Auditory and language outcomes in children with unilateral hearing loss

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Abstract

Objectives: Children with unilateral hearing loss (UHL) are being diagnosed at younger ages because of newborn hearing screening. Historically, they have been considered at risk for difficulties in listening and language development. Little information is available on contemporary cohorts of children identified in the early months of life. We examined auditory and language acquisition outcomes in a contemporary cohort of early-identified children with UHL and compared their outcomes at preschool age with peers with mild bilateral loss and with normal hearing.

Design: As part of the Mild and Unilateral Hearing Loss in Children Study, we collected auditory and spoken language outcomes on children with unilateral, bilateral hearing loss and with normal hearing over a four-year period. This report provides a cross-sectional analysis of results at age 48 months. A total of 120 children (38 unilateral and 31 bilateral mild, 51 normal hearing) were enrolled in the study from 2010-2015. Children started the study at varying ages between 12 and 36 months of age and were followed until age 36-48 months. The median age of identification of hearing loss was 3.4 months (IQR: 2.0, 5.5) for unilateral and 3.6 months (IQR: 2.7, 5.9) for the mild bilateral group. Families completed an intake form at enrolment to provide baseline child and family-related characteristics. Data on amplification fitting and use were collected via parent questionnaires at each annual assessment interval. This study involved a range of auditory development and language measures. For this report, we focus on the end of follow-up results from two auditory development questionnaires and three standardized speech-language assessments. Assessments included in this report were completed at a

median age of 47.8 months (IQR: 38.8, 48.5). Using ANOVA, we examined auditory and language outcomes in children with UHL and compared their scores to children with mild bilateral hearing loss and those with normal hearing.

Results: On most measures, children with UHL performed poorer than those in the mild bilateral and normal hearing study groups. All children with hearing loss performed at lower levels compared to the normal hearing control group. However, mean standard scores for the normal hearing group in this study were above normative means for the language measures. In particular, children with UHL showed gaps compared to the normal hearing control group in functional auditory listening and in receptive and expressive language skills (three quarters of one standard deviation below) at age 48 months. Their performance in receptive vocabulary and speech production was not significantly different from that of their hearing peers ($p<0.001$).

Conclusions: Even when identified in the first months of life, children with UHL show a tendency to lag behind their normal hearing peers in functional auditory listening and in receptive and expressive language development.

Key words: children, unilateral hearing loss, mild hearing loss, auditory function, language development

INTRODUCTION

Population-based newborn hearing screening aims to improve developmental outcomes for all children with hearing loss through early detection and intervention. There has been renewed interest in pediatric unilateral hearing loss (UHL) since universal newborn hearing screening (UNHS) has become standard care. Bess and colleagues drew attention in the 1980s to children with 'minimal' loss, both UHL and mild bilateral loss, showing that they were at risk for difficulties in language-related and other developmental areas (Bess, 1985; Bess et al. 1986). However, these results were generally based on children identified in the late preschool/school age years. Essentially, a 'new population' of early-identified children has now surfaced with little information about the impact of UHL when identified in the first months of life. In screening programs that specifically include milder hearing loss, the average age of diagnosis has been substantially reduced from school age to less than 1 to 2 years of age (Fitzpatrick et al. 2014; Ghogomu et al. 2014). Even when not specifically targeted, UHL is identified in infancy as a by-product of screening (Wood et al. 2015). Recent population-based data from one region of Ontario showed that 20.1% (108 of 537) of all children diagnosed with permanent hearing loss from 2003-2015 had UHL and children with congenital loss were diagnosed at a median age of 2.8 months (Fitzpatrick et al. 2017). Based on data from the Infant Hearing Program (IHP) in Ontario, the provincial early hearing detection and intervention program, Bagatto et al. (2016) reported that approximately 15% of children identified up to age 6 years had UHL.

Research in the 1980s and 1990s underscored some of the difficulties for children with UHL in the area of localization, listening in noise, language, and academic function (Porter et al. 2016). More recently, in a series of reports from a case-controlled study, comparing children with UHL to their siblings, Lieu and colleagues have shown that children with UHL are at risk for difficulties in communication and academic function (; Lieu et al. 2010, 2012). These school-aged children with UHL had lower oral language scores and were 2.5 times more likely to have received speech-language therapy than their siblings. Although language scores improved in later school years, academic and behavioral problems continued relative to their siblings and 51% required an Individualized Educational Plan (IEP) compared to a US average of 12.3% (Lieu et al. 2012). However, in these studies, the average age of diagnosis was 4.7 years (standard deviation [SD]: 2.6), relatively late compared to what might be expected in newborn screening cohorts. Furthermore, based on a meta-analysis, Borton et al. (2010) found that children with UHL had significantly lower quality of life scores particularly in the school domain. Another recent meta-analysis, that compared intelligence scores from children with UHL and those with normal hearing, concluded that children with UHL had lower IQ scores (difference of 6.4 points; 95% CI: -0.1, -3.5) (Purcell et al. 2016). However, most of the children included in these studies were diagnosed late relative to current expectations, and therefore this research shows the increased negative consequences of UHL when diagnosed at late preschool/school age.

Even when UHL is identified early, there is some evidence that children have difficulties in the area of communication. In a study of 26 children identified before 6 months of age, 17-23% assessed between age 2 to 5 years had language scores below the

10th percentile on parent-report measures compared to normative data for hearing children and one-third had mean length of utterance below the expected score for hearing peers (Sedey et al. 2005). Of 15 children in this study who had multiple measures, 27% had persistent language delay, all of whom reportedly had severe to profound loss in the impaired ear (Sedey et al. 2005; Yoshinaga-Itano et al. 2008). A study of 34 children with UHL not fitted with amplification, evaluated at a median age of 9.4 months, reported that infants were at high risk in auditory and oral communication behaviours. After adjusting for other communication risk factors, the study reported delayed auditory and preverbal vocalizations to be four and nine times more common than in these infants compared to a group without hearing loss (Kishon-Rabin et al. 2015).

