The Effects of Early Intervention on Language Growth after Age 3 for Children with Permanent Hearing Loss

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Abstract

Purpose: The overall goal of this study was to examine language performance in children with permanent hearing loss who were enrolled in a Listening and Spoken Language program. The influence of time spent in Early Intervention (EI) on language trajectories and ability to attain age-appropriate language skills was examined.

Method: Retrospective data were obtained from children (N = 48) who attended Central Institute for the Deaf (CID) for various lengths of time between 2004 and 2017. Children were grouped into those who had received EI prior to age 3 years versus those who did not. Standardized tests of receptive and expressive language were administered annually. Comparisons of language levels attained at the initial and final assessment were conducted and linear mixed model analyses examined language scores over time.

Results: Children receiving EI attained significantly higher levels of language than those receiving no EI. The rate of improvement over time in vocabulary scores was similar for both groups, however on a global language test that included morpho-syntax, children with EI made greater progress relative to age-matched peers than children without EI.

Conclusion: Children receiving EI exhibited a lasting advantage in the acquisition of spoken language over children who did not have access to EI.

Keywords: pediatric, early intervention, hearing loss, spoken language, hearing devices

Acronyms: BAHA = bone aided hearing aid; BI-CROS = Bilateral Contralateral Routing of Signal; CELF = Clinical Evaluation of Language Fundamentals; CI = cochlear implants; CID = Central Institute for the Deaf; EI = early intervention; EVT = Expressive Vocabulary Test; FM = Frequency Modulated; HA = hearing aid; HL = hearing loss; LSL = listening and spoken language; NBHS = newborn hearing screening; PHL = permanent hearing loss, PPVT = Peabody Picture Vocabulary Test; PTA = pure tone average; TH = typical hearing

Acknowledgment: The first author is a member of the Knowledge Implementation in Pediatric Audiology (KIPA) group sponsored by Oticon. The second and third authors report no relevant conflicts of interest related to this work. Portions of this research were supported by the NIDCD grant RO1 DC012778. This research was approved by the Human Studies Committee at Washington University School of Medicine (IRB ID 201803208). We would like to thank Michael Strube and Chris Brenner for statistical support and data analysis for this project.

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The overarching goal of Listening and Spoken Language (LSL) Early Intervention (EI) programs is to provide children with permanent hearing loss (PHL) the opportunity to develop spoken language skills that are commensurate with age-matched peers who have typical hearing (TH). These programs are a constellation of multidisciplinary services that include early hearing screening, confirmation of hearing loss, fitting and management of appropriate hearing technology and individualized family and small-group sessions for language instruction. The individualized sessions include optimizing language input by focusing on language acquisition, hearing loss, and hearing device use. Overall, exposure to newborn hearing screenings (NBHS) has had positive effects on language outcomes for children with PHL with the primary benefits related to early confirmation of hearing loss and subsequent receipt of hearing devices and services (Pimperton & Kennedy, 2012). In the United States, children with hearing loss have access to EI services from birth to 36 months through part C of the Individuals with Disabilities Educational Act (IDEA).

Prior to implementation of universal NBHS, Moeller (2000) found that children enrolled in EI services before 11 months of age had receptive language and verbal
In addition to the positive effects of EI factors, family, rehabilitation settings, these studies, since they occur simultaneously in most and audiological intervention are confounded in all of et al., 2018). The effects of educational intervention as age at hearing aid fit, age at confirmation of hearing instruction. Therefore, studies typically use variables such with signs or gestures) skills reveal positive outcomes related to earlier services (Ching et al., 2017; Meinzen- Derr et al., 2011; Vohr et al., 2011).

Studies that have examined the effects of EI using intensity of treatment as a metric have reached somewhat different conclusions. Geers et al. (2019) examined the effects of the amount of EI on spoken language and literacy outcomes for 50 children with PHL at preschool age and again at elementary age. Their variable of interest was the total hours (dose) of therapy in a listening and spoken language EI program (determined from billing records) between 0 and 36 months of age. They concluded that greater intensity of services during the birth to 36-month period was associated with higher spoken language and literacy scores at elementary age. These effects were apparent even after other contributing factors such as degree of hearing loss, nonverbal intelligence, and age at entry into services were controlled. Those children with poor early speech perception skills benefited the most from the increased dose provided by toddler classes beginning at 18 months. Alternatively, in a retrospective analysis of standardized receptive and expressive language scores of 40 children (mean age at test: 4.18 years) with cochlear implants (CIs), Chu et al. (2019) found that total dose of therapy, defined as the total number of parent-reported hours of therapy, was not associated with better spoken language outcomes. Moreover, for expressive language scores they found an inverse association between total dose of therapy and scores. That is, children with smaller total doses of intervention exhibited better language scores. These univariate analyses did not control for other contributing factors, such as hearing level, parent education, or intervention age. Notably, there was a significant association with age at CI and intensity of EI services, such that those children who received CIs at younger ages received less intensive services. Thus, those children receiving CIs at younger ages were likely to exhibit superior language skills and need less intensive therapy to achieve age-appropriate language levels.

Audiological interventions, such as confirmation of hearing loss and device fitting, typically co-occur with initiation of individual family sessions that focus on language instruction. Therefore, studies typically use variables such as age at hearing aid fit, age at confirmation of hearing loss, or age at CI as a proxy for initiation of EI services (Ching et al., 2018; Fulcher et al., 2012; Yoshinaga-Itano et al., 2018). The effects of educational intervention and audiological intervention are confounded in all of these studies, since they occur simultaneously in most rehabilitation settings.

In addition to the positive effects of EI factors, family, and child characteristics such as higher non-verbal intelligence, higher socio-economic status, less severe levels of hearing loss, female gender, and higher maternal education level contribute to positive outcomes for children with PHL (Ching et al., 2013; Ching et al., 2018; Wake et al., 2005; Yoshinaga-Itano et al., 2018). These studies have examined the effects of EI on spoken language skills at specific time points such as preschool and elementary age (see also Daub et al., 2017). Tomblin and colleagues (2015) analyzed longitudinal data from 414 children with mild to moderate hearing impairment to test whether language growth trajectories were associated with degree of residual hearing and whether aided hearing influenced language growth in a systematic manner. The degree to which language skills fell behind those of age-matched peers with TH increased with greater severity of hearing loss. Early fitting of hearing aids was associated with better early language achievement, but children fit after 18 months of age improved in their language abilities as a function of the duration of hearing aid use. Greater language delays were reported in the domain of morpho-syntax (more dependent on auditory phoneme discrimination) than in semantic abilities (presumably less reliant on audition).

A number of variables contribute to children missing the opportunity to receive audiological and language instruction services during the first three years of life (late identification of hearing loss, loss to follow-up, inconsistent audiological results, family attendance). This is reflected in statistics that reveal that as many as 40% of children referred for follow up testing for hearing loss do not meet the recommended guidelines for early identification and intervention (Centers for Disease Control and Prevention, 2015). As such, these children may receive little, if any, EI services and begin intensive instruction in spoken language and listening after the age of three years. As might be expected, children with amounts of EI varying from none to a maximum of near-36 months, enter LSL programs with vastly different spoken language skills. Recently, Soman and Nevins (2018) proposed three different performance profiles of language growth for children entering LSL programs, those who Keep Up, Catch Up, or Move Up. In general, those in the Keep Up category have the benefit of meeting EI milestones and attain spoken language skills that are at or near their age-matched peers who have TH. The goal for these children is early entry into general education classrooms and maintenance of age-appropriate language skills for academic and linguistic success. Children in the Catch Up category typically start with language skills below their age-matched peers with TH as a result of little or no EI services, late identification of hearing loss, or late receipt of devices. However, with intensive instruction, many of these children show improvement in language skills and some ultimately achieve age-appropriate language skills. Those in the Move Up category may have secondary diagnoses (e.g., attention deficit, learning disabilities) in addition to hearing loss that may preclude obtaining age-appropriate language skills. These children do, however, manage to make some progress and attain some
functional listening and communication skills (Soman & Nevins, 2018).

In the current study, the effects of EI during the birth to 36-month time period on longitudinal (i.e., annual) development of spoken language skills was measured for children with PHL attending an intensive LSL program. A primary goal of this study was to understand the benefit associated with greater amount of time spent in an EI program that included coordinated audiological and language services after controlling for age, gender, maternal education, and degree of hearing loss.

Tests of vocabulary and global language (including semantics and morpho-syntax) were administered on a yearly basis for children ranging in age from 3 to 9 years. First, overall language levels were determined for all children in the study. Second, language growth trajectories were compared for children who received some amount of EI services during the birth to 36-month EI period and those who received no EI services in that age range. Third, for the group that received some period of EI services, the effects of duration of time spent in EI were examined. Finally, differential effects of EI on vocabulary compared to global language skills were explored.

Language development of children with PHL in the following areas was examined: (a) attainment of age-appropriate language levels, (b) effects of maternal education, gender, and hearing level on language, (c) improvement of language skills with age relative to age-matched peers with TH, and (d) effects of EI on language level and language growth rates with age.

**Method**

Retrospective data were obtained from children (N = 48) who attended the school-age program at CID for various lengths of time between 2004 and 2017. Typical of all LSL programs, CID emphasizes intensive listening and spoken language instruction (Bradham et al., 2018; Estes, 2010; Soman & Nevins, 2018) beginning as young as possible. The EI programs serve children from diagnosis of hearing loss (HL) through 36 months of age. The school program at CID serves children starting at age 3 years and emphasizes individualized spoken language instruction, literacy, and social skills as well as age-appropriate academic instruction.

**Participants**

Table 1 describes sample demographics for the total group of 48 children, including maternal education, gender, unaided pure-tone average (PTA; 0.5, 1 & 2 kHz) for the better ear, age at hearing aid (HA) fitting and age the child enrolled in the school program at CID. In this study, we report age at HA fitting as opposed to age at confirmation of hearing loss, since age at HA fitting represents the initiation of access to sound. On average these children were fit with HAs at 18.7 months (SD = 14.4 months) with an average unaided PTA for the better ear of 66 dB HL. The average maternal education calculated as total years of education through college and beyond, was 13 years. Forty-four percent of the group were female, and the average age enrolled in the CID school was 3.8 years (SD = 1.2 years). The devices worn during the time attending CID school were as follows: 15 children wore two cochlear implants (CI), 13 wore binaural HAs, 11 wore a CI and a HA at the non-implanted ear (bimodal devices), 3 wore Bone Anchored Hearing Aid (BAHA), 3 used a Frequency Modulated device (FM), 2 wore a combination of HA/FM, and 1 wore a bilateral contralateral routing of signal device (BI-CROS).

Children were categorized into those who had received EI services during a period from birth to 36 months (n = 32) and those who did not receive EI (n = 16). Demographic characteristics of each group are in Tables 2a and 2b. The EI service model included confirmation of hearing loss and monitoring of hearing thresholds, provision and monitoring of hearing devices (i.e., HAs, CIs, FM, BAHA, BI-CROS) and instruction for families related to hearing loss and acquisition of language. Typical of most LSL programs,

**Table 1**

**Demographics of Children**

<table>
<thead>
<tr>
<th>Demographics (N = 48)</th>
<th>Mean</th>
<th>Standard Deviation</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at First Hearing Aid Fitting (months)</td>
<td>18.7</td>
<td>14.4</td>
<td>2–60</td>
</tr>
<tr>
<td>Maternal Education (years)</td>
<td>13.8</td>
<td>2.3</td>
<td>11–18</td>
</tr>
<tr>
<td>Unaided Pure Tone Average (Better Ear)</td>
<td>66.4</td>
<td>32.2</td>
<td>10–115</td>
</tr>
<tr>
<td>Age Began Central Institute for the Deaf School (years)</td>
<td>3.8</td>
<td>1.2</td>
<td>2.9–7.5</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female – 21 (44%)</td>
<td>Male – 27 (56%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**Table 2a**

**Demographics of Students with Early Intervention Services**

<table>
<thead>
<tr>
<th>Demographics (n = 32)</th>
<th>Mean</th>
<th>Standard Deviation</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at First Hearing Aid Fitting (months)</td>
<td>12.4</td>
<td>8.8</td>
<td>2–28</td>
</tr>
<tr>
<td>Maternal Education (years)</td>
<td>13.8</td>
<td>2.2</td>
<td>11–18</td>
</tr>
<tr>
<td>Unaided Pure Tone Average (Better Ear)</td>
<td>63.0</td>
<td>35.5</td>
<td>10–115</td>
</tr>
<tr>
<td>Age Began Central Institute for the Deaf School (years)</td>
<td>3.3</td>
<td>0.5</td>
<td>2.9–4.9</td>
</tr>
<tr>
<td>Duration of Early Intervention (months)</td>
<td>22.3</td>
<td>9.6</td>
<td>3–34</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female – 11 (34%)</td>
<td>Male – 21 (66%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Demographics of Students with no Early Intervention Services

<table>
<thead>
<tr>
<th>Demographics (n = 16)</th>
<th>Mean</th>
<th>Standard Deviation</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at First Hearing Aid Fitting (months)</td>
<td>30.9</td>
<td>15.5</td>
<td>3–60</td>
</tr>
<tr>
<td>Maternal Education (years)</td>
<td>13.8</td>
<td>2.5</td>
<td>11–18</td>
</tr>
<tr>
<td>Unaided Pure Tone Average (Better Ear)</td>
<td>73.3</td>
<td>23.7</td>
<td>31–115</td>
</tr>
<tr>
<td>Age Began Central Institute for the Deaf School (years)</td>
<td>4.9</td>
<td>1.5</td>
<td>3.0–7.5</td>
</tr>
<tr>
<td><strong>Count (%)</strong></td>
<td><strong>Count (%)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gender</td>
<td>Female – 10 (62%)</td>
<td>Male – 6 (38%)</td>
<td></td>
</tr>
</tbody>
</table>

The EI program at CID involves a multidisciplinary team of audiologists, teachers of the deaf and speech language pathologists specializing in developing listening and spoken language skills through device use and instructional strategies. For children attending CID during the time period listed above (2004–2017), the frequency of sessions with teachers of the deaf and speech language pathologists varied depending on the age of the child and the needs of the family. These sessions were conducted primarily through home visits, although some used a combination of homebased and center-based therapy depending on distance from the facility. Some small group instruction was included for children beginning at 24 months of age. The duration in EI was the number of months enrolled in EI services through 36 months of age. Most children were fit with HAs within 1 to 2 months of enrolling in EI services. The duration of time spent in EI varied from 3 months to 34 months (mean = 22 months). Children entered the CID school at an average age of 3.3 years. Most were enrolled near their 3rd birthday, however depending on the academic school calendar, some entered slightly before (~2.9 years). Four children entered the school at later ages (~4.0–4.9 years) due to a variety of family circumstances.

For children who did not receive EI services (n = 16), the average age of HA fitting was 31 months with a range from 3 months to 60 months. These participants entered the CID school at an average age of 4.9 years (SD = 1.5 years).

All children enrolled at CID receive norm-referenced standardized tests of receptive and expressive vocabulary and language annually. All measures were administered and scored according to the test manual by certified SLPs. The number of tests administered to a specific child varied based on student age at enrollment and length of enrollment. The average number of annual test sessions was 3 and ranged from 1–6.¹ The maximum age tested was 9 years old. The following tests were administered at each test session.

¹One child had only one test session. In this case, the child’s data point is used in the analysis to estimate the intercept.

Receptive Vocabulary

The Peabody Picture Vocabulary Test, Fourth Edition (PPVT-4; Dunn & Dunn, 2007), a measure of receptive vocabulary, was administered live voice in an auditory–visual mode. The examiner provided a target word and the child was asked to identify the correct picture from a closed set of 4 pictures.

Expressive Vocabulary

The Expressive Vocabulary Test, Second Edition (EVT-2; Williams, 2007), requires the child to provide either verbal labels or synonyms. The child is shown a colored picture and prompted by the examiner to provide a one-word response (e.g., “What is this animal?” or “Tell me another word for jacket.”).

Receptive and Expressive Language

Depending on the child’s age at test, either The Clinical Evaluation of Language Fundamentals Preschool-2 (CELF-P2; Wiig et al., 2004) or the Clinical Evaluation of Language Fundamentals–Fourth Edition (CELF–4; Semel et al., 2003) was administered to evaluate global language in both the receptive and expressive domains.

The CELF-P2 provides a core language score derived from three subtests: Sentence Structure, Word Structure, and Expressive Vocabulary. The Sentence Structure subtest evaluates a child’s ability to understand and process sentence formation rules. The child chooses one of four pictures which best represents sentences read by the examiner. Sentences vary in length and complexity (Wiig et al., 2004, p. 15). The Word Structure subtest evaluates the child’s ability to apply morphology rules and use appropriate pronouns. The child provides a missing word or phrase (e.g., preposition, pronoun, and various verb tense) in a sentence spoken by the examiner (Wiig et al., 2004, p. 18). The Expressive Vocabulary subtest evaluates the child’s ability to provide an appropriate label to describe pictures of objects and actions (Wiig et al., 2004, p. 22).

The CELF–4 is a global measure of language skills and provides a core language score derived from four subtests for children ages 5 to 8: Concepts and Following Directions, Word Structure, Recalling Sentences, and Formulated Sentences. The Concepts and Following Directions subtest is used to evaluate the child’s ability to interpret, recall, and execute oral directions of increasing length and syntactic complexity (Semel et al., 2003, p. 18). The Word Structure subtest evaluates a child’s ability to apply morphology rules and use appropriate pronouns. The child provides a missing word or phrase in a sentence spoken by the examiner (Semel et al., 2003, p. 22). The Recalling Sentences subtest evaluates the child’s ability to recall and reproduce sentences of varying length and syntactic complexity without altering word meanings, morphology or syntax. The child imitates sentences spoken by the examiner (Semel et al., 2003, p. 25). The Formulated Sentences subtest evaluates the child’s ability to formulate compound and complex sentences.
using target words or phrases, while using a picture as a reference (Semel et al., 2003, p. 33).

Data Analysis

Standardized scores were used to control for chronological age and compare a child’s performance to that of their age-matched peers with TH in each test’s normative sample. A standardized score of 100 reflects average age-appropriate performance, with a standard deviation of 15. Receptive vocabulary (PPVT), expressive vocabulary (EVT) and global language (Core Language Score from the CELF) scores from each successive annual evaluation were obtained from student records.

Linear mixed model analyses examined change in annual standardized language scores over time on a continuous rather than a fixed set of points and without having the same number of tests per subject. If a child makes age-appropriate progress over time, their standardized scores from year to year remain essentially the same. Thus, repeated assessments showing a year of language growth for an average hearing student would be shown by a flat line (i.e., a slope of zero). If the student makes more than yearly expected progress, the slope would be positive; if the child makes less than age-appropriate progress, the slope would be negative.

Predictor variables were entered in stages to test their independent contributions to language scores. Demographic variables were entered on the first step to determine the amount of variance in language outcome attributable to the child’s gender, mother’s education, degree of hearing loss (best-ear unaided PTA), and age at test. The categorical variable Early Intervention compared language levels achieved by those who received EI (coded as 2) and those who did not receive EI (coded as 1) after variance due to demographic variables had been accounted for in the first step. Duration of EI was entered as a continuous variable at subsequent stages to determine the effects of duration of EI within the group receiving services. Interactions between the EI variables of interest and age were also analyzed in subsequent steps of the regression. The sequential entry of variables was necessary to account for main effects of predictor variables before interpreting any interactions among variables of interest (age at test, receipt of EI services, and duration of EI). In these linear mixed models, age and the intercept were treated as random effects.

Results

The average age at the initial assessment was 4.57 years old and the average age at the final assessment was 6.75. Average language test scores are summarized in Table 3a, along with the percentage of children at each assessment session scoring within 1 SD of age-matched peers in the normative sample for each test. Pairwise comparisons of scores on each of the three measures are summarized in Table 3b. EVT scores were significantly higher than PPVT or CELF scores at both test sessions and scores on both vocabulary tests were significantly higher than scores on the global language measure (CELF). Mean language scores for the EI group (n = 32) and No EI group (n = 16) at the initial test and final test sessions are summarized separately in Table 4. Both groups made significant gains over time relative to age-matched peers with TH. Scores of the EI group exceeded those of the No EI group at both initial and final test sessions on all tests.

For the subsequent analyses, age at HA fitting is not included as a predictor variable. As one might expect age at HA fitting and duration of EI services were highly correlated (r = .68) since most children received their HAs when they entered EI, thus age at HA was not included in the demographic predictors.

The regression coefficients; standard error; t values; and significance values for the PPVT, EVT, and CELF language scores are shown in Table 5 (sections 5a, 5b and 5c respectively). The graphs in Figures 1–3 illustrate the relationship between each outcome variable (PPVT, EVT, and CELF Language respectively) score and age for the EI groups. The predicted average scores and 95th percentile confidence intervals (CEIs) from the model are plotted over ranges that represent the median first and last ages tested for each group. The No EI group is labeled and shown in green. To illustrate the effects of duration of intervention for the EI group, the duration variable is divided into two levels for each assessment session: 1) before variance due to demographic variables had been accounted for and 2) and those who did not receive EI (coded as 0) with No EI group (n = 16). To illustrate the effects of duration of intervention for the EI group, the duration variable is divided into two levels for each assessment session: 1) after variance due to demographic variables had been accounted for (coded as 2) and those who did not receive EI (coded as 1) at each assessment session on all tests.

Table 3a

<table>
<thead>
<tr>
<th>Test</th>
<th>Initial Test Session</th>
<th>Final Test Session</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean</td>
<td>Standard Deviation</td>
</tr>
<tr>
<td>PPVT</td>
<td>85.1</td>
<td>17.6</td>
</tr>
<tr>
<td>EVT</td>
<td>88.9</td>
<td>18.4</td>
</tr>
<tr>
<td>CELF</td>
<td>76.4</td>
<td>21.0</td>
</tr>
</tbody>
</table>

Note. There were 48 participants tested. WNL = within normal limits; PPVT = Peabody Picture Vocabulary Test; EVT = Expressive Vocabulary Test; CELF = Clinical Evaluation of Language Fundamentals.

*Mean = 100; Standard Deviation = 15

Table 3b

<table>
<thead>
<tr>
<th>Test</th>
<th>First Test Session</th>
<th>Last Test Session</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean Difference</td>
<td>Standard Error</td>
</tr>
<tr>
<td>PPVT</td>
<td>EVT</td>
<td>-.9</td>
</tr>
<tr>
<td>PPVT</td>
<td>CELF</td>
<td>9.3</td>
</tr>
<tr>
<td>EVT</td>
<td>CELF</td>
<td>13.2</td>
</tr>
</tbody>
</table>

Note. PPVT = Peabody Picture Vocabulary Test; EVT = Expressive Vocabulary Test; CELF = Clinical Evaluation of Language Fundamentals.
<table>
<thead>
<tr>
<th>Table 4</th>
<th>Standard* Test Scores for Early Intervention and No Early Intervention Groups</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Early Intervention (n = 32)</td>
</tr>
<tr>
<td></td>
<td>Initial</td>
</tr>
<tr>
<td>Age (years)</td>
<td>Mean</td>
</tr>
<tr>
<td></td>
<td>SD</td>
</tr>
<tr>
<td>PPVT</td>
<td>Mean</td>
</tr>
<tr>
<td></td>
<td>SD</td>
</tr>
<tr>
<td></td>
<td>%WNL</td>
</tr>
<tr>
<td>15.5</td>
<td>%WNL</td>
</tr>
<tr>
<td>EVT</td>
<td>Mean</td>
</tr>
<tr>
<td></td>
<td>SD</td>
</tr>
<tr>
<td></td>
<td>%WNL</td>
</tr>
<tr>
<td>CELF</td>
<td>Mean</td>
</tr>
<tr>
<td></td>
<td>SD</td>
</tr>
<tr>
<td></td>
<td>%WNL</td>
</tr>
</tbody>
</table>

Note: WNL = within normal limits; PPVT = Peabody Picture Vocabulary Test; EVT = Expressive Vocabulary Test; CELF = Clinical Evaluation of Language Fundamentals. *Mean = 100; Standard Deviation = 15

<table>
<thead>
<tr>
<th>Table 5a</th>
<th>Peabody Picture Vocabulary Test</th>
</tr>
</thead>
<tbody>
<tr>
<td>Predictor</td>
<td>Coefficient</td>
</tr>
<tr>
<td>Intercept</td>
<td>85.18</td>
</tr>
<tr>
<td>Gender</td>
<td>-3.76</td>
</tr>
<tr>
<td>Mom’s Education (in years)</td>
<td>1.49</td>
</tr>
<tr>
<td>Better Ear unaided PTA</td>
<td>-0.26</td>
</tr>
<tr>
<td>Age</td>
<td>2.16</td>
</tr>
<tr>
<td>EI-Yes</td>
<td>20.3</td>
</tr>
<tr>
<td>Age x EI-Yes</td>
<td>-0.48</td>
</tr>
<tr>
<td>EI-Yes x duration of EI</td>
<td>0.51</td>
</tr>
<tr>
<td>Age Squared</td>
<td>-1.06</td>
</tr>
<tr>
<td>Age x EI-Yes x duration of EI</td>
<td>-0.08</td>
</tr>
<tr>
<td>EI-Yes x Age Squared x duration of EI</td>
<td>-0.03</td>
</tr>
</tbody>
</table>

Note: The regression model summary has 48 subjects. EI = Early Intervention, PTA = Puretone Average, Std = standard, df = degrees of freedom. Bold text indicates statistically significant variables in the model.

<table>
<thead>
<tr>
<th>Table 5b</th>
<th>Expressive Vocabulary Test</th>
</tr>
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<tbody>
<tr>
<td>Predictor</td>
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<tr>
<td>Intercept</td>
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<tr>
<td>Gender</td>
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<tr>
<td>Mom’s Education (in years)</td>
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<tr>
<td>Better Ear unaided PTA</td>
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<td>Age</td>
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<tr>
<td>EI-Yes</td>
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<td>Age x EI-Yes</td>
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<tr>
<td>Age Squared</td>
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<td>Age x EI-Yes x duration of EI</td>
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<tr>
<td>EI-Yes x Age Squared x duration of EI</td>
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Note. The regression model summary has 48 subjects. EI = Early Intervention, PTA = Puretone Average, Std = standard, df = degrees of freedom. Bold text indicates statistically significant variables in the model.

<table>
<thead>
<tr>
<th>Table 5c</th>
<th>Comprehensive Evaluation of Language Fundamentals</th>
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<td>Intercept</td>
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<tr>
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<tr>
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<td>Age x EI-Yes x duration of EI</td>
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</tr>
<tr>
<td>EI-Yes x Age Squared x duration of EI</td>
<td>-0.01</td>
</tr>
</tbody>
</table>

Note. The regression model summary has 48 subjects. EI = Early Intervention, PTA = Puretone Average, Std = standard, df = degrees of freedom. Bold text indicates statistically significant variables in the model.
Receptive Vocabulary (PPVT)

The regression analyses for the PPVT revealed the following:

EI and No EI Groups (N = 48)

Step 1: Age at test and better ear unaided PTA were significant predictors of receptive vocabulary level (p = .003 and p = .002 respectively). Increases in age at test had a positive effect on PPVT standard score (children closed the vocabulary gap with age-matched peers with TH in the normative sample as they got older) and increases in PTA (poorer hearing) had a negative effect on scores. Children with greater hearing losses were further behind age-matched peers with TH in vocabulary development.

Step 2: The EI categorical variable (EI group vs. No EI group) was a significant predictor of vocabulary level after controlling for demographic variables entered at the first step of the analysis. Children in the EI group scored approximately 20 standard score points higher than those without EI, a significant difference (p = .0003).

Step 3: There was no significant interaction between test age and EI group indicating that the growth of receptive vocabulary relative to age-matched peers with TH is similar in both EI and No EI groups.

Step 4: The quadratic effect of age (age squared) at test was significant and negative (p < .01) showing that the standard score gains associated with increasing age diminish as age increases (i.e., rate of change over time levels off).

EI Group (n = 32)

Step 5: The effect of duration of intervention on PPVT scores within the EI group did not meet statistical significance (p = .058).

Steps 6 & 7: There were no significant interactions between age (linear or quadratic) and duration of EI within the group that received EI. The effects of age do not change with duration of EI.

The graph in Figure 1 illustrates the relationship between PPVT score and age for the EI groups. Note that the EI group achieves overall higher scores than the No EI group, however receptive vocabulary growth is similar for both groups.

Expressive Vocabulary (EVT)

The regression analyses for the EVT scores revealed the following:

EI and No EI Groups (N = 48)

Step 1: Age at test and better ear unaided PTA were significant predictors of EVT scores (p = .002 and p = .01 respectively) in the model. Increases in age at test had a positive effect on EVT standard score (children closed the vocabulary gap with age-matched peers with TH in the normative sample as they got older) and increases in PTA had a negative effect on scores. Children with greater hearing losses were further behind in expressive vocabulary development.

Step 2: After controlling for demographic variables, the EI categorical variable (EI group vs. No EI group) was a significant predictor of vocabulary level. Children in the EI group scored approximately 21 standard score points higher than those in the group without EI, a significant difference (p = .001).

Step 3: The interaction between EI group and age was not significant indicating that the rate of standard score change with age was not different between the EI and No EI groups.

Step 4: The quadratic effect of age (age squared) at test was significant (p = .002) indicating that the linear effect of age diminishes as age increases.

EI Group (n = 32)

Step 5: The duration effect within the EI group was significant (p = .02) indicating that for those who received some EI services, longer durations of EI resulted in better expressive vocabulary skills.

Steps 6 & 7: The linear effects of age and duration of EI within the EI group were significant (p = .04) indicating that the effects of duration of intervention diminish as age increases. The non-linear effects of age and duration of EI were not significant.

Figure 1

Peabody Picture Vocabulary Test (PPVT) Standard Scores

Note. PPVT standard scores are shown on the Y axis as a function of age at test in years on the X axis for the No Early Intervention (EI) Group, Low Duration EI Group and High Duration EI Groups (green, blue, and red respectively). The 95th percent confidence intervals are illustrated by gray shaded areas. The dashed line illustrates the minimum standard score (85) for the normative range.
The graph in Figure 2 illustrates the relationship between EVT and test age for the EI groups. Attendance in EI programs leads to higher expressive vocabulary scores as shown by the overall differences in the predicted absolute EVT standard score for the No EI group compared to the Low and High Duration EI groups. Overall, scores on the EVT increase with age, but eventually plateau. Within the EI group, longer duration of early intervention leads to better outcomes for expressive vocabulary. Expressive vocabulary growth over time was reflected in the duration of EI by age interaction within the EI group. Those children in the High Duration EI group started with overall higher EVT scores that plateaued with increasing age, while those in the Low Duration EI group started with lower overall scores and showed a linear increase in scores with increasing age.

**Figure 2**
Expressive Vocabulary Test (EVT) Standard Scores

<table>
<thead>
<tr>
<th>Age</th>
<th>Predicted EVT</th>
</tr>
</thead>
<tbody>
<tr>
<td>2</td>
<td>80</td>
</tr>
<tr>
<td>3</td>
<td>85</td>
</tr>
<tr>
<td>4</td>
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<tr>
<td>7</td>
<td>105</td>
</tr>
<tr>
<td>8</td>
<td>110</td>
</tr>
<tr>
<td>9</td>
<td>115</td>
</tr>
</tbody>
</table>

*Note.* Expressive Vocabulary Test (EVT) standard scores are shown on the Y axis as a function of age at test in years on the X axis for No Early Intervention (EI), Low Duration EI, and High Duration EI groups (green, red, and blue respectively). The 95th percent confidence intervals are illustrated by gray shaded areas. The dashed line illustrates the minimum standard score (85) for the normative range.

**Global Language ( CELF )**

The regression analyses revealed the following:

**EI and No EI Groups (N = 48)**

Step 1: Age at test and better ear unaided PTA were significant predictors ($p = .02$ and $p = .001$ respectively) in the model. Increases in age at test had a positive effect on CELF standard scores (children’s language skills more closely approximated age-matched peers who have TH in the normative sample as they got older) and increases in PTA had a negative effect on scores. Children with greater hearing losses were further behind in language development.

Step 2: After controlling for demographic variables entered at the first step of the analysis, the EI categorical variable (EI group vs. No EI group) was a significant predictor. Children in the EI group scored approximately 24 standard score points higher than those in the group without EI, resulting in a significant difference between means ($p = .001$).

Step 3: The interaction between EI group and age was significant ($p = .04$) indicating that improvement in language scores over time were greater for the EI group vs. the No EI group.

Step 4: The quadratic effect of age (age squared) at test was not significant indicating that language gains over time did not plateau.

**EI Group (n = 32)**

Step 5: The duration effect within the EI group was significant ($p = .01$) indicating that for those in the EI group, longer durations of early intervention resulted in better language skills.

Step 6: The interaction of age and duration of EI within the EI group was not significant. The rate of language growth over time was similar regardless of duration of EI.

Step 7: The interaction between EI duration and the quadratic effect of age of duration was not significant meaning that language growth did not plateau with increased duration of EI.

**Figure 3**
Clinical Evaluation of Language Fundamentals ( CELF ) Standard Scores

<table>
<thead>
<tr>
<th>Age</th>
<th>Predicted Language</th>
</tr>
</thead>
<tbody>
<tr>
<td>2</td>
<td>80</td>
</tr>
<tr>
<td>3</td>
<td>85</td>
</tr>
<tr>
<td>4</td>
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<tr>
<td>8</td>
<td>110</td>
</tr>
<tr>
<td>9</td>
<td>115</td>
</tr>
</tbody>
</table>

*Note.* CELF standard scores are shown on the Y axis as a function of age at test in years on the X axis for the No Early Intervention (EI) Group, Low Duration EI Group and High Duration EI Groups (green, blue, and red respectively). The 95th percent confidence intervals are illustrated by gray shaded areas. The dashed line illustrates the minimum standard score (85) for the normative range.
The graph in Figure 3 illustrates the relationship between CELF scores and age for the EI groups. As noted in these graphs, attendance in an EI program leads to higher language scores as shown by the overall differences in the predicted absolute CELF standard score for the No EI group compared to the Low Duration and High Duration EI groups. The effects of age are more apparent for the EI group as evidenced by the steeper increase in CELF scores over time for both the Low or High Duration EI groups compared to the No EI group.

Discussion

The overall goal of this retrospective study was to examine language growth profiles in children with PHL enrolled in an LSL program. Specifically, the influence of time spent in EI programs (birth to 36 months) on these children’s language trajectories and ability to attain age-appropriate language skills were examined. Prior to enrolling in CID’s school program, some children had spent varying amounts of time in an EI program where they received the benefit of early audiological management coupled with individual spoken language instruction. Due to a variety of reasons (e.g., loss to follow-up from NBHS, late confirmation of hearing loss) some children had received no EI services prior to enrolling into the CID school. A second aim of this study was to determine if the effects of EI were consistent across various language domains including receptive vocabulary, expressive vocabulary, and global language skills. The following questions were addressed in the data analysis:

Do scores of children with PHL reach age-appropriate levels during their years of enrollment in a LSL education program?

As a group, the average scores from these children with PHL were within 1 SD of age-matched peers with TH (i.e., within the normative test range, 85–115) in expressive and receptive vocabulary but not in global language skills at their initial assessment (mean age 4.5 years). Average scores at the last assessment (mean age 6.6) were within the normative range on all three measures. As noted in Table 3a, average performance for the EVT (standard score = 99.5) more closely approximated age-matched peers with TH (standard score = 100) than receptive vocabulary (93.6) and global language skills (85.2) at the last assessment.

