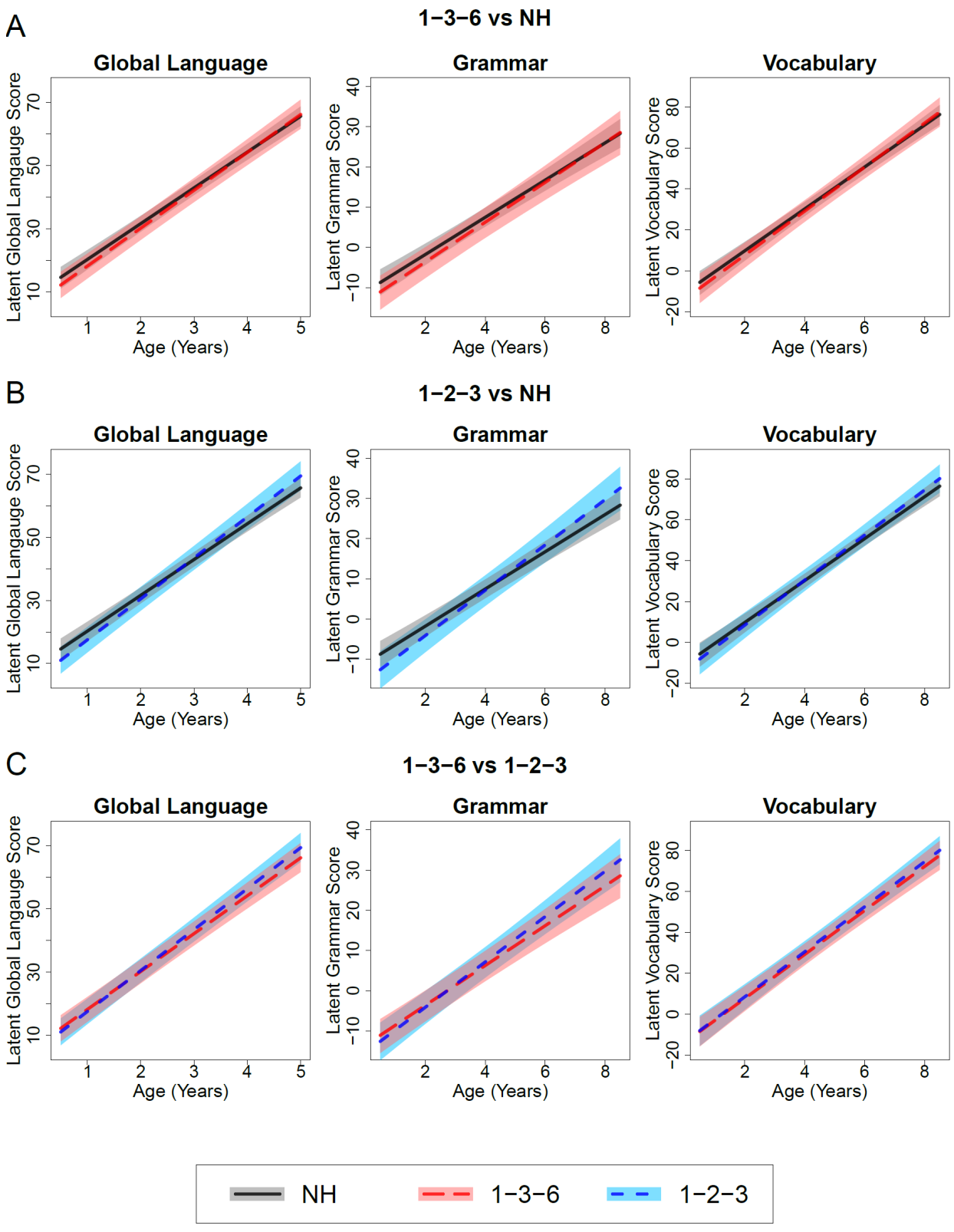
HL severity was significant in the global language model; more severe HL was associated with a reduction in a child's global language score, regardless of whether they met 1-2-3 or 1-3-6 benchmarks. HL severity was not significant in the grammar or vocabulary models. This lack of significance suggests that any degree of HL impacts grammar or vocabulary development. Maternal education level was significant in all three models. Regardless of group or language construct, as maternal education level increased, language scores were higher overall. This effect remained constant over time.

## Discussion

This paper is among the first to prospectively follow a well-described cohort of children with HL who have met 1-2-3 benchmarks or 1-3-6 benchmarks, using a comprehensive battery of parent-report and direct language assessments out to age 8 years. With this cohort, we were able to compare initial language abilities and longitudinal growth trajectories between these two groups, as well as relative to an age- and SES-matched group of children with NH. A major strength of our study is the Bayesian approach to longitudinal analysis, which allowed us to estimate

**Figure 1**

*Population Curves and 95% Credible Intervals Over Time*




*Note.* Group population curves and 95% credible intervals for global language (left), grammar (center), and vocabulary (right) constructs.

latent language traits using different tests and scales across a wide age range. The primary advantage to this approach was that we could avoid problems with measuring language growth using standard scores from norm-referenced tests. This also allowed us to avoid floor or ceiling effects because the test measures were developmentally appropriate at every visit.

Results indicate that children with mild to severe HL who are enrolled in EI by 6 months develop language skills that are on par with their hearing peers. Although this trend is seen in vocabulary for both groups, the children who met the 1-2-3 benchmarks had the additional advantage of showing steeper growth trajectories in global language and grammar compared to children with NH. The children who met the 1-3-6 benchmarks did not show a difference in growth trajectories for these constructs compared to the NH group.

Our findings of a positive effect of EI by 3 or 6 months of age is consistent with other cross-sectional reports. An Australian study compared global language scores for children with moderate to profound HL who received HAs by 3 or 6 months to children who were fit later (Ching et al., 2017). Although they did not specify if children were enrolled in EI at these ages, results indicated that earlier HA fitting led to better language outcomes at age 5, relative to later HA fitting. Further, children with moderate HL who were fitted with HAs by 3 or 6 months showed language scores within the average range for norm-referenced tests (Ching & Leigh, 2020).

Similarly, Yoshinaga-Itano et al. (2017) examined expressive vocabulary outcomes in a cross-sectional sample of 448 children with bilateral mild to profound HL. They found that children who met the 1-3-6 JCIH benchmarks had better vocabulary outcomes than children who experienced delays in diagnosis or early intervention, as measured by the parent-report MacArthur-Bates Communicative Development Inventories. These previous cross-sectional studies support the finding in this study that earlier ages of identification can positively impact global language abilities in children with HL.

The finding of steeper growth in global language and grammar for the 1-2-3 group than children with NH provides evidence that reaching intervention benchmarks at younger ages can result in improved outcomes. Specific to grammar, this finding is consistent with previous research indicating that structural aspects of language (i.e., form) may be a particularly challenging aspect of development for children with mild to severe HL (Tomblin, Harrison et al., 2015; Walker et al., 2020). Form is an especially vulnerable area of language for children with HL because it depends on the processing of fine phonetic details, which are difficult to perceive in the presence of degraded language input. Children enrolled in earlier intervention may experience a multitude of benefits including earlier family access to treatment, HA support, and informational counseling that protect against differential risk in grammar. The positive impact of these benefits is substantiated in the growth trajectories in grammar and global language for the children who received EI by 3 months.

In contrast to the grammar and global language measures, vocabulary growth trajectories did not show differences across groups. This finding may be because lexical measures, particularly assessments of vocabulary breadth, are protected by higher level factors such as contextual cues and redundancy in linguistic input (Moeller et al., 2015; Walker et al., 2019). As a result, the content domain (i.e., vocabulary) is less sensitive to the impact of cumulative auditory experience than structural aspects of language (i.e., morphosyntax).

Implementation of the 1-3-6 benchmarks for children with HL remains challenging. Forty-four percent of our full cohort did not meet all three benchmarks, a proportion that is similar to other studies (Yoshinaga-Itano et al., 2017). The lack of success in meeting diagnostic or EI benchmarks raises questions about whether implementing more stringent benchmarks would have an appreciable impact on language outcomes in children with HL. This analysis indicates that a subsample of children with HL were able to meet the 1-2-3 benchmarks, and there were benefits to language growth in global language and grammar. Setting earlier benchmarks for EI may decrease the number of children who meet the benchmarks, but it would also send a message that EI should proceed as quickly as possible to promote opportunities for language development.

### Limitations

Although this study is one of the first to contrast outcomes between children who met the current 1-3-6 benchmarks for early intervention with children who met an earlier 1-2-3 criterion, there are several important limitations that must be considered. The study sample was relatively homogeneous compared to groups of children with HL in previous studies or the general population. The sample included only children from English-speaking homes without additional disabilities. It should be noted that clinical caseloads are rarely this homogenous: Approximately 40% of children with HL have an additional disability and around 50% are from a culturally or linguistically diverse population (Gallaudet Research Institute, 2003). Further, the participants all had bilateral hearing loss in the mild to severe range. The exclusion of children with profound HL allowed us to control for the effects of device type (i.e., all children wore HAs; no children had cochlear implants) and communication mode (all children relied entirely on spoken English), but limited investigations of the full range of hearing levels. This is an ideal cohort for examining the effects of HL and EI on language outcomes with minimal confounds, but these results likely overestimate outcomes from more diverse populations.

### Conclusions

This study documents the positive impact of early diagnosis and intervention of HL on language outcomes in children with congenital HL. Children who received EI by 6 months of age were able to maintain language growth at a level commensurate to hearing peers. These parallel growth trajectories were evident in vocabulary for children who met the recommended 1-3-6 benchmarks as well as the accelerated 1-2-3 benchmarks. However, children who

met 1-2-3 demonstrated steeper growth in global language and grammar relative to the NH group. These findings provide preliminary evidence to support the potential effects of very early intervention in children with HL, but additional data are needed before we can encourage states to devote the time and resources needed to implement an accelerated EHDI timeline.

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# Hearing Screening in North Carolina's NICU and Well-Baby Nurseries: Impact of JCIH 2019 and COVID-19

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## Abstract

**Purpose:** Over an 18-month period in 2020–2021, the North Carolina Early Hearing Detection and Intervention (EHDI) program in collaboration with the North Carolina Leadership Education in Neurodevelopmental and Related Disabilities (LEND) program conducted a statewide examination of newborn hearing screening practices in North Carolina's 24 Neonatal Intensive Care Units (NICU) and 86 well-baby nurseries to determine how current protocols and procedures conform to those recommended by the Joint Committee on Infant Hearing (JCIH) in its Year 2019 Position Statement: Principles and Guidelines for Early Hearing Detection and Intervention Programs. The COVID-19 pandemic emerged during the study period and motivated a second aim, to examine the impact of the pandemic on infant hearing screening.

**Results:** Our findings revealed that the hospitals in North Carolina are fully committed to their hearing screening programs as demonstrated by a 100% response rate and numerous strengths in both the NICU and well-baby nurseries. Even so, for many hospitals we identified opportunities for program development or improvement based on JCIH 2019 recommendations, especially those concerning oversight of the screening program by a pediatric audiologist, direct referral to an audiologist for NICU babies who fail the in-hospital screening, and audiology referral for well babies who fail the outpatient rescreen. Following the investigation, the NC-EHDI program has worked in partnership with hospitals to provide information, technical assistance, and resources based on our findings and recommendations. The authors would be happy to share the survey instruments and other resources developed for this project with EHDI programs in other states interested in conducting a similar study.

**Keywords:** Hearing Screening, JCIH 2019, COVID-19, NICU, Well-Baby, Infant

**Acronyms:** AABR = automated auditory brainstem response; ANSD = auditory neuropathy spectrum disorder; cCMV = congenital cytomegalovirus; EHDI = Early Hearing Detection and Intervention; LEND = Leadership Education in Neurodevelopmental and Related Disabilities; OAE = otoacoustic emissions

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Permanent hearing loss[1] is the most common condition identified through newborn screening, detectable in 1.7 newborns per 1000 in the general population (CDC, 2019). The prevalence for both cochlear hearing loss and auditory neuropathy spectrum disorder (ANSD) is significantly higher for infants requiring hospitalization in the neonatal intensive care unit (NICU; Berg et al., 2005; Hille et al., 2007; Robertson et al., 2009; White et al., 1994; Xoinis et al; 2007). Accordingly, practice guidelines published by the Joint Committee on Infant Hearing (JCIH, 2019) recommend separate hearing screening protocols for the NICU and well-baby nurseries.

The JCIH was established in 1969 with representatives from audiology, otolaryngology, pediatrics, and nursing. Today, representatives to the JCIH include 13 organizations, each dedicated to ensuring early identification, intervention, and follow-up care for infants and young children with hearing loss. The Joint Committee's primary activity has been publication of position statements summarizing the status of infant hearing screening along with recommendations for preferred practice in early identification and intervention for newborns and infants with or at risk for hearing loss (CDC, 2021). Over its 50+ year history, the JCIH has published eight position statements. The current clinical practice guideline is the JCIH *Year 2019 Position Statement: Principles and Guidelines for Early Hearing Detection and Intervention Programs* (JCIH, 2019).

For hearing screening in the NICU, JCIH 2019 reaffirmed the Joint Committee's previous position statement (JCIH, 2007), which recommended automated auditory brainstem response (AABR) as the sole hearing screening technology for infants admitted to the NICU for more than 5 days. Also reaffirmed for NICU hearing screening was direct referral to an audiologist for rescreening and, if indicated, comprehensive audiological evaluation including diagnostic ABR for infants who fail the in-hospital screen. For hearing screening in the well-baby nursery, the JCIH currently recommends AABR and otoacoustic emissions (OAE) technologies, alone or in combination, and outpatient rescreening for babies who do not pass the in-hospital screen (JCIH, 2019). A notable change in JCIH 2019 is the recommendation regarding follow-up screening of well babies who do not pass an initial AABR. For infants in the well-baby nursery who fail an AABR screening, the previous position statement, JCIH 2007, recommended they *not* be rescreened and passed using OAE technology because of presumed risk for ANSD. Although AABR is still the preferred protocol in JCIH 2019, because of the low incidence of ANSD in the well-baby population and challenges associated with access to outpatient rescreening, JCIH currently advises that rescreening of well-babies may be accomplished using either OAE or AABR. These and other JCIH 2019 recommendations pertaining to hearing screening in the NICU and well-baby nurseries are summarized in Table 1.

An overarching theme within JCIH 2019 that applies to both settings is the recommendation for greater audiology oversight of hearing screening programs in all state/territory hearing screening programs, at both the systems level and the individual programs level (Table 2). JCIH 2019 recommends that an audiologist with experience in evaluating newborns and young children be involved in the development and oversight of each component of the hearing screening program, including selection of screening technology based on the population to be screened, with confirmation that equipment calibration performed by the manufacturer is completed in a manner consistent with purported screening parameters. JCIH 2019 also advises hospitals and agencies to designate a physician/provider to oversee the medical aspects of the EHDI program.

For decades, the practice guidelines published by JCIH have impacted hearing screening protocols throughout the United States and beyond. North Carolina's Early Hearing Detection and Intervention (NC-EHDI) Program was established in 2000, following a legislative mandate in 1999 requiring birthing hospitals to provide physiologic screening for hearing loss prior to discharge (Fort, 2017). Soon after the establishment of NC-EHDI, a group of stakeholders from across the state formed an advisory committee to guide the implementation, development, and coordination of EHDI services. Although initially focused on the implementation of newborn hearing screening, NC-EHDI and its advisory committee soon expanded its scope to include a variety of issues related to early identification, diagnosis, and intervention services for children with permanent hearing loss. NC-EHDI is now divided into 10 regions of the state, each served by one or more regional consultants.

For purposes of program evaluation and improvement, initially the primary aim of this study was to examine the current status of newborn hearing screening programs in the state's 24 Level III and Level IV NICUs and 86 well-baby nurseries to determine how current protocols and procedures conform to those recommended by JCIH 2019. The COVID-19 pandemic emerged during the NICU study period and motivated a second aim, to examine how the pandemic was impacting infant hearing screening in both the NICU and well-baby nurseries.

## Method

Data collection over an 18-month period involved collaboration between the NC-EHDI program and the North Carolina LEND (Leadership Education in Neurodevelopmental and Related Disabilities) Program. LEND is a federally funded, interdisciplinary program that provides graduate-level training, technical assistance, continuing education, and consultation to states regarding screening, diagnosis, advocacy, and treatment for neurodevelopmental and related conditions (HRSA, 2021). Eight LEND audiology trainees from the University

[1]The authors recognize the importance of culturally sensitive language when referring to content related to people who are deaf or hard of hearing. Consistent with JCIH 2019, the term *hearing loss* is used here to clearly convey audiological concepts and conditions. Also consistent with JCIH 2019, we use the term *fail* in reference to infants who do not pass their newborn hearing screening.

**Table 1**

*JCIH 2019 Recommendations for Hearing Screening in the NICU and Well-baby Nurseries*

| Summary of JCIH 2019 Newborn Hearing Screening Recommendations | |
|---|---|
| Well-Baby | NICU |
| Interpretive Criteria<br>• Criteria for hearing screening outcomes should demonstrate both sensitivity and specificity due to the prevalence of hearing loss in infants, manufacturer-reported test performance, and the goal of identifying elevated hearing thresholds that can affect spoken language development.<br>• Screening technology that automates results considering both sensitivity and specificity should be used to optimize consistency among tests. | |
| Calibration of hearing screening equipment<br>• Due to a lack of universal standard, calibration should be performed based on manufacturer specifications. | |
| Timing of newborn hearing screening<br>• Infants should have their hearing screened as close to discharge as is feasible. However, there should be ample time to perform a repeat screen should the infant not pass the first screen.<br>• If an infant fails the initial screen, the second screening should be performed at least several hours after the first screen.<br>• Infants that present with congenital aural atresia in one or both ears or with visible pinna/ear canal deformity such as stenosis or severe malformation should not be screened in either ear but should be referred for diagnostic audiologic evaluation immediately upon discharge. | Timing of newborn hearing screening<br>• Although infants can be tested while in the NICU, it is not always feasible for these children to be tested prior to 1 month of age. In these situations, arrangements should be made to test the infant as soon as medically possible. |
| Screening protocols in the well-baby nursery<br>• An acceptable pass result consists of a pass result for both ears in a single screening session using either technology prior to hospital discharge.<br>• Due to the low incidence of auditory neuropathy in the well-baby nursery, initial screening as well as any repeat screening (second in-hospital screen) can be performed with either OAE or AABR technology.<br>• However, the recommendation to rescreen using only AABR technology for infants who fail their initial screen performed with AABR technology is the Committee's preferred recommendation.<br>• Rescreening with OAE after a failed initial screen using AABR is acceptable, though an infant with auditory neuropathy in the well-baby nursery will be missed. | Screening protocols in NICU nursery<br>• Due to increased rates of hearing loss and auditory neuropathy in this population, screening should solely be performed using AABR.<br>• Although not recommended at this time it was noted that screening with both AABR and OAE can aid in preventing infants with mild-moderate hearing loss from being missed.<br>• If rescreening is necessary, patient should be referred directly to an audiologist for a comprehensive audiologic evaluation. |
| Communication and documentation of results<br>• Families should be informed in such a way that is comforting to the family while still emphasizing the importance of follow-up. It is recommended that this be done using the scripts composed by the National Center for Hearing Assessment and Management (NCHAM) or the state EDHI program.<br>• To aid in preventing loss to follow up, results (including the method of testing) should be given to the infant's medical home. | |
| Rescreening in the outpatient setting<br>• For well-infants, a single rescreening of both ears within the same session should be conducted within 1 month of age, or as soon as possible after discharge from the hospital.<br>• If the infant does not pass the rescreening, in either ear, the child should immediately be referred to a pediatric audiologist for diagnostic ABR testing. If the rescreening was performed by a pediatric audiologist, a diagnostic evaluation should be conducted within the same appointment. | |
| Rescreening in the medical home<br>• Screenings conducted within the medical home should be limited to a rescreening, as initial screenings should be completed at the infant's birthing center.<br>• Rescreening should be conducted in a quiet environment by a trained professional using approved manufacturer calibrated equipment (OAE/ABR).<br>• Rescreening should be performed on both ears in the same session regardless of initial screening results. | |
| Improving EHDI loss-to-follow-up/loss-to-documentation rates<br>• States should not only offer newborn hearing screening to all out-of-hospital births, but also be prepared to share results with neighboring states when necessary.<br>• When a child is transferred to a different hospital, appropriate documentation should be sent to the receiving hospital specifying if screening has been performed. In cases where the infant is discharged prior to screening an outpatient screening should be scheduled as soon as possible. | |

*Note.* JCIH = Joint Committee on Infant Hearing; EHDI = early hearing detection and intervention; AABR = automated auditory brainstem response; OAE = otoacoustic emissions.

**Table 2**

*Summary of Joint Committee on Infant Hearing (JCIH) 2019 Recommendations Regarding the Role of the Audiologist in Newborn Hearing Screening Programs*

**Systems Level Audiology Oversight**

- Periodic on-site and/or remote surveillance of individual hospital programs
- Oversight and participation in writing policies and procedures
- Monitoring of program statistics
- Development of referral pathways and timelines with community resources and the state Early Hearing Detection and Intervention (EHDI) program

**Hospital Level Audiology Oversight**

- Selection of screening technology
- Confirmation of equipment calibration
- Protocols for training and certifying competence of screeners
- Development of policies, procedures, and protocols
- Quality assurance procedures; program staffing requirements and relevant assignments of staff/team members
- Procedures for discharge or transfer plans; assurance of, "acceptable, independent, on-site oversight by an audiologist who is either employed by the hospital or is otherwise independent of the contracted entity in screening programs where services are contracted through an outside entity" (JCIH, 2019 p. 5-7).

of North Carolina's Doctor of Audiology (AuD) program and their faculty advisors worked with NC-EHDI staff and regional consultants to identify an appropriate individual from each hospital. Prospective participants were contacted by email or by phone in advance to confirm their participation. The LEND trainees also assisted with survey development, correspondence with NC-EHDI staff, data analysis, preparation of hospital reports, and manuscript preparation. Our goal was to recruit the participation of every NICU and well-baby nursery in the state. Some of the hospitals responded immediately, others within a few days. If there was no response after approximately two weeks, an email reminder was sent. If there was still no reply, a phone inquiry was made, and, in a few cases, the study team enlisted the assistance of the NC-EHDI regional consultant.

### NICU

In February 2020, a 25-item survey was pilot tested with personnel from two hospitals and distributed electronically to a representative from each of the 24 hospitals in North Carolina with a Level III or Level IV NICU using Qualtrics, a web-based survey tool (Qualtrics, Provo, Ut). Level III and IV NICUs were targeted because they care for the most critically ill newborns and those at highest risk for permanent hearing loss. Level III and IV NICUs provide care for babies born prematurely or with low birth weight, including those with critical illness or conditions requiring sustained life support. They also provide advanced imaging and a full range of respiratory support. Level IV NICUs care for the most complex and critically ill newborns including those requiring medical and surgical specialists (American Academy of Pediatrics, 2012).

The NICU survey included questions regarding screening personnel, technologies used for hearing screening, and protocols for referral and follow-up. It also included questions related to training and continuing education for screeners as well as challenges associated with hearing screening in the NICU. Additionally, the role of audiology in oversight of the hearing screening program was investigated, as was the impact of COVID-19. The hospital representatives (chosen based on recommendations from NC-EHDI regional consultants) included nurses, nurse managers, administrators, and audiologists. Because the COVID-19 pandemic began during the NICU study period and was not part of the initial survey, a follow-up study was conducted in January 2021, to investigate how the pandemic was impacting hearing screening in the NICU. The COVID-19 follow-up survey asked NICU representatives if the pandemic had affected newborn hearing screening and if so, to describe the effects.

### Well-Baby Nurseries

One year following distribution of the NICU survey a second phase of the project addressed North Carolina's 86 well-baby nurseries. In February 2021, following pilot testing in two hospitals, a 32-item Qualtrics (Provo, Ut) survey was distributed to all 86 birthing hospitals in North Carolina and again directed to an individual recommended by the hospital's NC-EHDI regional consultant. As with the NICU survey the participation of each hospital representative was confirmed prior to distribution. Because the COVID-19 pandemic was known to be impacting hearing screening in the well-baby nurseries, the survey included two parts. Part 1 consisted of 21 questions pertaining to hearing screening prior to the onset of the pandemic, and Part 2 included 11 questions related to the impact of COVID-19 on well-baby hearing screening. Survey questions for the well-baby nurseries included screening personnel, screening technologies, and protocols for referral and follow-up. Also included were questions related to training and continuing education for screeners as well as challenges associated with hearing screening. In addition, hospitals were asked if there was a protocol for referral of infants with aural atresia or other visible outer

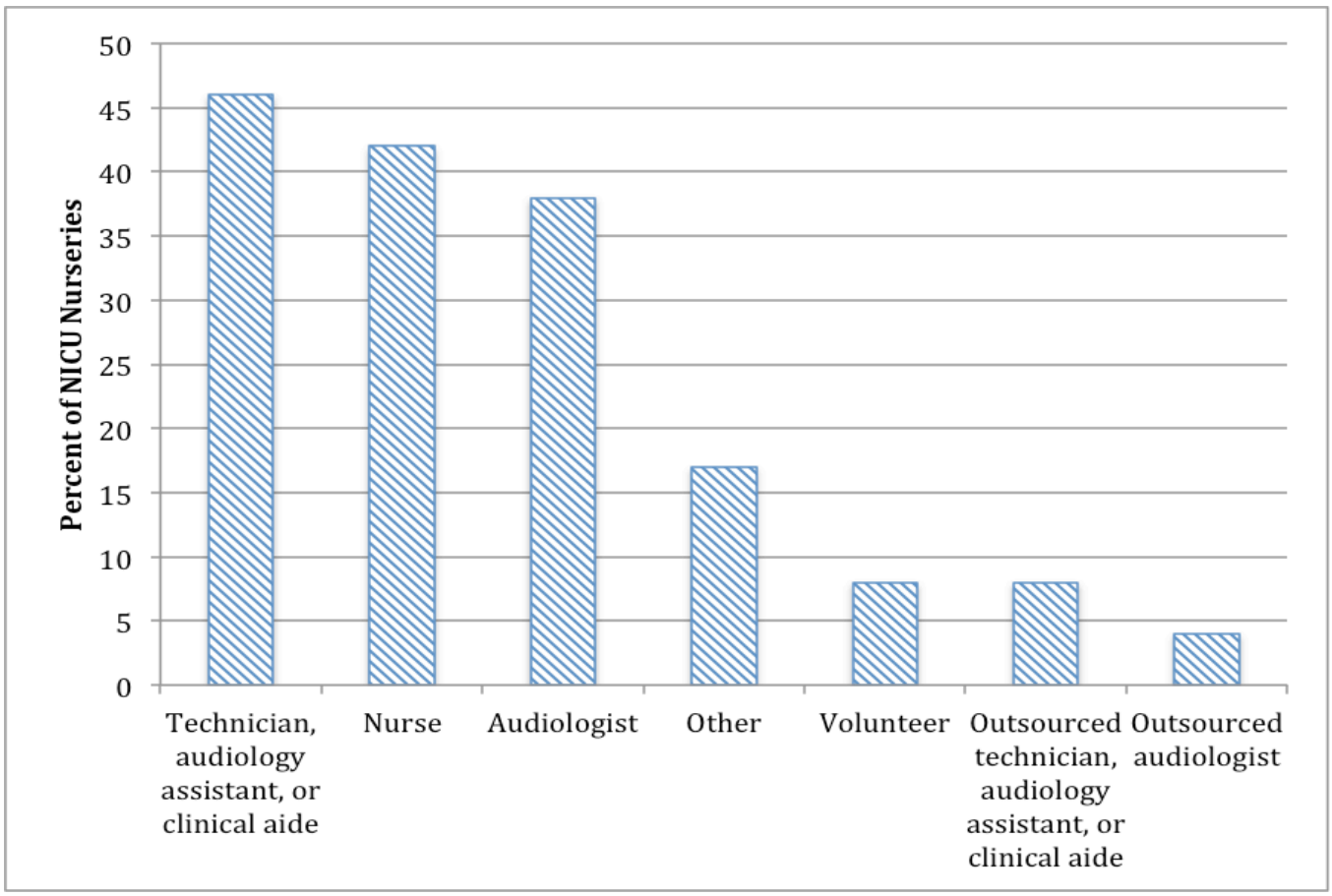
ear anomalies, and they were asked if they were currently providing or planning to conduct screening for congenital cytomegalovirus (cCMV). As with the NICU survey, hospital representatives were also asked if an audiologist provided oversight of the hearing-screening program.

## Results

The results of this investigation confirmed that North Carolina's hospitals are fully committed to their hearing screening programs. The information we requested was reported promptly and thoroughly with a 100% response rate for all 24 NICUs and all 86 well-baby nurseries.

**Figure 1**

*Personnel Responsible for Conducting Hearing Screening in the NICU*




*Note.* The total exceeds 100% because respondents could select more than one option.

## NICU Nurseries

### *Screening Personnel*

A hospital technician or assistant employed by the institution is most likely to administer the in-hospital screening (Figure 1).

### *Screening Technology*

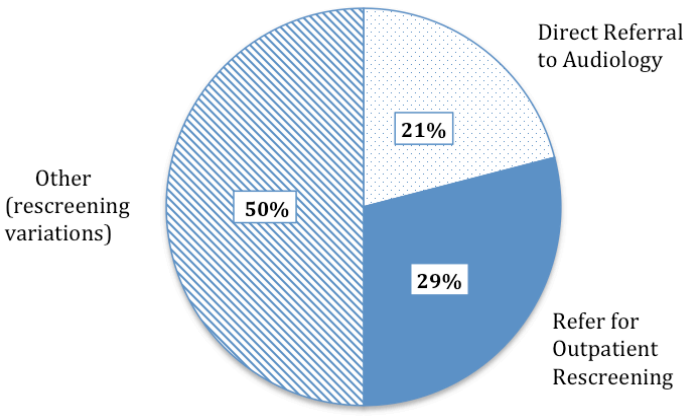
All 24 NICUs reported using AABR; however, two hospitals reported combined use of AABR and OAE. None of the NICUs reported using OAE only.

### *Referral and Follow-up*

For the 24 NICUs, five (21%) reported direct referral to an audiologist for babies who fail the hearing screening; seven (29%) reported referral for outpatient rescreening; and 12 (50%) reported a variety of other referral strategies (Figure 2).

**Figure 2**

*Referral of NICU Infants who Fail the Inpatient Hearing Screening*




For infants *readmitted* to the NICU with a condition or treatment associated with a risk factor for hearing loss, one NICU reported that all infants are rescreened prior to discharge, and 15 (63%) reported that infants may be rescreened prior to discharge based on certain conditions such as exposure to ototoxic medications, newly identified

risk factors, previous screening results, or physician orders. Survey respondents for the remaining eight (33%) were not aware of a rescreening protocol for readmitted infants.

### *Training and Continuing Education for Screeners*

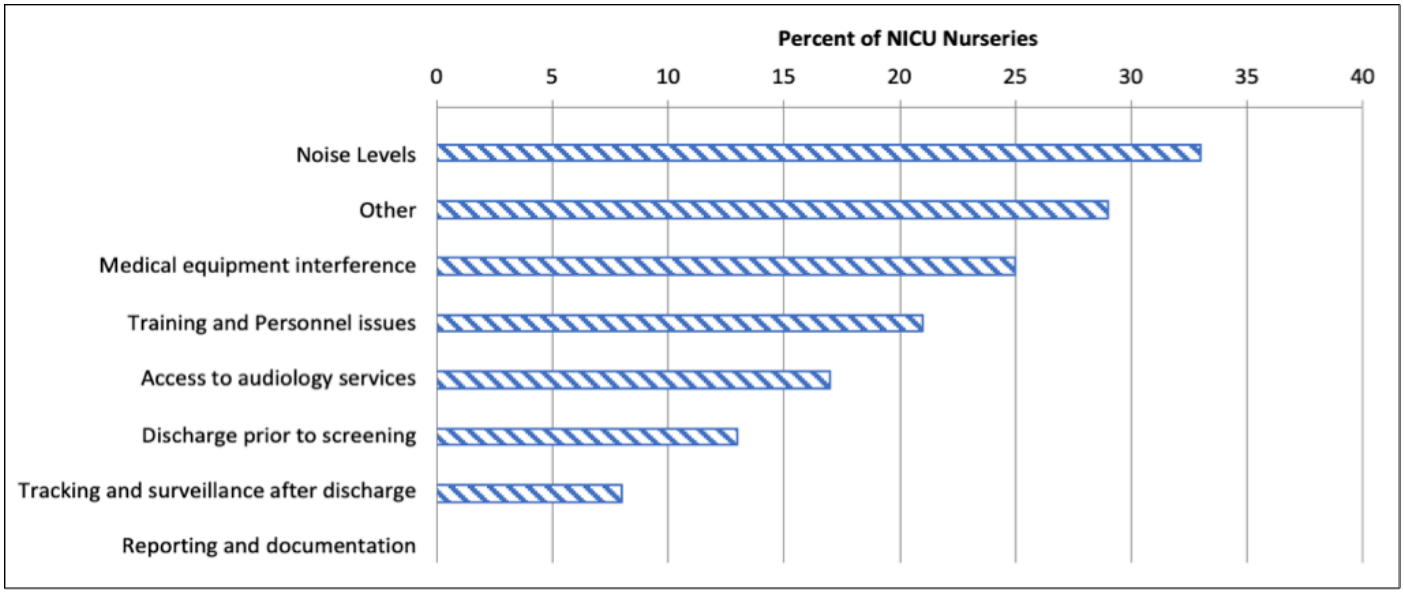
The frequency of training and continuing education among the 24 NICUs varied considerably. Eight hospitals (33%) reported 1 to 2 times per year and 13 (54%) reported no regular continuing education. The remaining three employed audiologists and/or contracted providers whose continuing education requirements are unknown.

### *Challenges*

Challenges associated with hearing screening in the NICU, summarized in Figure 3, included a variety of issues such as noise levels, medical equipment interference, training and personnel issues, limited access to audiology services, discharge prior to screening, and tracking/surveillance after discharge. None of the NICUs reported challenges related to reporting and documentation.

**Figure 3**

*Challenges Associated with Hearing Screening in the NICU*




*Note.* The total exceeds 100% because respondents could select more than one option.

### *Audiology Oversight*

Sixteen NICUs (67%) reported direct oversight of the screening program by an audiologist.

### *COVID-19*

The COVID-19 pandemic had not emerged when the planning began for the NICU project in the fall of 2019. Because of the potential impact of the pandemic on hearing screening in the NICU, a follow-up survey was conducted in November 2020. Responses from all 24 NICUs indicated that COVID-19 did not appreciably affect hearing screening in the NICU other than a few hospitals that noted a change in screening location for babies requiring a second in-hospital screen, or a delay in screening if the baby had been exposed to COVID-19 or was awaiting test results. One hospital reported that babies with COVID-19 positive mothers were required to wait 30-45 days before a hearing screening could be provided.

## Well-Baby Nurseries

### *Screening Personnel*

A nurse or hospital technician was most likely to administer the in-hospital screening (Figure 4) and a nurse or pediatrician was most likely to provide screening results to families and discuss recommendations for babies who failed the in-hospital screening (Figure 5). Hospital technicians, certified nursing assistants, administrative support staff, and audiologists were other providers who discussed screening results with families.

### *Screening Technology*

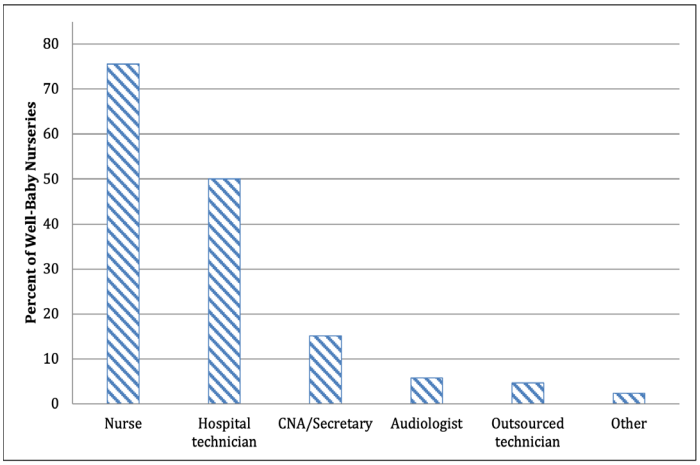
As summarized in Table 3, for the in-hospital screening, 76 (88%) of the well-baby nurseries reported using AABR only and five (6%) reported using OAE only. For infants requiring outpatient rescreening, 61 (71%) reported AABR and 10 (12%) reported OAEs. A few hospitals reported a combination of screening technologies or stated that outpatient rescreening was not conducted at their birthing hospital.

### *Referral and Follow-up*

As summarized in Figure 6, 51 well-baby nurseries (60%) reported direct referral to an audiologist following a failed outpatient rescreening; 20 (23%) reported referral to a pediatrician or other primary care provider; and seven (8%) reported referral to an ear nose and throat physician. The remaining eight nurseries (9%) reported some other protocol for referral of babies who fail the outpatient rescreen.

**Figure 4**

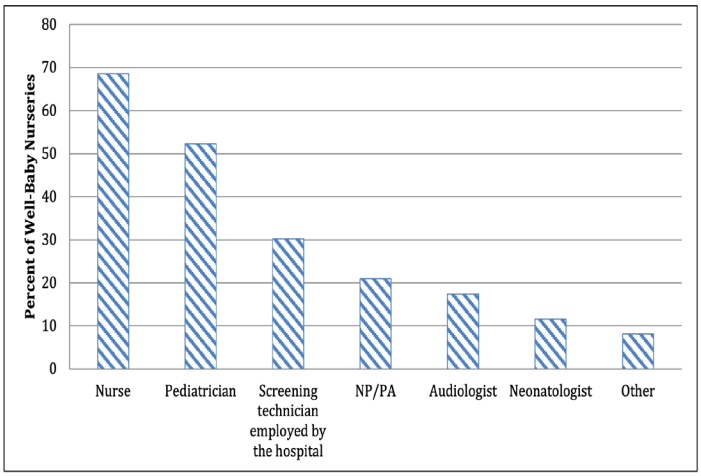
*Personnel Responsible for Conducting Newborn Hearing Screening in the Well-Baby Nursery*




*Note.* The total exceeds 100% because respondents could select more than one option.

**Figure 5**

*Personnel Responsible for Informing the Family of In-Hospital Screening Results for the Well-Baby Nurseries*




*Note.* The total exceeds 100% because respondents could select more than one option.
NP/PA = Nurse Practitioner/Physician's Assistant.

**Table 3**

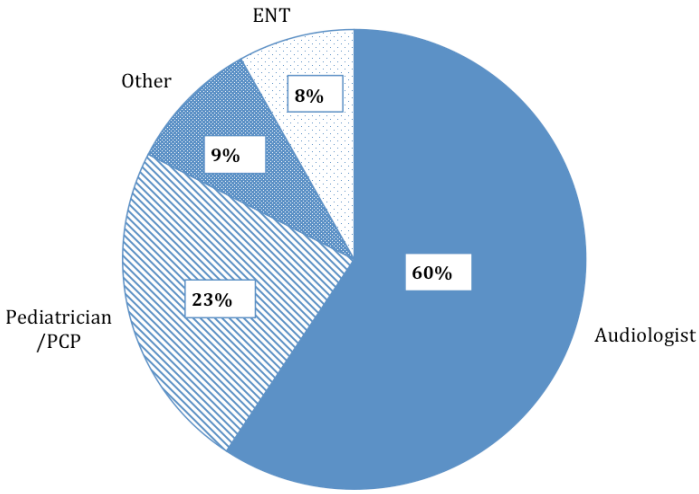
*Screening Technology Used in the Well-baby Nursery for Initial In-Hospital Screening and Outpatient Rescreens for Infants who Fail the In-Hospital Screen*

| | In-Hospital Screen | Outpatient Rescreen |
|---|---|---|
| **Auditory Brainstem Response (ABR) only** | 76 (88%) | 61 (71%) |
| **Otoacoustic emissions (OAE) only** | 5 (6%) | 10 (12%) |
| **OAE or ABR** | 3 (3%) | 1 (1%) |
| **OAE followed by ABR** | 9 (10%) | 0 |
| **ABR followed by OAE** | 0 | 2 (2%) |
| **Not applicable** | 1 (1%) | 12 (14%) |

*Note.* The total exceeds 100% because respondents could select more than one option.

**Figure 6**

*Referral of Well Babies who Fail the Outpatient Rescreen*




*Note.* ENT = ear, nose, and throat doctor; PCP = primary care physician.

### *Training and Continuing Education for Screeners*

Most well-baby nurseries (56%) reported annual continuing education; however, nearly half (43%) reported no regular continuing education for screening personnel. Of those reporting regular training, in-person was the most common method followed by online modules, electronic materials, and competency exams.

### *Challenges*

Nearly all well-baby nurseries reported challenges associated with hearing screening. The most frequently cited challenges were associated with equipment issues and tracking following discharge. A number of other challenges were also noted (see Figure 7).

### *CMV Screening*

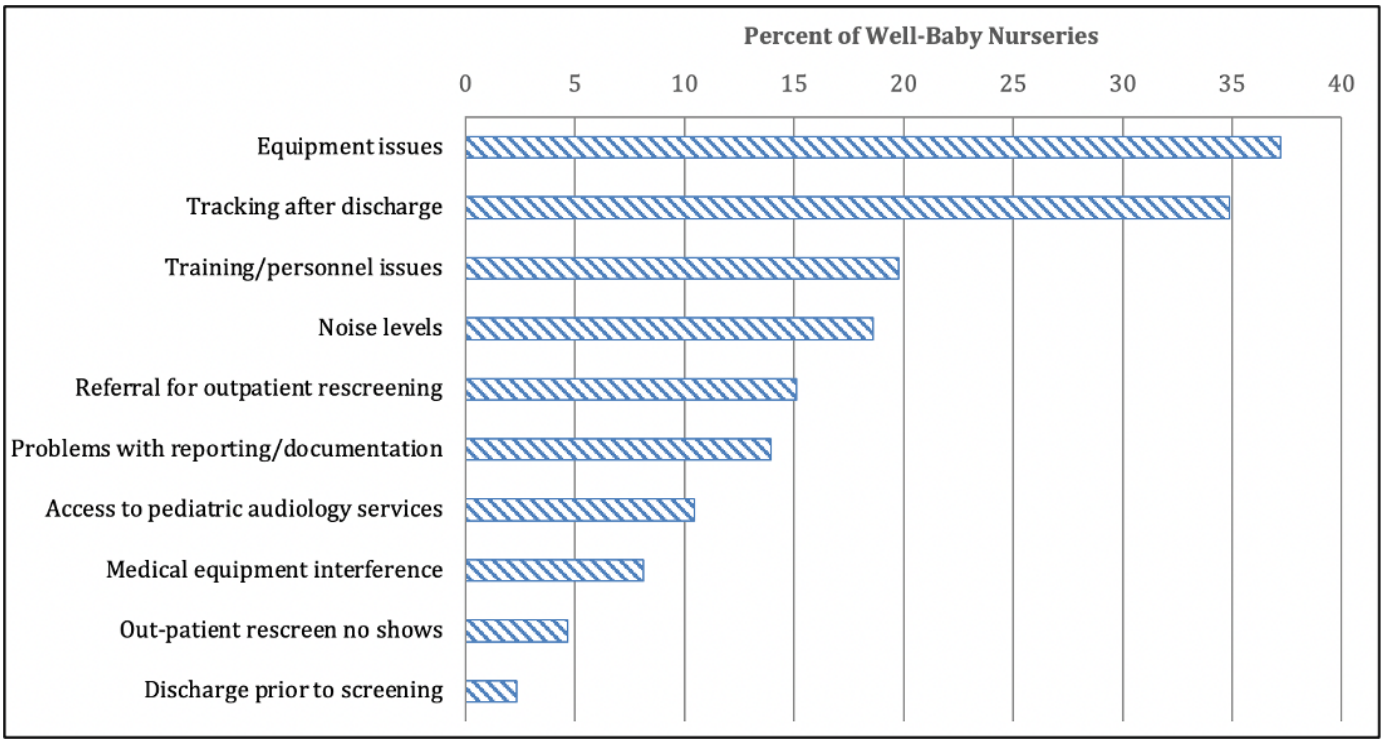
Eleven (13%) well-baby nurseries reported screening for CMV during the study period and seven (8%) indicated they were planning to implement CMV screening in the future.

### *Aural Atresia*

Twenty-seven (13%) well-baby nurseries reported a formal protocol for infants born with aural atresia and other visible ear anomalies. Protocols included referral to an

**Figure 7**

*Challenges Reported by Well-Baby Nurseries Prior to Onset of the COVID-19 Pandemic*




*Note.* The total exceeds 100% because respondents could select more than one option.

audiologist, pediatrician, or ENT regardless of screening outcome. Fifty-nine (69%) reported not having a formal protocol for referral of infants with aural atresia or other visible ear anomalies.

### *Audiology Oversight*

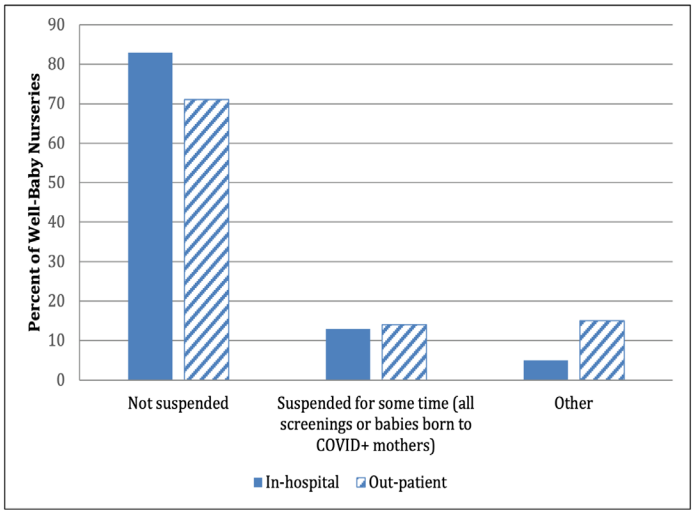
Twenty-six (30%) well-baby nurseries reported direct oversight of the screening program by an audiologist.