Although numerous studies have shown a range of negative consequences for children with UHL (Anne et al. 2017; Appachi et al. 2017), there have been a few contradictory results. For example, Keller & Bundy (1980) found no differences in children with UHL on standardized educational test scores compared to their normal hearing siblings and Hallmo et al (1986) reported no differences in language and academic function in children with UHL. Overall, current research continues to indicate that UHL affects development for at least some children (Vila & Lieu 2015). However, the bulk of the evidence comes from children assessed at school age, most being late-identified or having late-onset hearing loss. From these studies, it is not possible to determine whether late-identified hearing loss was a factor negatively impacting outcomes. Overall, the research related to outcomes in children identified in the first year or so of life is still quite sparse. Consequently, best practices related to intervention and technology have remained unclear.

Intervention with hearing technology for UHL has evolved from no treatment in the early years or an approach that involved monitoring the child (i.e., wait and see whether intervention is required) to the fitting of conventional and bone anchored implant systems (Snik et al. 2005) and more recently to cochlear implants for severe to profound unilateral loss (Arndt et al. 2015). However, research has also shown that fitting of hearing technology tends to be later than for peers with bilateral loss and that amplification use is challenging (Fitzpatrick et al. 2010, 2014). Delay to fitting appears to be associated with age at identification, with older children more likely to be fitted shortly after identification compared to younger children (Fitzpatrick et al. 2014). It is unclear whether this is due to hesitation on the part of both parents and audiologists. Our qualitative research has shown that the need for hearing aids emerged as one of the most confusing and uncertain areas for parents of young children with minimal hearing loss (Fitzpatrick et al. 2016). One reason for the uncertainty is the overall lack of evidence about the effectiveness of intervention and in parallel about the outcomes for these children, especially when UHL is identified early.

Essentially, audiologists and therapists are now confronted with a new generation of children with UHL. Given that about 1 in 5 to 1 in 7 (Bagatto et al. 2016; Fitzpatrick et al. 2017), children with permanent hearing loss can be expected to be diagnosed with UHL in the early years, it is important to collect contemporary data to guide parents about expectations for outcomes and to guide clinics in making decisions about whether children might be candidates for intervention services. The purpose of this inquiry was to examine auditory function and spoken language outcomes at 4 years of age in early-

identified children diagnosed with UHL. We compared their performance with that of children with mild bilateral hearing loss and children with normal hearing.

METHODS

Design and setting

This study is based on cross-sectional data collected as part of the Mild Bilateral and Unilateral Hearing Loss (MUHL) study, a multicenter observational longitudinal cohort study investigating developmental outcomes in preschool age children with minimal hearing loss (mild bilateral or unilateral hearing loss). During the study, measures were collected related to several aspects of auditory and communication development as well as amplification use and parent perspectives. For this study, our interest was in auditory and communication development of the group of children with unilateral hearing loss. For this report, outcomes were examined cross-sectionally at study end (approximately age 48 months) for two auditory outcome measures and three standardized language assessments.

Setting

The province-wide universal newborn hearing screening (UNHS) program in Ontario, Canada, known as the Infant Hearing Program (IHP) was implemented in 2002, and is comprised of screening, early communication development, and parent support, all publicly funded services, (Hyde et al. 2004; Brown and Mackenzie 2005). Ontario is Canada's largest province with a population of approximately 11 million people and approximately 350-400 children are diagnosed with hearing loss annually under the IHP.

Unlike some jurisdictions, the Ontario program specifically includes unilateral and mild bilateral hearing loss in the target disorder. Province-wide mandated audiologic protocols have been developed and updated (Ontario Ministry of Children and Youth Services 2014; Bagatto et al. 2016). Children are screened using a two-step process and those who do not pass screening, undergo diagnostic assessment at a designated pediatric audiology center. Children with permanent hearing loss, who are considered for hearing technology, are referred to an otolaryngologist to receive medical clearance if applicable. Through this publicly funded program, cchildren are typically provided with audiological follow-up at 3- and 6-month intervals respectively during the first and second year post-diagnosis, followed by annual visits up to 6 years of age (Ontario Ministry of Children and Youth Services 2014). Provision and duration of therapy is determined by the child's intervention program. In some regions, children are seen for intervention at the diagnostic audiology center whereas in others, children are seen in off-site community-based therapy and specialized educational programs; all children have a designated audiology center for ongoing follow-up.

Participants

Children in the MUHL study met the following inclusion criteria: 1) permanent unilateral or mild bilateral hearing loss, 2) less than age 3 years at enrolment, 3) English spoken as one of the languages in the home. We excluded children with a diagnosis of severe developmental delay who could not complete the study protocol, which involved multiple spoken language assessments.

The definition for unilateral and for mild bilateral hearing loss for this study was

applied from the National Workshop on Mild Bilateral and Unilateral Hearing Loss (2005), where unilateral hearing loss refers to hearing loss in one ear only with a pure-tone average ≥ 20 dB HL or > 25 dB at two or more frequencies above 2 kHz. Mild bilateral hearing loss refers to average pure-tone air conduction thresholds (at 0.5, 1, and 2 kHz) between 20 and 40 dB HL or thresholds > 25 dB HL at two or more frequencies above 2 kHz. For this study, mild bilateral loss was determined based on better ear hearing thresholds.