Do language scores for children with PHL improve with age?

Across all measures of language, there was a significant trend for their language delay to diminish over time. This trend reflects the positive effect of enrollment in an LSL education setting throughout the age range represented in the sample.

Do language scores improve with greater residual hearing?

Across all measures of language, children with greater amounts of residual hearing scored closer to age-matched peers with TH. The overall effects of residual hearing level are consistent with studies examining benefits of EI (Ching et al., 2017; Tomblin et al., 2015; Vohr et al., 2011). However, unaided PTA did not interact with test age, EI status, or duration of EI. The positive effects of longer duration of intervention on language were similar regardless of degree of hearing loss. This finding stands in contrast to earlier results showing that children with greater hearing loss benefitted more from greater doses of early intervention (Geers et al., 2019). This apparent contradiction may be associated with the different measures of EI, number of months enrolled compared to number of hours participated.

Are language scores affected by level of maternal education and by the child’s gender?

Unlike results observed in some other samples reported in the literature (Ching et al., 2018; Yoshinaga-Itano et al., 2018; Tomblin et al., 2015) maternal education level did not contribute significantly to variance in language performance. The current finding may be attributable to the relatively high mean education level and low variability observed in this sample. In contrast to showing language benefits for females (Ching et al., 2013), the current study found no significant effects related to gender.

Do language scores improve with EI? How much advantage does EI provide?

After controlling for age and hearing loss, children who received some amount of EI performed closer to TH age-appropriate levels than children who did not receive EI. The advantage was similar across the three language measures, ranging from 20 to 24 standard score points, an increase of more than one standard deviation for age-norms. This advantage was apparent in initial language level measured when each child began attending the school at CID as well in the final assessment.

Do language scores improve with longer EI?

With the exception of receptive vocabulary, where group differences did not meet statistical significance, longer durations of EI were associated with expressive vocabulary scores and language scores closer to age-appropriate levels. Duration of EI was highly correlated with age at HA fitting, suggesting that these factors overlap in affecting language levels. Therefore, the advantage associated with EI is associated with younger access to the auditory speech signal through the use of technology as well as greater duration of guided instruction. There was no attempt in this study to disentangle the effects of cumulative early auditory access with hearing devices from time spent in guided language instruction. During the EI period, families receive information related to hearing device function, device limitations, troubleshooting techniques and promoting consistent device use. Concurrently, they receive guided instruction related to spoken language development and strategies to foster language skills. The authors suggest that these functions must occur in tandem for children to receive optimal spoken language outcomes.

Does language growth rate improve with EI?

The rate of improvement over time in receptive and expressive vocabulary scores is similar in trajectory for children with and without EI. This result indicates that intervention in the school at CID produced similar gains...
in children who entered school scoring closer to age-appropriate levels after receiving EI and children who started school without EI and were thus further behind in language.

The results for the global language measure showed a different pattern. The children with EI exhibited substantial gains with age, while children without EI showed almost no change in standard score over time. While children with EI were in the process of catching up with age-matched peers with TH in global language skill, children without EI were maintaining a rate of growth similar to age-matched peers with TH, but without closing the gap. Tomblin et al. (2015) reported that the development of morpho-syntax was particularly susceptible to deficits for children with mild to severe hearing loss. We observe a similar trend in this study where children who did not receive EI were struggling to catch up with age-matched peers with TH when morpho-syntax skills were included.

Does language growth rate plateau with age?

The growth of receptive and expressive vocabulary scores with age plateaus over time, due in part to a number of children scoring within age-appropriate levels and reaching ceiling performance while attending CID. There was no significant tendency for growth in global language skills to level off during this period, indicating that global language skills continued to improve substantially with years spent in intensive LSL education.

Do the effects of EI diminish with age?

The advantage of EI over No EI for receptive and expressive vocabulary scores got smaller as children aged. This indicates that the No EI group learned vocabulary at a faster rate than the EI group, so that the effects of EI diminished with age. However, for global language, the advantage of EI persisted throughout the age range measured for this study indicating lasting effects for early services.

Conclusion

Reaching spoken language levels commensurate with age-matched peers with TH facilitates academic success in a general education setting and is a primary goal of LSL intervention programs for children with PHL. Results from the CID sample indicate that achieving this objective depends on the severity of a child’s hearing loss and how early auditory access to speech through amplification and spoken language intervention (typically occurring in tandem) are initiated. Regardless of their degree of hearing loss, children enrolled in EI before 3 years of age exhibited a lasting advantage in the acquisition of spoken language over children who did not have access to EI. Over 80% of children with EI reached language levels commensurate with age-matched peers who have TH by the time of their last test session (average age of 5.7 years). However, at an average age of 7.6 years, only 25% of students without EI caught up with age-matched peers with TH for global language, 31% for receptive, and 56% for expressive vocabulary respectively. Acquiring facility with English semantics, morphology, and syntax presented substantially greater difficulty than learning new vocabulary for students who did not receive EI. These global language skills contribute substantially to the development of reading comprehension, and therefore should receive increased instructional focus in intervention programs designed to prepare children with PHL for age-appropriate placement in regular education classrooms.

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[http://doi.org/10.1097/mao.0000000000001976](http://doi.org/10.1097/mao.0000000000001976)
Developing a Spoken Language Outcome Monitoring Procedure for a Canadian Early Hearing Detection and Intervention Program: Process and Recommendations

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Abstract

Purpose: Routine spoken language outcome monitoring is one component of Early Hearing Detection and Intervention (EHDI) programs for children who are hard of hearing and learning a spoken language. However, there is no peer-reviewed research that documents how spoken language outcome monitoring may be achieved, or what processes EHDI programs can use to develop these procedures. The present article describes the process used by a Canadian EHDI program and the final recommendations that were developed from this process.

Methodology: Through consultation with the program’s stakeholders, consideration of the Joint Committee on Infant Hearing’s recommendations, and drawing on our own expertise in spoken language assessment, we developed an overall framework for monitoring spoken language. Based on the needs of the EHDI program, we conducted a scoping review and critical appraisal of norm-referenced tests to identify candidate tests to use within this framework.

Results: We recommended a two-pronged assessment approach to measuring spoken language outcomes, including program-level assessment and individual vulnerability testing. We identified several tests that have been previously used to measure spoken language outcomes. There was little consistency in how tests were used across studies with no clear indicators as to which tests are the most appropriate to accomplish for which outcome monitoring purposes.

Conclusions: This article reports on the framework and tests used by a Canadian EHDI program to accomplish spoken language outcome monitoring. We highlight different factors that need to be considered when designing spoken language outcome monitoring procedures and the complexity in doing so. Future work evaluating the effectiveness and feasibility of our recommendations is warranted.

Keywords: Spoken language outcome monitoring; Program Evaluation

Acronyms: CASL = Comprehensive Assessment of Spoken Language; CDI = Child Development Inventory; CELF = Comprehensive Evaluation of Language Fundamentals; COSMIN = Consensus Based Standards for the Selection of Health Status Measurement Instruments; DEAP = Diagnostic Evaluation of Articulation and Phonology; EHDI = Early Hearing Detection and Intervention; EOWPVT = Expressive One Word Vocabulary Test; EVT = Expressive Vocabulary Test; GFTA = Goldman-Fristoe Test of Articulation; IHP = Infant Hearing Program; KLPA = Khan-Lewis Phonological Analysis; MBCDI = MacArthur Bates Communicative Development Inventories; (M)CDI = (Minnesota) Child Development Inventory; MSEL = Mullen Scales of Early Learning; PLAI = Preschool Language Assessment Inventory; PLS = Preschool Language Scale; PPVT= Peabody Picture Vocabulary Test; SLP = speech language pathologist; TACL = Test of Auditory Comprehension of Language, VABS = Vineland Adaptive Behavior Scales

Acknowledgements: The authors have no conflicts of interest to declare. This work was funded by the Ontario Ministry of Children, Community and Social Services.

The authors would like to thank the speech-language pathologists, audiologists, and program managers who contributed to the development of these procedures and recommendations. We would also like to thank Kelsi Breton for her work in evaluating articles for inclusion and exclusion.

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Early Hearing Detection and Intervention (EHDI) programs provide family centered support in the pursuit of typical language development (whether signed or spoken) for children who are deaf and hard-of-hearing (DHH; Moeller et al., 2013). For families who elect to pursue language in a spoken modality, EHDI programs have been demonstrated to improve spoken language outcomes (Ching, Day et al., 2013; Moeller, 2000;
Tomblin et al., 2015). Recent research has identified that interventions provided through EHDI programs such as early amplification, high levels of audibility, and support for consistent hearing aid use, are significant predictors of eventual spoken language outcomes and growth in spoken language over time (Tomblin et al., 2015). Comprehensive EHDI programs are gaining increasing international support, and international recommendations have been developed to guide their implementation (Moeller et al., 2013; Joint Committee on Infant Hearing, 2013, 2019). The Joint Committee on Infant Hearing (JCIH) has worked for many years to establish guidelines to ensure consistent and equitable service for children who are DHH and enrolled in different EHDI programs across the United States, and their work has set a standard for EHDI programs worldwide (e.g., the Canadian Infant Hearing Task Force endorses these recommendations). One of the committee’s activities has been the publication of position statements summarizing the current state of the evidence in infant hearing and providing preferred practice recommendations on early identification and intervention for children who are DHH.

Of interest to the present article are JCIH recommendations for routine outcome monitoring of children enrolled in EHDI programs, specifically the monitoring of language outcomes. Because a central aim of EHDI programs is to prevent developmental delays associated with permanent childhood hearing loss, the recommendation for routine monitoring of spoken language development (when this is the mode of communication chosen by the family) is intended to ensure that “a child’s developmental progress is comparable with his or her hearing peers” (JCIH, 2007, p. 909) and within 1 SD of their age or cognitive development on norm-referenced spoken language testing (JCIH, 2013). To meet this expectation, the JCIH recommends that policymakers, service providers, and family members use the results of routine spoken language outcome monitoring to support decision making. For instance, results from spoken language monitoring should be used to inform program evaluation and quality assurance at the program level, support comparison between EHDI programs using national databases, inform intervention planning at the level of the individual child and family, and determine whether a child is or is not meeting developmental milestones (JCIH, 2013, 2019).

However, there is no clear guidance on how EHDI programs ought to accomplish spoken language outcome monitoring, and the concept of spoken language outcome monitoring is poorly defined. Spoken language encompasses a wide range of inter-related skills, some of which a child may or may not struggle with at different ages. Nor do recommendations connect assessment purposes with tests or propose solutions to overcome the psychometric challenges associated with defining acceptable outcomes. Identifying the intended purpose(s) of conducting routine measurement of spoken language outcomes is an essential consideration in selecting the assessment approaches and which tests to use (Daub et al., in press), because different tests may be better suited to different purposes. Furthermore, not all tests are validated to support multiple decisions (Daub et al., 2019) and some assessment purposes are at psychometric odds with one another. For instance, the appropriate composition of a normative sample changes if the test is being used for absolute purposes (i.e., determining whether a child is below age expectations) or relative purposes (determining the severity of a spoken language disorder; Peña et al., 2006). As outlined by JCIH (2007, p. 909), “the primary purpose of regular developmental monitoring is to provide valuable information to parents about the rate of their child’s development as well as programmatic feedback concerning curriculum decisions.” These two decisions (i.e., information about rate of development and programmatic feedback) imply two conflicting purposes: measurement that is sensitive to an individual child’s growth over time and measurement that is comparable between all children in a program. In speech-language pathology, it is traditionally advised to avoid measuring growth with norm-referenced tests because these tests are inherently broad, robust, and stable measures of spoken language constructs that aren’t designed to be sensitive to change in language ability (McCauley & Swisher, 1984). However, relatively new statistics (e.g., item response theory derived scores such as growth scale values) that can be used to measure change over time are increasingly being reported in norm-referenced tests, although these are not yet commonplace (Daub et al., 2017; Daub et al., 2019). Comparing results between groups of children for the purpose of evaluating the broader EHDI program, however, requires that all children in the program are assessed at regular intervals with a consistent measure so that norm-referenced results can be compared.

The present project was born out of our efforts to support a Canadian EHDI program, the Ontario Infant Hearing Program (IHP), which serves children from birth to age 6, in developing a spoken language outcome monitoring procedure that would allow them to fulfill best-practice recommendations. The IHP was developed in 2002 and is a publicly funded EHDI program. The IHP provides universal newborn hearing screening services to all babies born in Ontario and intervention services to children with permanent hearing loss up to the age of 6 years. Spoken language development services for children in the IHP are provided by the publicly funded Ontario Preschool Speech and Language Program until they transition to school services, which can start as early as 3 or 4 years for those who attend junior kindergarten, but does not occur until 6 years of age for others. The IHP provides language development support in the primary language modality (either signed or spoken) as chosen by the family (Moeller et al., 2013) and may include technological intervention (e.g., hearing aids), sign language consultation, or spoken language intervention through speech-language pathology services. However, it is not the case that families are committed to selecting one language modality. Rather, given the publicly funded nature of the program, the
IHP provides funding for families to access services to support a primary language modality and families may pursue additional, privately funded services if, for instance, they wish to raise their child in a bimodal bilingual environment. Similarly, children in the IHP who are learning spoken language may also be raised in homes with two spoken languages. In cases where cochlear implantation is indicated, families access support through a collaboration with a separate publicly funded program and may not be followed by the IHP specifically. As a result, the present article focuses specifically on children who are hard of hearing (HH) and not children who are candidates for cochlear implantation. The IHP aligns its expectations closely with the recommendations put forth by the Canadian Infant Hearing Task Force and the JCIH. Currently, Ontario is one of six Canadian provinces/territories judged to be sufficiently meeting EHDI program standards (Canadian Infant Hearing Task Force, 2019).

Since 2009, spoken language outcome monitoring in the IHP has been conducted using the Preschool Language Scale, 4th ed (PLS-4; Zimmerman et al., 2006) every 6 months (JCIH 2007; 2013). Outcomes were to be tracked for all children for whom families selected spoken language as a primary language modality. This group can include children learning spoken language only or in conjunction with a signed language. Our research team was previously contracted by the IHP to evaluate outcomes using PLS-4 data from two birth cohorts in the program (Daub, 2016; Daub et al., 2017) and were therefore familiar with the previous process, as well as elements of data collection and reporting that were inconsistently implemented across the program. For example, less than 50% of the children in the birth cohorts analyzed did not have PLS-4 scores in the database, and PLS-4 scores were inconsistently scored across children (Daub, 2016). Because the nature of our involvement with the PLS-4 data was post-hoc, it was unclear whether data collection issues stemmed from issues with administration of the PLS-4, data entry/management errors, or errors in extraction from the data management system. The amount of data that were missing for undocumented reasons highlighted the importance of improving upon the previous procedure to support program evaluation.

Around the same time that our team was involved in evaluating the outcome data from previous cohorts, the PLS-4 fell out of print in favor of the Preschool Language Scale, 5th ed (PLS-5; Zimmerman et al., 2011). As a result, the IHP sought to confirm that the PLS-5 would be an adequate replacement, and to evaluate and reconsider their procedure if necessary. At the same time, speech-language pathologists (SLPs) raised concerns about the appropriateness of the PLS-4/PLS-5 and questioned the rationale for its selection.

This article reports on a series of program evaluation and quality improvement projects we conducted to facilitate the IHP’s decision-making about a new spoken language outcome monitoring procedure. These projects began in 2014, and our initial recommendations were shared with the IHP in 2017. We begin by orienting the reader to the overall process we used to develop the procedure (see Figure 1). This includes identifying the IHP’s assessment purposes, developing a framework for assessing outcomes, and identifying tests to use in the framework. We then report on how we identified tests that appropriately fit within the framework, while also balancing needs at the level of both the program and the individual service providers and families.

**Figure 1**

**Process for Developing Proposed Outcome Monitoring Process**

**Step 1: Identifying Assessment Purposes**

**The IHP’s Assessment Purposes at the Program Level**

Our main priority was to collect and maintain data within a provincial database that was appropriate for (a) evaluating the overall expressive and receptive spoken language outcomes of children in the IHP as a group to demonstrate the effectiveness of the IHP, (b) modeling children’s spoken language growth over time to identify ages/stages of development where additional support might be needed, (c) identifying predictors of better, or worse, spoken language outcomes to support quality improvement initiatives, and (d) identifying whether there are differences in outcomes across regions of the province to support resource allocation. IHP management was also cognizant of the importance of clinician’s assessment purposes and minimizing the time and financial burden of spoken language outcome monitoring on service providers to the greatest extent possible. They were also interested in a procedure that could provide clinically useful data about individual children in addition to program-level evaluation.
The IHP’s Assessment Purposes for Individual Children and Families

At the level of the individual child and family, routine assessment of speech and language development should (a) identify children who are performing below age expectations and thus require speech-language development services, (b) allow profiling areas of relative strength and weakness in individual children, thus enabling clinicians to set goals and tailor interventions to meet individual needs at different stages of the child’s development, and (c) allow for evaluation of school readiness and anticipation of academic supports needed to ensure success upon school entry. Because children with permanent hearing loss have ongoing inconsistent access to auditory information, they are at greater risk for difficulties in certain areas of spoken language than others (Moeller, Tomblin, et al., 2007), even if they perform within age expectations on omnibus spoken language tests. Therefore, developing a procedure that is informative to intervention planning for individual children required an approach that probed more deeply than overall spoken language outcomes, specifically those domains of language that are (a) known to be at particular risk in children with permanent hearing loss and (b) predictive of future spoken language outcomes. For children with moderate to severe hearing loss, who are served by the IHP, there are certainly gaps in knowledge about development of specific spoken language domains (Moeller, Tomblin, et al., 2007), but some of the most vulnerable domains in children from birth to 6 years appear to be related to inconsistencies in auditory access, including:


3. Morphosyntactic difficulty, which is suspected to stem from underlying concerns with articulation and phonology (Moeller, Tomblin, et al., 2007)

4. Phonological awareness in the preschool/Kindergarten period (Moeller, Tomblin, et al., 2007)

Matching the Assessment Purpose with the Assessment Method

Achieving individual level purposes requires different assessment approaches and tests than achieving program level purposes. Individual level evaluation requires different tests measuring different vulnerabilities at different stages of development. Program level evaluation requires the same metric and the same or similar tests across programs and over time. To fulfill both of these sets of purposes, it became immediately apparent that there was no single test that would be sufficient.

As a result, we suggested a two-tiered outcome monitoring framework for the IHP: (a) monitoring overall receptive and expressive language development for program-level evaluation purposes using a single test, and (b) targeted individual monitoring of selected areas of speech/language vulnerability (see Figure 2). Although we recognize that concerns in any of these domains do not clearly begin or end at any age, we recommended limiting monitoring to selected areas of speech/language vulnerability using only one or two tests at any one of three developmental time points to minimize the clinical burden of the process. This process was not intended to replace SLPs’ current practices of collecting the information they need to set goals and monitor progress for individual children on their caseload. Our next step was to identify which norm-referenced tests were best equipped to measure overall expressive and receptive spoken language and each of these domains.

Figure 2
Proposed Outcome Monitoring Process

Overall Language Assessment (Birth – Program Discharge):
Goal: Assess overall receptive and expressive language development with an omnibus tool for program outcome evaluation

Key Vulnerability Monitoring (Birth – Program Discharge):
Goal: Measure specific domains of spoken language known to be at risk in children who are hard of hearing

1. Key Vulnerabilities in Infants
   - Goal: Monitor vocalization and babble development for predictors of speech-language delay in individual children

2. Key Vulnerabilities in Toddlers
   - Goal: Monitor speech-language development with attention to speech and first words to identify individual children in need of additional speech-language development support

3. Key Vulnerabilities in Preschoolers/Kindergarteners
   - Goal: Monitor morpho-syntact and emergent literacy/phonological awareness development to tailor intervention and identify additional supports needed for school success in individual children

Step 2: Selecting Tests for Outcome Monitoring

Step 2a) Scoping Review of Norm-Referenced Tests

The purpose of the scoping review was to identify which norm-referenced tests have been previously used in studies of children who are HH and the results obtained using each of these tests. In developing our recommendations, we sought to select amongst tests that have a documented history of use in the peer-reviewed
literature as preliminary evidence that the tests (a) have some ability to differentiate between children who are HH and children with typical hearing thresholds and (b) are sensitive to change over time. Although the original purpose of these studies was not to document test sensitivity to group differences per se, there is a dearth of norm-referenced tests designed specifically to capture the spoken language outcomes of children who are HH. Thus, our scoping review served as our closest approximation of whether a test was likely to be sensitive enough to allow the IHP to detect group differences and change over time, should those differences or changes occur. Our expectation was that narrowing our consideration of norm-referenced assessments to only those that have been documented in the peer-reviewed literature would provide the IHP with benchmarks for spoken language outcomes, and some context to interpret their program's results. We were cognizant that if we selected a set of tests that were not sensitive to group differences, or have not previously been used with children who are HH, then we ran the risk of overestimating the outcomes of children who are HH in the IHP. Inversely, if we selected tests that were very sensitive to the spoken language vulnerabilities of children who are HH, without appropriate research context to demonstrate that these results are reasonable, we ran the risk of underestimating the outcomes of children who are HH. Although age-appropriate outcomes are appropriate goals for individual children who are HH, as a group they have been demonstrated to statistically perform below their same-aged peers but within age-expectations (e.g., Ching et al., 2013). This is not to say that EHDI programs should not strive for spoken language outcomes on par with children who are typically hearing, per JCIH recommendations (2013). However, we did not want to over- or under-estimate the IHP's impact based on artefacts of test selection.

Although EHDI intervention programs provide services to children and families electing to pursue spoken and signed language, and children who are (or are not) amplified with hearing aids or cochlear implants, our scoping review focused on articles reporting results of children who are HH who have been fitted with hearing aids and are learning a spoken language. In Ontario, cochlear implant candidacy represents a unique population who often receive services from a different publicly funded program and their outcomes are not routinely tracked by the IHP. We also restricted our review to outcomes measured in children who are HH from birth to 6 years of age to capture the language development of children who are HH in the program. Our initial review took place in 2016 across three databases (SCOPUS, CINAHL, and PubMed), but we conducted a more recent review across a modified set of databases for the purposes of this article to capture the most up-to-date publications. The results of this review were consistent with our prior review (Oram Cardy & Daub, 2017). Our review was guided by the following research questions:

1. Which tests have been used to measure spoken language in children who are HH and who have been fitted with hearing aids between birth and 6 years?

2. Which tests have been used to compare children who are HH and children with typical hearing, or subgroups of children who are HH? Which tests have detected group differences?

3. Which tests have been used to measure change over time in children who are HH? Which tests have detected change over time?

**Search Strategy**

Five databases were searched in October 2018: CINAHL, Pubmed, EMBASE, ERIC, and PsycInfo. Search terms were developed with the assistance of a subject librarian (see Appendix A for an example search). The search was restricted to include only studies published between 1990 and 2018 to capture research completed during the time in which the evidence supporting universal newborn hearing screening and EHDI programs began to accumulate. Following the search, the titles, abstracts, and full texts of articles were screened for several criteria. First, the article must have been published in English. Second, the article needed to have measured spoken language using a commercially available, English, norm-referenced test. Third, the study was required to report outcome data for children who are HH who wore hearing aids separately from data for children who wore cochlear implants and needed to report data for, at a minimum, a subgroup of children between birth and 6 years, 11 months. Case studies of individual children where group data was not reported were also excluded.

Title, abstract, and full text screening from articles identified through the initial database search were completed by the first author and a trained research assistant to identify articles for full review. All eligibility disagreements were resolved through discussion. Title, abstract, and full text screening from articles identified through forward and backward searching was completed by the first author using the same set of criteria previously described. This process was repeated until no new publications were identified.

The first author extracted from each eligible article: (a) the demographic characteristics of the study population; (b) the norm-referenced test(s), including test version, used; (c) whether group comparisons were made and the results of these comparisons; and (d) whether change over time was evaluated and the results of these evaluations. At this stage, studies were excluded if the norm-referenced test was out of print (i.e., studies using only the Reynell Language Developmental Scales; Reynell & Gruber, 1990). Older versions of tests were included if there is a more recent version available for purchase. Study quality was not evaluated as the purpose of our scoping review was to capture the breadth of tools used with children who are HH and the results found with them.
**Scoping Review Results**

We identified 12,084 non-duplicate articles. Of those, 195 articles were retrieved after title and abstract screening. Finally, data were extracted from 36 articles (see Figure 3, and Supplemental Materials in Appendix B for the data extraction). From these 36 articles, 16 commercially available, norm-referenced tests across multiple versions were identified as having been previously used to measure spoken language outcomes in English-speaking children who are HH. Six of these tests were omnibus language measures, four were language or communication development subscales of broader developmental tests, three were measures of vocabulary, and three were measures of articulation and phonology.

**Figure 3**

*Articles Included for Evaluation*

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<tr>
<th>Database</th>
<th>1990-2018</th>
<th>Citations</th>
</tr>
</thead>
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</tr>
<tr>
<td>Eric</td>
<td>1043</td>
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</tr>
<tr>
<td>CINAHL</td>
<td>1990-2018</td>
<td>1992 Citations</td>
</tr>
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<td>PsychInfo</td>
<td>1990-2018</td>
<td>3403 Citations</td>
</tr>
<tr>
<td>Pubmed</td>
<td>1990-2018</td>
<td>5870 Citations</td>
</tr>
<tr>
<td>Forward &amp; Backward Search</td>
<td>1990-2018</td>
<td>3350 Citations</td>
</tr>
</tbody>
</table>

12084 Non-duplicate citations screened

Inclusion/exclusion criteria applied:
- Measured spoken language using an English, norm-referenced assessment
- Study reported spoken language data for children between birth and 6 years of age
- Study reported spoken language data for children fitted with hearing aids
- Study reported primary research data (i.e., not a review paper)

11889 Articles excluded after title/abstract screen

195 Articles retrieved

Inclusion/exclusion criteria applied:
- Measured spoken language using an English, norm-referenced assessment
- Study reported spoken language data for children between birth and 6 years of age
- Study reported spoken language data for children fitted with hearing aids
- Study reported primary research data (i.e., not a review paper)

157 Articles excluded after full text screen

2 Articles excluded during data extraction (test out of print)

36 articles included

For each test, the following was charted: the number of studies (out of 36) that used the test, whether any study used the test to make group comparisons (regardless of the results of the comparison), whether group differences were detected (out of the number of studies that used the test to evaluate group differences), whether any study used the test to measure change over time, and whether the test detected changes over time (out of the number of studies that used the test to evaluate group differences; see Table 1). Studies varied widely with respect to the ages of children included in the sample, the frequency with which they were assessed, the severity of hearing loss, characteristics of hearing aid amplification, and the demographics of comparison groups (see Supplemental Materials for further details). We identified a distinct lack of overlap in our studies in that no two studies evaluated the same outcomes in similar groups of children who are HH.

Of the 36 studies identified, 30 used 16 different norm-referenced tests to compare spoken language outcomes to other children (i.e., children with typical hearing, with cochlear implants, or with different amplification technologies) or the test’s normative mean. Ten studies evaluated change over time using a variety of analyses (e.g., growth scale values, rates of language development, or linear regression). Six studies evaluated spoken language outcomes using composite scores from multiple tests using factor analyses or multivariate analyses. Only 8 out of the 16 tests were used for both comparing spoken language outcomes to other groups of children and measuring change over time and none of the 8 tests consistently identified both differences between groups and change over time.

**Scoping Review Implications**

The scoping review provided 16 candidate tests for measuring each of the spoken language domains within the outcome monitoring process (see Figure 2). However, one of the tests (i.e., the *Wechsler Preschool and Primary Scales of Intelligence*; Wechsler, 2002) does not primarily...
measure spoken language, and largely measures domains that fall outside SLPs’ scope of practice in the province of Ontario. Therefore, it was excluded from future evaluations. Additionally, the Expressive Vocabulary Test (Williams, 2007) was used once in previous studies as a part of a composite score and was not used in studies making group comparisons or evaluating change over time. Given the lack of data about the Expressive Vocabulary Test’s performance on its own, we excluded it from future evaluations. Our next step was to examine the psychometric properties of each of the 14 candidate tests to determine which ones would be psychometrically appropriate to meet the IHP’s assessment purposes.

Step 2b) Critical Appraisal of Norm-Referenced Tests
After completing the initial 2016 scoping review, the most recent versions of the 14 tests, regardless of whether they were the versions used in studies included in the scoping review, were evaluated using the 2012 version of the Consensus Based Standards for the Selection of Health Status Measurement Instruments (COSMIN; Mokkink et al., 2012) checklist. The COSMIN checklist was developed using an International Delphi study method where experts in fields related to measurement (e.g., epidemiology and statistics) iteratively responded to a series of questions about which measurement properties ought to be evaluated in test design (specifically Health-Related Patient Reported Outcomes, but with application to other tests) and the statistics that should be used to report them. Consensus (greater than 67% agreement) was reached on most major terms (with the exception of structural validity), definitions of each property, and on the taxonomy’s organization. From this taxonomy, the COSMIN team developed quality criteria for both

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Norm-Referenced Test Use in Research with Children who are Hard of Hearing</th>
</tr>
</thead>
<tbody>
<tr>
<td># of studies that used the test for any purpose</td>
<td># of studies that used tests to compare groups</td>
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<tr>
<td>Omnibus language tests</td>
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<td>PLS</td>
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<td>MBCDI</td>
<td>9/36</td>
</tr>
<tr>
<td>CASL</td>
<td>4/36</td>
</tr>
<tr>
<td>PLAI</td>
<td>4/36</td>
</tr>
<tr>
<td>CELF</td>
<td>3/36</td>
</tr>
<tr>
<td>TACL</td>
<td>1/36</td>
</tr>
<tr>
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</tr>
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<td>WPPSI</td>
<td>1/36</td>
</tr>
<tr>
<td>Vocabulary tests</td>
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</tr>
<tr>
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<td>17/36</td>
</tr>
<tr>
<td>EVT</td>
<td>1/36</td>
</tr>
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<td>DEAP</td>
<td>6/36</td>
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<td>KLPA</td>
<td>1/36</td>
</tr>
</tbody>
</table>

Note. Six of the 36 reviewed studies used composite scores as an outcome measure. Multiple editions/versions of tests are combined. CASL = Comprehensive Assessment of Spoken Language; CELF = Comprehensive Evaluation of Language Fundamentals; DEAP = Diagnostic Evaluation of Articulation and Phonology; EOWPVT = Expressive One Word Vocabulary Test; EVT = Expressive Vocabulary Test; GFTA = Goldman-Fristoe Test of Articulation; KLPA = Khan-Lewis Phonological Analysis; MBCDI = MacArthur Bates Communicative Development Inventories; (M)CDI = (Minnesota) Child Development Inventory; MSEL = Mullen Scales of Early Learning; PLAI = Preschool Language Assessment Inventory; PLS = Preschool Language Scale; PPVT = Peabody Picture Vocabulary Test; TACL = Test of Auditory Comprehension of Language, VABS = Vineland Adaptive Behavior Scales.
the methodological quality of studies designed to collect data information about measurement properties, and the measurement properties themselves (Terwee, 2011). For the purposes of developing our recommendations, we focused our evaluation on the quality of the measurement properties reported in the examiner’s manual, but not the methodological quality of the studies designed to report the measurement properties, as it was quite likely that not all examiner’s manuals would report sufficient detail to adequately appraise the quality of the methods themselves.

Critical Appraisal Analysis

To appraise each test, we used a revised version of the COSMIN quality criteria in which we excluded four criteria that were included in the original checklist (criterion validity, cross-cultural validity, responsiveness, and measurement error). Although we agree that these criteria are important to consider, upon review it became clear that the statistics required to evaluate these criteria (e.g., differential item functioning analyses between multiple language versions) were very rarely evaluated in any of the included tests, and evaluating these criteria would not support us in choosing a test amongst the 14 tests we identified. Therefore, each of the 14 tests were appraised with respect to the following: internal consistency, reliability, content validity, construct validity (hypothesis testing), and construct validity (structure). Each domain was assigned one of three ratings (positive, indeterminate, negative) according to the operationalizations of each criterion in the COSMIN checklist. For example, a test was rated as having positive evidence for structural validity if factors explained 50% or more of the variance, indeterminate if explained variance was not evaluated/discussed, or negative if factors explained 49% or less of the variance. For our purposes, we considered a test to have met reasonable criteria if they received a positive rating in at least 4 of the 5 categories.

Critical Appraisal Results

Only eight of the 14 tests met acceptable criteria in 4 of the 5 appraised COSMIN domains (see Table 2). Within each of the test categories (omnibus/language scale, vocabulary, phonology/articulation; Table 2), at least one test met acceptable criteria in 4 of the 5 COSMIN domains. Most tests (12 of the 14) met acceptable criteria for reliability, and all tests reported at least one measure of reliability. Only one test reported weak evidence for validity domains, but most tests were missing validity information. Information about tests’ internal structure was the least frequently reported (only two of the 14 tests) in examiner’s manuals.

Critical Appraisal Implications

Based on our appraisal, we identified eight norm-referenced tests that were largely psychometrically acceptable to select for the spoken language outcome monitoring process. There was not one test with clearly better measurement properties over the others. Our next step was to summarize the administration properties of each of these tests.

Step 2c) Consideration of Administration Properties

We considered various administration properties in summarizing the candidate tests including: the age ranges for which each test had normative data; whether the test covered overall language abilities or subskills; the types of scores that could be calculated (e.g., percentile ranks and/or growth scale values), who was required to administer the test (clinician or caregiver), and the amount of time each test took to administer. Each of the eight acceptable tools had various administration properties that might make the test more, or less, attractive to individual EHDI programs (Table 3). For instance, the PLS-5, Clinical Evaluation of Language Fundamentals, Preschool, 2nd ed. (CELF-P2; Semel et al., 2004) and Comprehensive Assessment of Spoken Language, 2nd ed. (CALS-2; Carrow-Woolfolk, 2017) were all acceptable omnibus language measures, but the PLS-5 provides scores that support measuring change over time (i.e., growth scale values), the CELF-P2 supports profiling different domains of language, and the CASL-2 measures a broader range of language abilities and is appropriate at older ages than either the PLS-5 or CELF-P2. Therefore, consideration of these properties presented us with flexibility in which test(s) to propose. For the purpose of the IHP, tests like the PLS-5 had administration properties that would enable the IHP to achieve more of their outcome monitoring purposes. Specifically, the PLS-5 reported normative data for all age ranges served by the program and also reported growth scale values, which would enrich program level evaluation of growth over time. However, other tests had other relative advantages over the PLS-5. For instance, the MacArthur-Bates Communicative Development Inventories, 2nd ed. (MBCDI-2; Fenson et al., 2007) could be completed by parents without SLPs’ support, and the CELF-P2 supported profiling. Our next step was to triangulate the administrative properties and relative advantage of each test with the evidence for the quality of each test to develop a set of options. We then shared these initial recommendations with the IHP and a panel of expert SLPs who had volunteered their time to provide feedback on the clinical feasibility of our recommendations.

Step 3) Integrating the Evidence into Recommendations

Recommendations for Overall Spoken Language Outcome Monitoring

In accordance with JCIH recommendations, we proposed that all children in the IHP be tested with a standardized measure that compares their spoken language development to that of same-aged children with typical hearing every 6 months during the first 3 years of life, and every year thereafter. Triangulation of the evidence from our scoping review, critical appraisal, and summary of administration properties indicated that the following three measures had the strongest evidence supporting their selection as a measure of overall language abilities: PLS-5, MBCDI-2, and CELF-P2. Both the PLS-5 and CELF-P2 offer the additional advantages of having diagnostic accuracy information with cut-point scores.
Table 2

<table>
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<tr>
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</table>

Note. Ratings included positive evidence (+), indeterminate (?), and negative evidence (-) in meeting COSMIN Criteria. +/- indicates that some, but not all, subtests meet acceptable criteria. Shaded tests received a positive rating in at least 4/5 of the categories.