### *COVID-19*

The COVID-19 pandemic had already emerged at the beginning of the well-baby screening phase of the study and it impacted both in-hospital screening and outpatient rescreening. As summarized in Figure 8, the outcomes clustered into three categories. Seventy-one hospitals (83%) reported no COVID-19 related suspension of in-hospital hearing screening and 61 hospitals (71%) reported no suspension of outpatient rescreening. Temporary suspension of in-hospital hearing screening was reported by 11 hospitals (13%) and by 12 (14%) for outpatient rescreening. The remaining hospitals (Other) reported suspension of initial inpatient hearing screening if the mother was found to be COVID positive. In those cases, an infant was usually scheduled for later outpatient screening. Many well-baby nurseries implemented additional precautions to enable screening of babies with COVID-positive mothers, and some hospitals suspended outpatient screening temporarily but with added protocols to mitigate delays or loss to follow-up.

**Figure 8**

*Impact of the COVID-19 Pandemic on Hearing Screening in the Well-Baby Nurseries*




### Recommendations to Hospitals

Many strengths, reflected by protocols and procedures consistent with JCIH 2019 recommendations, were noted for all screening programs and for some there were no recommendations for improvement. For many of the NICUs, however, the findings resulted in one or more specific recommendations.

### NICU Nurseries

In November 2020, the study team contacted each NICU representative to thank them for their participation and provide two documents: a statewide summary of aggregate findings and an individualized report with recommendations, if any, for each hospital. Recommendations were made for 20 (83%) of the 24 NICUs. The statewide aggregate report included a summary of screening technologies employed; audiology oversight of screening programs; screening personnel; challenges associated with NICU hearing screening; and next steps after a failed in-hospital screening. The individualized reports highlighted areas perceived to be strengths of the program, as well as any recommendations for programmatic modification based on JCIH 2019 recommendations. This information was also provided to the NC-EHDI regional consultant for each hospital and to the NC-EHDI Coordinator. In February 2021, a final report was submitted and presented to the NC-EHDI advisory committee. The study team considered all recommendations to be important but identified three as immediate priorities: (a) babies who do not pass the in-hospital hearing screening should be referred directly to a pediatric audiologist for follow-up, (b) clarification should be sought regarding how a few of the NICUs were using OAEs in conjunction with AABR, and (c) need for confirmation of rescreening for infants readmitted to the NICU or pediatric intensive care unit who are at risk for permanent hearing loss. Recommendations also included greater oversight of the screening program by an audiologist if needed, and more systematic and ongoing continuing education for screening personnel along with suggested resources such as those developed by NCHAM. With submission of the final report, the study team concluded the NICU study. The NC-EHDI regional consultants, each of whom provided the contact person for the 24 NICUs, have since communicated directly with the hospitals in their regions to offer guidance, technical assistance, and resources.

### Well-Baby Nurseries

Because of the large number of well-baby nurseries, variability in the contact person/s for some hospitals, and the potential for ongoing changes related to COVID-19, a separate report was not sent to each hospital as was done for the NICUs. Instead, the study team summarized key findings for NC-EHDI and its regional consultants to share with well-baby nurseries in each region. In addition to a summary of overall findings, the report highlighted the following needs for some hospitals based on JCIH 2019 recommendations: (a) direct referral to an audiologist following a failed outpatient rescreening, (b) regular educational in-service training for program personnel, (c) oversight of the program by an audiologist with experience in evaluating newborns and young children, and (d) implementation of a protocol for referring infants with congenital aural atresia or visible pinna/ear canal deformities for audiologic assessment. The report also emphasized the need for ongoing monitoring of potential impacts from the COVID-19 pandemic.

## Discussion

The primary aim of this investigation was to assess newborn hearing screening practices in North Carolina's NICU and well-baby nurseries, and to determine how current protocols and procedures compared to those recommended by JCIH 2019. A second aim was to assess the impact of the COVID-19 pandemic on infant hearing screening.

### Hearing Screening in the NICU

As expected, NICU hearing screening personnel included a variety of healthcare providers such as hospital-based technician/assistants, nurses, or audiologists. We were also interested in the screening technology employed in our NICUs and found, unsurprisingly, that nearly all NICUs reported using AABR only, with none using OAE as the sole screening technology. However, two NICUs reported using AABR and OAE. Although some NICU infants are not at risk for neural hearing loss, JCIH recommends AABR as the sole hearing screening technology because of its ability to detect ANSD, a condition known to be substantially more prevalent in this population (JCIH, 2007, 2019). This finding provided an opportunity for NC-EHDI consultants to remind NICUs in their regions of this longstanding JCIH recommendation.

An important finding related to NICU screening was that many hospitals were not directly referring to a pediatric audiologist when an infant fails the NICU hearing screening. Because of the high prevalence of sensorineural hearing loss in the NICU population, and the importance of timely diagnosis and intervention, JCIH, in both the 2007 and 2019 position statements, recommends direct referral of infants who fail their NICU hearing screening to an audiologist for rescreening and, if indicated, for a diagnostic ABR evaluation (JCIH, 2007, 2019). Although this requires the infant to be medically stable, direct referral to an audiologist is needed as soon as possible to promote early diagnosis and intervention, which in some cases can begin while the infant is still in the NICU (Grosnik & Baroch, 2020). Sapp et al. (2020) found that hearing screening and diagnostic evaluations are often delayed for NICU infants because of medical factors and lengthy NICU admissions, noting that specific clinical guidelines should be considered for this population to facilitate the timing and delivery of hearing healthcare. Fortunately, a revised protocol resulting in direct referral to an audiologist should be straightforward to implement if NICUs choose to do so. The need for direct referral to an audiologist was cited as a top priority in our report to the NICUs, and according to the NC-EHDI manager, many hospitals that were not following this JCIH recommendation have since modified their referral criteria. On a related topic, although many hospitals reported screening of infants *readmitted* for a condition or treatment associated with a risk factor for hearing loss, some appeared to lack specific protocols. NC-EHDI has also worked with hospitals to address this issue.

Our findings also revealed a perceived need among many NICUs for more systematic training and continuing

education related to hearing screening. Hospitals are required to assume responsibility for ensuring the qualifications of their screening personnel and most appear to be making a deliberate effort to do so. We have observed anecdotally, however, that some hospitals are unaware of training materials available to hearing screeners such as the Newborn Hearing Screening Training Curriculum (NHSTC) developed by NCHAM and recently updated in 2020. The NHSTC is an online interactive competency-based course available at no charge and designed to provide a thorough understanding of the components necessary for conducting quality newborn hearing screening based on JCIH recommendations (NCHAM, 2020).

Regarding challenges encountered with NICU hearing screening, we were surprised by the number and variety of issues. Excessive noise was cited most frequently, but the reported challenges included a range of other issues. The current study did not permit exploration of details associated with these challenges, but the information has been used by NC-EHDI for further inquiry and follow-up.

**Hearing Screening in the Well-Baby Nursery**

As with the NICUs, personnel consisted of a variety of healthcare providers. More than 80% of the nurseries reported that a hospital technician or nurse provides the screening. Also noted for approximately half of the well-baby nurseries, if a baby does not pass the in-hospital screen, a nurse or pediatrician is the professional most likely to discuss recommendations with the family. Communication with families regarding screening outcomes is known to have a significant effect on follow-up (Pynnonen et al., 2016). JCIH 2019 states that results of hearing screening should be conveyed immediately to the family so that they are aware of the screening outcome and the importance of follow-up when indicated. Also included in the JCIH 2019 position statement are resources and specific recommendations for documentation and communication with families.

Regarding choice of screening technology, most of the well-baby nurseries reported using AABR for in-hospital screening and for outpatient rescreening. Although JCIH 2019 endorses both technologies, AABR has the potential for detecting ANSD and related retrocochlear dysfunction. Also noted in JCIH 2019, however, is evidence of OAE screening having the potential for greater sensitivity to mild hearing losses. Although an ideal protocol might involve both technologies, practical considerations associated with multiple technologies are acknowledged by the Joint Committee. Even so, considering the high prevalence of sensorineural hearing loss in the NICU population and the relatively small number of NICU nurseries compared to well-baby nurseries, a dual screening protocol that includes both OAE and AABR is worthy of consideration.

Training and continuing education for screeners are critical components of any screening program, and for many are ongoing challenges. Still, we were surprised that more than 40% of the hospital representatives reported a need for more systematic training and continuing education related to hearing screening. As noted earlier in reference to NICU screening, training materials are available from NCHAM and other organizations. NC-EDHI is working with hospitals interested in obtaining additional resources.

Considering the many details associated with hearing screening of newborns (Winston & Roush, 2016) we were not surprised to see that nearly all well-baby nurseries reported specific challenges that included equipment maintenance, tracking and follow-up after hospital discharge, and excessive noise. As with NICU screening, the current study did not permit exploration of details associated with these challenges, but the information has been used by NC-EHDI for inquiry and follow-up.

Approximately 1 in every 6000 babies is born with visible evidence of external ear anomalies, ranging from mild deformities of the pinna to microtia and aural atresia (Brent, 1999). JCIH 2019 recommends that infants with congenital aural atresia in one or both ears, or with visible pinna/ear canal deformities such as stenosis or severe malformation, not be screened in either ear but instead referred for diagnostic audiologic evaluation immediately upon hospital discharge. JCIH 2019 further states that diagnostic audiologic evaluation for these infants may be accomplished while the infant is in the NICU or other inpatient hospital unit. We are confident that hospitals included in this study report these conditions in the baby's birth history and discharge summary but found that fewer than one-third of the nurseries reported having a formal protocol as recommended by JCIH 2019. In addition to the recommendations of JCIH, organizations like *Ear Community* (earcommunity.org) based in Denver, Colorado, provide information and advocacy related to aural atresia and microtia.

Congenital CMV (cCMV) is the leading cause of non-genetic permanent hearing loss in children (Doutre et al, 2016; Rawlinson et al, 2018). As a result, some states are moving toward cCMV screening, especially for newborns who fail their hearing screen. Because cCMV can result in late-onset sensorineural hearing loss (Cannon et al., 2014), JCIH recommends that infants who test positive on a neonatal screen for CMV receive periodic monitoring by an audiologist, with appropriate hearing technology and early intervention if indicated. In this study, only 12 well-baby nurseries (14%) in North Carolina reported screening for CMV during the study period although seven indicated they were considering implementation of CMV screening in the future. We are unable to report details associated with CMV screening in this study; however, a follow-up investigation is currently underway as part of another NC-LEND/NC-EHDI collaboration. Also, NC-EHDI convened a CMV workgroup in 2019 that includes parent advocates, pediatric infectious disease and primary care physicians, audiologists, research and public health stakeholders with a mission to determine collaborative approaches to support the prevention and reduction of CMV infections in women and newborns; to ensure access to care for affected children, and to perform outreach and education on congenital CMV for patients,

providers, and the general public. The ongoing outreach and educational efforts of this workgroup have contributed to more hospitals implementing or considering the implementation of CMV screening.

### The Role of Audiology in Newborn Hearing Screening and Follow-up

Among the most significant and potentially consequential recommendations included in JCIH 2019 is greater audiology oversight of hearing screening programs in both the NICU and well-baby nurseries. As summarized in Table 2, audiology oversight is recommended for all state and territory hearing screening programs at both the systems level and at the individual program level. Our findings revealed that only two-thirds (66%) of the NICUs in North Carolina had direct oversight by an audiologist, and fewer than one-third (30%) of the well-baby nurseries reported oversight of the screening program by an audiologist. Anecdotally, we have observed that many of the larger hospitals or healthcare systems that already employ audiologists are more likely to have direct involvement with the screening programs. In North Carolina, few of the well-baby nurseries are in hospitals that employ audiologists, although some may have contractual arrangements with consulting audiologists. The implementation of audiology oversight, if not already provided, has many potential benefits but will require advocacy and additional financial resources. States whose EHDI programs employ audiologists may have the potential to further develop their consulting roles with hospitals, and in some states it may be possible to expand the role of educational audiologists in providing outpatient rescreening and assessments in regions with limited access to comprehensive services (Sapp et al., 2021). As more hospitals become consolidated within health systems there may be cost-efficient opportunities to expand audiology oversight of hearing screening in both the NICU and well-baby nurseries.

### COVID-19

Early hearing detection and intervention, like many healthcare practices, has been significantly affected by the COVID-19 pandemic (Yoshinaga-Itano, 2020). In response to concerns raised by clinicians and public health officials, NCHAM has compiled several COVID-19 resources and documents; among them, a statement from the American Academy of Pediatrics (2020) noting that continuation of newborn hearing screening amid COVID-19 "is essential to ensure healthy and appropriate development." According to the CDC (2020), vertical transmission of COVID-19 is rare between mother and baby, but all providers who encounter the newborn were advised to take infection control measures. AAP furthermore recommended that "healthcare workers should use gowns, gloves, standard procedural masks, and eye protection (face shields or goggles) when providing care for well babies. When this care is provided in the same room as a mother with COVID-19, healthcare workers may opt to use N95 respirators in place of standard procedural masks, if available" (NCHAM, 2021). The pandemic emerged and intensified during the NICU study period and as noted earlier, a decision was made to include questions related to the impact of the pandemic in the survey of well-baby nurseries, and to add a follow-up NICU survey in January 2021. For the NICUs, we were pleased to find that COVID-19 did not appreciably affect hearing screening in North Carolina other than a few hospitals noting a change in screening location for babies requiring a second in-hospital screen, or a delay in screening if the baby had been exposed to COVID-19 or was awaiting test results. One hospital reported that COVID-positive mothers and babies were required to wait 30 to 45 days for hearing screening. In the well-baby nursery, most hospitals continued to screen babies, both inpatient and outpatient; however, issues associated with COVID-positive mothers were frequently cited as reasons why hospitals had to modify or halt their screening programs. For hospitals electing to screen babies with COVID-positive mothers, special precautions were taken during screening, including use of PPE (personal protective equipment) and other hygienic procedures. Typically, these precautions also involved thorough cleaning of equipment. Most hospitals screened the baby in the mother's room, although a few conducted screenings in an isolation area. Some hospitals reported waiting to perform the screen until the last day of the infant's hospital stay or waiting until the end of the day to screen the baby. It is important to emphasize that the impact of the pandemic may vary significantly across the country based on multiple factors. Blaseg et al. (2021) in a retrospective study of how COVID-19 has impacted newborn hearing screening in six western states, reported significant disruptions including decreased rates of screening by one month of age, screening overall, and referral for early intervention services. The authors note that these disruptions may have important long-term consequences that warrant continued investigation of COVID-19 and its impact on newborn hearing screening. At the time of this writing, the Delta and Omicron variants have caused a resurgence of COVID-19 in some regions. Until the pandemic ends, EHDI programs and providers will need to closely monitor and mitigate any impact of COVID-19.

### Strengths and Limitations

An important strength of this study was the full participation of birthing hospitals in North Carolina, which resulted in a 100% response rate from all 24 NICUs and all 86 well-baby nurseries. This outcome is a testament to the dedication of our hospital nurseries and to the perseverance of our research team, and it enabled our EHDI program to assess the current status of infant hearing screening and make specific recommendations statewide. Several potential limitations must also be acknowledged. Our findings are based on responses from a single representative from each hospital with no means of checking the accuracy of the information provided. To help mitigate this concern, hospital representatives were chosen based on the recommendations of NC-EHDI regional consultants, all of whom were familiar with screening personnel in their regions. There was also variability in the respondents' professional disciplines and backgrounds that may have affected their familiarity with some of the technical aspects

of the newborn hearing screening program. To address this concern, the study team and the NC-EHDI regional consultants were available to support hospital personnel if they had questions or needed assistance when completing the survey. Finally, it is important to acknowledge that the NC-EHDI program, as with most state healthcare agencies, can make recommendations to hospitals regarding clinical practice, but it does not have the authority to prescribe policies and procedures.

**Summary and Future Directions**

The hospitals in North Carolina are fully committed to their hearing screening programs, as demonstrated by numerous strengths in both the NICU and well-baby nurseries. Even so, for many hospitals we identified opportunities for program development or improvement based on JCIH 2019 recommendations. For the NICU nurseries, our recommendations emphasized the importance of direct referral to a pediatric audiologist for babies who do not pass the in-hospital hearing screening. Also highlighted was the importance of rescreening infants readmitted to the NICU or pediatric intensive care unit with a condition or treatment associated with a risk factor for hearing loss. For the well-baby nurseries, our recommendations underscored the importance of direct referral to an audiologist following a failed outpatient rescreening. Also emphasized was the importance of direct referral to an audiologist and otolaryngologist for babies with visible signs of external ear anomalies. For both the NICU and well-baby nurseries, JCIH 2019 recommends systematic and ongoing continuing education for screening personnel and oversight of the screening program by an audiologist with experience in evaluating newborns and young children. Following the completion of these studies, NC-EHDI has worked in partnership with hospitals to provide additional resources and technical assistance. As a result of this collaborative effort, many programmatic improvements have occurred statewide.

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## EHDInfo

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# "It Gives Me Confidence": Caregiver Coaching from the Perspective of Families of Children who are Deaf or Hard of Hearing

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## Abstract

Caregiver coaching is used in early intervention services with families of children who are deaf or hard of hearing to increase caregivers' skills and confidence in supporting their child's language development, but few studies have examined coaching from the perspective of the caregivers. The purpose of this study was to increase understanding of caregivers' experiences of coaching in the context of listening and spoken language intervention services. Using semi-structured, qualitative interviews, this study examined 13 caregivers' perspectives at three intervention sites in the United States and Canada. Results indicate that caregivers perceive that practitioner characteristics, expectations, and the evolution of the coaching relationship over time contribute to a positive caregiver coaching relationship. This study contributes to the understanding of the caregiver coaching experience and has implications for new and experienced practitioners working to improve their practice by establishing and strengthening collaborative caregiver coaching relationships with the families they serve.

**Keywords:** deaf or hard of hearing, early intervention, listening and spoken language, caregiver coaching

**Acronyms:** DHH = deaf or hard of hearing; EI = early intervention; LSL = listening and spoken language

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Caregiver coaching is a process designed to empower caregivers by building their capacity, competence, and confidence to support their child's development within naturally occurring daily routines (Dunst & Trivette, 2009; Dunst et al., 2007; Rush & Shelden, 2011; Sukkar et al., 2016; Woods et al., 2011). Caregiver coaching is widely considered best practice in early intervention (EI) for families of children with disabilities, including children who are deaf or hard of hearing (DHH; Division for Early Childhood, 2014; Moeller et al., 2013). For families pursuing listening and spoken language (LSL) for their children who are DHH, timely diagnosis, appropriate audiologic management (including hearing technology), and early enrollment in specialized EI services provide much-needed support for families (Ching & Leigh, 2020; Durieux-Smith et al., 2008; Holzinger et al., 2011; Moeller et al., 2013). Through caregiver coaching, families learn LSL strategies to support their child's learning and development.

EI in general, and LSL practice specifically, has an imperative to include caregivers as active participants, and caregiver coaching is one of the primary approaches for achieving this goal (Rush & Shelden, 2005, 2011; Shelden & Rush, 2005). This is particularly relevant for families of children who are DHH, because research indicates that caregiver involvement in EI is linked to positive outcomes for children (Allegretti, 2002; DesJardin et al., 2006; Spencer, 2004; Zaidman-Zait & Young, 2008), particularly in communication development (Calderon, 2000; Moeller, 2000; Yoshinaga-Itano, 2003). Recent research indicates that early amplification and participation in EI resulted

in a higher likelihood of reaching language scores commensurate with typical hearing peers (Ching & Leigh, 2020; Davidson et al., 2021), and these benefits increased with greater intensity of EI services and greater levels of hearing loss (Ching et al., 2017; Geers et al., 2019).

Recent research has begun to examine the effectiveness of coaching for caregiver learning (Ciupe & Salisbury, 2020; Sone et al., 2021); however, incongruence persists in definition, terminology, and framework. Improving specificity is critical to inform robust evaluations of the processes, intermediate outcomes (e.g., caregiver learning), and eventual outcomes (e.g., communication outcomes for children) of caregiver coaching. In a research synthesis on coaching in EI, Kemp and Turnbull (2014) found no common definition or description of coaching, and practices ranged from *relationship-driven* on one end of the spectrum to *intervener-directed* on the other. Relationship-driven practices involved practitioners collaborating with caregivers on planning and decision-making, and intervener-directed practices involved a more prescribed approach for caregivers to follow. A more recent systematic review in Australia indicated a persistent lack of an operationalized definition of caregiver coaching, inconsistencies in reporting of how practitioners learn and implement coaching practices, and a lack of outcome measures to determine its effectiveness with families of children at risk for disabilities (Ward et al., 2020).

Listening and spoken language practitioners abide by principles that emphasize the importance of caregiver coaching when working with families of children who are DHH (AG Bell Academy for Listening and Spoken Language [AG Bell Academy], 2017; Kendrick & Smith, 2017; Moeller et al., 2013); however, these practices are not well-defined. Practitioners are expected to *guide and coach parents* to become the primary facilitators of their child's communication development and integrate listening and language into all areas of the child's life (AG Bell Academy, 2017; Estabrooks et al., 2016). Widely recognized best practice principles for family-centered EI provide guidance for coaching caregivers of children who are DHH, including the development of collaborative partnerships characterized by open communication, shared tasks, and mutual trust. Coaching helps teach caregivers new skills through the use of adult learning strategies and builds on existing knowledge and skills (Moeller et al., 2013). Additional guidance indicates that practitioners are expected to develop proficiency in parent guidance, including family coaching and adult learning (AG Bell Academy, 2017, p. 20). Although these constructs are essential components of LSL practice, professional guidance documents lack clarity regarding the elements of coaching and how it should be implemented with families of children who are DHH.

Few empirical studies have examined caregiver coaching in this population. Recent reviews of the literature highlight this dearth of evidence. Shekari et al. (2017) identified 22 studies for inclusion in a systematic review of the role of parents and the effectiveness of EI for children who are DHH, but none were directly related to caregiver coaching. The review found that family participation in EI is an important factor in a child's outcomes; however, how caregivers learn skills in the context of intervention was not examined. In a systematic review of coaching practices in EI for children at risk of developmental delay, only one of the 18 included papers was directly related to the impact of parent coaching versus therapist-delivered intervention (Ward et al., 2020). The authors concluded that although caregiver coaching is widely accepted, there is a need for studies measuring the impact of caregiver coaching on parent capacity and self-efficacy. Our scoping review on caregiver coaching in LSL EI services included 22 articles, six of which were primary research studies but only one was peer-reviewed (Noll et al., 2021). Our results indicated that caregiver coaching should be individualized, context-driven, collaborative, and strengths-based (Noll et al., 2021). We consolidated eight models of coaching and a variety of coaching practices found in the literature to propose a model of caregiver coaching in LSL practice.

There is limited evidence that parent training is effective in teaching caregivers to implement language strategies with their children who are DHH (Nicastri et al., 2020; Roberts, 2019). In a small randomized-controlled trial, Roberts (2019) found that caregivers ($n$ = 9) increased their use of communication support strategies following training, and this resulted in significant gains in prelinguistic speech skills in their children, compared to a control group ($n$ = 10) who did not receive training. In a small prospective clinical study, Nicastri et al. (2020) studied the long-term effects of a parent training program focused on increasing language facilitation skills in 14 parents of children with cochlear implants. Parental interaction and child language results were measured immediately following the parent training, and again three years later. Parents improved the quality of their interactions and the children in the treatment group showed a significant improvement in linguistic skills compared to the control group. This study indicates that parent training can be an effective tool for improving parents' use of communication strategies; however, parents learned new skills through a predetermined group curriculum, rather than through individualized caregiver coaching.

Although the EI literature supports caregiver coaching and LSL guidelines suggest its use as a standard of practice, current literature lacks a clear description of caregiver coaching with families of children who are DHH, and little is known about caregivers' experiences with coaching. As such, the purpose of this qualitative study was to broadly examine and increase understanding of caregivers' experiences with coaching in EI services for their children who are DHH and suggest steps practitioners can take to establish a positive caregiver coaching relationship.

## Method

This qualitative research study involved semi-structured interviews with caregivers receiving LSL language EI services at one of three sites and was informed by the principles of interpretive description (Teodoro et al., 2018;

Thorne, 2016; Thorne et al., 1997, 2004). This methodology is well-suited to our purposes because the foundation of this applied qualitative approach is the investigation of a clinically relevant phenomenon to identify themes and patterns from subjective perceptions and generate an interpretive description to inform clinical understanding (Burdine et al., 2020; Thorne et al., 2004). This study received research ethics approval from the University of Ottawa and the CHEO Research Institute in Ottawa, Ontario. Consent was obtained prior to each interview.

### Sampling

Participants were purposely selected from one early intervention program in Canada and two programs in the United States, representing diversity in geographical location, service delivery models, and exemplary LSL services. Site 1 offers services on-site, Site 2 primarily offers home-based services, and Site 3 offers a combination of site-based and home-based intervention services. Eligible participants included caregivers who: (a) participated in LSL services for a child who is DHH, ages birth to 3 years within the previous six months, and (b) were able to communicate in English. Caregivers were invited to participate by their practitioners, and each practitioner was asked to recruit 1 to 2 caregivers, at their discretion. This sampling strategy allowed the practitioners to choose caregivers who could meaningfully inform an understanding of the research problem and provide valuable information to help answer the research questions (Creswell & Poth, 2018).

The aim was to identify recurrent patterns while also capturing diversities in the experiences among caregivers participating in LSL services in different contexts (Braun & Clarke, 2021; Burdine et al., 2020; Thorne et al., 2016). Aligned with the principles of interpretive description, we identified commonalities while acknowledging that the coaching relationship is unique to each caregiver/practitioner dyad. We obtained a deeper understanding of caregivers' experiences, while still recognizing that variations will always exist in applied practice (Abdul-Razzak et al., 2014; Burdine et al., 2020; Thorne, 2016). The resulting commonalities provide new and clinically applicable understanding of the experience of caregiver coaching.

### Data Collection and Analysis

Individual, semi-structured interviews were conducted at a convenient location for the caregivers, including on-site, in the family's home, and, for one family, via Zoom video conferencing software. Caregivers were asked to describe their overall experience participating in LSL EI services, with a particular focus on their relationship with their practitioner and how they learn within the context of an intervention session. The interviewer explained coaching to the caregivers as "a provider teaching the parent, rather than teaching the child" (see Appendix for interview guide). Interviews were audio recorded, transcribed verbatim, and verified before being uploaded into NVivo (12.6.0), a qualitative data analysis software used to organize and facilitate analysis. To preserve confidentiality in the final report, we removed participant and site names and assigned pseudonyms for reporting.

Interview data were analyzed using reflexive thematic analysis, which uses an inductive, iterative six-phase process: (a) familiarization, (b) generating codes, (c) constructing themes, (d) reviewing themes, (e) defining and naming themes, and (f) producing the report (Braun & Clarke, 2006; Braun et al., 2019; Terry et al., 2017). This method of analysis acknowledges and values the researchers' experience and perspective and is well-suited to applied qualitative research that answers clinically relevant questions (Campbell et al., 2021).

To ensure rigor and trustworthiness and account for potential bias (Holmes, 2020), the primary researcher critically reflected on her positionality, participated in reflexive memoing throughout data collection and analysis, maintained detailed field notes and an audit trail, and met with other members of the research team throughout to challenge assumptions, debrief, reflect, discuss, and refine codes and themes.

The primary researcher who conducted and analyzed the interviews is the parent of a child who is DHH and an experienced LSL EI practitioner with experience in collaborative caregiver coaching. This dual perspective affords the researcher a unique perspective on issues of clinical significance in LSL practice and informed the design of this research project.

## Results

Thirteen interviews were completed with one father, nine mothers, and three sets of both parents (see Table 1 for demographic information). All families but one had a child currently receiving LSL EI services; one child transitioned out of EI four months prior to the interview. Four of the participants reported working with more than one practitioner while in EI, and two participants had two children who have received LSL EI services, both of whom worked with a single practitioner. The distribution across sites was as follows: Site 1, $n = 3$; Site 2, $n = 6$; Site 3, $n = 4$.

Overwhelmingly, caregivers reported positive experiences with coaching throughout the course of their early intervention experience. Several discussed feeling hesitant, uncertain, or guarded in the beginning, which changed over time as they established a trusting relationship with their practitioner.

Cumulatively, the caregivers described coaching as a positive experience, and we identified three overarching themes that contribute to this positive experience, from the caregivers' perspective: (a) it takes a special kind of person, (b) building on expectations, and (c) figuring it out along the way. See Table 2 for a description of themes, sub-themes, and codes, along with supporting quotes from the interview data.

**Table 1**
*Demographics*

| Variable | Number | Percentage |
|---|---|---|
| Interview participant(s) | | |
| Mother | 9 | 69.2% |
| Father | 1 | 7.7% |
| Both parents | 3 | 23.1% |
| Age of child at time of interview | | |
| 12–18 months | 2 | 15.4% |
| 18–24 months | 2 | 15.4% |
| 24–30 months | 5 | 38.5% |
| 30–36 months | 3 | 23.1% |
| > 36 months | 1 | 7.7% |
| Age at diagnosis | | |
| < 6 months | 13 | 100% |
| Degree of hearing loss | | |
| Mild | 2 | 15.4% |
| Moderate | 1 | 7.7% |
| Severe | 2 | 15.4% |
| Moderately-severe | 3 | 23.1% |
| Profound | 5 | 38.5% |
| Age at service initiation | | |
| < 6 months | 11 | 84.6% |
| 7–12 months | 1 | 7.7% |
| 13–24 months | 1 | 7.7% |
| Device type | | |
| Hearing aid(s) | 5 | 38.5% |
| Cochlear implant(s) | 6 | 46.2% |
| Both | 2 | 15.4% |
| Frequency of services | | |
| 1x/week | 6 | 46.2% |
| 2x/month | 5 | 38.5% |
| 1x/week (onsite), 2x/month (home) | 2 | 15.4% |

### Caregiver Coaching is a Positive Experience

*"So, coaching is very positive. Strong reinforcement with the things we're doing right, and then guidance on the things we're doing wrong." (Henry)*

### It Takes a Special Kind of Person

*"You really have to be interested in helping these kids and the parents." (Ashley)*

All of the caregivers talked about their relationship with their practitioner as an impactful part of the coaching relationship, using a variety of adjectives to describe positive attributes (see Figure 1). Some caregivers worked with multiple practitioners over the course of their time in early intervention and used positive language to describe all of them. All the caregivers talked about the importance of establishing a meaningful relationship as a foundational aspect of their overall positive experience.

**Figure 1**
*Caregivers' Description of Practitioners*


*Note.* Word size represents frequency (created on wordart.com).

In addition to highlighting positive personality characteristics, caregivers also described a warm relationship with their practitioner, using phrases such as "familial," "like a friend," and "a professional friendship." After describing her practitioner as supportive, Chelsea described their relationship in this way: "I would say that our relationship is like a family member but also kind of like a teacher—that you really want to please and that you don't want to disappoint."

When asked what they thought was most important for establishing the caregiver-practitioner relationship, some caregivers referred to practical factors, such as practitioners' preparedness, expertise, and time; however, most also cited positive personality traits and the primacy of establishing trust as the building blocks for the coaching relationship.

### Building on Expectations

"*Expectations have to be clear." (Henry)*

When describing their experiences, caregivers talked about practitioner expectations as a fundamental component in a positive coaching relationship. Expectations were either explicitly or implicitly established by the practitioner at the beginning of the coaching relationship, and this set the tone for how caregivers viewed their role and the role of the practitioner. These expectations established the foundation for how caregivers experienced the coaching relationship over time and included three elements: (a) the caregivers' expectations of the practitioner, (b) the caregivers' expectations of themselves, and (c) how caregivers expected to see progress as a result of coaching.

Eight of the caregivers described an explicit manner in which their practitioners established expectations for their role in the coaching relationship, while five described a more implicit approach. The explicit approach included clearly outlining the role of the caregiver from the very

**Table 2**
*Description of Themes and Supporting Evidence*

| Theme | Sub-theme | Description | Codes and Quotes as Evidence |
|---|---|---|---|
| It Takes a Special Kind of Person | | Practitioner characteristics reported as important for fostering the coaching relationship | "I mean, obviously you have to have a certain demeanor to be that type of profession." (Ashley) |
| Building on Expectations | Expectations of Practitioner | How caregivers view their practitioners' role in the coaching relationship | *Practitioner-as-expert:* "But she is the, at the end of the day, she's the professional in this. She feels that that's, that's where we need to be going, okay, that's where we're gonna go." (Matthew)<br>*Practitioner-as-partner:* "I don't know, she feels like a partner. It's kind of fun. Like, compared to some of the other therapists, like physical therapy and occupational therapy, it's a little more them directing everything and them doing everything and just kind of talking me through stuff. Where I feel like with (Practitioner), it's kind of like, I don't know, we're doing it together." (Julie) |
| | Expectations of Self | How caregivers view their role in the coaching relationship | *Being an observer*: "So, you know, that's what I take away from my role: observing what they're doing." (Ashley)<br>*"I'm the student":* "But yeah, I do feel like a student. I'm learning new things and I feel like every session I'm learning something different." (Jane)<br>*"It's all on me"*: "I'm the everything." (Henry) |
| | Expectations of Success | How caregivers view progress as a result of coaching | *Caregiver learning as a measure of success:* "I wanted her to see that we were learning, and we were trying and that we were applying the things that we were learning." (Chelsea)<br>*Child performance as a measure of success:* "And then she turned 18 months and her language just exploded. I felt so confident after that. That everything they said, 'Oh, work on this,' I would work on it for like a day and (Child) would have it down. And I would, I would be like, 'Oh my gosh, this is amazing!'" (Sarah) |
| Figuring it Out Along the Way | Establishing a Foundation | The foundation of the coaching relationship is built during a vulnerable time in caregivers' lives and involves a high need for information and establishing trust. | *Building trust:* "I would also say that you just have to immediately establish this trust, which is not something you can teach, it just kind of happens." (Chelsea)<br>*Establishing expectations*: "One of the very first things she said to me was, 'This is going to be as good as you, as you want it to be. And it's going to be as much as you're engaged in it.'" (Henry)<br>*Information sharing*: "When he was younger, we – it was a lot about how to deal with his equipment…it was more informative for us." (Ashley)<br>*Overwhelming at times:* "I remember at the beginning, it was so overwhelming for all of us…and she…would take the time to explain what is now, what will be, and give us all the information in between." (Isabelle) |

**Table 2 (cont.)**
*Description of Themes and Supporting Evidence*

| Theme | Sub-theme | Description | Codes and Quotes as Evidence |
|---|---|---|---|
| | Ongoing Trust and Unguardedness | Trust and unguardedness are needed for the entirety of the coaching relationship. | *Mutual respect:* "When there were things that we questioned, I felt like our relationship made it so that we could bring things up, or I never felt like I could ask a dumb question or anything like that, and I think it's just because we've had that mutual respect." (Chelsea)<br>*Openness:* "And it's really, you just got to let your walls down and trust someone else." (Cynthia)<br>*Rapport:* "…if you don't make that connection, it's not going to work." (Michael)<br>*Transparent communication:*<br>I: "So, what would you say is, is the most important thing for a good provider and parent relationship?"<br>Mary: "I would say transparency and being able to listen to one another…" |
| | Shared Development of Knowledge and Skills Leads to Empowerment | Practitioners equip caregivers over time by providing information and developing skills; as a result, caregivers take on more responsibility and need less support. | *Explaining the "why":* "She was, from beginning to end, step by step, we knew why we were doing it from the beginning and what result we were going to have at the end." (Michael)<br>*"I've learned a lot":* "I learn what I need to know. I mean, I feel like it's an accomplishment, like 'oh, oh!'" (Rebecca)<br>*"It makes me feel empowered and confident"*: "So I can try their new suggestions and, yeah, it makes me feel, like, empowered and more confident as a parent." (Mary) |

beginning of the coaching relationship and reiterating the importance of the caregiver's role over time. A more implicit approach involved demonstrating for the caregiver without explicitly outlining the importance of his or her involvement in planning and during sessions.

### *Expectations of Practitioner*

Although all the caregivers acknowledged and respected the practitioner's expertise in LSL, some deferred to the practitioner as the primary expert and others saw the practitioner as more of a partner whose role it was to collaborate with them as the experts on their child. Some caregivers vacillated between the two, while others generally fit into one category or the other.

### *Practitioner-as-Expert*

Caregivers who viewed their practitioner as the primary expert tended to describe themselves as less important partners in the coaching relationship. They relied on the practitioner to problem-solve, provide resources, and plan goals and activities for intervention sessions, and were less likely to describe the relationship as collaborative than caregivers who considered their practitioners as a partner. For example, when asked about her role in deciding what to work on with her child, Rebecca shared that she would feel comfortable bringing up concerns with her practitioner, but "I probably wouldn't make a suggestion because I feel like I'm not the expert."

### *Practitioner-as-Partner*

Alternatively, caregivers who viewed their practitioner as more of a partner considered their role in the coaching relationship as pivotal for their child's progress. These caregivers described setting goals in partnership with their practitioner because they know their child best and understand what will work in the context of their daily lives. Chelsea described it as "shoulder-to-shoulder learning together," and stated, "I like working alongside someone." Some caregivers reported choosing activities and goals for the sessions themselves, others worked together with their practitioner to decide what to target during intervention sessions, and some reported a combination of both approaches.

### *Expectations of Self*

Caregivers described their expectations of themselves in the context of their role in the coaching relationship. These expectations ranged from taking full responsibility during sessions and in-between, to being a learner who takes an active role in intervention sessions following practitioner demonstrations, to being an observer and primarily watching the practitioner working with their child. How caregivers viewed their role in the coaching relationship was tied to how they talked about their practitioners' role—those who saw themselves as observers were more likely to defer to their practitioner as *the expert*, and those who talked about their own role as primary in the relationship

viewed their practitioners as *a partner*. The following codes represent this continuum.

**It's All on Me.** Five of the caregivers described themselves as highly involved in the coaching relationship, because the outcome depended on them learning and implementing strategies with their children in their everyday lives. Henry described the significance of his role in the coaching relationship this way: "I'm the everything. I mean, (Practitioner) is really just giving us the framework." He went on to say that although he sees his practitioner for 45 minutes to an hour twice a month, it's what he does in-between that makes the difference, and that he and his wife want to make sure they are doing everything possible to ensure their child's progress.

**I'm the Student.** Six of the caregivers saw their role as students, learning from the practitioners' expertise, but also willing to actively participate and practice skills after a model during the coaching exchange. Mary described a typical session in which she observes as her practitioner demonstrates a strategy with her child, then she takes a turn and her practitioner offers feedback. She may try again and then they will discuss how she did and what she might do differently the next time. "She'll pull out her activity, she'll tell me what she expects (Child) to say from it. She'll, she'll say it and then she'll pause and wait for him to do it, and then she'll ask me to try it out."

**I'm an Observer.** Two caregivers described their participation as primarily watching and learning and not necessarily taking a turn during the session. Lauren described her role as "an observer and taking it in." She described intervention sessions in which she watches and learns while her practitioner interacts with and teaches her child. She described hesitation to actively participate during sessions because, as she states, "I'm not good at demoing with somebody watching me….but if I can gather all the information and watch you do it, then I can do it later."

### *Expectations of Success*

Caregivers revealed the ways in which they measured success, separate from traditional indicators of progress, such as assessments. They talked about things that made them feel like caregiver coaching was successful, either in terms of their child's progress or their own learning. Some caregivers indicated that both these factors contributed to what they considered a successful coaching experience.

**Child Performance as a Measure of Success.** Caregivers indicated that their child's speech and language growth played a role in determining whether coaching was working. Julie talked about her child's progress as a motivating factor for continuing to implement the strategies she was learning:

> I think at first, too, it was hard because he really was not turning to anything, so it's, it's hard to be motivated when you're not seeing direct results of it. Once we started really seeing the changes happening, then it was like, ok this is, this is real.

**Caregiver Performance as a Measure of Success.** Six caregivers considered their own growth in understanding and implementing LSL strategies with their child as an indicator of success. Henry referred to his own learning as a measure of progress: "I'm reading to her, I'm always making sure I'm beside, like, and it, there's times where I'll realize, I'm like, holy smokes, she trained me!"

### Figuring It Out Along the Way

> *"It's a process, it's a journey, you figure it out along the way - what works and what doesn't." (Sarah)*

The coaching relationship changes over time in response to the changing needs of the caregivers. The caregivers described their emotional state and needs in the beginning as very different than what they needed as services progressed, and suggested that by adapting to their needs, practitioners contributed to a positive coaching experience overall.

### *Establishing a Foundation*

The foundation of the coaching relationship is built during a vulnerable period in caregivers' lives. Caregivers reported feeling overwhelmed and in need of information and emotional support. This vulnerable time period is when trust and expectations must be established. Cynthia described the beginning of the coaching relationship in this way: "They come into your life in such a vulnerable place. And it's really, you just got to let your walls down and trust someone else." According to caregivers, the time and effort that practitioners spend in the beginning laying the foundation helps to establish a positive and meaningful coaching relationship. Establishing a foundation includes building trust, establishing expectations, and sharing information, and caregivers often described this as overwhelming at times.

### *Ongoing Trust and Unguardedness*

The ongoing coaching relationship also requires trust and unguardedness, and caregivers shared that mutual respect, rapport, transparent communication, and openness contribute to a positive coaching experience. All of the caregivers described a level of comfort with their practitioners that allowed them to freely ask questions, share concerns, and communicate openly without fear of judgement. They expressed relief to have someone supporting them and providing reliable information, and the confidence that was gained in the beginning provided the foundation upon which the ongoing relationship was built. Gina described this progression of trust: "I always felt like I had to be so defensive about him and stuff, where, after a while, she just made it really comfortable, and I didn't feel like I had to have a guard up anymore."

Cynthia highlighted the willingness to be open and vulnerable as a necessary component in a coaching relationship that may involve difficult conversations at times:

> I think there needs to be an element of accepting and giving of, like, critical information. If you can't receive information from them that is hard to hear…it's a level of vulnerability that's kind of

> required…if you can't receive or give information back and forth, without open communication, and there's a lot of walls up, it's just, it's not going to be a good relationship…

Gina and Michael talked about what they considered to be the most important component of the ongoing coaching relationship: trust.

> We trusted that we could ask her a question and she trusted that she could ask us or tell us something and it would not change anything… so even when we did not get the best news or you get the good news, she's always there to help you and guide you.

Caregivers described several ways in which practitioners established trust, including being a reliable source of information, being supportive and non-judgemental, establishing a personal connection with them and their child, and actively listening to their concerns. They also indicated that time was a factor, both in the amount of time they spent with their practitioner, and the timing of the onset of the relationship, when they needed information, support, and encouragement.

**Shared Development of Knowledge and Skills Leads to Empowerment**

Over the course of the coaching relationship, the shared development of knowledge and skills leads to a transfer of responsibility and empowerment from the practitioner to the caregiver. Sarah described how her level of confidence has changed over time: "I always leave, especially now, feeling really confident in what (Child) is doing…Knowing that, that I get it, that I can, that I can help my child." Ashley talked about having so many questions in the beginning, especially with regard to how to help her child, but then, over time, using LSL strategies has become second nature: "I've started doing things that I don't even notice that I'm doing…it's become the norm."

Although all of the caregivers described an evolution of the coaching relationship over time, the progression was not necessarily linear. Caregivers described times when they felt overwhelmed, even after the intensity of the early stages of their child's diagnosis and beginning EI. They reported feeling more empowered as they learned skills and built confidence, but there were times when they still needed extra support. Ashley explained one example of this: "I think, personally, like, with early intervention and with parents that are, like, overwhelmed—like, right now we are going into the transition stage and that's very overwhelming to me. I don't want to leave the comfort of here."

## Discussion

This research is novel in that it examines caregivers' perspectives specific to coaching in LSL EI services, increases understanding of how caregivers experience coaching, and highlights how practitioners can establish and maintain an effective coaching relationship. Caregivers of children who are DHH viewed coaching as a positive experience; however, because practitioners recruited caregivers, it is possible that these data reflect only meaningful coaching relationships. The caregivers conceptualized coaching in different ways, according to their experience, and some conflated caregiver coaching with the entirety of the EI experience. This suggests one of two things: that the LSL practitioners integrated coaching seamlessly with families in the context of their intervention, or that practitioners did not always take a collaborative approach to caregiver coaching. This study reveals three factors that contribute to a positive coaching experience, according to caregivers: practitioner characteristics, how expectations are set and maintained, and coaching that adapts to changing caregiver needs over time.

Our findings indicate that characteristics of the practitioner play an important role in a positive caregiver coaching relationship. Caregivers used a variety of descriptors to describe their practitioner as warm, caring, and trustworthy. Interestingly, Tattersall and Young (2006) also found that professional communication and manner were the most important influences on parents' experiences during the audiologic diagnostic process. The perspective of the caregiver has been underrepresented in both the general EI and LSL literature, and, as such, this insight highlights the importance of demeanor and the establishment of trust in creating a positive coaching partnership, which can, in turn, lead to growth. This finding aligns with perspectives of coachees in an early childhood setting, who reported that that they valued their relationships with their coaches and this positive partnership led to growth and change (Knoche et al., 2013). Other studies have indicated that caregivers were satisfied with their family-centered intervention services (Stewart et al., 2020) and that a collaborative and supportive relationship was important for their learning (Salisbury et al., 2018); however, our study extends the understanding of specific characteristics that may lead to a supportive relationship between caregivers and practitioners. Caregivers' experiences with coaching may in part determine the uptake of intervention and their engagement as well as their perceptions of the quality of intervention, which in turn can influence their child's developmental outcomes.