Children and their families were enrolled in the study between 2010 and 2014. Respecting research ethics requirements, children with hearing loss were recruited through their clinical providers from three regions in Ontario (Ottawa, Toronto, and Southern Ontario). Clinical providers were asked to invite all eligible families by sharing written information on the study. However, no information could be collected on the number of families approached compared to the number who accepted to participate. Children entered the study at various ages between 12 and 36 months and were followed up to study end, which for most children was age 48 months.

Children with normal hearing were recruited through Parent and Baby drop-in play groups at Early Years Centers (public early childhood education centers) situated in four different areas of the region in Ontario (n=24) as well as through the provincial Infant Hearing Program Community Screening Clinics (n=27). For the former group, researchers visited the education centers and presented information about the study to parents. For the latter group, professionals in the screening clinics were asked to distribute information about the study to parents after the child received a pass on the

hearing screen. Four additional children were recruited through contact with providers at the various sites.

The Children's Hospital of Eastern Ontario (CHEO) (file #09-64X) Research Institute (main study site), and the University of Ottawa (file #H10-09-11), approved the study and the study met ethics requirements for all of the clinical programs involved in inviting participants. Informed consent was obtained from all parents who participated in the study.

Procedures

For this report, our focus is on auditory and spoken language outcomes for children at the end of the study, planned at age 48 months, but which varied from age 36 to 48 months due to study design. Baseline characteristics related to the child and family were collected through an intake form at study enrolment. At enrolment and annually, families also completed a study-specific intervention questionnaire, which addressed changes in hearing, amplification recommendations and use, as well as type of therapy services. With parental consent, information about the diagnosis and degree of hearing loss as well as amplification was collected annually from the child's audiology center to confirm parent reports.

For this study, we analyzed data at study end (typically 48-month test interval) from two parent-administered auditory questionnaires as well as standardized language assessments. Language assessments were administered by speech-language pathologists or listening and spoken language specialists with experience in test administration. All test measures were double-scored by a second researcher prior to analysis.

Description of measures

In this article, data were analyzed for the auditory outcomes measures, Parents' Evaluation of Aural/Oral Performance of Children (PEACH) (Ching & Hill, 2007), and the Children's Home Inventory for Listening Difficulties (CHILD) (Anderson & Smaldino 2011). The PEACH is a questionnaire designed to assess the performance of children with hearing loss (ranging from mild to profound degrees), ranging in age from 4 weeks to 18 years, in everyday life situations based on parents' observations. Research has supported the reliability of the PEACH and has provided some normative data (Ching & Hill 2007). In this study, we asked parents to complete the short form of the PEACH, which consists of 11 questions and compared scores to those we collected on children with normal hearing in our study. We recorded and analyzed PEACH percentage score (maximum score=100%) for the PEACH-quiet and PEACH-noise subscales. The Children's Home Inventory for Listening Difficulties – CHILD (Anderson & Smaldino 2011) was developed as a family-centered clinical tool for parents of children from age 3 to age 12. Parents are asked to observe and rate their child's abilities (on an 8-point Likert scale) for 15 different listening situations in the home environment. The score recorded was the average of the responses (the total sum of the ratings divided by 15). To our knowledge, there are no published normative means for the CHILD; we therefore compared the results to the group score for children with normal hearing assessed in our study. According to the authors, the CHILD is designed to assist in identifying the effects of 'subtle' hearing loss on communication areas such as attention span, following directions and behaviour.

Speech-language measures administered to the child included: the Peabody Picture Vocabulary Test (PPVT-4) (Dunn & Dunn, 2007), Preschool Language Scale (PLS-5) (Zimmerman et al. 2011), and the Goldman-Fristoe Test of Articulation Sound-in Words subtest (GFTA-2) (Goldman & Fristoe, 2001). All of these are norm-referenced tests that are widely used in clinical and research assessments. All have mean standard scores of 100 and a standard deviation of 15. All results are reported as standard scores for this study. Results were compared with mean scores obtained for children with normal hearing assessed in this study.

The PPVT-4 (Dunn & Dunn, 2007) is a norm-referenced test for ages 2.5 to 90 years, which is widely used to assess receptive vocabulary. Individuals are required to point to one of four pictures on an easel board to indicate understanding of the word produced by the tester. The PLS-5 (Zimmerman et al. 2011) measures receptive and expressive language from birth to age 6 years, 11 months. The test has receptive and expressive language tasks and includes a picture book and toys as stimuli. The test provides a standard score for two subscales, Auditory Comprehension (AC) and Expressive Communication (EC). The GFTA-2 (Goldman & Fristoe, 2001) is a measure of speech production ability for ages 3 years to adult. The Sounds-in Words subtest was administered in this study to measure children's articulation skills. The test consists of pictures that are used to elicit targeted speech sounds from the child in initial, medial, and final positions in words.

Data analysis

Statistical analysis was carried out using SPSS Version 24 (IBM Corporation).

Sociodemographic and baseline clinical characteristics of the participants were summarized by group (unilateral, mild bilateral, normal hearing) using descriptive statistics and included frequency counts as well as means and standard deviations or medians and interquartile ranges (IQR), as appropriate. Data were visually inspected to examine normality of the distribution of variables. Statistical tests were conducted to compare differences between groups using chi-square tests for categorical variables and Mann-Whitney U or ANOVA tests for continuous variables as appropriate.

Using Analysis of Variance tests (ANOVA), we examined auditory (PEACH and CHILD scores) and language outcomes (PPVT, PLS, GFTA) in children with UHL and compared their scores to children with mild bilateral hearing loss and with normal hearing assessed in our study group. For the auditory questionnaires, PEACH and CHILD, given that a small number of children (n=9) had completed their assessment between 36 and 48 months of age, rather than at the 48-month follow-up interval, we added age as a covariate in the ANCOVA to adjust for age at assessment, i.e., to assess whether age affected auditory questionnaire scores for any of the groups. Post-hoc Tukey tests were carried out to examine differences between groups.