The PLS-5 covers the full 0 to 6 year age range serviced by the IHP, while the CELF-P2 covers 3 to 6 years, and the MBCDI includes three separate forms that cover 8 to 18 months (MBCDI Words and Gestures), 16 to 30 months (MBCDI Words and Sentences), and 30 to 37 months (MBCDI III). Therefore, the most parsimonious approach would be to use the PLS-5 across the entire age span of the program. However, we have encountered SLPs and scientific experts in the field of permanent childhood hearing loss (e.g., Dr. Mary Pat Moeller, personal communication) who have expressed concerns about the sensitivity of the PLS-5 in the first two years of life. These concerns are consistent with the diagnostic accuracy data reported in the examiner’s manual (Zimmerman et al., 2011). That is, the PLS-5’s diagnostic accuracy does not meet acceptable criterion (≥ 0.80; Plante & Vance, 1994) for detecting language delays in children under 2 years for any cut-score. Therefore, although using the PLS-5 would allow the IHP to evaluate whether children were making significant progress over time, SLPs would be unable to accurately determine whether children were obtaining age-appropriate outcomes and the PLS-5 posed greater clinical burden (i.e., longer administration time) than other candidate tests.

An alternative option could be to use the three separate forms of the MBCDI-2 in the first three years of life and the CELF-P2 thereafter. However, because the subtests and scores on the three MBCDI-2 forms are different, this would prohibit future analysis of developmental growth over time, which “can only be analyzed if the child is assessed with at least some instruments that can be
Table 3
Administration Properties for Currently Available Versions of Psychometrically Suitable Norm-Referenced Tests

<table>
<thead>
<tr>
<th>Age range</th>
<th>Language Areas</th>
<th>Scores Available</th>
<th>Examiner/Respondent</th>
<th>Time (min)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Overall</td>
<td>Subskills</td>
<td>SS</td>
<td>GSV</td>
</tr>
<tr>
<td>PLS-5</td>
<td>0-7 years</td>
<td>✓</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>MBCDI-2</td>
<td>8-18, 16-30, 30-37 months</td>
<td>✓</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>CASL-2</td>
<td>3-6 years</td>
<td>✓</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>CELF-P2</td>
<td>3-6 years</td>
<td>✓</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>PPVT-4</td>
<td>2;6-90 years</td>
<td>✓</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>GFTA-3</td>
<td>2-21 years</td>
<td>✓</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>DEAP</td>
<td>3-8 years</td>
<td>✓</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>KLPA-3</td>
<td>8-21 years</td>
<td>✓</td>
<td>✓</td>
<td>✓</td>
</tr>
</tbody>
</table>

Note. AE = age equivalent; GSV = gross scale value; PR = percentile rank; SS = standard score.

Preschool Language Scale (PLS-5; Zimmerman et al., 2011); MacArthur Bates Communicative Development Inventories (MBCDI-2; Fenson et al., 2007); Comprehensive Assessment of Spoken Language (CASL-2; Carrow-Woolfolk, 2017); Comprehensive Evaluation of Language Fundamentals (CELF-P2; Semel et al., 2004); Peabody Picture Vocabulary Test (PPVT-4; Dunn & Dunn, 2007); Goldman-Fristoe Test of Articulation (GFTA-3; Goldman & Fristoe, 2015); Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd et al., 2006); Khan-Lewis Phonological Analysis (KLPA-3; Khan & Lewis, 2002).

Figure 4
Final Recommendation

<table>
<thead>
<tr>
<th>Program Monitoring</th>
<th>Individual Vulnerability Testing</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>Vocalization/Babbling/Articulation/Phonology</td>
</tr>
<tr>
<td>0.5-1</td>
<td>MBCDI-2 Words &amp; Gestures* (Scores for: Words Understood, Words Produced, Phrases Understood, and Gestures Produced)</td>
</tr>
<tr>
<td>1-1.5</td>
<td>GFTA-3 (Scores for Sounds-in-Words)</td>
</tr>
<tr>
<td>1.5-2</td>
<td>CELF-P2 (Scores for Word Structure) or CASL-2 (Scores for Grammatical Morphemes)</td>
</tr>
<tr>
<td>2-2.5</td>
<td></td>
</tr>
<tr>
<td>2.5-3</td>
<td></td>
</tr>
<tr>
<td>3-4</td>
<td></td>
</tr>
<tr>
<td>4-5</td>
<td></td>
</tr>
<tr>
<td>5-6</td>
<td></td>
</tr>
</tbody>
</table>

Note. CASL-2 = Comprehensive Assessment of Spoken Language; CELF-P2 = Comprehensive Evaluation of Language Fundamentals; EOWPVT = Expressive One Word Vocabulary Test; GFTA-3 = Goldman-Fristoe Test of Articulation; MBCDI-2 = MacArthur Bates Communicative Development Inventories; PLS-5 = Preschool Language Scale.
repeated throughout the target age range” (JCIH, 2013, p. e1334). An additional concern is that only the MBCDI Words and Gestures form includes evaluation of both receptive and expressive language (along with gestures); the remaining MBCDI-2 forms only assess expressive language.

A third option included using the MBCDI-2 Words and Gestures form until 18 months of age, and the PLS-5 thereafter. This would provide scores on the same measure (the MBCDI-2) for the first two testing sessions at the 6-month testing interval, and then PLS-5 scores for all 6-month and 12-month testing intervals beyond 18 months. Under this option, the program would be able to make direct comparisons of growth across all time points except for the one point of transition between the MBCDI-2 and PLS-5 around 18 to 24 months. We felt that this was a reasonable compromise to have a more clinically accepted tool in the earliest years of development, and thus this third option formed the basis for our final recommendation.

**Recommendations for Individual Vulnerability Testing**

Our scoping review and critical appraisal identified norm-referenced tests that have been used with children who are HH and that measure areas that are particularly vulnerable for them. Based on the results of our scoping review and critical appraisal, we recommended a two-pronged approach to assessment for the purposes of supporting individual child/family needs. We recommended that SLPs include assessment of key vulnerabilities associated with the child’s particular age/stage of development (see Figure 4) alongside of their administration of the program-level test of overall language abilities. To reduce the time associated with assessment, and to prevent children from being assessed with more than two norm-referenced tests at a single session, we recommended assessing one area of key vulnerability at each age, even though the ages at which different skills (e.g., articulation and phonology) can be assessed may overlap with other key vulnerabilities. Additionally, in our scoping review we were unable to identify any commercially available test of early vocal development, although some articles (e.g., Ambrose et al., 2014) report on experimental tests that are currently in development. In this regard, we were unable to recommend a specific test for the IHP to use for monitoring early vocal development. In short, we recommended that the IHP provide a set of recommended tests from which SLPs are advised to select. This would support consistency across regions and ensure that only those tests with the strongest evidence are used to assess these key vulnerability areas.

**Consultation with Stakeholders**

We summarized the overall process (program level monitoring and individual vulnerability testing) as well as the three options for overall outcome monitoring and our recommendations for individual vulnerability testing (described above), in a formal written report (Oram Cardy & Daub, 2017). This report was shared with IHP audiological policy development, IHP government leaders, and a team of SLPs who formed an advisory panel. All parties provided written feedback on the report and discussed the recommendations at length through teleconference meetings. Following the revisions to the recommendations, all parties reached agreement on a final procedure (see Figure 4). This procedure included program-level outcome monitoring and individual vulnerability testing. Following final discussion via teleconference, the managerial team ultimately adopted the final spoken language outcome monitoring procedure for implementation in the IHP.

**Discussion**

The present article describes our process for developing a set of spoken language outcome monitoring recommendations to support a Canadian EHDI program, the Ontario IHP, in fulfilling best practice recommendations. To date, there has been limited guidance in the literature on (a) the best way to approach the development of a spoken language outcome monitoring process or (b) how to accomplish all of the facets of spoken language outcome monitoring in a way that provides statistically appropriate evidence, is implementable across entire EHDI programs, and meets the competing needs of different stakeholders. Our expectation is that documenting our steps in this process and the recommendations that resulted will not only provide a general framework and example for other EHDI programs, but also highlight the previously undiscussed challenges of designing such a procedure.

Our process was grounded in the initial JCIH (2007, 2013) recommendations for spoken language as well as consideration of the International Consensus work on best practice principles (Moeller et al., 2013). From this foundation, we considered the purposes of spoken language outcome monitoring from the perspective of various IHP stakeholders to clarify the assessment purposes our process would need to fulfill. Using these purposes, we conducted a scoping review to identify a set of candidate norm-referenced tests that have been previously used to fulfill these assessment purposes and appraised the psychometric quality of the most recent versions of these tests. We then considered the administration properties of the tests that we rated as psychometrically acceptable and integrated all sources of evidence with our originally described assessment purposes. This allowed us to develop a set of recommendations to share with IHP stakeholders, who ultimately decided to adopt them. We expect that our work will be of interest to other EHDI programs and service providers who work with children who are DHH by documenting our process in developing our recommendations, the recommendations themselves, and the final procedure adopted by the IHP. Our results highlight the unique challenges faced when trying to develop a process for spoken language outcome monitoring, guide future research designed to refine the development process, and contribute to a body of literature that provides guidance for EHDI programs looking to fulfill best practice recommendations.
Our next step is to design implementation materials and conduct pilot projects to evaluate the new procedures for both overall spoken language monitoring and individual vulnerability monitoring. These pilot projects are intended to identify barriers and facilitators to implementing the new recommendations in clinical practice, and to allow us to refine our process into one that is most sustainable and clinically feasible before program-wide launch. We anticipate that the results of these pilot projects will similarly support discussions of spoken language outcome monitoring in EHDI programs and highlight the inherent complexity in accomplishing these goals.

We do not intend to assert that our process or final recommendations are a gold standard for spoken language outcome monitoring and should be adopted by other EHDI programs. Rather, we believe that our work uniquely highlights the challenges in accomplishing spoken language outcome monitoring and may be a valuable foundation for EHDI programs looking to develop, or refine, their spoken language outcome monitoring procedures. Our projects were developed through the lens of the Ontario IHP, and other EHDI programs might have different priorities for spoken language outcome monitoring, amongst other needs. In our case, the IHP sought a process that would allow them to use the data to evaluate whether children across the province are making progress in their spoken language over time, whether they are meeting age-appropriate expectations by the time they are discharged from the program, and whether they have the spoken language skills they need at discharge to be prepared for school. Necessarily, fulfilling these purposes required the use of multiple tests that are sensitive to multiple domains of language, and that were norm-referenced to establish whether a child was performing within or below age-expectations.

An additional priority was selecting norm-referenced tests from those that have been previously used in research with children who are HH to contextualize the outcomes in the IHP with the peer-reviewed literature. The Ontario IHP is publicly funded and managed under a larger provincial division also responsible for the allocation of resources across multiple programs from a single budget. We were wary of selecting norm-referenced tests without a documented history of use in the literature because it has been demonstrated that children who are HH often score within age-expectations (and close to the test’s normative mean of a standard score of 100), but statistically lower than matched groups of children with typical hearing (e.g., Tomblin et al., 2015). In this case, using a standard score cut-off recommended by a norm-referenced test was not sufficient to describe program outcomes. We were aware that spoken language outcome data could be used by policy makers to make funding decisions and that there was a risk of misinterpreting program level outcomes as being insufficient to continue funding. We were also aware that EHDI programs are precariously positioned in Canada: many EHDI programs are in development, and some have seen declines in support from previous years (Canadian Infant Hearing Task Force, 2014; 2019). In the Canadian context, statistically sound outcome data from one EHDI program has the potential to provide evidence to influence other provincial or national funding priorities. Therefore, it was critical to develop a process that we could connect to the peer-reviewed literature to evaluate whether the IHP was performing on par with documented outcomes in other EHDI programs.

Even within the context of the Ontario IHP, our recommendations remain limited in a number of respects. Canada has two official languages (English and French) and many regions in the province are densely populated, multicultural areas where residents speak languages other than these. We focused our reviews and recommendations on measuring outcomes for children who are HH from English speaking families, in part, due to a dearth of norm-referenced tests that have been validated in other languages to include in our scoping review and critical appraisal. Certainly, many (but not all, i.e., the MBCDI-2) of the tests we selected for our current recommendations have not been normed in French, even if there are translated versions (i.e., the PLS-5). To fulfill clinical assessment needs, we have advised SLPs to continue using the tools they typically would for children for whom English is not a primary language, although their outcomes will not be able to be evaluated at the program-level in the provincial database. This raises concerns about equitable service provision—regardless of the language their child is learning, families deserve to know whether their child is progressing as expected in response to intervention. Solutions and next steps, such as collecting local normative data on translated versions, are under discussion. Until norm-referenced assessments for these groups of children exist, EHDI programs will need to identify other creative solutions to evaluate spoken language outcomes and rely on less formal assessments. Our general framework could be modified to support identifying informal assessments or interview tools, although a different process for critically appraising the approaches would be needed.

It is likely that there are other important considerations requiring attention in other EHDI programs that we did not account for in our process for the Ontario IHP. For example, EHDI programs in which outcome data are not likely to be used to support funding decisions may feel comfortable considering the use of norm-referenced tests without a history of previous peer-reviewed use. Additionally, our process did not consider the spoken language outcomes of children with cochlear implants because many are served by a different program in the province of Ontario, but other EHDI programs may wish to do so. Furthermore, our process did not attend to the sensitivity and specificity cut-off scores for language impairment on the tests we evaluated because there is no mandate in Ontario for children to perform below a certain threshold (e.g., -2 SD below the mean) to be considered eligible for receiving SLP services outside of EHDI programming. This is certainly the case in some American state education departments (Spaulding et al., 2012), thus, EHDI programs located in regions with similar...
Future work could extend the methods used here to developmental (Cupples, Ching, Crowe, Day, et al., 2014) and developmental delay) may further impact language to 40% of children who are HH who have additional challenges to stakeholders looking to interpret their psychometric properties of the tests they are using or risk performing within age-expectations. Therefore, procedures, the role of norm-referenced tests versus other sources of assessment information (e.g., criterion referenced testing for goal setting), and ways to ensure equity in how these sources of information are collected and used across programs. First, outcomes from two norm-referenced tests are not directly comparable and the operationalization of “within age-expectations” is entirely dependent on the statistical properties of the norm-referenced test in question. Although the JCIH recommends that children who are HH should score within -1 SD of the mean or higher on norm-referenced tests (2013), this recommendation does not acknowledge the unique sensitivity and specificity of individual tests at individual scores (Spaulding et al., 2006). For example, both the PLS-5 and the CELF-P2 have the greatest diagnostic accuracy at -1 SD (Zimmerman et al., 2011; Semel et al., 2004), but the GFTA-3 maximizes diagnostic accuracy at -1.5 SD (Goldman & Fristoe, 2015). As such, children with typical hearing thresholds and typical language development can be expected to score between -1.49 and -1 SDs below the mean on the GFTA-3. If stakeholders apply the -1 SD cut-off as the expectation on tests that are less accurate at -1 SD, they may be inadvertently holding children who are HH to a higher standard than their peers with typically developing hearing. In other words, defining age-appropriate outcomes for individual children, and appropriate outcomes for children who are HH as a group, is confounded with the psychometric properties of norm-referenced tests (Spaulding et al., 2006). These confounds pose significant challenges to stakeholders looking to interpret their population level outcome data. A program that elects to use the PLS-5 to measure outcomes might appear to have better outcomes (i.e., within -1 SD of the mean) than a program that elects to use a test with a -1.5 SD cut-off, even though the children in both programs might be performing within age-expectations. Therefore, procedures for measuring outcomes must consider the unique psychometric properties of the tests they are using or risk generating data that suggests their program is failing to meet JCIH benchmarks.

These concerns with defining age-appropriate outcomes and interpreting results are compounded when we consider applying spoken language outcome monitoring to different groups of children, including those 20% to 40% of children who are HH who have additional diagnoses, some of which (e.g., autism, cerebral palsy, and developmental delay) may further impact language development (Cupples, Ching, Crowe, Day, et al., 2014). Future work could extend the methods used here to identify studies examining language outcomes in children with an additional diagnosis, with and without hearing loss. This would provide context to any program looking to report on the results of children who are HH with additional disabilities.

A second challenge with accomplishing spoken language outcome monitoring pertains to the clinical feasibility of accomplishing all necessary assessment purposes. Many norm-referenced tests are not developed to serve multiple assessment purposes, and their use is best restricted to interpreting whether a child is, or is not, within age-expectations. This creates challenges for accomplishing the diverse purposes that spoken language outcome monitoring is intended to fulfill (e.g., treatment planning and evaluating EHDI programs broadly). Some of these purposes can certainly be accomplished through other forms of assessment (e.g., criterion referenced assessment, language sample analysis), and neither we, nor the JCIH (2013), argue that norm-referenced assessments should be the only component of a spoken-language outcome monitoring battery. Certainly, SLPs will need to rely on other sources of information to develop their therapy plans. However, the addition of a standard norm-referenced process to fulfill program-level evaluation goals adds lengthy tasks to SLPs’ assessment time and it is unknown whether it is feasible for SLPs to collect, interpret and integrate all of the necessary sources of information needed to fulfill spoken language outcome monitoring recommendations. It is widely accepted that whether research evidence or new recommendations will be successfully used in clinical practice is influenced by numerous factors within the clinical context (e.g., Dobrow et al., 2004; Graham et al., 2006) such as time, caseload, and clinician factors (e.g., beliefs, knowledge, skills) above and beyond the quality of the research evidence or recommendation itself. Accomplishing spoken language outcome monitoring in EHDI programs is complicated not only by limited evidence to guide development of procedures, but also by a lack of evidence to support implementation of these procedures. To our knowledge, there is only one peer-reviewed paper, published by our research group (Cunningham et al., 2019) that has evaluated SLPs’ perceptions of the barriers to implementing spoken language outcome monitoring in an EHDI program. In Cunningham’s investigation, time for additional testing was a primary concern. Additional work is needed to evaluate the feasibility of our recommendations specifically, and spoken language outcome monitoring broadly, as well as to develop implementation interventions that result in effective, sustained uptake of spoken language outcome monitoring procedures.

Conclusion

Guidance for how to best implement spoken language outcome monitoring recommendations (JCIH 2007; 2013) is lacking, and EHDI programs face significant barriers to developing procedures that fulfill best-practice recommendations. The present article describes a series of projects, conducted as part of program evaluation and
quality improvement for the Ontario IHP, to develop a spoken language outcome monitoring procedure using a scoping review and critical appraisal of candidate norm-referenced tests. We expect that the process we used, the recommendations we developed, and the challenges we encountered, will be informative to other EHDI programs looking to develop their own procedures. Final recommendations included developing a two-tiered assessment battery measuring overall spoken language outcomes and key areas of spoken language vulnerability. Future work evaluating the appropriateness of these recommendations, whether the data collected is sufficient to fulfill our intended purposes, the feasibility of our recommendations, and ways to implement them into clinical practice are needed.

References
Appendix A

CINAHL Search Strategy

#1 (MH “Outcome Assessment”) OR (MH “Outcomes (Health Care)”) OR (MH “Treatment Outcomes”)

#2 (MH “Child, Disabled”) OR (MH “Child, Preschool”) OR (MH “Child Health”) OR (MH “Child Development Disorders”)


#4 (MH “Language”) OR (MH “Speech and Language Assessment”) OR (MH “Rehabilitation, Speech and Language”) OR (MH “Language Disorders”)

#5 (MH “Outcome Assessment”) OR (MH “Outcomes (Health Care)”) OR (MH “Treatment Outcomes”)

#6 (MH “Child, Disabled”) OR (MH “Child, Preschool”) OR (MH “Child Health”) OR (MH “Child Development Disorders”)


#8 (MH “Language”) OR (MH “Speech and Language Assessment”) OR (MH “Rehabilitation, Speech and Language”) OR (MH “Language Disorders”)

#9 S5 AND S6 AND S7 AND S8

#10 (MH “Clinical Assessment Tools”) OR (MH “Speech and Language Assessment”) OR (MH “Outcome Assessment”) OR (MH “Functional Assessment”)

#11 (MH “Instrument Validation”)

#12 (MH “Clinical Assessment Tools”)

#13 (MH “Language Tests”)

#14 ((MH “Language Tests”)) AND (S1 OR S10 OR S11 OR S12 OR S13)

#15 (((MH “Language Tests”)) AND (S1 OR S10 OR S11 OR S12 OR S13)) AND (S5 OR S14)
Appendix A (cont.)

#16 (((MH “Language Tests”) AND (S1 OR S10 OR S11 OR S12 OR S13)) AND (S5 OR S14)) AND (S6 AND S7 AND S8 AND S15)

#17 (MH “Measurement Issues and Assessments”)

#18 (((MH “Measurement Issues and Assessments”) AND (S1 OR S5 OR S10 OR S11 OR S12 OR S13 OR S17))

#19 ((MH “Measurement Issues and Assessments” OR S1 OR S5 OR S10 OR S11 OR S12 OR S13 OR S17)

#20 (((MH “Measurement Issues and Assessments” OR S1 OR S5 OR S10 OR S11 OR S12 OR S13 OR S17)) AND (S3 AND S4 AND S6 AND S19)

#21 (((MH “Measurement Issues and Assessments” OR S1 OR S5 OR S10 OR S11 OR S12 OR S13 OR S17)) AND (S3 AND S4 AND S6 AND S19)

#22 (MH “Infant”) OR (MH “Infant Development”)

#23 (MH “Early Childhood Intervention”)

#24 ((MH “Early Childhood Intervention”)) OR (S6 OR S22 OR S23)

#25 ((MH “Early Childhood Intervention”)) OR (S6 OR S22 OR S23)) AND (S7 AND S8 AND S19 AND S24)
Appendix B

Supplemental materials describing the 36 studies can be found on Open Sciences Framework https://osf.io/ncm23/?view_only=1455217c19c44e3881e4628ed252fe3a

Details such as study authors, tests used, sample characteristics, and study purposes are laid out in an easy-to-read table. We also list whether the authors included composite scores, made group comparisons, noted informal differences, and evaluated change over time. Finally, we noted if the study had statistically significant or significant results or if they included other analyses.

Supplemental Materials References


It may bring comfort to know what specific things your provider (audiologist, health care clinician, early intervention specialist, etc.) is doing to keep you and your baby safe. Your provider may also ask you to take certain steps to keep them and their staff safe. Many providers are calling families prior to their appointment to discuss safety.

**Emerging Solutions: How to Keep You, Your Baby, and Your Provider Safe During COVID-19**

**Staying Safe During Your Appointment**

During the call with your provider, consider asking:

1. If doing a hearing screening only, do you have screening options other than us entering the building (e.g., screening in car)?
2. If there is paperwork to be filled out, can you send it to me ahead of time?
3. When I arrive, are there specific instructions (e.g., phone before I enter the building)?
4. Is there a limit to who can come to the appointment with me and my child?
5. Is there a limit to the number of people who can be in the waiting area?
6. Are there health screenings (e.g., temperature) of patients upon arrival?
7. How are public areas being cleaned (e.g., waiting rooms, restrooms, food service areas) and how often?
8. How do you screen yourself or staff for wellness (e.g., temperature)?
9. What protective gear (e.g., gloves, masks) does the provider and his/her staff use?
10. How is equipment (e.g., screening, diagnostic) cleaned or replaced between patients?
11. How can I help keep you and your staff safe?
   - Would you like me to wear a face mask?
   - If the clinic serves both sick and well patients, how will you handle that?
   - Anything else?

**If You Decide to Cancel or Reschedule**

Even though your provider is taking steps toward safety, if you still do not feel comfortable with an in-person appointment, you may want to think about and/or take action in the following ways:

1. Have you talked to your provider about:
   - Your safety concerns?
   - Additional safety strategies that would make you more comfortable to attend an appointment?
2. Would it help to talk to another parent who has recently had the experience of an in-person appointment?
3. If you plan to cancel or reschedule, and you have an appointment scheduled, please call and let your provider know at least 48 hours in advance (or within the timeframe outlined by your provider). Not showing up impacts the schedule of the provider and his/her staff.
4. If you plan to reschedule your appointment:
   - Ask your provider how far out they are scheduled.
   - Have you balanced your concerns with safety with the amount of time that will pass until you are able to be seen by your provider?
   - Does the delay in going to the appointment impact the services your child needs?
5. Ask your provider if they can do a video visit by a secured system.

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**Additional Resources**

- [https://handsandvoices.org/fl3/topics/fam-fam-support/need-support.html](https://handsandvoices.org/fl3/topics/fam-fam-support/need-support.html)
- [http://www.infanthearing.org/COVID-19/index.html#support](http://www.infanthearing.org/COVID-19/index.html#support)

**We went to the audiologist at our CI Center last week, and I’ve been VERY anxious about COVID. It was a VERY comfortable experience!! The CI Center called us when they were ready to re-open. They were very transparent about the new policies (masks, temp checks, etc.) and wanted me to know that I could cancel at the last minute if I wasn’t comfortable. There was no waiting room—only waiting in the vehicle was allowed. There were cones lined up in the parking lot with phone numbers and spot numbers on them. You let them know what spot you were parked at, and they came out, with PPE on, with extra masks if we didn’t have any. They took our temperatures and asked us some questions. They gave us hand sanitizer, and we went into the appointment. LOTS of sanitizer was used by the audiologists, and everything that was touched was thrown away or set aside for sterilization. We didn’t need to check out. Everything was done over the phone after the appointment. It was a LOVELY experience for this COVID-anxious mama!!

—Michelle Thomas, Parent, Michigan


Evaluating Pennsylvania’s Newborn Hearing Screening Program

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²Division of Newborn Screening and Genetics, Pennsylvania Department of Health, Harrisburg, PA

Abstract

Scope: Pennsylvania’s Newborn Hearing Screening (NBHS) program is a critical state-run program that is imperative for the goal of early identification of children with hearing loss. The purpose of this study was to evaluate Pennsylvania’s administration of the NBHS, as well as analyze Pennsylvania’s adherence to the Joint Committee on Infant Hearing (JCIH) 1-3-6 Guidelines.

Methodology: Records from 131,832 newborns born in 2018 were analyzed for this study. Descriptive statistics were used to determine outcomes related to the JCIH guidelines. Prevalence of hearing loss and odds ratios were calculated to determine risks of hearing loss in the 2018 newborn population.

Conclusions: The findings suggest that Pennsylvania has a strong adherence to the 1-3-6 guidelines, with an average timeframe of 3.04 days from birth to screening, 75.39 days from birth to diagnosis, and 174.2 days from birth to early intervention enrollment. The information from this study will be used for future program development, as well as to identify areas of improvement within the Commonwealth.

Keywords: newborn hearing screening, state outcomes, JCIH guidelines

Acknowledgment: The authors have no relevant conflicts of interest.

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Since the 2000 recommendation by the Joint Committee on Infant Hearing (JCIH); a guideline recommending that all infants born in the United States are screened for hearing loss by one month of age, diagnosed by three months of age, and enrolled in early intervention (EI) by six months of age; the number of infants screened has increased dramatically. The Centers for Disease Control and Prevention reported that in 2016, roughly 98% of infants born in the United States underwent a newborn hearing screening at birth. Although this statistic is very reassuring, there remain gaps in data related to diagnostic assessments and later EI enrollment. These gaps are often attributed to incomplete or inconsistent local data (Alam et al., 2016).

In a study by Uhler et al. (2014), Early Hearing Detection and Intervention (EHDI) coordinators from across the United States were surveyed on the state or territory structures in place to track diagnostic, amplification, EI, and medical outcomes in children screened for hearing loss. Their results found that only 31.25% of those surveyed had a database in place that contained information regarding assessment and audiology follow-up data. The researchers attribute difficulties following up with screened infants to limited staff capacity as well as limitations in obtaining funding for database creation and maintenance. In addition to the findings by Uhler et al. (2014), Shulman et al. (2010) identified communication between hospitals and newborn hearing screening (NBHS) staff as a major challenge in optimizing the EHDI reporting program throughout the United States. In this study, staff from NBHS programs were asked to rank the quality of data reported from hospitals. The researchers found that staff largely reported that data was poor or good compared to very good or excellent.

In 2001, the Pennsylvania State Assembly passed the Infant Hearing Education, Assessment, Reporting and Referral (IHEARR) Act. This act required providing newborn hearing screenings to all infants within the Commonwealth as well as programs for follow-up services. Newborn hearing screenings were implemented statewide in July 2002. The IHEARR Act additionally called for the creation of a newborn hearing screening advisory board, consisting of organizations, stakeholders, and professionals to monitor hearing health outcomes for all children born within the Commonwealth.
The Pennsylvania Division of Newborn Screening and Genetics implemented a policy shift to track the outcomes of all babies screened in the state through a centralized, web-based monitoring system, called iCMS. All activities related to the NBHS, including screening results, tools used, diagnostic evaluation results, and EI enrollment, were tracked through iCMS. This system was fully implemented to track the outcomes of children born in 2018 and later. The purpose of this study is to assess the implementation of Pennsylvania’s NBHS program and its adherence to the JCIH 1-3-6 guidelines, using the data received through the iCMS system.

**Methods**

For this study, infant records of those born between 1/1/2018 and 12/31/2018 were assessed through the iCMS system. Inclusion for this study was limited to babies native to Pennsylvania, as identified by maternal zip code and county. Descriptive analyses were used to determine outcomes related to screening, diagnosis, and EI enrollment. All statistical analyses were completed using R statistical analysis software (R Core Team, 2019). Apparent prevalence of hearing loss was calculated using 89% sensitivity and 92% specificity, the most conservative estimates from Butcher et al. (2019).

**Results**

A total of 131,832 (67,746 males, 64,083 females, 3 unspecified) newborn screening records were analyzed for this study. Of the 131,832 total records; 125,381 infant records reported information regarding birth setting. Of those born in Pennsylvania, 125,627 infants (95.3%) were seen in inpatient settings, and 6205 (4.7%) were assessed in outpatient locations. Families of infants who did not pass the initial hearing screening prior to discharge from the birthing center were instructed to follow-up at an outpatient clinic. Infants were further followed through the iCMS system, where each case was kept open until a final diagnosis was rendered in the case of normal hearing, or the infant was enrolled in EI services, in the case of a diagnosis of permanent hearing loss. Outpatient centers and midwifery services were further used for infants born at home.

**Screening Results**

Figure 1 illustrates the ultimate screening results for newborns born in Pennsylvania in 2018. Of all screenings, 119,683 (90.1%) occurred in well-baby nurseries, while 11,884 (9.0%) occurred in the neonatal intensive care unit (NICU). Pennsylvania had an overall pass rate of 96.9%, with 127,694 babies passing bilaterally. A total of 1148 babies, less than 1%, referred on the screening in at least one ear. Roughly 2% (2,439) of babies recorded did not have a completed screen. The largest reason for this lack of screen can be attributed to parent refusal. Babies who had their final screening in inpatient settings tended to have a higher rate of passing (97.8%) compared to those who were screened in outpatient settings (77.9%). Table 1 illustrates the difference in outcomes based on screening setting. Although babies screened in the outpatient setting tended to have a higher refer rate (5.0%) than those tested in inpatient screenings (0.7%), there was a substantially higher percentage of children who ultimately were not able to complete the screen in outpatient. Most significantly, the parent refusal rate for outpatient screens was 10.9% compared to 0.6% in inpatient screenings. There was also a marked difference in the time it took to obtain the newborn hearing screen.

Initial inpatient screenings were conducted an average 2.59 (± 9.32) days following birth. Outpatient initial screens were conducted an average of 10.62 (± 18.73) days after birth. The length of time from initial to final screenings was substantially different for inpatient screenings when compared to outpatient screenings. On average, the final inpatient hearing screen took place 3.04 (± 11.22) days after birth, while it took 32.36 (± 46.67) days to complete the final hearing screen on outpatient infants. Pennsylvania’s average time frame was 4.39 days (± 16.06) from birth to final screening completion for all babies, regardless of screening setting.

![Figure 1](image.png)

**Total Results of 2018 Newborn Hearing Screening Program in Pennsylvania**
Table 1
Newborn Hearing Screening Outcome Based on Screening Setting

<table>
<thead>
<tr>
<th>Outcome</th>
<th>Count (n)</th>
<th>Percent (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Inpatient</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pass</td>
<td>122,799</td>
<td>97.7%</td>
</tr>
<tr>
<td>Refer</td>
<td>840</td>
<td>0.7%</td>
</tr>
<tr>
<td>Parent Refusal</td>
<td>769</td>
<td>0.6%</td>
</tr>
<tr>
<td>Not Screened (Other)</td>
<td>602</td>
<td>0.5%</td>
</tr>
<tr>
<td>Expired</td>
<td>617</td>
<td>0.5%</td>
</tr>
<tr>
<td><strong>Outpatient</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pass</td>
<td>4,794</td>
<td>77.3%</td>
</tr>
<tr>
<td>Refer</td>
<td>308</td>
<td>5.0%</td>
</tr>
<tr>
<td>Parent Refusal</td>
<td>679</td>
<td>10.9%</td>
</tr>
<tr>
<td>Not Screened (Other)</td>
<td>408</td>
<td>6.6%</td>
</tr>
<tr>
<td>Expired</td>
<td>16</td>
<td>0.3%</td>
</tr>
</tbody>
</table>

Some infants (5,482) had their first screening completed in an inpatient setting and had a follow-up screening completed at an outpatient facility. This number includes infants that eventually passed their newborn hearing screening but may have referred on their first screen. On average, it took 35.1 (±48.51) days for these babies to receive a final screening outcome. Babies who were screened in outpatient settings were over 10 times more likely to refer on their final screen compared to those screened in an inpatient setting (OR = 10.46, 95% Confidence Interval: [CI] 9.13–11.97).

Of the 119,683 babies screened in well-baby nurseries, 97.1% passed their newborn hearing screening and 0.7% referred. This pass rate was higher compared to those screened in the NICU, who had a 94.1% pass rate and 2.2% referral rate. Those screened in the NICU were over three times more likely to refer on the NBHS compared to those screened in well-baby units (OR = 3.28, 95% CI: 2.86–3.77). There was no association between well-baby nursery screening and referral on the NBHS (OR = 1.0). A total of 2,405 babies were screened using midwife services in 2018. Of those infants, 1,380 (57.4%) passed their screening, 10 (0.4%) referred on their final screening, and 1,015 (42.2%) did not complete a final screen due to parent refusal (n = 763), missed appointments (n = 246), or similar reasons. Six infants had no information regarding their screening status.

**Diagnostic Assessment Results**

Of the 1,067 babies who referred on their newborn hearing screen, 884 (82.8%) were seen for a diagnostic follow-up. Of these infants, 664 received a final diagnosis of normal bilateral hearing. This finding suggests a false positive rate of 0.5%. Table 2 shows the diagnostic outcomes for those found to have permanent hearing loss either unilaterally or bilaterally. The prevalence of permanent hearing loss among newborns was 1.76 per 1000 (95% CI: 1.5–2.0) in 2018, with 233 children diagnosed with hearing loss by their final evaluation. Bilateral and unilateral hearing diagnoses were equally common, with 108 (46.4%) children diagnosed with a bilateral hearing loss, compared to 110 (47.2%) children diagnosed with a unilateral hearing loss. The average length of time from birth to the completion of the diagnostic assessment was 75.39 (±72.3) days. Analysis of the severity of hearing impairment showed the highest representation of hearing loss as either a moderate (21.7%) or profound (21.1%) hearing loss among those with a classified severity. In total, 176 of the 233 (75.5%) infants diagnosed with permanent hearing loss had a severity classified in at least one ear. For 38 (22.9%) of these infants, the classified severity was unknown, indicating that more diagnostic testing was necessary before making a final classification. Table 3 demonstrates the severity rating for children with both unilateral and bilateral permanent hearing loss.