An interesting finding from this study was that expectations were a strong underlying factor in a positive coaching partnership. Caregivers' expectations of their practitioners were connected to their view of their own role in the partnership, with those who described their practitioners as partners taking a more active role in the coaching process during EI sessions. Consistent with previous literature, our study showed that clear expectations and mutually agreed upon goals are important for establishing a partnership, leading to a positive and successful coaching relationship where partners play a vital role (Rush et al., 2003; Rush & Shelden, 2011; Workgroup on Principles and Practices in Natural Environments, 2008). As active caregiver participation is understood as an important component in the coaching process (Noll et al., 2021), a lack of engagement precludes a bidirectional, collaborative exchange between caregiver and coach. This

balance of power is an important consideration. Balanced partnerships between families and practitioners are considered best practice in family-centered EI for children who are DHH, according to an international consensus statement (Moeller et al., 2013).

Our findings indicated that this partnership is established at the beginning of the coaching relationship and is reinforced through joint planning and active participation in individual sessions. Caregivers who consider themselves observers and the practitioner as expert do not enter into a reciprocal coaching exchange where the caregivers actively contribute and participate; rather, the practitioner primarily chooses goals and activities and instructs the caregivers, with or without opportunities to practice skills within the context of a session. This level of caregiver participation represents more of a practitioner-directed style of intervention and does not represent a balanced partnership, therefore highlighting a potential obstacle in establishing a collaborative coaching relationship. Ambiguities in the EI literature suggest that caregiver coaching is not always differentiated from parent training; the difference lies in the extent of the caregiver's role in decision-making and goal setting and a truly collaborative partnership between caregiver and coach (Kemp & Turnbull, 2014; Ziegler & Hadders-Algra, 2020). Most caregivers in our study described an active role and hands-on practice during sessions with their child; however, two caregivers described their role primarily as observers. Although all three intervention sites espouse caregiver coaching, this indicates that at least some of the time with some caregivers, more traditional intervention that does not incorporate caregiver coaching is used. This may be due to personality characteristics of the caregivers or may be linked to the expectations established and maintained by the practitioners throughout the EI process.

Our results highlight that practitioners need to explicitly establish expectations and partner with families in ways that will encourage active participation and allow for a reciprocal coaching relationship to develop. Caregivers were more likely to view their practitioner as the expert (vs practitioner as partner) when expectations were established implicitly rather than explicitly. Additionally, the ways in which caregivers talked about their expectations of progress provides insight into their perception of success. Caregivers who view progress as their own mastery of LSL strategies, rather than solely based on their child's progress, understand how critical their role is in the coaching process, and take responsibility for learning and implementing LSL strategies with their child beyond the context of the intervention session.

Results of our study indicate that caregivers' needs change over time, and practitioners who adjust their coaching in response contribute to a positive coaching relationship. The goal of the coaching relationship is to build expertise to enable the caregivers to become skilled facilitators of speech and language with their children. The practitioners scaffold their coaching by gradually increasing the caregivers' responsibility and ownership as they gain skills. This is accomplished by ensuring that the caregivers understand the reasoning behind the strategies they are learning, co-creating goals, continuing to build on what they are learning over time, and giving them opportunities to feel successful and confident in their newfound expertise. Previous studies have indicated that the provision of information is important for meeting the needs of caregivers of children who are DHH (Decker & Vallotton, 2016; Fitzpatrick et al., 2008; Roberts et al., 2015; Stewart et al., 2020), and our study suggests that this need is greatest in the beginning of the coaching relationship. Previous research suggests that caregivers initially experience shock, but it gets easier over time with information and support provided by EI professionals (Haddad et al., 2019). Additionally, caregivers have reported that they find the initial decisions related to intervention such as communication modality and device use stressful, and the support of LSL practitioners is invaluable (Gilliver et al., 2013; Roberts et al., 2015). In our study, caregivers reported this as a time of trust-building that formed the foundation of the coaching relationship, so, although it was a stressful time, ultimately it solidified their confidence in their practitioner.

Not only does the type of information caregivers need change, the amount of support changes as caregivers gain knowledge and confidence in implementing LSL strategies. One goal of family-centered EI is for caregivers to gain proficiency in implementing LSL strategies with their children. According to the caregivers in this study, practitioners who scaffolded their support built the caregivers' confidence and made them feel empowered. Empowerment resulted in caregivers taking a more active role in the coaching process, and in some cases independently setting goals and implementing strategies with feedback from the practitioner. Our finding supports recent research that indicates that caregivers gain skills over time as a result of focused LSL EI (Josvassen et al., 2019). Our finding also supports research in the general EI literature that found that practitioners' use of caregiver coaching strategies decreased over time, resulting in caregivers taking the lead in sessions with less support (Ciupe & Salisbury, 2020). An interesting direction for future research would be to examine the effectiveness of coaching practices—whether coaching (process) indeed leads to measurable skill development (outcome) for families participating in EI services.

This study adds to recent research aiming to better understand the experiences of caregivers receiving family-centered EI, including coaching. Studies have indicated that caregivers report being told that taking an active role in the intervention process is essential for their child's development (Decker & Vallotton, 2016), which is aligned with recommended EI practices and essential for caregiver coaching (Division for Early Childhood, 2014; Moeller et al., 2013). Families of children who are DHH find coaching beneficial for learning LSL strategies (Josvassen et al., 2019), and report satisfaction overall with the family-centered services they receive (Fitzpatrick et al., 2008; Josvassen et al., 2019; Roberts et al., 2015; Stewart et al.,

2020). Our results align with recent survey research that indicated that caregivers considered coaching a positive experience (Josvassen et al., 2019; Salisbury et al., 2018).

Our study extends this understanding by examining the experiences of caregivers receiving LSL EI services and suggests specific factors that practitioners can incorporate to contribute to a positive coaching relationship in their work with families. First, there is benefit to setting clear expectations and parameters for caregiver participation as partners in the coaching relationship from the very beginning. Also, recognizing that caregivers' needs change over time and that they have a high need for information and support in the beginning, practitioners can build trust by being a credible source of information and offering support with kindness and empathy. Another consideration is that families who start the process later, resulting in less time in EI, will likely still need the trust-building that sets the stage for the remainder of the coaching relationship. Once trust is established and the foundation is set, practitioners can adapt to the changing needs of the caregivers over the course of their time together. Finally, practitioners can scaffold their coaching strategies, including modeling and demonstrating in the early stages of learning, with the goal of transferring responsibility to the caregiver as skills and confidence increase. Caregiver coaching is a capacity-building practice, intended to build knowledge and skills to a level of mastery that empowers caregivers in their interactions with their children (Dunst et al., 2014; Dunst & Trivette, 2009; Rush & Shelden, 2011, 2019). This goal should be explicitly shared with the caregivers from the beginning to establish the expectation for active participation in the coaching relationship and to empower them as capable agents of change in their child's LSL development.

The variability with which the caregivers talked about coaching highlights differences in coaching practices among practitioners. In particular, caregivers described their role in intervention on a continuum from observation to active participation in all aspects of the coaching exchange. These differences in expectations and practice may reflect discrepancies in practitioners' training and preparation for coaching, as some may have been trained to teach children who are DHH rather than to coach their caregivers. This is an important consideration, because although best practices indicate that children who are DHH should receive services from highly trained practitioners (Moeller et al., 2013), this does not account for the specialized skills needed to engage with and teach adult learners. Additionally, because there is a lack of a consistently used model of caregiver coaching in LSL services (Noll et al., 2021), it cannot be assumed that all practitioners are adequately trained to implement evidence-based coaching practices with families. This indicates a need for the development of standards of practice for coaching caregivers and pre-service and in-service training to increase the likelihood that practitioners will consistently implement these coaching practices.

This study was not without limitations. Caregivers were invited to participate by their practitioners, who may have chosen *ideal* families that do not necessarily represent the diversity of viewpoints and experiences of all families on their caseload. This is especially important to consider since all participants considered caregiver coaching a positive experience. It is also important to note that differences in caregiver demographics were not addressed in this study due to small numbers; however, this presents an opportunity for future exploration. In addition, although a strength of this study was the inclusion of three different models of service provision, the experiences of relatively few caregivers may not be transferable to experiences of the broad range of caregivers receiving LSL EI services across North America, much less globally. This limitation provides direction for future research to elicit the voices of caregivers from a variety of cultures and backgrounds, in a range of settings, in the broader context of LSL EI services. In addition, the design of the study and the number of participants precluded meaningful comparison between sites offering different models of service provision. However, it would be interesting to further explore these differences with a larger group of caregivers. Examining the views of LSL practitioners in future research will also enhance understanding of the caregiver coaching process. Finally, interpretive description necessitates that the researcher uses reflexivity to continually evaluate their response during data collection, analysis, and writing. The researcher's own positionality, pre-understandings, and experiences are considered by some to be integral to the research process and these important considerations should be identified and disclosed as a means to enhance the credibility of the study (Agrey, 2014; Berger, 2015; Holmes, 2020). Interpretation from the lead researcher's perspective as a parent of a child who is DHH and an LSL practitioner becomes a common ground from which to hear, co-construct meaning and learn from others. While I employed reflexivity throughout this work, my belief in collaborative caregiver coaching as an effective and family-centered approach to LSL EI services informed the research design and analysis and therefore may have impacted the results.

Caregiver coaching in LSL practice is a means by which caregivers learn to use enhanced language interactions to improve their child's language outcomes, ultimately resulting in self-efficacy and carryover of intervention strategies into their daily routines (Noll et al., 2021). This study is unique in that it explores from the perspectives of caregivers how LSL coaching influences their active role in communication intervention and achieving positive outcomes for their child and family. This work has the potential to help current and future caregivers of children who are DHH advocate for a partnered, collaborative approach to caregiver coaching. Additionally, this study provides insight for practitioners working to establish and maintain positive caregiver coaching relationships, including understanding the role of practitioner characteristics, explicitly establishing expectations, and adapting their coaching over time. This insight has the potential to impact the work of practitioners currently coaching caregivers as well as pre-service professionals learning the art and science of LSL caregiver coaching.

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# Improving Newborn Hearing Screening Through Collaboration and Communication

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## Abstract

**Purpose:** Hearing loss is the number one birth defect among children. There are significant consequences of delayed diagnosis and failure to obtain timely intervention, particularly for a child's speech and language development. Design and implementation of successful newborn hearing screening (NHS) programs can be challenging. The purpose of this paper is to demonstrate improved efficiency and effectiveness of a large NHS program through the implementation of a team approach engaging both ambulatory and hospital services.

**Methodology:** A Strengths, Weaknesses, Opportunities, and Threats (SWOT) analysis was used to develop an improved NHS program focused on improving patient care. The SWOT analysis outcomes were used to determine several key factors to be implemented, including dedicated technicians solely assigned to the NHS program and purchase of new equipment to improve accuracy and reduce disposable costs. In addition, a two-tiered approach was implemented whereby the dedicated technicians performed initial screenings, with all rescreens performed by an audiologist.

**Results:** Implementation of the new NHS program demonstrated numerous successes including a significant reduction in the failure rate, improved care coordination, and increased communication between ambulatory and hospital services.

**Keywords:** Newborn hearing screening, interdisciplinary collaboration, care coordination, congenital CMV

**Acronyms:** AABR = automated auditory brainstem response; ABR = auditory brainstem response; CMV = cytomegalovirus; EHDI = Early Hearing Detection and Intervention; NHS = newborn hearing screening; JCIH = Joint Committee on Infant Hearing; SWOT = Strengths, Weaknesses, Opportunities, and Threats

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Between 1 and 3 of every 1000 live newborns are identified as having congenital hearing loss, making it the most common birth defect (Centers for Disease Control and Prevention [CDC], 2018; Vohr, 2003). It is well established that untreated hearing loss in the first few years of life can have devastating consequences on a child's speech and language development. Hearing loss can also have significant psychosocial and academic ramifications for children (Tomblin et al., 2020; Wong et al., 2017). Key to improving these outcomes is prompt diagnosis and intervention to maximize early access to listening and spoken language through hearing aids and cochlear implants or to visual language via manual communication (Sininger et al., 2010; Yoshinaga-Itano et al., 1998).

Universal Newborn Hearing Screening (NHS) is a vital hospital program designed to ensure all infants with hearing loss are quickly identified. NHS programs also provide a framework for flagging infants at heightened risk for both congenital and late onset hearing loss, so they may be referred for more thorough testing and monitoring. This includes infants with craniofacial malformations, certain genetic syndromes, family history, in utero infections, other

serious health conditions such as meningitis, and history of medical treatments known to be associated with hearing loss (Joint Committee on Infant Hearing [JCIH], 2019). At notably increased risk are infants who receive care in the neonatal intensive care unit (NICU), a population whose rate of hearing loss has been reported as approximately 3% (Chang et al., 2020; Hille et al., 2007).

National Early Hearing Detection and Intervention (EHDI) guidelines from the JCIH (2019) state that all newborns should be screened for hearing loss before age 1 month, receive a diagnosis before age 3 months, and begin early intervention before age 6 months. Meeting this critical "1-3-6" timeline is associated with improved language outcomes for children with hearing loss and earlier activation of cochlear implants in deaf children (Yoshinaga-Itano et al., 2018). Although screening by age 1 month is the target, the JCIH advocates it be completed prior to hospital discharge. The recommended standard for newborns referred for diagnostic assessment is less than 4% (JCIH, 2007). There are considerable challenges to developing and implementing NHS programs to effectively meet these goals. Obstacles may include financial, technological, organizational, logistical, and human resource needs (Winston-Gerson & Ditty, 2021).

At the University of Kentucky Medical Center (UKMC) 2500 infants are screened for hearing loss every year on average. All screenings are conducted prior to hospital discharge, using automated auditory brainstem response (AABR) equipment. UKMC does not conduct outpatient rescreens after discharge, only full diagnostic auditory brainstem response (ABR) evaluations. Screening failure rate in the 2017–2018 fiscal year was 18% for initial screens and 5% for repeat screens. In addition to exceeding the limit of the national failure benchmark, the program's high rate of initial failures resulted in inflated program cost and time investment. Screenings were conducted by a large team of general medical technicians who had numerous job responsibilities and minimal NHS training. Limited program oversight and poor interdepartmental communication left gaps in patient care, particularly for infants with complex needs. Through the implementation of a team approach engaging both ambulatory and hospital services, the present project aimed to improve the efficiency and effectiveness of the UKMC NHS program.

## Method

An interdisciplinary team was formed, including hospital leadership and specialists from audiology, neonatology, otolaryngology, and infectious disease. Additionally, the chief of audiology was in communication with the Kentucky EHDI Board regarding the program redesign. Collaborators met over the course of a year to design and implement the new NHS program, with the goal of reducing the screening failure rate and improving overall patient care. A SWOT (Strengths, Weaknesses, Opportunities, and Threats) analysis was conducted. The results are outlined in Table 1. Based on the team's discussion of the SWOT, it was determined that the fundamental weaknesses affecting screening rate and patient care were inadequate staffing, older and inefficient equipment, and poor ambulatory/hospital communication in cases of screening failure. To address these weaknesses, a list of potential solutions was generated. Cost versus benefit analysis of each solution was discussed with regard to financial, time, and human resource ramifications. Feedback from patient care technicians, care team members, and families was taken into consideration. It was ultimately decided that several key changes would be enacted.

**Table 1**

*SWOT Analysis of Pre-Existing Newborn Hearing Screening (NHS) Program*

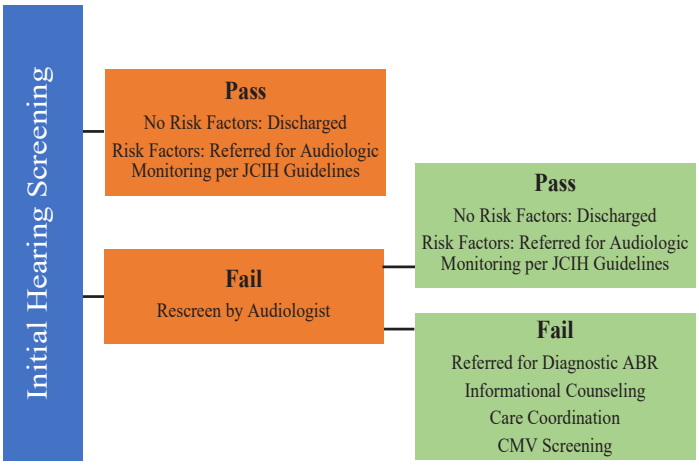
| Strengths | Weaknesses |
|---|---|
| • Large program with low rate of missed screenings<br>• Audiology department with nine licensed audiologists on site<br>• Access to large medical center resources such as patient care coordinators, social workers, etc.<br>• Support from leadership for programmatic improvements | • High screening failure rate compared to national standard<br>• High rate of missed diagnostic audiology follow-up appointments<br>• Poor communication between medical services for newborn nursery and complex NICU patients<br>• Disconnect between hospital and ambulatory care<br>• Inadequate staffing for screenings<br>• Lack of screening technicians with appropriate training and experience<br>• High cost of disposable materials |
| **Opportunities** | **Threats** |
| • Interdepartmental collaboration<br>• Advance technology and medicine<br>• Meet or exceed national screening guidelines<br>• Improve patient care<br>• Decrease loss to follow-up | • Pressure to reduce costs while maintaining high standard of care<br>• Time constraints of audiologists<br>• Non-NHS responsibilities and time constraints of screening technicians |

*Note. SWOT = Strengths, Weaknesses, Opportunities, and Threats.*

First, two medical technicians were hired as dedicated hearing screeners who were solely responsible for completing and documenting the screenings. They were specially trained on EHDI principles, equipment use, and troubleshooting. The program also purchased new AABR screening equipment with improved accuracy and significantly lower disposable cost. Lastly, a two-tiered screening approach was implemented in which a newborn who failed the initial screening was rescreened by an audiologist from UKMC (see Figure 1). All babies who failed the second screening were referred for a diagnostic ABR evaluation in the outpatient audiology clinic. Babies who passed the screening but had risk factors for delayed onset or progressive hearing loss were referred for audiologic monitoring as recommended by JCIH 2019 guidelines. Babies who passed with no risk factors were discharged from audiology service.

**Figure 1**

*New Two-Tiered Approach to Hearing Screenings*




*Note.* ABR = auditory brainstem response; CMV = cytomegalovirus; JCIH = Joint Committee on Infant Hearing.

NHS program changes initiated with the 2018–2019 fiscal year and were compared with the 2017–2018 fiscal year to determine the impact of the program redesign on screening failure rate. Pass/fail data were collected by the hearing screening technicians and audiologists. Feedback regarding care coordination and interdepartmental communication was collected from providers, technicians, and families by their respective interdisciplinary team members.

## Results

In the 2018–2019 fiscal year a total of 2,386 newborns received a newborn hearing screening at UKMC, including 1513 infants in the newborn nursery and 873 infants in the NICU (Figure 2). Of those screened, 9% failed the initial screening and 2% failed the second screening (Figure 3). This demonstrates a substantial decrease in the rate of failed screenings compared to the 2017–2018 fiscal year, in both the nursery and NICU populations (Figure 4). New NHS equipment achieved a disposable cost reduction of $11 per screening, resulting in approximately $30,000 savings to UKMC.

**Figure 2**

*Total Number of Hearing Screenings Conducted*

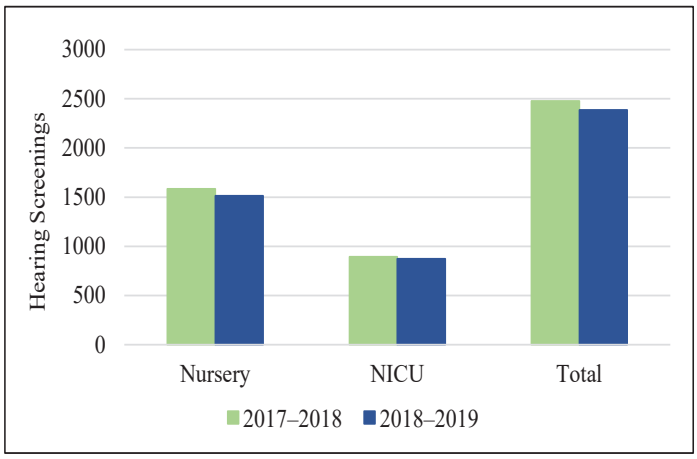



**Figure 3**

*Failed Rescreens Prompting Diagnostic Referral*

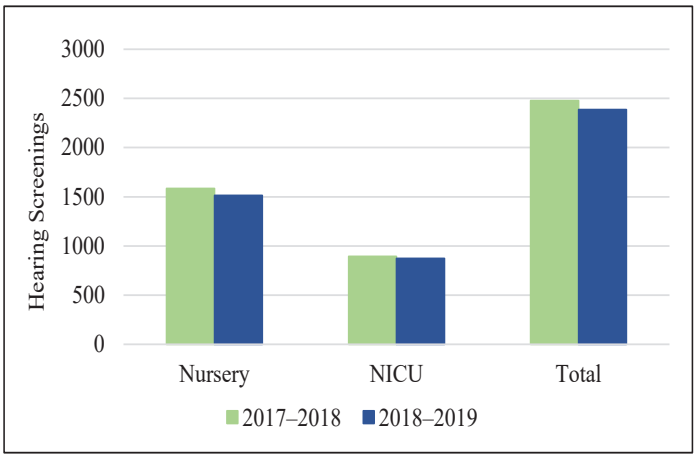
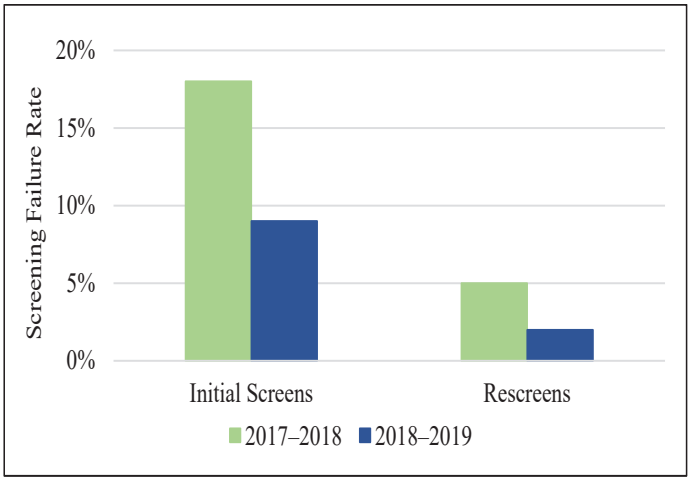



**Figure 4**

*Screening Failure Rates*




Under the new screening program, clinical audiologists completed all NHS program rescreens prior to newborn discharge. Audiologists were scheduled to take call for the NHS program for one week at a time, on a rotating schedule. They conducted screenings during

administrative or unstructured periods to ensure no loss of revenue-generating clinic time. The process was typically completed in 30 minutes or less. Their responsibilities included: scheduling the outpatient diagnostic ABR appointment immediately upon second screening failure, counseling parents on the failed screening results, providing pretest instructions for the ABR appointment, and communicating the screening results and appointment dates with other key professionals involved in the infant's care. For infants in the newborn nursery this included an infectious disease physician and NHS program director. For infants in the NICU, a neonatology physician and patient care coordinators were also informed.

A targeted congenital cytomegalovirus (CMV) infection screening program was initiated for all infants in the newborn nursery who failed the second hearing screening. This policy change did not impact patients in the NICU, as all newborns treated in this unit are screened for CMV upon admission.

## Discussion

The collaborative redesign of the NHS program has yielded numerous successes in the areas of patient care, care coordination, and value. A major improvement with respect to patient care is the significant decrease in both initial and secondary screening failures; both rates have been reduced by approximately 50%. Ongoing analyses indicate this improvement has been sustained even in the face of the COVID-19 pandemic, during which the UKMS NHS program has remained in full operation. The program continues to exceed its goal of meeting the JCIH quality benchmark of 4% or less of final referrals for diagnostic audiological testing. Although superior technical accuracy of the new AABR equipment cannot be overlooked, it is suspected that strengthened program staffing also plays a significant role in achieving this goal. The quality of screenings improved by hiring two technicians designated as hearing screeners, modifying their clinical responsibilities to ensure adequate time for screenings, and training them in principles of newborn hearing, equipment use, and troubleshooting.

Introduction of experienced audiologists to the secondary screening phase has also been essential to improving patient care. The audiologists provide expertise in hearing assessment and advanced equipment troubleshooting as needed. Perhaps more importantly, they also provide individualized counseling to families whose newborns fail the rescreen. The focus of counseling was basic principles of newborn hearing screening and diagnostic testing, and pre-test instructions to support a successful outpatient ABR. Informational counseling from a clinical audiologist was included in the program redesign with the hope of improving caregiver knowledge and reducing loss to follow-up. UKMC is currently collecting and analyzing loss to follow-up data to determine if the program changes positively impacted loss to follow-up rates.

Another success of the redesigned NHS program is improved care coordination and communication between ambulatory and hospital services. Ensuring proper time for conducting and communicating results of a hearing screen can be challenging amidst a newborn's many initial evaluations. This is particularly true for medically fragile babies who require lengthy hospital admissions and for whom hearing screening is not a priority. Typical communication and planning channels can also be interrupted by instability of a newborn's social situation, such as changes in custody or foster care. Deploying UKMC's audiologists as hearing care coordinators seeks to overcome these challenges. Immediately scheduling outpatient appointments, communicating results with families, and directly contacting medical team members ensures all stakeholders are informed of needed follow-up. Appointments can be scheduled on a timeline sensitive to the caregivers' logistical needs, which is another factor believed to play a significant role in loss to follow-up (Ravi et al., 2016). Designation of an NHS program director has also proven essential to care coordination and communication. The program director serves as a chief point of contact between hospital and ambulatory services, coordinates any emergent testing needs (e.g., congenital CMV or meningitis), oversees program implementation and statistic tracking, and resolves any programmatic issues or concerns that arise.

Lastly, NHS program changes have achieved significant time and financial savings to UKMC. The use of dedicated hearing screeners has allowed nursery and NICU directors to redeploy non-screening technicians to their primary job duties. Experienced screeners using new equipment with greater accuracy has greatly reduced the number of repeat screenings required, cutting both physical costs and time investment. The new equipment also requires significantly less disposable costs, at only $2.50 per screening. The original AABR equipment required nearly $14 in supplies per screening. Taking into account both initial screenings and rescreens, it is estimated that this substantial cost reduction has saved UKMC approximately $30,000 annually since the changes were implemented. Such savings have more than accounted for the initial cost of new equipment.

An unanticipated outcome of the program redesign has been development of a targeted CMV screening policy in the newborn nursery. CMV is the most common congenital viral infection in the United States, affecting approximately 1 in 200 newborns (CDC, 2020). It can result in significant central nervous system deficits, including congenital and delayed onset hearing loss. Early diagnosis allows for swift initiation of antiviral therapy, which may improve hearing outcomes in some affected newborns (CDC, 2020). Additionally, early diagnosis allows for initiation of close audiologic monitoring during a child's critical developmental years. This new collaborative screening policy elevates the standard of care provided to newborns at UKMC, in addition to bolstering interdisciplinary team involvement in the NHS program. It is the authors' hope that early identification of both congenital CMV infection and hearing loss will result in earlier involvement of other critical medical specialties and intervention services.

Future directions for this project will include outcome measures beyond hospital screenings, to see if successes in this phase do in fact result in lowered age of hearing loss

diagnosis and initiation of intervention at UKMC. Also of interest is probing the efficiency of the program's diagnostic phase, through measures such as outpatient ABR no-show rates and number of appointments required to obtain a diagnosis. Additionally, after the successful establishment of a working relationship between audiology and infectious disease, the authors are looking to expand partnerships with other key services to develop standardized hearing care paths for infants at higher risk of hearing loss.

## Conclusion

The NHS program at UKMC was collaboratively redesigned using results from a SWOT analysis completed by a multidisciplinary team. Key changes included designating two medical technicians as the sole hearing screeners, purchasing new equipment with improved technical accuracy and lower disposable costs, and implementing a two-tiered screening protocol by which audiologists completed all repeat screenings and provided information counseling and hearing care coordination. These changes resulted in decreased screening failure rates, increased communication between hospital and ambulatory services, improved care coordination, significant cost savings, and a new targeted CMV screening protocol in the newborn nursery.

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# Adaptation of the Conditioned Assessment of Speech Production in Spanish

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## Abstract

**Purpose:** The purpose of this article is to describe the adaptation of the Spanish version of the Conditioned Assessment of Speech Production (CASP).

**Method:** The authors adapted each segment into Spanish, then had 41 participants complete a survey to determine if each adapted segment was representative of the Spanish phonologic system. Thirty-six children (half with typical hearing, half with hearing loss) completed the CASP in English and Spanish. Paired samples *t*-tests were run to compare English and Spanish CASP scores between children with hearing loss and those with typical hearing.

**Results:** All segments were adapted as needed into Spanish. There was no statistical difference between the English CASP scores (18.61 ± 2.03) and Spanish CASP scores (18.78 ± 1.99) for the children with typical hearing. Similarly, there was no statistical difference between the English CASP scores (16.78 ± 3.44) and Spanish CASP scores (16.67 ± 3.41) for the children with hearing loss. Children with typical hearing scored statistically significantly higher on the English and Spanish CASP than children with hearing loss.

**Discussion:** The CASP-S is an appropriate Spanish adaptation of the CASP, which has been field-tested for use with young Spanish-speaking children with hearing loss.

**Keywords:** CASP-S, Spanish early speech production, speech assessment

**Acronyms:** AF = Advanced Forms; BCS = Basic Canonical Syllables; CASP-S = Conditioned Assessment of Speech Production–Spanish; CI = cochlear implant; EHDI = early hearing detection and intervention; JCIH = Joint Committee on Infant Hearing; PC = Precanonical; SAEVD-R = Stark Assessment of Early Vocal Development-Revised

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Early speech production prepares young children motorically to build their repertoire for early language development (Vihman et al., 1985). This motoric patterning leads to more advanced speech, which lays the building blocks for early vocabulary in young children. Children with hearing loss are at risk for speech delays due to limited auditory access (Joint Committee on Infant Hearing [JCIH], 2019; Oller & Eilers, 1988). Early identification and sensory aid use (e.g., hearing aids and cochlear implants) can counteract delays in speech production, and rapid development of early speech sounds typically occurs when sensory aid use begins (Apuzzo & Yoshinaga-Itano, 1995; Robinshaw, 1995). Assessing the early speech productions of children with hearing loss is crucial to identify those who are at-risk or already delayed so they can begin targeted therapeutic interventions (Ambrose et al., 2014; Eilers & Oller, 1994; Moeller et al., 2007a; Yoshinaga-Itano et al., 2017). Although there are assessments that assess early vocalization of English-speaking children, there are few for children with hearing loss and even less so for children with hearing loss who speak Spanish.

The Conditioned Assessment of Speech Production (CASP) is an efficient tool to assess the early vocal productions of young children with hearing loss who speak English (Ertmer & Stoel-Gammon, 2008). Due to the differences between English and Spanish phonological systems, the CASP is not an adequate tool to use with Spanish-speaking children with hearing loss. The Conditioned Assessment of Speech Production-Spanish (CASP-S) was adapted as a more appropriate assessment tool to document the early vocal productions of young children with hearing loss who speak Spanish. The purpose of this article is to describe the adaptation and initial field-testing of the CASP-S. The CASP-S was first adapted into Spanish segments by the authors. Then surveys were presented to Spanish-speaking speech-language pathologists and graduate students to identify appropriateness of the segments selected. Finally, field testing was conducted with 18 pairs of age- and gender-matched young Spanish-speaking children, half of whom have hearing loss.

**Speech Development in Children with Typical Hearing**

There is a large body of research that describes in depth the speech development of children who speak English (Poole, 1934; Prather et al., 1975; Sander, 1972; Templin, 1957) from which general guidelines were established for expected development of English speech production. Of particular importance for early vocalizations is the onset of canonical babbling, which is typically developed by 10 months of age (Nathani et al., 2007; Stark et al., 1993), and is universal across different languages (Ertmer & Moreno-Torres, 2009). This knowledge assists in understanding and identifying typical versus atypical development in even the earliest expected developing vocalizations in young children, regardless of language.

Spanish speech development has some distinctions from English speech development (Canfield, 1981; Dalbor, 1980; Gildersleeve-Neumann et al., 2008; Jimenez, 1987; Navarro, 1968). For example, Spanish does not contain all the same phonemes as English. Spanish has fewer phonemes than English, has some phonemes not represented in English, and uses its consonants (C) in phonologically different ways, even when those consonants are shared with English (Acevedo, 1993; Goldstein, 2015; Jimenez, 1987). Additionally, the majority of Spanish words end in vowels (V) and there are only 5 consonants that are used in the final position of words (i.e., /n/, /s/, /l/, /r/, and /d/) (Gildersleeve-Neumann et al., 2008). Spanish has a smaller number of initial consonant clusters, /s/ is never combined with another consonant in an initial cluster in Spanish, and Spanish has two types of "r" sounds (a tap /ɾ/ that is similar to an English flap /d/ and a trilled /r/), neither of which are produced like the English retroflex /ɹ/. Additionally, although English and Spanish share most of their phonemes and thus their ages of acquisition are very similar, there are more late-acquired fricative sounds in English than in Spanish; thus, Spanish consonants are typically mastered much earlier than English consonants (Acevedo, 1993). Due to these differences between English and Spanish speech development, English normative data for speech production beyond the earliest vocal productions cannot be applied to Spanish and Spanish-specific normative data have been developed (Acevedo, 1993; Goldstein, 2015; Jimenez, 1987).

**Vocal Development in Children with Hearing Loss**

The first months of vocal development, including crying, are very similar between children with and without hearing loss (Oller & Eilers, 1988; Stoel-Gammon & Otomo, 1986). Changes begin with vocal play and children with hearing loss will have delayed or deviant vocal development without the use of sensory aids. Several studies have confirmed that improved auditory access through sensory aids is associated with improvements in speech development in English similar to typically hearing peers. As Universal Newborn Hearing Screening has become the norm for infants born in the United States, more infants are being identified with hearing loss at earlier ages than before (JCIH, 2019). The Joint Committee on Infant Hearing emphasizes the importance of Early Hearing Detection and Intervention (EHDI) activities for identification of hearing loss as early as birth. The goal of their efforts has led to earlier identification of hearing loss and, subsequently, earlier entrance into early intervention. The JCIH's specific recommendations are known as 1-3-6 Goals, wherein all infants should have their hearing screened by no later than one month of age, hearing loss should be confirmed by three months of age, and early intervention services should begin as soon as diagnosis but no later than six months of age. Longer length of time of sensory aid use is associated with better speech outcomes, including more prelinguistic vocalizations, more complex structures, and faster prelinguistic/speech development when compared to children who are identified later and begin use of sensory aids later (Ambrose et al., 2014; Binos et al., 2013; Eilers & Oller, 1994; Fagan, 2014; Fulcher et al., 2012; Moeller et al., 2007a; Moeller et al., 2007b; Pratt et al., 2007; Salas-Provance et al., 2014; Tomblin et al., 2008; Tomblin et al., 2014; von Hapsburg & Davis, 2006). These findings demonstrate the importance of earlier identification and earlier use of sensory aids.

Several studies address early vocal development for young children with cochlear implants (CIs) and found that the use of precanonical vocalizations decreased as they produced more advanced speech-like vocalizations, and that vocal development milestones were typically reached with fewer months of hearing experience than for children with typical hearing (Ertmer et al., 2007; Ertmer, et al., 2013; Ertmer & Jung, 2012a, 2012b). Children with CIs likely achieve vocal developmental milestones with fewer months of hearing than hearing peers because they are older when they begin hearing. Cognitively, they are ready for word learning and they already have semantic concepts (visual representations or signs) to associate with a spoken label (Ertmer et al., 2007). This may be why children with cochlear implants "skip" the babbling stages. Additionally, it is important to stress assessment of early speech sound production to identify children with hearing loss who may be at risk for delays in speech development (Ambrose et al., 2014; Moeller et al., 2007b; Eilers & Oller, 1994). However, assessment tools are needed to assess early speech sounds in the target language.

In Spanish, there is extremely limited research on the early speech production of Spanish-speaking children with hearing loss who use sensory aids. Sosa and Bunta (2019) found that children with CIs had lower consonant and vowel accuracy and whole-word variability than peers with typical hearing. However, there were no differences between those rates in bilingual and monolingual children who were matched by hearing status. Additionally, bilingual language exposure did not appear to have a negative effect on the phonologic development of children with CIs. Moore et al. (2006) documented the early Spanish speech development of a toddler who had a CI activated at 20 months of age. They found that early speech production was similar to CI recipients learning English, but that post-implant overall production accuracy was greater than for English-speaking peers. Finally, Moreno-Torres (2014)

studied 8 Spanish-speaking children with hearing loss who were implanted before the age of 24 months. He found that the children's first words were similar to the types of babbling they were using and that their more advanced productions were constrained by Spanish prosodic structures. Taken all together, these few studies highlight two important findings. First is the urgent need for more research in the area of early vocal productions of Spanish-speaking children with hearing loss. Second is the need to consider that since the phonological systems of English and Spanish differ, it is necessary to produce language-specific norms, assessments, and interventions in Spanish for Spanish-speaking children with hearing loss.

**Test Adaptations**

Assessments are being translated and adapted at a higher rate than they were before (Matsumoto & van de Vijver, 2011). Test adaptations involve deciding whether the assessment can measure the same constructs in a different language, selecting appropriate items to translate, deciding on appropriate changes to be made in preparing a test for a second language, adapting it, and ensuring both forms of the assessment are equivalent. Assessments need to be adapted to facilitate comparative studies of achievement across cultural and language groups, can be more cost-effective than developing new tests, and can achieve fairness in assessment methods through establishment of equivalence of scores (Hambleton et al., 2012). Adaptations require significantly more than the translation of literal words from one language to another and are more highly involved with ensuring that they address the same concepts, words, and expressions that are culturally and linguistically equivalent in a second language and culture (Hambleton et al., 2012).

**Adaptation of the CASP**

The Conditioned Assessment of Speech Production (CASP) was developed to be a useful criterion-referenced vocal stimuli test that assesses vocal development in English-speaking children with hearing loss between the ages of 18 and 48 months (Ertmer & Stoel-Gammon, 2008). The CASP has been used to monitor vocal development of children with hearing loss through imitative and prelinguistic speech patterns, but these speech stimuli solely test English phonology. It is a time-efficient tool that allows quick regular clinical use (Ertmer & Jung, 2012a). It was developed on the premise that advancements in auditory access allow for improvements in vocal development for children with hearing loss. The benefits of the sensory aids are demonstrated when children's imitations and vocalizations become more complex, phonetically varied, and speech-like. Additionally, it was demonstrated that young children could be conditioned to imitate speech stimuli from a familiar person during a game-like activity.

The CASP used two published investigations as the basis for its development, both of which used the Stark Assessment of Early Vocal Development-Revised (SAEVD-R; Nathani et al., 2006), which classifies prelinguistic utterances of typically developing infants and toddlers during play with their mothers. The SAEVD-R was developed to use perceptual and articulatory characteristics of vocalizations to capture infant vocal productions. In the first study, Nathani et al. (2006) examined 30 infants (from 2 weeks to 20 months of age), recording their representative sound production behaviors 5 times within their age-group time span. From that, five levels of vocalizations were identified that describe typical infant and toddler vocalization in English-speakers that progress developmentally with age: Level 1: reflexive sounds, Level 2: control of phonation, Level 3: expansion, Level 4: basic canonical syllables, and Level 5: advanced forms.

In the second study, Ertmer et al. (2007) followed 7 children (4 girls and 3 boys) with hearing loss longitudinally. These children ranged from 10 to 36 months at the time they received CIs. Children were seen for two 30-minute data collections within 2 months before activation of their CIs, and at monthly intervals following CI activation until they met the criteria for completing vocal development on the SAEVD-R. Sessions were audio- and video-recorded and utterances were counted in each 10-minute segment. Results indicated longer periods of vocal development for children who were younger and that, typically, younger children completed vocal development earlier than children who were older when implanted. Five of the 6 children followed the expected hierarchical sequence of the SAEVD-R. Of particular interest in this study was the length of time it took for children to establish adultlike vocalizations (Level 4, basic canonical syllables and Level 5, advanced forms). Four of the 5 children who had not yet reached Level 4 at the beginning of the study were able to complete it within 17 months of CI activation. Six of the 7 children who had not yet reached Level 5 at the beginning of the study were able to do so within 11 months after CI activation.

Assessment tools like the CASP have allowed clinicians to assess early vocalizations for young children with hearing loss who speak English. However, appropriate assessment of children from homes that speak other languages than English is not possible with the CASP. Eighteen percent of the current U.S. population (325+ million individuals) is estimated to be Hispanic or Latino (United States Census Bureau, n.d.a), which represents the largest minority group in the United States. Additionally, over 21% (71+ million) of the population speaks a language other than English, with more than 27 million individuals reporting speaking English "less than very well". The Hispanic/Latino population is also expected to triple in size, making up 29% of the U.S. population by 2050 (Passel & Cohn, 2008). Hispanics are known to have a higher prevalence of hearing loss when compared to non-Hispanic Whites and non-Hispanic Blacks (Goman & Lin, 2016; Mehra et al., 2009), and about 1.8 million of the 11 million U.S. children under age 18 with at least 16 dB hearing loss are Hispanic (Niskar et al., 1998; United States Census Bureau, n.d.b). While the number of bilingual English/Spanish speakers continues to grow in the United States, the research on

bilingual (English/Spanish) and Spanish monolingual speech development in young children with hearing loss is extremely limited.

Since it is known how important it is to monitor progress in spoken language development soon after fitting of sensory aids, there is a pressing need to develop tools for children who are from Spanish-speaking homes. As the CASP only assesses English phonological systems, it is not an appropriate assessment for testing the emerging phonological system of Spanish-speaking children. Therefore, an appropriate assessment for Spanish-speakers is needed.

**Rationale for the Adaptation of the CASP-S**

The CASP-S is a Spanish adaptation of the CASP developed by Ertmer and Stoel-Gammon (2008). In line with the CASP, the CASP-S is a time efficient, game-like activity that measures prelinguistic vocal development in children with hearing loss by having them produce 10 different vocal utterances that follow a hierarchical sequence of development. These utterances move through the final 3 levels of vocal development of the SAEVD-R, namely the Precanonical (PC) level, the Basic Canonical Syllables (BCS) level, and the Advanced Forms (AF) level.

**Administration and Scoring of the CASP-S**

Administration of the CASP-S is the same as the CASP, and in-depth procedures can be found in Ertmer and Stoel-Gammon (2008). The clinician engages the parent to model for their child by providing models of the 10 utterances. Initially, the clinician role-plays with the parent by modeling the utterance for the parent in the game-like activity. The parent listens and repeats while the child observes the interaction. The parent's imitation is reinforced by having them stack a ring on a ring stacker toy. Following the clinician-parent interaction, the parent models the same utterance for the child and encourages the child to imitate. Having the parent model the utterance is advantageous for the child because a familiar partner is being used as the source of the stimulus. In sum, the CASP-S follows a clinician to parent, parent to child sequence of events per item. Complete instructions are given in Appendix A. The child's imitative response is then scored using a graduated scoring scale: 0 = *no attempt*, *not a close match*, 1 = *partially acceptable match*, and 2 = *fully acceptable match*. Criteria for each CASP-S item are included on the score sheet (Appendix B).

## Method

The adaptation of the CASP-S was completed in three phases. For the adaptation phase, the specific segments were adapted as needed to accurately represent Spanish phonological development. During the construct validity phase, the adapted segments were presented to a panel of native Spanish-speakers to identify which segments were the best representations of Spanish phonology. Finally, the validated segments were field-tested with children with hearing loss and age- and gender-matched peers.

**Segment Adaptation Phase**

***Segment Rationale for Changes from CASP to CASP-S***

To determine Spanish-appropriate segments, each item of the CASP was reviewed and adapted as needed by the authors based on general Spanish phonology. The following adapted segments for CASP-S moved to the validation stage (see Table 1). For vowels, Spanish has a basic five-phonemic vowel system of /i/, /e/, /a/, /o/, /u/, as opposed to English, which has a larger number of vowels. Due to the difference in the number of vowels, several vowel changes were required in the adapted version and all 5 vowels are represented in CASP-S. For consonants, the English consonants used in the CASP (i.e., /b/, /m/, /w/, /s/, /k/, /n/) are consonants used in Spanish and are expected to be mastered by 4 years and 6 months in typically developing Spanish-speaking children (Acevedo, 1993). Therefore, these consonants did not require adaptations and are all represented in CASP-S. The following vowels and consonants were used in each of the 10 total segments plus warm-up sounds:

Warm-up Sounds (open vowels for imitation and conditioning practice, elicited as a warm-up activity before the administration of CASP-S): the visually salient high back vowel /u/ and the mid back vowel /o/ are both found in Spanish and were not changed.

For Level 1 PC: precanonical vocalizations lack phonetic content and adult-like timing of true syllables. Because these vowels are not visually salient, they require the child to rely mainly on auditory information for imitation. The original CASP uses the mid-central /ʌ/, which is not in the vowel repertoire in Spanish. Therefore, the mid-low vowel /a/ was used. For item 2, the CASP used /i/, which is represented in the Spanish vowel repertoire. Consequently, that vowel was not changed for the CASP-S. For item 3, the CASP uses the low-front /æ/, which is not in the vowel repertoire in Spanish and the mid-front [e] replaced it.

For Level 2 BCS: basic canonical syllables consist of consonant-vowel (CV) syllable shapes with adult-like timing. Two kinds of canonical syllables are presented in the CASP, 3 CV syllables with highly visible consonants and 2 CV syllables containing consonants with minimal speech reading cues. The highly visible consonants emerge early in life, and in contrast, the less visually salient consonants represent later emerging sounds. As the consonants did not change, the only change made to the CASP-S was for the vowel. The low-back vowel /ɑ/ changed to the mid-low vowel /a/ for all segments.