In univariate analysis, we examined whether there was an association between sex or maternal education and language outcomes. Maternal education was not associated with any outcome; sex was associated with only one language measure and showed a different trend depending on the groups. Therefore, these variables were not included in the analyses. For the children with UHL, we also explored the effect of amplification use using the Mann-Whitney test as well as the association between degree of hearing loss in the impaired ear at diagnosis (three frequency 0.5, 1k, 2 kHz pure-tone average) and

outcome score using Pearson correlation. For all statistical tests, significance was accepted at the p=0.05 level and all p-values were two-sided.

RESULTS

Clinical characteristics of participants

Table 1 provides the characteristics of all of the participants in this study. As noted earlier, children were invited to enrol in the study from the time of diagnosis of hearing loss and entered the study at different ages up to age 3 years. The children with UHL were enrolled at a median age of 14.2 months (IQR: 9.0, 26.4) and those with bilateral loss at 19.3 months (IQR: 10.8, 28.1). Children with normal hearing were recruited through early education and newborn screening centres and enrolled at a median age of 5.7 months (IQR: 3.5, 13.5); children started the study at varying ages between 12 and 36 months of age depending on age at enrolment. As shown in table 1, median age at final assessment, which was the interest in this study, was similar across all groups at 47.8 to 48.1 months. Four children in the UHL group and 3 in the mild bilateral and 8 in the normal hearing group discontinued assessments prior to completion of the study. There was no significant difference between groups in the proportion of families who did not complete the study (Fisher's Exact Test = 0.75, p=.73). Although efforts were made to complete all assessments in a timely manner, for various reasons (e.g., family illness, relocation to another city), not all children completed every measure at each test interval. Final assessments analyzed for this report were available for 33 children in the UHL group, 27 in the mild bilateral group, and 42 in the normal hearing group). There was no significant difference between the groups in the percentage of children who completed

the assessments analyzed for this report (Fisher's Exact Test = 4.557, p=0.335). Age at final assessment for these children was also not significantly different across the three groups [F (2,10) =1.78, p=0.17].

This was an early-identified group of children with 91.3% of the UHL group and 90.3% of the mild bilateral group referred through newborn screening. There was no significant difference in age of diagnosis (Mann-Whitney U = 512.50, p=0.45); the 38 children with UHL were diagnosed at a median age of 3.4 months (IQR: 2.0, 5.5) and the mild bilateral group at 3.6 months (IQR: 2.7, 5.9). The overwhelming majority (92.1%) of children with UHL presented with congenital hearing loss. In the UHL group, at initial diagnosis, significantly more children (36.8%, n=14) had permanent conductive loss (structural in nature, 13 with atresia and/or microtia) compared to 9.7% of the children with mild bilateral loss ($X^2(1)=6.80$, p = 0.01). Etiology was unknown for the majority of children (50% - UHL to 64.5% - mild bilateral) with hearing loss. At the time of this study, genetic screening and cytomegalovirus (CMV) screening, which has been identified as the most frequent nongenetic cause of UHL in children (Nance, 2007), now approved as part of the Ontario IHP protocol, were not part of standard care for these children. Of the 38 children with UHL, 23 (60.5%) had three frequency pure-tone average less than or equal to 70 dB with the remaining 39.4% having severe (n=11) or profound (n=4) loss.

Amplification was fitted on 26 of 38 children with UHL (68.4%) at a median age of 12.2 months (IQR; 7.2, 29.9). A total of 8 children in this group were fitted with softband bone conduction hearing devices. Four children had amplification recommended but never acquired it. Consistent amplification use was reported for 14 of

these 26 (53.8%) children and the remaining reported inconsistent or no use. For the children with mild bilateral loss, 27 of 31 (87.1%) were fitted with amplification at a median age of 10.5 months (5.2, 21.7). Consistent use was reported for 25 of the 27 (92.6%) children. There was a significant difference in the consistency of amplification use between groups with the mild bilateral group showing greater use ($X^2(1)=10.23$, p=0.002).

By study end, based on audiological reports, 6 of the 38 children in the UHL group at enrolment showed progressive hearing loss, including 3 who had deterioration of ≥ 20 dB HL in hearing levels in the impaired ear and 3 who progressed to bilateral hearing loss (2 of these 3 also lost hearing in the impaired ear). Three children in the mild bilateral group also showed deterioration in thresholds. All children with UHL remained in the initial allocation group (that is, based on UHL at diagnosis) for all analyses.

Auditory function

In this report, we examined outcomes from the PEACH and CHILD at the final test interval. A one-way between-groups analysis of covariance was conducted to compare the difference between groups for scores on the PEACH-quiet and PEACH-noise subscale and the CHILD. Child age at the time the PEACH was completed was entered as a covariate. The UHL group completed the PEACH at a median age of 47.9 months (IQR: 47.1, 48.5), the mild bilateral at 48.1 months (IQR: 47.7, 48.7) and the normal hearing group at 48.0 months (IQR: 47.5, 48.8). Figure 1 shows the results of the PEACH-quiet and PEACH-noise subscale scores as a function of group. After adjusting

for age at assessment, there was no significant difference between groups for the PEACH-quiet subscale score [F (3,91) =1.92, p adjusted=0.12].