Of the 884 infants that completed a diagnostic assessment, 683 were born in well-baby nurseries. Of these infants, 78.7% were diagnosed with normal hearing, and 21.1% were diagnosed with some form of hearing loss in at least one ear. For those screened in the NICU (n = 199), 62.3% had normal hearing, while 37.7% were diagnosed with some level of hearing loss in at least one ear. Those born in well-baby nurseries were nearly half as likely to be diagnosed with a hearing loss compared to those screened in the NICU (OR = 0.44, CI: 0.31–0.62).

**Early Intervention**

As of May 2020, 180 of the 233 children diagnosed with a hearing loss from the newborn hearing screen in 2018 were referred for EI services. At this time, 137 (76.1%) children have been enrolled in EI. Table 4 illustrates the status of children being followed for early intervention services. Data from the 137 children suggests that the average length of time from birth to the generation of an EI referral is 121.4 (±107.1) days. The average length of time from birth to the enrollment in early intervention services is 174.2 (±116.5) days.

Table 2
Count of Children Diagnosed with Unilateral/Bilateral Permanent Hearing Loss (HL) in 2018

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>Right Ear HL Only</th>
<th>Left Ear HL Only</th>
<th>Bilateral HL</th>
<th>Total</th>
<th>Prevalence (per 1000)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Auditory Neuropathy</td>
<td>1</td>
<td>1</td>
<td>4</td>
<td>6</td>
<td>0.045509709</td>
</tr>
<tr>
<td>Mixed Loss</td>
<td>7</td>
<td>6</td>
<td>6</td>
<td>19</td>
<td>0.144114078</td>
</tr>
<tr>
<td>Permanent Conductive Loss</td>
<td>8</td>
<td>5</td>
<td>4</td>
<td>17</td>
<td>0.128944175</td>
</tr>
<tr>
<td>Sensorineural Loss</td>
<td>35</td>
<td>33</td>
<td>98</td>
<td>166</td>
<td>1.259101942</td>
</tr>
<tr>
<td>Unknown Loss</td>
<td>3</td>
<td>5</td>
<td>17</td>
<td>25</td>
<td>0.189623786</td>
</tr>
</tbody>
</table>
Table 3
Degree of Permanent Hearing Loss in Study Sample

<table>
<thead>
<tr>
<th>Degree</th>
<th>Unilateral</th>
<th>Bilateral</th>
<th>Total</th>
<th>Percent</th>
</tr>
</thead>
<tbody>
<tr>
<td>Slight</td>
<td>2</td>
<td>1</td>
<td>3</td>
<td>1.8%</td>
</tr>
<tr>
<td>Mild</td>
<td>4</td>
<td>22</td>
<td>26</td>
<td>15.7%</td>
</tr>
<tr>
<td>Moderate</td>
<td>11</td>
<td>25</td>
<td>36</td>
<td>21.7%</td>
</tr>
<tr>
<td>Moderately-</td>
<td>16</td>
<td>11</td>
<td>27</td>
<td>16.3%</td>
</tr>
<tr>
<td>Severe</td>
<td>10</td>
<td>27</td>
<td>11</td>
<td>6.6%</td>
</tr>
<tr>
<td>Profound</td>
<td>8</td>
<td>27</td>
<td>35</td>
<td>21.1%</td>
</tr>
<tr>
<td>Unknown</td>
<td>9</td>
<td>29</td>
<td>38</td>
<td>22.9%</td>
</tr>
</tbody>
</table>

Note. Percent indicates percent of total diagnosed.

Table 4
Status of Children Monitored for Early Intervention Services

<table>
<thead>
<tr>
<th>Status</th>
<th>Count</th>
<th>% Followed</th>
<th>% PHL</th>
</tr>
</thead>
<tbody>
<tr>
<td>Enrolled</td>
<td>137</td>
<td>76.1%</td>
<td>58.8%</td>
</tr>
<tr>
<td>Pending</td>
<td>12</td>
<td>6.7%</td>
<td>5.2%</td>
</tr>
<tr>
<td>Refused</td>
<td>4</td>
<td>2.2%</td>
<td>1.7%</td>
</tr>
<tr>
<td>Unknown</td>
<td>27</td>
<td>15.0%</td>
<td>11.6%</td>
</tr>
</tbody>
</table>

Note. PHL = permanent hearing loss.

Discussion
The findings from this analysis suggest that Pennsylvania largely meets the JCIH 1-3-6 guidelines. Newborns screened in-hospital usually receive an initial hearing screen within the first 12 to 24 hours after birth, a number that is reflected in the average in-patient screening time of 3.28 days. This number increases significantly and exceeds the target of screening by one month of age in the outpatient screening population. This increase in screening time, as well as the increased no-screen rate among outpatient events can potentially be attributed to the geographic makeup of the state. Pennsylvania is largely stratified between large urban centers in the east and west of the state, and more suburban and rural communities within the center of the state. According to the Center for Rural Pennsylvania, as of 2018, roughly 26% of the population of Pennsylvania lives in a rural community (Center for Rural Pennsylvania, n.d.). These communities generally have more limited access to healthcare services.

Low compliance in outpatient screenings is unfortunate, not unusual. A study by Griz et al. (2009) found that lower maternal education level, socioeconomic status, and rural living all demonstrate lower compliance with attending outpatient screening events. In 2018, the Pennsylvania Department of Health (DoH) reported that there were 66 general hospitals with 7,265 beds, (2.14 beds per 1000 residents) in rural Pennsylvania, with seven counties having no hospital at all. Additionally, these rural areas tended to demonstrate a higher poverty level (12.7%) compared to more urban areas (12.1%; Semega et al., 2019). Low compliance for outpatient screenings may also be attributed to the number of screenings and births provided by midwives throughout the state. According to Goedert et al. (2011), most midwives do not view newborn hearing screening as a responsibility and do not have the knowledge to provide information related to the NBHS program. In our study, we found that over 40% of babies screened using midwife services did not have a final NBHS result. Given our findings and previous literature, it is essential to educate midwife service providers on both the importance of the NBHS program and the role that these service providers play in conducting this vital service.

Currently in Pennsylvania, programs have been designed to increase midwife and outpatient education in NBHS. Further studies should evaluate the effectiveness of these training programs.

The mean duration from birth to diagnostic assessment result fell within the JCIH 1-3-6 guidelines. On average, infants were provided a final diagnosis approximately 75 days after birth. This is well within the guidelines suggested by the JCIH, which is that a final diagnosis occurs by three months of age. The Pennsylvania prevalence rate of 1.76 per 1,000 infants aligns with the national prevalence rate of 1.7 (Centers for Disease Control [CDC], 2017). Though these numbers appear to agree with published data, further study into the impact of loss to follow-up (LFU) on this prevalence would be beneficial. LFU is a major concern with any screening program. Presently, nearly 87% to 95% of newborns undergo a newborn hearing screening shortly after birth (Gaffney et al., 2010; Mehl & Thomson, 1998, 2002). Gaffney et al. (2010) assessed nationwide LFU on those who referred their newborn hearing screening and suggested that nearly a third of those identified with a hearing impairment at birth could go without hearing loss identification.

The false positive rate of 0.5% agrees with the hypothesized false positive rate of Clemens et al. (2000). In their study, the research team analyzed the false-positive rate of newborns during the initial screening (Stage 1) and found a false positive rate of 1.9%. The team notes that if they completed the rescreening process, which they called State 1b, the false-positive rate would be 0.5% overall. Our study confirms this estimation.

Additionally, the timeline for EI enrollment fell within the JCIH guidelines. The mean duration from birth to EI enrollment was approximately 175 days, just shy of the six-month JCIH recommendation. Adherence to the 1-3-6 guidelines is linked to increased vocabulary development in children, including better receptive and expressive language abilities, as well as a higher level of speech.
outreach programs for optimizing outcomes. Although the EI enrollment dates for Pennsylvania fall within the JCIH guidelines, there is still a lapse of time between final diagnosis and EI referral of approximately 46 days. Further research should explore reasons for this gap, though data from nation-wide studies suggest that the delay can often be attributed to agreements that states make with birthing centers related to timeliness. Sanchez-Gomez et al. (2019) note that states that require data collection within two weeks of screening have better follow-up rates than those who require collection within one month. This should be considered in evaluating the time lapse from screening to diagnosis, as well as from diagnosis to EI enrollment.

The change of policy requiring submissions to iCMS, the Pennsylvania newborn screening system, mandates all NBHS submitters (hospitals, birthing centers, or midwives) to report individual-level hearing screening results for all babies. This includes those who were unable to be screened due to parent refusal, missed screening, and transferring to hospitals outside of the state. This change came into effect in full for all babies born on January 1, 2018. Although this mandatory reporting has many benefits, communication between birthing centers and NBHS programs still faces some difficulty. One limitation is in considering that data input was completed by individual stakeholders throughout the process. Those stakeholders include nurses, audiologists, social workers, and early interventionists, as well as staff within the Pennsylvania DoH. Although it is important to have a variety of inputs for tracking and normalizing purposes, the variety in personnel inputting the data leads to the possibility of human error. For example, 212 children of the 882 children seen for a diagnostic assessment had an unknown or no-indicated hearing severity in their final report. There is no state-wide standard as to who must provide this data to the PA DoH, therefore it may be possible that this number can be attributed to human error. It may also be reflective of an aspect of the iCMS system that may need to be improved and standardized for more universal understanding among stakeholders.

The purpose of this study was to assess Pennsylvania’s compliance to the JCIH recommendations of screening by one month of age, diagnosis of hearing loss by three months of age, and early intervention enrollment by six months of age. Of interest, was the analysis of this adherence as it pertains to the policy shift of 2018, requiring all information to be stored within a centralized databank. The findings from this study suggest that Pennsylvania largely adheres to the JCIH guidelines and that use of a centralized database allows for intensive analysis into the NBHS program implementation. These findings will be used for future program improvement in Pennsylvania, specifically for outpatient screening improvement. Further research analyzing the specific outcomes related to race and region can provide deeper insight into the program’s efficacy, as well as identify outreach programs for optimizing outcomes.

References
universal newborn hearing screening and intervention program. *Pediatrics*, 126(Supplement 1), S19–S27.


**EHDInfo** [Can be downloaded here for distribution.]

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Wearing Hearing Aids improves language skills IF WORN 10 HOURS OR MORE EACH DAY.

Put hearing aids on

In the car

When awake

At play

Every day

Talk to your audiologist for support to meet the individual needs of your family.

The Role of Early Intervention in Parental Self-Efficacy for Families of Children Who Are Deaf or Hard-of-Hearing

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Kristina Bowdrie, AuD
Rachael Frush Holt, PhD

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Abstract

Purpose: To investigate the effects of age at enrollment in early intervention (EI) and dosage of EI services (frequency and intensity) on parental self-efficacy (PSE) and to determine whether parents with better PSE demonstrate more involvement in deciding Individualized Family Service Plan (IFSP) goals, services, and amount of services.

Method: Sixty-five parent-child dyads were included in this retrospective between-subjects study. PSE was measured using the Scale of Parental Involvement and Self-Efficacy (SPISE; DesJardin, 2003). Dosage of EI services and parent/professional involvement in IFSP decision-making were measured using a Child Demographic Questionnaire.

Results: Statistically significant correlations were not found between age at EI enrollment and SPISE subscales. Statistically significant correlations were not found based on frequency or intensity of EI services. Mixed results were found regarding level of parent involvement in decision-making of IFSP goals, kinds of services, and amount of services.

Conclusions: Findings demonstrate the complexities in determining the effects of age at EI enrollment, EI dosage, and central elements of the IFSP on self-efficacy in parents of children who are deaf or hard-of-hearing. Future studies are needed to validate these findings and further the knowledge base about the role of EI in supporting parents’ sense of self-efficacy in supporting their child’s development.

Keywords: parental self-efficacy, early intervention, deaf or hard-of-hearing

Acronyms: CDQ = Child Demographic Questionnaire; CI = cochlear implant; DHH = deaf or hard of hearing; EI = early intervention; FLT = facilitative language technique; HA = hearing aid; IFSP = Individualized Family Service Plan; PSE = parental self-efficacy; SPISE = Scale of Parental Involvement and Self-Efficacy

Acknowledgements: We have no known conflict of interest to disclose. This research was supported in part through a grant from the National Institutes of Health (NIDCD R01 DC014956).

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A fundamental goal of early intervention (EI) is to foster parental self-efficacy (Moeller et al., 2013). Grounded in Bandura’s social learning theory, parental self-efficacy is the belief that one is capable of positively impacting child development and confident in carrying out parenting tasks to do so (Bandura, 1989). Parental self-efficacy has been identified as a predictor of parental functioning and can mediate the effects of infant temperament and social support on postpartum depression (Coleman & Karraker, 1998). Research demonstrates the benefits of positive self-efficacy for both parents and children, including markers of healthy parent-child relationships, such as parental responsiveness (Teti et al., 1996), having home routines, and setting appropriate developmental goals (Albanese et al., 2019).

Parents who are self-efficacious have the knowledge to set appropriate goals for their child, as well as the tenacity to carry out the requisite tasks to help their child achieve those goals. Conversely, parents who doubt their ability to support their child’s development might be less likely to acquire new knowledge, or apply the knowledge they have. To feel confident and competent, parents must: (a) be knowledgeable about various childcare responses (i.e., setting appropriate limits for preschool-age child), (b) be confident in their ability to carry out such tasks; and (c) hold the belief that their child will respond contingently (Coleman & Karraker, 1998). Self-efficacy is considered a dynamic process, not a fixed trait; when new situations arise, it is possible for individuals to
acquire the knowledge to face those situations and develop the confidence to do so. In the case of parents, it is possible to gain knowledge and acquire new skills, thereby bolstering confidence in parenting. In fact, experiencing success is one of four primary sources of self-efficacy (Bandura, 1989). Conversely, experiencing failures—especially multiple failures—can result in low self-confidence. A second source of self-efficacy is social modeling. For families of children who are deaf or hard-of-hearing (DHH), interacting with and observing other parents of children who are DHH might boost parents’ sense that they can successfully raise their child. A third source is social persuasion. For example, a parent of an infant who did not pass their newborn hearing screening may feel encouraged and empowered to follow through with diagnostic audiological testing after talking with a parent who has experienced this process. And finally, emotional arousal, or feelings of stress, can be a source of self-efficacy, or inefficacy. Parents who feel especially anxious about a particular situation may experience feelings of fear and doubt, and subsequently inaction. For instance, the parent who finds early intervention sessions stressful due to worries about having a messy house may be less inclined to fully participate in those sessions.

**Parental Self-Efficacy and Children Who Are Deaf or Hard-of-Hearing**

DesJardin and colleagues have conducted several foundational studies on the role maternal self-efficacy plays in supporting their children’s language development, as well as managing use of sensory devices (e.g., DesJardin, 2005; DesJardin, 2006; DesJardin & Eisenberg, 2007). Using a measurement of parental self-efficacy developed for parents of children who are DHH, the Scale of Parental Involvement and Self-Efficacy (SPISE; DesJardin, 2003), these studies have revealed several important findings. A newly revised version—the SPISE-R—offers updated items and an expanded number of sections, including Parent Beliefs, Knowledge, Confidence, and Actions (Ambrose et al., 2020). Results from the original SPISE indicate that better self-efficacy is positively associated with maternal linguistic input, specifically use of facilitative language techniques (FLTs; DesJardin, 2006; DesJardin & Eisenberg, 2007). FLTs are markers of quality parental language input. Higher-level FLTs (e.g., parallel talk, expansion, recast, open-ended questions) promote more complex language in young children at risk for either a delay due to a disability that may interfere with typical development (Baumwell et al., 1997) or an impoverished language environment (Hart & Risley, 1999). In contrast, lower-level FLTs (e.g., labeling, imitating, linguistic mapping, close-ended questions) are less effective than higher-level FLTs at promoting spoken language skills in children who are DHH (Cruz et al., 2013). More precisely, maternal use of open-ended questions was found to be positively associated with children’s expressive language skills, and maternal recast was positively associated with children’s receptive language skills.

In addition to maternal self-efficacy and involvement being related to quality of parental input, quantity (e.g., mean length of utterances, total word-types) of parental linguistic input supports children’s spoken language development (DesJardin & Eisenberg, 2007). As Cruz et al. (2013) found, recast and open-ended questions (higher-level FLTs) were predictors of expressive language growth and associated with children’s better receptive language abilities. Moreover, longer utterances and a greater number of word types used were positively related to children’s spoken language. Considering the variability in outcomes for children who are DHH who use cochlear implants (CIs; Niparko et al., 2010), parental self-efficacy is a source of individual differences in child language development worth further investigation because it likely is malleable through early intervention.

Mothers of children who are DHH indicate that they feel more capable and comfortable in managing their child’s hearing aid (HA) and/or CI than supporting their child’s language development (DesJardin, 2005; DesJardin & Eisenberg, 2007). This may be due to the more straightforward nature of checking batteries and conducting daily listening checks compared to the unexpected task of actively supporting their child’s speech and language development. Additionally, it could be due to the importance placed on effective device use by their audiologist and early interventionists. To more fully enhance children’s language growth, parents also need to feel confident in their role as language models. This requires a shift in terms of how parents view their role in their children’s language development and, thus, the need for supporting parents early in their journey through education and coaching (DesGeorges, 2016).

Parental self-efficacy has been reported to differ between mothers of children with HAs and mothers of children with CIs (DesJardin, 2005). Specifically, relative to mothers of children with HAs, mothers of children with CIs perceived themselves as being more involved in managing their child’s device, in particular carrying out a daily listening check with their child. Mothers of children with CIs also reported more involvement in supporting their child’s spoken language development, including feeling included and comfortable participating in EI sessions, as well as engaging in language activities at home. Additionally, according to DesJardin (2005), mothers of children with HAs who entered EI earlier reported feeling more competent and confident in managing their child’s device and more involved in their child using their device compared to those who enter EI later (although earlier and later were unspecified). This suggests that early entrance into intervention might be particularly important for supporting parents’ development of self-efficacy when their child has a less severe loss and are likely receiving less-frequent intervention compared to parents of children with CIs.

Although research consistently demonstrates benefits of early enrollment in EI for children who are DHH in terms of language development (Yoshinaga-Itano, 2003; Yoshinaga-Itano et al., 1998), less attention has been paid to the effects of early enrollment on parent self-efficacy. Evidence shows that quality EI services can positively influence growth across developmental domains, particularly language. For children who are DHH, early identification and timely enrollment are related to better expressive (Pipp-Siegel et al., 2003; Yoshinaga-Itano et al., 2010) and receptive language
outcomes (Kennedy et al., 2006). Moreover, timely diagnosis and enrollment in EI are strong predictors of expressive and receptive language in children across the range in hearing levels (Holzinger et al., 2011). It is possible that an aspect of the advantage of early enrollment in EI is that it facilitates parent self-efficacy.

Another question related to EI and self-efficacy in parents of children who are DHH is *how much* EI service matters. Do more frequent visits and visits that last longer support parents’ perceptions of self-efficacy? Traditional measures of EI dosage have been in terms of duration (e.g., time spent receiving EI services from enrollment to transition), intensity (e.g., number of hours an EI provider works with a family), and comprehensiveness (e.g., number of types of services provided, such as occupational therapy or vision services; Guralnick, 1989.) The current investigation takes a slightly different approach to quantifying dosage by focusing on frequency of EI services per month and duration of sessions. Presently, there are no empirically supported recommendations for EI dosage, however, general trends in frequency of sessions fall between once a week and once a month, or based on family need. Duration of EI sessions typically fall between 30 to 90 minutes.

A further consideration regarding parent self-efficacy is the role of parent involvement in developing the driver of EI, the Individualized Family Service Plan (IFSP). Developing the IFSP is a collaborative effort between families and professionals; beginning with identifying the child’s strengths and the family’s resources, priorities, and concerns. These discussions, along with evaluation and assessment information, guide the IFSP team (e.g., parents/family members, family advocate, service coordinator, EI providers, and other professionals as needed) in determining IFSP goals. Setting goals leads to determining other key elements of the IFSP, including kinds of services (e.g., speech-language, occupational therapy, physical therapy), and intensity (e.g., frequency and length of sessions).

Furthermore, we do not know if parents with better ratings of self-efficacy are more involved in determining critical aspects of the IFSP. Therefore, the current study was motivated by the following research questions:

(1) What effect does age at enrollment in EI services have on parental self-efficacy (PSE)?

(2) What effect does dosage of EI services (frequency and intensity) have on PSE?

(3) Do parents with better self-efficacy demonstrate more involvement in deciding IFSP goals, services, and amount of services?

Materials and Method

Participants

Parents/Caregivers

A total of 65 parent-child dyads from a larger longitudinal study investigating the role of the family environment on spoken language and executive function outcomes in children who are DHH were included in this investigation. The data used in this investigation constitute those obtained from families of children who are DHH at their first of three visits who were enrolled at the time the data were analyzed. The vast majority of parents/caregivers were female (n = 61). From this point forward the term parent(s) will be used to encompass mothers, fathers, and other caregivers. Over half of the parents had earned a four-year college or graduate degree and the majority reported a household income of $50,000 or more. All of the parents were hearing and used English in the home. See Table 1 for parent demographic information.

Children

Children had prelingual bilateral sensorineural hearing loss ranging from moderate to profound with no additional neurodevelopmental disabilities directly related to deafness. All of the children used HAs (n = 29) or CIs (n = 36) in accordance with their degree of hearing loss. The average chronological age of the children was 6.25 years; and 37 were girls and 28 were boys. All children’s hearing loss was identified by 3 years of age, with the vast majority being identified through newborn hearing screening. All children received EI services by age 3 years (M = 8.44, range 1–28 months at EI enrollment) and those with cochlear implants were implanted by age 3.5 years. Most of the children were White with small numbers identifying as Black, Asian, or biracial (e.g., Black/White, Asian/White, Native American/White). Child demographics are presented in Table 2.

Table 1  
Parent/Caregiver Demographics

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>N</th>
<th>Percent</th>
<th>Frequency</th>
</tr>
</thead>
<tbody>
<tr>
<td>Highest Education Level</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>High School graduate</td>
<td>65</td>
<td>12.3</td>
<td>8</td>
</tr>
<tr>
<td>Associate’s degree</td>
<td></td>
<td>10.8</td>
<td>7</td>
</tr>
<tr>
<td>Some college</td>
<td></td>
<td>21.5</td>
<td>14</td>
</tr>
<tr>
<td>Bachelor’s degree</td>
<td></td>
<td>32.8</td>
<td>21</td>
</tr>
<tr>
<td>Master’s/PhD/Professional</td>
<td></td>
<td>23.1</td>
<td>15</td>
</tr>
<tr>
<td>Annual Household Income</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Under $5,500–$24,999</td>
<td></td>
<td>10.9</td>
<td>7</td>
</tr>
<tr>
<td>$25,000–$49,999</td>
<td></td>
<td>15.6</td>
<td>10</td>
</tr>
<tr>
<td>$50,000–$94,999</td>
<td></td>
<td>31.6</td>
<td>20</td>
</tr>
<tr>
<td>$95,000 and over</td>
<td></td>
<td>42.2</td>
<td>27</td>
</tr>
</tbody>
</table>

Measures

Child Demographic Questionnaire (CDQ)

The CDQ consists of two sections. The first section (CDQ1) collects basic demographic information about the family
and child. The second section (CDQ2) collects information pertaining to the child’s hearing loss, including age at diagnosis, age at sensory device fitting, and aided word recognition. Also included are questions about the child’s EI and education history, including frequency and length of EI sessions, as well as identifying who made decisions related to IFSP goals, kinds of services received, and frequency and duration of services. The CDQ1 was mailed to families prior to a home visit to collect further data about the family’s home environment for the larger, ongoing research study. Parents completed the CDQ2 with the clinical researcher during the home visit. Both parts of the CDQ were collected from the family at the home visit.

**Scale of Parental Involvement and Self-Efficacy (SPISE)**

The SPISE (DesJardin, 2003) is a self-report questionnaire designed to measure parents’ perception of self-efficacy and involvement related to managing their child’s amplification use and supporting their child’s speech-language development. The questionnaire consists of three sections: Demographic Information, Self-Efficacy, and Parental Involvement. In lieu of having families complete the demographic section of the SPISE, the CDQ was used to collect pertinent demographic information. The remaining two sections of the SPISE, Self-Efficacy and Parental Involvement, are each divided into two subscales: Child Amplification Use and Speech-Language Development. The Self-Efficacy section includes five questions about parents’ ability to manage and maintain their child’s sensory device and the extent to which they feel like they can affect their child’s language development. The Parental Involvement subscale consists of five questions about device maintenance and seven questions about affecting language development. All items use a 7-point Likert rating scale. Descriptive statistics were calculated for SPISE outcomes for each of the four subscales: (a) Self-Efficacy: Amplification Use; (b) Self-Efficacy: Speech-Language Development; (c) Parental Involvement: Amplification Use; and (d) Parental Involvement: Speech-Language Development.

**Procedures**

Families were recruited from two universities and their respective partner children’s hospitals, as well as through community groups and word of mouth. Two clinical researchers with extensive experience working with children who are deaf and their families visited families’ homes to carry out behavioral testing. One researcher worked with the child and one with the parent. Visits lasted up to 2.5 hours (these data constitute a subset of what was collected at the visit). In addition, primary caregivers were mailed a packet of questionnaires, including the CDQ1 and the SPISE, to complete prior to the home visit. All research was approved by the local IRB.

**Data Preparation and Analyses**

Due to lack of normal distribution, frequency and length of EI sessions were divided into two categories. Frequency of EI sessions per month were categorized as 1–2 visits or >3 visits. Visit length was categorized as 30–45 minutes and >45 minutes. IFSP/service plan variables (who decided goals, kinds of services, and amount of services) also were divided into two categories: my family/my family and a professional made these decisions, or the professional made these decisions.

Of note, three parents reported that their children began EI services much later than the rest of the children in the current sample. These participants were removed from the sample due to their age at enrollment falling more than three standard deviations above the mean. One child entered EI at 28 months, which is less than a year from exiting EI services at the standard 3 years of age. Two children actually entered EI after the standard EI timeframe, birth to 3 years. Age at enrollment among these three participants stand in contrast to a mean age of 8.77 months ($SD = 7.4$) for the remaining participants.

**Results**

**Descriptive Statistics**

Table 3 displays descriptive statistics for each of the SPISE subscales. Average scores on the Self-Efficacy of Amplification Use and Speech-Language Development subscales fell on the high end of the 7-point Likert scale. Average scores on the Parental Involvement in Speech-Language Development subscales were somewhat lower and were quite a bit lower for Parental Involvement in Child’s Amplification Use.

No statistically significant differences were found between parents of children with HAs and parents of children with CIs on three of the SPISE subscales, including Self-Efficacy.
of Speech-Language Development (mean HA = 5.9; mean CI = 6.2), Parental Involvement in Child’s Amplification Use (mean HA = 3.8; mean CI = 3.6), and Parent Involvement in Speech-Language Development (mean HA = 5.0; mean CI = 5.1). Parents of children with HAs ($M = 5.1$, $SD = .61$) had significantly lower scores than those of children with CIs ($M = 6.34$, $SD = 1.03$) on Self-Efficacy of Amplification Use, $t(58) = 6.04$, $p < .001$. Age at enrollment in EI also was not significantly different ($p = .655$) between children with HAs (9.0) and those with CIs (8.1).

Descriptive statistics were also calculated for frequency and length of EI sessions (see Table 4). Note that a small number of parents did not complete the questions about frequency and length of EI session. Just over half of families reported that they received EI services 3 or more times per month; the remaining families received EI services 1 to 2 times per month or did not respond. The majority of families reported that EI sessions were longer than 45 minutes, with a small percentage reporting participating in EI sessions that lasted 30–45 minutes. No statistically significant differences were found between parents of children with HAs versus CIs on frequency ($p = .203$) or length of EI sessions ($p = .736$).

Table 5 displays the descriptive statistics for who made decisions regarding IFSP goals, kinds of services, and amount of services. Of the 65 responses, the majority of parents reported that either their family or their family in collaboration with professionals determined IFSP goals. The responses to who decided the kinds of EI services and amount of services was split almost evenly between (a) families who reported that their family or their family with a professional made these decisions, and (b) those reporting that the professional alone made the decision.

An independent samples $t$-test was conducted to compare each of the three aspects of decision-making based on type of device (e.g., HA or CI). No statistically significant differences were found between parents of children with HAs versus CIs regarding who decided goals ($p = .780$) or kinds of services ($p = .778$). A statistically significant difference was found between parents of children with HAs and parents of children with CIs regarding deciding the amount of services, $t(63) = 2.43$, $p = .018$. Compared to parents of children with CIs (36%), more parents of children with HAs (66%) reported that the professionals determined the amount of services.

Correlation Analyses: Age at Enrollment in EI Services and Parental Self-Efficacy

There were no statistically significant correlations between age at enrollment and any subscale of the SPISE: Self-Efficacy of Device Use ($p = .987$), Self-Efficacy of Speech-Language Development ($p = .672$), Parental Involvement in Device Use ($p = .756$), and Parental Involvement in Speech-Language Development ($p = .831$). See above for values of each $p$.

Table 3
Descriptive Data for the Scale of Parental Involvement and Self-Efficacy

<table>
<thead>
<tr>
<th>Subscales</th>
<th>N</th>
<th>Mean</th>
<th>SD</th>
</tr>
</thead>
<tbody>
<tr>
<td>Self-efficacy</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sensory aid use</td>
<td>65</td>
<td>5.8</td>
<td>1.0</td>
</tr>
<tr>
<td>Speech-language development</td>
<td>65</td>
<td>6.1</td>
<td>0.8</td>
</tr>
<tr>
<td>Parental involvement</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sensory aid use</td>
<td>65</td>
<td>3.7</td>
<td>0.6</td>
</tr>
<tr>
<td>Speech-language development</td>
<td>65</td>
<td>5.1</td>
<td>0.9</td>
</tr>
</tbody>
</table>

Table 4
Descriptive Data for Early Intervention (EI) Dosage

<table>
<thead>
<tr>
<th>Variable</th>
<th>N</th>
<th>Percent</th>
<th>Frequency</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of EI visits per month</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1–2 visits/month</td>
<td>40</td>
<td>24</td>
<td></td>
</tr>
<tr>
<td>3+ visits/month</td>
<td>60</td>
<td>36</td>
<td></td>
</tr>
<tr>
<td>Average length of EI sessions</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>30–45 minutes</td>
<td>16.4</td>
<td>10</td>
<td></td>
</tr>
<tr>
<td>More than 45 minutes</td>
<td>83.6</td>
<td>51</td>
<td></td>
</tr>
</tbody>
</table>

Table 5
Descriptive Data for Individualized Family Service Plan (IFSP) Decision-Making

<table>
<thead>
<tr>
<th>Variable</th>
<th>N</th>
<th>Percent</th>
<th>Frequency</th>
</tr>
</thead>
<tbody>
<tr>
<td>Who decided the goals or outcomes for your child on their IFSP or Service Plan?</td>
<td>65</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mostly my family/our family and professionals together</td>
<td>83.1</td>
<td>54</td>
<td></td>
</tr>
<tr>
<td>Mostly the professionals</td>
<td>16.9</td>
<td>11</td>
<td></td>
</tr>
<tr>
<td>Who decided the kinds of services for your child on their IFSP or Service Plan?</td>
<td>65</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mostly my family/our family and professionals together</td>
<td>53.8</td>
<td>35</td>
<td></td>
</tr>
<tr>
<td>Mostly the professionals</td>
<td>47.7</td>
<td>31</td>
<td></td>
</tr>
<tr>
<td>Who decided on the amount of services for your child on their IFSP or Service Plan?</td>
<td>65</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mostly my family/our family and professionals together</td>
<td>50.8</td>
<td>33</td>
<td></td>
</tr>
<tr>
<td>Mostly the professionals</td>
<td>49.2</td>
<td>32</td>
<td></td>
</tr>
</tbody>
</table>
Relation Between Dosage of EI Services and Parental Self-Efficacy

Independent samples t-tests were conducted to compare group means on each of the SPISE subscales between families who received on average 1 to 2 EI sessions per month and those who received 3 or more visits per month. No statistically significant differences were found between the two groups on any of the SPISE subscales: Self-Efficacy of Device Use for families receiving 1 to 2 EI sessions (M = 5.68, SD = .99) and 3 or more EI sessions (M = 6.04, SD = .92) per month, t(58) = -1.422, p = .160; Self-Efficacy of Speech-Language Development for families receiving 1 to 2 EI sessions (M = 6.06, SD = .93) and 3 or more EI sessions (M = 6.10, SD = .73) per month, t(58) = -.182, p = .856; Parental Involvement in Sensory Device Use for families receiving 1 to 2 sessions (M = 3.80, SD = .68) and 3 or more sessions (M = 3.65, SD = .57) per month, t(58) = .957, p = .342; and Parental Involvement in Speech-Language Development for families receiving 1 to 2 sessions (M = 5.14, SD = 1.03) and 3 or more sessions (M = 4.98, SD = .89) per month, t(58) = .619, p = .538.

Independent samples t-tests also were conducted to compare means on each of the SPISE subscales between families whose EI sessions ranged from 30 to 45 minutes and those who received visits that lasted more than 45 minutes. As with frequency of EI services, there were no significant differences on SPISE subscales between these two groups: Self-Efficacy of Sensory Device Use for families receiving 30–45 minute EI sessions (M = 5.62, SD = 1.13) and those receiving EI sessions lasting more than 45 minutes (M = 5.88, SD = 1.03), t(59) = - .742, p = .461; Self-Efficacy of Speech-Language Development for families receiving 30–45 minute EI sessions (M = 5.74, SD = .98) and those receiving EI sessions lasting more than 45 minutes (M = 6.09, SD = .82); t(59) = -1.222, p = .226; Parental Involvement in Sensory Device Use for families receiving 30–45 minute EI sessions (M = 3.66, SD = .61) and those receiving EI sessions lasting more than 45 minutes (M = 3.74, SD = .62); t(59) = -.404, p = .688, and Parental Involvement in Speech-Language Development for families receiving 30–45 minute EI sessions (M = 4.73, SD = .88) and those receiving EI sessions lasting more than 45 minutes (M = 5.1, SD = .98), t(59) = -1.142, p = .258.

Parental Self-Efficacy and Involvement in IFSP Decision-Making

Independent samples t-tests were conducted to compare group means on each of the SPISE subscales for two groups of families: those who reported that their family or their family with a professional decided IFSP goals, services, and amount of services; and families who reported that the professionals decided on these aspects of the IFSP. Independent samples t-tests were also conducted to compare group means of parents of children with HA’s and parents of children with CI’s on each of the aforementioned variables.

Who Decided: IFSP/Service Plan Goals

There was not a statistically significant difference in Self-Efficacy for Amplification Use or Speech-Language Development (p ≥ .454) between families reporting that they alone or with the help of professionals decided IFSP goals and families reporting that professionals decided goals. In contrast, there was a statistically significant difference in Parent Involvement in Amplification Use, t(63) = -2.41, p = .02, with families who reported that professionals decided goals (M = 4.16, SD = .66) having higher levels of involvement in their child’s sensory aid than families reporting that they alone or they with professionals decided IFSP goals (M = 3.63, SD = .57). There also was a significant difference in Parent Involvement in Speech-Language Development, t(63) = 2.93, p = .005, with families reporting that they alone or they with professionals decided IFSP goals (M = 5.2, SD = .92) having higher levels of parent involvement in speech-language than those reporting that professionals decided goals (M = 4.18, SD = .86).