For Level 3: Advanced Forms utterances include a consonant plus a diphthongized vowel syllable and a CVC syllable. Speechreading cues are minimal in these stimuli, thus requiring children to rely mainly on their auditory perception ability. The segment [naɪ] was judged an appropriate segment for Spanish and was not changed. By the age of 2, almost half the syllable types produced by Spanish speaking children are CV syllables. Accordingly, the consonants in CASP-S mostly appear in CV syllable

**Table 1**

*Adaptations to the Conditioned Assessment of Speech Production (CASP)*

| | CASP | CASP-S |
|---|---|---|
| Warm-up Sounds | /u/ and /o/ | /u/ and /o/ |
| **Level 1: Precanonical vocalizations** | | |
| 1. prolonged central vowel in isolation | 1. /ʌ/ | 1. /a/ |
| 2. two high-front vowels | 2. /i/ /i/ | 2. /i/ /i/ |
| 3. three low-front vowels | 3. /æ/ /æ/ /æ/ | 3. /e/ /e/ /e/ |
| **Level 2: Basis Canonical Syllables** | | |
| 4. CV syllable with bilabial stop consonant | 4. /bɑ/ | 4. /ba/ |
| 5. CV syllable with bilabial nasal | 5. /mɑ/ | 5. /ma/ |
| 6. CV syllable with bilabial glide | 6. /wɑ/ | 6. /wa/ |
| 7. CV syllable with velar stop | 7. /kɑ/ | 7. /ka/ |
| 8. CV syllable with lingua-alveolar fricative | 8. /sɑ/ | 8. /sa/ |
| **Level 3: Advanced Forms** | | |
| 9. C+ diphthong syllable | 9. /nai/ | 9. /nai/ |
| 10. CVC | 10. /tʌk/ | 10. /kon/<br>/don/<br>/tok/ |

*Note.* Adaptations are shown from CASP (English version) to CASP-S (Spanish version). C = consonant; V = vowel.

structures. Given the phonotactic constraints of Spanish, it should be noted that the diversity of consonants in CVC syllables is limited, and selecting a representative CVC sequence was the most challenging aspect of adapting and validating the CASP-S. For this final item, three segments were selected as appropriate: [kon], [don], and [tok].

## Construct Validity of CASP-S

### *Participants*

Following approval from Florida International University's Institutional Review Board, 44 participants who self-identified as native Spanish speakers listened to a presentation about CASP and CASP-S in the construct validity phase of the CASP-S. They were then asked to complete a survey about the representativeness of the Spanish segments selected for Advanced Form Level of the CASP-S. Participants included 37 speech-language pathology graduate students, 2 professors in the Communication Sciences and Disorders Department at Florida International University and 5 speech-language pathologists in Miami. Participants ranged in age from 21 to 70 years of age. Participants were given a scale to self-rank their Spanish proficiency on the Interagency Language Roundtable (ILR) Speaking Skill Scale. Criteria to be considered sufficiently proficient to participate in the study was to be at a level 2 or above on the ILR scale. Three participants did not meet criteria for participation in the study and were not included in the data analysis as 2 failed to state their level of proficiency and one had a proficiency level below 2. Forty-one total participants' data were analyzed. Spanish dialects represented by the 41 participants were Cuban, Colombian, Dominican, Venezuelan, Uruguayan, Panamanian, Argentinian, Nicaraguan, Peruvian, and Mexican. Thirteen of the 41 participants were immigrants.

### *Construct Validity Results*

Forty-one out of 44 surveys were considered when determining the representativeness of the Spanish segments proposed in CASP-S. The segments chosen for Level 1 and Level 2 of CASP-S were determined to be representative of an emerging Spanish phonological system. For Level 3, all participants agreed that the segment in the original English CASP for "alveolar nasal plus diphthong" /naɪ/ was an appropriate equivalent in Spanish and thus, should remain on the CASP-S. For the CVC item, 56% agreed that /kon/ was the most representative CVC segment, 27% agreed that /don/ was the most representative, 10% agreed that /tok/ was the most representative, and the remaining 7% agreed that a combination of either /kon/ and /tok/ or /don/ and /tok/ were equally the most representative. Seventeen percent felt that /tok/ was unrepresentative, 2% felt that /don/ was unrepresentative, and 0% felt that /kon/ was unrepresentative. In sum, /kon/ was determined to be the most representative CVC sample.

### Field Testing

Eighteen pairs of English-Spanish bilingual children were administered the CASP and the CASP-S, all of whom were from bilingual homes per parental report. Half of the children had hearing loss and the other half had typical hearing. Inclusion criteria for children with hearing loss was as follows: identified with moderate to profound hearing loss by 10 months of age, began wearing sensory aids (hearing aids or CIs) by 17 months of age, had no additional disabilities, were in schools where spoken language was used, and were not exposed to sign language. Inclusion criteria for children with typical hearing was as follows: had typical speech, language, and hearing development, and were matched to the children with hearing loss by gender and by age, within 4 weeks of age. Eighteen children with hearing loss and 18 children with typical hearing met inclusionary criteria and participated in the study.

Paired samples *t*-tests were run to determine if there were differences between English and Spanish scores for the children with typical hearing, between English and Spanish scores for children with hearing loss, in English scores between children with hearing loss and children with typical hearing, and in Spanish scores between children with hearing loss and children with typical hearing. There was no statistical difference between the English CASP scores (18.61 ± 2.03) and Spanish CASP scores (18.78 ± 1.99) for the children with typical hearing. Similarly, there was no statistical difference between the English CASP scores (16.8 ± 3.44) and Spanish CASP scores (16.67 ± 3.41) for the children with hearing loss. Children with typical hearing scored significantly higher (18.61 ± 2.03) on the English CASP than children with hearing loss (16.78 ± 3.44), a statistically significant increase of 1.83, $t(17) = 2.829$, $p < .05$. Children with typical hearing also scored higher (18.78 ± 1.99) on the Spanish CASP than children with hearing loss (16.67 ± 3.41), a statistically significant increase of 2.11, $t(17) = 2.801$, $p < .05$.

## Discussion

The CASP-S is an efficient, easy to administer adaptation of the CASP. The adaptation was completed by making changes to accurately represent Spanish phonology, validating the changes through field testing by native Spanish-speaking speech-language pathologists and graduate students, and field testing with 18 pairs of young English and Spanish-speaking children. The results indicate that the CASP-S was able to capture the early Spanish speech vocalizations in young children with hearing loss and was sensitive enough to identify statistically different productions in a similar way as the English CASP. Additionally, it was able to identify statistically different performance between children with typical hearing and children with hearing loss (face validity). These results demonstrate that the CASP-S is an appropriate measure to assist clinicians' ability to accurately document production and detection of early vocalizations and can be used to monitor changes in prelinguistic speech development in young Spanish-speaking children with hearing loss with repeated administration. This adaptation is a step forward that helps fill the gap of limited assessment procedures for young Spanish-speaking children with hearing loss. Future studies should be completed to measure the validity and reliability of the CASP-S, as well as to establish expected scores by age to use this assessment as a criterion referenced tool. This would allow clinicians to more specifically identify an age-level for a child's vocalizations, which could then be compared to both the child's chronological age and hearing age. This information would then assist in shaping individualized intervention goals for Spanish-speaking children with hearing loss.

### Limitations

There is little research on the vocal development of Spanish-speaking children with hearing loss. Additional research, test development, and test adaptations should be conducted in this area to better serve this growing population. This study was limited in size and geographical area, and therefore, the results may not be generalizable to all Spanish-speaking populations. This study may be used as the impetus for future test adaptations.

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## Appendix A

Directions for Administering and Scoring the Conditioned Assessment of Speech Production – Spanish (CASP-S)
Alliete R. Alfano, Daniel Gonzalez, and David J. Ertmer

1. Warm-up Items
   a. After getting the child's attention, the clinician models the first warm-up vocalization (/u/) while holding a toy reinforcer (e.g., ring piece for the ring-post toy) next to her mouth. Models are spoken at slightly louder than conversational intensity level and without unusual visual or intonation cues. The clinician says /u/ or "Say /u/" while looking at the parent.
   b. The parent imitates the modeled vocalization. The parent is given the reinforcer and places it on the post. Parent and clinician respond enthusiastically as the ring is placed on the post.
   c. The parent gets the child's attention and models the same vocalization (i.e., /u/ or "Say /u/") while holding the toy reinforcer next to his or her mouth and looking at the child. When the child vocalizes, he or she is praised and is allowed to place a ring on the post. Any vocalization is reinforced. To maintain a game-like situation, the child is allowed to place the ring on the post even if he or she has made no attempt to imitate.
   d. If the child does not respond to /u/, repeat steps a–c with a warm-up vocalization /o/. If the child attempts to imitate either warm-up item, move to level 1.

NOTE: Clinicians may choose to modify these procedures if the child is familiar with a different, previously established routine for eliciting speech (e.g., if the reinforcer is typically given to the child before an imitative attempt). If the child responds more consistently to the clinician than the parent, the clinician and the parent roles can be reversed. Two familiar clinicians can also administer the CASP-S if parents are unavailable; however, the parent should participate in the process whenever possible. Three adult models are given before the child is expected to imitate each item.

2. Testing
   a. The clinician models the first vocalization of level 1 for the parent as described in step 1a.
   b. The parent imitates the vocalization and receives a ring reinforcer.
   c. The parent turns to the child, gets his or her attention, and models the vocalization while holding the star next to his or her mouth. The child imitates the model.
   d. All of the child's imitative attempts are praised and reinforced immediately. The clinician transcribes the child's response in the space provided on the score sheet.
   e. If the child's production is fully acceptable (receives 2 points), go to the next stimulus item and repeat steps 2a–d.
   f. If the child does not respond or the imitative response is not fully acceptable, note NR (no response) or transcribe the child's original attempt on the first line under the stimulus item.
      - Repeat steps 2a–d with the same stimulus to give the child a second chance. Transcribe and score the child's second attempt.
      - Only one repetition is allowed for each stimulus item.
      - The ring reinforcer is given even if the child does not respond.
   g. Continue introducing other stimulus items as in steps 2a–d until all the items at level 1 (precanonical) have been presented to the child.
   h. Present stimulus items for level 2 (basic canonical syllables) using the procedures in steps 2a–g.
   i. If the child scores at least 1 point on level 2, present stimulus items from level 3 (advanced forms) following steps 2a–g. Testing may be discontinued if the child does not receive any points on level 2 and the parent reports that the child rarely produces canonical (CV) syllables. If the child is reported to produce canonical syllables, present all stimulus items.

3. Scoring
   a. Scoring criteria are given on the score sheet.
   b. If more than one imitation is elicited, score only the most acceptable imitative response (i.e., the response with the highest score).
   c. Compare the child's productions with the parent/clinician's model. For example, an imitative production can be fully acceptable if it matches a model that was slightly different from the intended target (e.g., Mother says /kan/ instead of /kon/ and child says /kan/).
   d. Add up the number of points for the total score.

4. Repeat Testing

The CASP-S can be given at 2-, 3-, or 4-month intervals. Compare results with the previous scores for the same child.

## Appendix B

### The Conditioned Assessment of Speech Production - Spanish (CASP-S)

**Alliete R. Alfano, Daniel Gonzalez, and David J. Ertmer**

**Child's Name**_____ **DOB**_____ **CA**_____ **Date**_____

**Parent**_____ **Clinician**_____ **Sensory aid type**_____ **Months of sensory aid use**_____

**Directions for parents: I am going to say some sounds for you to imitate. Then you will say the same sounds for your child to imitate. Try to say the sounds in the same way and at the same loudness level that I use. We will give (child's name) toys and praise for playing this game with us.**

**Instrucciones para los padres: Voy a decir algunos sonidos para que me imites. Después vas a decir esos mismos sonidos para que su hijo/a los imite. Intente decir los sonidos de la misma manera y volumen de voz que yo. Vamos a premiar y darle juguetes a (nombre del cliente) por jugar con nosotros este juego.**

**Warm-up Sounds:**

/u/: Child imitates readily _____ Imitates after pause_____ No Response_____
/o/: Child imitates readily _____ Imitates after pause_____ No Response_____

Level 1: Precanonical Vocalizations

| Stimuli for Models Transcribed Responses | 0 points | 1 point | 2 points | Score |
|---|---|---|---|---|
| 1. Prolonged central vowel in isolation: /a/<br>1. _____<br>2. _____ | 1. No response<br>2. Two or more vowels that do not match target<br>3. Response is not a vowel (e.g., squeal, raspberry, click, /m:/, /s:/)<br>4. CV syllable(s) without target vowel (e.g., /bu/) | 1. Two or more vowels that match target<br>2. Single vowel that is not /ə/<br>3. CV syllable containing target vowel (e.g., /ba/) | 1. One central vowel (i.e., /a/) | |
| 2. Two high-front vowels: (/i/ /i/)<br>1. _____<br>2. _____ | 1. No response<br>2. Response is not a vowel<br>3. Syllables with vowels that do not match target (e.g., /bu/) | 1. Single vowel that matches target<br>2. Two vowels that are not /i/<br>3. Two vowels, only one of which matches the target (e.g., /i/ /ə/)<br>4, CV syllables containing target vowel (e.g., [bibi]) | 1. Two high front vowels (i.e., /i/) | |
| 3. Three mid-front vowels: (/e/ /e/ /e/)<br>1. _____<br>2. _____ | 1. No response<br>2. Response is not a vowel<br>3. Syllables with vowels that do not match target (e.g., /bu/) | 1. Single /e/<br>2. Two matching vowels (e.g., /e/ /e/)<br>3. Three vowels, only one /e/<br>4. Two or three non-matching vowels (i.e., none are /e/)<br>5. CV syllables containing target vowel (e.g., [bebebe]) | 1. Three mid front vowels (i.e., /e/) | |

Level 2: Basic Canonical Syllables

| Stimuli for Models<br>Transcribed Responses | 0 points | 1 point | 2 points | Score |
|---|---|---|---|---|
| 4. CV syllable with bilabial stop consonant: [ba]<br>1. _____<br>2. _____ | 1. No response<br>2. Vowel without consonant | 1. CV syllable in which only the C or the V match the model (e.g., [bi] or [ka])<br>2. Two or more matching CVs (e.g., [bababa] or [papə])<br>3. CVC syllable with matching C or V | 1. A single CV with a bilabial stop consonant and /a/ or /ə/ (i.e., [pa], [bə], or [pə]) | |
| 5. CV syllable with bilabial nasal: [ma]<br>1. _____<br>2. _____ | 1. No response<br>2. Vowel in isolation<br>3. Consonant in isolation | 1. CV syllable in which only the C or the V match the model<br>2. Two or more matching CVs (e.g., [mamama] or [məmə])<br>3. CVC syllable with matching C or V | 1. A single CV with a bilabial nasal consonant and /a/ or /ə/ (i.e., [ma] or [mə]) | |
| 6. CV syllable with labiovelar glide: [wa]<br>1. _____<br>2. _____ | 1. No response<br>2. Vowel in isolation<br>3. Consonant in isolation | 1. CV syllable in which only the C or the V match the model<br>2. Two or more matching CVs (i.e., [wawawa] or [wəwə])<br>3. CVC syllable with matching C or V | 1. A single CV with a labiovelar glide /w/ and /a/ or /ə/ (i.e., [wa] or [wə]) | |
| 7. CV syllable with velar stop: [ka]<br>1. _____<br>2. _____ | 1. No response<br>2. Vowel in isolation<br>3. Consonant in isolation | 1. CV syllable in which only the C or the V match the model<br>2. Two or more matching CVs (i.e., [gagaga] or [kəkə]) | 1. A single CV with /k/ or /g/ and /a/ or /ə/ (i.e., [ka], [ga] or [kə], [gə]) | |
| 8. CV syllable with alveolar fricative: [sa]<br>1. _____<br>2. _____ | 1. No response<br>2. Vowel in isolation<br>3. Consonant in isolation | 1. CV syllable in which only the C or the V match the model<br>2. Two or more matching CVs (i.e., [səsəsə] or [zaza])<br>3. CVC syllable with match C or V | 1. A single CV with /s/ or /z/ and /a/ or /ə/ (i.e., [sa], [za] or [sə], [zə]) | |

Level 3: Advanced Forms

| Stimuli for Models<br>Transcribed Responses | 0 points | 1 point | 2 points | Score |
|---|---|---|---|---|
| 9. C + diphthong syllable: [naɪ]<br>1. _____<br>2. _____ | 1. No response<br>2. Isolated vowel<br>3. Isolated C (e.g., /m/)<br>4. CV without a diphthong<br>5. Non-matching diphthong (e.g., /ui/) | 1. Matching diphthong in isolation<br>2. /n/ + non-matching diphthong (e.g., [nɔɪ])<br>3. Non-matching C with matching diphthong (e.g., [maɪ])<br>4. /n/ plus vowel (e.g., [na])<br>5. CVC syllable with /n/ and /aɪ/ (e.g., [naɪk]) | 1. /n/ plus matching diphthong (i.e., [naɪ]) | |
| 10. CVC: [kon]<br>1. _____<br>2. _____ | 1. No response<br>2. Vowel in isolation<br>3. Isolated consonant (e.g., /s/)<br>4. VC or CV syllable | 1. CVC syllable with non-matching Cs and V (e.g., [pip])<br>2. CVC syllable with one or two segmental errors (e.g., [kop]) | 1. CVC syllable with initial /k/ or /g/ and final /n/ combined with /o/ or /a/ (e.g., [kon], [gon], [kan], [gan]) | |

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# Association Between Craniofacial Anomalies and Newborn Hearing Screening Fail Rate

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## Abstract

**Purpose:** Increased knowledge of the prevalence of various craniofacial anomalies and their associated risks for hearing loss can help (a) guide the development of evidence-based practice regarding detection and documentation of risk factors at birth, and (b) health care professionals make appropriate recommendations for follow-up testing and monitoring.

**Method:** Records were reviewed for 39,813 infants born at two hospitals between January 1, 2014 and December 31, 2019 to determine the association between the presence of craniofacial anomalies and newborn hearing screening fail rates. Prevalence of confirmed hearing loss for infants born with and without risk factors were also examined. Additionally, surveys were sent to state EHDI programs and newborn hearing screening program coordinators across the United States to determine how facilities document risk factors for hearing loss, specifically craniofacial anomalies.

**Conclusions:** Study outcomes revealed four primary conclusions: (a) Infants with craniofacial anomalies are at a greater risk for failing their newborn hearing screening; (b) There is a need to better delineate craniofacial anomaly risk factors into subgroups; (c) Follow-up audiologic evaluations are not warranted for infants with preauricular sinuses/tags and; (d) A universal protocol needs to be developed for recording risk factors for all infants and for training newborn hearing screening (NBHS) staff to identify such risk factors.

**Keywords:** newborn hearing screening, risk factors, craniofacial anomalies, hearing loss, Early Hearing Detection and Intervention, Neonatal Intensive Care Unit

**Acronyms:** AABR = automated auditory brainstem response; CFA = craniofacial anomalies; DPOAE = distortion product otoacoustic emissions ECMO = extracorporeal membrane oxygenation; EHDI = early hearing detection and intervention; NBHS = newborn hearing screening; OAE = otoacoustic emissions; TEOAE = transient-evoked otoacoustic emissions; WBN = Well-Baby Nursery

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Universal newborn hearing screening (NBHS) programs have been established to provide the early detection of, as well as guide intervention for, hearing loss in newborns (Joint Committee on Infant Hearing [JCIH], 2019). One of the primary goals of NBHS is to "maximize linguistic competence and literacy development for children who are deaf or hard of hearing" since they are more likely to fall behind their hearing peers in communication, cognition, reading, and social-emotional development (p. 898, JCIH, 2007) without early intervention. The 1-3-6 Early Hearing Detection and Intervention (EHDI) model recommends that all infants be screened by one month of age, identified with hearing loss by three months of age, and receive intervention by six months of age (JCIH, 2019) so that they have the best chance to reach their potential.

The Joint Committee on Infant Hearing (JCIH) currently identifies 12 risk indicators that are associated with congenital, late onset, or progressive hearing loss in newborns: family history of childhood hearing loss, neonatal intensive care unit (NICU) stay of greater than five days, hyperbilirubinemia with exchange transfusion, treatment with ototoxic medications for greater than five days, asphyxia or hypoxic ischemic encephalopathy, extracorporeal membrane oxygenation (ECMO), in-utero infections, craniofacial conditions and physical conditions associated with hearing loss, syndromes associated with hearing loss, perinatal or postnatal bacterial and/or viral meningitis or encephalitis, events associated with hearing loss, and family/caregiver concern. It is imperative that infants identified as having one or more of these risk factors be closely monitored and re-evaluated routinely to rule out later onset or progressive sensorineural, mixed, or conductive hearing loss, regardless of NBHS screening results. The specific timing and number of these evaluations vary for each individual based on the identified

risk factor(s) and clinical judgment of the audiologist and/or primary care provider. It is recommended, however, that infants who have a craniofacial anomaly (CFA), regardless of the type, be re-evaluated by nine months of age (JCIH, 2019). It is the responsibility of the pediatrician or primary care provider (also known as *the medical home*) to monitor these risk indicators to ensure that audiological evaluations are completed as recommended (JCIH, 2019).

Research suggests that there is a lack of knowledge among healthcare professionals as to which of the aforementioned risk factors are "discoverable, predictive, and useful" (Karace et al., 2014, p. 262). This lack of knowledge lessens the effectiveness of initial screening and the impact of the JCIH guidelines, as well as the occurrence of follow-up testing, because medical professionals and newborn hearing staff are often unable to recognize the need for follow-up hearing testing when a specified risk factor is present (Hutt & Rhodes, 2008). Increasing knowledge about the various risk factors and their associated risk for hearing loss will help guide evidence-based practice and policy development regarding the detection and intervention of hearing loss in infants (Hutt & Rhodes, 2008).

The present study addresses NBHS outcomes for infants who have a documented CFA. CFAs are defined as those that include microtia/atresia, ear dysplasia, oral facial clefting, white forelock, microphthalmia, congenital microcephaly, congenital or acquired hydrocephalus and/or temporal bone abnormalities, and skull malformations (JCIH, 2019). CFAs are also found within certain syndromes such as: Trisomy 21, Treacher Collins syndrome, Waardenburg syndrome, CHARGE Association, Crouzon disease, Klippel-Feil syndrome, DiGeorge syndrome, Goldenhar syndrome and Pierre Robin syndrome (Greydanus et al., 2007). There is substantial evidence that the CFA risk factor group yields the highest prevalence of hearing loss in infants and children, however, the prevalence of hearing loss associated with each specific craniofacial disorder has not been consistently reported in the literature (Appelbaum et al., 2018; Cone-Wesson et al., 2000; Dumanch et al., 2017; Yelverton et al., 2013). For example, the published prevalence of hearing loss in cleft lip and cleft palate ranges from 26% to 82% (Chen et al., 2008; Viswanathan, Vidler, & Richard, 2008) and .3% to 18% for preauricular sinuses and tags (Firat et al., 2008; Kankkunen & Thiringer, 1987; Roth et al., 2008). These variations appear to be due to different methods for ascertaining the presence of hearing loss and risk factors including newborn hearing screenings, retrospective review of medical charts, and auditory brainstem response threshold assessments. There is also insufficient data on prevalence figures for other CFAs such as malformed ears, microtia, and skull malformations. Consequently, the exact association between each specific CFA and hearing loss risk at birth is unknown. Risk factors are only as "useful as their predictive power" (Karace et al., 2014, p. 262); therefore, it is imperative to determine the associated risk for hearing loss at birth for each of these disorders. In turn, this could lead to the development of effective follow-up guidelines and recommendations appropriate to each CFA. Since not every CFA has the same incidence/prevalence of congenital, progressive, or late-onset hearing loss, this clarification is crucial.

Aside from a lack of knowledge concerning the prevalence of each specific CFA and their respective contributions to NBHS fail rates, there is also a lack of documentation in state databases regarding risk factor information from hospitals in the country (Hutt & Rhodes, 2008; White, 2014). It is a guideline, not a requirement, to record risk factors in the NBHS databases (JCIH, 2019). Current risk factor registers are designed to ensure that newborn infants who need evaluation and follow-up are identified, however, these registers often lack specific/universal criteria, are under-utilized, and NBHS programs likely underreport various risk factors associated with hearing loss in their databases (Hutt & Rhodes, 2008). Without documentation of risk factors, healthcare providers are unable to ensure efficient, effective, timely and appropriate follow-up recommendations. For example, in a study examining birth certificate records, Purcell and colleagues (2018) "found that only 39% of children with cleft palates were correctly identified as having a craniofacial risk factor [for hearing loss] at the time of hearing screening", which ultimately led to a delayed diagnosis of hearing loss for many of these children (p. 26). That is, many of these children may have initially passed their newborn hearing screening but developed progressive/late onset hearing loss that was not caught. This delayed diagnosis of hearing loss may have been due to a lack of follow-up and monitoring for these children. Determining which CFAs result in a child having a greater risk for childhood hearing loss, in turn, ensures adequate and appropriate follow-up and intervention.

The following were the specific research questions examined in this study:

1. What is the association between the presence of CFAs (as a general category) and NBHS fail rates for infants born at two hospitals, Adventist HealthCare Shady Grove Medical Center (AHC SGMC) and Adventist HealthCare White Oak Medical Center (AHC WOMC) in the Greater DC area?
2. What is the association between specific CFAs and NBHS fail rates for infants born at AHC SGMC and AHC WOMC?
3. What is the prevalence of confirmed hearing loss (conductive, mixed, or, sensorineural) for infants born at AHC SGMC and AHC WOMC who failed the NBHS, with or without risk factors?
4. What information does each state's EHDI program require from individual screening programs as it pertains to the documentation of CFAs? How successful are the EHDI programs in obtaining such information?
5. How well are NBHS programs across the country recording and documenting information about the presence of risk factors for hearing loss and CFAs?

## Method

This study was approved by the Institutional Review Boards at Adventist HealthCare (2019-29) and Gallaudet University (Legacy-IRB-FY20-04). Descriptive statistics were used to report the outcomes obtained from this study.

### Participation

To answer the first three research questions, records were reviewed for 39,813 infants born at AHC SGMC and AHC WOMC over a six-year period between January 1, 2014 and December 31, 2019; reporting requirements remained consistent during this time frame. Both hospitals provide newborn hearing screening services, but not diagnostic evaluations.

The following data were retrieved from the Maryland Early Hearing Detection and Intervention (MD EHDI) program database, also known as OZ: (a) risk factor(s) present for hearing loss, (b) NBHS outcomes, (c) outpatient screening outcomes (if applicable and if available), and (d) diagnostic outcomes (if applicable and if available). If specific CFA information was missing, or if confirmation of OZ was needed, the hospitals' electronic medical records were subsequently reviewed. A review of both hospitals' NBHS program department records was also conducted to obtain information about specific CFAs for each infant, and when available, to clarify any discrepancies in OZ, as well as identify any risk factors for hearing loss that were incorrectly documented in OZ. Incorrect documentation could include omissions of risk factors in the state database and inaccurately recorded results in the hospital records.

Both hospitals in this study used a two-step screening protocol. All babies in the Well-Baby Nursery (WBN) without a risk factor for hearing loss were tested using either transient otoacoustic emissions (TEOAE) or distortion product otoacoustic emissions (DPOAE). If an infant failed the initial screening, a second OAE test was performed the next day. If the infant failed the second OAE, then an automated auditory brainstem response (AABR) screening was performed. All babies born with a risk factor for hearing loss (with the exception of preauricular pits and preauricular tags) or treated in the NICU were screened using AABR. If the infant failed the initial AABR, a subsequent and final AABR was performed, time permitting.

All OAE and AABR equipment were calibrated annually according to manufacturers' guidelines. AHC WOMC uses the Otodynamics Otoport for portable, bedside TEOAE screening, while AHC SGMC uses the Maico EroScan DPOAE for bedside DPOAE screening. For AABR and additional TEOAE screening, both hospitals use the Intelligent Hearing Systems (IHS) Smart Screener Plus.

To answer research questions 4 and 5, two separate surveys were distributed to: (a) State EHDI leaders and (b) NBHS program coordinators throughout the United States. The EHDI state leader survey can be found in Appendix A, while the NBHS Coordinator Survey can be found in Appendix B. Participants for both surveys signed an informed consent form and remain anonymous. The surveys were designed to be completed easily and quickly by participants using multiple choice, multi-answer, yes/no, and open-ended questions. Sample surveys were piloted with two independent audiologists (one state leader and one NBHS program coordinator) to ensure ease of completion, address any ambiguities, and determine the time needed to complete the surveys.

Surveys were designed and posted on a secure online platform (REDCap), where participants were able to access and complete the survey anonymously. Requests for participation for each survey were distributed electronically in three ways. The first was through two professional audiology groups (American Speech-Language-Hearing Association and the American Academy of Audiology). The second was through posting on two audiology Facebook pages (Audiology Antics and Anecdotes- for professionals only, and Audiology Happy Hour). The last was by distributing two different emails to all state EHDI leaders. The first email requested their participation in the State EHDI Leader Survey; this email included a link to the REDCap survey. The second email requested that they forward via email a description of, and a link to, the NBHS Coordinator Survey to NBHS coordinators in their state. To complete the NBHS Coordinator Survey, the participants had to currently be in charge of a NBHS program at a hospital, birthing center, NICU or other facility providing NBHS. A follow-up email was distributed to EHDI state leaders four weeks after the first email was sent if they had not yet completed the survey.

For the EHDI state leader survey, data from 13 states was received. Participating states are not identified in this paper to protect the privacy of those who responded. A total of 90 participants responded to the NBHS program coordinator study with a total of 18 states being represented across various regions of the United States; one participant did not report in which state they practiced. Descriptive statistics were used to analyze responses from both surveys.

## Results

### Presence of Any CFA and NBHS Fail Rates

There were 39,813 infants born at AHC SGMC and AHC WOMC between January 1, 2014 and December 31, 2019. A total of 2.05% ($n$ = 817) of all infants born during this period had a CFA that was documented in the department's paper records and/or hospital electronic medical records. The hearing screening fail rate for those identified with any CFA was 4.41% ($n$ = 36), compared to the overall fail rate of 0.74% ($n$ = 293) for all newborns, with or without a risk factor for hearing loss. The majority of the 293 infants who failed the NBHS had no risk factors for hearing loss (64.51%, $n$ = 189), while 12.29% ($n$ = 36) had a CFA, and 23.20% ($n$ = 68) had other risk factors for hearing loss.

### Association Between Specific CFAs and NBHS Fail Rates

As described earlier, a total of 817 of the 39,813 infants seen for a newborn hearing screening were identified as having a CFA. This group of infants was further categorized based on the specific anomalies present. The syndromes (associated with hearing loss) identified in our population were: Trisomy 13, Trisomy 18, 13q syndrome, Osteogenesis Imperfecta, Achondroplasia, Waardenburg syndrome, Goldenhaar syndrome, Pallister Killian syndrome, Beckwith Wiedemann syndrome, Turners syndrome, Albinism, and Cornelia de Lange syndrome. Other CFAs present included preauricular sinuses/tags, atresia/microtia, malformed ears, skull malformations, and cleft lip/palate. The prevalence for each of these specific CFAs and their associated NBHS fail rates are reported in Table 1. It is important to also note that two infants in the cleft lip/palate category were counted in two categories; once in the skull malformation category and once in the syndrome category.

**Table 1**

*Prevalence of Specific Craniofacial Anomalies (CFAs) at Birth and the Associated Newborn Hearing Screening (NBHS) Fail Rate*

| Category of CFA | Total Percentage of CFA (*n* = 819)[a] % *(n)* | NBHS Fail Rate (*n* = 36) % *(n)* |
|---|---|---|
| Syndrome associated with hearing loss | 9.4% (77) | 18.18% (14) |
| Ear Sinuses/Tags | 82.25% (672) | 0.74% (5) |
| Atresia/Microtia[b] | 1.47% (12) | 100% (12) |
| Malformed Ears, other | 1.35% (11) | 36.36% (4) |
| Skull Malformations | 0.61% (5) | 20% (1) |
| Cleft Lip/Palate | 4.16% (34) | 5.8% (2)[a] |
| Other | 0.98% (8) | 0.00% (0) |

[a]includes two babies with other CFAs: Syndrome & Skull Malformations

[b]babies were tested with Automated Auditory Brainstem Response only

Considering that 82.25% of babies with a CFA had preauricular sinuses and tags, this category was further analyzed. Table 2 shows the prevalence of, and the fail rate for, each sub-category of preauricular sinuses and tags. Regardless of whether a sinus and/or tag was unilateral or bilateral, the NBHS fail rate was less than 1%.

In reviewing the records of infants with a CFA who had failed their NBHS (*n* = 36), 58.33% (*n* = 21) did not exhibit any additional risk factors, while 41.66% (*n* = 15) did have an additional risk factor. Of the latter, a total of 22.22% (*n* = 8) of these infants spent greater than five days in the NICU; 11.11% (*n* = 4) had two other risk factors (e.g. NICU stay greater than five days and ototoxic medications greater than five days; ECMO and a NICU stay greater than five days); and 8.33% (*n* = 3) had three other risk factors present (cytomegalovirus, NICU stay greater than five days, and ototoxic medications greater than five days; ototoxic medications greater than five days, NICU stay greater than five days, and ventilator; or, ototoxic medications greater than five days, NICU stay greater than five days, and loop diuretics).

**Table 2**

*Prevalence of Unilateral vs Bilateral Preauricular Sinuses/ Tags and the Associated Fail Rate*

| Category of CFA | Prevalence *(n)* | NBHS Fail Rate *(n)* |
|---|---|---|
| Unilateral Preauricular Tags | 30.95% (208) | 0.96% (2) |
| Unilateral Preauricular Sinuses | 51.34% (345) | 0.29% (1) |
| Bilateral Preauricular Tags | 2.68% (18) | 0.00% (0) |
| Bilateral Preauricular Sinuses | 14.43% (97) | 0.21% (2) |
| Preauricular Sinuses and Preauricular Tags | 0.61% (4) | 0.00% (0) |

*Note*. NBHS = Newborn Hearing Screening; CFA = Craniofacial Anomaly.

### Prevalence of Confirmed Hearing Loss for Infants Who Failed the Newborn Hearing Screening, With or Without Risk Factors

Data from the outpatient hearing screenings, as well as diagnostic outcomes for infants who had failed their newborn hearing screening, were accessed through OZ. For the 293 infants who failed the newborn hearing screening, 70.99% (*n* = 208) were referred for a follow up screening, and were seen either internally or at an outside clinic. Of these infants, 69.23% (*n* = 144) passed the rescreening, 14.90% (*n* = 31) failed the rescreening, and 15.87% (*n* = 33) were lost to follow-up. The 31 infants who failed their rescreening were referred to a local children's hospital for diagnostic testing. Those results revealed that 43.75% (*n* = 13) infants had normal hearing acuity, 46.88% (*n* =15) were diagnosed with hearing loss, and 9.38% (*n* = 3) were lost to follow-up. Most of the infants who failed their initial screening and had a risk factor for hearing loss (*n* = 85) were referred directly to a pediatric audiologist for diagnostic evaluation. Hearing loss was identified in 48.23% (*n* = 41) of these infants, while normal hearing acuity was found in 30% (*n* = 26), and 21.12% (*n* = 18) were lost to follow-up. Note that the definition of *normal hearing* and *hearing loss*, as well as the degree and type of hearing loss, were not provided in OZ, so parameters are unknown.

A risk factor was present in 76.79% (*n* = 43) of the 56 infants who were diagnosed with hearing loss, while no risk factors were present in 23.21% (*n* = 13). A CFA was identified in 33.93% (*n* = 19) of the infants diagnosed with hearing loss; that is, 19 of the total number of infants identified with a risk factor and subsequently identified with a hearing loss had a CFA (i.e., 19/43 = 44.2%). Of these infants, unilateral hearing loss was found in 63.16% (*n* = 12), while 36.84% (*n* = 7) were diagnosed with bilateral hearing loss. For those infants who did not have a CFA (66.07%; *n* = 37), a unilateral hearing loss was diagnosed in 18.92% (*n* = 7) and a bilateral hearing loss was diagnosed in 81.08% (*n* = 30).

### State EHDI Program Survey Results

As previously described, a total of 13 EHDI state leaders (22.03%) responded to the EHDI State Leader Survey. Results from this survey are presented in Table 3; the numbering beside each question corresponds to the specific question found in Appendix A.

**Table 3**

*Results from the Early Hearing Detection and Intervention State Leader Survey*

| Survey Questions from EHDI State Leaders | Yes | No |
|---|---|---|
| 3) Does your state mandate newborn hearing screenings in all, or most, birthing facilities? | 84.62% (*n* = 11) | 15.38% (*n* = 2) |
| 5) Are facilities required to document specific risk factors in the state EHDI database? | 46.15% (*n* = 6) | 53.85% (*n* = 7) |
| 8) If a CFA is reported, do you request additional information about the specific CFA from program coordinators? | 23.08% (*n* = 3) | 76.92% (*n* = 10) |
| 9) Are you satisfied with the documentation completed by the facilities in your state regarding the risk factors for hearing loss? | 38.36% (*n* = 5) | 61.54% (*n* = 8) |
| | **Yes, most facilities do** | **Some facilities do, some do not** | **No, most facilities do not** |
| 4) Do facilities in your state submit newborn hearing screening data for all infants born? | 100% (*n* = 13) | 0% (*n* = 0) | 0% (*n* = 0) |
| 6) Do facilities in your state submit risk factor information, as required? | 15.38% (*n* = 2) | 61.54% (*n* = 8) | 23.08% (*n* = 3) |
| 7) If an infant is identified as having a CFA, do facilities in your state record the specific anomaly present? | 38.46% (*n* = 5) | 38.46% (*n* = 5) | 23.08% (*n* = 3) |

*Note.* EHDI = Early Hearing Detection and Intervention; CFA = Craniofacial Anomaly.

Multiple state leaders had suggestions for NBHS programs in response to question #10 "Do you have any suggestions for how to improve the recording, reporting or follow-up process for infants with risk factors for hearing loss, including infants with craniofacial anomalies?" Top responses included (a) involve primary care physicians, (b) increase the education and training for staff involved in NBHS programs, (c) include a system that automatically links data from the hospital/facility's electronic health records system to the state database, and (d) utilize the Centers for Disease Control and Prevention Birth Defects Registry.

### NBHS Program Coordinator Survey Results

A total of 90 newborn hearing screening program coordinators responded to our survey. Appendix C1 demonstrates a breakdown of the professions of the program coordinators, Appendix C2 shows the professions of those conducting the newborn hearing screenings, and the breakdown of states in which the respondents practice is shown in Appendix C3.

When surveyed, 75.56% (*n* = 68) of the program coordinators reported that information regarding risk factors for hearing loss were collected at their facility. This information was collected in multiple ways (Question 9 from survey): by asking hospital staff, 28.89% (*n* = 26); checking infant medical records, 62.22% (*n* = 56); asking the infant's mother case history questions, 60.00% (*n* = 54); and collecting risk factor information by another method, 3.90% (*n* = 3).

In some facilities, information concerning risk factor information was obtained from multiple sources, resulting in the total percentage exceeding 100%.

Table 4 displays responses to other questions (corresponding to the numbered questions in Appendix B) that were posed to the NBHS program coordinators. In addition to the information contained in Table 4, nine of the 90 NBHS coordinators (10%) also reported their newborn hearing screening fail rate for infants with CFAs.

**Table 4**

*Results from the Newborn Hearing Screening Program Coordinator Survey*

| Survey Questions from Program Coordinator Survey | Yes | No |
|---|---|---|
| 6) Does your facility employ audiologists to oversee the program? | 24.44% (*n* = 22) | 75.56% (*n* = 68) |
| 7) Is your staff trained to identify the different risk factors associated with childhood hearing loss? | 83.33% (*n* = 75) | 16.67% (*n* = 15) |
| 8) Is information regarding risk factors for childhood hearing loss collected prior to or following each screening? (Questions 10 through 13 were recorded only if answered 'yes' for question 8) | 75.56% (*n* = 68) | 24.44% (*n* = 22) |
| 10) Does your program record the type of risk factor(s) in the hospital medical records | 77.94% (*n* = 53) | 22.06% (*n* = 15) |
| 11) If an infant is identified as having a risk factor, does your program report the information to the state EHDI program? | 76.47% (*n* = 52) | 23.53% (*n* = 16) |
| 12) If an infant is identified with a CFA, does your program record the specific type of CFA present? | 76.47% (*n* = 52) | 23.53% (*n* = 16) |
| 13) Do you report the specific CFA to the state EHDI program? | 69.23% (*n* = 36) | 30.77% (*n* = 16) |
| 14) Do you feel as though you are getting enough guidance from your state EHDI program on how to document and report risk factors? | 58.89% (*n* = 53) | 41.11% (*n* = 37) |

*Note.* EHDI = Early Hearing Detection and Intervention; CFA = Craniofacial Anomaly.

## Discussion

The overall newborn hearing screening fail rate measured at the two study hospitals (0.74%) was lower than the national fail rate of 4% (ASHA, n.d.). This lower fail rate may be attributed to the two-step screening protocol used at these two hospitals, as described in the Method section. A failed OAE followed by an AABR may reduce false positive rates since the AABR is less sensitive to vernix or debris in the ear canal which is a significant cause of failed screenings (American Speech-Language-Hearing Association [AHSA], n.d.). Without the follow-up AABR, there would be a much higher false-positive rate. Reducing false-positive rates (a) allows infants at risk to be more accurately targeted for follow-up testing, and (b) reduces the wait time and workload for pediatric audiologists by reducing the demand for diagnostic testing.

The hearing screening fail rate for those identified with a CFA (4.4%) is six times higher than the overall fail rate of 0.74% for all newborns. This supports the current JCIH inclusion of CFA on the list of risk factors for hearing loss. In examining the NBHS fail rates for the seven CFA subgroups identified in this study, the fail rates vary greatly, from 0% to 100%. This suggests the need to further delineate the craniofacial risk factor category into subgroups, with follow-up evaluation recommendations based on the specific CFA, rather than CFA category as a whole. For example, an infant with only a preauricular sinus (.25% fail rate) should not receive the same follow-up recommendations as an infant with microtia/atresia (100% fail rate). Currently, the JCIH recommends audiological follow-up/re-evaluation by nine months of age, regardless of the type of CFA present (JCIH, 2019). However, appropriate follow-up evaluations should be recommended based on the specific needs of an infant to ensure that the infant has the best opportunity for early detection and intervention or plan for monitoring in case of a possible progressive or late-onset hearing loss. These findings suggest that JCIH should consider refining their list of risk factors for hearing loss to include separate recommendations for the specific CFAs.

Our screening fail rate of 0.74% for infants with preauricular sinuses and/or tags, in the absence of other physical findings, indicates that routine audiological reevaluation is not warranted for this population. After subdividing the preauricular sinuses/tag group into more specific sub-groups, we determined that infants with either a preauricular sinus or tag, whether unilateral or bilateral, exhibited similar NBHS fail rates (i.e., all less than 1%). A progressive hearing loss would not be expected if these were the only anomalies, however, it is recommended that these anomalies continue to be documented since they can be associated with various syndromes that do have a higher likelihood of hearing loss, such as Trisomy 21, Treacher Collins syndrome, CHARGE Association, Waardenburg Syndrome, Crouzon disease, and so on (Greydanus et al., 2007). Accurate documentation of preauricular sinuses and preauricular tags will assist the medical home in monitoring for any additional signs and/or symptoms associated with such syndromes. Research suggests that skin tags can also be associated with maternal diabetes and may not be related to any ear issues at all; therefore, it is important to obtain a mother's medical history to determine if maternal diabetes is a possible underlying cause for the preauricular tags (Grix et al., 1982; Johnson, Fineman & Opitz, 1982; Sait et al., 2019).

Based on the findings from the two hospitals, of the 56 infants subsequently diagnosed with a hearing loss following their birth screening, infants with a risk factor for hearing loss (43/56) were 3.3 times more likely than infants without a risk factor (13/56) to be diagnosed with a hearing loss following their birth screening. And, of the infants with a risk factor(s) that were identified with a hearing loss, 19 of 43 (44.2%) of them had a CFA. This data provides strong evidence of the need for follow-up evaluations for all infants with a CFA, except for infants with preauricular sinuses and tags due to the low prevalence of NBHS fail rate.

Multiple errors were discovered when comparing the hospital records to OZ. Information pertaining to preauricular sinuses and tags that had been documented appropriately in the hospital records was omitted 154 times in the state database. In addition, one infant's NBHS result was entered incorrectly into the state database as having passed the NBHS when, in fact, the infant had actually failed in both ears. When errors were discovered, they were corrected in the database. The two NBHS programs in this study are managed by audiologists and have a well-developed protocol for documenting and recording risk factors, yet, errors still occurred. Programs without such audiology oversight and thorough protocols could potentially have even more documentation errors.

Based on the survey findings, it appears that a majority of the NBHS program coordinators are documenting and reporting the presence of risk factors in their hospital medical records (see Table 4), however, the majority of the EDHI State Leaders indicated that newborn hearing screening programs were failing to document this information in their state EHDI records (see Table 3). In addition, responses suggest that close to 2/3 of the EHDI NBHS state leaders who responded (61.54%; $n = 8$) were not satisfied with how facilities in their state document and record risk factors for hearing loss. Currently, JCIH has a list of guidelines that each newborn hearing screening program should follow. Because the guidelines are suggestions, not policy, and because each program documents and reports risk factors differently, measuring prevalence figures for hearing loss risk is a challenge. If the findings from this study are indeed reflective of most NBHS programs, then these guidelines should become protocol. Having a universal protocol (including training of staff) would reduce the chances of NBHS programs overlooking risk factors, which would increase appropriate referrals for diagnostic testing and early intervention.