For the PEACH-noise score, there was a significant difference between groups [F (3,91) =3.84, p adjusted=0.01] after adjusting for age at assessment. The UHL group achieved a mean score of 76.9% (SD 15.7), which, based on posthoc analyses, was significantly lower than the normal hearing group score of 86.9% (SD 11.5, p=0.002) While the unilateral group scored lower than the mild bilateral group (83.8%, SD 15.6) the difference did not reach statistical significance p=0.05). There was no significant difference in scores between the mild bilateral and normal hearing groups (p=0.34).

There was considerable variability on PEACH-noise scores for the children with hearing loss. Figure 2 shows the individual scores for the UHL and mild bilateral groups, compared to the mean score of 86.9% (SD 11.5) for the children with normal hearing. As shown, 21 of 29 in the UHL group obtained scores below the mean obtained for the normal hearing group, with scores ranging from 45 to 100%. As seen in Figure 2, 13 of 29 children in the UHL group had scores more than 1 SD below the mean obtained for the normal hearing children in this study.

Similarly, for the CHILD, after adjusting for age at assessment, there was a significant difference between groups [F (3,91) = 7.69, p adjusted <0.001]. Posthoc analyses showed that the UHL group had significantly lower scores than the normal hearing group (p<0.001) with a mean of 6.1 (SD 1.0) compared to 7.2 (SD 0.66) for the normal hearing group. The UHL group did not differ significantly (p=0.11) from the mild bilateral group which had a score of 6.6 (SD 0.77). On this measure, the mild bilateral group also performed significantly below the normal hearing group (p=0.01).

Speech-language outcomes

For this study, we performed one-way ANOVAs to compare standardized assessments at study end across the three groups. Age was not entered as a covariate because standardized scores based on age-appropriate normative samples were used in these analyses. In univariate analysis, we examined whether there was an association between sex or maternal education and language outcomes. Sex was not significantly associated with any of the language outcome measures [PPVT, t(85) = 0.18, p=0.91; GFTA, t (87)=-1.64, p=0.38; PLS-AC, t(78) = -2.14; p=0.04; PLS-EC, t (77) = 1.62, p=0.11], except one subtest, the PLS-AC. Given these results and that the trend was different depending on the groups (males lower scores for mild bilateral group only and females lower for UHL and normal hearing groups), we did not include sex as a covariate to allow comparison between the ANOVA results. There was no significant association between maternal education and any language outcome (PPVT, r=0.08, p=0.48; GFTA, r=0.13, p=0.19; PLS-AC, r=-0.15, p=0.20; PLS-EC, re=0.22, p=0.05).

As shown in Figure 3, the PPVT showed no significant difference across groups [F (2,84)=1.78, p=0.18]. Standard scores ranged from a mean of 108.4 (SD 15.8) for the mild bilateral group to 112.6 (SD 16.1) for the UHL and a mean of 115.8 (SD 14.5) for the normal hearing groups.

For the PLS-AC (receptive language), there was a significant difference between the three groups [F (2,77) = 4.32, p=0.02]. Posthoc Tukey tests showed that the UHL group (M =103.4, SD 15.8) performed poorer than the normal hearing group (M=114.6, SD 12.2; p=0.02) but not differently than the mild bilateral group (p=0.89). Similarly, there was a significant difference [F (2.76) = 4.73, p=0.01] between the three groups on

the PLS-EC subtest. Again, the UHL group (*M*=103.9, SD 12.5) performed lower than the hearing control group (*M*=115.1, SD 18.2; p=0.04) but not differently than the mild bilateral group (p=0.99). Overall, this represented a difference in standard scores of 11-12 points between the UHL and normal hearing control groups on each PLS subtest.

Speech production measured by the GFTA, showed no significant difference between the three groups [F (2,77) = 2.12, p=0.13] with the UHL group showing a mean standard score of 111.1 (SD 7.3) compared to a mean score of 103.8 (SD 17.0) and 109.3 (SD 10.0) for the mild bilateral and normal hearing groups.

Given that these measures were standardized norm-reference language assessments, we also compared mean scores to the normative means (*M=100, SD 15)* using one sample t-tests. The children with normal hearing in our control group scored significantly above the normative mean on all standardized assessments. [PPVT: t(33)=6.35; PLS-AC: t(32)= 6.90; PLS-EC, t(32)=4.79; GFTA, t(32)=5.35; p <0.001 for all tests]. For the UHL group, scores were not significantly different from the normative mean on receptive language [PLS-AC: t(24) = 1.087, p=0.29] and expressive language measures [PLS-EC: t(23)=1.515, p=0.14], however, the group mean was significantly higher than the normative mean on both receptive vocabulary [PPVT: t(27)=4.14, p <0.001] and articulation (GFTA: t(27)=7.30, p<0.0001). Scores for the mild bilateral group were higher than the normative mean only for receptive vocabulary [PPVT: t(24)=2.65, p=0.01] and not significantly different for any other measures [PLS-AC: t(21)=1.43, p=0.17; PLS-EC: t(21)=0.92, p=0.37; GFTA: t(18)=0.99, p=0.34].

An examination of individual scores on the PLS showed more children with hearing loss below test norms. For example, four children (16%) in the UHL group and

three in the mild bilateral group scored below a standard score of 85 (1 SD below the normative mean of 100), considered clinically to be below average, while no children in the normal hearing control group scored below the average range; only 1 child having a score of 88 fell below 0.5 SD of the test mean.

We explored the effects of amplification use on PLS-AC scores for the UHL group as this subtest showed the greatest difference compared to the normal hearing control group. We examined the association between scores and treatment followed/not followed (i.e., used amplification consistently if recommended or did not use amplification recommended). There was no significant difference between the treatment followed group (PLS-AC: *M*=101.7, SD 15.9) and the treatment not followed group [PLS-AC: *M*=106.0, SD 15.9; t(23)=0.65, p=0.52]. The small number of children not using amplification in the mild bilateral group precluded any analysis related to the effects of amplification. An examination of the effect of degree of hearing loss for the children with UHL did not reveal any association with outcomes (r =-0.25, p=0.12).