Who Decided: Kinds of Services

Self-Efficacy for Amplification Use and Speech-Language Development approached significance based on who decided the kinds of EI services (p = .57). In contrast, there was a statistically significant difference in Parent Involvement in Amplification Use, t(64) = -2.13, p = .04, with families who reported that professionals decided kinds of services (M = 3.97, SD = .61) having higher levels of involvement in device use than families reporting that they alone or with professionals decided the kinds of services (M = 3.61, SD = .58). However, there was not a significant difference in Parent Involvement regarding Speech-Language Development between the two groups (p = .32).

Who Decided: Amount of Services

Families who reported that they alone or with the help of professionals decided the amount of services (M = 6.07, SD = .82) had statistically higher levels of Self-Efficacy for Amplification Use than families reporting that professionals decided the amount of services (M = 5.49, SD = 1.14), t(63) = 2.17, p = .023. Self-Efficacy for Speech-Language Development approached significance based on who decides the amount of services (p = .07) with families who reported that they alone or with professionals decided the amount of services having more involvement (M = 6.25, SD = .69) compared to families who reported that the professional decided amount of services (M = 5.88, SD = .95).

Concerning parent involvement, families reporting that professionals decided the amount of services (M = 4.0, SD = .61) had significantly higher levels of involvement regarding Amplification Use than families reporting that they alone or they with professionals decided amount of services (M = 3.45, SD = .46), t(63) = -4.10, p = .001. In terms of Parent Involvement in Speech-Language Development, families reporting that they alone or with professionals decided the amount of services (M = 5.32, SD = .89) had significantly higher levels of involvement compared to families reporting that professionals decided amount of services (M = 4.8, SD = .96), t(63) = 2.234, p = .029.

Discussion

The purpose of this study was to investigate parental self-efficacy relative to age at entry into EI, EI dosage (frequency
and length of EI sessions), and level of parental involvement in IFSP decision-making. Our findings indicate no statistically significant correlation between parental self-efficacy and children’s age at enrollment in EI. Moreover, parental self-efficacy did not differ based on frequency and length of EI sessions. Finally, mixed results were found regarding whether parents with better self-efficacy demonstrate more involvement in deciding IFSP goals, services, and amount of services.

Overall, SPISE outcomes for the current sample of parents are comparable to outcomes from previous studies on self-efficacy in parents of children who are DHH (e.g., DesJardin, 2005; DesJardin & Eisenberg, 2007). Previous studies found that parents report better self-efficacy in managing their child’s device rather than supporting their language development. In the current study, parents also reported higher levels of self-efficacy regarding device use than supporting their child’s language development.

**Age at EI Enrollment and Parental Self-Efficacy**

DesJardin (2005) found that for mothers of children with HAs, but not for those of children with CIs, early enrollment in EI correlated with better perceptions of self-efficacy and involvement in supporting their child’s language development and device management. By comparison, the current study found a correlation between age at EI enrollment and self-efficacy for sensory device use, but not speech-language development, for parents of children with HAs. Similar to DesJardin (2005), we did not find a correlation between age at enrollment into EI and SPISE outcomes for parents of children with CIs.

Comparing SPISE subscale mean scores collapsed across device group in the current study with mean scores from DesJardin (2005) indicates slight differences in three of the subscale scores, and a larger difference for one of the subscales. Parents in the current study reported slightly lower self-efficacy regarding device use (difference of .47 points) and slightly higher self-efficacy regarding speech-language development (difference of .64 points) than those in the Desjardin (2005) study. Also, parents in the current study reported lower parent involvement regarding device use compared to those in the 2005 study, with a difference of 2.63 points, and very similar scores (a difference of .07 points) on the parent involvement regarding speech-language development subscale. Of note, average scores for both groups on each subscale were rather high, ranging between 6 and 7.

One potential explanation for the relatively high subscale scores in the DesJardin (2005) study and the current study is related to the psychometrics of the tool. In the current study, the level of sensitivity in the version of SPISE administered may have been insufficient to capture the degree of parents’ sense of self-efficacy. In fact, Coleman and Karraker (1998) identified several factors that have inhibited investigations of parental self-efficacy, one of which is the lack of psychometrically sound measures of the construct. Although there is a relatively long history of acknowledging parental self-efficacy in the literature as an important variable in effectively and successfully parenting children with disabilities, there are some criticisms of the measurement tools that have been used. Most measures of parental self-efficacy, including the SPISE, are minimally validated and include rather vague descriptions of certain concepts related to self-efficacy (Coleman & Karraker, 1998).

Furthermore, translating a complex human construct like self-efficacy into a quantifiable unit is an enduring challenge (Cook & Bechman, 2006). Likert scales are a frequently-used method of capturing strength of human attributes, such as attitudes, opinions, and perceptions, but as with all forms of measurement, they come with advantages and disadvantages (Joshi et al., 2015). A major advantage of Likert scales is the ubiquity with which they are used—most people are familiar with completing them. A major disadvantage is that they are an indirect measure of multidimensional constructs (Hasson & Arnetz, 2005). Perhaps a slightly wider range in scale would provide a clearer understanding of the relation between age at EI enrollment and parent involvement and self-efficacy. Future studies might investigate parental self-efficacy longitudinally. Perhaps parents demonstrate greater self-efficacy in relation to age at enrollment further into their parenting journey beyond the birth to three years. The absence of a relationship between age at EI and parental self-efficacy may be explained by the limited variability in age at enrollment. Greater variability in age at EI, (i.e., 2 months to 3 years), might yield a different outcome.

**Effect of Dosage of EI Services**

The current study is the first to investigate the effects of EI dosage on parental self-efficacy among parents of children who are DHH. No significant differences in self-efficacy were found between parents who participated in EI sessions more or less frequently (e.g., 1–2 per month or ≥ 3 per month), or for shorter or longer sessions (e.g., 30–45 minutes or > 45 minutes). There are a couple considerations that should be made in explaining this null finding. The first consideration is the demographics of the current sample, which included children who are DHH without additional diagnoses. Most children were from relatively resource-rich households with college-educated parents. Hallam and colleagues (2009) indicated that Medicaid status, access to third party insurance, and children’s developmental abilities influence level and intensity of EI services. If the current sample were more diverse demographically, perhaps a different outcome would have emerged. Future studies might implement research-supported strategies for recruiting and retaining underrepresented populations, including collaboration with community partners (Brannon et al., 2013; Wallerstein & Duran, 2010) and employing a dedicated staff member to walk families through the study consent process, assist in completing paperwork, and mentorship (Brannon et al., 2013; Flores et al., 2017).

The second consideration is how dosage is quantified in the current study compared to methods of quantification in previous studies (e.g., Hallam et al., 2009). Hallam and colleagues measured dosage by the number of events (e.g., visits), units (e.g., total hours within a 6-month period), and services (i.e., speech-language services, physical therapy,
Parental Self-Efficacy and Involvement in IFSP Decision-Making

Analyses of parental self-efficacy and involvement in IFSP decision-making produced mixed results, some of which are seemingly counterintuitive. There was not a statistically significant difference in either type of self-efficacy between parents who were involved in determining IFSP goals or kinds of EI services and parents who were not involved. Parents perceived themselves as competent and confident in these two areas, regardless of involvement in establishing goals and determining the appropriate services.

However, differences were found between the two groups in terms of self-efficacy in determining amount of services. Goals and kinds of services may be more salient to parents compared to amount of services. Parents may feel more capable of identifying what they want for their child considering language, sensory technology, and social-emotional health, but feel less knowledgeable about how much will be required to achieve their goals. In partnership with EI professionals, particularly professionals with expertise in deafness, parents of children who are DHH may also have an easier time identifying the kinds of services needed. This may be due to severity of deafness or presence of a condition that makes the case for kinds of services more obvious. It may be the case that this element of services is less salient to parents, thus requiring more input from professionals.

Regarding parent involvement in device use and speech-language development, families reporting that professionals decided EI goals had higher levels of involvement in device use compared to families reporting that they decided IFSP goals or they worked with professionals to decide on goals. This finding indicates a relationship between level of parent involvement in determining IFSP goals and involvement in their child’s device use, but runs counter to the expectation that the more parents are involved in developing their child’s EI services, the more they would be involved in their child’s use of a device. Perhaps parents, at this early point in their journey, rely on professionals to guide them in their process of setting IFSP goals and that support results in them feeling involved in managing their child’s HAs or CIs. Or, perhaps the explanation lies in the training and experience of the EI providers: It is possible that highly qualified EI providers are skilled at guiding the development of IFSP goals while at the same time actively engaging parents in managing their child’s devices.

There was a statistically significant difference based on level of parent involvement regarding device use (e.g., daily listening checks with the device, putting on the device, and attending scheduled audiology and speech-language appointments) in relation to determining kinds of EI services. Families reporting that professionals decided kinds of services had higher levels of involvement in their child’s device use than families reporting that they alone or they with professionals decided kinds of services. This finding is somewhat counterintuitive, warranting further examination. It would be expected that the more involved parents are in the development of their child’s IFSP, the more involved they would be in their child’s use of a sensory device, or vice versa. There was a significant difference in parent involvement regarding speech-language development between the two groups in determining the amount of EI services. Families reporting that they were involved in the decision about the amount of services had higher levels of involvement regarding device use than families reporting that the professionals decided amount of services.

To better understand the mixed results between parent involvement in IFSP development and parental self-efficacy, the authors offer two areas for consideration. First, parental temperament or personality might be contributing to the relationship between self-efficacy and IFSP decision-making. It is possible that, depending on temperament, some parents feel quite comfortable deferring IFSP decision-making to professionals yet view themselves as engaged in the process. Some parents may feel more involved in their child’s device use and more apt to follow the professional’s instructions on managing their child’s device use when professionals take the lead in these matters.

A second consideration is parents’ views on the relationship between sensory devices and spoken language development. Parents may view setting IFSP goals as more closely related to speech-language development than to managing technology. Parents who report less involvement in EI may have greater sense of reliance on the device to help their child acquire spoken language. Parents who are more involved in EI may view themselves, as parents, as having a larger impact on their child’s spoken language development. Both considerations should drive future directions of research on the relationship between parental self-efficacy and involvement in EI.

Conclusions

Previous studies on self-efficacy among mothers of children who are DHH demonstrate the importance of building families’ sense of competence and confidence in the early years so that they can better support language development and manage sensory device use (DesJardin, 2005, 2006; DesJardin & Eisenberg, 2007). Research also demonstrates the benefits of early enrollment in EI for this population of children (e.g. Moeller, 2000; Yoshinaga-Itano, 2003). Furthermore, EI best practices call for providers to directly involve parents in developing the IFSP and developing a partnership to achieve IFSP goals. However, findings from the current study demonstrate the complexities in determining the relation between these variables. In light of these findings, EI
providers and clinicians should continue to coach parents and caregivers on implementing facilitative language techniques and emphasize the important role they play in between EI sessions. Future studies are needed to validate these findings and further the knowledge base about the role of EI in supporting parents' sense of self-efficacy in supporting their child's development.

References


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Evaluation of a Group Music Intervention to Support School-Readiness Skills in Preschool Children with Hearing Loss

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Abstract

Although children with hearing loss are now often integrated into mainstream classrooms, many do not begin school with age-appropriate school-readiness skills. Traditional therapies in early listening and spoken language programs may not focus on developing the social skills, executive functions, and motor abilities needed for the typical classroom environment of friends, academics, and play. This study was developed to better understand how to incorporate group activities into traditional therapies to build skills in these areas, and whether or not the use of music and its social aspects could support this. A quasi-randomized, group, facilitated, music intervention was conducted to help support school readiness skill development in preschool-aged children with hearing loss. Standardized testing was used to measure outcomes, and although improvement in skills was observed during the intervention, all test results were nonsignificant. Families reported overall improvement in skills and enjoyment of the intervention. Questions arise regarding the limits of standardized measures and the possibility of adding observational assessments for studies measuring function in social settings to better capture change.

Keywords: school readiness, hearing loss, formalized assessment, music and movement, integration

Acronyms: ASI = auditory skills intervention; AVT = Auditory-Verbal Therapy; CB = craft-based; EF = executive functions; HL = hearing loss; IHP = Infant Hearing Program; M&M = music and movement

Correspondence concerning this article should be addressed to: Glynnis E. DuBois, MHSc, Department of Speech Language Pathology 10th floor, University of Toronto, 500 University Ave., Toronto, ON, Canada M5G 1V7 2. Email: glynnis.dubois@mail.utoronto.ca.
Although auditory-oral therapies have had good success with language outcomes (Fairgay et al., 2010; Fulcher et al., 2015), other studies report continued delays (Meinzen-Derr et al., 2018) with approximately 50% of children having language levels below those of their typically hearing peers at school entry (Geers et al., 2015; Niparko et al., 2010; Wei, 2010). Other developmental areas related to literacy, social, and executive functions may not typically be included in auditory oral therapy and may also be impacted. These all depend, at least in some part, upon age-appropriate language including vocabulary for their continued development. Also, due to the nature of hearing loss and its association with the vestibular system, balance is a challenge for many children with HL (Cushing et al., 2008; Livingstone & McPhillips, 2011) and can be an impediment to social games and play.

For all children, the cascading influence of various areas of development on overall success is important to understand and is a good starting point to address some of the challenges children with a hearing loss face. A lag in any area of skill may influence development in others (Hoffman et al., 2014).

### Areas of Challenge for Children with Hearing Loss

#### Language

Language outcomes of children with HL continue to be a challenge as the population is varied and consistent access to speech and language is a key factor. The reason for and degree of hearing loss, presence of residual hearing (Niparko et al., 2010), age at diagnosis, technology support (Stika et al., 2015), type of therapy (Dettman et al., 2013), and other diagnoses all contribute to the overall outcomes of children with hearing loss. Combined, this diversity greatly impacts outcomes, and reporting on children with hearing loss as a group may not accurately reflect all areas needing support.

Listening and spoken language therapies focus on language development using a one-on-one, structured hierarchy of strategies and parental coaching to enable parents to use these strategies during all daily activities (A.G. Bell, 2011). Therapists model and coach as the child, the parent, and the therapist interact through listening and language-based activities. Although reports cite positive outcomes for listening and spoken language therapies, children can continue to have language delays by school entry (Wei, 2010). Data from some studies predict that these children may not catch up to their peers until 8 years of age or later (Leigh et al., 2013). As language proficiency impacts other areas of development (Rinaldi et al., 2013), it is imperative that these gaps are closed as quickly as possible.

#### Literacy

The ability to decode written language plays a large part in the school curriculum. From early on, children are expected to be able to move through the steps needed to attain this milestone. Mastering literacy skill is paramount to ultimate success in school as all subsequent learning depends on the ability to read and understand written material.

Preliteracy skills including phonological awareness impact the development of skills needed for reading (von Muenster & Baker, 2014). These involve the ability to rhyme, segment sentences and words into syllables, and later, delete and blend sounds. Delays in this area for children with hearing loss are related to ongoing challenges with speech perception and language skills (Ching et al., 2014). Children with hearing loss often do not perform at the same level as their peers with typical hearing in pre-literacy skills and there can be a significant lag in their development (Goldberg & Lederberg, 2015; Harris et al., 2017; Nittrouer et al., 2012; Webb & Lederberg, 2014). Test scores of children with HL continue to be one standard deviation below their peers who have typical hearing (Ambrose et al., 2012; Ching et al., 2014; Goldberg & Lederberg, 2015) and these scores correlate with receptive and expressive language as well as speech perception scores (Ambrose et al., 2012).

### Social Skills

Skills related to social interactions with both peers and others is another very important aspect of development. Social skills incorporate all abilities to communicate, negotiate, and participate successfully in the activities of the day. Consequently, language also plays a large part in the development of social skills. Although children with HL initiate interactions as often as children with typical hearing, they may not be as readily accepted into the play group (DeLuzio & Girolametto, 2011). This may be due to challenges with language; either issues with intelligibility or lack of age-appropriate vocabulary, a possible result of the inability to overhear peer interactions (DeLuzio & Girolametto, 2011). Related challenges have also been seen in the delayed development of pragmatics (Rinaldi et al., 2013), emotional perception and production in speech (Chatterjee et al., 2019; Van De Velde et al., 2019), and overall emotional understanding (Wiefferink et al., 2013). Some have emphasized that a focus on language development along with social skills should be stressed when developing strategies for supporting children with hearing loss (Hoffman et al., 2014; Wong et al., 2017), along with the suggestion of developing a truly inclusive environment in the classroom where children are part of the classroom community and not just present in the class (Xie et al., 2014).

Children with hearing loss are also at a greater risk of having mental health issues related to loneliness (Most et al., 2011), and depression (Brown & Cornes, 2015; Idstad et al., 2019; Jiang et al., 2020; Theunissen et al., 2014). Interviews and surveys have concluded that issues around making friends and challenges understanding nuanced communication add to the hurdles faced by children with HL (Punch & Hyde, 2011). These all illustrate the importance of early supports for social skill development in children with HL in order to have continued success and happiness.
Executive Functions

Another aspect of development influenced by language is executive functions (EF). These play an important role in behaviors such as inhibition, flexibility, problem solving, planning, focus, and working memory. As a whole, EF may be influenced or their development interrupted by challenges such as a language delay (Beer et al., 2014; Kaushanskaya et al., 2017). Some question whether it is the executive functions that contribute to the language delay or the language delay that impedes the development of executive functions (Beer et al., 2014). Children with hearing loss tend to score significantly lower on EF skills related to inhibition, concentration, and working memory (Beer et al., 2014; Kronenberger et al., 2013). Children with lower language abilities tend to also have more EF difficulties (Hintermair, 2013). Some posit that in order to best support development in the area of executive functions, one must take a holistic view of the child and activities should include aspects of social, emotional, and physical development (Diamond & Lee, 2011).

Balance

The ability to interact and play with peers in a competent, confident manner is paramount to success, both in the classroom and on the playground. For children with hearing loss this is a two-fold challenge as both language delays and balance play a role. Due to the anatomy of the inner ear, the cochlea has two related but separate functional areas, the auditory and the vestibular systems. Hearing loss can have a great impact on the vestibular system due to its close proximity and often overlapping structural or functional issues (Cushing, Chia, et al., 2008; Cushing, Papsin, et al., 2008; Livingstone & McPhillips, 2011).

Twenty to seventy percent of children with hearing loss have vestibular deficits (Cushing, Chia, et al., 2008) that can impact other multisensory processing systems (i.e., tactile and motor function also involved in play; Bharadwaj et al., 2012; Fellinger et al., 2015) further affecting engagement with peers. Children with HL would also benefit by making motor skills an aspect of habilitation.

The Role of Music

Several areas in development are dependent on the ability to perceive sounds in the environment accurately and in a timely fashion to maintain context and synchrony with others. Many have reviewed the literature and commented on the use of music to assist in the development of processing, audition, and language (Brandt et al., 2012; Francois et al., 2015; Shahin, 2011). Evidence has supported the use of musical experience to scaffold development in these areas in children with typical hearing. The rhythmical quality of both music and language, demonstrated in children’s nursery rhymes, engages children in a number of ways: emotionally through the enjoyment of the sounds, neurologically through entrainment to the beat, and socially through aspects of language use and sharing of the activity. Preliteracy skills may be built on the ability to entrain (or engage both the auditory and motor neural pathways) to a rhythm as this allows for the development of segmentation of both sentences and words, tasks necessary ultimately for reading (Degé & Schwarzer, 2011). Music experience can support social skills as it is often enjoyed in a group setting. Children’s music groups from early on have demonstrated the ability to support positive social engagement behaviors described as prosocial (Cirelli et al., 2014; Gerry et al., 2012). During these social interactions, other aspects of development can also be supported and practiced.

Music and children with HL

The use of music and movement for children with hearing loss comes from a logic based on evidence that increasing the complexity of listening exercises can build auditory skills. This then may influence all other skills dependent on the ability to access and process auditory input accurately and finely. The ability of music and movement to scaffold these skills has been demonstrated in numerous outcomes related to speech perception, language, social skills, and executive functions (Gfeller, 2016). Although the limitations of hearing technology are well known regarding certain aspects of music (Hsiao & Gfeller, 2012; See et al., 2013), the question arises as to whether or not early training and experience may be able to fine-tune the auditory pathways and support skill development. Understanding the areas of strength both in the technology and neural pathways, makes the use of music and movement in the early years a possible strategy for skill development in preschool children with hearing loss.

Research Questions

This study used a twelve-week, group music intervention to investigate two questions.

1. Will the outcomes in areas of school-readiness skills (language, literacy, social competence, executive functions, and balance) be significantly improved in the intervention group compared to the control group?

2. Will the outcomes between the music and movement and craft-based groups be significantly different?

Method

A quasi-randomized music intervention was conducted with 12 weekly, facilitated, group sessions. Each child had one parent participate with them during the intervention.

Participants and Recruitment

Children with bilateral, permanent, sensorineural hearing loss, using hearing technology consistently, and in an English listening and spoken language program, were recruited for this study. School boards, listening and spoken language practitioners, and community support groups were all approached to identify potential participants. All children were between the ages of 3 and 5 years and were screened using the Nipissing District Developmental Screen (NDDS, 2011) to exclude any...
children who might have developmental conditions that would preclude their participation in the intervention programs, including those with auditory neuropathy spectrum disorder.

If the child met the inclusion criteria, parents signed a participation consent form and completed a demographic questionnaire containing information regarding general developmental milestone attainment, hearing tests and technology, and any previous involvement in music lessons. Families were subsequently put into one of three groups: music and movement (M&M), craft-based (CB), or control. The control group was offered a series of twelve 45-minute music and movement sessions after their post testing with the understanding that they would act as late entry participants and would be tested a third time. The children were quasi-randomized for age and sex only with each group having both sexes and different ages represented whenever possible. Each child participated with a parent/caregiver in twelve 45-minute, weekly sessions. Two sites for the intervention were selected to support attendance of all interested families. A total of 15 children were recruited for the interventions: eight for M&M (two late-entries) and seven for CB (two late-entries).

**Intervention**

Each intervention curriculum was developed based on activities to support school-readiness skills including language, listening, phonological awareness, social skills, executive functions, and balance. Using aspects of entrainment theory and a focus on school-readiness skills, the goal was to support development in these important areas and better prepare children with hearing loss for an integrated classroom setting. Twelve sessions were organized with a weekly theme (e.g., transportation, under the sea, superheroes), a book, and activities to reinforce the theme (see Tables 1 and 2). Groups consisted of between two and five children with one accompanying parent/caregiver who also participated in the activities. All intervention groups were facilitated by a speech-language pathologist specializing in HL who had had no previous interactions with the participating families. The two groups were chosen to attempt to distinguish between group effect and music effect as both could contribute to overall outcomes.

All music used for the sessions was made available to the families for use at home during practice time through a link to a YouTube channel that was sent to each family after the first two classes. The same pieces of music were used in both intervention groups and consisted of a selection of both classical and children’s music. None of the music used had lyrics. The M&M sessions had activities facilitating movement to the music whereas the CB sessions had the music playing in the background while crafts were being completed.

Attendance was taken each week and a portable sound field amplification system was used by the facilitator at each session to ensure optimal auditory access for all participants (a sound field amplification system is made up of a microphone worn by the facilitator, an amplifier, and a built-in speaker which makes the facilitator’s voice more intense than the ambient noise in the room).

**Table 1**

**Sample Curriculum for Music and Movement**

<table>
<thead>
<tr>
<th>Activity</th>
<th>Goals</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Warm up: Done in a circle and will include various stretches of the legs, arms, and torso. Each stretch will have a set song/rhyme to accompany it.</td>
<td>self-regulation, listening, vocabulary, rhyming, active use of language, cooperation, singing, memory</td>
</tr>
<tr>
<td>2. Follow the leader: Children form a line, remain in that line for the completion of the song and move to the beat of the music in one of three ways (march, gallop, or tip toe). The music will be chosen based on its rhythm and tempo.</td>
<td>self-regulation, cooperation, listening and moving to the beat, motor coordination and balance</td>
</tr>
<tr>
<td>3. Sleeping game: Children sleep while they listen to the rhyme that tells them what they will be when they wake up. Various props are utilized in this activity (e.g., scarves, bean bags, bells).</td>
<td>pretend play and imagination, self-regulation, vocabulary, rhyming, language use, negotiation</td>
</tr>
<tr>
<td>4. Story time: A different nursery rhyme is read each session and the children are encouraged to act out the story with scaffolding by the instructor.</td>
<td>self-regulation, cooperation, imagination and pretend play, vocabulary, language use, rhyming</td>
</tr>
<tr>
<td>5. Stop and go: Various types of music will be played with differing aspects such as rhythm (gallop, march, skip, bounce, skate/slide), high/low, fast/slow, quiet/loud, happy/sad. Children will interpret the music freely but will need to listen for when it starts and stops to regulate their own dancing. Reminders will be given before the activity starts regarding when to stop and when to go.</td>
<td>listening, self-regulation, cooperation, focus</td>
</tr>
<tr>
<td>6. Bird on a wire: This activity requires the children to form a line side by side to watch a demonstration of steps as well as say thank you and curtsey/bow. It is begun with a request for bird on a wire and a countdown is done from 3 to 1.</td>
<td>listening, self-regulation, cooperation, focus, memory</td>
</tr>
</tbody>
</table>
Table 2
Sample Curriculum for Craft-Based Group

<table>
<thead>
<tr>
<th>Activity</th>
<th>Goals</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Warm up: Introduce the theme of the class by reading a story and discussing content and vocabulary.</td>
<td>self-regulation, listening, vocabulary, active use of language, cooperation, memory</td>
</tr>
<tr>
<td>2. Follow the directions of the facilitator in making the craft by using various fine motor skills related to cutting, pasting, crayons, stickers, and lacing.</td>
<td>self-regulation, cooperation, listening, motor skill</td>
</tr>
<tr>
<td>3. The children will interact using their completed craft in show and tell and nursery rhyme activities.</td>
<td>pretend play and imagination, self-regulation, vocabulary, language use</td>
</tr>
<tr>
<td>4. Clean-up Routine: Craft area will be tidied and cleaned up as a group.</td>
<td>listening, self-regulation, cooperation, focus</td>
</tr>
</tbody>
</table>

coloring or cutting and pasting. Parents were also asked to keep track of any other behaviors from the sessions that were initiated by their child. Again, this might be songs/movements from class, rereading the book, or doing a craft. These sheets were collected each week.

Data Collection

All assessments used in this study were selected as they each reported both reliability and validity. Each test provided either a Standard Score or a T score and had been normed on a population of typically developing children. All pre-testing was done within one month prior to the beginning of the intervention. Testing consisted of the Preschool Language Scale 4th edition (PLS-4; Zimmerman et al., 2002), the Phonological Awareness Test 2nd edition (PAT-2; Robertson & Salter, 1997), the Peabody Developmental Motor Scales (PDMS; Follo & Fewell, 2000), the Social Skills Rating Scale Parent and Teacher (SSRS; Gresham & Elliot, 1990), and the Behavioural Rating Inventory of Executive Function Preschool Version (BRIEF-P; Gioia et al., 2002). Two subtests of the PAT-2 (Rhyming Discrimination and Production, and Segmentation for Words and Sentences) and the PDMS (Stationary and Locomotion) were used.

Testing took approximately one hour for each child. A speech-language pathologist with more than 10 years of experience working with children with hearing loss was hired by the researcher and completed all testing for this study. Each child was tested using the PLS-4, the PAT, and the PDMS and one parent completed the SSRS (parent version) and the BRIEF-P. The two tests for teachers, SSRS teacher and the BRIEF-P were given to the parent for their child’s teacher along with an envelope and directions regarding how the teacher was to return the completed forms to the researcher. Participants then attended twelve 45-minute, weekly sessions of either M&M or CB or waited the 12 weeks if in the control group. Post-testing was completed within one month of the final intervention class or after the 12-week waiting period. All post-testing was completed by the same speech-language pathologist in the same location as for pre-testing. Parents and teachers (when possible) also completed the same tests post intervention (SSRS, BRIEF-P). The speech-language pathologist completing the testing was not aware of the intervention group to which each child had been assigned. Families in the control group completed testing at baseline and then three months later using the same protocol as the intervention groups.

Parents in the intervention group also participated in a semi-structured interview with the researcher during post-testing that explored the experience of the sessions by both the parent and the child, specific behaviors during and between sessions related to intervention activities, and any final comments. Results of this qualitative analysis are presented elsewhere (DuBois et al., 2020).

The facilitator was videotaped during sessions to assess her consistent interaction and engagement with the children between the M&M and CB interventions to avoid possible bias in facilitation. The storybook reading section of each video was selected, cut, and randomly assigned to a folder. Eight folders with three videotaped sections were created to ensure that each video clip would be evaluated a minimum of four times. Eight students from the Department of Speech-Language Pathology were recruited and assigned one folder each to watch and evaluate the videos using a Likert Scale based on agreement (strongly disagree to strongly agree).

Data Analysis Plan

Assessment outcome values were calculated into Standard Scores for each individual test. Standard Scores were then changed to categorical outcomes based on whether scores increased or decreased for each participant post-intervention or post 12 week waiting period for the control group.

Ethical Considerations

Ethics approval was obtained from the University of Toronto and all school boards involved in recruitment for the study.

Results

Five children were lost to the interventions due to family circumstances (4 CB and 1 M&M); however three of these families agreed to be controls only (1 CB and 2 of the late-entry CB), and two were lost completely (1 CB and 1 M&M); therefore, the final data set was comprised of ten participants in the intervention data group (8 direct entry and 2 late entry) and five in the control data group (3 controls and 2 late-entry; Table 3). All children had their hearing loss identified during the newborn screening period except one whose hearing loss was not identified until two years of age. Eight mothers and two fathers participated. All families attended a minimum of 9 sessions during the intervention, with one family attending 9 of 12 sessions and 9 families attending 10, 11, or 12 of 12.
sessions. Homework sheets were collected from nine of the 10 families during the intervention. All families recorded that they had practiced activities from the group intervention twice during the week between sessions as requested by the facilitator. Families also listed instances when their child initiated activities spontaneously and what these activities were. Overall, all participants initiated activities on their own a minimum of one to two more times during the week. No post-intervention test results were available from the teachers as the interventions ran through the summer term.

**Data Analysis**

All children were post tested within one month of completing the intervention sessions. The formalized tests were scored according to their respective manual protocols and standard scores were collected in preparation for analysis. All standard scores were evaluated based on whether the score had increased or decreased post intervention and these values were used in a 2x2 chi square (intervention X control and decrease X increase) to assess change between the intervention and control groups. As the chi square assumptions were not met due to the small number of participants, a Fischer’s Exact test was used to correct for this. Results for all assessments were nonsignificant using a two-sided test and a significance level of .05 (range 0.075–1.00).

These same parameters were then used to compare the intervention groups and the controls in a descriptive manner comparing increases in standard scores. More children in the music and movement group improved post intervention in preliteracy (Table 4). Although both intervention groups had the same rhyming books read to them each week, the warmup for the M&M group involved rhymes with finger play or actions. Added to this, their activities involved moving to music throughout the sessions, whereas the craft-based group had only music playing in the background during their craft activities. The influence of moving to the rhymes influenced the impact of the rhythms as they became a whole-body experience rather than being solely auditory. Also of note are the scores of the intervention groups when compared to those of the control group. Overall, 90% of the intervention participants improved in their rhyming scores compared with 40% of the controls.

**Table 3**

*Participant Characteristics*

<table>
<thead>
<tr>
<th>Parent</th>
<th>Child age (years at recruitment)</th>
<th>Child Sex</th>
<th>Child hearing device technologya</th>
<th>Group (M&amp;M/CB)b</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. parent</td>
<td>5</td>
<td>female</td>
<td>CI</td>
<td>M&amp;M</td>
</tr>
<tr>
<td>2. parent</td>
<td>5</td>
<td>male</td>
<td>HA</td>
<td>M&amp;M</td>
</tr>
<tr>
<td>3. parent</td>
<td>5</td>
<td>female</td>
<td>HA</td>
<td>M&amp;M</td>
</tr>
<tr>
<td>4. parent</td>
<td>5</td>
<td>male</td>
<td>CI</td>
<td>M&amp;M</td>
</tr>
<tr>
<td>5. parent</td>
<td>5</td>
<td>female</td>
<td>HA</td>
<td>M&amp;M</td>
</tr>
<tr>
<td>6. parent</td>
<td>3</td>
<td>female</td>
<td>CI</td>
<td>M&amp;M (late entry)</td>
</tr>
<tr>
<td>7. parent</td>
<td>3</td>
<td>female</td>
<td>HA</td>
<td>M&amp;M (late entry)</td>
</tr>
<tr>
<td>8. parent</td>
<td>5</td>
<td>male</td>
<td>HA</td>
<td>CB</td>
</tr>
<tr>
<td>9. parent</td>
<td>5</td>
<td>male</td>
<td>HA</td>
<td>CB</td>
</tr>
<tr>
<td>10. parent</td>
<td>3</td>
<td>male</td>
<td>HA</td>
<td>CB</td>
</tr>
<tr>
<td>11. parent</td>
<td>4</td>
<td>female</td>
<td>CI</td>
<td>Control</td>
</tr>
<tr>
<td>12. parent</td>
<td>3</td>
<td>female</td>
<td>CI</td>
<td>Control</td>
</tr>
<tr>
<td>13. parent</td>
<td>3</td>
<td>male</td>
<td>HA</td>
<td>Control</td>
</tr>
</tbody>
</table>

aCI = Cochlear Implants; HA = Hearing Aids
bGroups were divided into Movement & Music (M&M), Craft-based (CB), and Control

**Table 4**

*Preliteracy: Phonological Awareness Test (PAT-2) Rhyming (Discrimination &/or Production subtests)*

<table>
<thead>
<tr>
<th>Group</th>
<th>Percentage of participants with increased standard scores post test</th>
</tr>
</thead>
<tbody>
<tr>
<td>Music and Movement</td>
<td>100%</td>
</tr>
<tr>
<td>Craft Based</td>
<td>66%</td>
</tr>
<tr>
<td>Controls</td>
<td>40%</td>
</tr>
</tbody>
</table>

The social skills scores demonstrated an increase in pro-social behaviors in the intervention group, but not in the controls (Table 5). This adds support to the idea that being in group activities with peers allows for opportunities to practice peer-to-peer interactions in natural, but supportive conditions. In the case of this intervention, a facilitator and a parent were able to both model and scaffold appropriate behaviors in a multitude of situations during the intervention making it a rich environment for watching, learning, and practicing.

**Table 5**

*Social Skills: Social Skills Rating Scale (SSRS)*

<table>
<thead>
<tr>
<th>Group</th>
<th>Percentage of participants with increased standard scores post test</th>
</tr>
</thead>
<tbody>
<tr>
<td>Music and Movement</td>
<td>71%</td>
</tr>
<tr>
<td>Craft Based</td>
<td>66%</td>
</tr>
<tr>
<td>Controls</td>
<td>0%</td>
</tr>
</tbody>
</table>

Language outcomes improved for all groups with the intervention groups having a higher percentage of participants with increased standard scores (Table 6).
Balance scores increased for both intervention groups only (Table 7); however as discussed, balance is variable in children with hearing loss making these outcomes difficult to measure and comment on with any certainty. Executive function scores improved more for the control group than for the intervention groups (decreased standard scores for the combined intervention groups was 40%, Table 8).