Results from the NBHS Coordinator Survey also indicate that some programs (16.67%; $n = 15$) do not train their newborn hearing screeners to identify the different risk factors for hearing loss. While conducting

the hospital records review for this study, it was noted that pediatricians at the hospitals often neglected to document the presence of preauricular sinuses and tags as well as other ear malformations during the newborn's physical exam, and that our trained hearing screeners were often the ones to identify the presence of such abnormalities. Correctly and accurately identifying infants with risk factors for hearing loss, such as CFAs, helps guide referrals for follow up care. In addition to detecting physical anomalies associated with possible hearing loss, newborn hearing screening staff should also be trained to review hospital records and question parents to identify other possible risk factors for hearing loss, including a family history of childhood hearing loss. The NBHS Program Coordinator survey suggests that roughly a quarter of newborn hearing screening programs do not collect general risk factor information, as recommended by JCIH guidelines. Additionally, 31% of the newborn hearing screening programs do not report CFAs to their state database (see Table 4). This information is vital because without such information, infants with these risk factors may not receive the appropriate follow-up recommendations and referrals.

Responses from this survey also revealed that 75% ($n$ = 68/90) of facilities do not employ an audiologist to oversee the newborn hearing screening program. Having audiologist oversight, as recommended in the new JCIH 2019 guidelines, would increase the probability that staff are trained properly to identify risk factors for hearing loss, and ensure appropriate referrals and recommendations. In addition to having audiologist oversight, it is important for NHBS programs to identify the newborn's medical home, and communicate the hearing screening results with them, if the infant failed their screening (ASHA, n.d.).

Approximately 40% ($n$ = 37) of the NBHS Program Coordinators reported that they did not feel they had received enough guidance from their state EHDI program as to how they should document and record risk factors for hearing loss. EHDI programs are designed to maintain a coordinated, statewide screening and referral system for those infants who do not pass the newborn hearing screening. Without strong guidance from their state EHDI program, infants are at risk for not receiving adequate and appropriate follow-up care (EHDI, 2020). Thus, based on the findings in this study, JCIH should consider developing a universal newborn hearing screening program protocol that all birthing facilities must follow.

**Study Limitations**

The first limitation of this study concerns the number of responses from NBHS program coordinators and state leaders of EDHI programs. It is hard to generalize our conclusions with responses from only 13 state leaders and 90 program coordinators from 18 different states. Increased participation would have allowed for a better representation of how NBHS programs across the country are recording and documenting risk factors for hearing loss.

A second limitation of the study was that the population from the two hospitals lacked geographic diversity. Both hospitals are located in suburban neighborhoods outside of Washington, DC. It is possible that the incidence of CFAs and the screening/documentation protocols may differ from birthing centers in other regions. Therefore, additional data are needed from birthing centers throughout the United States of America before our findings can be generalized.

A third limitation is that the technicians who performed the newborn hearing screenings could have omitted or incorrectly documented risk factors for hearing loss, particularly preauricular sinuses and preauricular tags, as well as other ear malformations. If these risk factors were missed or entered incorrectly, then the hearing screening fail rate relative to the possible risk factors and CFA type could be inaccurately represented.

A fourth limitation of this study was that some of the department paper records for a period of a few months were lost when one of the two study hospitals moved to a new location, while the other was not in possession of some of their records from one of the six years being studied. Therefore, the ability to cross-check information from the department records with OZ was impacted, leading to possible missed identification of infants with risk factors for hearing loss, and potentially influencing the screening fail rate that was obtained.

**Future Directions**

In this study, seven CFA sub-groups were described based on the anomalies present in the infants at the two study hospitals. Development of a standardized list of specific CFAs and their associated NBHS fail rates is needed to maintain consistency across and, in turn, guide NBHS programs in the United States. Because the findings in this study were obtained from only two hospitals, a larger scale study would provide information that could facilitate the development of policies to address the specific hearing needs of different sub-groups of the CFA group.

Having described NBHS fail rates in infants with different CFAs in this paper, future research should focus on describing the degree and type of hearing loss associated with this population. Because specific diagnostic audiology outcomes are not reported in OZ, we were unable to obtain and analyze this information. Collaboration with diagnostic testing facilities would allow access to this data. In addition, it would be beneficial for NBHS programs to document the specific diagnostic outcomes for infants who are diagnosed with a hearing loss in their state database. Such information would be beneficial for the medical home, as well as audiologists, as it would help guide appropriate monitoring and intervention.

Further research should aim to gather information about how risk factor information is being entered and stored into the various databases from a larger proportion of states. In addition, future studies should also examine how NBHS sites are conducting their screening protocols (such as a one-step versus two-step screening approach as used in

the two hospitals in this study), as that may influence the pass/fail rate. The protocol that NBHS sites use as a pass/fail criteria should be looked at as well. Getting all of this information would help determine areas of weakness in NBHS policy. This would help result in solutions that could be universally applied to guide policy making, such as ensuring accurate entry of information (e.g., for CFA and its specific subtypes).

In the current study, 17.41% ($n$ = 51) of infants who failed the NBHS were lost to follow-up. This rate is less than the national average of 31.3% (Subbiah et. al., 2018), presumably due to audiology oversight of the NBHS programs described in this paper. There have been studies that explore reasons for loss to follow-up in newborn hearing screening programs (ASHA, 2008), however, there are no specific protocols in place for guiding these programs. It is important to better understand the underlying causes of loss-to follow-up rates so that JCIH can make recommendations and guidelines for improving service to infants and families.

## Conclusions

Results from this study revealed that children with CFAs were six times more likely to fail their NBHS when compared to the fail rate for all infants at the two participating hospitals.

Audiological follow-up and monitoring is not warranted for infants with preauricular sinuses and tags unless the infant exhibits other features associated with a syndrome that has an associated risk for hearing loss. It is important to document these anomalies in the state EHDI database so that the medical home can monitor for any additional signs and/or symptoms associated with such syndromes and make appropriate referrals to other medical professionals.

Because the NBHS fail rates in this study varied greatly for the different CFAs, further research should be completed to determine if these findings can be replicated. If so, JCIH should consider updating the list of risk factors for hearing loss to delineate the current CFA category into different subgroups. Along with updating the list of risk factors. JCIH should also consider updating the recommendations for each specific CFA, as infants in this risk factor group should be followed based on their specific anomaly rather than the group as a whole.

Results from the two surveys in this study demonstrate that NBHS programs are not recording and documenting risk factor information adequately and consistently. If our findings are representative of the other state EDHI programs that did not respond to the survey, this would suggest a need for changes to the existing NBHS protocol to include programs that train staff to identify and document risk factors for hearing loss. This training would improve the chances that risk factors for hearing loss are being identified and properly documented. Correct documentation of risk factors would also provide medical professionals the information they need to appropriately refer infants for follow-up evaluations, monitoring, and early intervention services so that they have the best opportunity to maximize their potential. This is imperative because, without appropriate and timely referrals, children are more likely to fall behind their hearing peers in communication, cognition, reading, and social-emotional development (JCIH, 2007).

Lastly, it would be advantageous to change the NBHS guidelines to protocols to ensure consistency across all programs. It is crucial that all NBHS programs throughout the country follow the same procedures to improve recommendations for follow-up care in a timely manner.

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## EHDInfo

## Appendix A

## EHDI State Leader Survey

1) If you agree with the informed consent above, please add a signature.
2) Which state do you represent?
3) Does your state mandate newborn hearing screenings in all, or most, birthing facilities?
    a) Yes
    b) No
4) Do facilities in your state submit newborn hearing screening data for infants born at that facility?
    a) Yes, most facilities do
    b) Some facilities do, some facilities do not
    c) No, most facilities do not
5) If an infant is identified with a specific risk factor associated with childhood hearing loss including: family history of hearing loss, NICU stay of greater than 5 days, hyperbilirubinemia with exchange transfusion, ototoxic medications for greater than 5 days, asphyxia or hypoxic ischemic encephalopathy, ECMO, in-utero infections, craniofacial anomalies, syndromes associated with hearing loss, and significant head trauma (re: JCIH 2019 Risk Factor Indicators), are facilities in your state required to document this information in the state EHDI database?
    a) Yes
    b) No
6) Do facilities in your state submit this documentation, as required (re: question 5)?
    a) Yes, most facilities do
    b) Some facilities do, some facilities do not
    c) No, most facilities do not
7) If an infant is identified as having a craniofacial anomaly, do facilities in your state record the specific anomaly present (e.g., cleft lip and palate, preauricular sinus and tags, microtia, atresia, and/or syndromes such as Trisomy 21, Treacher Collins syndrome, CHARGE Association, Crouzon disease, Klippel-Feil syndrome, Goldenhar syndrome, Pierre Robin syndrome, etc.)?
    a) Yes, most facilities do
    b) Some facilities do, some facilities do not
    c) No, most facilities do not
8) If a craniofacial anomaly risk factor is reported, but the specific anomaly is not recorded, do you request additional information from the program coordinator about the specific craniofacial anomaly?
    a) Yes
    b) No
9) Are you satisfied with the documentation completed by the facilities in your state regarding the risk factors for hearing loss?
    a) Yes
    b) No
10) Do you have any suggestions for how to improve the recording, reporting or follow-up process for infants with risk factors for hearing loss, including infants with craniofacial anomalies?
11) Would you like the final results of the study sent to you?
    a) Yes, electronically
    b) Yes, via mail
    c) No
12) If 'yes', where should the results be sent? (email address or mail address)

## Appendix B

### Newborn Hearing Screening Program Coordinator Survey

1) If you agree with the informed consent above, please add a signature.
2) What is your profession?
3) In which state is your facility located?
4) Who performs the newborn hearing screenings? (Mark all that apply)
    a) Audiologists
    b) Nurses/Nurse Techs
    c) Technicians hired specifically to perform the screenings
    d) Other; please specify in the next question
5) If your answer was 'other' from the previous question, please specify.
6) If an audiologist does not perform the hearing screenings, does your facility employ an audiologist to oversee the program?
    a) Yes
    b) No
    c) N/A
7) Is your staff trained to identify the different risk factors associated with childhood hearing loss including: family history of hearing loss, NICU stay of greater than 5 days, hyperbilirubinemia with exchange transfusion, ototoxic medications for greater than 5 days, asphyxia or hypoxic ischemic encephalopathy, ECMO, in-utero infections, craniofacial anomalies, syndromes associated with hearing loss, and significant head trauma (re: JCIH 2019 Risk Factor Indicators)?
    a) Yes
    b) No
8) Is information regarding risk factors for childhood hearing loss collected either prior to or following each newborn hearing screening? (if no skip to question 14).
    a) Yes
    b) No
9) If yes (re: question 8), how is this information collected? (Mark all that apply)
    a) Ask hospital staff about risk factors
    b) Check infant's medical records (e.g., admission reports, lab reports, physician, nurse and/or social worker assessments, etc.)
    c) Ask the mother case history questions at the time of the hearing screening
    d) Other
10) Does your program record the type of risk factor(s) in the hospital medical records (re: question 8)?
    a) Yes
    b) No
11) If an infant is identified as having one of the risk factors (re: question 7), does your program report this information to your state Early Hearing Detection and Identification (EHDI) program?
    a) Yes
    b) No
12) If an infant is identified as having a craniofacial anomaly, does your program record the specific type of craniofacial anomaly present (e.g., cleft lip and palate, preauricular sinus and tags, microtia, atresia, and/or syndromes such as Trisomy 21, Treacher Collins syndrome, CHARGE Association, Crouzon disease, Klippel-Feil syndrome, Goldenhar syndrome, Pierre Robin syndrome, etc.)?
    a) Yes
    b) No

13) If you answered 'yes' for the previous question, do you report the specific craniofacial anomaly to the state EHDI program?

    a) Yes
    b) No

14) Do you feel that you are getting enough guidance from your state EHDI program on how to document and report risk factors, including craniofacial anomalies?

    a) Yes
    b) No

15) Are you able to access the newborn hearing screening pass/fail data for infants with craniofacial anomalies tested at your facility?

    a) Yes
    b) No

16) If you answered 'yes' for the previous question, what is the refer rate for these infants for the period of January 2017 through December 2019?

17) Would you like the final results of the study sent to you?

    a) Yes, electronically
    b) Yes, via mail
    c) No

18) If 'yes', where should the results be sent? (email address or mail address)

## Appendix C

**Table C1**
*Profession of Participating Newborn Hearing Screening (NBHS) Program Coordinators*

| Profession | Total Percentage (*n*) |
|---|---|
| Nurse | 54.44% (49) |
| Audiologist | 37.66% (29) |
| NBHS Coordinator | 4.44% (4) |
| Nurse Practitioner | 1.11% (1) |
| Operations Coordinator | 1.11% (1) |
| Perinatal Safety Specialist | 1.11% (1) |
| Unit Secretary | 1.11% (1) |
| Practice Manager | 1.11% (1) |
| Administrative Assistant | 1.11% (1) |
| Hearing Technician | 1.11% (1) |
| Did Not Answer | 1.11% (1) |

**Table C2**
*Profession of Those Conducting NBHS*

| **Profession**[a] | **Total Facilities** |
|---|---|
| Audiologists | 20/90 facilities |
| Nurses | 61/90 facilities |
| Technicians (hired specifically for the hearing screening) | 25/90 facilities |
| Others: Physician assistants, nurse practitioners, trained volunteers and student interns | 4/90 facilities |

*Note.* NBHS= Newborn Hearing Screening
[a]Some facilities employ multiple professionals to perform the screenings.

**Table C3**
*States Represented in the Newborn Hearing Screening Program Coordinator Survey*

| **Profession** | **Total Percentage (*n*)** |
|---|---|
| Arkansas | 2.22% (2) |
| California | 1.11% (1) |
| District of Columbia | 2.22% (2) |
| Illinois | 1.11% (1) |
| Louisiana | 1.11% (1) |
| Maryland | 12.22% (11) |
| Michigan | 36.67% (33) |
| Missouri | 1.11% (1) |
| North Carolina | 1.11% (1) |
| Nebraska | 14.44% (13) |
| New Jersey | 6.67% (6) |
| New York | 3.33% (3) |
| Nevada | 1.11% (1) |
| Ohio | 5.55% (5) |
| Oregon | 1.11% (1) |
| South Carolina | 1.11% (1) |
| Tennessee | 1.11% (1) |
| Virginia | 5.55% (5) |
| Blank Responses | 1.11% (1) |

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# Usability and Feasibility of a Spoken Language Outcome Monitoring Procedure in a Canadian Early Hearing Detection and Intervention Program: Results of a 1-Year Pilot

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## Abstract

**Purpose**: Best practice recommendations for Early Hearing Detection and Intervention (EHDI) programs include routine spoken language outcome monitoring. The present article reports on pilot data that evaluated the usability and feasibility of a spoken language outcome monitoring procedure developed for Ontario's Infant Hearing Program (IHP). This procedure included both Program-level monitoring using omnibus language tests from birth to 6 years of age and individual vulnerability monitoring of key domains of spoken language known to be at risk in children who are deaf or hard of hearing.

**Methodology**: Speech-language pathologists (SLPs) in the IHP piloted the new procedures for one year and provided feedback on the procedure through surveys at the end of the pilot.

**Results**: Data was suggestive that the Program-level procedure might be sensitive to change over time and known predictors of spoken language outcomes. Some, but not all, Program-level test scores were predicted by the presence of additional developmental factors. None of the test scores were significantly predicted by severity of hearing loss. Depending on the tests and scores used, some aspects of the Program-level procedure were sensitive to change over time. There was insufficient evidence to support individual vulnerability monitoring. SLPs reported significant concerns about the time involved in implementing both procedures.

**Conclusions**: This article describes preliminary evidence suggesting that the Program-level procedure might be feasible to implement and useful for evaluating EHDI programs. Future evaluations are needed to determine whether the procedure can be accurately implemented to scale in the IHP, and whether the data that results from the procedure can meaningfully inform stakeholders' decision-making.

**Keywords:** Spoken language outcome monitoring; Program evaluation

**Acronyms:** BEPTA = better-ear pure-tone average; CASL-2 = Comprehensive Assessment of Language Fundamentals, 2nd ed.; CELF-P2 = Clinical Evaluation of Language Fundamentals; dB HL = decibels Hearing Loss; DHH = deaf or hard of hearing; EHDI = Early Hearing Detection and Intervention; EOWPVT = Expressive One Word Picture Vocabulary Test; GFTA-3 = Goldman-Fristoe Test of Articulation, 3rd ed.; MBCDI-2 = MacArthur-Bates Communicative Development Inventories, 2nd ed.; IHP = Infant Hearing Program; IVT = Individual Vulnerability Test; Preschool – 2nd ed.; JCIH = Joint Committee on Infant Hearing; OMRU = Revised Ottawa Model of Research Use; PLS = Preschool Language Scale; PTA = pure-tone average; SII = Speech Intelligibility Index; SLP = speech-language pathologist

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Best practice recommendations for Early Hearing Detection and Intervention (EHDI) programs include routine spoken language outcome monitoring for infants who are born deaf or hard of hearing (DHH) and are learning a spoken language (Joint Committee on Infant Hearing [JCIH], 2007, 2013; Moeller et al., 2013). Routine spoken language outcome monitoring is intended to provide various stakeholders (i.e., administrators, clinicians, educators, families) with regular feedback on a child's development, and to support program evaluation and intervention planning. Stakeholders should expect that children who are DHH will progress toward age-appropriate spoken language outcomes regardless of the severity or type of hearing loss because hearing loss is not a language learning disorder (Moeller & Tomblin, 2015). Research has repeatedly demonstrated that when infants who are born DHH have adequate access to spoken language they perform, as a group, within age-expectations, but statistically below their

peers, on norm-referenced tests of overall spoken language ability (Ching et al., 2017; JCIH, 2019; Tomblin et al., 2015).

Despite the clear recommendations and rationale for spoken language outcome monitoring, there is limited evidence to support best practice recommendations for EHDI programs, and the clinical barriers and facilitators to implementing spoken language outcome monitoring procedures are not well understood. Daub and Oram Cardy (2021) provided the first report of the process used by one EHDI program, the Ontario Infant Hearing Program (IHP), to develop a standard spoken language outcome monitoring procedure. The IHP was launched in 2001 and provides comprehensive EHDI programming guided by JCIH best practice recommendations (JCIH, 2007, 2013, 2019). In the Canadian context, Ontario is one of the provinces/territories that continually provides adequate EHDI services through its IHP (Canadian Infant Task Force, 2014, 2019), including universal newborn hearing screening as well as intervention services to over 11,000 children who are DHH across the province annually. The IHP is a publicly funded program managed by the Ontario Ministry of Children, Community and Social Services. Children enrolled in the IHP access speech-language pathology supports from a related Ministry program—the Preschool Speech and Language Program, which is a publicly funded speech-language pathology program that serves all preschoolers in Ontario with speech, language, and communication needs. The Preschool Speech and Language Program employs more than 400 speech-language pathologists (SLPs) and provides services to more than 60,000 children with a wide range of needs (i.e., SLPs do not exclusively serve children who are DHH) each year. Preschool Speech and Language Program services are delivered in various contexts including designated clinics, childcare centers, in children's homes, and at fly-in clinics for families living in remote areas. The IHP previously tasked SLPs to use the Preschool Language Scale, 4th edition (Zimmerman et al., 2002) to monitor spoken language outcomes. Under this procedure, implementation between regions varied and SLPs tended to collect outcome data for children receiving IHP services *only* if the child was actively receiving Preschool Speech and Language Program services, that is, the outcome data were mostly focused on those children who were DHH for whom there were concerns about their spoken language development. When the Preschool Language Scale, 4th edition fell out of print, the IHP contracted the authors to support the development of a new procedure.

In developing a new spoken language outcome monitoring procedure, the authors and the IHP prioritized identifying a process for modelling growth in spoken language using norm-referenced tests that had previously been used in the peer-reviewed literature to evaluate children who are DHH. Based on the results of a scoping review, critical appraisal, and consultation with IHP managers and SLPs, a two-tiered assessment approach was recommended (Daub & Oram Cardy, 2021). In Tier 1, it was recommended that SLPs measure spoken language every six months from birth to 3 years, and annually thereafter (JCIH, 2007, 2013). Between birth and 1 year 6 months, SLPs were advised to use the MacArthur-Bates Communicative Development Inventories, 2nd edition (MBCDI-2; Fenson et al., 2007) Words and Gestures form, and from 1 year 7 months to 6 years, the Preschool Language Scale, 5th edition (PLS-5; Zimmerman et al., 2011) was recommended. The PLS-5 was selected based on its suitability for children within IHP age eligibility (up to 6 years), its psychometric appropriateness, and its Growth Scale Values, which are more sensitive to measuring change in language abilities than traditional norm-referenced scores (i.e., standard scores; Daub et al., 2017). Initial recommendations included using the PLS-5 right from birth, but concerns voiced by various stakeholders about the long administration time, lower diagnostic accuracy, and limited clinical value of the PLS-5 for children under 18 months of age, motivated the recommendation for use of the MBCDI-2 at the earliest ages. The purpose of the Tier 1 assessment was to collect data on children's spoken language outcomes that could be entered into a provincial database and used to facilitate program evaluation and planning (see Figures 1 & 2). Planned analyses for program evaluation included fitting growth curves of children's spoken language development and identifying factors predictive of growth in spoken language that could inform IHP curriculum development.

**Figure 1**

*Overall Outcome Monitoring Process (from Daub & Oram Cardy, 2021)*

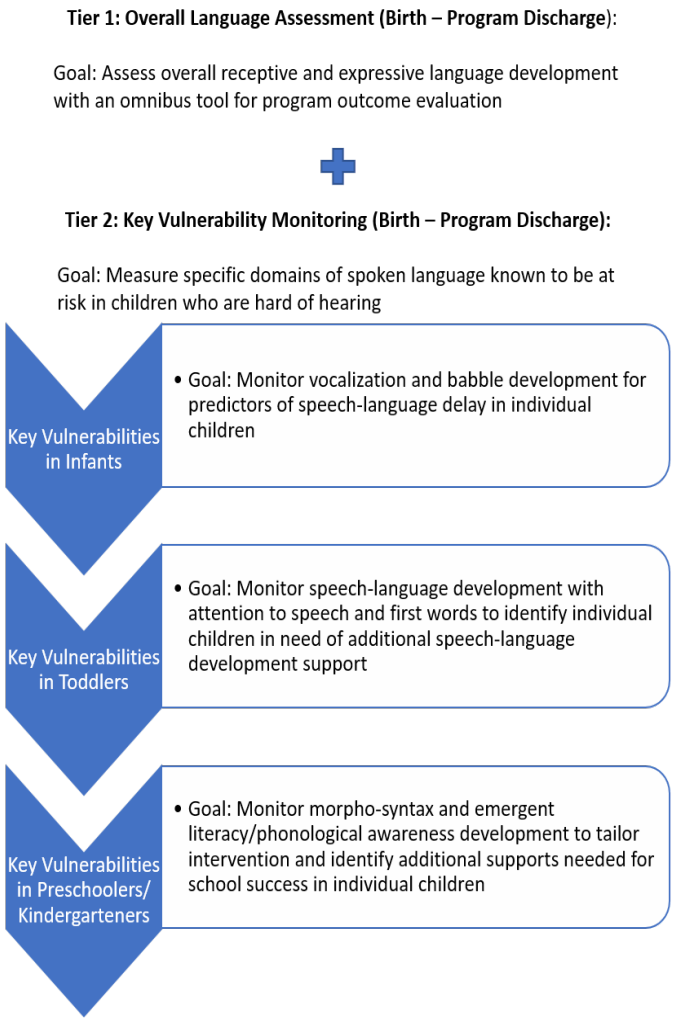


**Figure 2**

*Tests Used in Outcome Monitoring Process (from Daub & Oram Cardy, 2021)*

<table>
<tr><th></th><th>Program Monitoring</th><th colspan="3">Individual Vulnerability Testing</th></tr>
<tr><th>Age (years)</th><th></th><th>Vocalization/Babbling/ Articulation/Phonology</th><th>Words/Grammar</th><th>Emergent literacy/ Phonological awareness</th></tr>
<tr><td>0.5–1</td><td rowspan="2">MBCDI-2 Words & Gestures* (Scores for: Words Understood, Words Produced, Phrases Understood, and Gestures Produced)</td><td rowspan="3">Vocal development tests require further evaluation</td><td rowspan="2">(MBCDI-2 Words & Gestures)</td><td rowspan="4"></td></tr>
<tr><td>1–1.5</td></tr>
<tr><td>1.5–2</td><td rowspan="5">PLS-5 (Scores for: Auditory Comprehension & Expressive Communication)</td><td rowspan="2">MBCDI-2 Words & Sentences <u>or</u> EOWPVT-4</td></tr>
<tr><td>2–2.5</td></tr>
<tr><td>2.5–3</td><td rowspan="2">GFTA-3 (Scores for Sounds-in-Words)</td></tr>
<tr><td>3–4</td><td rowspan="3">CELF-P2 (Scores for Word Structure) <u>or</u> CASL-2 (Scores for Grammatical Morphemes)</td></tr>
<tr><td>4–5</td><td rowspan="2"></td><td rowspan="2">CELF-P2 (Scores for Pre-literacy Rating Scale) <u>or</u> CELF-P2 (Scores for Phonological Awareness Subtest)</td></tr>
<tr><td>5–6</td></tr>
</table>

*Note.* GFTA-3 = Goldman-Fristoe Test of Articulation, 3rd ed.; CASL-2 = Comprehensive Assessment of Spoken Language; CELF-P2 = Clinical Evaluation of Language Fundamentals, Preschool – 2nd ed.; MBCDI = MacArthur-Bates Communicative Development Inventories; PLS = Preschool Language Scale; EOWPVT = Expressive One Word Picture Vocabulary Test.

In Tier 2, it was recommended that SLPs assess key spoken language domains for which children who are DHH are at ongoing risk due to limitations with auditory access (see Figures 1 & 2). This tier was recommended as an improvement to the existing common practice whereby children were discharged from services when SLPs and families were not concerned about spoken language development. Tier 2 monitoring was recommended because permanent childhood hearing loss imposes lifelong limitations to auditory access, and it is therefore possible that delays in spoken language could still emerge despite overall age appropriate spoken language development being measured in a Tier 1 assessment. Tier 2 assessment recommendations included a list of tests SLPs could select from to measure each of three key individual vulnerabilities (see Figure 2). It was recommended that SLPs track key vulnerabilities at the same intervals as overall spoken language (every six months from birth to 3 years of age and annually afterward). For SLPs, the purpose of Tier 2 was to provide them with clinically useful information about a child's developmental status, facilitate intervention planning, and clarify the links between delays in different domains of spoken language development and overall spoken language performance. For the IHP, the purpose was to track key vulnerabilities to allow the program to model the development of three language domains for children who are DHH, and document agreement in disorder classification between omnibus spoken language assessments (Tier 1 MBCDI or PLS-5) and assessments specific to individual language domains (Tier 2 assessments).

Tier 1 and 2 recommendations were made based on the best available empirical and clinical evidence (Daub & Oram Cardy, 2021). However, evidence was still needed to confirm that these tiers resulted in usable data and were feasible to implement in the real-world. Although each of the tests included in the procedure were selected based on their alignment between psychometric properties and the IHP's program evaluation goals (Daub & Oram Cardy, 2021), it is possible that the data may not be sufficient when collected in practice. Whether the data can address the questions they are intended to answer depends on SLPs' ability to administer the procedure and enter the data into provincial databases. It must also be possible to extract the relevant data from the provincial database and prepare it for analysis.

This paper reports data from two pilot studies that were initiated to evaluate the usability and feasibility of both assessment tiers prior to program-wide implementation as a proof of concept that the data collected and entered conformed to our theoretical expectations when developing the procedure. These pilot projects were part of a series of program evaluation projects initiated by the IHP for which Western University provided methodological and statistical support. In Pilot Study 1, SLPs working in the IHP implemented the Tier 1 procedure for a one-year period and provided feedback through surveys on their perceptions of the procedure at the end of the pilot. In Pilot Study 2, a subset of SLPs from Pilot Study 1 simultaneously implemented the Tier 2 procedure and provided feedback at the end of the pilot. The current study addressed the following questions for the Tier 1 pilot:

1) Is the procedure sensitive to known predictors of spoken language outcome?
2) Is the procedure sensitive to change over time?
3) What are the barriers that SLPs experienced in implementing the procedure?
4) What modifications can be made to the procedure to improve its clinical feasibility?

The Tier 2 testing procedure was developed with the intention to provide information about key domains of spoken language to inform service provision for individual children. This study addressed the following questions for the Tier 2 pilot:

1) Does the procedure provide unique information beyond the Tier 1 procedure?
2) Do SLPs believe that the procedure is clinically useful?
3) What barriers did SLPs experience in implementing the procedure?
4) What modifications would improve the procedure's clinical feasibility?

## Pilot Study 1: Tier 1 Program-level Outcome Monitoring

### Method

#### *Ethical Approval*

Both pilots were Program Evaluation and Quality Improvement projects with the Ontario Ministry of Children, Community and Social Services. These projects were reviewed by the Western University Research Ethics Board (REB). The REB considered the projects not to be research as described in the Canadian Tri-Council Policy Statement V.2 (Research Exempt from REB Review, Article 2.4) and therefore they were not considered to fall under the purview of the REB.

#### *Procedure*

Prior to implementing the pilot program, participating SLPs ($N = 56$) from 11 regions in Ontario completed an online learning module designed to introduce and support implementation of the new spoken language outcome monitoring procedures (see Cunningham et al., 2021). SLPs implemented the recommended procedures in practice, routinely assessing the spoken language of all IHP children on their caseloads for one year (data collection completed in July 2019). At each assessment point, SLPs entered de-identified data into a secure REDCap database on a local server including test scores, age, and unique IHP identification number. SLPs also reported additional factors they believed influenced the child's scores (e.g., a comorbid diagnosis) or performance (e.g., distractibility). The first author (O.D.) then used the data in the REDCap database for analysis and checked all test scores for typographical or scoring errors by comparing the test scores SLPs entered into the database against the child's age using the examiner's manuals. Unique identification numbers were used to extract additional clinical information (i.e., child's sex, audiological variables) from the IHP database. The first author (O.D.) then used each child's identification number to link the demographic and audiological data with the pilot data. The final dataset was used to assess whether the procedures were sensitive to change over time and to predictors of spoken language outcomes. Note that the IHP database was managed by the IHP for clinical, not research, purposes and we did not have access to complete clinical charts or all variables that may impact children's language. Similarly, we did not have access to SLPs' clinical charts, and so we were unable to identify whether the data entered in the REDCap database represented *all* children on SLPs' caseloads who were eligible to be assessed with the procedure. These data, therefore, represented an opportunity to broadly investigate whether the outcome monitoring procedure conformed to our expectations.

To identify barriers to implementation and modifications required to improve feasibility, SLPs completed surveys designed to evaluate potential barriers to future implementation of the procedures at the end of the one-year pilot. Surveys were designed based on the Revised Ottawa Model of Research Use (OMRU; Graham &

Logan, 2004) and modelled after surveys used in the design of procedures to monitor auditory based outcomes for pediatric audiologists (Moodie et al., 2011). The OMRU is a framework to guide implementation of new innovations (in our case, spoken language outcome monitoring procedures) including assessing influential barriers and supports (i.e., features of the innovation, potential adopters, and the practice environment) related to implementing the innovation. Once implementation has begun, the OMRU recommends ongoing monitoring to generate evidence of the innovation's adoption and impact. Our feasibility analysis is positioned within the *assess* stage of the OMRU and our surveys were designed to understand factors about the innovation, potential adopters, and practice environment that may influence future implementation efforts.

### *Participants Assessed in Pilot Study 1*

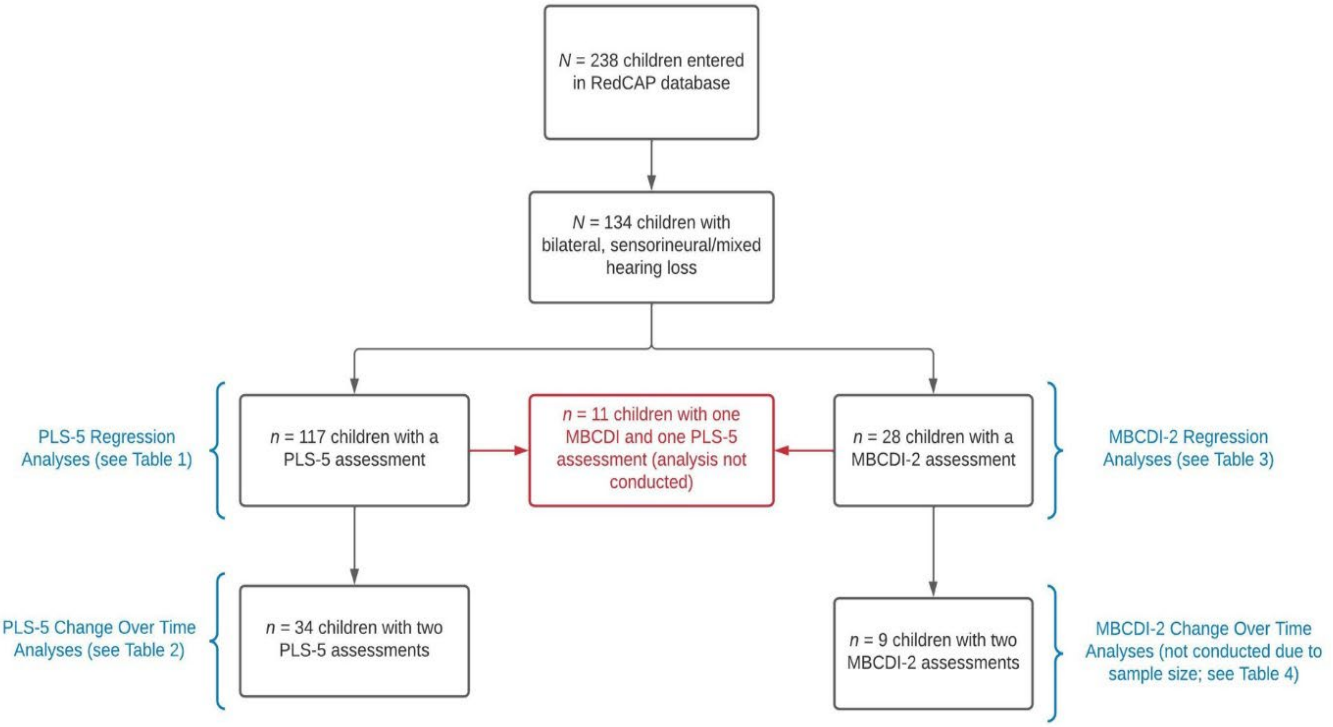
At the end of the pilot, data were available in REDCap for 238 different children. These children had a range of audiological profiles, including unilateral or bilateral, conductive or sensorineural, and ranging from mild to severe in degree. During the pilot study, SLPs were instructed to assess all children enrolled in the IHP at the recommended age ranges and enter the assessment results into the REDCap database. However, we did not have access to the caseload records of the pilot sites, and therefore cannot confirm whether there were children who were DHH for whom SLPs should have conducted an assessment but did not. Therefore, it was unknown whether the children in our database represent all children for whom the procedure should have been used or whether there are groups of children for whom the procedures were not administered.

We can confirm one instance whereby the identification number reported by the SLP could not be linked to an identification number in the program database, and this child was excluded from our analyses. Three children were removed from all analyses for having normal hearing thresholds. In these cases, children were previously under investigation for hearing loss (and so they were assessed by SLPs) but follow-up assessment confirmed normal hearing thresholds.

The analyses for this pilot are based on the entire subset of 134 children who had bilateral sensorineural or mixed hearing losses (see Figure 3). Although the purpose of the Tier 1 outcome monitoring procedure is to document outcomes for all children who receive services from the IHP, very little is known about how unilateral (José et al., 2014) and conductive losses influence spoken language development. There are some data suggesting that children with unilateral losses have poorer spoken language and academic outcomes than children with typical hearing thresholds, although children in these studies tended to be identified later than is the case in the IHP (Fitzpatrick et al., 2019). Similarly, children with conductive losses have a healthy cochlea and their outcomes could reasonably be expected to be different from children with sensorineural losses. Because the primary purpose of this pilot study was to determine whether data generated by the Tier 1 procedure was sensitive to known predictors of spoken language outcomes, we elected to focus our analyses on the groups of children for whom there was the most peer-reviewed data to contextualize our outcomes—children with bilateral sensorineural or mixed hearing losses.

**Figure 3**

*Children from the Overall Sample Included in Pilot Study 1 Analyses*




*Note.* MBCDI = MacArthur-Bates Communicative Development Inventories; PLS = Preschool Language Scale.

After excluding children with normal hearing thresholds, and unilateral and conductive losses, data were available for 117 children with at least one assessment with the PLS-5 (see Table 1) and 34 had data for two assessments (see Table 2). Twenty-eight children had data for at least one assessment with the MBCDI-2 (see Table 3) and nine had data for two assessments (see Table 4). Two children with PLS-5 assessments were fitted with cochlear implants, and 98 were fitted with hearing aids in at least one ear at the time of their language assessment (87 were binaurally fitted, 11 were monaurally fitted). One child with a MBCDI-2 assessment was fitted with a cochlear implant and 19 were fitted with a hearing aid in at least one ear (17 were binaurally fitted, two were monaurally fitted). As a group, children's hearing aids were well-fitted (see Supplemental Materials 1-4 for a comparison of aided Speech Intelligibility Index (SII) to Better Ear Pure Tone Average [BEPTA] to norms reported in Moodie et al., 2017). The decision to fit an ear with a hearing aid is complex and influenced by various factors including the configuration and severity of the child's hearing loss in each ear, and the family's readiness for amplification. Therefore, it is not the case that children in our sample who were not fitted with hearing aids in one, or both ears, *should* have been fitted. Rather, children's audiological profiles at the time of language assessment reflect the family-centered, clinical decision-making of the child's team at the time of their language assessment.

**Table 1**

*Demographics of Children with Data for One PLS-5 Assessment*

| | ***Children without Additional Factors (n = 75)*** | | ***Children with Additional Factors (n = 41)*** | |
|---|---|---|---|---|
| Variable | *M* (range) | SD | *M* (range) | SD |
| Age (months) | 38.3 (19–71) | 7 | 35.2 (19–71) | 12.24 |
| BEPTA (dB HL) | 53.2 (17.5–107.5) | 23.2 | 56.49 (26.25–113.33) | 22 |
| Better Ear SII (Conversational Speech) | 72.5 (5–95) | 22.59 | 68.45 (2–95) | 23.88 |
| Better Ear SII (Quiet Speech) | 64.29 (2–97) | 24.83 | 66.33 (11–96) | 22.1 |
| Expressive Communication (Standard Score) | 100.92 (50–150) | 20.5 | 79.67 (53–118) | 13.73 |
| Auditory Comprehension (Standard Score) | 98.96 (50–137) | 19.81 | 74.49 (50–104) | 16.6 |

*Note*. BEPTA = better-ear pure-tone average; dB HL = decibels Hearing Loss; PLS = Preschool Language Scale; SII = Speech Intelligibility Index, the proportion of the speech signal that is audible when the child is wearing their amplification.

**Table 2**

*Demographics of Children with Data for Two PLS-5 Assessments*

| | ***Children without Additional Factors (n = 24)*** | | ***Children with Additional Factors (n = 9)*** | |
|---|---|---|---|---|
| Variable | *M* (range) | SD | *M* (range) | SD |
| Age at first PLS-5 (months) | 26.96 (19–38) | 6.17 | 28.56 (19–40) | 6.1 |
| Age at second PLS-5 (months) | 34.76 (24–48) | 7.04 | 34.89 (26–45) | 5.8 |
| BEPTA (dB HL) | 55.55 (20–107.5) | 25.04 | 67.27 (35–113.33) | 27.03 |
| Better Ear SII (Conversational Speech) | 69.61 (5–95) | 26.13 | 58.86 (2 – 86) | 32.57 |
| Better Ear SII (Quiet Speech) | 61.11 (13–97) | 26.71 | 60.33 (11–83) | 29.79 |
| First Expressive Communication (Standard Score) | 103 (73–123) | 14.07 | 79.63 (68–88) | 7.09 |
| Second Expressive Communication (Standard Score) | 101.9 (74–122) | 14.95 | 82.75 (72–95) | 8.68 |
| First Expressive Communication (Growth Scale Value) | 382.25 (297–448) | 36.99 | 328.13 (297–348) | 17.73 |
| Second Expressive Communication (Growth Scale Value) | 412.5 (314–507) | 43.80 | 362.13 (319–390) | 28.22 |
| First Auditory Comprehension (Standard Score) | 104.35 (81–127) | 13.94 | 70.88 (53–100) | 16.65 |
| Second Auditory Comprehension (Standard Score) | 103 (65–123) | 14.72 | 71.75 (54–95) | 14.79 |
| First Auditory Comprehension (Growth Scale Value) | 394.45 (324–450) | 34.27 | 334.13 (261–392) | 41.85 |
| Second Auditory Comprehension (Growth Scale Value) | 426.74 (352–504) | 36.1 | 360.63 (304–414) | 43.39 |

*Note.* BEPTA = better-ear pure-tone average; dB HL = decibels Hearing Loss; PLS = Preschool Language Scale; SII = Speech Intelligibility Index, the proportion of the speech signal that is audible when the child is wearing their amplification.

**Table 3**

*Demographics of Children with Data for One MBCDI Assessment*

| | ***Children without Additional Factors (n = 19)*** | | ***Children with Additional Factors (n = 9)*** | |
|---|---|---|---|---|
| Variable | *M* (range) | SD | *M* (range) | SD |
| Age (months) | 12.37 (8–18) | 3.14 | 12.56 (9–18) | 2.5 |
| BEPTA (dB HL) | 56.23 (31.25–95) | 19.77 | 51.74 (25–95) | 24.83 |
| Better Ear SII (Conversational Speech) | 72 (21–91) | 22.77 | 60.5 (25–86) | 26.29 |
| Better Ear SII (Quiet Speech) | 64.17 (6–88) | 27.31 | 67 (56–76) | 10.15 |
| Phrases Understood (Percentile Rank) | 37.5 (10–75) | 19.8 | 19.11 (< 5–65) | 19.89 |
| Words Produced (Percentile Rank) | 32.78 (< 5–85) | 29.67 | 21.67 (< 5–45) | 16.96 |
| Words Understood (Percentile Rank) | 42 (10–99) | 25.85 | 20.22 (< 5–45) | 19.26 |
| Gestures (Percentile Rank) | 39.67 (< 5–80) | 22.61 | 12.33 (< 5–45) | 13.32 |

*Note*. BEPTA = better-ear pure-tone average; dB HL = decibels Hearing Loss; MBCDI = MacArthur-Bates Communicative Development Inventories; SII = Speech Intelligibility Index, the proportion of the speech signal that is audible when the child is wearing their amplification.

### *Analyses*

**Data Usability.** There were two primary analytic purposes of the Tier 1 pilot. The first was to evaluate whether the Program-level scores (PLS-5 and MBCDI-2) were sensitive to predictors known to influence spoken language outcome in children who are DHH. The second analytic purpose related to data usability was to evaluate whether Program-level scores were sensitive to change for children who had a second assessment using the same test.

The predictors we evaluated for our first purpose included the severity of hearing loss and the presence/absence of additional factors influencing performance. Additional factors were broadly defined as any factor that a SLP believed influenced the child's performance on the test, above and beyond their hearing loss. These additional factors included comorbid diagnoses, social factors such as inconsistent hearing aid use, or children's inability (or unwillingness) to engage in testing. Given the relatively

**Table 4**

*Demographics of Children with Data for Two MBCDI Assessments*

| | ***Children without Additional Factors (n = 5)*** | | ***Children with Additional Factors (n = 4)*** | |
|---|---|---|---|---|
| Variable | *M* (range) | SD | *M* (range) | SD |
| Age at first MBCDI (months) | 10.5 (8–14) | 2.65 | 11.6 (9–14) | 1.95 |
| Age at second MBCDI (months) | 15.25 (14–17) | 1.26 | 17.4 (16–19) | 1.14 |
| BEPTA (dB HL) | 54.5 (31.25–90) | 25.6 | 52.33 (31.67–95) | 27.48 |
| Better Ear SII (Conversational Speech) | 78 (71–85) | 7 | 41.5 (25–58) | 23.33 |
| Better Ear SII (Quiet Speech) | 75.67 (64–82) | 10.15 | 69 (69–69) | NA |
| First Phrases Understood (Percentile Rank) | 28.75 (14–45) | 13.77 | 23.6 (< 5–65) | 24.99 |
| Second Phrases Understood (Percentile Rank) | 28.75 (15–40) | 11.09 | 17.5 (5–40) | 15.55 |
| First Words Produced (Percentile Rank) | 50 (5–80) | 31.88 | 12 (5–30) | 10.95 |
| Second Words Produced (Percentile Rank) | 30 (25–40) | 7.01 | 13.75 (5–30) | 11.09 |
| First Words Understood (Percentile Rank) | 43.75 (20–55) | 16.01 | 23.6 (< 5–45) | 20.6 |
| Second Words Understood (Percentile Rank) | 30 (10–50) | 16.83 | 8 (< 5–20) | 8 |
| First Gestures (Percentile Rank) | 36.25 (5–60) | 22.23 | 16.6 (< 5–45) | 16.8 |
| Second Gestures (Percentile Rank) | 37.5 (15–50) | 15.55 | 13.5 (< 5–20) | 7.89 |

*Note*. BEPTA = better-ear pure-tone average; dB HL = decibels Hearing Loss; MBCDI = MacArthur-Bates Communicative Development Inventories; SII = Speech Intelligibility Index, the proportion of the speech signal that is audible when the child is wearing their amplification.

large number of possible factors that could influence language development, it was beyond the scope of the present study to identify which additional factors were differentially associated with language development (e.g., Cupples et al., 2014). For our purposes, we used the presence of additional factors as a coarse indicator that the procedures could be sensitive to additional factors if implementation was scaled up across the province. Follow-up work exploring children's outcomes at the population level would better differentiate the impact of various factors on children's spoken language outcomes.