DISCUSSION

Early identification and management of children with UHL is a relatively recent consideration as a consequence of newborn screening programs. Our findings showed that by the end of their preschool years, at age 48 months, children with UHL on average had scores that were lower than the normal hearing peer group in this study on some measures of auditory function and language. Auditory function was behind when compared to the control group using the PEACH-noise scores and CHILD scores. Although, children with UHL scored within test norms for children with normal hearing

on standardized language assessments, on average, their receptive and expressive language skills demonstrated a gap of almost a standard deviation (11-12 points) when compared with children with normal hearing in our study drawn from the same population. Their scores on receptive vocabulary and speech production were however aligned with both their peers with normal hearing and those with mild bilateral loss. While both children with UHL and mild bilateral loss tended to show gaps compared to the normal hearing study group in most areas measured, the UHL group showed greater differences. They also demonstrated somewhat lower scores than their peers with mild bilateral loss, although generally the differences were not statistically significant.

An examination of factors including sex, maternal education level and amplification use (for the UHL group) did not shed any further light on which children were more likely to experience difficulty with language acquisition. Examination of outcomes for the UHL group also did not show any association with degree of hearing loss.

Our results for these young children with UHL based on a functional auditory measure (PEACH-noise score) are consistent with reports for older children with UHL based on direct speech perception measures. Studies have shown that children with UHL have more difficulty than their normal hearing peers on speech recognition tasks in adverse acoustic conditions (Bess et al. 1986; Rothpletz et al. 2012). For example, Bess et al. (1986) found that for children with UHL, there was about a 40% difference in scores compared to normal hearing children on a nonsense syllable test when noise was presented to the good ear and speech to the impaired ear (~40% vs. 80% correct).

At first glance, our findings in speech-language acquisition are encouraging in

that they show that early identified children with UHL obtain scores on standardized measures that are within the mean for test normative data. However, as pointed out by Tomblin et al. (2015), it is likely more meaningful to compare outcomes with hearing peers from the same population as standardized test scores may provide an underestimate of their actual potential. In a previous study from our lab, we also found that while children with hearing loss ranging from mild to profound obtained scores within test norms, they were well behind their local normal hearing counterparts based on a study control group (Fitzpatrick et al. 2011). Recently, for a large cohort of 290 children with mild to severe hearing loss, Tomblin et al. (2015) reported that, although they were within a third of a SD of test norms, they were actually two-thirds of a SD behind hearing peers from the same SES group. As noted previously, average standard scores for children with normal hearing in the current study were well above standardized test norms of 100 and much closer to standard scores in the 115 range for language and vocabulary tests. Consequently, when compared with outcomes for their local population, our findings indicate that preschool age children with UHL are at risk for delayed auditory behaviours and receptive and expressive language development. However, their functioning in receptive vocabulary and speech production were comparable to peers in the normal hearing study group and above normative test means. Similarly, scores for children with mild bilateral loss, although showing smaller gaps, suggest that careful monitoring and long-term follow-up are required to prevent gaps from widening compared to their normal hearing age-mates. These results also point to the need for more in-depth language analyses beyond standardized measures to examine grammatical morphemes to better understand the gaps in children's language. Results

from the Tomblin et al. (2015) study suggested that at age 4 years, morphosyntax was more affected than lexical development in children with hearing loss.

Our findings, when compared to the study control group, are in line with those of Sedey et al. (2005) who reported that about one-third of preschool children with UHL lagged behind their peers in language acquisition. Findings from early research in the 1990s by Bess and colleagues showed that about a third of children with minimal hearing loss, who were much later-identified (UHL and mild bilateral) had language and/or academic difficulties and repeated a year in school (Bess et al. 1998). Despite very early age at identification of hearing loss of about 3.5 months for the children in our study, they had not entirely closed the gap in some areas of linguistic development when compared to the normal hearing study group.

Strengths of this study include the multi-center and prospective nature of the research, which allowed us to collect detailed baseline characteristics of these children and outcome data in a standard format, rather than relying on clinical data. It is an advantage that children were serviced in different clinical programs as it provides a broader representation of this clinical population. A second important strength is the use of a local comparison group of children with normal hearing, which we believe to be more representative than depending on test norms. Furthermore, all children were managed through the Ontario Infant Hearing Program (Ontario Ministry of Children and Youth Services 2014), which has established provincial protocols for diagnosis and management. All services are publicly funded, eliminating to some degree the variable of access to services Most importantly, this cohort was unique in that the very young age of the children at diagnosis allowed us to examine the effect of UHL when identified

and managed early.

However, our study has some limitations, the most important one being the small number of children with UHL and mild bilateral loss that were recruited over a 5-year period, which precluded analyses of certain factors affecting outcomes. Our previous studies have shown that approximately 20% of children with permanent hearing loss are initially diagnosed with UHL (Fitzpatrick et al. 2017). This represents an estimated 100-150 children diagnosed over the study recruitment period in the three regions where parents were invited to participate in this study (these regions include a population of approximately 7 million), therefore we achieved about a 30% enrolment rate. Compared to our previous studies with children with hearing loss, recruitment was particularly challenging and may reflect the lesser degree of concern of providers and perhaps parents when hearing loss is unilateral or mild. Secondly, like other longitudinal studies of this type, we tended to attract families from higher socio-economic status backgrounds, however, this was the case for both hearing loss and normal hearing groups. It is quite possible that the higher SES levels account for the results in the normal hearing control group that placed them well above test normative means. However, it also seems reasonable to assume that if children from higher socioeconomic (SES) backgrounds are below their normal hearing peers, those from less advantaged backgrounds may be even more vulnerable to difficulties. Further population-level studies are required to improve the transferability of the results.