Table 7
Balance: Peabody Developmental Motor Scales (PDMS-2)
Locomotion and Stationary

<table>
<thead>
<tr>
<th>Group</th>
<th>Percentage of participants with increased standard scores post test</th>
</tr>
</thead>
<tbody>
<tr>
<td>Music and Movement</td>
<td>71%</td>
</tr>
<tr>
<td>Craft Based</td>
<td>66%</td>
</tr>
<tr>
<td>Controls</td>
<td>0%</td>
</tr>
</tbody>
</table>

Although outcomes were not statistically significant, these data support the idea that the addition of group activities is promising and may help to demonstrate a positive trend in outcomes for preliteracy, social skills, language, and possibly balance.

Intervention Video Evaluations

The videotapes of the sessions were initially recorded to measure consistent facilitation between the intervention groups. As the intervention outcome scores were grouped together, the variable of possible bias in facilitation was no longer relevant. Consequently, results from the student-evaluated Likert scales is not reported here as they do not add pertinent information.

Discussion

Many studies have demonstrated benefits when music and movement are used in areas of school readiness skills such as: language (Chobert et al., 2014), preliteracy (Degé & Schwarz, 2011), social (Kokal et al., 2011), executive function (Zachariou & Whitebread, 2015), and balance (Fernandes et al., 2015). Surprisingly, this study did not demonstrate significant outcomes in any of the areas of interest during the standardized testing, despite evidence of improvements in all areas during the intervention observed by both parents and the facilitator.

Limitations of this study that affected these overall outcomes statistically may have been the small intervention group (10 children), which resulted in very little power, and the length of the overall intervention (12 weeks) as compared to previous studies. Many of the music interventions previously cited included sessions over an entire school year rather than the 3 months used in this study.

A larger component of the outcome results may have been the scope of the tests used. Although all were chosen due to their reported validity and reliability in the individual specialties, their sensitivity to real life situations and function may not have been adequate for this study. Balance was one such area. Although testing did not demonstrate a significant change in balance, observations during the intervention belied the scores. As it was an easily observed change in skill during the intervention sessions, the test scores were surprising. The children walked a tape line a number of times each week in the session room, competing against both themselves and one another. By the end of the sessions, each child was able to walk the line much more easily and often very accurately to the end of the tape. They did, however, need a few practice runs to allow for precision. The test for balance did not allow for any practice and therefore did not truly represent the balance capability of each child. As balance in play, sports, or physical education has many opportunities for practice, improvements are more obvious as more practice occurs. Also, as children become more adept at these skills, practice is more satisfying. As was observed in the sessions; when each child saw improvement in their skill on the tape line, they tried harder to be better—success drove the practice, in turn supporting the use of activities to build confidence and skills in this area.

The same occurred in the area of language as test scores did not show any significant changes in language development, but there was observable change during the sessions. As the test used a particular selection of vocabulary and language skills for each age group, there was no opportunity to expand on any of the areas during testing. During the sessions, children were exposed to many new vocabulary words. Each book brought a new set of words but also different situations for language use and form (polite forms, tenses, descriptives, poetry), expansion of known vocabulary (unusual farm animals, sea creatures, baby animals), and scaffolding for skills such as how to ask a question, how to kindly help a peer, or how to ask for help.
giving clear information. The children demonstrated both vocabulary and language use gains during the sessions which are both very important language skills. Both skills, however, can be difficult to capture during a standardized test.

Another area that showed promise during sessions was that of preliteracy. Although children did not demonstrate significant improvements in rhyming ability on the test, during the intervention many of the children had great fun trying to make up words that rhymed. They would bounce ideas off one another and compete to see who could make up the most words. As all of the warm-up songs/rhymes and many of the books read in the intervention had rhyming components, the children had ample opportunity to play with rhyming. Parents reported that their children spent time both in the car ride home and with siblings playing rhyming games. This use of rhyming as a game allowed the children to expand their skill and build confidence in an area of literacy preparation. Again, although the test had sections for both discriminating whether or not two words rhymed and producing a word that rhymed with the one given by the tester, it had a set list of words to be tested and no room for expansion, thereby limiting the child’s opportunities. Word and sentence segmentation added another unforeseen challenge for the children. Children with coordination challenges were not able to demonstrate their abilities well because this test relied on clapping or tapping to demonstrate the various segments of a sentence or word. As has been discussed, children with HL often have motor challenges (Livingstone & McPhillips, 2011) which take some time to mature possibly making their test results under representative of ability.

Social skills were also difficult to test. A number of challenges arose; (a) the test was a parent questionnaire possibly adding bias to the answers given, (b) a second bias related to exposure to a group, and (c) despite the test including a Teacher Questionnaire component, teacher evaluation was not able to be accessed due to timing of the intervention. The value of teacher input may also not have been representative of the child’s social skills, however, since the difficulties of assessing one child’s peer-to-peer interactions in a busy classroom or playground setting would be challenging. Parents completed the questionnaire before the intervention began and based their answers on observed behaviors of their child at home. It was later divulged to the facilitator that many of the parents had never seen their child interact with peers, only siblings. Consequently, many of the participating children scored lower in social skills after the interventions possibly based on parents’ perception of their child’s behaviors when compared to that of their group mates’. Once again, many improvements in social skill development were observed during the sessions. The facilitator used scaffolding to help children during interactions intrinsic to the activities (sharing, taking turns, requesting), and in peer-to-peer discussions during story time or joining and leaving the group. As the sessions progressed, the children were able to consistently use the skills practiced with their peers, helping to build confidence for further practice and use in the classroom. The two children whose scores decreased the most in the post test according to their parents, actually improved the most during the sessions with evidence of greater consideration of their peers. Unfortunately, this was not demonstrated in their post intervention scores.

The final area of challenge for testing was executive functions (EF). Although other more objective tests have been used in research (e.g., Go-No Go, Dimensional Change Card Sort, Marshmallow Test) they do not test function in real life situations. Therefore, like social skills, EF was tested using a parent questionnaire. This questionnaire had the same possible biases as social skills test; it too depended on parent judgement of the child before and after participation in the intervention. Once again, the input from the teacher component was not accessible due to timing. The teacher’s evaluation of peer-to-peer use of EF may not have been representative in this case. Focus, memory, and flexibility in the classroom, however, may have shed some light on academic areas of development. During the intervention, many instances of improvement were observed. Children were often corrected by their peers if they were being disruptive. This resulted in an immediate change in behavior, supporting the idea that children are often able to support and model appropriate behaviors with their peers. Each group demonstrated this with different children being the model or enforcer at different points in the intervention. It was also observed that children reacted very differently when a peer gave the correction as compared to when the parent gave it. The children seemed to understand that it was important to behave in a particular manner to be part of the group. This ability to self-regulate for inclusion is important in the classroom and the children were able to watch and learn as well as practice strategies during the group sessions.

As skill development was observed during sessions, it was surprising when test results did not reflect this. Most were not measurable in testing as there was no method to observe how skills were used in context during the standardized tests. Parents also commented that the sessions provided a safe environment for their children and might have supported growth as they all understood that they had HL and felt part of a common group. The children helping each other was also observed in multiple instances during the intervention sessions (e.g., initial sound in words, getting a friend’s attention, supporting successes, competing on the taped line). It is clear therefore that it is important to gain a more complete picture of the child; within their own world of family, school, and other activities; when deciding how best to support development.

Parent involvement in sessions is also important to consider. Parents have reported a need for more information and ongoing support for their children (Jackson, 2019). Adapting the modeling, strategies, and advocacy (for self and teaching modelling for child) to real life situations helps both parents and children use the demonstrated skills on a daily basis. Because there is typically no way of measuring what is practiced and reinforced day-to-day, the homework sheets used in this study demonstrated
that children practiced skills from sessions at home, both alone and with family members, each week. Hopefully parents saw the benefits of joint participation and continued to encourage and support these activities at home by participating with their child even after the sessions were completed.

Conclusion
As this study demonstrated, being able to measure abilities in functional settings is paramount to ultimate success for this population. Using purely formalized testing did not show improvement even though observations during intervention sessions showed a few examples or at times multiple instances of skill development. One suggestion of how to glean a clearer view of the child in his or her world would be to use behavioral observations along with formalized testing. This would allow for a more complete evaluation of the child and his or her challenges, thereby allowing for a more appropriate and individual set of goals. In the case of this study, outcome measures would have benefitted from an observer scoring a set of criteria related to social skills and executive functions as well as balance that could have supplemented what was seen in the standardized testing. Observations in areas such as peer-to-peer interactions (initiation, sharing, vocabulary and language use, empathy, self-regulation, listening strategies, and advocacy) would have given a more complete idea of areas for future support and scaffolding for each child. This would, in turn, allow for the creation of goals related to areas needing support which could then be incorporated into real life activities with opportunities for practice.

Behaviors are complex and dynamic, making it imperative that their assessments reflect this. Helping children with HL to catch up to their peers and continue to build school-readiness skills needs accurate observation and continued evaluation so that skills can move on the same trajectory as classmates. Although standardized tests accurately assess the child’s ability with regards to the specific test and in those particular circumstances, they may not access the child’s full potential or flag challenges not addressed by the assessment tool. Those working with this population and assessing their progress would have a more comprehensive view of outcomes if functional measures of skill were assessed. This would then ensure that outcomes were not solely based on test scores, but rather on a more complete picture of the child in a functional role. Consideration of the child as a member of society trying to learn how to function and be successful in all aspects of life (i.e., family, academics, social, and self-regulation abilities) must be the goal. Representative outcomes guiding functional habilitation is the means to the attainment of ultimate success both in the classroom and beyond.

References


The development and distribution of this material was supported in part by the Maternal and Child Health Bureau (MCHB) of the Health Resources and Services Administration (HRSA) as part of award U52MC0439, totaling $3,400,000; and as part of award 2UJ1MC30748 04 00, totaling $1,800,000. The contents are those of the author(s) and do not necessarily represent the official views of, nor an endorsement by, HRSA, HHS, or the U.S. Government.

From the American Academy of Pediatrics (AAP):

From the National Center for Hearing Assessment and Management (NCHAM):
- https://www.infanthearing.org/components/


Diagnostic Infant Auditory Brainstem Response Testing Via Telehealth: A Survey of Professional Opinions and Current Barriers

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Douglas P. Sladen, PhD¹

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Abstract

**Purpose:** To examine the barriers to Washington State audiologists adopting telehealth as a means of improving accessibility to diagnostic audiology for infants.

**Methods:** A Qualtrics survey was distributed via e-mail and social media. Survey participants were required to be audiologists practicing in Washington State. The sixteen-question survey consisted of topics related to participant demographics, previous telehealth experience, and barriers to the use of telehealth for diagnostic infant auditory brainstem response (ABR) testing. A total of 17 participants completed the survey.

**Results:** Survey responses indicated that Washington State audiologists are largely neutral or disagree with telehealth being an effective means of performing remote diagnostic ABRs. Participants primarily identified equipment cost as a barrier, and had varying opinions regarding insurance reimbursement, internet connection, privacy, and ability to counsel.

**Conclusions:** This study identified several barriers to the implementation of remote diagnostic ABR testing in Washington State. The neutral and negative view of telehealth for diagnostic infant ABR points to the need for education among Washington State audiologists. Disseminating information on the efficacy of telehealth to audiologists is a likely next step in reframing the current attitude toward remote diagnostic ABR and working toward reducing loss to follow-up rates for rural families.

**Keywords:** telehealth, infant, hearing loss, diagnosis, loss to follow-up, auditory brainstem response (ABR)

**Acronyms:**
- ABR = auditory brainstem response
- JCIH = Joint Committee on Infant Hearing

**Acknowledgements:** We have no known conflict of interest to disclose.

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Undiagnosed childhood hearing loss inhibits the development of spoken language, social skills, and cognition. To mitigate the negative impact of hearing loss on child development, the Joint Committee on Infant Hearing (JCIH, 2019) recommends a “1-3-6” approach for early intervention; infants should: (a) be screened for hearing loss by one month of age, (b) if hearing loss is present, receive diagnosis by three months of age, and (c) if hearing loss is present, receive early intervention services by six months of age. In 2018, 25.3% of Washington State infants were lost to follow-up after a refer on newborn hearing screening (Center for Disease Control and Prevention, 2018). This percentage varies greatly among screening centers, with as many as 44% to 100% of infants remaining undiagnosed after a refer on newborn hearing screening at centers across the state (Washington State Department of Health Early Hearing Detection and Diagnosis and Intervention, 2019; Figure 1).

The wide variance in loss to follow-up rates across the state is likely due, in part, to the issue of the health service disparity between urban and rural communities, as evidenced by lower loss to follow-up rates in densely populated counties (e.g., King, Pierce, Snohomish, Spokane), and higher loss to follow-up rates in sparsely populated counties in Central Washington (e.g., Yakima, Douglas, Okanogan; Washington State Department of Health Early Hearing Detection and Diagnosis and Intervention, 2019; Figure 1). Families in rural areas experience barriers to hearing health services such as travel distance and access to specialized pediatric audiologists (Hatton et al., 2019). These barriers may prevent families from receiving appropriate diagnostic services, including diagnostic auditory brainstem response (ABR) testing following a refer on newborn hearing screening. Previous studies have suggested telehealth as a viable means of service provision in rural communities (Hatton et al., 2019; Stuart, 2016). However, there has been limited progress toward implementing telehealth for diagnostic audiology in Washington State.
performed remote diagnostic ABR testing on 22 infants with a referred hearing screening. Among these infants, 59.1% were diagnosed with some form of permanent or transient hearing loss. Overall, none of the infants were lost to follow-up, compared to the 22% loss to follow-up rate previously recorded in that region. This indicates that telehealth is a powerful tool in reducing loss to follow-up rates (Dharmar et al., 2016).

Together, these studies confirm the feasibility of remote diagnostic ABR testing and support the idea that telehealth lowers loss to follow-up rates in rural areas (Dharmar et al., 2016; Hatton et al., 2019; Stuart, 2016). Despite the success of remote diagnostic ABR programs, the uptake of telehealth for audiology has been limited, due to the lack of published literature, high equipment costs, and inconsistencies in internet connection (Polovoy, 2008). Audiologists themselves have identified infrastructure, training, and reimbursement as major barriers to the use of teleaudiology (Ravi et al., 2018). However, there is limited information on clinician perceptions of the applications of telehealth in audiology. Examining these barriers and perceptions among audiologists will assist in understanding why telehealth has not been adopted for remote ABR testing.

Research Questions

Several challenges have affected implementation of remote ABR testing in Washington State and across the nation. Barriers including costs, professional opinions, technical effectiveness, privacy, and counseling all require additional research (Ravi et al., 2018). The primary purpose of the present study was to investigate the lack of movement toward telehealth as a means of improving accessibility to diagnostic
audiology for infants in Washington State. Specifically, the study aimed to answer the two following questions.

1. Would professionals use telehealth for diagnostic ABR testing if made available?
2. Do professionals believe a telehealth model would improve service provision for rural families in the region?

Method

Participants

Participants included Washington State audiologists who perform pediatric ABR testing. Though the exact number of pediatric ABR providers in Washington State is unknown, the Washington State Department of Health (2020) reports 29 diagnostic audiology clinics for infants. Participant information related to years of experience, geographic location, number of diagnostic infant ABRs performed in a month, and number of infants lost to follow-up at their place of work in 2018 was collected.

Survey

Survey questions were developed based on the available literature identifying barriers to the use of telehealth in audiology. The survey consisted of two questions required for participation in research, two questions related to demographics, two questions surrounding infant ABR experience, one question regarding previous telehealth experience, and nine questions related to opinions and barriers to the use of telehealth for diagnostic infant ABR, for a total of 16 questions (see Appendix A). Among these questions were six multiple-choice questions, one drop-down menu question, nine Likert scale questions, and an additional optional text-box to give participants the opportunity to submit any questions or comments regarding the survey content. Once participants began the survey, they were given two weeks to complete it. During this two-week period, participants were able to save their progress and return later. The survey was available for 15 weeks, between December 19th, 2019 and April 4th, 2020.

Procedure

This study was approved by the Western Washington University Institutional Review Board (IRB#: 3351EX19). The survey was developed using Qualtrics, an online survey-building program licensed through Western Washington University. Participants accessed the survey through a secure and anonymous link that was distributed through social media and e-mail. The reusable link and scripted instructions were posted on December 9, 2019 and approximately one month later on January 7, 2020, to several audiology Facebook pages and emailed directly to various Washington State audiologists. In accordance with the Western Washington University Human Subjects Research Protocol, an informed consent statement was included at the beginning of the survey to inform participants of their rights and the nature of the study. All participants indicated that they read the informed consent statement and agreed to participate in the survey.

Results

A total of 45 participants opened the survey and a total of 17 participants completed it. The final responses came from King, Spokane, Whatcom, Clark, Pierce, San Juan, and Snohomish Counties. Years of experience varied greatly with 23.5% (4) of the participants reporting 0–5 years of experience, 29.4% (5) reporting 6–10 years of experience, 23.5% (4) reporting 11–15 years of experience, 11.8% (2) reporting 16–20 years of experience, and the remaining 11.8% (2) reporting greater than 20 years of experience. On average, the survey took three minutes to complete.

The majority (58.8%) of the participants reported performing 1–5 diagnostic infant ABRs per month on average, with 11.8% (2) performing 6–10, 5.9% (1) performing more than 15, and 23.5% (4) performing none, which may mean they only perform a few in any given year or previously performed ABR testing and do not do so now. When asked to report how many infants were lost to follow-up at their place of work in 2018, 53.3% (8) reported 1–10 infants lost to follow-up, 26.7% (4) reported no infants lost to follow-up, 13.3% (2) reported 11–30 infants lost to follow-up, and 6.7% (1) reported 31–50 infants lost to follow-up. Only 17.6% (3) of the participants reported using telehealth to provide audiologic services prior to taking the survey.

Participants responded to the following statement “I view telehealth as an effective means of performing diagnostic infant ABRs.” Just over half (52.9%, 10) of participants were neutral regarding their opinion of the efficacy of telehealth for diagnostic infant ABRs or did not know enough to make an informed decision. Among the rest of the participants, 35.2% (6) either disagreed or strongly disagreed with this statement, and the remaining 11.8% (2) either agreed or strongly agreed. The majority of participants (64.7%, 11) disagreed or strongly disagreed that many infants in their community are lost to follow-up because they do not have access to diagnostic ABR. A small portion (23.5%, 4) were neutral with this statement, and only 11.8% (2) of participants agreed.

The participants were asked to rate their opinion of various barriers to the use of telehealth, including insurance reimbursement, equipment cost, internet connection, privacy, and ability to counsel remotely (Figure 2). Regarding insurance reimbursement, 52.9% (9) of the participants were neutral, 35.3% (6) agreed, and 11.8% (2) strongly agreed. The majority of the participants (64.7%, 11) either agreed or strongly agreed with the statement, “Equipment cost is a barrier to the use of telehealth for remote diagnostic infant ABR,” with 29.4% (5) being neutral and the remaining 5.9% (1) disagreeing with the statement. A large portion (47.1%, 8) of participants were neutral about internet connection being a barrier to the use of telehealth for remote diagnostic ABR, with the rest of the responses divided almost evenly between those who agreed (29.4%, 5) and those who either disagreed or strongly disagreed (23.5%, 4) with internet connection being a barrier. In response to the statement “Privacy is a barrier to the use of telehealth for remote diagnostic infant ABR,” the participants were split evenly across responses with 29.4% (5) agreeing, 29.4% (5) disagreeing, and 29.4% (5) being neutral. The remaining 11.8% (2) of participants strongly disagreed with this statement. When asked to respond to the statement “Ability to counsel remotely is a barrier to the use of telehealth for remote diagnostic infant ABR,” 47.1% (8) of the participants...
Figure 2
Participants' Opinions of Various Barriers to Auditory Brainstem Response (ABR) Testing Via Telehealth

<table>
<thead>
<tr>
<th></th>
<th>Strongly Disagree</th>
<th>Disagree</th>
<th>Neutral</th>
<th>Agree</th>
<th>Strongly Agree</th>
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<td>20% (4)</td>
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<td>6% (1)</td>
</tr>
<tr>
<td><strong>Equipment Cost</strong></td>
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<td>40% (7)</td>
<td>20% (4)</td>
<td>10% (2)</td>
<td>6% (1)</td>
</tr>
<tr>
<td><strong>Internet Connection</strong></td>
<td>30% (5)</td>
<td>40% (7)</td>
<td>20% (4)</td>
<td>10% (2)</td>
<td>6% (1)</td>
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<tr>
<td><strong>Privacy</strong></td>
<td>30% (5)</td>
<td>40% (7)</td>
<td>20% (4)</td>
<td>10% (2)</td>
<td>6% (1)</td>
</tr>
<tr>
<td><strong>Ability to Counsel</strong></td>
<td>30% (5)</td>
<td>40% (7)</td>
<td>20% (4)</td>
<td>10% (2)</td>
<td>6% (1)</td>
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</tbody>
</table>

**Discussion**

Overall, results of the present study indicate that Washington State audiologists are largely neutral or disagree with remote ABR testing being an effective diagnostic tool for assessing hearing loss in infants. Admittedly, there are limited peer-reviewed studies on the applications of telehealth in audiology, which may contribute to the misconception or ambivalence among audiologists. However, the available literature supports the efficacy of a telehealth approach for infant ABRs and confirms that remote diagnostic ABR yields comparable results to traditional, face-to-face versions (Hatton et al., 2019; Stuart, 2016).

**Equipment Cost**

Still, many barriers obstruct the widespread use of telehealth in Washington State. One of the primary barriers identified by audiologists sampled in the current study was equipment cost. Particularly in rural communities, in which audiologists would otherwise incur travel costs to conduct ABRs, remote ABR models provide direct travel cost savings (Hatton et al., 2019). In the study design used by Hatton et al. (2019), the cost to equip a complete telehealth ABR system was $9000, indicating that this approach can be highly cost effective.

**Insurance Reimbursement**

Most participants of the current study stated they were neutral or did not know enough information to make an informed decision about insurance reimbursement. Though many other fields use telehealth throughout the course of diagnosis and treatment, there are no current federal or Washington State standards for reimbursement of remote audiology services. Rather, the individual payer determines reimbursement (Polovoy, 2008; ASHA, n.d.). As a result, audiologists are largely restricted to providing face-to-face services, posing a significant barrier to the use of telehealth in the field of audiology.

Currently, many audiologists are not able to provide in-person services due to the COVID-19 pandemic. In response, the Centers for Medicare & Medicaid Services released an update on April 30th, 2020 that includes audiologists as eligible providers for reimbursement of certain telehealth services.

**disagreed**, 29.4% (5) either agreed or strongly agreed and, the remaining 23.5% (2) were neutral. Approximately half of participants (47.1%; 8) agreed or strongly agreed with the statement “If the technology and training were made available for my workplace, I would feel comfortable diagnosing an infant with hearing loss remotely.” A large portion of participants (35.3%; 6) disagreed or strongly disagreed, and the remaining 17.6% (3) were neutral.

**Internet Connection**

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**Ability to Counsel**

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(ASHA, 2020a, 2020b). This expansion is retroactive to March 1st, 2020 and will continue for the duration of the public health emergency. To date, however, ABR testing has not been listed as a covered service under the Medicare telehealth benefit. According to the American Academy of Audiology (2020) this lack of coverage does not necessarily mean audiologists are prohibited from providing remote ABR services. Patients are able to reimburse the audiologist directly for uncovered services. Though this is an imperfect solution, it is promising that professional organizations are lobbying for audiologists to be included in coverage for telehealth services.

Internet connection

The use of telehealth has also been hampered by the internet capacity required for remote ABR technology, and its availability in rural communities. The audiologists surveyed in the present study were largely neutral regarding the issue of internet connection. In a study conducted by Hatton et al. (2019), the authors used the previously existing broadband infrastructure to conduct remote ABR testing. Reportedly, the authors did not encounter connectivity issues (Hatton et al., 2019). However, additional research is needed to determine the necessary network requirements for remote diagnostic ABR testing, particularly for a combined synchronous and asynchronous approach.

Privacy

Privacy issues may be one of the most challenging barriers to the use of telehealth, especially in cases where audiologists use video interface technology. Audiologists are bound by the Health Insurance Portability and Accountability Act of 1996 (HIPAA), as well as individual state privacy requirements (Denton & Gladstone, 2005). Though HIPAA-compliant video interface platforms are available, one must ensure that all transactions of personal health information are secured when being transmitted electronically. The respondents in the present study were split evenly between being neutral, agreeing, and disagreeing with the concept of privacy as a barrier.

Considering the recent COVID-19 pandemic, the U.S. Department of Health & Human Services (HHS; 2020) issued a statement temporarily waiving the enforcement of HIPAA requirements for the duration of the federally declared national emergency (ASHA, 2020a, 2020b). According to the HHS Office, providers can use any non-public facing video or audio communication product (e.g., Zoom, Skype, Google Hangouts) to provide telehealth during the national emergency.

Despite these recent developments, the perception of privacy issues may also limit families from wanting to partake in a telehealth model. In a study conducted by Dharmar et al. (2016), the participating parents were surveyed and overwhelmingly reported to be comfortable discussing their child’s hearing status remotely. However, further research is needed to determine patient and provider perception of privacy issues and potential safeguards.

Counseling

Finally, in the case that hearing loss is diagnosed, there must be a tactful approach to counseling remotely. Polovoy (2008) interviewed William Campbell, the Infant Hearing Program audiologist at the Thunder Bay District Health Unit in Ontario. Campbell’s program uses both synchronous video conferencing and a data stream, which allows the audiologist to control the remote ABR equipment. Campbell discussed the challenges of diagnosing a hearing loss remotely and how it may not be appropriate to discuss sensitive news in a telehealth format. To address this issue, social workers at the Thunder Bay District Health Unit are collaborating with audiologists to develop a protocol in the case of a remote hearing loss diagnosis.

One participant in the present study wrote, “For me, counseling via video would be the most difficult barrier to overcome. In my position, I have needed to use video interpreters for families on occasion, and these have been the most challenging counseling sessions by far. However, if a family did not have another choice, I would much rather offer telehealth service and diagnose a baby than miss them.” Diagnosing a permanent childhood hearing loss during face-to-face appointments must be done clearly and empathetically. The same level of care must be achieved during remote diagnostic appointments as well.

It is promising that many audiologists responded that they would feel comfortable diagnosing a hearing loss remotely if the technology and training were made available. However, the majority of participants were either neutral or disagreed, further emphasizing the varied attitudes of audiologists toward a telehealth approach to diagnostic ABR testing and counseling.

Equipment set-up

One topic not included in this survey was audiologists’ opinion on collaboration with support staff for equipment set-up (e.g., scrubbing, electrodes and impedance, filters). Multiple participants addressed this issue in their response: “There are so many nuances to performing ABR on infants. Doing this remotely would require a highly trained person on the other end [and] does not negate the need for expensive equipment”; “Through Telehealth, who will prep the infant and apply electrodes and ear inserts?”; “Electrode montage setup and proper placement of earphones cannot be done remotely. At a minimum a highly trained and competent technician would need to be with the infant in person.” Certain programs have successfully employed local support personnel or technicians to place the transducers and electrodes required to record an ABR. A model described by Polovoy (2008) sends the necessary equipment to a technician at the local health center or hospital, who then connects the infant. At that point, the remote audiologist will take control of the computer, complete an impedance check, interact with the family and conduct the ABR once the infant settles or falls asleep. In this model, the technician only requires minor supplemental training, indicating that this approach can be effective even with limited resources (Polovoy, 2008).

Limitations and Future Research

The present study has several limitations. Primarily, the small sample size means the findings cannot be generalized to reflect the opinions of all audiologists in Washington State.
Future studies may be able to gather more information from a larger group of audiologists. Likewise, the majority of participants were from King County, which incorporates some of the more populated areas in Washington State. Therefore, it is not surprising that most of the participants did not identify access to diagnostic ABR testing as a major barrier. It would be beneficial to focus on gathering responses from rural communities, who tend to see more issues with loss to follow-up.

Despite the limitation of a small sample size, these data are relevant in terms of informing what to do next. The neutral and negative view of telehealth for diagnostic infant ABR points to the need for education among Washington State audiologists. Disseminating information on the efficacy of telehealth to audiologists is a vital step in reframing the current attitude toward remote diagnostic ABR and working toward reducing loss to follow-up rates for rural families.

**Conclusion**

Remote diagnostic infant ABR testing is an evidence-based way to diagnose infants with hearing loss in rural communities and reduce loss to follow-up. However, several barriers remain in its implementation in Washington State, including the negative view audiologists have toward telehealth and its applications. Once these barriers are addressed, the audiology community can promote the uptake of remote diagnostic ABR and working toward reducing loss to follow-up rates in Washington State and beyond.

**References**


Appendix A

Western Washington University: A Telepractice Model for Diagnostic Infant ABR Testing: Professional Opinions and Current Barriers

Welcome!

We are asking you to take part in a research study. Participation is voluntary. The purpose of this form is to give you the information you will need to help you decide whether to participate. Please read the form carefully. You may ask questions about anything that is not clear. When we have answered all of your questions, you can decide if you want to be in the study or not. This process is called "informed consent."

The aim of this survey is to evaluate the reasons why telepractice has not been adopted to improve accessibility to diagnostic audiology for infants in Washington State. A secondary aim of the survey is to answer whether audiologists would use telepractice for diagnostic ABR if made available and further, if they believe a telepractice model would improve service provision for rural families.

Your perspective as an audiologist is valuable to this topic. Your responses in this survey may reveal patterns related to service provision for rural communities across Washington State.

- The survey will take less than 5 minutes to complete.
- You may use the back button to visit earlier questions.
- You will have the option to save your progress, exit, and return to complete the survey later.
- None of your personal information will be collected in this survey.
- The data collected here will be kept secure and will not be traceable back to you.
- There is no predicted risk or discomfort related to these questions.
- You may choose to NOT answer any question or exit the survey at any time. If you do not know the answer to a question, you can leave it blank.

If you have any questions, please contact us directly. Haley Prins, prinsh@wwu.edu or Douglas Sladen, douglas.sladen@wwu.edu.

If you have any questions about your rights as a research participant, you can contact the Western Washington University Office of Research and Sponsored Programs at compliance@wwu.edu or (360) 650-2146. Thank you for your time!

You can download a copy of this form to print for your records using the following link: Consent Form
Q1 I have read the above information and I agree to participate in this survey.

○ Yes, I agree to participate.
○ No, I do not agree to participate.

Q2 Are you at least 18 years of age?

○ Yes
○ No

Q3 How long have you worked as an audiologist?

○ 0-5 years
○ 6-10 years
○ 11-15 years
○ 16-20 years
○ > 20 years

Q4 In which county do you work? Please select an option
▼ Adams (1) ... Yakima (39)

Q5 On average, how many diagnostic infant ABRs (following a referred NBHS) do you perform in a month?

○ 0
○ 1-5
○ 6-10
○ 7-15
○ > 15

Q6 At your place of work, how many infants were lost to follow up following a failed NBHS in 2018?

○ 0
○ 1-10
○ 11-30
○ 31-50
○ > 50
Q7 Have you used telehealth to provide any audiologic services before?

- Yes
- No

Q8 Please respond to the following statements.

<table>
<thead>
<tr>
<th>Strongly agree (1)</th>
<th>Agree (2)</th>
<th>Neutral, I don't know enough about it to make an informed decision (3)</th>
<th>Disagree (4)</th>
<th>Strongly disagree (5)</th>
</tr>
</thead>
<tbody>
<tr>
<td>I view telehealth as an effective means of performing diagnostic infant ABRs.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Many infants in my community are lost to follow up because they do not have access to diagnostic ABR.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Insurance reimbursement is a barrier to the use of telehealth for remote diagnostic infant ABR.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Equipment cost is a barrier to the use of telehealth for remote diagnostic infant ABR.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Internet connection is a barrier to the use of telehealth for remote diagnostic infant ABR.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Privacy is a barrier to the use of telehealth for remote diagnostic infant ABR.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ability to counsel remotely is a barrier to the use of telehealth for remote diagnostic infant ABR.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>The use of telehealth for remote diagnostic infant ABR would improve service provision to families in my community.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>If the technology and training were made available for my workplace, I would feel comfortable diagnosing an infant with hearing loss remotely.</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Q9 Please use the space below to write any questions or comments regarding this survey.

_____________________________________________________________________

Note. ABR = auditory brainstem response; NBHS = newborn hearing screening.
Visual Reinforcers Designed for Children with Developmental Disabilities

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Abstract
A library of visual reinforcers has been created to facilitate visual reinforcement audiometry (VRA) testing in children with developmental disabilities. The library includes 45 reinforcer sets—photos or videos grouped by a common theme—that were created based on commonly reported interests of children with developmental disabilities. Each reinforcer set contains a minimum of 20 unique photo or video files that can be downloaded in two formats: one for commercially available VRA reinforcement systems and another for a custom setup. The library is freely available for download online under a Creative Commons License (Creative Commons Attribution-NonCommercial 4.0 International License). Use of these materials has the potential to improve behavioral testing outcomes for children with developmental disabilities, including children with restricted interests. Future research is needed to determine the effectiveness of implementing these materials in clinical settings.

Keywords: developmental disabilities, visually reinforced audiometry, hearing test, restricted interests, autism, pediatric audiology

Acronyms: ABR = auditory brainstem response; ASD = autism spectrum disorder; CPA = conditioned play audiometry; OAE = otoacoustic emissions; VRA = visual reinforcement audiometry; VROCA = visually reinforced operant conditioning audiometry

Acknowledgment: This work was supported by the American Speech-Language-Hearing Foundation (awarded to Angela Y. Bonino) and the Leadership Education in Neurodevelopmental Disabilities Audiology Supplement under Award T73MC11044 (Awarded to Principal Investigator, Sandra Friedman) from the Health Resources and Services Administration (HRSA) of the U.S. Department of Health and Human Services (DHHS). The content is solely the responsibility of the authors and does not necessarily represent the official view of, nor an endorsement by, the HRSA, the U.S. DHHS, or the U.S. Government. Thanks are extended to Emma Carpra, Madison Graham, Emily Nightengale, and L. J. Werner for their assistance during the development of this project.

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For infants (> 6 months) and children, an audiogram is the gold standard of hearing health care (American Academy of Audiology [AAA], 2020) and is the cornerstone upon which a correct differential diagnosis and access to the appropriate interventions are built. However, audiologists often report that it is difficult to obtain accurate behavioral thresholds for children with developmental disabilities (e.g., Gans & Gans, 1993; Widen, 1990). Developmental disabilities are a group of conditions that result in impairments in physical, language, learning, or behavior functioning and are estimated to occur in 8.4% of children under 5 years worldwide (Global Research on Developmental Disabilities Collaborators, 2018). When children’s developmental profiles are mismatched with the developmental demands of the behavioral testing method, thresholds may not be obtained or it may require multiple visits to complete an audiogram. In this situation, audiologists may heavily rely on physiological measures (e.g., otoacoustic emissions [OAE] and auditory brainstem response [ABR]) to determine hearing status. Although these tests are vital components of the assessment battery, physiological measures only provide partial information about the auditory system’s integrity which limits their ability to determining hearing abilities in children with auditory neuropathy spectrum disorder or other neural hearing losses (e.g., Berlin et al., 2010). OAEs are prone to missing mild hearing loss cases (Johnson et al., 2005); and although ABR thresholds generally predict behavioral thresholds by 5 to 10 dB, they can be misaligned for some children (e.g., McCreery et al., 2015). Furthermore,
children with developmental disabilities often require general anesthetics or sedation drugs for ABR testing (Rumm et al., 1990; Valenzuela et al., 2016) which may be contraindicated because of underlying medical conditions or concern of developmental harm (U.S. Food and Drug Administration, 2016). Limitations in our current behavioral testing methods leave children with developmental disabilities vulnerable to delays in the differential diagnosis process and in the enrollment of appropriate, targeted intervention services. Moreover, for children with permanent hearing loss, difficulty obtaining reliable behavioral thresholds can introduce error in the quality of the hearing aid fit—a key predictor of language outcomes (Tomblin et al., 2015)—placing children with developmental disabilities at increased risk for poor outcomes.