Prior to analysis, the first author (O.D.) checked the scores recorded in REDCap against the scores reported in the examiner's manuals for the child's recorded chronological age. This process was done to ensure that raw scores were consistently converted to normative scores amongst clinicians, as there is some latitude (particularly with the MBCDI-2) with which to assign percentile ranks. O.D. also checked each child's thresholds from their closest audiology appointment to (but not later than) the Program-level assessment in the IHP database. This was done to determine the child's audiological profile at the time of the language assessment.

***Tests' Associations with Predictors***. We conducted two direct entry linear regression models to evaluate each test's association with two independent variables (a) severity of hearing loss and (b) the presence/absence of additional factors that SLPs believed may have influenced a child's performance. Severity of hearing loss was conceptualized as the child's Better Ear Pure Tone Average (BEPTA). The presence/absence of additional factors was represented using a dichotomous coding of whether SLPs indicated that they believed factors may have influenced a child's performance as independent variables. Within the IHP, audiometric thresholds must be obtained at 500, 2000, and 4000 Hz in each ear (1000 Hz is discretional; Bagatto et al., 2020; Scollie et al., 2019). Audiologists will attempt to measure all four frequencies in each ear at each assessment, although this may not be possible for various reasons (e.g., child's engagement in testing). Each model's conformity to linear regression assumptions was evaluated using the Global Validation of Linear Models Assumptions, v. 1.0.0.3 in R-Studio (Pena & Slate, 2019).

The first regression model evaluated the association between standard scores for the PLS-5 subtests (auditory comprehension and expressive communication) and the independent variables. The first regression was done using data for a subsample of children who had a PLS-5 assessment. The second regression model evaluated the association between percentile ranks for the MBCDI-2 subtests and the independent variables.

***Tests' Sensitivity to Change Over Time.*** Sensitivity to change over time was coarsely evaluated using paired *t*-tests to compare scores between the first and second assessment intervals. For PLS-5 scores, change was evaluated separately using standard scores and growth scale values, as it has been demonstrated that growth scale values are more sensitive to gains in skills over short intervals (Daub et al., 2017). For the MBCDI-2, change was evaluated using percentile ranks as the test does not report standard scores or growth scale values. We corrected for multiple comparisons using Bonferroni's correction.

**Procedure Feasibility**. Surveys (see Supplemental Material 5) were designed to identify potential barriers and facilitators to successful implementation. Surveys included 75 questions and asked SLPs to rate their perceptions of the new procedures; their knowledge, skills, and abilities in using the recommended tools; and their opinions on implementation materials and suggestions to improve them. Questions were either in yes/no format or used 5-point Likert scales to measure the strength of SLPs' agreement with statements. Results are reported descriptively.

## Results

### *Data Usability*

**Tests' Associations with Predictors.** All regression analyses met assumptions of normality, independence, homoscedasticity, and linearity with the exception of the PLS-5 Expressive Communication models, which were significantly heteroscedastic. PLS-5 standard scores for both the Auditory Comprehension and Expressive Communication scales were negatively predicted by the presence of additional factors but not BEPTA [auditory comprehension: $F(2, 104) = 21.87$, $p < 0.001$; expressive communication: $F(2,100) = 16.8$, $p < 0.001$; see Table 5]. The combination of BEPTA and the presence of additional factors accounted for 28% and 24% of the variance in children's Auditory Comprehension and Expressive Communication standard scores, respectively (as indicated by R-squared). In both cases, the presence of additional factors was the only significant predictor.

**Table 5**

*Association Between PLS-5 Standard Scores and Predictors*

| | PLS-5 Standard Score at First Assessment | | | |
|---|---|---|---|---|
| | Auditory Comprehension | | Expressive Communication | |
| Predictor | $R^2$(adj) | b | $R^2$(adj) | b |
| Model | 0.28* | | 0.235* | |
| Better Ear Pure Tone Average (dB HL) | | 0.263 | | -0.1 |
| Presence of additional factors affecting outcome | | -24.13* | | -20.79* |

*Note*. dB HL = decibels Hearing Loss; PLS = Preschool Language Scale.

*$p < 0.001$

The model of the influence of BEPTA and the presence of additional factors on gestures was the only significant model of the MBCDI-2 subtests, $F(2,24) = 5.32$, $p < 0.05$, [phrases understood: $F(2,24) = 2.57$, $p > 0.05$: words produced: $F(2,24) = 0.77$, $p > 0.05$: words understood: $F(2,23) = 2.45$, $p > 0.05$; see Table 6]. Regardless of significance testing, the combination of BEPTA and the presence of additional factors did not explain a large proportion of variance for the phrases understood (11% of variance explained), words produced (-2% of variance explained, indicating exceptionally poor model fit) or words understood (10% of variance explained). The combination of BEPTA and the presence of additional factors accounted for 25% of the variance in children's percentile ranks on the Gestures Produced subtest, although the presence of additional factors was the only significant predictor.

**Table 6**

*Association Between MBCDI-2 Percentile Ranks and Predictors*

| | MBCDI-2 Percentile Rank at First Assessment | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| | Phrases Understood | | Words Produced | | Words Understood | | Gestures | |
| Predictor | $R^2$(adj) | b | $R^2$(adj) | b | $R^2$(adj) | b | $R^2$(adj) | b |
| Model | 0.11 | | -0.02 | | 0.10 | | 0.25* | |
| Better Ear Pure Tone Average (dB HL) | | 0.07 | | -0.17 | | -0.09 | | 0.01 |
| Presence of additional factors affecting outcome | | -17.97* | | -12.1 | | -22.44* | | -27.27** |

*Note*. dB HL = decibels Hearing Loss; PLS = Preschool Language Scale.
*$p < 0.001$

**Tests' Sensitivity to Change Over Time.** With regard to change over time, PLS-5 standard scores did not differ significantly between first [auditory comprehension; $M = 94.26$: expressive communication; $M = 96.04$) and second [auditory comprehension; $M = 93.73$: expressive communication; $M = 96.43$) assessments for either scale [auditory comprehension: $t(26) = 1.5623$, $p > 0.0125$; expressive communication: $t(26) = -0.15823$, $p > 0.0125$]. However, growth scale values were higher at second assessments [auditory comprehension; $M = 405.79$: expressive communication; $M = 396.89$] than first assessments [auditory comprehension; $M = 373.8$: expressive communication; $M = 363,73$) for both subtests [auditory comprehension; $t(26) = 11.623$, $p < 0.0125$: expressive communication; $t(26) = 10.589$, $p < 0.0125$].

We were underpowered to statistically evaluate whether change over time occurred for the MBCDI-2 scores as there were only nine children with data for repeat assessments (see Table 4).

### *Procedure Feasibility*

Fifty-eight SLPs responded to the end of pilot survey, 18 of whom indicated they did not apply the procedure over the one-year pilot. The results for the 40 eligible SLPs are summarized in Appendices A–D. Overall, the majority of SLPs (> 60%) were confident in their knowledge, skills, and abilities to implement the new Program-level outcome monitoring procedures and were confident that they had the physical resources and support from management to do so. There was a lack of strong agreement (< 60%) amongst SLPs that the procedures themselves would be useful within clinical practice and to families. As a group, the majority of SLPs did not agree that the time to administer the Tier 1 procedures either in isolation, or in conjunction with Tier 2 individual vulnerability testing procedures, was appropriate for clinical practice.

## Pilot Study 2 – Tier 2 Individual Vulnerability Testing

### Method

### *Procedure*

The decision to participate in the Tier 2 individual vulnerability testing pilot during the Tier 1 Program-level pilot was left to the discretion of regional management. Ten of the eleven volunteer sites from Pilot Study 1 agreed to participate in the additional individual vulnerability testing pilot and implement both procedures at the same time. Twenty-three SLPs collected data for the Tier 2 procedure and completed post-pilot surveys to identify barriers and facilitators to implementation.

### *Participants Assessed in Pilot Study 2*

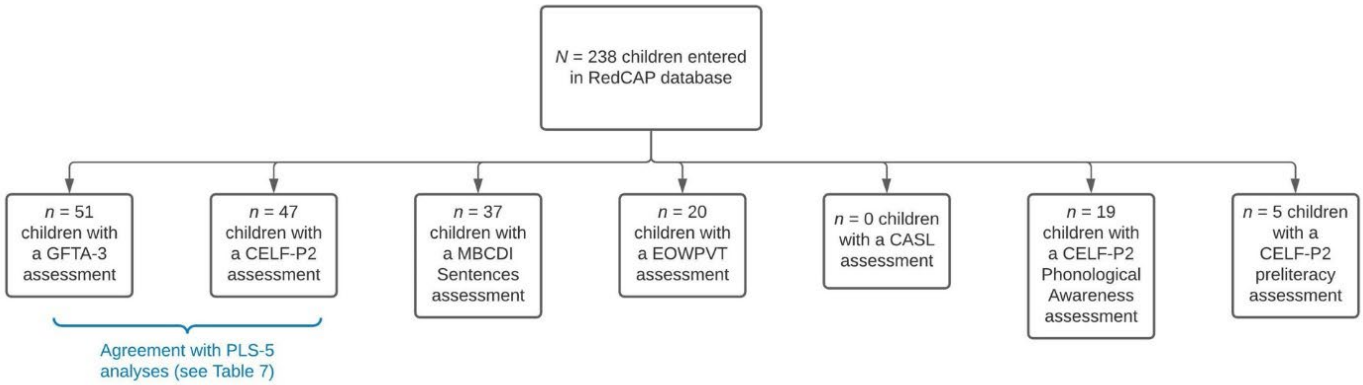
Over the course of the pilot, our team noticed a trend for SLPs from regions that we believed were involved in Pilot Study 2 to flag many children ($n = 72$ of 238) as not being involved in the pilot (i.e., they were only completing assessments from Pilot Study 1). The lack of data for Pilot Study 2 assessments in the REDCap database suggested there was a miscommunication of pilot procedure expectations. It is unclear why this miscommunication occurred, because the decision for a region to participate in Pilot Study 2 was left to regional coordinators. It is possible there was a miscommunication between our research team and the coordinators, between coordinators and SLPs, or a lack of clarity in the implementation materials provided by our team to SLPs.

Once the error was identified, our team reconnected with regional coordinators to confirm their participation and additional communication was provided to SLPs. Following this re-communication, our team observed that SLPs who originally indicated they were not involved in the Tier 2 pilot began to enter individual vulnerability data. However, a significant number of children from these regions were not

assessed with the tests from the Tier 2 procedure before expectations were recommunicated. As a result, a significant amount of expected data (57% of children in piloting regions) was not collected ($n$ = 126 of 238). Reasons for missing data included issues surrounding the original miscommunication ($n$ = 72), and practical limitations ($n$ = 10). Reasons were unknown in 44 cases. Moreover, assessment data for all tests were not reported because the procedure did not require SLPs to administer all tests, but rather gave them choices. The amount of missing data limited our ability to fulfill our primary analytic purposes, but some preliminary hypotheses were developed based on the available data. Our analyses were based on data that were available for children who were assessed using the Goldman-Fristoe Test of Articulation, Third Edition (Goldman & Fristoe, 2015; GFTA-3) and the Clinical Evaluation of Language Fundamentals, Preschool, Second Edition (CELF-P2; Semel et al., 2004; see Figure 4 and Table 7). We included data for all children for whom data were entered in the REDCap database. Children were included regardless of audiological profile (i.e., we included children with unilateral and conductive losses), as our primary aim was to explore whether the Tier 2 tests agreed in their characterization of whether a child had an impairment based on Tier 1 testing regardless of hearing characteristics.

**Figure 4**

*Children from the Overall Sample Included in Pilot Study 2 Analyses*




*Note.* GFTA-3 = Goldman-Fristoe Test of Articulation, 3rd ed.; CELF-P2 = Clinical Evaluation of Language Fundamentals, Preschool, 2nd ed.; MBCDI = MacArthur-Bates Communicative Development Inventories; EOWPVT = Expressive One Word Picture Vocabulary Test; PLS-5 = Preschool Language Scale, 5th ed. CASL-2 = Comprehensive Assessment of Language Fundamentals, 2nd ed.

**Table 7**

*Demographics of Children Included in Pilot Study 2*

| | ***Children with GFTA-3 Sounds-in-Words Assessments (n = 48)*** | | | ***Children with CELF-P2 Word Structure Assessments (n = 46)*** | | |
|---|---|---|---|---|---|---|
| Variable | *n* | *M* (range) | SD | *n* | *M* (range) | SD |
| Presence of Additional Factors | 16 | | | 10 | | |
| Bilateral Hearing Loss | 31 | | | 34 | | |
| BEPTA (dB HL) | | 51.47 (18.75–98.33)* | 24.17 | | 47.43 (17.25–92.5)* | 23.18 |
| Unilateral Hearing Loss | 17 | | | 12 | | |
| PTA (dB HL) | | 47.74 (26.25–81.25) | 18.84 | | 47.96 (28.75–83.75) | 16.6 |
| Conductive Hearing Loss | 4 | | | 4 | | |
| Age at PLS Assessment (months) | | 43.54 (31–71) | 8.58 | | 48.63 (34–71) | 9.48 |
| Expressive Communication (Standard Score) | | 103.85 (64–150) | 19.12 | | 103.53 (54–150) | 22.14 |
| Auditory Comprehension (Standard Score) | | 103.34 (73–150) | 16.67 | | 101.55 (51–150) | 20.26 |
| Age at IVT Assessment (months) | | 44 (31–71) | 6.68 | | 49.19 (37–72) | 9.27 |
| GFTA-3 Sounds-in-Words (Standard Score) | | 89.32 (42–123) | 18.2 | | | |
| CELF-P2 Word Structure (Percentile Ranks*) | | | | | 42.2 (0.1-99) | 35.74 |

*Note.* GFTA-3 = Goldman-Fristoe Test of Articulation, 3rd Ed.; CELF-P2 = Clinical Evaluation of Language Fundamentals, Preschool – 2nd Ed.; BEPTA = better-ear pure-tone average; dB HL = decibels Hearing Loss; PLS = Preschool Language Scale; PTA = pure-tone average; IVT = Individual Vulnerability Test. Pure tone averages < 25 db were the result of high or low frequency hearing losses, where the child experienced hearing losses at some, but not all, frequencies. Standard scores are not available for the CELF-P2 subtests.

## Analysis

### *Data Usability*

We had intended to develop a structural equation model to map the relations between overall spoken language assessment data (collected using the Tier 1 PLS-5 and MBCDI-2) and different domains of spoken language (data collected using the Tier 2 individual vulnerability testing procedures), but we were underpowered to perform these analyses because of the large amount of missing data. Instead, we explored the level of classification agreement (whether a child performed below age expectations on a given test) between the two most frequently completed tests (the GFTA-3 Sounds-in-Words subtest, which measures accuracy of articulation of consonants and consonant clusters during single word production, and the CELF-P2 Word Structure subtest, which measures accuracy of grammar use) and scores on the PLS-5. As in Pilot Study 1, the first author (O.D.) corrected the GFTA-3 standard scores and CELF-P2 percentiles to ensure consistency and developed audiological profiles for each child. For both subtests, few corrections were required (14% for the GFTA-3, and 2% for the CELF-P2).

Children's scores for the GFTA-3 Sounds-in-Words and CELF-P2 Word Structure subtests and both scales of the PLS-5 were categorized as *within*, *borderline*, or *below* age expectations based on the cut-score with the highest diagnostic accuracy as well as a 90% confidence interval around that score. For instance, the PLS-5 has the highest diagnostic accuracy when a cut-score of -1*SD* below the mean is used. In this case, we categorized a score as *below* age-expectations when the child's standard score was lower than the cut-score *and* the upper-bound of the 90% confidence interval was also below the cut-score. Similarly, a score was considered *within* age expectations when the child's score was above the cut-score and the lower bound of the 90% confidence interval was above the cut-score. In cases where the child's score was diagnostically ambiguous (the upper and lower bound of the confidence interval fell both above and below the cut-score), the child was categorized as *Borderline*. Instances were then tallied when the categorization of the PLS-5 was higher, the same as, or lower than the diagnostic categorization of the companion tests. Finally, we evaluated agreement between the PLS-5 auditory comprehension and expressive communication scales and GFTA-3 and CELF-P2 subtests using a Kendall's Coefficient of Concordance.

### *Procedure Feasibility*

Survey data were analyzed descriptively as in Pilot Study 1.

## Results

### *Agreement between Tier 1 and Tier 2 Tests in Diagnostic Categorizations*

The proportions of children considered *within, borderline,* or *below* age expectations for each test are reported in Tables 8–11. Children's categorization on both PLS-5 auditory comprehension and expressive communication scales agreed with one another, and with diagnostic categorization on the GFTA-3 ($Wt(46) = 0.71$, $p < 0.05$) and CELF-P2 ($Wt(43) = 0.73$, $p < 0.05$). Analyses were not repeated for scores on the other tests included in the Tier 2 procedure because of the small amount of data available for each assessment and a lack of sensitivity/specificity data to define within/borderline/below age expectations for the MBCDI Words and Sentences form.

**Table 8**

*Agreement Between PLS-5 Auditory Comprehension and GFTA-3 Sounds-in-Words Subtest*

| | GFTA-3 | | |
|---|---|---|---|
| **PLS-5** | **Within** | **Border** | **Below** |
| Within | 32 | 4 | 1 |
| Border | 4 | 0 | 2 |
| Below | 0 | 0 | 5 |

*Note.* PLS = Preschool Language Scale; GFTA-3 = Goldman-Fristoe Test of Articulation, 3rd ed.

**Table 9**

*Agreement Between PLS-5 Expressive Communication and GFTA-3 Sounds-in-Words Subtest*

| | GFTA-3 | | |
|---|---|---|---|
| **PLS-5** | **Within** | **Border** | **Below** |
| Within | 31 | 3 | 2 |
| Border | 4 | 1 | 4 |
| Below | 0 | 0 | 2 |

*Note. PLS = Preschool Language Scale; GFTA-3 = Goldman-Fristoe Test of Articulation, 3rd ed.*

**Table 10**

*Agreement between PLS-5 Auditory Comprehension and CELF-P2 Words Structure Subtest*

| | CELF-P2 | | |
|---|---|---|---|
| **PLS-5** | **Within** | **Border** | **Below** |
| Within | 17 | 12 | 4 |
| Border | 0 | 5 | 2 |
| Below | 0 | 0 | 5 |

*Note. PLS = Preschool Language Scale; CELF-P2 = Clinical Evaluation of Language Fundamentals, Preschool, 2nd ed.*

**Table 11**

*Agreement Between PLS-5 Expressive Communication and CELF-P2 Words Structure Subtest*

| | CELF-P2 | | |
|---|---|---|---|
| **PLS-5** | **Within** | **Border** | **Below** |
| Within | 16 | 12 | 3 |
| Border | 1 | 5 | 2 |
| Below | 0 | 0 | 5 |

*Note. PLS = Preschool Language Scale; CELF-P2 = Clinical Evaluation of Language Fundamentals, Preschool, 2nd ed.*

### *Procedure Feasibility*

At the end of the pilot, 36 SLPs completed online surveys to provide feedback on the new Tier 2 procedures. Thirteen SLPs indicated that they did not use the individual vulnerability testing procedure at all over the course of the pilot, and therefore did not complete the remaining survey questions. Summaries of the remaining 23 SLPs' responses are outlined in Appendices E–I.

As was the case with the Tier 1 Program-level outcome monitoring procedures, the majority (> 60%) of SLPs were confident in their knowledge, skills, and abilities to implement the Tier 2 individual vulnerability testing procedures, with the notable exception of the *Comprehensive Assessment of Language Fundamentals*, *2nd edition* (CASL-2; Carrow-Woodfolk, 2017). The majority of SLPs also reported that they had resources such as test manuals (except for the CASL-2) and managerial support. Most SLPs agreed or strongly agreed that results from the Tier 2 testing supported their clinical decision making and could be used to improve services for families of children who are DHH.

Although SLPs reported that the individual vulnerability test process provided valuable information, there was a lack of consensus about whether the amount of time required to implement was feasible. The percentage of SLPs who reported being able to consistently implement the Tier 2 process was also divided, and 78% of respondents reported that additional administrative support or time release from other clinical duties would be helpful for implementing it. In open-ended comments, some SLPs reported concerns that the combination of Tier 1 and Tier 2 testing was overly burdensome for children, families, and themselves. Finally, when asked whether it would be helpful to forgo Tier 2 testing altogether, 47% of SLPs reported feeling neutral, and the remaining SLPs were divided between agreeing and disagreeing.

## Discussion

These two pilot studies present preliminary evidence for the usability and feasibility of the spoken language outcome monitoring procedure developed by Daub and Oram Cardy (2021). For program evaluation purposes, repeated assessment using a narrow set of omnibus language tests (i.e., the MBCDI-2 and the PLS-5) was expected to support group level analysis of outcomes for children who are DHH. By using the same tests over time, we expected that any changes we observed would be attributable to the child's development, rather than changes in the psychometric properties of the assessment tools. This is the first account, to our knowledge, of an effort to evaluate a spoken language outcome monitoring procedure for an EHDI program. Although the need for routine spoken language outcome monitoring is clear (JCIH, 2007, 2013, 2019; Moeller et al., 2013), there is limited guidance for how to accomplish the diverse assessment purposes proposed under these recommendations.

### Data Usability

Data from Pilot Study 1 suggest that the PLS-5 might be appropriate for fulfilling program evaluation purposes, however there was less evidence to support use of the MBCDI-2. PLS-5 growth scale values were sensitive to change over time (Daub et al., 2017) and standard scores were predicted by additional factors, so in this regard, the PLS-5 conformed to our prediction that it would capture growth in children's spoken language skills. The MBCDI-2 did not conform to our prediction, although we did not have a large enough sample of children with two MBCDI-2 assessments to adequately evaluate whether the MBCDI-2 scores changed over time.

There are several possible explanations for the lack of evidence to support using the MBCDI-2. First, it is possible that the impact of hearing loss on the aspects of language measured by the MBCDI-2 Words and Gestures form is not observed in very young children ($M < 14$ months, in our sample). Without data to compare performance on the PLS-5 in children under 18 months, we cannot be assured that the PLS-5 would have been any more informative at this young age. Our findings might also be explained by the scoring characteristics of the MBCDI-2 itself: it has been well documented that there is a wide range of typical variation associated with MBCDI-2 scores, particularly with regard to words produced in children younger than 18 months (Fenson et al., 2000; Feldman et al., 2000). Further, a single total number of words can correspond to a wide range of percentile ranks and small changes in total scores can dramatically influence a child's percentile rank. For example, for an 8-month-old boy who produces no words, a percentile rank of between 5 and 55 can be assigned, whereas an 8-month-old boy producing a single word corresponds to a percentile rank of either 65 or 70 (Fenson et al., 2007, p. 120). Therefore, the scoring properties of the MBCDI-2 may mean that it is not sensitive enough to use as a Program-level outcome measure in young children.

Why neither test was predicted by severity of hearing loss (BEPTA) is less immediately clear. The lack of an effect is particularly surprising for the PLS-5 for several reasons. First, the use of standard scores rather than percentile ranks allows for more precise scoring than the MBCDI-2. Second, we used the PLS-5 for a much broader age range than the MBCDI-2 and the lack of effect cannot be accounted for by the age of the children in our sample. We also had a much larger sample for the PLS-5 analyses than the MBCDI-2 and the lack of effect cannot be explained by a lack of power. Finally, we had a wide range of both PLS-5 scores (e.g., between 50 and 150) and BEPTA (e.g., 20–107.5). For both variables, we had data representing the full range of possible values and our null finding cannot be accounted for by range restriction of either variable. Interestingly, the average PLS-5 scores in our sample (for children without additional factors) were higher than what is typically reported in outcome studies (e.g., Tomblin et al., 2015) and approximate a normal distribution, which has a mean standard score of

100 and standard deviation of 15. In our data, children without additional factors (as a group) had a mean of 100.92 (*SD* = 20.5) on the expressive communication scale and a mean of 98.98 (*SD* = 19.81) on the auditory comprehension scale (see Supplemental Material 5). This raises the possibility that perhaps the lack of influence of BEPTA on PLS-5 scores accurately reflects children's spoken language outcomes. All children in our sample were receiving comprehensive EHDI services and wearing well-fitted hearing aids (see Supplemental Materials 1–4). If an EHDI program's goal is to support age-appropriate language outcomes by providing children with consistent access to auditory information, then it is reasonable to expect that severity of hearing loss should not predict outcomes but other variables (e.g., additional factors influencing performance) would. In our data, additional factors were broadly defined as any factor SLPs believed may influence a child's performance on the test, above and beyond their hearing loss. Once those factors were statistically controlled for (by entering the variable into our regressions), severity of hearing loss did not uniquely contribute to children's performance.

It may be the case that our data is preliminary evidence that the IHP is achieving their goal of ameliorating the impact of inconsistent auditory access on spoken language outcomes. That is, perhaps the impact of severity of hearing loss on spoken language development is mitigated by response to intervention. This idea is consistent with previous work suggesting that children with lower language skills and more severe hearing loss show greater gains in PLS-5 growth scale values after amplification (Daub et al., 2017). This idea also aligns with research showing children with permanent hearing loss catch up to their peers with typical hearing thresholds over time as a function of access to auditory information (conceptualized as consistent hearing aid use and quality of hearing aid fit; Tomblin et al., 2015). However, we remain cautious in our interpretation of the data. Without access to SLPs' caseloads to ensure that *all* children in the IHP were reflected in our data, we cannot confirm that our sample is representative of the IHP. Future work using population-level data from the IHP will model how children's spoken language outcomes change over time, and as a function of intervention characteristics such as quality of hearing aid fit. We are also cautious in our interpretations because our sample was insufficient to identify whether some additional factors differentially interacted with severity of hearing loss in predicting spoken language outcomes. There is some evidence that certain comorbid diagnoses (e.g., autism, cerebral palsy, developmental delay) are particularly influential in spoken language outcomes of children who are DHH (Cupples et al., 2014). It is also possible that some performance factors (e.g., inattention during testing) influenced children's hearing thresholds. Future work evaluating the outcomes of children across the entire IHP is warranted to identify whether the lack of effect of BEPTA on children's spoken language outcomes holds for children with, and without, additional complicating factors.

Pilot Study 2 was conducted to evaluate the usability of data from an individual vulnerability testing procedure. Because children who are DHH have ongoing inconsistent access to auditory information, it has been documented that they continue to struggle in certain domains of spoken language (e.g., Moeller et al., 2007) even when they may perform within normal limits on omnibus measures. As a result, an outcome monitoring procedure that only reports on spoken language outcomes broadly has the potential to over-estimate children's abilities and miss opportunities to develop additional supports for specific domains of spoken language development. Due to missing data, we were unable to fulfill our planned analyses, however, preliminary analyses exploring the agreement between overall language comprehension and use of language (PLS-5) with articulation (GFTA-3) and grammar (CELF-P2) indicated that diagnostic categorizations largely agreed. Our data were insufficient to report on whether the individual vulnerability testing procedure provided unique clinical information. Note that these analyses do not account for all domains of language that we planned to measure, nor do they account for longitudinal relationships between measures. Future, longitudinal research evaluating this procedure on a larger and more representative sample of children who are DHH is needed to draw definitive theoretical and clinical conclusions.

**Procedure Feasibility**

Both pilot studies evaluated the feasibility of the recommended procedures through a descriptive evaluation of SLPs' survey responses. For both the Tier 1 Program-level outcome monitoring procedure and the Tier 2 individual vulnerability testing procedure, SLPs reported a high degree of confidence in their knowledge and skills to implement the procedures accurately. In both pilot studies, SLPs flagged concerns about the amount of time it took to complete the procedures. Note that most SLPs participating in Pilot Study 1 were also participating in Pilot Study 2. Therefore, we are unable to identify whether SLPs' perceptions of the amount of time each procedure took was a true reflection of each procedure independently *or* if completing both procedures simultaneously impacted their perceptions.

The key difference in SLPs' perceptions between the two pilot studies related to clinical relevance. As a group, SLPs were less convinced of the value of the Tier 1 Program-level outcome monitoring procedure than they were of the Tier 2 individual vulnerability testing procedure. Although we are cautious in the generalizability of this finding because of the small number of SLPs who completed surveys in Pilot Study 2, it is not necessarily surprising. The Tier 1 Program-level outcome monitoring procedure was intended to support program evaluation and we know that many children who are DHH perform within normal limits on omnibus language assessments but still have needs in certain domains of language. Although our usability data for the individual vulnerability testing pilot was insufficient to make recommendations for EHDI programs and to determine whether tests provided unique

predictive information, SLPs' feedback indicates that valuable clinical information may be gained from the Tier 2 procedure. Future work is warranted where administration of tests can be more closely controlled to evaluate the relation between the proposed measures in the individual vulnerability testing procedure.

**Limitations & Future Directions**

Naturally, the results of our feasibility analyses are heavily dependent on the IHP's context and may not necessarily generalize to other EHDI programs. However, our results provide preliminary evidence that the procedures recommended in Daub and Oram Cardy (2021) are possible to implement, and are largely perceived as informative by SLPs. In addition to the findings reported here, our surveys (see Supplemental Materials) can support other EHDI programs in evaluating their own procedures.

Finally, it is unknown whether the procedures we evaluated are implementable at the scale of an entire EHDI program, whether appropriate implementation is sustainable over time and survives staff turnover, and whether the data collected here can be used to benefit programs, families, and children who are DHH. Future work will monitor use of the procedures over time and document the impact of data on program planning and services.

Taken together, results highlight the importance of carefully considering the questions EHDI programs seek to answer with spoken language outcome monitoring and the methods they use to answer these questions. Testing is not a neutral activity. There are costs associated with engaging in testing including using limited resources to test rather than allocating those resources elsewhere (e.g., intervention). There are also costs for children and their families who engage in testing such as time and emotional impact of engaging in repeated testing (e.g., frustration with their child's progress; Daub et al., 2021; Messick, 1993). Risks associated with testing for families and children who are DHH are another factor that must be considered. If inappropriate tests are used, or data are misinterpreted, SLPs may draw erroneous conclusions about the effectiveness of an intervention, or about children who are DHH themselves. If the data that are collected during spoken language outcome monitoring cannot answer the questions they were intended to, then the costs and risks are not justified. If the procedure used to collect data is too burdensome to be implemented consistently and accurately, then the resulting data may become unusable and testing is similarly unjustified. The data reported here suggest that our proposed Tier 1 Program-level procedure may result in data appropriate for our intended purposes, but we have insufficient evidence to justify the implementation of the Tier 2 individual vulnerability testing procedure in clinical practice. In presenting these findings to the IHP, we recommended adoption of the Program-level procedure as originally defined with regular data monitoring for the first two years to verify whether the data are suitable at the scale of the entire program. For the individual vulnerability testing, we recommended sharing the tests we selected for Tier 2 monitoring with SLPs (Daub & Oram Cardy, 2021), and the rationale for monitoring key areas of vulnerability in children who are DHH. This would leave SLPs free to use the recommended Tier 2 tools when they identify a need in clinical practice, rather than mandating it program-wide.

Although spoken language outcome monitoring is predicted to support various stakeholders' decision-making (JCIH, 2007, 2013, 2019), if spoken language outcome monitoring procedures fail to improve programs or children's outcomes in practice, then the efforts spent regularly assessing children's spoken language development might be better spent elsewhere. As interdisciplinary professionals invested in improving outcomes for children who are DHH, it is imperative that we grapple with these psychometric and implementation issues in the design and evaluation of EHDI programs.

**Conclusions**

This paper summarizes preliminary evidence of the usability and feasibility of a spoken language outcome monitoring procedure for EHDI programs. This evidence suggests that the Tier 1 Program-level procedure may be feasible to implement and result in usable data, although future work is needed to evaluate whether the data are sufficient to address program evaluation needs once implemented across the IHP. There was insufficient evidence to recommend the use of the Tier 2 individual vulnerability testing procedures to implement in EHDI programs at this point. Future work will evaluate whether the procedure can be accurately implemented, whether accurate implementation can be sustained over time, and whether the procedure influences decision-making to improve program and children's outcomes.

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## EHDInfo

**IOWA EHDI Program recognized as the best website among the website of the year awards for the past 12 years.**

## Appendix A

Speech-Language Pathologists' Opinions on the Tier 1 Procedure Statement

| Statement | Strongly Disagree *n* (%) | Disagree *n* (%) | Neither agree nor disagree *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (Range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| The IHP's new Program-level Outcome Monitoring Procedure was useful for my clinical practice. | 1 (2.5%) | 4 (10%) | 17 (42.5%) | 15 (37.5%) | 3 (7.5%%) | 3 (1–5) | 0 (0%) |
| I was able to consistently implement the new Program-level Outcome Monitoring recommendations in my practice. | 1 (2.5%) | 14 (35%) | 7 (17.5%) | 17 (42.5%) | 1 (2.5%) | 4 (1–5) | 0 (0%) |

*Note.* IHP = Infant Hearing Program.

**Appendix B**

Speech-Language Pathologists' (SLP) Opinions of their Capacity to Implement the Tier 1 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neither agree nor disagree *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (Range) | Not answered *n* (%) |
|---|---|---|---|---|---|---|---|
| Over the past year I felt I had the clinical skills required to implement the new Program-level Outcome Monitoring Procedures. | 0 (0%) | 0 (0%) | 3 (7.5%) | 26 (65%) | 11 (27.5%) | 4 (3–5) | 0 (0%) |
| I am familiar with the administration of the MacArthur-Bates Communicative Development Inventories- Words & Gestures (MBCDI). | 1 (2.5%) | 1 (2.5%) | 2 (5%) | 25 (62.5%) | 10 (25%) | 4 (1–5) | 1 (2.5%) |
| I was able to accurately score and use the norms tables for the MacArthur-Bates Communicative Development Inventories Words & Gestures (MBCDI). | 2 (5%) | 1 (2.5%) | 5 (12) | 19 (47.5%) | 9 (22.5%) | 4 (1–5) | 4 (10%) |
| I am familiar with the administration of the Preschool Language Scales-5th Edition. | 0 (0%) | 0 (0%) | 0 (0%) | 21 (52.5%) | 19 (47.5%) | 4 (4–5) | 0 (0%) |
| I was able to accurately score and use the norms tables for the Preschool Language Scales-5th Edition. | 0 (0%) | 0 (0%) | 0 (0%) | 18 (45%) | 21 (52.5%) | 5 (4–5) | 1 (2.5%) |
| The new Program-level Outcome Monitoring Procedures have helped me with my clinical decision-making. | 3 (7.5%) | 7 (17.5%) | 12 (30%) | 14 (35%) | 3 (7.5%) | 4 (1–5) | 1 (2.5%) |
| The new Program-level Outcome Monitoring Procedures have helped parents with their decision-making. | 1 (2.5%) | 4 (10%) | 21 (52.5%) | 9 (22.5%) | 3 (7.5%) | 3 (1–5) | 2 (5%) |
| Repeat administration of the Program-level Outcome Monitoring tools to the same child 6–12 months later benefited the families and children that I serve. | 1 (2.5%) | 3 (7.5%) | 15 (37.5%) | 12 (30%) | 4 (10%) | 3 (1–5) | 5 (12.5%) |
| Repeat administration of the Program-level Outcome Monitoring tools to the same child was useful for my own clinical practice. | 1 (2.5%) | 3 (7.5%) | 11 (27.5%) | 18 (45%) | 3 (7.5%) | 4 (1–5) | 4 (10%) |

**Appendix C**

Speech-Language Pathologists' (SLP) Opinions on the Practice Environment and the Tier 1 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neither agree nor disagree *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (Range) | Not answered *n* (%) |
|---|---|---|---|---|---|---|---|
| The length of time it took to administer the recommended Program-level Outcome Monitoring tests was appropriate for incorporation into routine clinical practice. | 5 (12.5%) | 10 (25%) | 7 (17.5%) | 15 (37.5%) | 2 (5%) | 4 (1–5) | 1 (2.5%) |
| The length of time it took to score and interpret the results of the recommended Program-level Outcome Monitoring tests was appropriate for incorporating into routine clinical practice. | 3 (7.5%) | 4 (10%) | 13 (32.5%) | 19 (47.5%) | 2 (5%) | 4 (1–5) | 1 (2.5%) |
| The length of time it took to talk with parents about results of the recommended Program-level Outcome Monitoring tests was appropriate for incorporation into clinical practice. | 1 (2.5%) | 4 (10%) | 13 (32.5%) | 19 (47.5%) | 2 (5%) | 4 (1–5) | 1 (2.5%) |
| The time it took to do the recommended Program-level Outcome Monitoring and reporting did NOT negatively impact other areas of my practice. | 2 (5%) | 9 (22.5%) | 12 (30%) | 14 (35%) | 2 (5%) | 4 (1–5) | 1 (2.5%) |
| The environment in which I worked made it difficult for me to implement the recommended Program-level Outcome Monitoring. | 7 (17.5%) | 15 (37.5%) | 8 (20%) | 7 (17.5%) | 0 (0%) | 2 (1–4) | 3 (7.5%) |
| I had the supplies I needed (e.g., test forms) to implement the new Program-level Outcome Monitoring. | 0 (0%) | 2 (5%) | 0 (0%) | 19 (47.5%) | 19 (47.5) | 5 (2–5) | 0 (0%) |
| When I had a question about the Program-level Outcome Monitoring Procedures, I consulted with my colleagues. | 0 (0%) | 3 (7.5%) | 4 (10%) | 24 (60%) | 5 (12.5%) | 4 (2–5) | 4 (10%) |
| When I had a question about the Program-level Outcome Monitoring Procedures, I consulted with my managers/ administrators. | 1 (2.5%) | 8 (20%) | 7 (17.5%) | 16 (40%) | 1 (2.5%) | 4 (1–5) | 7 (17.5%) |
| When I had a question about the Program-level Outcome Monitoring Procedures, I consulted the "Pilot Implementation Q&A" section of Western's OWL site. | 1 (2.5%) | 8 (20%) | 4 (10%) | 20 (50%) | 4 (10%) | 4 (1–5) | 3 (7.5%) |

**Appendix C (cont.)**

Speech-Language Pathologists' (SLP) Opinions on the Practice Environment and the Tier 1 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neither agree nor disagree *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (Range) | Not answered *n* (%) |
|---|---|---|---|---|---|---|---|
| I had the resources I needed (e.g., administrative support for scheduling, data entry) to do the new Program-level Outcome Monitoring Procedures. | 4 (10%) | 13 (32.5%) | 8 (20%) | 11 (27.5%) | 3 (7.5%) | 2 (1–5) | 1 (2.5%) |
| I had permission from my manager to take the time I needed to complete Program-level Outcome Monitoring Procedures. | 0 (0%) | 0 (0%) | 5 (12.5%) | 27 (67.5%) | 6 (15%) | 4 (3–5) | 2 (5%) |
| Getting timely feedback from experts (i.e., the research team at Western University) helped me to implement the new Program-level Outcome Monitoring Procedures. | 0 (0%) | 2 (5%) | 15 (37.5%) | 13 (32.5%) | 5 (32.5%) | 3 (2–5) | 5 (12.5%) |
| The SLPs I worked with were excited about the new Program-level Outcome Monitoring Procedures. | 5 (12.5%) | 8 (20%) | 18 (45%) | 4 (10%) | 2 (5%) | 3 (1–5) | 3 (7.5%) |
| Managers/administrators I worked with were supportive of the new Program-level Outcome Monitoring Procedures. | 0 (0%) | 0 (0%) | 10 (25%) | 24 (60%) | 4 (10%) | 4 (3–5) | 2 (5%) |
| The parents I worked with were interested in the results of the new Program-level Outcome Monitoring Procedures. | 5 (12.5%) | 2 (5%) | 21 (52.5) | 10 (25%) | 1 (2.5) | 3 (1–5) | 1 (2.5%) |
| The task of completing the MBCDI was not too difficult for parents (respondents) to perform. | 5 (12.5%) | 3 (7.5%) | 7 (17.5%) | 19 (47.5%) | 3 (7.5%) | 4 (1–5) | 3 (7.5%) |
| The task of completing the MBCDI was not too time consuming for parents (respondents) to perform. | 3 (7.5%) | 9 (22.5%) | 9 (22.5%) | 15 (37.5%) | 1 (2.5%) | 4 (1–5) | 3 (7.5%) |

*Note.* MBCDI = MacArthur-Bates Communicative Development Inventories.

**Appendix D**

Speech Language Pathologists' (SLP) Opinions on the Quality of the Tier 1 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neither agree nor disagree *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (Range) | Not answered *n* (%) |
|---|---|---|---|---|---|---|---|
| The new Program-level Outcome Monitoring Procedures were similar to the previous outcome monitoring procedures for the IHP. | 1 (2.5%) | 5 (12.5%) | 10 (25%) | 23 (57.5%) | 2 (2.5%) | 4 (1–5) | 0 (0%) |
| The new Program-level Outcome Monitoring Procedures were an improvement over the current procedure. | 2 (5%) | 4 (5%) | 17 (42.5%) | 13 (32.5%) | 5 (12.5%) | 3 (1–5) | 1 (2.5%) |
| I found the MacArthur-Bates Communicative Development Inventories Words & Gestures to be a high-quality clinical outcome evaluation tool. | 1 (2.5%) | 6 (15%) | 12 (30%) | 17 (42.5%) | 1 (2.5%) | 4 (1–5) | 3 (7.5%) |
| I found the MacArthur-Bates Communicative Development Inventories Words & Gestures to be a valid and reliable tool for preschoolers with permanent hearing loss. | 2 (5%) | 4 (10%) | 14 (35%) | 15 (37.5%) | 2 (5%) | 4 (1–5) | 3 (7.5%) |
| I felt the MacArthur-Bates Communicative Development Inventories Words & Gestures was the right choice for evaluating spoken language outcomes for the IHP's youngest children. | 2 (5%) | 3 (7.5%) | 10 (25%) | 17 (42.5%) | 5 (12.5%) | 4 (1–5) | 3 (7.5%) |
| I found the Preschool Language Scales-5th Edition to be a high-quality clinical outcome evaluation tool. | 1 (2.5%) | 6 (15%) | 14 (35%) | 14 (35%) | 3 (7.5%) | 3 (1–5) | 2 (5%) |
| I found the Preschool Language Scales-5th Edition to be a valid and reliable tool for preschoolers with permanent hearing loss. | 1 (2.5%) | 6 (15%) | 14 (35%) | 14 (35%) | 3 (7.5%) | 4 (1–5) | 2 (5%) |
| I felt the Preschool Language Scales-5th Edition was the right choice for evaluating spoken language outcomes for older children in the IHP. | 1 (2.5%) | 8 (20%) | 13 (32.5%) | 14 (35%) | 2 (5%) | 4 (1–5) | 2 (5%) |
| I do not have concerns about the validity/reliability of the Preschool Language Scales-5th Edition | 3 (7.5%) | 8 (20%) | 11 (27.5%) | 14 (35%) | 3 (7.5%) | 4 (1–5) | 1 (2.5%) |
| I feel that implementing the new Program-level Outcome Monitoring Procedures will result in a systematic evaluation of spoken language outcomes in children with hearing loss in the IHP. | 2 (5%) | 1 (2.5%) | 13 (32.5%) | 19 (47.5%) | 4 (10%) | 4 (1–5) | 1 (2.5%) |

*Note.* IHP = Infant Hearing Program.

## Appendix E

Speech Language Pathologists' (SLP) Opinions on the Tier 2 Procedure Statement

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neutral *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (Range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| The IVT procedures were useful for improving services for families of children with hearing loss. | 2 (9.5%) | 1 (4.7%) | 4 (19%) | 13 (61%) | 0 (0%) | 4 (1–4) | 3 (12.5%) |
| The IVT procedures were useful for my clinical practice. | 2 (9.5%) | 2 (9.5%) | 2 (9.5%) | 14 (66.7%) | 0 (0%) | 4 (1–4) | 3 (12.5%) |
| I was able to consistently implement the IVT procedures in my practice. | 3 (13.6%) | 5 (22.7%) | 3 (13.6%) | 10 (45.5%) | 0 (0%) | 4 (1–4) | 2 (8.3%) |

*Note.* IVT = Individual Vulnerability Test.