Another limitation is that we were dependent on several audiology clinics to provide updated audiological information, and while we are confident that our information on the child's degree of hearing loss is accurate, we were unable to document

specific information about hearing aid use, (e.g., exact age when hearing aids were discontinued). However, audiological reports were used in combination with parent intervention questionnaires to log these details as accurately as possible. Etiology was unknown for the majority of children (50.0% UHL to 64.5% mild bilateral) with hearing loss, which likely reflects the fact that genetic screening and CMV screening were not required as part of the IHP protocol at the time of this study. These have since been integrated into the protocol and further research will be better able to examine whether there is any relationship between outcomes and etiology.

There appears to be uncertainty about the benefits of intervention and whether children with UHL should be enrolled in intervention programs, leading some programs to not include children with UHL or mild loss in the target disorder for newborn hearing screening programs (Wood et al. 2015) or to not provide them with specialized intervention (Yoshinaga-Itano et al. 2008). Our findings lead us to conclude that these children should at a minimum be carefully and systematically monitored for language development throughout early childhood to minimize the chances of falling behind their peers, particularly in receptive and expressive language. Further analysis is required to examine other aspects of language not measured in standardized tests. We are continuing to follow this group of children into the school years, which should provide valuable information about the effects of hearing loss on language and academic, and social functioning as they progress through school.

In this study, we did not find any substantial differences between children with UHL who followed the recommended treatment program, i.e., using amplification if recommended, or not using amplification. However, as noted, the relatively small

number of children available for analysis for any particular outcome weakened the analysis and may have precluded meaningful conclusions. For the children with UHL, although amplification was recommended for 79% (n=30), less than 50% (14 of 30) of parents reported that they acquired and used it consistently. Our qualitative interviews exploring early experiences with 20 families from the study, half of whom had children with UHL, clearly showed that they found the hearing aid experience to be challenging and confusing. Parents indicated that they were uncertain about the benefits of amplification and how much to invest in encouraging hearing aid use (Fitzpatrick et al. 2016). Controlled studies with larger cohorts of early-identified children with UHL would be valuable in helping to determine the effects of amplification on language outcomes.

Conclusions

This study adds to the growing literature base on UHL in this new era of NHS and contributes information specific to an early-identified cohort of children with unilateral hearing loss. Given that about 1 in 5 children identified with permanent hearing loss initially present with UHL, decisions need to be made about the need for intervention. Our results, raise some red flags and support the need for careful attention to be accorded to children with UHL in the early years as they experience difficulty in some areas of auditory function and are at risk of lagging behind children with normal hearing from their local peer group in receptive and expressive language skills.

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Conflicts of interest: None

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Contributors

EMF conceived the overall project. EMF, IG, ADS, and DC developed the methods and procedures and provided input throughout the study. JW managed the data collection and data entry and JW and FN carried out data verification and statistical analyses. IG oversaw statistical analysis and was involved in the interpretation of the results. EMF drafted the first version of the manuscript. All authors reviewed and approved the final manuscript.

Figure Legend

Figure 1. Results for children by group on the PEACH auditory questionnaire.

Figure 2. Individual PEACH-noise scores for children with hearing loss. Children with normal hearing obtained a mean score of 86.9% (SD: 11.5). The mean and 1 SD are indicated by the horizontal lines on the graph.

Figure 3. Results for children by group on speech-language assessments.