There are a variety of potential factors—related to the audiologist, the child, and the test method—that can make it difficult to collect reliable behavioral thresholds from children with developmental disabilities (McTee et al., 2019). First, some audiologists have limited training and/or experience with developmental disabilities (e.g., Dittman & Brueggegan, 2003; Peter et al., 2019; Peterman et al., 2018). Audiologists with limited experience may have difficulty instructing the child, training the child to perform the task, or judging the child’s responses which may be atypical or inconsistent. Second, children may have specific conditions or challenges that make it difficult to have a successful testing session. Children with developmental disabilities can have sensory sensitivities, transition difficulties, or perceive aspects of the testing protocol or environment to be aversive (e.g., American Psychological Association [APA], 2013; Gomes et al., 2004; Richier et al., 2007). Furthermore, some developmental conditions are associated with a high prevalence of anxiety as a secondary diagnosis (e.g., White et al., 2009), which can result in children being anxious if working with an unfamiliar person and/or in a new setting. Finally, current behavioral methods are based on the assumptions of typical child development and auditory behavior (Diefendorf & Tharpe, 2017), making them not well-suited for children with diverse or complex developmental profiles.

One example of a clinical method that is based on the assumptions of typical development is visual reinforcement audiometry (VRA). VRA is the recommended method for obtaining behavioral thresholds from infants and children with a developmental age of 5 to 24 months (AAA, 2020). In this method, children are taught to make a head-turn response toward a visual reinforcer—mechanical toy or brief video—based on the observation that typically-developing infants make a reflexive head-turn response to sound (Muir et al., 1989; Widen, 1993). However, preschool children with autism spectrum disorder (ASD) are less likely than neurotypical, mental age-matched peers to orient to sounds in their environment (Dawson et al., 2004). A second example of a misalignment between the demands of the task and the abilities of children with developmental disabilities is that VRA requires frequent shifts in attention (e.g., shifting between the test assistant and the reinforcer). However, this is often an area of concern for children with intellectual disabilities and/or ASD (Liss et al., 2006). Another example, which will be discussed in detail below, is that the reinforcers commonly used in clinics may not align with the interests of some children with developmental disabilities, reducing their effectiveness. These examples highlight the limitations of VRA for evaluating hearing in children with developmental disabilities and may, at least in part, explain the reported challenges for measuring thresholds with VRA in this population (e.g., Gans & Gans, 1993; Greenberg et al., 1978; Meagher et al., 2020; Nightengale et al., 2020).

The motivation of this article is to improve behavioral assessment of children with developmental disabilities by creating a library of visual reinforcement materials that are tailored to the needs of this population. VRA data from typically-developing infants and young children have well established that the quality of the reinforcement affects the number of trials that are performed prior to habituation (Moore et al., 1975, 1977; Primus & Thompson, 1985). The above studies demonstrate a clear advantage for complex and/or novel reinforcement, with mechanical toys and brief videos being equally effective in clinical settings (Doggett et al., 2000; Lowery et al., 2009; Schmida et al., 2003). However, no VRA studies have compared the effectiveness of different reinforcement types for children with developmental disabilities. Because the reinforcers used in commercially available VRA reinforcement systems were designed for typically-developing infants, these reinforcers may not be appropriate for children with developmental disabilities for two reasons. First, because of their developmental abilities or sensory sensitivities, some children with developmental disabilities are tested with VRA outside of the chronological age range recommended for VRA. Current clinical audiologists may not be engaging or motivating for some chronologically older children: they require reinforcers that are aligned with their developmental interests, not those of infants. Second, some children with developmental disabilities have restricted interests. Children with restricted interests demonstrate a strong or intense preoccupation with one or more specific topics or objects (APA, 2013). Having restricted interests is a hallmark feature of ASD (e.g., Richier et al., 2007) but is seen in other developmental conditions, including Down syndrome (e.g., Evans et al., 2014). Although circumscribed interests are unique to an individual, interests do vary in type and degree between developmental profiles and a child’s interest(s) can change over time (Evans et al., 2014; Joseph et al., 2013; Richier et al., 2007). Previous ASD intervention research has demonstrated that using objects or games related to a child’s circumscribed interests improves outcomes (e.g., Baker et al., 1998; Boyd et al., 2007; Kryzak et al., 2013; Kryzak & Jones, 2014). Drawing on this research, audiologists may be able to obtain more thresholds from children if the reinforcer is related to a child’s circumscribed interest(s). Thus, we created a library of visual reinforcers that are based on commonly reported interests of children with developmental disabilities to facilitate behavioral testing. Here we provide a collection
of 45 reinforcer sets comprised of either photos or videos. A description of the material generation process, implementation recommendations, and access to the materials follows.

**Creation of Materials**

**Selection of Themes for Reinforcer Sets**

A list of potential themes for the reinforcers was generated based on commonly reported circumscribed interests for children with developmental disabilities by clinicians or in the literature (Anthony et al., 2013; Caldwell-Harris & Jordan, 2014; Klin et al., 2007; Turner-Brown et al., 2011). This list was reviewed by all authors, as well as by an additional external reviewer. Three of the authors and the external reviewer all have more than 10 years of experience working with children, including children with developmental disabilities, as an audiologist (n = 2), developmental psychologist (n = 1), or early childhood educator (n = 1). Reviewers were asked to provide feedback on the list of themes. Additionally, reviewers were asked to identify other topics that they thought would be appropriate based on their professional experience, especially for children with restricted interests. This process resulted in a list of 63 potential themes for the reinforcers.

**Selection of Photos or Videos**

Potential digital materials for each theme were identified using three open-access, online depositories: www.flickr.com, www.pexels.com, and www.pixabay.com. Based on the amount and quality of materials identified, a decision was made to use either photos or videos for each reinforcer set. For each reinforcer set, 20 to 40 unique photos or videos were selected. The one exception is that only 14 videos were identified for the theme of flushing water (e.g., toilets and drains). All digital materials were required to be of high-quality and be in the public domain or hold a Creative Commons (CC), Pexels, or Pixabay license that allowed us to freely build upon, enhance, or reuse the original work. Based on their cultural background and experience, two audiology students reviewed all selected digital materials to verify that they were appropriate for the reinforcer set and for children. The end result of this process was that materials were generated for a total of 45 reinforcer sets (representing 43 unique themes). Table 1 provides the theme, digital material type, and the number of unique photos or videos for each reinforcer set.

**Format of Materials in Reinforcer Sets**

All photo and video files were edited to be of standard properties. The minimum size of individual .jpg photo files was 1000X1000 or 1280X780 ppi. To be compatible with commercial systems, Microsoft Photos (version 2020.20090.1002.0) was used to convert individual photo files to 4-second or 10-second .mp4 files with a resolution of 1080 p. Videos were edited in Microsoft Photos and a 4-second or 10-second segment was selected that was judged to have a natural start and stop point. Individual videos were saved as an .mp4 file with resolution that ranged from 720 to 1080 p across videos. These individual files are stored by reinforcers set and can be downloaded from our library.

To facilitate the use of our materials in clinics or laboratories that do not have a commercial system, we provide slideshows for all reinforcer sets that can be used in a custom setup. Details on the custom setup can be found in the implementation section below. For each reinforcer set, a single slideshow (.pptx format) was created in Microsoft PowerPoint (version 16.0). All available photos or videos were compiled in the slideshow. A single, full-screen photo or video is provided per slide. Prior to each photo or video is a background slide that is solid black in color. To display the next photo or video, the slideshow must be manually advanced. To be consistent with commercial systems, the default display time for all reinforcer sets is 4 seconds, but this duration can be customized in PowerPoint. To assist the audiologist, the slideshow displays a running count for the number of photos or videos shown and an additional visual alert on the final 3 slides for the set. The slideshow will automatically restart when the last photo or video in the set is shown. Further details about this process and a blank template can be found in our online resource.

**Online Access to the Materials**

The library described here is being made freely available for access and download under a Creative Commons Attribution-NonCommercial 4.0 International License (CC BY-NC 4.0) at https://osf.io/bk6rc/ (Hemann et al., 2020). The library consists of 45 reinforcer sets and can be downloaded as either a Microsoft PowerPoint slideshow or as a folder of individual .mp4 files. The site also provides the associated metadata (e.g., citations for all photos or videos) and supplemental documentation for the material generation process and implementation strategies. A bulk export of the entire library can be performed to allow for rapid download.

**Clinical Implementation of the Materials**

Reinforcers can be implemented in commercially available VRA reinforcement systems or through a custom setup. In general, the same procedure used for installing custom videos should be followed here. Audiologists interested in integrating these materials in a commercial system can find support documentation on our online OSF resource for two commercial systems: Flex and Intelligent VRA systems. Audiologists using other systems are advised to contact their system’s manufacturer if they need support. For clinics that do not have a commercial system, audiologists can use a custom setup with widely available and inexpensive technology. This set-up is achieved by connecting a computer in the control room to a secondary monitor that is mounted on the wall in the booth. Requirements of the computer are (a) appropriate hard drive storage for the files, (b) software to run the .pptx files (i.e., Microsoft PowerPoint), and (c) the ability to display to a second monitor. Any appropriately sized monitor can be used, but the ability to play sound may be desirable for some of the video reinforcers. Slide advancement can be

The Journal of Early Hearing Detection and Intervention 2020: 6(1)
**Table 1**

*Description of the Individual Visual Reinforcers by Theme Category*

<table>
<thead>
<tr>
<th>Category</th>
<th>Theme of Reinforcer Set</th>
<th>Media Type</th>
<th>Number of Images</th>
</tr>
</thead>
<tbody>
<tr>
<td>Activities</td>
<td>Cleaning</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Cooking</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Shopping</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Sports</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td>Animals</td>
<td>African safari animals</td>
<td>Photo</td>
<td>33</td>
</tr>
<tr>
<td></td>
<td>Big cats (e.g., lions, tigers, cougars)</td>
<td>Photo</td>
<td>33</td>
</tr>
<tr>
<td></td>
<td>Birds</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Birds of prey</td>
<td>Photo</td>
<td>40</td>
</tr>
<tr>
<td></td>
<td>Bugs</td>
<td>Photo</td>
<td>34</td>
</tr>
<tr>
<td></td>
<td>Butterflies and moths</td>
<td>Photo</td>
<td>40</td>
</tr>
<tr>
<td></td>
<td>Cats (domestic)</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Dinosaurs</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Dogs</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Farm animals</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Forest animals</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Frogs and toads</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Reptiles</td>
<td>Photo</td>
<td>35</td>
</tr>
<tr>
<td></td>
<td>Sea creatures (e.g., fish, whales, dolphins, turtles)</td>
<td>Photo</td>
<td>31</td>
</tr>
<tr>
<td></td>
<td>Snakes</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Zoo animals</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td>People</td>
<td>Babies</td>
<td>Photo</td>
<td>37</td>
</tr>
<tr>
<td></td>
<td>Faces</td>
<td>Photo</td>
<td>40</td>
</tr>
<tr>
<td>Science and Letters</td>
<td>Alphabet (i.e., street topography)</td>
<td>Photo</td>
<td>26</td>
</tr>
<tr>
<td></td>
<td>Space</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Waterfalls</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Weather</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td>Sensory</td>
<td>Bubbles</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Clocks, timers, and counters</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Fans and windmills spinning</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Items moving or spinning (abstract)</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>People in motion</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Rainbows and other colorful images</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Reflections</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Rides at amusement parks</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Water spinning (e.g., toilets, drains)</td>
<td>Video</td>
<td>14</td>
</tr>
<tr>
<td>Transportation and Equipment</td>
<td>Airplanes</td>
<td>Photo</td>
<td>31</td>
</tr>
<tr>
<td></td>
<td>Boats</td>
<td>Photo</td>
<td>31</td>
</tr>
<tr>
<td></td>
<td>Emergency vehicles</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Farm equipment</td>
<td>Photo</td>
<td>26</td>
</tr>
<tr>
<td></td>
<td>General transportation vehicles</td>
<td>Video</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Race cars</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Semi-trucks and heavy construction equipment</td>
<td>Photo</td>
<td>32</td>
</tr>
<tr>
<td></td>
<td>Trains</td>
<td>Photo</td>
<td>31</td>
</tr>
</tbody>
</table>
implemented in many of the common behavioral methods
supplemental reinforcement in CPA to guard against
The visual reinforcers provided here can be used as
monitor or a tablet in the booth using the custom setup.
by having the child respond by pushing a large button and
paradigm can be easily implemented with our materials
for a correct response (e.g., Thompson et al., 1989). This
the signal is heard, then visual reinforcement is provided
of VROCA involves training a child to push a lever when
the reinforcement sets can be used in visually reinforced
children with restricted interests, it may
be particularly motivating to continue with the original
reinforcer sets contain sound. Audiologists may want to
deactivate sound for children who have inadequate access
and/or sensory sensitivities to auditory input. Third, for
children with restricted interests, it may be desirable to
reduce the number of photos or videos in a reinforcer set
to increase the alignment of the available images with the
child’s circumscribed interest. For some children, it may be
preferred to only use the photos or videos that correspond
to their circumscribed interest. However, for other children,
especially those with high cognitive abilities, it may be
particularly motivating to continue with the original
reinforcer set and to instruct them to look for the slides in
that set that correspond to their circumscribed interest.
One final feature of these materials is that they can be
used for behavioral methods other than VRA. Specifically,
the reinforcement sets can be used in visually reinforced
operant conditioning audiometry (VROCA) or conditioned
play audiometry (CPA). The traditional implementation
of VROCA involves training a child to push a lever when
the signal is heard, then visual reinforcement is provided
for a correct response (e.g., Thompson et al., 1989). This
paradigm can be easily implemented with our materials
by having the child respond by pushing a large button and
the visual reinforcers can be displayed on the secondary
monitor or a tablet in the booth using the custom setup.
The visual reinforcers provided here can be used as
supplemental reinforcement in CPA to guard against
habitation (e.g., Bonino et al., 2019; Primus & Thompson,
1985). Thus, the materials provided here can be
implemented in many of the common behavioral methods
for measuring hearing in children.

Summary
A large library of video reinforcer sets is available for
clinical and research use. Based on the design of these
materials we expect that they will facilitate behavioral
hearing evaluations of children with developmental
disabilities, including children with restricted interests.
Support for this idea comes from two lines of research
from children with ASD. First, because children with
ASD often struggle with in-person, social interactions,
treatments that use videos have been shown to be
effective (e.g., McCoy & Hermansen, 2007; Wang et al.,
2011). For example, children with ASD have longer visual
attention to a puppet show presented as a video compared
to in person (Cardon & Azuma, 2012). Second, improved
outcomes—social interaction and behaviors of joint
attention—are observed if the intervention uses objects
or games that are related to the child’s circumscribed
interests (e.g., Baker et al., 1998; Boyd et al., 2007).
Moreover, children with ASD look longer and visually
explore an object in a more detail-oriented manner if it
is related to their circumscribed interests compared to
an object that is not (e.g., Sasson et al., 2008; Thorup et
al., 2017). For VRA, maximizing the child’s looking time
at the reinforcer may facilitate the audiologist’s ability to
judge the child’s response. Plus, if the child is motivated
and engaged by the reinforcer, it would be expected that
training would be faster, the risk of habituation would
be reduced, and on-task behavior would be improved.
Data from Chebli and Lanovaz (2016) is consistent with
this idea: children with ASD sat in their chair longer if
viewing their more preferred videos compared to their less
preferred videos. For these reasons, selecting a reinforcer
set that is related to a child’s circumscribed interests (like
the ones presented in our library) is expected to result in
an increased collection of behavioral data. Improving the
number of thresholds obtained per encounter is expected
to reduce the number of visits required to determine
hearing status in children with developmental disabilities.
In turn, reducing the number of visits has the potential to
lower medical expenses, reduce family stress, and provide
earlier access to intervention. For children with hearing
aids, obtaining a complete audiogram means better
fitting quality of their devices, which is a key predictor
of language outcomes. Another possible benefit of our
library of reinforcer sets is that it may facilitate behavioral
testing of 18- to 36-month-old children that are typically
developing: an age group that is notoriously difficult to
test with current methods. The library shared here has the
potential to improve clinical care, but further research is
needed to verify the efficacy of our reinforcer sets and to
evaluate our recommended implementation procedures
with children with developmental disabilities in a clinical
setting.
References


Canadian Families’ Decisions of Communication Options* for Children Who are Deaf or Hard of Hearing: An Initial Exploration

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Abstract

Communication is an essential aspect of human interaction and helps connect us to the people around us. The majority of children who are deaf or hard of hearing are born to hearing parents who are likely unfamiliar with hearing loss. These parents are then asked to make critical decisions about communication options for their children. It can be a challenging process, but one that needs to be done quickly to capture the critical language development period. Little research has explored the factors associated with parents’ decisions about communication options for their children who are deaf or hard of hearing and no studies have been done specifically with Canadian parents. This exploratory survey design study examined the factors which influence Canadian parents’ decisions relative to communication options for their children who are deaf or hard of hearing. Results indicate that parents’ personal judgement and a desire for their child to be able to communicate with their family and be happy in their own unique lives were driving forces behind the decisions that were made. Confirming research conducted in other countries, Canadian parents use a combination of their own judgement, professionals’ opinions, the needs of their child, and internal values to make communication option decisions. Implications of these results are discussed as they pertain to parent-professional partnerships and family-centered services.

Keywords: Canada, Families, Communication, Deaf, Survey

Acronyms: ASL = American Sign Language; CIHTF = Canadian Infant Hearing Task Force DHH = deaf or hard of hearing; EHDI = Early Hearing Detection and Intervention; IDEIA = Individuals with Disabilities Education Improvement Act; LSQ = la Langue des Signes Quebecoise

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*The term communication options is used in this article in place of communication mode/modality and is inclusive of listening, spoken languages, and signed languages.

“Well, the doctor told us we shouldn’t sign and to send him to the program in (city). Back then parents just did what the professionals thought best and we didn’t question it.” Parent statement regarding her deaf son born in 1980 (Pedersen, personal communication, December 14, 2019).

When a child is born with hearing loss, the need to provide early and appropriate intervention to avoid language deprivation and its consequences is urgent (Cole & Flexer, 2020; Yoshinaga-Itano et al., 1998). It is vital for families to make communication decisions as soon as possible because “effective communication supports cognitive development as well as social development, including the ability to develop positive relationships with others” (Decker et al., 2012, p. 326). The decisions families must make regarding communication options for their children who are deaf or hard of hearing (DHH) will significantly impact their children and ultimately who and how others will communicate with them (Kluwin & Gaustad, 1991).

However, these important and urgent decisions can be difficult. More than 90% of children who are DHH are born to parents with typical hearing; the family may have very little or no previous experience with hearing loss. Moreover, strong emotions and differences of opinion related to the use of spoken languages and signed language, despite the lack of empirical evidence proving a superior method (Gardiner-Walsh & Lenihan, 2019), are longstanding and add to the complexity of communication decisions for parents. Upon diagnosis, the family will usually meet with a professional who will explain the procedures and options available to the family. Professionals are defined as social workers, intake service counselors, medical personnel (e.g., audiologist and ear nose and throat physician), and educational personnel (e.g., teacher of the deaf and speech language pathologist; Crowe et al., 2014b). Eleweke and Rodda (2000) found that: 
The parents were strongly influenced by the information they received, especially in the period immediately after the hearing loss was diagnosed. This was because the information given to the parents might be either balanced (with detailed information provided on all available options) or not balanced (with only limited information provided, and with the expectations that the parents would follow it without consideration of other options. (p. 377)

Clearly parents rely on information shared with them by professionals; however, these professionals may not share information in an unbiased manner and may not be fully aware of all the options available, especially if a team approach is not in place (Eleweke et al., 2008; Crowe et al., 2014a). It is critical that professionals in both medical and educational fields understand the importance of factors that influence families’ decision making to support these family decisions and to better deliver family-centered support services.

**Communication Options in DHH Education History**

In the most basic of terms, communication options for people who are DHH can be separated into oral/spoken languages (used by the hearing population in that area) and visual/signed/manual languages. These origins are traced back to France and Spain for signed languages and Germany and Great Britain for oral languages. From its inception, the field has been shaped by polarizing views about these two approaches to communication. The first school for the deaf in North America began in 1817 in Connecticut and used sign language. By 1867, schools for the deaf that employed oral methods were established. Tensions between manualists like Edward Miner Gallaudet and oralists like Alexander Graham Bell continued to build. A landmark event known as the Milan Conference took place in 1880 in Milan, Italy during which sign language was outlawed in the education of the deaf. Consequently, during the first half of the 20th century, it was most common for children who were DHH to be educated primarily using oral methods—with varying degrees of success. In the United States, passing of PL 94-142 in 1975 and its reauthorizations, most recently the Individuals with Disabilities Education Improvement Act (IDEIA) of 2004, began a shift in segregated education for children with disabilities, including those who were DHH. A key tenant of IDEIA is free, appropriate public education in the least restrictive environment. Subsequent federal guidance on IDEIA for children who are DHH does not specify a communication modality that is most appropriate nor least restrictive and, despite the strong opinions in the field, research has not proven a superior method (Gardiner-Walsh & Lenihan, 2019). Although this is most likely due to the numerous individual variables that contribute to communication success for each child, this ambiguity can result in additional stress for parents and families about how and what to choose.

**Variations in Communication Options**

If communication options are conceptualized as a continuum, with oral methods at one end and signed methods at the other, there would be a number of sub-methods and variations that can be used in combination and are ever evolving. In general, current terminology describes the main communication options beginning with listening and spoken language (LSL) and ending with American Sign Language/Bi-Lingual Bicultural. Some common terms can be summarized as follows¹ (Anderson, 2011; Hands & Voices, 2020):

- **Auditory Verbal**
  
  Listening and Spoken language is generally how babies without hearing loss learn language.

- **Auditory Oral**
  
  Language can be spoken and heard. It can also be visual. When we watch someone talking we are getting some clues about what they are saying, even if it is noisy and we can’t hear them well. This is called lipreading or speechreading. But not all speech sounds can be seen on the face so speechreading doesn’t allow a child to fully catch language. Listening, talking, speechreading, using facial expressions, and gestures are all considered auditory oral communication approaches.

- **Cued Speech**
  
  It is also possible to make spoken language into a visual form through Cued Speech, which provides hand shapes for the speech sound combinations.

- **Simultaneous Communication**
  
  This involves people signing words or concepts at the same time as they are talking. It may also be called SimCom or Manually Coded English (MCE).

- **Total Communication**
  
  This refers to a philosophy of educating children with hearing loss that incorporates all means of communication: formal signs, natural gestures, fingerspelling, body language, listening, lipreading, and speech.

- **American Sign Language (ASL)**
  
  ASL is a true language. It has a sign for every language concept. Because it is a different language than English, the order of the concepts is not the same as English word order, so you can’t talk and use ASL at the same time. In Canada there are two recognized spoken languages, English and French, and two recognized sign languages which are American Sign Language (ASL) and la Langue des Signes Quebecoise (LSQ; Canadian Association of the Deaf [CAD], 2015).

**Early Hearing Detection and Intervention (EHDI)**

The field of education of children who are DHH has experienced unprecedented change during the last two decades. Many helpful infographics are available and provide more detailed descriptions of the aspects of these various terms (e.g., [https://sound-advice.ie/wp-content/uploads/2014/07/sound-advice-comm-options-infographic.pdf](https://sound-advice.ie/wp-content/uploads/2014/07/sound-advice-comm-options-infographic.pdf)).
decades. This is primarily due to technological advances of universal newborn hearing screening and sophisticated digital hearing aids and implantable devices such as cochlear implants (Strickland et al., 2011). Seminal research in the field (Yoshinaga-Itano et al., 1998) found that the language and communication outcomes of children who are DHH that received EHDI services by six months of age were far superior to those of children receiving services later in childhood; these gains held true across a number of variables including socio-economic status, degree of hearing loss, and presence of additional disabilities. Consequently, current best practice in EHDI world-wide dictates a 1-3-6 rule meaning screening should occur within one month of birth, a diagnosis confirmed by 3 months of age, and intervention implemented by 6 months of age (National Center for Hearing Assessment and Management, 2020). In Canada, through a joint effort of Speech-Language & Audiology Canada and the Canadian Academy of Audiology, a group of national experts formed the Canadian Infant Hearing Task Force (CIHTF) to monitor and oversee EHDI efforts. Consistent with the International Consensus Statement on Best Practices in Family-Centered Early Intervention for Children Who Are Deaf or Hard of Hearing (Moeller et al., 2013), the CIHTF cites five core goals for Canadian EHDI programs:

1. Universal hearing screening of all newborns
2. Identification of babies with permanent hearing loss
3. Intervention services which include support for technology and communication development
4. Family support
5. Monitoring and evaluation of the program

The smaller national population of Canada spread out over a much larger geographical land mass poses unique challenges to achieving the goals of EHDI. The CIHTF issued a Canadian EHDI report card in 2019 and ranked achievement as insufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 provinces and territories receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13 receiving a score of sufficient overall. Individual provinces and territories varied in their ranking with only six of the 13

The Journal of Early Hearing Detection and Intervention 2020: 6(1)

Previous Research on Parental Decisions on Communication Options

Early research examining this topic conducted by Kluwin and Gaustad (1991) found that “the mother appears to be the primary decision-maker for the family’s mode of communication. Influenced by her own educational sophistication, she will base her decision on the child’s degree of impairment and the nature of available services” (p. 33). More recently, the idea that family culture plays a role in communication decision making is also present in the research. Borum (2012) recommends that professionals working with families who have children who are DHH need to be more understanding of cultural perspective and ideas when providing resources and supports to families. Guiberson (2013) and Matthijs et al. (2017) also support these findings by indicating that family involvement, family beliefs and values, and culture are important factors and influences in the decision-making process for families who may be bi- or multi-lingual. In such cases, adding another language such as ASL may be more natural than for monolingual families.

A recent systematic literature review on the topic of parental decision making and children who are DHH (Porter et al., 2018), found only 37 peer reviewed studies. The two most common focus areas related to parental decision making were implantable devices and communication modality. Porter et al.’s (2018) study revealed only nine of the 37 studies pertained to communication modality and none of them took place with Canadian parents. Table 1 summarizes the characteristics of these nine studies.
The timeline of these studies is consistent with important advances in the field mentioned earlier, including newborn hearing screening and advances in hearing technology. Prior to these events, the average age of identification of profound hearing loss in children was 12 months, and 18–24 months for milder degrees of hearing loss (Norman & Heffernan, 2017). Often communication option decisions were dictated by the degree of hearing loss, medical models of hearing loss, and limitations of hearing technology.

Table 1

<table>
<thead>
<tr>
<th>Author</th>
<th>Country</th>
<th>Sample</th>
<th>Method</th>
</tr>
</thead>
<tbody>
<tr>
<td>Borum (2012)</td>
<td>US</td>
<td>14 parents</td>
<td>Qualitative</td>
</tr>
<tr>
<td>Bruin and Nevoy (2014)</td>
<td>Norway</td>
<td>27 parents</td>
<td>Qualitative</td>
</tr>
<tr>
<td>Crowe et al. (2014a)</td>
<td>Australia</td>
<td>177 parents</td>
<td>Quantitative</td>
</tr>
<tr>
<td>Crowe et al. (2014b)</td>
<td>Australia</td>
<td>177 parents</td>
<td>Qualitative descriptive</td>
</tr>
<tr>
<td>Decker et al. (2012)</td>
<td>US</td>
<td>36 parents</td>
<td>Quantitative descriptive</td>
</tr>
<tr>
<td>Eleweke and Rodda (2000)</td>
<td>UK</td>
<td>2 families</td>
<td>Qualitative</td>
</tr>
<tr>
<td>Li et al. (2003)</td>
<td>US</td>
<td>83 parents</td>
<td>Quantitative</td>
</tr>
<tr>
<td>Matthijs et al. (2017)</td>
<td>Belgium</td>
<td>5 parents</td>
<td>Qualitative</td>
</tr>
<tr>
<td>Wheeler et al. (2009)</td>
<td>UK</td>
<td>12 parents</td>
<td>Qualitative</td>
</tr>
</tbody>
</table>

The nine studies identified by Porter et al. (2018) have several common features relative to the findings on parental decisions of communication options. The exploratory study conducted by Eleweke and Rodda (2000) identified themes of the influence of information that was provided to parents and the attitudes of the professionals providing the information. They further found that parents’ expectations about the child’s hearing technology and the availability of resources were factors parents considered. The contribution of parental values was identified in several studies. Parental views about what they wanted the future to look like for their child who is DHH were associated with their choice of communication modality. Parents whose values most closely aligned with the medical model of hearing loss tended to select communication options that included spoken language, while parents who valued a socio-cultural model of hearing loss tended to support communication options that included sign language (Borum, 2012; Decker et al., 2012; Li et al., 2003). This association was also evident in relation to the child’s hearing device. Parents who chose cochlear implants for their child also selected communication options that included spoken language and more often, exclusively spoken language (Wheeler et al., 2009). The need for parents to receive unbiased information from a collaborative team was very evident (Decker et al., 2012; Eleweke & Rodda, 2000; Li et al., 2003; Matthijs et al., 2017).

The Current Study

Some research has been done regarding how families make communication decisions about their children who are DHH, but none of them have been conducted with Canadian parents; in fact, little research is available relative to families of children who are DHH in Canada. One qualitative study conducted by Fitzpatrick et al. (2008) explored the needs of Canadian parents after receiving their child’s hearing loss diagnosis. Service coordination and lack of access to information was cited by parents as problematic aspects of early intervention. Fitzpatrick et al. (2008) called for further research into understanding the needs and actions of Canadian parents of children with hearing loss in a variety of settings and across variables to better support healthy family outcomes. Adding support to Fitzpatrick et al.’s (2008) call, the 2019 Report Card on Canadian EHDI Programs issued by the CIHTF graded Canada’s status as insufficient. Beyond universal newborn screening and identification, the CIHTF lists support for communication development and family support as two of its five core goals (CIHTF, 2019). Further, the International Consensus Statement on Best Practices in Family-Centered Early Intervention for Children Who Are Deaf or Hard of Hearing (Moeller et al., 2013) cites (a) informed choice and decision making and (b) parent-professional partnerships as two of its 10 principles. One thing is clear from the available literature—professionals must seek to thoroughly understand factors in parents’ decision making for communication to offer truly family-centered services.

Parents of children who are DHH must make many critical decisions regarding communication for their child that parents of hearing children do not encounter. These critical decisions are complex, controversial, and need to be made in a timely manner for the child to receive maximum benefit of EHDI. Professionals are charged with providing evidence-based and non-biased information to empower parents to make educated decisions for their children who are DHH; however, scarce information is available regarding how Canadian parents make these decisions, what factors influence them, and what types and sources of information are most effective. The current study aims to explore various factors and influences that contribute to Canadian parents’ decisions regarding communication with their child who is DHH. Using a survey design, the current study seeks to answer the research question, “What are the factors associated with the decision-making process of Canadian parents regarding communication option(s) for their children who are DHH?”

Method

Participants

The study sample was drawn from the population of Canadian parents of children who are DHH. Twenty-one parents who had a child who is DHH completed the survey. Ten of the families resided in Manitoba, two in...
Institutional Review Board for Human Subjects (Protocol and Audiology Canada. Following approval from the provincial schools for the Deaf and Hard of Hearing, the Canadian Hard of Hearing Association, including the Alexander Graham Bell Association, searches of professional organizations and their affiliates children with hearing loss were identified through internet Canadian organizations that support families who have communication technologies, and family demographics. The second part of the survey asked parents to identify the importance or significance that various factors and influences played on the communication options for their child. The final section contained Likert items regarding the degree to which parents perceive the importance of statements related to their child’s future. Per Decker et al. (2012) and Crowe et al. (2014a, 2014b), these questions were designed to gather information regarding parental values and hopes for the future of their child, which may also influence their communication decisions. Finally, the survey had one open ended item allowing parents to comment on any aspect of the study topic if they wished. The survey instrument is contained in Appendix A.

Data Collection and Analysis

Canadian organizations that support families who have children with hearing loss were identified through internet searches of professional organizations and their affiliates including the Alexander Graham Bell Association, Canadian Hearing Services, the Hearing Foundation of Canada, the Canadian Hard of Hearing Association, provincial schools for the Deaf and Hard of Hearing, the Canadian Association of the Deaf, and Speech-Language and Audiology Canada. Following approval from the Institutional Review Board for Human Subjects (Protocol # 2017), an invitation containing informed consent, a brief explanation of the study, and a link to the survey was posted to social media pages and/or emailed to Canadian organizations that serve children who are DHH and their families. A snowball procedure was used as the survey requested that the invitation be forwarded to that recipient’s contacts, thus increasing the number of potential parents to participate in the study. The survey was available for a total of four weeks with a second round of postings and emails done after the first three weeks. Once the survey was closed, the raw data was exported from Microsoft Forms® into an Excel spreadsheet. Descriptive statistics in the form of percentages, tables and pie charts were used to represent the data and draw conclusions. Participant responses to the open-ended survey question were examined individually to determine if or how they aligned with each participant’s quantitative responses as well as with the sample as a whole.

Results

DHH Children Demographics

Current Age and Age at Identification

Parents were asked both the current age of their child and the age at which their hearing loss was identified. Current ages of their child who was DHH indicate 16 were school age with seven children between six and 10 years old and six children between 11 and 18 years old. Three children were preschool age, between three and five years old, and one child was less than two years old. Four parents reported they had adult children who are DHH. The age at which their child’s hearing loss was identified varied, with four children identified prior to six months old, seven children identified between six and 12 months, three between 13 and 24 months, three children between 25 and 36 months, and three children were identified between the ages of four and five years old. One child’s hearing loss was identified at older than five years of age.

Hearing Loss Levels and Technology

Nineteen participants indicated that their child had a bilateral loss while two had unilateral losses. Standard audiological hearing loss level categories were offered as a forced choice question. The majority (n = 13) of children had profound losses. Two had severe, five had moderate-severe losses, and one had a moderate loss. Parents were asked about their child’s assistive listening technology. Results indicated eight children used hearing aids, eight used cochlear implants, one had a bone anchored hearing aid, and three used an FM system. The remaining four parents indicated their children used another listening technology device but did not specify. Parents could select more than one choice, so it appears some children used more than one assistive listening device.

Early Intervention (EI)

Participants were asked to rate the quality of their EI services and nine thought their services were excellent and seven reported their services were adequate. Four parents believed their EI services were unsatisfactory. One parent indicated they did not receive EI services. The
majority (n = 12) of parents indicated that they were not at all familiar with hearing loss prior to their child being identified. Seven parents reported they were a little familiar and two parents were very familiar with hearing loss prior to their child’s diagnosis.

Sources of Information
Parents were asked from what sources they sought information when they first learned that their child had hearing loss. Table 2 displays the percentage of parents seeking information from each source. The primary sources of information used by parents were medical professionals and audiologists/speech-pathologists. The next most often used sources of information by parents were the internet, books/magazines, and community agency professionals.