**Appendix F**

Speech Language Pathologists' (SLP) Perceptions of Time Involved in Tier 2 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neutral *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (Range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| The length of time it took to administer the Individual Vulnerability Tests was appropriate for incorporation into routine clinical practice. | 5 (22.7%) | 3 (13.6%) | 4 (18.2%) | 9 (40.9%) | 0 (0%) | 4 (1–4) | 2 (8.3%) |
| The length of time it took to score and interpret the results of the Individual Vulnerability Tests was appropriate for incorporating into routine clinical practice. | 2 (9.1%) | 4 (18.2%) | 3 (13.6%) | 12 (54.5%) | 0 (0%) | 4 (1–4) | 2 (8.3%) |
| The length of time it took to talk with parents about results of the Individual Vulnerability Tests was appropriate for incorporation into clinical practice. | 2 (9.5%) | 2 (9.5%) | 6 (28.6%) | 10 (47.6%) | 0 (0%) | 4 (1–4) | 3 (12.5%) |
| The time it took to do the Individual Vulnerability Testing and reporting negatively impacted other areas of my practice. | 0 (0%) | 8 (36.4%) | 8 (36.4%) | 4 (18.2%) | 1 (4.5%) | 3 (2–5) | 2 (8.3%) |

**Appendix G**

Speech Language Pathologists' (SLP) Perceptions of Practice Environment for the Tier 2 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neutral *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (Range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| The environment in which I work will made it difficult for me to implement the IVT procedures. | 0 (0%) | 14 (63.6%) | 6 (27.3%) | 1 (4.5%) | 0 (0%) | 2 (2–4) | 2 (8.3%) |
| I had the supplies I needed (e.g., test forms) to implement the new IVT procedures. | 0 (0%) | 1 (4.5%) | 0 (0%) | 12 (54.5%) | 8 (36.4%) | 4 (2–5) | 2 (8.3%) |
| When I had questions about the IVT procedures, I consulted my colleagues. | 0 (0%) | 1 (5.3%) | 3 (15.8%) | 12 (63%) | 2 (10.5%) | 4 (2–5) | 5 (20.8%) |
| When I had questions about the IVT procedures, I consulted my manager/ administrators. | 5 (19%) | 1 (5.3%) | 7 (36.8%) | 3 (15.8%) | 7 (36.8%) | 2 (1–4) | 5 (20.8%) |
| When I had questions about the IVT procedures, I consulted the "Pilot Implementation Q&A" section of Western's OWL site. | 1 (4.5%) | 4 (18.2%) | 4 (18.2%) | 9 (40.9%) | 3 (13.6%) | 4 (1–5) | 2 (8.3%) |
| I had the resources I needed (e.g., administrative support for scheduling, data entry) to do the IVT Procedures. | 3 (21%) | 1 (4.8%) | 6 (28.6%) | 7 (33.3%) | 1 (4.8%) | 4 (1–5) | 3 (12.5%) |
| I had permission from my manager to take the time I needed to complete IVT Procedures. | 0 (0%) | 0 (0%) | 5 (23.8%) | 13 (61.9%) | 2 (9.5%) | 4 (3–5) | 3 (12.5%) |
| Getting timely feedback from experts (e.g., the research team at Western University) helped me to implement the IVT Procedures. | 0 (0%) | 1 (5.3%) | 10 (52.6%) | 6 (31.6%) | 1 (5.3%) | 3 (2–5) | 5 (20.8%) |
| The SLPs I work with were excited about the new IVT Procedures. | 6 (27.3%) | 3 (13.6%) | 7 (31.8%) | 4 (18.2%) | 1 (4.5%) | 3 (1–5) | 2 (8.3%) |
| Managers/ administrators I work with were supportive of IVT procedures. | 0 (0%) | 0 (0%) | 5 (23.8%) | 14 (66.7%) | 1 (4.7%) | 4 (3–5) | 3 (8.3%) |

**Appendix G (cont.)**

Speech Language Pathologists' (SLP) Perceptions of Practice Environment for the Tier 2 Procedure

| Statement | Strongly Disagree *n* (%) | Disagree *n* (%) | Neutral *n* (%) | Agree *n* (%) | Strongly Agree *n* (%) | Mode (range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| The parents I worked with were interested in the results of IVT procedures. | 2 (9.1%) | 3 (13.6%) | 9 (41%) | 6 (27.3%) | 1 (4.5%) | 3 (1–5) | 2 (8.3%) |
| The task of completing the MacArthur-Bates Communicative Development Inventories - Words & Sentences was not too difficult for parents (respondents) to perform. | 0 (0%) | 4 (21%) | 3 (15.8%) | 11 (57.9%) | 0 (0%) | 4 (2–4) | 5 (20.8%) |
| The task of completing the MacArthur-Bates Communicative Development Inventories - Words & Sentences was not too time consuming for parents (respondents) to perform. | 0 (0%) | 5 (26%) | 2 (10.5%) | 11 (57.9%) | 0 (0%) | 4 (2–4) | 5 (20.8%) |
| The task of completing the CELF-P2 Pre-literacy Rating Scale was not too difficult for parents (respondents) to perform. | 0 (0%) | 0 (0%) | 3 (42.8%) | 2 (28.6%) | 1 (14.3%) | 3 (3–5) | 17 (70.8%) |
| The task of completing the CELF-P2 Pre-literacy Rating Scale was not too time consuming for parents (respondents) to perform. | 0 (0%) | 1 (14.3%) | 3 (42.9%) | 1 (14.3%) | 1 (14.3%) | 3 (2–5) | 17 (70.8%) |

*Note.* IVT = Individual Vulnerability Test; CELF-P2 = Clinical Evaluation of Language Fundamentals.

**Appendix H**

Speech Language Pathologists' (SLP) Opinions of their Capacity to Implement the Tier 2 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neutral *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| Over the past year I felt I had the clinical skills required to implement the new IVT procedures. | 0 (0%) | 0 (0%) | 3 (13%) | 12 (52%) | 7 (30%) | 4 (3–5) | 1 (4.2%) |
| I am familiar with the administration of the Goldman Fristoe Test of Articulation (GFTA-3). | 0 (0%) | 0 (0%) | 0 (0%) | 9 (39%) | 13 (56.5%) | 5 (4–5) | 1 (4.2%) |
| I was able to accurately score and use the norms tables for the Goldman Fristoe Test of Articulation (GFTA-3). | 0 (0%) | 0 (0%) | 1 (5.3%) | 8 (42.1%) | 9 (47.4%) | 5 (3–5) | 5 (20.8%) |
| I am familiar with the administration of the MacArthur-Bates Communicative Development Inventories - Words & Sentences | 0 (0%) | 0 (0%) | 5 (21.8%) | 12 (52.2%) | 5 (21.7%) | 4 (3–5) | 1 (4.2%) |
| I was able to accurately score and use the norms tables for the MacArthur-Bates Communicative Development Inventories - Words & Sentences | 0 (0%) | 3 (15.8%) | 4 (21.1%) | 8 (42%) | 3 (15.8%) | 4 (2–5) | 5 (20.8%) |
| I am familiar with the administration of the Expressive One Word Picture Vocabulary Test-4th Edition (EOWPVT-4). | 0 (0%) | 0 (0) | 4 (20%) | 12 (60%) | 3 (15%) | 4 (3–5) | 4 (16.7) |
| I was able to accurately score and use the norms tables for the Expressive One Word Picture Vocabulary Test-4th Edition (EOWPVT-4). | 0 0%) | 0 (0%) | 2 (13.3%) | 8 (53%) | 4 (26.7%) | 4 (3-5) | 9 (37.5%) |
| I am familiar with the administration of the Clinical Evaluation of Language Fundamentals, Preschool-Second Edition (CELF-P2) Word Structure subtest. | 0 (0%) | 0 (0%) | 0 (0%) | 8 (34.8%) | 14 (61.9%) | 5 (4–5) | 1 (4.2) |
| I was able to accurately score and use the norms tables for the Clinical Evaluation of Language Fundamentals, Preschool-Second Edition (CELF-P2) Word Structure subtest. | 0 (0%) | 0 (0%) | 0 (0%) | 7 (26.8%) | 11 (57.9%) | 5 (4–5) | 5 (20.8%) |

**Appendix H (cont.)**

Speech Language Pathologists' (SLP) Opinions of their Capacity to Implement the Tier 2 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neutral *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| I am familiar with the administration of the Comprehensive Assessment of Spoken Language-Second Edition (CASL-2) Grammatical Morphemes subtest. | 3 (20%) | 7 (46.7%) | 1 (6.7%) | 3 (20%) | 0 (0%) | 2 (1–4) | 9 (37.5%) |
| I was able to accurately score and use the norms tables for the Comprehensive Assessment of Spoken Language-Second Edition (CASL-2) Grammatical Morphemes subtest. | 0 (0%) | 1 (14.3%) | 3 (42.8%) | 2 (28.6%) | 0 (0%) | 3 (2–4) | 17 (70.8%) |
| The new IVT procedures helped with my clinical decision-making. | 2 (9.5%) | 2 (9.5%) | 4 (19.05%) | 9 (42.9%) | 3 (14.3%) | 4 (1–5) | 3 (12.5%) |
| The new IVT procedures helped parents with their decision-making. | 2 (9.5%) | 3 (14.3%) | 8 (38.1%) | 7 (33%) | 0 (0%) | 3 (1–4) | 3 (12.5%) |
| Repeat administration of the Individual Vulnerability tests to the same child 6–12 months later benefited the families and children that I serve. | 1 (5.3%) | 4 (21%) | 4 (21%) | 7 (37%) | 2 (10.5%) | 4 (1–5) | 5 (20.8%) |

*Note.* IVT = Individual Vulnerability Test.

**Appendix I**

Speech Language Pathologists' (SLP) Perceptions of the Quality of the Tier 2 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neutral *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| I found the assessment tools required for the IVT to be high quality clinical outcome evaluation tools. | 0 (0%) | 1 (4.5%) | 3 (13.6%) | 14 (64.6%) | 3 (13.6%) | 4 (2–5) | 2 (8.3%) |
| I felt the MacArthur-Bates CDI Words and Gestures "Words Produced" was the right choice for evaluating vocabulary vulnerability in children with permanent hearing loss (8–18 months). | 2 (10%) | 2 (10%) | 4 (20%) | 8 (40%) | 3 (15%) | 4 (1–5) | 4 (16.7%) |
| I felt the MacArthur-Bates CDI Words and Sentences "Words Produced" was the right choice for evaluating vocabulary vulnerability in children with permanent hearing loss (19–30 months). | 3 (15%) | 2 (10%) | 4 (20%) | 7 (35%) | 3 (15%) | 4 (1–5) | 4 (16.7%) |
| I felt the Expressive One Word Picture Vocabulary Test (EOWPVT-4) was the right choice for evaluating vocabulary vulnerability in children with permanent hearing loss (24–35 months). | 0 (0%) | 1 (6.3%) | 6 (37.5%) | 7 (43.8%) | 1 (6.3%) | 4 (2–5) | 8 (33.3%) |
| I felt the CELF-P2 Word Structure subtest was the right choice for evaluating grammar vulnerability in children with permanent hearing loss (3–6 years). | 0 (0%) | 1 (5.3%) | 4 (21%) | 10 (52.6%) | 3 (15.8%) | 4 (2–5) | 5 (20.8%) |
| I felt the CASL-2 Grammatical Morphemes subtest was the right choice for evaluating grammar vulnerability in children with permanent hearing loss (3–6 years). | 0 (0%) | 0 (0%) | 4 (66.7%) | 1 (16.7%) | 0 (0%) | 3 (3–4) | 18 (75%) |
| I felt the Goldman Fristoe Test of Articulation, Third Edition (GFTA-3) - Sounds in Words subtest was the right choice for evaluating vocabulary and syntax vulnerability in children with permanent hearing loss (30–48 months). | 1 (5.6%) | 3 (16.7%) | 3 (16.7%) | 7 (38.9%) | 3 (16.7%) | 4 (1–5) | 6 (25%) |

**Appendix I (cont.)**

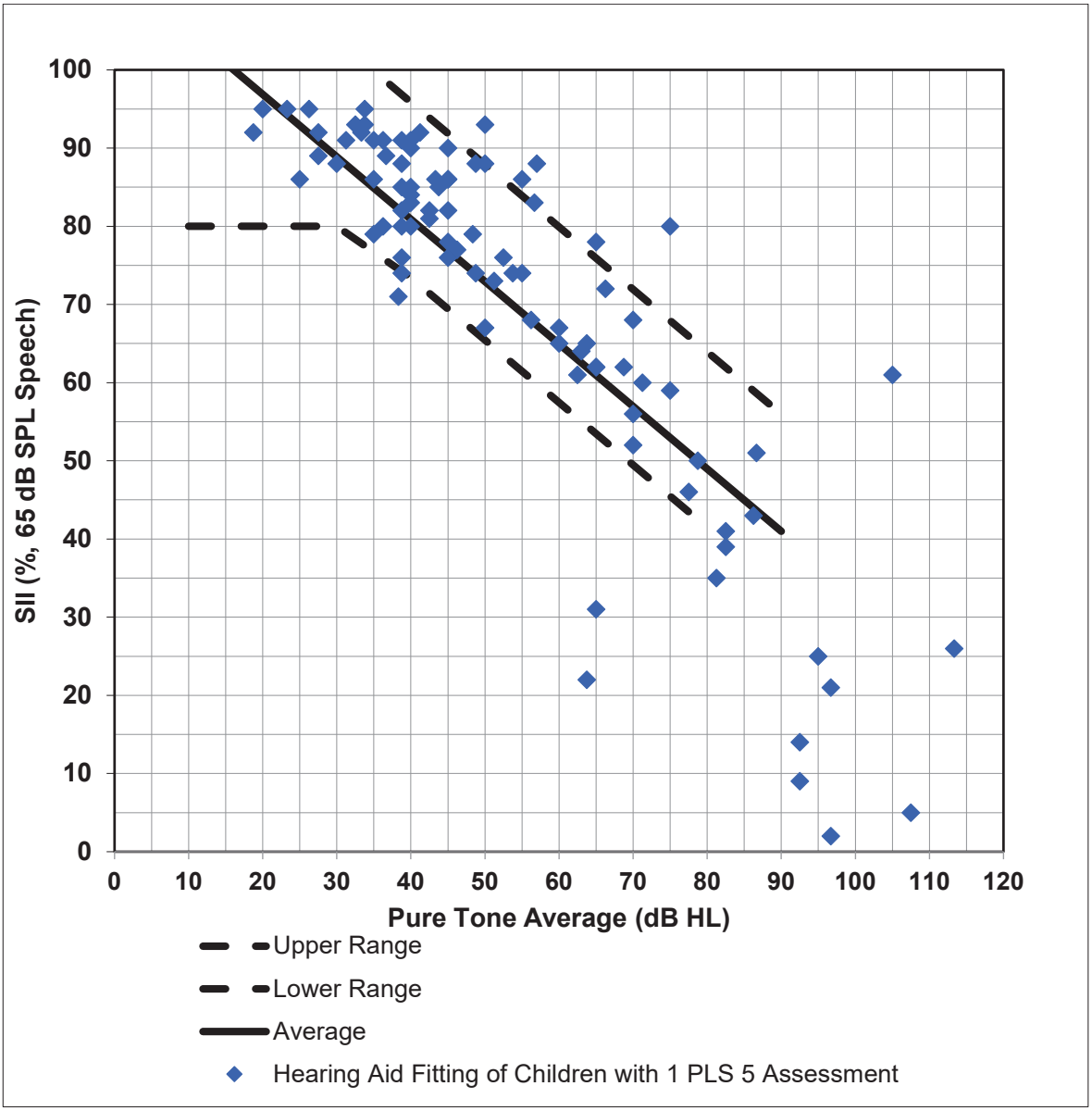
Speech Language Pathologists' (SLP) Perceptions of the Quality of the Tier 2 Procedure

| Statement | Strongly disagree *n* (%) | Disagree *n* (%) | Neutral *n* (%) | Agree *n* (%) | Strongly agree *n* (%) | Mode (range) | Not applicable *n* (%) |
|---|---|---|---|---|---|---|---|
| I felt the CELF-P2 Pre-literacy rating scale was the right choice for evaluating emergent literacy/ phonological awareness vulnerability in children with permanent hearing loss (4–6 years). | 0 (0%) | 1 (7.7%) | 6 (46%) | 5 (38.5%) | 0 (0%) | 3 (2–4) | 11 (45.8%) |
| I felt the CELF-P2 Phonological Awareness subtest was the right choice for evaluating emergent literacy/phonological awareness vulnerability in children with permanent hearing loss (4–6 years). | 0 (0%) | 0 (0%) | 6 (37.5%) | 8 (50%) | 1 (6.25%) | 4 (3–5) | 8 (33%) |
| I feel the implementation of IVT helped me to identify impairments in children with permanent hearing loss that were missed through Program Level Outcome Monitoring. | 4 (20%) | 2 (10%) | 1 (5%) | 10 (50%) | 2 (10%) | 4 (1–5) | 4 (16.7%) |

*Note.* IVT = Individual Vulnerability Test.

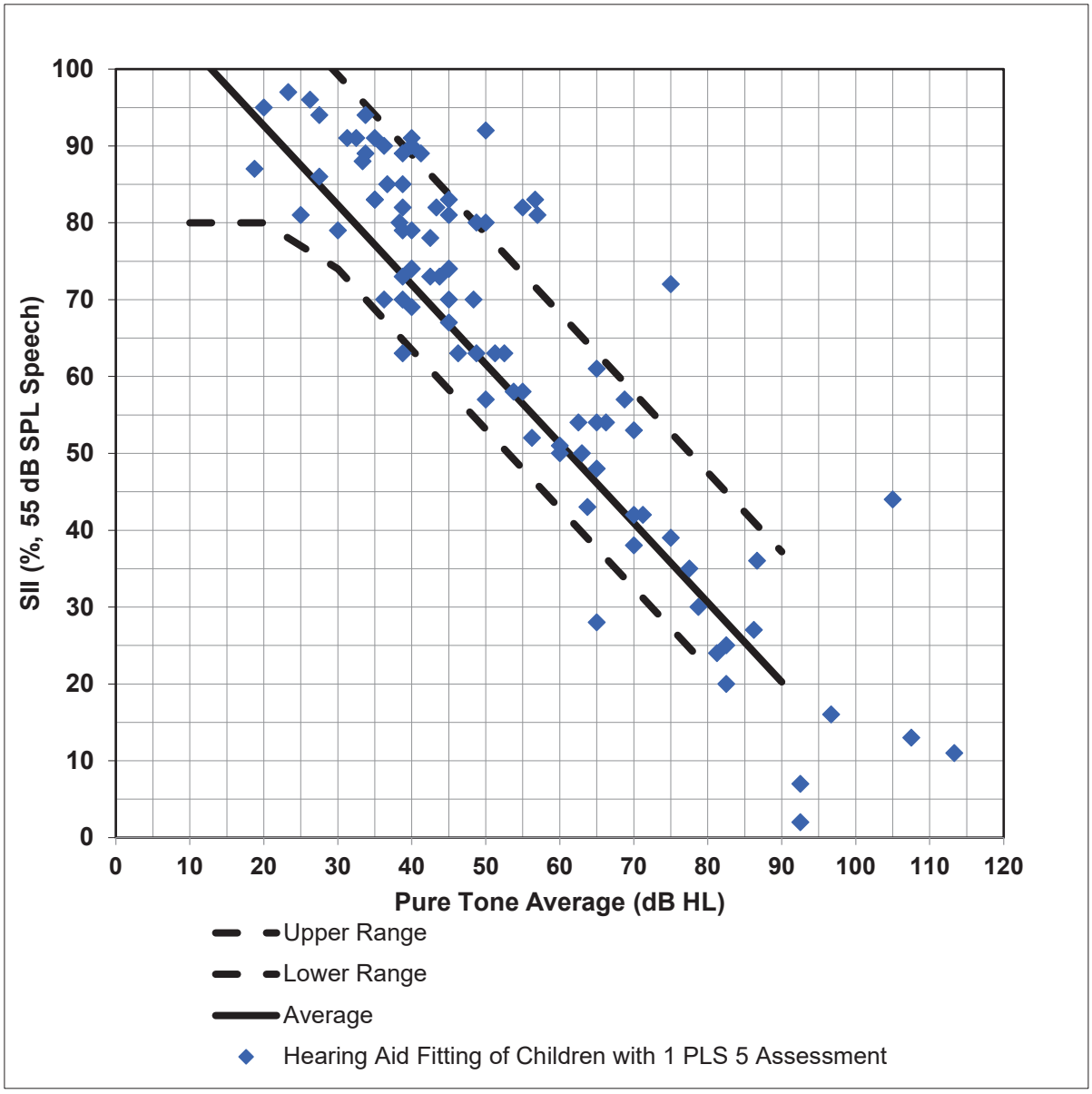
**Supplemental Material 1**

Hearing Aid Speech Intelligibility Index (SII) at 65 dB compared to Moodie et al., 2017 normative data: Children with data from the Preschool Speech & Language Scale (PLS), 5th ed



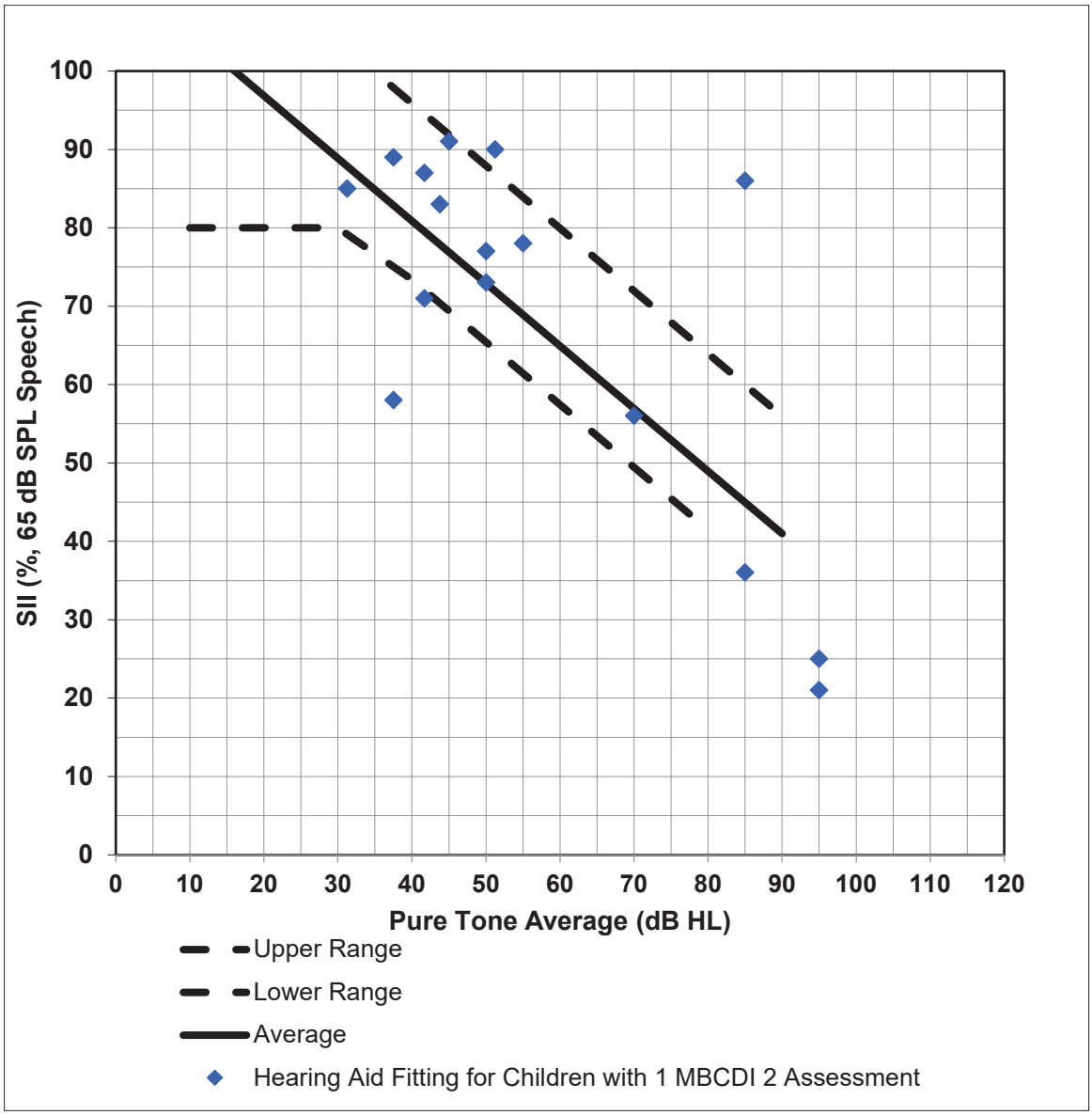
**Supplemental Material 2**

Hearing Aid Speech Intelligibility Index (SII) at 55 dB compared to Moodie et al., 2017 normative data: Children with data from the Preschool Speech & Language Scale (PLS), 5th ed



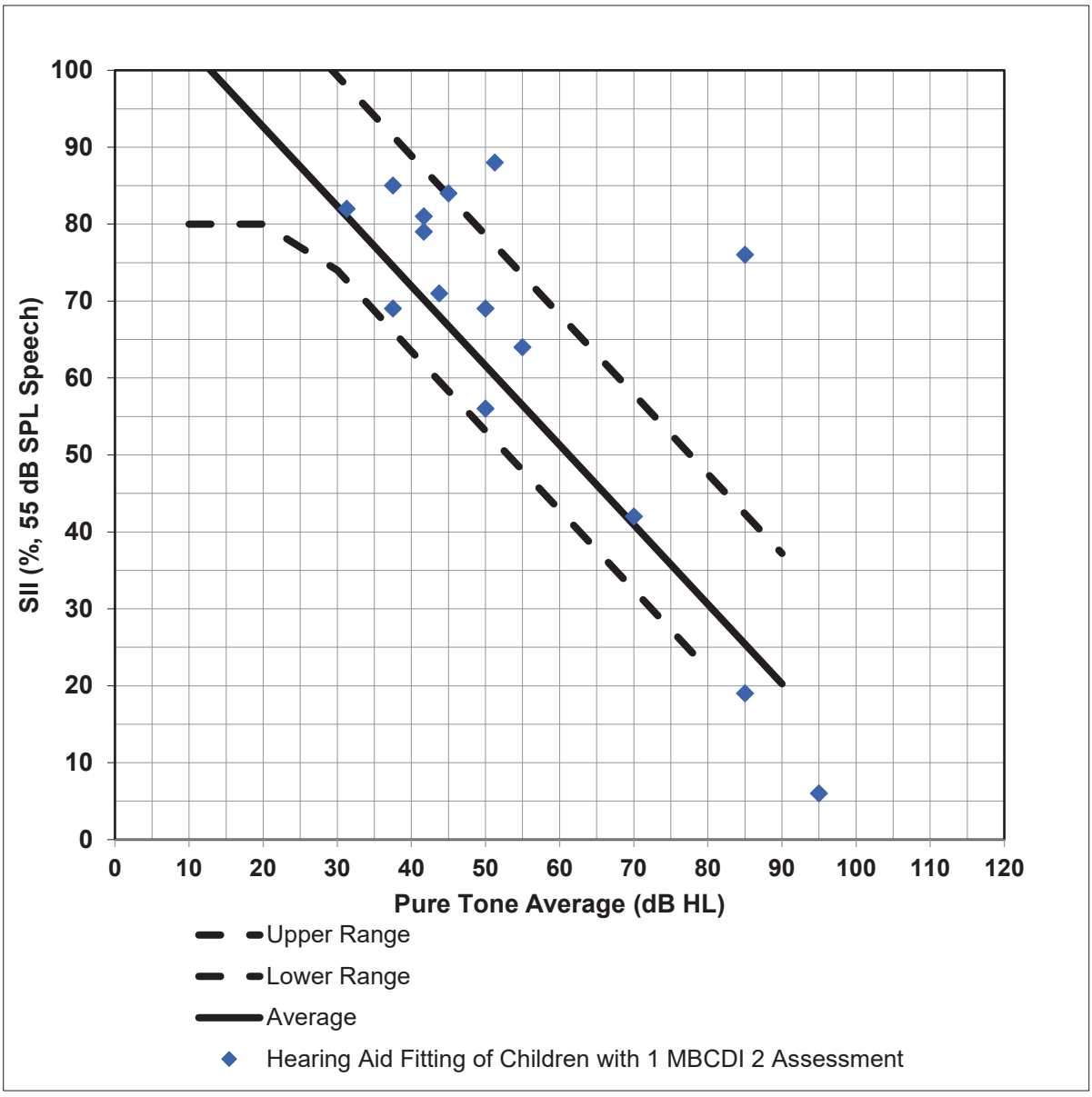
**Supplemental Material 3**

Hearing Aid Speech Intelligibility Index (SII) at 65 dB compared to Moodie et al., 2017 normative data: Children with data from the MacArthur-Bates Communicative Development Inventories, 2nd ed. (MBCDI-2)



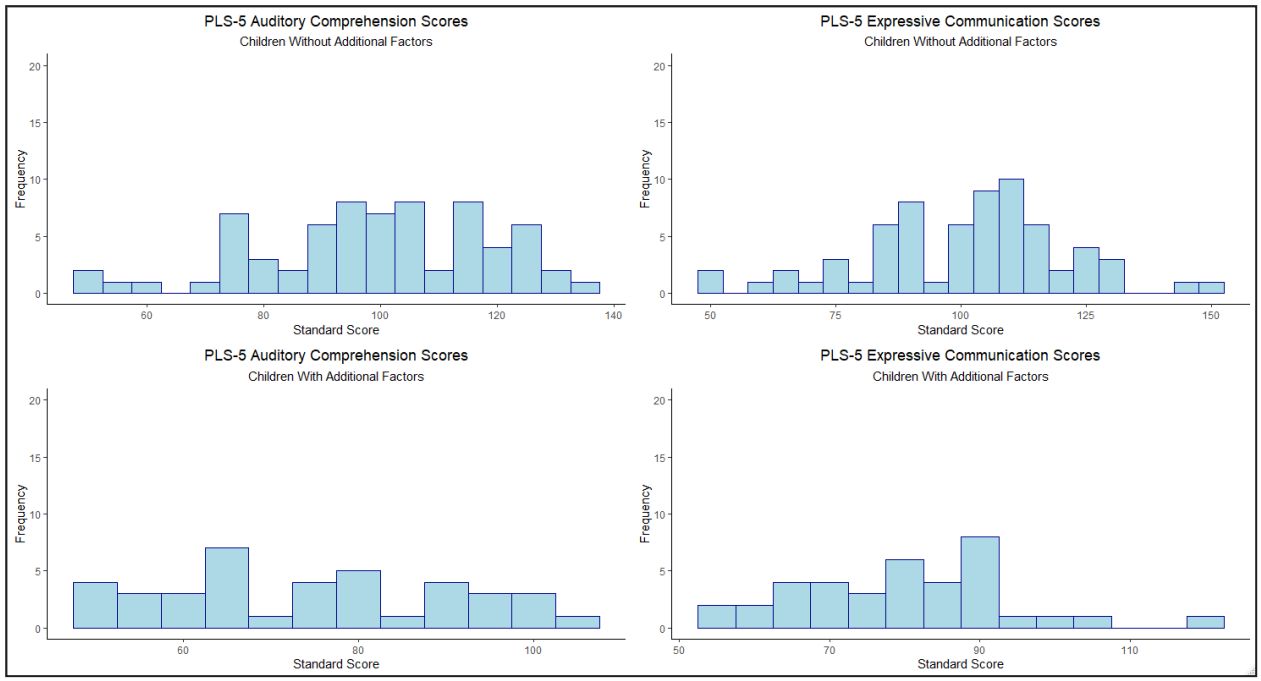
**Supplemental Material 4**

Hearing Aid Speech Intelligibility Index (SII) at 65 dB compared to Moodie et al., 2017 normative data: Children with data from the MacArthur-Bates Communicative Development Inventories, 2nd ed. (MBCDI-2)



## Supplemental Material 5

Distributions of Preschool Language Scale, 5th ed. (PLS-5) Scores



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# Outcome of a Newborn Hearing Screening Program in a Tertiary Care Center, South India

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## Abstract

**Objective:** Using data from a four year period, the authors evaluated the Universal Newborn Hearing Screening (UNHS) Program in the Government Medical College Manjeri, South India. The prevalence of hearing loss (HL) among screened children, description of confirmed case characteristics, and documentation of speech and language development acquired by children at follow up are discussed.

**Design:** Hospital based retro-prospective study.

**Method:** Data were collected from all newborns who underwent UNHS from November 2014 to October 2018. Confirmed HL cases were studied by pre-structured questionnaire and telephone interview. Speech and language assessments of 10 confirmed cases were conducted after an intervention period.

**Results:** 16,625 of 17,260 babies were screened (96.3%). Thirteen infants had confirmed HL (prevalence rate = 0.08%) and 61.5% of those with HL did not have risk factors. Median confirmation age was 6 months with an Interquartile Range (IQR 4–12). Median age of speech therapy and hearing aids was 17.5 months (IQR 13–25) and the median duration of intervention before assessment was 30 months (IQR 17–43). Three children were lost to follow up. The remaining 10 children received speech therapy; five children used hearing aids, five required cochlear implants at a median age of 24 months (IQR 17.5–33). Eight children showed a lag in speech and language development after assessment, with a median delay of 19.3 months (IQR 2–34.5).

**Conclusions:** Program coverage was optimal, with most newborns successfully screened. More than half of the confirmed children did not exhibit risk factors for HL and might not have been identified early without UNHS. The observed median age of starting intervention for confirmed cases was higher than the age recommended by AAP guidelines and most of the children had language development below those of children with typical hearing after months of intervention.

**Key words**: Universal Newborn Hearing Screening, outcome, otoacoustic emission, hearing loss, neonates

**Acronyms**: ABR = auditory brainstem response; DPOAE = distortion product otoacoustic emissions; HL = hearing loss; ISD = Integrated Scale of Development; REELS = Receptive Expressive Emergent Language Scale; UNHS = universal newborn hearing screening

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Hearing loss is one of the most common congenital conditions seen in newborns. One to three per 1000 newborns and 2 to 4% in Newborn Intensive Care Unit (NICU) have hearing loss (Dedhia et al., 2018; Erenberg et al.,1999; Parving et al., 2003). According to the World Health Organization, it is estimated that about 7.5 million children around the world have a significant problem with hearing (Dedhia et al., 2018). Hearing loss can have a great impact on a child's development. Failure to identify newborn babies with hearing loss early in life may result in delayed development of speech and language, poor academic achievements, and deficient social and emotional development (Haddad et al., 2019; Stevenson et al., 2010, Yoshinaga-Itano et al.,1998).

The peripheral auditory organs are completely developed at birth, but proper development of the auditory cerebral cortex requires appropriate sound stimulation especially in the first 2 to 3 years after birth. After this period, regardless of hearing rehabilitation, the brain's plasticity starts decreasing and the development of spoken language is limited (Ruben & Rapin, 1980). It has been shown that children diagnosed with hearing loss at an earlier age of about 6 months followed by early interventions including speech therapy, hearing aid, and cochlear implantation, often achieve normal or near normal spoken language development (Yoshinaga-Itano, 2004). Without universal screening for hearing loss, hearing problems may not be detected early for many children and once hearing loss is detected, it may be too late for them to receive optimal benefit from intervention.

Universal Newborn Hearing Screening (UNHS) helps to ensure early detection of hearing loss and to execute effective interventions as early as possible (Yoshinaga-Itano, 2003). The American academy of Pediatrics (AAP) recommends screening for hearing loss be completed by 1 month of age, confirmation by 3 months of age, and early intervention by the age of 6 months (AAP, 2010; Joint Committee on Infant Hearing [JCIH], 2007, 2019; Mehl, & Thomson, 2002). According to the JCIH 2019 Position statement, it is recommended that those states who have achieved the benchmark of 1-3-6 months, should try to achieve the 1-2-3 months timeline (JCIH, 2019).

For the last two decades, UNHS has been initiated in many countries in the world as a cost effective practice for standard newborn care (Korver et al., 2017). The implementation of UNHS in developed countries is extensive. Approximately 98% of newborn babies are screened in United States (Centre for Disease Control and Prevention, 2019). A retrospective study in England analyzing the screening for nine years showed that 98.9% of infants are screened by 3 months of age (Wood et al., 2015). Many studies conducted around the world have shown that UNHS helps in improving the early detection of hearing loss (Dedhia et al., 2018).

According to the World Health Organization (WHO), the majority of children who suffer from hearing loss are from developing countries of South Asia, Sub Saharan Africa and Asia Pacific (World Health Organization, 2018). African countries lack mandatory screening strategies at present and there is only 24% reported coverage for hearing screening (Theunissen & Swanepoel, 2008). Thus, implementation of UNHS in resource-constrained and developing countries is very important, but at the same time challenging. A staged approach should be adopted for implementation of universal screening by initially targeting the coverage of high risk groups, followed by universal screening (Das et al., 2020).

Nationwide UNHS is not yet started in many developing countries. In India, at present, it is done at sub-national level or district wise only (Singh, 2015) and not implemented uniformly across the country (Galhotra & Sahu, 2019). Another study revealed that only 38% of medical colleges in India have a newborn screening program (Kumar & Mohapatro, 2011).

In 2006, the Government of India launched the National Program for Prevention and Control of Deafness (NPPCD). The main goal of this program was to prevent and control hearing loss and to rehabilitate people of all age groups with hearing problems. Institution-based and community-based screening programs are being implemented in several districts of the country under this program (Galhotra & Sahu, 2019). Community-based programs are mainly targeting those babies born at home. Rashtriya Bal Swasthya Karyakram (RBSK) is another program launched in 2013 which is an important initiative involving child health screening and early intervention services for children 0 to 18 years of age for birth defects including congenital hearing loss, developmental delays, and other disabilities (Galhotra & Sahu, 2019).

A centralized screening facility for universal hearing screening was established in Cochin, Ernakulam district of Kerala, South India in 2003, which included 20 major hospitals (Paul, 2011, 2016). UNHS started in the Government Medical College Manjeri, Kerala in November 2014 and has been continuing successfully until now. The current study was planned to look at the coverage and gaps in implementation of the program, and provide corrective measures for improvement.

**Objectives**

The objectives of this study were to determine the coverage and the outcome of the Universal Newborn Hearing Screening Program at Government Medical College, Manjeri, Kerala, South India and to determine the prevalence of hearing loss among those babies who were screened. Secondary objectives were to assess the characteristics of confirmed cases, the interventions carried out, and the status of speech and language development after intervention had been initiated.

**Materials & Method**

This was a hospital based retrospective study. Distortion product otoacoustic emissions (DPOAE) was used for screening in our hospital. In DPOAE, frequency specific pure tone stimuli is delivered to the ear through the instrument probe. The frequency range of 2kHz to 5kHz was used. The model of instrument used in our hospital was Interacoustics Titan. A signal to noise ratio (SNR) of > 6 dB in 3 out of 4 frequencies tested was labeled as *pass*. Those cases who failed the screening were labeled as *refer*. For confirmation, we used Intelligent Hearing Systems (IHS) Solo ABR (auditory brainstem response). The stimuli used were clicks and tone burst at a rate of 11.1 and at 500Hz, 30dB nHL was taken as threshold for HL, and a filter of 30 to 3000Hz was used.

A trained nurse conducted hearing screening. The protocol followed for newborn hearing screening in our institution was to complete DPOAE for all newborns admitted in the hospital (both inborn and outborn) 24 hours after birth, but before discharge from the hospital. Those who passed screening with no risk factors for hearing loss (JCIH, 2019) were discharged. If they had risk factors, they were advised to repeat DPOAE every 6 months until 3 years of age.

The refer cases were called back at 6 weeks of age and the screening test was repeated. To decrease the dropout rate, the screening test was completed at routine immunization so that an extra hospital visit for hearing screening was avoided.

Those babies who failed the second screening test (refer) were sent to the Audiology Department of our institution for confirmatory test by ABR and once the hearing loss was confirmed, babies were referred to other facilities for specific interventions like cochlear implantation, which was not available in our hospital. The details of all the

confirmed cases, including the interventions done, were maintained by a separate registry in our hospital, and the Audiology Department followed up with them.

During the 3 month study period of October 2019 to December 2019, the authors collected data from babies born during a 4 year span (November 2014 to October 2018). The follow-up of confirmed cases was completed during the month of December 2020. Permission to conduct the study was obtained from the hospital ethical committee (Ref No: IRC/GMCM/33(2)). Data included total number of deliveries, screened babies, missed cases, follow up cases, total number of pass or refer cases, total number of high-risk babies screened and number of babies who failed the screening among high-risk groups. Details of the confirmed cases were collected from the hospital follow up registry, including phone numbers of the parents. The parents were contacted by telephone. The questionnaire included patient's demographic details, time of confirmation of diagnosis, and time and type of interventions and risk factors for hearing loss as per the AAP guideline (JCIH, 2019).

The cases were reviewed during the month of December 2020. There was a delay in getting these families to the hospital because of the prevailing Covid-19 pandemic. Children were assessed using the Integrated Scale of Development (ISD; Cochlear, 2010) and the Receptive Expressive Emergent Language Scale (REELS; Bzoch & League,1971; Nair et al., 2013) with the help of an audiologist. These scales were used to assess the speech and language development of children in the Audiology Department. The speech and language development of each child at the time of follow-up was documented.

ISD incorporates different stages of development of listening, receptive and expressive language, speech, cognition, and social communication. Using this scale, children were assessed to discover the language development achieved at the time of follow up. REELS assesses different aspects of linguistic behavior which include receptive language and expressive language. Receptive Language Age , Expressive Language Age, and Combined Language Age of each child was calculated with REELS and compared with the chronological age of the child. As per our institutional policy, any delay of more than 6 months from chronological age was taken as a significant delay.

The data collected were statistically analyzed with the help of a statistician. Outcome was measured in terms of coverage of screening, prevalence of hearing loss, and percentage of cases identified as refer cases at each screening. The prevalence of hearing loss among the study group was calculated. For confirmed cases, median age of detection of hearing loss, median age of diagnosis confirmation, median age of starting interventions, median age of cochlear implantation, and median age of delay in speech and language development were determined. Qualitative variables were summarized as frequency and percentages and presented in Tables 1 through 3. Quantitative variables were summarized as Median and Inter quartile range (IQR).

## Results

A total of 17,260 babies were born during the study period, out of which 16,625 babies were screened (96.3%) through the newborn hearing screening program in our hospital. Among 16,625 newborns, there were 1057 (6.4%) refer cases after the first screening. Out of these 1057 babies, 998 (94.4%) were followed up and 59 were lost in follow up. Out of 998 babies, the second screening yielded 16 (1.6%) refer cases. Three out of 16 refer cases subsequently tested normal by auditory brainstem response and 13 babies were confirmed to have hearing loss (0.08%). Eight out of 13 confirmed cases (61.5%) did not have any risk factors for hearing loss.

The main risk factors for hearing loss identified in this study are shown in Table 1 and included the following: family history of childhood hearing loss, NICU admission and use of aminoglycosides, and neonatal jaundice treated by exchange transfusion.

**Table 1**
*Risk Factors Identified in Confirmed Cases of Hearing Loss (n = 13)*

| Risk factor identified | Total no. | Percentage (%) |
|---|---|---|
| Family history of hearing loss | 2 | 15.4 |
| NICU admission | 2 | 15.4 |
| Newborn jaundice treated by exchange transfusion | 1 | 7.7 |
| No risk factors | 8 | 61.5 |

The clinical characteristics of the patients with confirmed hearing loss is shown in Table 2. The majority (84.6%) were full-term babies, with no gender preponderance. No risk factors for hearing loss were evident in 61.5% of cases.

**Table 2**
*Clinical Characteristics of Confirmed Cases (n = 13)*

| Characteristics | | Number (%) |
|---|---|---|
| Gender | Male | 6 (46.2) |
| | Female | 7 (53.8) |
| Birth weight | ≥ 2.5kg | 7 (53.8) |
| | < 2.5kg | 6 (46.2) |
| Gestational Age | Term (≥ 37wks) | 11(84.6) |
| | Preterm (< 37wk) | 2 (15.4) |
| High risk group | Yes | 5 (38.5) |
| | No | 8 (61.5) |

Median age of confirmation by ABR was 6 months (IQR 4–12). The lower age limit of confirmation was 2 months, and the upper age limit was 14 months. The median age of starting interventions like speech therapy and hearing aids was 17.5 months (IQR 13–25). The lower and upper age limit of starting interventions were 12 months and 26 months respectively. Median age for cochlear implants among confirmed cases was 25 months (IQR 17.5–33). The lower age limit was 17 months and upper age limit was 41 months. Median duration of intervention at the time of assessment was 30 months (IQR 17–43).

Five out of 13 children (38.5%) were managed by cochlear implant and speech therapy, free of cost using Government funds. Five children (38.5%) were managed by hearing aids and speech therapy only. Two children (15.3%) with mild hearing loss were managed by speech therapy alone. One child with global developmental delay, was bedridden and managed conservatively by physiotherapy alone, without any intervention for hearing loss.

Out of the 13 children with hearing loss, only 10 (76.9%) children turned up for review and language assessment. Those included four cases with cochlear implant, three cases with hearing aid and speech therapy, and two cases who received speech therapy alone. The remaining one child had global developmental delay along with hearing loss, and did not receive any type of intervention. Three were lost to follow up (23.1%). After assessing the speech and language, it was noted that eight children showed a lag in speech and language development as evidenced by a delayed combined language age. The language assessment using REELS and the ISD scale is given below (Table 3). The results of ISD were similar to REELS. There was a median delay of 19.3 months in language development (IQR 2–34.5).

**Table 3**

*Speech and Language Assessment in Confirmed Cases (n = 10)*

| Age | Sex | Diagnosis | Interventions Done | Assessment by REELS (months) | | | ISD (months) |
|---|---|---|---|---|---|---|---|
| | | | | RLA | ELA | CLA | |
| 5 years 4 months | M | B/L profound HL | Cochlear Implant | 33–36 | 33–36 | 33–36 | 31–36 |
| 3 years 5 months | F | B/L profound HL | Nil | 0–3 | 0–3 | 0–3 | 0–3 |
| 5 years 6 months | M | B/L profound HL | Cochlear Implant + Speech Therapy | 42–48 | 42–48 | 42–48 | 31–36 |
| 3 years 7 months | M | B/L severe-profound HL | Hearing Aid + Speech Therapy | 24–27 | 24–27 | 24–27 | 16–18 |
| 5 years 1 month | M | B/L mild HL | Speech Therapy | 54–60 | 54–60 | 54–60 | --- |
| 3 years 10 months | F | B/L profound HL | Cochlear Implant + Speech Therapy | 30–33 | 30–33 | 30–33 | 31–36 |
| 3 years 9 months | M | B/L severe-profound HL | Hearing Aid + Speech Therapy | 20–22 | 20–22 | 20–22 | 16–18 |
| 4 years 8 months | F | B/L severe HL | Hearing Aid + Speech Therapy | 18–20 | 18–20 | 18–20 | 16–18 |
| 6 years | M | B/L profound HL | Cochlear Implant + Speech Therapy | 42–48 | 42–48 | 42–48 | 31–36 |
| 1 year 9 months | F | (R) mild HL | Speech Therapy | 22–24 | 22–24 | 22–24 | 19–24 |

*Note.* HL = hearing loss; B/L = bilateral; REELS = Receptive Expressive Emergent Language Scale; RLA = receptive language age; ELA = expressive language age; CLA = combined language age; ISD = Integrated Scale of Development.