Factors Influencing Parents’ Communication Decisions
Parents in the study reported that 13 of their children currently used listening and spoken language, six used ASL, and two used total communication. A list of potential influences which contributed to the decision made about their child’s communication was presented to participants. They were asked to rate each factor on a four-point Likert scale from having no influence to having a lot of influence. Figure 1 illustrates the data on these items.

Of the 12 factors, the top four in descending order that parents ranked as having a lot of influence in their decision about communication mode were the parent’s own judgement, the ability to communicate within the family’s home community, the child’s ability to communicate like the rest of the family, and their spouse’s or child’s other parent’s opinion. In contrast, the factors rated as having no influence for most parents on their communication modality decision were the cost of the services, the recommendation of a family member or friend, and their personal knowledge or experience with hearing loss. Sixty-two percent of parents indicated information found on the internet as having little or no influence on their decision of communication modality choice.

Parental Values Related to Communication
Participants were asked to rank statements reflective of their values about their child’s communication on a four-point Likert scale from very important to not important.

Table 2
Parental Sources of Information

<table>
<thead>
<tr>
<th>Information Source</th>
<th>N</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Medical professionals</td>
<td>17</td>
<td>81</td>
</tr>
<tr>
<td>Audiologist/speech pathologist</td>
<td>16</td>
<td>76</td>
</tr>
<tr>
<td>Community agency professionals</td>
<td>10</td>
<td>48</td>
</tr>
<tr>
<td>Books/magazines</td>
<td>9</td>
<td>43</td>
</tr>
<tr>
<td>The internet</td>
<td>9</td>
<td>43</td>
</tr>
<tr>
<td>People I know who are DHH</td>
<td>6</td>
<td>29</td>
</tr>
<tr>
<td>School/education program</td>
<td>4</td>
<td>19</td>
</tr>
<tr>
<td>Teachers/school personnel</td>
<td>4</td>
<td>19</td>
</tr>
<tr>
<td>Family members/close friends</td>
<td>3</td>
<td>14</td>
</tr>
<tr>
<td>Other parents I know</td>
<td>2</td>
<td>9</td>
</tr>
<tr>
<td>I don’t know/don’t remember</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>I didn’t seek additional information</td>
<td>1</td>
<td>5</td>
</tr>
</tbody>
</table>

Note. DHH = deaf or hard of hearing.
Figure 2 summarizes the parent responses to these value statements. One hundred percent of the parents indicated that it was very important to them that their child lived the life that was right for their child and were less concerned with their child having a normal life. Ninety-five percent of parents indicated that the parent-child relationship was very important to them as was their child’s ability to communicate as early as possible in their life. In a similar manner, 95% of the parents said it was more important for their child to have opportunities and experiences that met their child’s unique needs than for their child to have the same opportunities and experiences as other children. Parents further indicated it was more important to them that their child fit in with their peers who were also DHH than with their hearing peers.

Parent Comments
At the end of the survey parents were given the opportunity to provide comment on any aspect of the decision-making process for the communication modality for their child who is DHH. Sixteen of the 21 participants provided additional comments. The number of comments did not allow for thematic analysis; they are analyzed descriptively below. The verbatim comments are contained in Appendix B. Four of the 16 comments pertained to challenges faced by rural families such as access to the Deaf community and quality intervention. For example, one parent said,


We didn’t actually have a choice. We were told that the school system we were in only used SEE [Signed Exact English]. This choice has been a huge disservice to my child, I believe that if a child is learning SEE for reading and writing they should also be taught ASL so they can communicate with other DHH persons as well. As it stands today my child doesn’t fit in in the hearing world of his peers nor the peers in the Deaf community.

Another five comments expressed concerns and frustrations from parents on the real or perceived bias they felt from professionals. For example, one commented:

It was a very difficult decision for us and the fact that professionals were implying we had to choose one or the other made it harder and took us longer to decide. I wish we had support right from the start with choosing both ASL and spoken English via cochlear implant. With our second daughter we decided to use ASL right away which enabled us to communicate with her from the age of 6 months.

Discussion
Results of the current study were similar in many ways to the results found in previous studies from Decker et al. (2012) and Crowe et al. (2014a). Firstly, Canadian parents’
top sources of information after their child’s diagnosis came from medical, speech-language and hearing, and other professionals. Parents in this study also sought information from the internet and books, but to a lesser degree than in previous studies by Decker et al. (2012) and Crowe et al. (2014a). Canadian parents received information primarily from medical and speech-language and hearing professionals; however, this did not appear to be the primary influence on parental decision making. Yet, parents did note that professional bias was still present in their experiences as one parent remarked, “Non-bias in both (or all) directions should be emphasized in communication choices.” Parents may certainly weigh advice from professionals and incorporate it into their decisions, parents in this study indicated their own judgement and their values relative to communication for their child appeared to be most influential. This does indicate a shift from earlier studies (Kluwin & Gaustad, 1991; Eleweke & Rodda, 2000), in which parents tended to follow professional recommendations. This may mean that the professionals involved in supporting families with DHH children have evolved and adopted more family-centered approaches. In the context of this study, separating the direct influence from a source of information from the indirect influence that source may have on parent’s decisions is not possible to determine. It is possible that parents may have perceived that a decision was based on their own judgement, but information obtained from other sources may have influenced this judgment. Similar results were found by Decker et al. (2012) who also suggested that parents may internalize the opinions of professionals, which underlines the importance of providing unbiased information to families. Additionally, parent’s judgments may also be reflective of intuition, or a feeling that the selected communication modality is a good fit for their child and family. Further exploration of the role of intuition and parent self-efficacy regarding communication options could add to the knowledge base about parent decision making.

The sample size used in this study did not allow for analysis of the relationship of parental values directly to the specific communication modality chosen as done in previous studies (Decker et al., 2012; Crowe et al., 2014a). However, insight into Canadian parents’ values about communication for their children who are DHH was gained. Parents primarily valued their relationship with their child and ensuring that the individual and unique needs of their child were met rather than their child being normal. Further, parents in this study placed a greater value on their child fitting in with their peers who are DHH than peers with typical hearing. This may be reflective of greater appreciation of diversity and acceptance of hearing loss as a difference rather than a disability. This possibility is also strengthened by the fact that 29% of parents in this study indicated that people who are DHH were sources of information they sought regarding communication options for their children. EHDI efforts have recently focused on bringing the voice of individuals who are DHH to the EHDI discussion and ensuring that perspectives of these vital stakeholders are available to parents of children who are DHH (Benedict et al., 2015). This aspect of parental decision-making warrants further examination.

Finally, although parents in the current study did not identify access to services as a top influencing factor, 25% of the comments made by parents did pertain to frustrations with poor or unavailable access to support their communication choice. The field should continue to address innovative methods for increasing access to a range of services for families that include children who are DHH, particularly for families in rural areas as recommended by Sibon-Macarro et al., 2014.

Limitations and Future Directions

The current study was exploratory as there were no previous studies found to have been conducted with Canadian parents. Although generalization is limited due to the small sample size, these results can form the basis for future study using a larger sample. In Canada there is not federal legislation mandating universal newborn hearing screening nor EHDI services; consequently, the experiences of parents receiving a diagnosis of hearing loss may vary widely from province to province and from residence to residence. Canada’s large geographic area also poses challenges to service delivery, particularly in rural and remote locations. A larger sample size could allow for a more rigorous statistical analysis of the relationship of parental values to the particular communication option(s) they chose for their child. Additionally, more in-depth mixed-methods research designs such as those conducted by Crowe et al. (2014a, 2014b) could yield a deeper understanding of parental decision making and recommendations for support directly from parents. Also, future studies on this topic should give extra effort to recruiting diverse participants to ensure results are representative of the multicultural nature of Canadian families. Kluwin and Gaustad (1991) found that mothers were the primary decision maker in families with children who are DHH. All parents in the current study were their child’s mother; yet, almost half of them indicated their spouse or child’s father’s opinion was very important in their decision. Although not specifically explored in previous research relative to this topic, the literature on families of children who are DHH is still heavily weighted to mothers’ perspectives. Given the increasingly active roles that contemporary fathers have in their child’s life, further work needs to be done to gather perceptions of fathers regarding their involvement in the decision-making process (Pedersen & Othoff, 2019). Finally, although one parent commented that parent-to-parent support was important to her family, the influence of parent-to-parent support was not specifically addressed in the current study. A growing body of evidence indicates that parental support from other parents who have similar experiences is a powerful tool for families with children who are DHH (Friedman Narr & Kemmery, 2015; JCIH, 2013; Moeller et al., 2013; Ward et al., 2019). Future studies should include this component.

Conclusion

The ultimate goal that all parents expressed was for their child to be happy and successful in whatever path they choose in life. Parents wanted to select a communication option(s) that was right for their child. The current study supports the importance of professionals who offer unbiased
and up-to-date information to the families they serve. Professionals working in their specific areas also need to be aware of the geographical area that they are serving and know what sources of support and resources are available to parents so that they can direct parents on where to go and also be open to changing their decision as time goes on. The national parent-support organization for families with children who are DHH is Hands and Voices, whose motto is “What works for your child is what makes the choice right.”

Co-founder LeeAnn Seaver (2004) gives professionals this advice for supporting families through the communication modality decision-making process:

> When we have shifted from appropriately sharing the benefit of our experience and knowledge into intentionally manipulating a family, we’ve crossed the line into bias. Ultimately, we’ll experience greater trust in the relationship with the family when we approach them with an open mind. Encouraging their independent thought serves the greater good: increased sensitivity and awareness of this child-driven process, deeper investment and ownership of their choices, and more effective advocacy for their child. (p. 4)

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References


Appendix A
Survey Items

1. Which province/territory do you live in? (Forced-choice list)
2. Person completing this survey: a) child’s mother; b) child’s father
3. What is the population category where you live? (Forced-choice list)
4. How do you describe the primary ethnicity of your family? (Forced-choice list including other and prefer not to answer)
5. What is your family’s annual income? (Forced-choice list including prefer not to answer)
6. What languages are used in the home? a) spoken English; b) spoken French; c) American Sign Language (ASL), d) Langue des signes du Québec (LSQ)
7. What is the highest level of schooling in your household? (Forced-choice list)
8. What is the current age of your child who is deaf or hard of hearing (DHH)? (Forced-choice list)
9. At what age was your child’s hearing loss diagnosed? (Forced-choice list)
10. My child’s hearing loss is: a) unilateral (in one ear only); b) bilateral (both ears)
11. My child’s hearing loss can be described as: a) Slight/Mild (15-40 dB); b) Moderate (41-55 dB); c) Moderately-Severe (56-70 dB); d) Severe (71-90 dB); e) Profound (90+ dB)
12. What is your child’s primary communication mode? a) Listening & Spoken Language; b) American Sign Language (ASL); c) Langue des signes du Québec (LSQ); d) Total Communication (mix of talking, signing, lipreading etc.); e) Cued Speech; f) Other
13. What assistive listening technology does your child use? Check all that apply. a) hearing aids; b) cochlear implants; c) bone anchored device; d) FM/Remote microphone; e) other
14. Prior to becoming the parent of a child who is deaf or hard of hearing, my familiarity with hearing loss was: a) very familiar; b) somewhat familiar; c) a little familiar; d) not at all familiar
15. The early intervention services our family receives/d to support my child with hearing loss are/were: a) excellent; b) adequate; c) unsatisfactory; d) we did not receive early intervention services
16. When I first learned my child had a hearing loss, I sought information from (Check all that apply): a) Medical professionals; b) Community agency professionals or personnel; c) Family members/close friends; d) Other parents I know; e) Teachers/school personnel; f) A school/educational program for the Deaf; g) Audiologist/speech pathologist; h) People I know who are DHH or have a child who is DHH; i) The internet; j) Books or magazines; k) I didn’t seek additional information; l) I don’t know/don’t remember
17. The following factors influenced my decision about my child’s communication mode (Likert Scale: a lot of influence, some influence, a little influence, no influence): a) Recommendation of an audiologist; b) Recommendation of a family member or friend; c) Internet resources/information; d) My spouse’s/my child’s other parent’s opinion; e) My own judgement; f) Cost of the therapy/services; g) Availability of support close to home; h) Recommendation of an early intervention professional; i) Ability to communicate like the rest of the family; j) Ability to communicate within our home community; k) Personal knowledge and experience with people who are Deaf/Hard of Hearing; l) Ability to attend our local school
18. Please rate how important each of the following statements are for you (Likert Scale: very important, important, a little important, not important): a) When my child is of school age, it is most important that my child be able to fit in with his/her peers; b) When my child is of school age, it is most important that I have a good relationship with my child; c) It is important to me that my child lives a normal life, a life like everyone else; d) It is important to me that my child lives the kind of life that is right for him/her; e) It is important to me that my child has all of the opportunities and experiences that other children have; f) It is important to me that my child has opportunities that fit his/her own unique talents and limitations; g) The language that my child learns early in life should prepare him/her to more easily fit in with his/her peers when they are older; h) The language that my child learns early in life should help him/her and me communicate earlier in his/her life; i) When my child is of school age, it will be very important for him/her to fit in with his/her hearing peers and communicate effectively with those peers; j) When my child is of school age, it will be very important for him/her to fit in with his/her deaf or hard of hearing peers and communicate effectively with those peers.
19. Is there anything else you would like to say about the decision-making process of your family regarding communication choices for you child who is DHH? (Open comment box)
1. Right now he is with a great teacher who is knowledgeable of [deaf or hard of hearing] DHH and on how to work with my child.

2. Gave the best of both worlds with CIs [cochlear implants] and ASL [American Sign Language]. Then it’s her choice when she’s older, but she has all the tools, and perfect speech.

3. I think that my past personal experience was important. When I was in high school I was in the debate club and regularly travelled to our university to research in the libraries. One day a group of teens got on my bus; they were so animated! I watched, fascinated by their expressions, body language, and signing (I figured out that they were deaf and signing). I enrolled in a sign language class at the school for the deaf. Unfortunately, after the class ended it was summer break and I couldn’t take another class nor find any deaf people to practice with; I forgot everything by fall and was too disheartened to start all over again. I think having an ESL background matters too. Having English as my second language has made me fascinated in learning languages. I had taken Mandarin and Japanese in university before I had my daughter. I encourage her to pursue other languages too. She is interested in learning other sign languages and written forms of German and Mandarin.

4. I answered cost of services had no influence but not sure if I should have selected a lot of influence! Services in (my province) are free so cost of services was not a barrier to our choices.

5. Families facing this need to receive unbiased, neutral information right from the outset. This is not a tragedy, but a difference. Parents need options available to them that are easily accessible, free, and flexible. Parent-to-parent support is invaluable, and should be provided and encouraged automatically starting from day one, and continuing on through the school years, far beyond early intervention. Parents shouldn’t necessarily have to make choices; there shouldn’t be a divide. Non-bias in both (or all) directions should be emphasized in communication choices. Opportunities for connecting the children to others like them and mentors like them (not only Deaf, but also hard of hearing) should be provided to every family. Opportunities for continuing your education about your child’s hearing loss should be available as well. Hearing devices should be covered by our health care system. You shouldn’t be non-eligible for the disability tax credit because you wear cochlear implants and “can hear”. The decision we made around our communication choice for our child was not an easy one, and one which we continue to grapple with to this day, more than 10 years later. We are extremely proud of the hard work and outcomes that auditory oral language therapy has elicited for our child. We do recognize, however, that our child is and always will be deaf and hearing through a mechanical device using a damaged auditory system. This is something that we try never to forget and educate people in his life about. It is a gift, but it is far from perfect. We have seen now, as our child gets older, that he struggles with feelings of loneliness and isolation which we attribute to his feeling different in the “hearing world,” though puberty probably has something to do with it too. This is hard to bridge, but we are working through it with him. Over the years we have continued to give our son opportunities to learn sign language, but up to now, the programs for signing have seemed restrictive since he is a new signer. This has been discouraging for him. It’s like the opposite discrimination or bias occurs. I find this a tragedy. We use some basic sign and gestures at home when he is without implants. We participate in and have always participated in the hard of hearing community in our area so he maintains some ties to other oral deaf and hard of hearing kids. Upon identification, our audiologist did not persuade us to choose a listening and spoken language outcome, but she did almost immediately suggest that we should seek cochlear implantation for our child. The structure and proactive approach to auditory oral therapy was something that appealed to us right away. In retrospect, adding some visual aids would have benefited our son. We were also fortunate to be able to pay for additional private speech therapy and could afford my leaving work to be at home and work with our child all day every day on language learning and enrichment. It is probably the most important work I’ve done in my life, regardless of whether it was spoken or signed.

6. I think it is important to take into consideration how available support is in that person’s area. We live in a Rural community with no other deaf or hoh [hard of hearing] individuals. As well as no one to teach us or our child ASL. . . I had to try to teach myself to the best of my abilities in order to teach him.

7. We don’t have a Deaf community where we live. We wanted to give our daughter the best communication skills possible. We also want her to have independence. She is absolutely thriving.

8. Went through cochlear implant assessment and was not a fit. Decision accepted and continued with ASL.

9. The (province) deaf community is more than just a linguistic community. It is a social community which is extremely difficult to engage with when you are not deaf. They are kind and nice people but they are also insular. I found in teaching our son sign language as a child before he was verbal that the easiest tool was to use a phone app with signs - but these are not (PROVINCE) SIGNS and some signs he learned were ridiculed and I was pressured to use the (PROVINCE) sign resource - a duotang with illustrations. This simply does not cut it as a resource. I would have
been happy to continue longer with a bilingual approach with sign and spoken language but the community (despite kindness and great motivations I am sure) was not ultimately providing what we needed. My child soon preferred spoken language mostly out of a desire to be like his peers and not stand out, and as we were a verbal family at home, we allowed sign to essentially die out as a home language.

10. It was a very difficult decision for us and the fact that professionals were implying we had to choose one or the other made it harder and took us longer to decide. I wish we had support right from the start with choosing both ASL and spoken English via cochlear implant. With our second daughter we decided to use ASL right away which enabled us to communicate with her from the age of 6 months.

11. The only thing that matters is him being able to express himself and be happy.

12. We used ASL as well as cued speech initially to communicate. He is bilingual in both English and ASL. Due to distance away from families and medical issues with his grandparents we started English. Moved to ASL in school. Went to public school.

13. I was surprised and disappointed that the medical community still pushes oral communication above the use of ASL and spoken language. We try to use ASL at home and are in college programs to help support that. There was little support around the family learning ASL once we decided the oral communication was important to us too. Most ASL supports are in (large city) and make it difficult for us to attend.

14. The decision to pursue Cochlear Implants was greatly influenced by our ENT doctor’s recommendations.

15. I have 2 children ages 9 and almost 11.

16. We didn’t actually have a choice. We were told that the school system we were in only used SEE [Signed Exact English]. This choice has been a huge disservice to my child, I believe that if a child is learning SEE for reading and writing they should also be taught ASL so they can communicate with other DHH persons as well. As it stands today my child doesn’t fit in in the hearing world of his peers nor [with] the peers in the Deaf community.

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**Louisiana EHDInfo**

**Louisiana EHDInfo Program wins 2021 EHD Informational Program of the Year Award!**

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**The Louisiana Early Hearing Detection and Intervention (LA EHDI) Program**

**Communication is Key**

The Louisiana Early Hearing Detection and Intervention (LA EHDI) program supports coordinated systems of care that ensure families of babies and children who are deaf or hard-of-hearing (DH) receive appropriate and timely services. These services include hearing screening, diagnosis, early intervention (EI) and family to family support.

It is important to identify a baby’s hearing level as early as possible so families can figure out the best way to communicate with them. Hearing screening is the first step in finding out if a child is deaf or hard of hearing. Louisiana law mandates that all babies be screened prior to hospital discharge. EHDI follows the Joint Committee on Infant Hearing’s Position Statement and national benchmarks for hearing screening no later than 1 month of age, diagnosis no later than 3 months of age for infants who did not pass the screening, and enrollment in early intervention services no later than 6 months of age for those identified as DH. Enrollment in early intervention as soon as possible plays an important part in helping children who are deaf or hard of hearing reach their full potential.

**EHDInfo**

For more information on how to improve EHDI Websites visit [https://infanthearing.org/webguide/](https://infanthearing.org/webguide/)
Abstract

Objective: The purpose of this scoping review was to provide information about the research base related to psychosocial experiences of parents of young children who are deaf or hard of hearing (DHH) and use hearing devices. A scoping review identifies trends and gaps in available evidence. This information can be used to inform practice and identify areas in need of further research.

Design: A scoping review was conducted in June 2020 to identify English-language peer-reviewed journal articles published through May 31, 2020.

Study sample: Nine articles were found that investigated psychosocial factors of parents of children birth through five years who are DHH and use a hearing device.

Results: Four psychosocial areas were explored in the identified studies: stress \((n=5)\), self-efficacy \((n=2)\), depression \((n=1)\), and depression/psychological flexibility \((n=1)\). None of the studies investigated an intervention to address parent psychosocial factors interfering with treatment adherence.

Conclusions: There is a scarcity of research related to psychosocial barriers experienced by parents of young children who use hearing devices. Research is needed to identify effective interventions and to demonstrate the effect of addressing parent psychosocial barriers on spoken language outcomes for children. Providers have opportunities to use validated screening tools to assess for parent barriers and to individualize support for parents within the care plan for children identified with hearing loss.

Keywords: psychosocial, parents, pediatric, hearing device

Hearing loss is a common condition affecting approximately 34 million children worldwide (World Health Organization, 2020). For many children, their hearing loss is identified during the first few months of life through objective newborn hearing screening measures. This early identification provides children with the opportunity to receive early intervention critical for supporting developmental milestones and school readiness (Joint Committee on Infant Hearing, 2013). Consistent use of well-functioning hearing devices is a foundational aspect of the intervention process for spoken language development—better language outcomes have been found for children who wear their hearing aids 10 or more hours per day (Tomblin et al., 2015). Parents have a central role in intervention; however, it can be difficult for parents to be consistent in integrating evidence-based intervention routines (e.g., hearing aid listening checks) in their daily lives (Muñoz et al., 2019) and this can interfere with meeting intervention goals.

It is understandable that parents encounter challenges with daily intervention routines, such as having their child wear the devices consistently, checking function of the device regularly, and incorporating language strategies to provide a language-rich environment. For many parents, their child’s diagnosis is their first experience with hearing loss, as most parents have normal hearing themselves (Mitchell & Karchmer, 2004). Hearing loss degree varies among children (White, 2018), and parents may struggle with perceptions of what it means for their child to have a hearing loss and use a hearing device (Ambrose et al., 2020). Parents are faced with learning new information, new systems of care, and new skills.
that can feel overwhelming to navigate. Every family is unique and support needs may differ based on various factors including their beliefs, how they learn, their support network, and their psychosocial experiences. Family dynamics and parental coping strategies can influence engagement in the intervention process, and it is reasonable to expect support be provided differently based on individual parents’ strengths and needs.

Partnering with parents requires professionals to comprehensively consider factors, including parent thoughts, feelings, and behaviors, that are influencing parent engagement. Various psychosocial factors (e.g., depression) may influence how effectively parents cope with the addition of intervention demands in their daily lives as a result of the identification of their child’s hearing loss. This can signal the need for different or additional support. Considering and incorporating parent needs within the intervention process can provide protective factors to support progress toward intervention goals. For example, Cross et al. (2018), in a systematic review, found that person-centered care to promote caregiver well-being within the care plan is needed when working with dementia patients. In another systematic review, Borghi et al. (2019) found that psychosocial factors were important for parent adjustment to and treatment of phenylketonuria.

Psychosocial factors can influence intervention whether or not they are recognized by the audiologist and early intervention providers and considered in the overall care plan. Therefore, the purpose of this scoping review was to provide information about the research on psychosocial experiences of parents of young children who are deaf or hard of hearing (DHH) and use hearing devices. A scoping review identifies trends and gaps in available evidence and this information can be used to inform practice and identify areas in need of further research.

Method

Procedure

A scoping literature review was completed in June 2020 using the PRISMA extension (Tricco et al., 2018). Scoping reviews follow a systematic process to examine a broad area and can be used to identify main concepts and gaps in research. The purpose of a scoping review is to identify what kind of evidence is available, not necessarily to provide a critical appraisal of the evidence. The Joanna Briggs Institute provides a detailed description of the purpose and process for conducting scoping reviews (Aromataris & Munn, 2017).

For inclusion in the review, the articles needed to address psychosocial factors of parents of children birth to five years who are DHH and use hearing devices within their research question. This age range was selected because young children require help from their parents to access sound consistently through their hearing devices, and parent challenges can interfere with amplification management adherence. Peer-reviewed journal articles published prior to May 31, 2020 were included. Research articles were excluded if child age could not be determined, if the children did not have hearing devices, if no data were collected related to parent psychosocial factors, or if they were not in English.

To identify potentially relevant articles, three databases were searched by the authors (MEDLINE, CINAHL Complete, PsycINFO via EBSCOhost) using the following key words ([hearing loss OR deaf OR hearing impairment OR hearing disorder] AND [hearing aid OR cochlear implant] AND [adaptation OR psychological OR psychosocial OR coping OR quality of life OR mental health] AND [parent OR caregiver OR mother OR father]).

Two of the authors jointly developed a data charting form prior to completing the search and calibrated the search by working together before continuing the search independently. First, article titles and abstracts were reviewed. Second, a full text review was completed, followed by discussion to finalize article selection. Finally, reference lists of included articles were reviewed to identify further articles for consideration. Nine articles met the inclusion criteria (see Figure 1 for article inclusion flowchart). The primary reason for article exclusion was age of the child. Included articles were analyzed to identify psychosocial factors and findings were synthesized to provide a narrative overview.

Results

The scoping review identified nine peer-reviewed research articles, published through May 2020, that investigated psychosocial factors of parents of children birth through five years who are DHH and use a hearing device. Of these, four psychosocial areas were explored (see Table 1 for study details): stress (n = 5), self-efficacy (n = 2), depression (n = 1), and depression/psychological flexibility (n = 1).

Stress

Meadow-Orlans and colleagues published three articles (Meadow-Orlans, 1994, 1995; Meadow-Orlans & Steinberg, 1993) from one study that explored parental stress. The results were part of a larger longitudinal study (MacTurk et al., 1993) that investigated the development of infants that had moderate to profound hearing loss and used hearing aids at four time points (i.e., 9, 12, 15, and 18 months). No differences on the Parenting Stress Inventory (PSI) were found between mothers of infants who were DHH and mothers of infants with typical hearing. The PSI score at nine months, however, was strongly correlated with the mother’s behavior at 18 months for mothers of infants who were DHH, and social support had a significant positive effect on mothers’ behavior with their children who were DHH (Meadow-Orlans & Steinberg, 1993). The authors suggested that when mothers received support closer to the time of hearing loss identification, it had a greater impact on their behaviors at 18 months. Meadow-Orlans (1994) found that there were no difference in stress levels between mothers and fathers. They also found that PSI scores were significantly related to the Life Stress Index for mothers, but not the fathers. PSI sub-scales revealed fathers of children who
were DHH felt less attached to their children compared to their wives, whereas mothers were more depressed than their husbands (Meadow-Orlans, 1995). The authors encouraged professionals to include family support within their services, with particular attention to fathers, stating that families vary and individualization of services for each family member is critical.

Dirks and colleagues (2016) found that mothers of toddlers with bilateral hearing loss had comparable levels of stress compared to mothers of children with typical hearing on the Nijmegen Parenting Stress Index. Children of parents with higher stress levels had poorer social-emotional functioning and language ability, and parents who received less social support reported higher levels of stress. The authors indicated that professionals have a role in being aware of signs of parental stress and should pay attention to social support and social networks of parents.

Jean and colleagues (2018) interviewed mothers of children with severe-to-profound hearing loss to explore their experience with parenting stress and two themes emerged: contextual stressors and stress-reducing resources. Contextual stressors included distress related to hearing devices and intervention services that often contributed to delays in intervention and a lack of commitment to the intervention program. Parents described that the process of having to gain new
knowledge and apply new learning was stressful. Parents expressed distress from not meeting their own personal expectations, concerns about their child’s future, and experiencing negative social attitudes from others about their child’s hearing; including from family, friends, and strangers. Maternal coherence was the core social process that emerged from the interviews and it appeared to influence how mothers view and experience their parenting stress and their overall sense of wellbeing and parenting control. Mothers who perceived that they had control over the context stressors indicated that they felt more confident and motivated to engage in the intervention process.

**Self-efficacy**

Self-efficacy is broadly described as perceived estimations of an individual’s competence and confidence to perform a task. Desjardin (2005) created the Scale of Parental Involvement and Maternal Self-Efficacy (SPISE) and used it to assess maternal self-efficacy for mothers of young children with profound hearing loss who use hearing aids or cochlear implants. The findings revealed mothers of children with cochlear implants had higher self-efficacy than mothers of children with hearing aids. Furthermore, mothers of children with cochlear implants perceived themselves to be more involved in their child’s device use (i.e., checking device function on a daily basis and supporting speech-language development). The author described technical training and support needs critical for parents to learn new information and skills.

Ambrose and colleagues (2020) used a revised version of the SPISE (SPISE-R) to explore self-efficacy for parents of children birth to 36 months who use cochlear implants or hearing aids. Findings revealed that parents of children with cochlear implants reported higher knowledge scores than parents of children with hearing aids, and that mothers reported higher confidence than fathers. Furthermore, scores on knowledge and confidence were significantly correlated with parent action and hearing device use, and confidence scores were significantly related to language scores. The authors concluded that to provide comprehensive support for families it is important to assess parents’ perceptions, knowledge, confidence, and actions; as this information will help early intervention professionals identify parents’ strengths and areas in which they may need additional support and guidance.

**Psychological Flexibility**

Psychological flexibility describes a thought process used to respond effectively to difficult internal experiences by being open and nonjudgmental of these experiences in the present moment and able to successfully take personally meaningful action (Hayes et al., 2006). Muñoz and colleagues (2014) explored hearing aid management challenges for mothers and fathers of children birth to three years. Using a general instrument, Acceptance and Action Questionnaire (AAQ-II; Bond et al., 2011), Muñoz et al. surveyed psychological flexibility and depression (described in the next section) on hearing aid management. Ninety-six percent of the parents had low scores on the AAQ-II, indicating that psychological inflexibility was not a factor. A limitation was that this instrument was not specific to parents of children who are DHH, and challenges with psychological flexibility in the context of hearing aid management may have been missed by using the general AAQ-II questionnaire.

<table>
<thead>
<tr>
<th>First Author</th>
<th>Year</th>
<th>Sample Size</th>
<th>Demographics</th>
<th>Psychosocial Aspect</th>
<th>Hearing Device</th>
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<tr>
<td>Meadow-Orlans</td>
<td>1993; 1994;1995</td>
<td>40</td>
<td>Mothers and fathers; primarily white</td>
<td>Stress</td>
<td>Hearing aid</td>
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<td>2005</td>
<td>54</td>
<td>Mothers; 63% white</td>
<td>Self-efficacy</td>
<td>Hearing aid; cochlear implant</td>
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<td>2014</td>
<td>55</td>
<td>Mothers and fathers; 91% white</td>
<td>Psychological flexibility; depression</td>
<td>Hearing aid</td>
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<td>2016</td>
<td>30</td>
<td>Not reported</td>
<td>Stress</td>
<td>Hearing aid</td>
</tr>
<tr>
<td>Caballero</td>
<td>2017</td>
<td>42</td>
<td>80% mothers; Hispanic</td>
<td>Depression</td>
<td>Hearing aid</td>
</tr>
<tr>
<td>Jean</td>
<td>2018</td>
<td>15</td>
<td>Mothers; Malaysian</td>
<td>Stress</td>
<td>Hearing aid</td>
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<td>Hearing aid; cochlear implant</td>
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Depression

In a study done by Muñoz et al. (2014), depression was explored using the Patient Health Questionnaire (PHQ-9; Pfizer, 1999). Twenty-two percent of the parents were experiencing mild to severe symptoms of depression, and of those, 40% indicated the depression was influencing their ability to manage. The authors stated that comprehensive care includes addressing needs of mothers and fathers. In a similar study, Caballero and colleagues (2017) explored hearing aid management, including symptoms of depression, for mothers using the Spanish version of the PHQ-9. Forty-four percent of the participants reported minimal to moderately severe symptoms of depression.

Discussion

Parents of children who are DHH experience treatment adherence challenges for hearing device management. This is a critical issue as auditory access is essential for spoken language development. The model of cumulative auditory experience includes consistent use of well-functioning hearing aids and states that intervention for malleable factors are needed to reduce barriers that interfere with auditory access (McCreery & Walker, 2017). Parent psychosocial experiences can interfere with auditory access and it is important to recognize that psychosocial issues can be positively influenced with appropriate support. The purpose of this scoping review was to provide information about the research on psychosocial experiences of parents of young children who are DHH and use hearing devices. This scoping review revealed research gaps and opportunities to expand services to include identifying and addressing the needs of parents when children are identified with hearing loss—parents are essential intervention partners and their engagement is critical for optimizing child outcomes.

Parents can experience both practical and emotional barriers related to hearing aid management; however, there is a scarcity of research on parent psychosocial factors that interfere with and/or facilitate engagement in hearing device management for parents of children birth to five years of age. All of the studies identified in this review voiced the importance of and the need for parents to receive support that is individualized to their specific needs. This support may help them address challenges that interfere with effective engagement in the intervention process.

The gaps in the research conducted to date present limitations. The study samples represent a narrow demographic—primarily English-speaking, white mothers with a college education. Relatively little research has been done related to the psychosocial considerations for fathers and other caregivers. No intervention studies have been done to address parent psychosocial challenges. Research is needed to understand the needs of a broader demographic of parents, including those that do not speak English, and to provide more depth of understanding of the work parents must engage in to successfully implement treatment recommendations. Additionally, research is needed to identify effective approaches for addressing parent psychosocial barriers to treatment adherence within the intervention process, including interprofessional collaboration.

There are important clinical implications to consider when a child is identified with hearing loss. Current practice guidelines address parent adjustment (American Academy of Audiology [AAA], 2013); however, specific implementation guidance is lacking. There are validated screening tools clinicians can include as part of routine practice to assist them in determining when parents are struggling with internal distress (e.g., depression) that is interfering with treatment adherence. To address parental struggles, providers need to understand the extent of parent struggles, the intervention options available within their scope of practice, and how to recognize when referrals are indicated for professional counseling. Without practice guidelines that address the issue of how to support parents, the extent and scope of services provided will likely be insufficient and widely variable.

Informational counseling has received more attention in guidance documents (AAA, 2013) and clinicians have reported being more comfortable with providing information than addressing parent emotional barriers (Meibos et al., 2017). Providing parents with information is important; however, it is not sufficient to support the behavior change parents must engage in to become proficient in their role. Support for health behavior change, which includes addressing parent psychosocial challenges, is a component of service delivery that is largely missing from pediatric audiology practice and is understudied as it relates to pediatric hearing loss. Partnering with parents requires a responsiveness to the practical and emotional work they must navigate to fulfill their role, and this includes individualized support. Parents are their child’s most important teacher and are the people most invested in their child’s future.

This scoping review revealed a scarcity of research related to psychosocial barriers that parents of young children who use hearing devices experience. Research is needed to identify effective interventions to support parents in reducing barriers to auditory access for children, and to demonstrate the effect of addressing parent psychosocial barriers on spoken language outcomes for children. This review brought to light important opportunities. Providers can use validated screening tools to assess for parent barriers, and providers can incorporate individualized support for parents within the care plan for children identified with hearing loss when parents are struggling.

References