## Discussion

Coverage of the UNHS program in this study is 96.3% which is optimal per AAP guidelines. It is comparable to a study from Malaysia in which the coverage was around 98% (Ahmad et al., 2011). Like our study, Ahmad et al. (2011) was a hospital-based study and screening was done by DPOAE. But our coverage is higher compared to a study from China which was a population-based study in the rural areas of China where the coverage of screening was 89.2%. The screening method used in that study was transient evoked OAE (TEOAE; Guanming et al., 2012).

There was a high refer rate after the first screening in our study. This may be due to the fluid in the middle ear cavity (middle ear effusion) or residual debris which is normally seen in ears of newborns as observed in many other studies (Boone et al., 2005, Boudewyns et al., 2011). Referred cases were less in our study compared to the study from China (Guanming et al., 2012).

The prevalence of hearing loss in this study was low compared to the study from Germany which was a population-based study in which the prevalence was 2.32 per 1000 newborns (Rissmann et al., 2018) and also

compared to another hospital-based study from Benin city, where the prevalence was 6.5% (Amina et al., 2010). It was less when compared to the Hearing Screening and Follow-up Survey (HSFS) in United States, in which the prevalence rate was 1.7 per 1,000 babies screened (CDC, 2019). In our study we had 59 (5.6%) refer cases who were lost after the first screening. They might have done the repeat OAE at 6 weeks from their nearby hospital and been diagnosed as HL elsewhere. This could be a reason for low prevalence in our study.

The percentage of confirmed cases with no risk factors for hearing loss was 61.5%. In a similar study from the Ernakulum district, Kerala, India, only 29.6% cases did not exhibit risk factors (Paul, 2011). This was low compared to our study and shows the importance of universal screening of all babies early in their life for detection of hearing loss since hearing loss can be present without risk factors.

In our study there was a delay in the age of confirmation by ABR and age of starting intervention of confirmed cases when compared to the recommended AAP guidelines (JCIH, 2019). Similar findings were obtained in a study done from Saudi Arabia in which mean age of confirmation was 20 months and mean age of intervention was 25 months (Alshawi et al., 2019). But in a study from Germany, median age of confirmation was 3 months of age and median age of starting intervention was 4 months of age (Rissmann et al., 2018).

This delay in age of confirmation and age of starting intervention may be due to the inadequate human resources available for newborn hearing screening and follow up in the public sector and limited availability of screening equipment. More orientation programs for health workers and social workers are needed to educate stakeholders about the importance of proper follow up of confirmed cases. The success of any screening program depends mainly on the early intervention and management of diagnosed cases. Also, procurement of more equipment and timely maintenance of the same is very crucial. More detailed studies should be conducted to find out the exact reason for this delay.

The treatment and rehabilitation of all the confirmed cases were free of cost, under Government plans. Five children received cochlear implants, 5 received hearing aids and all these 10 were enrolled in speech therapy. Three children among the total 13 were lost to follow up. Of the remaining 10 children, eight had not achieved age-appropriate speech and language after a median duration of intervention of 30 months. There was a gross delay when compared to the Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study which included 470 deaf Australian children whose hearing loss was diagnosed by newborn hearing screening. In the LOCHI study, 72% of the screened group who received early intervention in the form of hearing aids before 6 months of age had better language outcome at the age 5 years (Ching & Leigh, 2020). Finally, in our study two children, who had mild hearing loss, received speech therapy alone and had normal speech. The remaining one child had global developmental delay; parents were not that keen and motivated to go for any treatment.

**Limitations of the Study**

Data was gathered from a single Centre; hence, the prevalence may not be a true representation of the population. Availability of single machine and single personnel for the screening was a major technical limiting factor for timely completion of the hearing screening.

**Conclusion**

The coverage of the newborn hearing screening program in our hospital was optimal. The prevalence of hearing loss in our study was 0.08%. The study highlights the importance of universal screening for hearing loss, because the majority of the confirmed cases in this study did not have the risk indicators associated with hearing loss. Children with hearing loss usually appear normal at birth without any complaints. They could be identified only because of the universal screening of all newborns. The study also emphasizes the importance of proper follow up of the confirmed cases as there was a time delay of about 12 months from the time of confirmation to the time of intervention. Also, eight children who had undergone interventions, did not achieve age-appropriate speech and language development. Thus, this study also emphasizes the importance of timely intervention following confirmation of the cases. We have to ensure adequate human resources and proper infrastructure. A multidisciplinary team of Neonatology, Pediatrics, Otorhinolaryngology, Audiology, Auditory verbal, and speech therapy should be available for different stages in the screening process and management to insure the timely identification, diagnosis, and management of children with hearing loss. Regular follow up of these children, including regular assessment of speech and language development, is also important.

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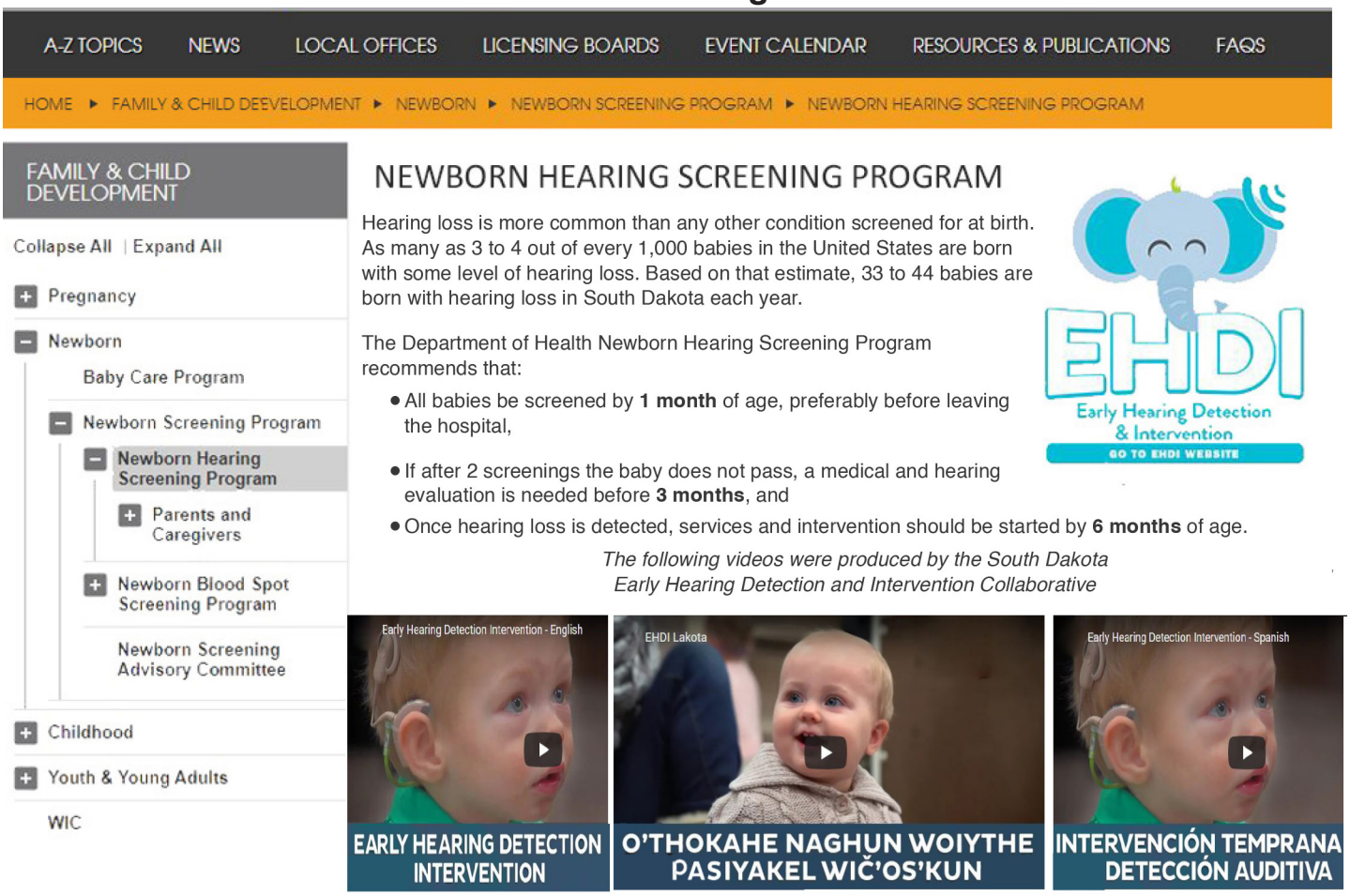

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# Deaf Early Intervention in Puerto Rico: A Qualitative Study

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## Abstract

Deaf children can develop similarly to hearing children with appropriate intervention. However, when Deaf and hard of hearing children have deferred access to services, they can experience significant delays in language, socioemotional development, and cognition that can lead to problematic behaviors. Although early intervention services are free in the United States starting at birth, there is often a lag in Deaf and hard of hearing children receiving services, especially when residing in U.S. territories such as Puerto Rico. The current qualitative study was to explore the lived early intervention experiences of three parents and three professionals of Deaf and hard of hearing children under the age of six years old. Questions explored the lived experiences and perceptions of both professionals and parents regarding their access and delivery of early intervention services in Puerto Rico for Deaf and hard-of-hearing toddlers. Several salient themes emerged to include support for sign language, barriers to services, and family support.

**Keywords:** Culture, Deaf, Early Childhood Development, Early Hearing Detection and Intervention (EHDI), Puerto Rico

**Acronyms:** ASL = American Sign Language; EHDI = early hearing detection intervention; IEP = individualized education plan; IFSP = individualized family service plan; IPA = interpretative phenomenological analysis

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Infants are born primed and ready to learn. However, research suggests that when an infant is born Deaf[1] or with a hearing status outside of a typical hearing range, parents may have a difficult time adjusting to their child's communication needs (Ebrahimi et al., 2017; Hardonk et al., 2011), and this may ultimately impact the child's language development. Early access to language is critical for linguistic, cognitive, and socioemotional development in infants and young children. Language provides children with opportunities to develop critical thinking skills and build socially reciprocal relationships. According to research on fetal and infant development, the auditory system develops by the 29th week of gestation (Graven & Browne, 2008), which means that most fetuses from this point forward have preliminary access to sounds including spoken language. However, infants who are born Deaf or with a hearing difference, experience various degrees of language deprivation because they have limited to no access to auditory language while in utero and for the early months to years of postnatal development.

The development of Deaf and hard of hearing children is influenced by early communication between parent and child. It has been estimated that over 90% of Deaf infants are born to hearing parents (Mitchell & Karchmer,

[1]To be inclusive of the heterogeneity Deaf people (e.g., Deafblindness or auditory access levels), the authors use the capitalization of the word Deaf to represent all identities, cultures, and medical experiences of people across Deaf communities. The authors are also using Identity First language as it is the preference of the Deaf Community.

2004). Many infants who are born Deaf or hard of hearing experience delays in language exposure when they are born into families who do not have fluency in a visual language such as American Sign Language (ASL; W. C. Hall et al., 2017). Deaf and hard of hearing infants and young children who do not have access to a full spoken language due to hearing differences have limited opportunities for incidental learning when compared to their hearing counterparts (Hauser et al., 2010), which can have a long-term developmental impact. Data on children born Deaf who gained access to language after the first year of life suggest that later language acquisition and challenges with fluency may lead to poorer developmental outcomes than Deaf children who had access to language at birth (e.g., children born to Deaf parents who were already fluent in a signed language; M. L. Hall et al., 2016; Netten et al., 2015). This delay in language exposure is referred to as language deprivation (W. C. Hall et al., 2017).

Screening and detection of a hearing difference is vital for understanding early intervention for Deaf children. In the United States, currently 43 states and the District of Columbia and Puerto Rico have mandates and guidelines for when hearing screening should occur. The Joint Committee on Infant Hearing (JCIH) recommends newborn screening at birth. If the inpatient screening detects a hearing difference, the newborn is then referred to an outpatient re-screening to be completed within a month (JCIH, 2019). If anomalies are found in the re-screening, the infant is then referred to complete an outpatient audiological evaluation by three months of age. Subsequently, JCIH (2019) suggests that early intervention services be implemented in a family-centered manner before the infant turns six months old. The Early Hearing Detection Intervention (EHDI) project reported 2019 data from 49 states and 7 U.S. territories to the Centers for Disease Control and Prevention (CDC, 2019). From the total births, 98.4% ($n$ = 3,545,388) of all newborns had documented hearing screenings. From these, 1.7% ($n$ = 65,475) of infants were referred to be further screened with 9.7% ($n$ = 5,934) being diagnosed with a hearing difference. Upon the infant being diagnosed with a hearing difference, 84.8% ($n$ = 5,034) of these families were referred to early intervention services; only 61.7% ($n$ = 3,662) enrolled in services.

Research has found that some parents of Deaf or hard of hearing infants exhibit emotional distress when they first learn of their child's diagnosis (Hardonk et al., 2011; Quittner et al., 2010; Zaidman-Zait et al., 2016). Because Deaf and hard of hearing children present with unique developmental needs and considerations in the areas of identification, diagnosis, and intervention, their parents have to learn to navigate services and programs that may have otherwise been foreign to them. Thus, parents of Deaf and hard of hearing children are thrust into learning about what it means to be Deaf from a variety of conflicting perspectives including cultural, medical, and federal (Flaherty, 2015; Luckner, 2011; Zaidman-Zait et al., 2016). In addition, since differences in hearing status are a low incidence diagnosis (Institute on Disability, 2019), sometimes general practitioners and mainstream early childhood care providers are unaware of the needs of Deaf and hard of hearing children (Flaherty, 2015).

There is a significant gap in the early intervention literature examining the needs of Latinx[2] Deaf and hard of hearing infants and appropriate service delivery to their families. More specifically, there is a lack of narrative, representation, and perspectives including Puerto Rican families with Deaf and hard of hearing infants and early intervention providers. Therefore, it is likely that Deaf and hard of hearing children and their families receive services that are both culturally biased and exclusive of research containing Deaf intersectional communities.

Early intervention programs rely on evidence-based practices but more research needs to be conducted to consider the impact of services on children and families from diverse cultural backgrounds. For example, although Puerto Rico is a territory of the United States that adheres to federal laws, such as the Individuals with Disabilities Education Act (IDEA), the unique cultural needs of Puerto Rican children and their families, such as family structure, are not considered when implementing early intervention programs. The Health Department of Puerto Rico, Law #311 (P. del S. 2404) established in 2003, states that children who have a hearing difference must be screened, diagnosed, and provided with early intervention by six months of age (LexJuris, n.d.a). However, many Latinx families who have a Deaf or hard of hearing child, including Puerto Rican families, move to the U.S. with hopes of high-quality services for their child with a hearing difference because Puerto Rican early intervention services are not currently meeting the needs of children and families (Steinberg et al., 2003). This is significant because it is estimated that between 135,000 and 185,000 individuals in Puerto Rico are Deaf or hard of hearing with limited access to high-quality services (Quintero, 2013). Yet, limited research among Puerto Rican families with Deaf and hard of hearing children makes it difficult to ascertain the strengths, challenges, and needs of this population.

Although Puerto Ricans have a strong adherence and respect for their own culture, they are at times heavily influenced by U.S. customs and behaviors due to Puerto Rican territorial status, required bilingual education, and federal laws (Capielo Rosario et al., 2018). Therefore, considerations of Puerto Rican families who have Deaf and hard of hearing children should support a bicultural perspective. Latinx hearing families tend to embrace multilingualism, such as teaching Spanish and other native languages and passing down Latinx customs and traditions to their Deaf Latinx children (Lopez, 2014). These bicultural and multilingual values could be used when

[2]The authors used the gender expansive term, Latinx. It is understood that there is currently no consensus on the use of this term. Therefore, Latinx should be complementary to other ethnic identities like Latine, Latina, Latino, or Hispanic (Mora et al., 2022). The authors understand that the 'x' violates Spanish orthography; however, it is commonly used to represent all genders, and has been seen in Puerto Rican scholarly work as a gender expansive term (Logue, 2015).

working with Puerto Rican families with Deaf and hard of hearing children. For instance, this view of biculturalism may be demonstrated when the Deaf child exists in a family that has a balanced perspective of both Deaf and hearing cultures. Families that adopt a bicultural model for their Deaf or hard of hearing child, tend to both embrace ASL through formal language acquisition, while also supporting written English (Gravel & O'Gara, 2003). In this perspective, families encourage interaction within the Deaf and hearing communities for their children and may also choose to use assistive technology devices (e.g., hearing aids, cochlear implants, bone conduction hearing aids).

There have been few studies examining early intervention in Puerto Rico among families with Deaf and hard of hearing children. For example, Pérez Rodríguez (2014) found that (a) families supported assistive technology with the hope that their children might be able to speak, and (b) families with Deaf children tend to have high expectations for their children's ongoing usage of cochlear implantation and speech. Families who seek services, whether for children ages birth to 3 years old (early intervention) or ages 3 years old and up, tend to establish a good relationship with professionals. However, many professionals do not provide families with a variety of communication alternatives, potentially leaving parents with limited knowledge about what it means to have a child with a hearing difference (Pérez Rodriguez, 2014). The same is true for the Puerto Rican Department of Education such that families view the Puerto Rican Department of Education as providing them with very limited services and information regarding what to do about their children's diagnoses (Marrero Vélez, 2014). In contrast, Marrero Vélez explored the perspectives of health professionals in Puerto Rico and found that although they are often lacking information when it comes to comorbid diagnoses, like Deafblindness, families continue to feel supported by them regarding guidance for Deaf children. With this information in mind, the current study explored how professionals and parents of Deaf and hard of hearing children view, navigate, and experience early intervention systems in Puerto Rico.

## Method

A misconception in research is that philosophical stance does not matter when deciding how to conduct research. Contrary to this misconception, philosophical stance directly influences scientific methodology. The reported study was conducted in Puerto Rico and rooted in a transformative paradigm. A transformative stance allowed for the investigations of marginalized communities and for an increase in awareness of social justice and human rights topics with the research (Mertens, 2009). Moreover, proponents of transformative research explain that this stance allows for addressing societal problems; issues of power, discrimination, and oppression; and allows for changes in society (Mertens, 2009). The current study addressed the transformative paradigm by disseminating information about the perceptions of early intervention status in a marginalized population within the United States. This form of research warranted for the investigators to refer to various assumptions: the ethical nature of the research (axiology), the perception of reality of the research (ontology), the understanding of the relationship between the researcher and participants and the understanding of the knowledge (epistemology), and the approach to how the research will be conducted (methodology).

As it pertains to the assumptions, the researchers followed principles of respect, beneficence, and justice to the community researched (Mertens, 2019). The authors unpacked and recognized their relationship with the Deaf community and Puerto Rican community. The investigators also maintained that there are many realities and perspectives to the research. For example, while paper-pencil questionnaires provide a quicker and more removed data collection experience for the researcher, face-to-face interviews with marginalized communities can often serve as a more human approach to data collection. The methodology selected was a qualitative approach based on the lack of literature in this community, the importance of having and maintaining an egalitarian relationship with the participants, and valuing the active involvement of participants in theme development. Finally, this approach prevented data manipulation by the authors, and it allowed for the data to be clearly understood during the analysis phase.

### Research Questions and Procedures

The research was reviewed and approved by the Institutional Review Board at the authors' institution. The primary author also applied and received a small grant to fund travel costs and participant stipends. The purpose of the current study was to better understand early intervention services in Puerto Rico by answering the following two research questions:

1. What are the perspectives and experiences of Puerto Rican parents accessing early intervention services for their Deaf and hard of hearing child?
2. What are the perspectives and experiences of Puerto Rican providers regarding early intervention services in Puerto Rico?

#### *Data Collection*

Data were anticipated to be collected via three sources: interviews of parents, interviews of professionals, and participant journals (see Appendices A and B for the Semi-structured Interview Guides). However, none of the participants completed their journals. As part of the transformative paradigm, the researchers followed up with the participants regarding their journals on two occasions across three months to no avail.

#### *Recruitment and Participants*

Recruitment was completed via social media, provider referral, and word of mouth. Eligibility criteria included the following: all parent participants had to be residents of Puerto Rico, have a Deaf child between the ages of one to five years old, and receive early intervention services. Providers had to provide early intervention services to Deaf children ages one to five years old in any discipline.

Six participants joined the study; three parents and three providers. Three parents had Deaf children under the age of five who received early intervention services in the island within one to two years of the interviews. One parent lived in an urban area of Puerto Rico and two lived in rural parts of Puerto Rico. One of the parents had a master's degree and two of them held high school diplomas. One parent self-identified as trilingual (Spanish, English, and ASL), another parent self-identified as bilingual (Spanish and ASL), and the last parent self-identified as monolingual (Spanish). Regarding socioeconomic status, two of the families self-identified as being of low socioeconomic status and one of the families self-identified as middle class. All of the parents in the study were married and identified as cisgender women.

Three professionals (an audiologist, a teacher/educational therapist, and a special education teacher) who provided early intervention services in Puerto Rico participated in the study. Two worked in urban cities of Puerto Rico and one lived in a rural town of Puerto Rico. The two educators held master's degrees in education/pedagogy and the audiologist held a doctorate in audiology. All professionals self-identified as trilingual, cisgender women, and married. The researchers paid participants $20 USD at the conclusion of the study for their participation. Phenomenological research suggests no minimum number of participants with research ranging from 1 to 325 participants (Creswell & Poth, 2018).

### *Phenomenological Analysis and Approach*

The researchers used interpretative phenomenological analysis (IPA) to analyze the data. This methodological analysis is experiential in nature, and it is used to learn what each participant is thinking about through various perspectives not limited to affective, cognitive, physical, and societal (Smith et al., 2009). As a tenet of IPA, the authors used thematic analysis to make sense of the participants' experiences and to find general commonalities.

Participants selected their preferred location for their face-to-face interviews. The primary researcher, who is a native Spanish speaker from Puerto Rico, conducted all interviews in Spanish. The researcher is also a certified trilingual (i.e., Spanish, ASL, English) interpreter, and has training and experience in language translation. The researchers conducted and recorded semi-structured interviews that lasted 45 minutes to 70 minutes. Post data collection included the interviewer listening to each recording once prior to analysis as the first step to experience the complete narrative with suspended judgment. The researchers used Dedoose version 8.0.35 to analyze the data (Dedoose, 2018), and they coded directly in the audio stream without transcribing separately to ensure fidelity to the Spanish language prior to translation. The native Spanish speaker author translated all of the selected quotes from Spanish into English.

The first step to coding included exploratory comments throughout the data. This notation allowed the researchers to highlight anything of interest (Smith et al., 2009). These comments allowed for deductive and inductive commentary to identify rich points of the data (Mertens, 2019). During this phase, the researchers observed the participants' language use, their concerns about their experiences with early intervention, and the associated themes.

The researchers maintained data integrity through the process of epoché, which allowed for the researchers to analyze their preconceived notions. The epoché initially allowed the participants to gain an understanding of the preconceptions, reducing as much bias as possible. The primary author used journaling as a tool to monitor prejudice, favoritism, and bias. Upon completion of journal entries, the researcher discussed self-reflective assumptions identified in the journaling process with an expert early intervention researcher in Deaf and hard of hearing populations with more than 20 years of research experience. The process allowed for an additional layer of ethical rigor as a technique of credibility for research trustworthiness.

Moreover, after each interview, the researchers bracketed powerful recollections that occurred during the interview with the participant interviewed. Again, this process was ongoing and continued to take place until the data was fully analyzed. This part of the analysis also served as a criterion for quality, allowing researchers to monitor subjectivity as an ongoing process by using notes. Through member checks, which included sending typed transcripts of the interviews to participants one to three months post interview via electronic delivery, all participants accepted and approved their interview transcripts as transcribed by the Spanish-speaking researcher.

## Results

Three major and two minor themes emerged during the analysis of the data (see Figure 1). The analysis also identified several subthemes under the category of *Barriers to Services*. In the following section each theme is described and supported by selected direct quotes from the interviews.

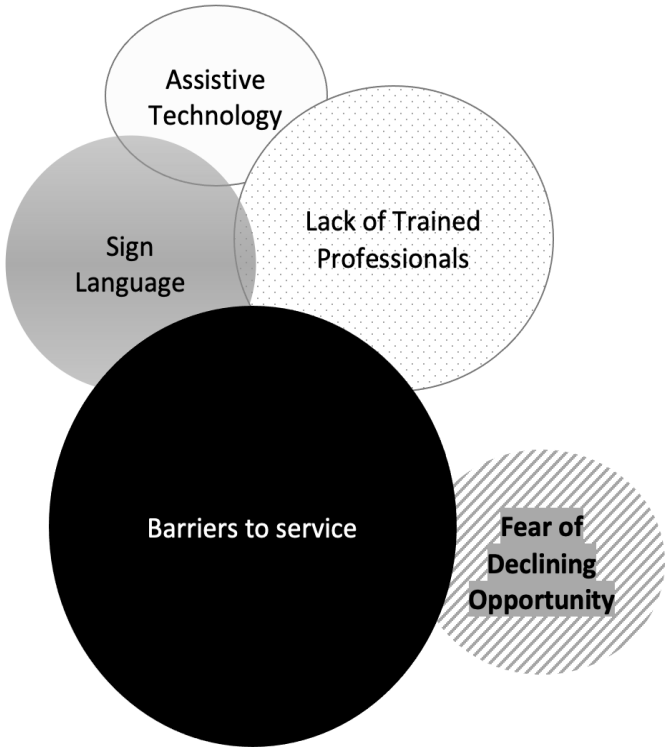
### Major Theme 1: Barriers to Services

The most prominent theme that emerged in the data suggested both parents and professionals experienced barriers in early intervention. Despite both groups experiencing difficulties, each group reported distinct barriers based on their specific role as either a caregiver or as an early intervention specialist.

### *Accommodations*

A shared concern between groups was the overall lack of accommodations for toddlers and young children in early intervention. Both parents and professionals discussed frustrations with advocating for interpreters in educational programs and often not having an individualized family service plan (IFSP). In fact, none of the parents interviewed in this study reported having an IFSP for

**Figure 1**
*Barriers to Service*




their child and the professionals shared that it was a rarity for Deaf and hard of hearing children to receive an IFSP. Parents and professionals reported that most Deaf and hard of hearing children who are eligible for early intervention services may attend an early head start program without appropriate communication access, such as an interpreter.

> Parent 2: *It is really sad that my child was placed in Head Start. No one knows what to do with a Deaf child in Head Start here. People do not know what to do. There is no interpreter, no language, no access! At one point, I became my own child's assistant in school.*

> Professional 2: *The established educational and legal system are hindering Deaf children's development in Puerto Rico. There are no interpreters or accommodations provided to families and children. When I became an itinerant teacher for children from ages 0-5, I told the parents what was going on. I explained the importance of IFSP and IEP [individualized education plan] to parents because the Department of Education here in my opinion takes advantage of families.*

### *Family Financial Burdens*

Another significant obstacle identified by parents of Deaf and hard of hearing children was financial hardships. Families had to make major financial decisions, such as having to sell some of their assets (e.g., houses, cars) to defray the expenses incurred in their children's medical appointments and other needs. These out-of-pocket costs were often made in private healthcare and educational settings to avoid being placed on long waitlists in government-sponsored programs. Moreover, data revealed that Puerto Rican family values of caring for their children was of most importance, which resulted in two of the mothers having to quit their jobs to care for their young children.

> Parent 2: *Many doctors in various specialties do not accept the government insurance I had, or they have longer wait lists for people like us with government-sponsored insurance. I ended up selling my house because we could not afford the doctors and I was concerned that something could worsen with my child.*

### *Advocacy*

Families reported challenges understanding how to appropriately navigate educational and healthcare systems and finding appropriate, high-quality services near their home. Parents reported frequently encountering inflexible government schedules that did not align with their child's or family needs. Furthermore, only parents with strong advocacy skills and those who were knowledgeable about their children's rights were able to access ongoing early intervention services and local educational programming.

> Parent 1: *They wanted to only offer speech services. I called my local early Head Start and the school complained about my child [being Deaf]. They said they had no service. I was then told to call this lady in a private Deaf preschool program. I called immediately, and they said you need to call the Department of Education for permission. I went to the Department of Education and got her enrolled really fast.*

### *Government Funding*

Early intervention specialists discussed how the current sociopolitical climate in Puerto Rico was what primarily impacted early intervention services on the island. All professionals mentioned the lack of governmental funding for Deaf specific early intervention and educational programs. Additionally, professionals explained that all Deaf services in Puerto Rico are currently private or government-subsidized, which contribute to the difficulties in families obtaining timely services. Furthermore, these professionals also disclosed how well-established Deaf programs on the island have dwindled in number due to the general lack of support from administrators and their misunderstanding of the needs of Deaf infant and toddler programs. Professionals mentioned how training and workshops for professional development in Deaf early intervention are inaccessible in the workplace also due to limited government funding.

> Professional 2: *The Department of Education is awful right now. We do not have any public Deaf schools in Puerto Rico. That is horrible and everyone is being mainstreamed. That hinders development on so many levels.*

> Professional 3: *When I started teaching, we had a Deaf preschool here... As numbers and funding went down, the Deaf preschool and program were eliminated. The problem with this is that a regular mainstream teacher is the one providing work and accommodations to the Deaf kids in our school with no one specializing in Deaf education, not even me because there is no Deaf program... The point is Deaf kids on the island are all over the place and no one is supporting them.*

The group of professionals also unanimously discussed how integrated programs are grouping Deaf early intervention services with early intervention services for other populations that may have distinct needs such as children with neurodevelopmental disabilities (e.g., autism, intellectual disability) or sensory disabilities (e.g., blindness).

> Professional 3: *My master's degree is in special education for children with autism, so they moved everyone into my autism specific classroom. So, kids are all over the place and no one is supporting them.*

**Major Theme 2: Sign Language**

All participants discussed their perspective on sign language. The majority of participants reported being a proponent of sign language. No one was against the use of sign language, but some reservations were made by one of the parents.

Two of the three parents reported using sign language with their children through total communication at the time of the interview. They catered to their child's preferred method of communication which included using a combination of speech, sign language, or both speech and sign language (SIMCOM). These two parents also reported being well integrated with the Deaf community in Puerto Rico after the diagnosis of their children. The third parent reported considering sign language with her child. However, she indicated that the biggest concern is her lack of knowledge of sign languages and how they may further hinder speech development. She indicated that her child's audiologist has suggested the use of sign language and was looking for sign language classes for her and her child despite her fear.

> Parent 3: *My speech-language pathologist wants me to learn sign language. I am unsure if I will teach him sign language because I think that would be good for him. However, if I teach him sign language, would he keep learning spoken language? The audiologist says sign language is the way to really go with him, so that he could have both. I can do both. At home, we communicate well, he communicates with his own signs with our family. He hasn't learned sign language, but hopefully he will.*

All professionals reported supporting the use of sign language with Deaf children. Two of the early intervention providers discussed how making sign language an official language in Puerto Rico might help develop better programs for Deaf children in early intervention centers and in public schools. The professionals also discussed the importance of how sign language can be used as a foundational language and a building block for spoken and written languages, such as Spanish or English. Furthermore, they expressed how the lack of early language exposure can cause delays in language, cognition, and socioemotional development. Sign language was framed as an accessible language that supports typical development. Lastly, the professionals specified the need for more professions and families to serve as sign language models for Deaf infants and toddlers.

> Professional 2: *When they go to first grade… Their role model in sign language is from the interpreter. That is also not appropriate language development for them… In public schools, we are seeing how a child just learns language from one person their whole life...This is a problem affecting Deaf culture because children no longer have access to their Deaf peers and teachers who know sign language. We are starting to close down schools for the Deaf or Deaf-specific programs without other avenues to facilitate Deaf culture.*

**Major Theme 3: Lack of Professionals Trained in Working with Deaf Communities**

All participants discussed the struggles they faced finding well-trained professionals in Deaf and hard of hearing practices. Parents reported that they want to have accessible early intervention services in sign language for their children, healthcare providers who know how to work with culturally Deaf young children, and educational staff who are competent in the area of hearing difference and sign language.

> Parent 1: *I keep fighting with the early intervention specialist because I request specialists that know about Deaf culture and ASL. The problem is that so many people do not know ASL. What if my daughter chooses to only sign? What if her hearing aids do not work? I have appealed and requested ASL fluent professionals. I need competent individuals. I have been waiting for a year!*

Professionals discussed that not having a wide pool of professionals whether early interventionists, educational staff, or health care providers impact the continuity of

services and sociocultural development of Deaf children. Further, the professionals discussed that having providers working with Deaf children who are not specialists in this area may lead to misdiagnosis, either over pathologizing or missing weaknesses. Most professionals raised the importance of at least speech-language pathologists, pediatricians, or teachers in being trained in cultural Deaf practices and being proficient in sign language to assist with adequate referral sources for services.

> Professional 1: *Pediatricians are the medical home for Deaf children. They need to learn how to work with Deaf families. They cannot use the same skills they use with children with autism and intellectual disabilities. This is different [for Deaf children] because they are needing to focus on attachment, development including language, cognitive, and social aspects. Again, these medical doctors focus only on the physical part. Deafness does not make [someone] a disabled person unless healthcare and educational providers hinder development, [thus] making [someone] disabled.*

**Minor Theme 1: Use of Assistive Technology**

Assistive technology presented as a less saturated theme despite it being generally endorsed by all parents and professionals. Although all parents considered cochlear implants for their children, ultimately, they elected for their children to have hearing aids. Parents' health literacy varied greatly on the topic of assistive technology. They preferred approaches including hearing aids, sign language, and speech/language therapy; in some cases, based on the belief that the time invested in cochlear implantation and habilitation could be better allotted toward allowing children access to the Deaf community.

> Parent 2: *My daughter has been very successful using her hearing aids. She can speak clearly and can hear some. She loves music and watching tv with what she can hear. I do have a big concern with hearing aids and that is with the financial aspect. Here in Puerto Rico, audiologists charge a lot for hearing aid appointments. I was lucky that I befriended an audiologist [who] gave me a discount. I considered a cochlear implant for my daughter, but I don't think she will benefit from it at this point. However, professionals really would like for my daughter to get one.*

All professionals supported children using assistive technology whether hearing aids or cochlear implants. Early interventionists believed that using assistive technology with sign language allows for optimal social and linguistic development.

> Professional 2: *I think that hearing aids and cochlear implants are crucial for Deaf children. Using technology with sign language will only maximize the child's development. However, I want parents to know of all the options they have.*

**Minor Theme 2: Fear of Declining Opportunity**

Worry for future discrimination emerged as the second minor theme for parents. Parents discussed their fears regarding having their children grow up and face discrimination by the larger society due to their hearing difference. Most of the fear stemmed from how others will perceive Deaf and hard of hearing children in Puerto Rico. However, one parent expressed concern of an inability to parent her Deaf child through later developmental stages.

> Parent 1: *My worries for her in the future is that society doesn't open their minds. That she will be shunned and marginalized because she is Deaf. I would hate if she did a job interview and prejudice takes over the interviewer thinking that she has intellectual deficits. I do not want people to discriminate against her. I want her to be happy! I have been teaching her that everyone is different. I tell her you are Deaf, and I am fat. People will judge us but you can still do anything you set your mind to. It can be hard feeling like you are the only one like you.*

On the other hand, professionals worried about the future of their professions. Sociopolitical issues were highlighted at the government level (e.g., senators and legislators not supporting Deaf rights), professional level (e.g., lack of advocacy within the field of early intervention), and the individual level (e.g., families demanding rights).

> Professional 3: *I am not sure what will happen to our profession. If it were for me, I would start the Deaf education and Deaf early intervention training again.*

## Discussion

The current study explored provision of early intervention services for Deaf and hard of hearing children in Puerto Rico from both parent and professional perspectives. The researcher maintained validity of this study by conducting a one-step member check process, involving a qualitative peer researcher, journaling, and by having a native Spanish speaker author from Puerto Rico, who is a nationally certified ASL interpreter, and a trained Spanish to English translator. These steps allowed the findings to be aligned with participants' intended expressions.

Participants in this study varied in terms of socioeconomic status, educational background, and understanding of what Deaf and hard of hearing infants, toddlers, and preschoolers need for early intervention. However, they shared many experiences and perceptions of what it means to have a child with a hearing difference in Puerto Rico or being an early intervention service provider of Deaf and hard of hearing children. Both parents and professionals identified prominent themes regarding accessing Deaf-specific early intervention, sign language,

and lack of trained professionals. Moreover, a couple minor themes emerged including lack of assistive technology and a fear of declining opportunities in the Deaf community and profession. All presented themes were related to developmental, family, and cultural needs in Puerto Rico.

Both parents and professionals discussed the multitude of barriers to receiving early intervention services for infants, toddlers, and young children in Puerto Rico and fear of declining opportunities for children and professionals. Public accommodations and appropriate placement for children were highlighted as a significant concern including the lack of access to language via an interpreter or a provider fluent in sign language. These findings are similar to the results from the Gerner de García and colleagues (2011) study that highlighted the lack of qualified early intervention professionals in Puerto Rico to work with young Deaf children. Many of these barriers would be nullified if agencies followed federal and local guidelines and mandates, including IFSPs and IEPs that require appropriate services and placements for these children. Moreover, local ordinances such as the Ley de Orientación sobre los Servicios Multidisciplinarios de Intervención Temprana en Puerto Rico ([P. de la C. 1469]; 2014, ley 200) stipulates Puerto Rico's Department of Health establish a strategic health plan for all children at-risk for any developmental concerns, which includes Deaf and hard of hearing children. Parents and professionals in early intervention in Puerto Rico are encouraged to advocate for these laws to be implemented according to their families' rights. Current advocacy strategies for children in the field are being driven by fears that Deaf and hard of hearing children will not have favorable long-term trajectories without the fervent intervention of adults.

Family advocacy will also need to include early intervention programming specificity. Study participants raised concerns about the lack of Deaf-specific early intervention programming. For example, instead of Deaf-specific programs and classrooms, Deaf and hard of hearing toddlers in Puerto Rico are being clustered with children who have neurodevelopmental needs, such as severe autism based on major classification of diagnoses (i.e., the International Classification of Diseases [ICD] and the Diagnostic Statistical Manual of Mental Disorders [DSM]). Some children with severe autism are unable to communicate using the full syntax, phonemes, morphemes, and context, which are needed in language development. Typically, signing communication systems that are used with children who have significant neurodevelopmental challenges are not fully formed languages. Therefore, this type of integration contributes to noteworthy language deprivation among Deaf and hard of hearing children because the needs of Deaf and hard of hearing children significantly differ from the needs of children with neurodevelopmental concerns, yet they are not being addressed in these programs.

Furthermore, the family's organizational structure continues to be an important factor for early intervention service providers in Puerto Rico due to *familismo,* a cultural practice. Familismo is a central heteronormative cultural value in the Latinx community, which refers to the importance of family interdependence, loyalty, and placing the family's needs before any other areas of importance (Sabogal et al., 1987). Therefore, professionals should provide early intervention services using a family-centered approach (Störbeck & Young, 2016). A family-centered approach seeks to understand the family's strengths, priorities, and resources through thoughtful collaboration with the family to best meet the needs of the child. When a child has a different ability, the family prioritizes the child's needs. In familismo, the female figure, or the mother, becomes the primary caregiver and implementer of services. This change in the family often results in the mother having to redirect her efforts away from working outside of the home (Kelly, 2009, Magaña & Smith, 2006). Although the mother carries the brunt of the child-related services in these cases, the family as a unit continues to make healthcare and educational decisions for the child. A family-centered approach includes all individuals who are identified as *family* members (e.g., immediate versus extended family). The dynamic of familismo and the framework of family-centered approach was discussed throughout the findings of the current study especially in the area of barriers to service.

Limited funding, another barrier to service, appears to be a common reason for inadequate early intervention services in Puerto Rico. The impact of government funding to early intervention in Puerto Rico can be attributed to the fact that Puerto Rico is a colonized territory of the United States that has poor government administration of educational and health programs (Denis, 2015). Puerto Rico currently has a major education crisis with teachers inconsistently receiving pay increases and having poor professional development opportunities, as well as staff having limited resources (Onieva López, 2015). Since the passing of the Puerto Rico Oversight Management and Economic Stability Act (PROMESA) which required an American appointed oversight board to manage the island's budget, Puerto Rico has not managed their own finances (Villanueva, 2019). The PROMESA, established a year prior to the completion of the current study, has implications on early intervention difficulties that both providers and parents reported in this study. In fact, the government budget for special education services to provide therapy services in 2018, which includes early intervention services for Deaf and Hard of Hearing young children, was cut by $78 million (Rivera Sánchez, 2018).

A significant finding in the sample was the limited use of sign language for Deaf children by both the parents and professionals. The findings of the current study align with previous research demonstrating that Deaf children who are exposed to a sign language develop in a typical manner (M. L. Hall et al., 2016). Research has found that many Puerto Ricans may not have access to accurate information regarding how a hearing difference may negatively impact typical social and language development when access to sign language is denied (Gerner de García et al., 2011).

However, no published research has been identified that demonstrates the amount of support for signed languages on the island of Puerto Rico. Related to language access, professionals and parents alike supported the use of assistive technology in Deaf young children as an opportunity for children to learn spoken and written English and Spanish. Unlike previous research, non-invasive technology (e.g., hearing aids) were the primary supported technology in the current study. Past investigations in Puerto Rico reflected how assistive technology, particularly invasive technology (e.g., cochlear implants), was important to Deaf children on the island (Peréz Rodríguez, 2014).

### Limitations

Phenomenological research allows for a rich and in-depth understanding of a specific phenomenon within a population. Although routinely the data collected in a study of this nature attempts to capture the experiences of a variety of people from the general population, the current study only provides experiences from a racially homogeneous perspective. However, the Puerto Rican community is a cultural group of people from many different racial backgrounds. The current study missed data from people who immigrated to the island or who identified as Black Puerto Rican or other racial backgrounds. This is problematic because Black Deaf individuals maintain a double marginalized status globally (Nelson Schmitt & Leigh, 2015; Foster & Kinuthia, 2003). Overall, racial identities and Deaf cultural identities tend to intersect and be multiplicative in nature for people from historically racialized groups. Therefore, representations of Deaf individuals are an important consideration for Deaf infants, toddlers, and children's development and in the services received. Furthermore, all participants, whether parent or professional, identified as cisgender women and had a marital status of married. Lastly, while there are few Deaf children in Puerto Rico who are enrolled in early intervention services, none of the children discussed in this study were receiving early intervention services at the times of the interviews.

### Recommendations and Future Studies

More research is needed in Deaf early intervention services in Puerto Rico, including the intersections of race that are reflective of the island. As with many of the studies, the current study had a sample population that was homogeneous, (i.e., White). A study that focuses on or includes the lived experiences of Black or Asian, for example, Puerto Rican families with Deaf or hard of hearing children would significantly contribute to the literature of these marginalized communities.

Future research could also focus on the language outcomes of Deaf and hard of hearing children in integrated classrooms in Puerto Rico compared to children in Deaf-specific programs. These findings may contribute to the understanding of how these classrooms affect the development of Deaf and hard of hearing children compared to programs tailored to Deaf children. In addition, retrospective accounts of Puerto Rican Deaf adults' views of their early childhood educational careers would contribute to a foundational understanding of changes in Deaf-specific services and experiences for children in Puerto Rico.

This study opens the door for future studies examining the efficacy of early intervention services in Puerto Rico on the development of the young Deaf children being served, as well as replication studies with similar goals as the current research. Future research should consider using the qualitative model of participatory action research (PAR). In PAR research, parents, professionals, and Deaf community members would serve as the main stakeholders to develop best practice guidelines for Deaf early intervention services in Puerto Rico for young children. Furthermore, research needs to include advocacy efforts of families for culturally responsive and collaboration for high-quality early intervention services (e.g., interpreters, appropriate placements and programing, trained Deaf educators, and attention to familismo). To further explore access to early intervention services across the island, researchers should consider the feasibility and efficacy of virtual service delivery. Finally, research needs to be conducted on the continued effect of PROMESA and funding decisions on federally required services to examine the long-term impact on Deaf and hard of hearing children.

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## Appendix A

Semi-structured Interview Guide—Providers

1. Tell me about your background and how you became involved in early intervention with children who are Deaf or hard of hearing.
2. What is your perception of Early Intervention services in Puerto Rico?
3. What are your experiences working in Early Intervention services in Puerto Rico?
4. What information do you provide families about their children's hearing status (e.g. hearing difference, Deaf, hard of hearing)? How about communication?
5. What type of services do you provide to Deaf and hard of hearing children and their families?
6. When are children usually referred to you?
7. What type of interdisciplinary work do you usually do when working with families?
8. What guidelines do you follow when working with Deaf children and their families?
9. What type of support do you receive to provide your services?
    a. From the mentioned above (e.g. supervisor, etc.), what are their strengths and how does this improve your services?
    b. From the mentioned above (e.g. supervisors, etc.), what do you wish they could support you better with?
10. How is the Deaf community involved in your program?

## Appendix B

Semi-structured Interview Guide—Families

1. Tell me about your and your family's thoughts and feelings when you were first told that your child was Deaf (or hard of hearing)? How old was your child?
2. What type of supports have you had and from whom?
3. What type of communication do you use at home and how did you decide on the communication approach to use with your child?
4. What type of information have you received to understand your child's strengths and needs?
5. What type of early intervention have you and your child received?
6. What progress has your child made since starting early intervention services?
7. Have you and/or your child met Deaf adults? If yes, who did you meet and why did you meet them? If no, why have you and/or your child not met Deaf adults?
8. What services have you received that have helped your child and your family?
9. What services or resources do you wish you had for your Deaf child?
10. What advice do you have for the professionals who work with families like you who have a Deaf child?
11. In 10 years, what do you hope your child will be doing?