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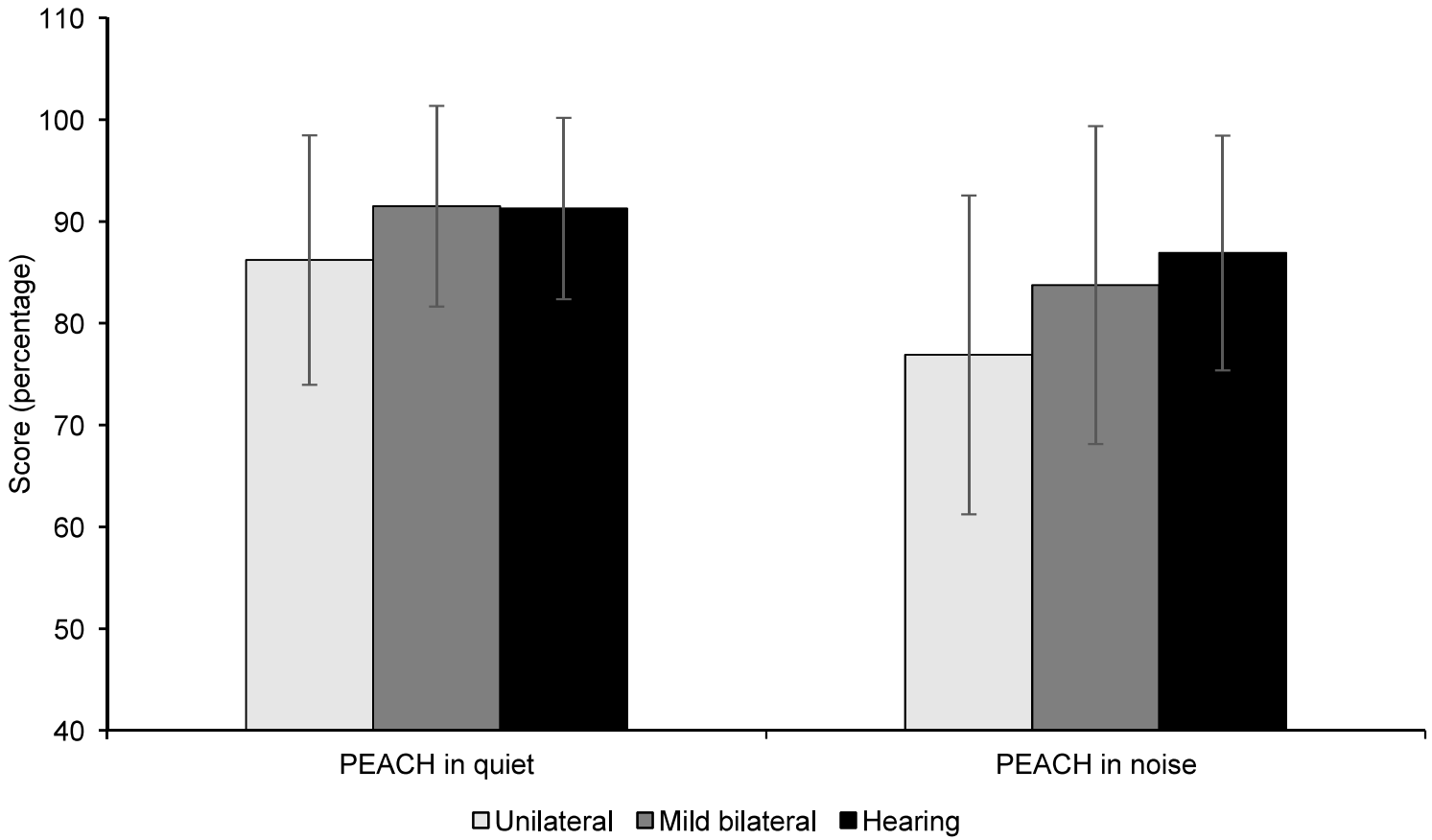
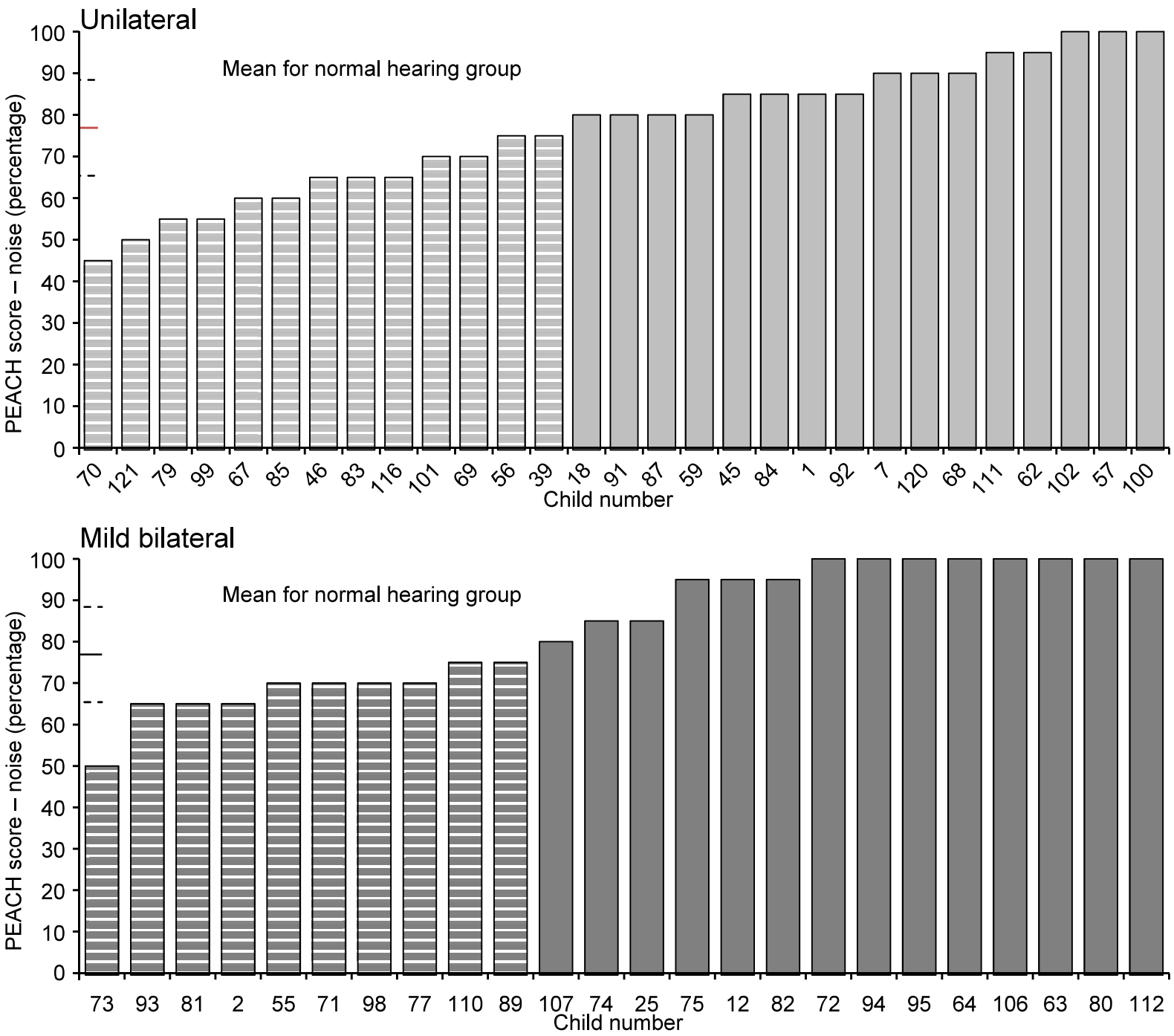

110
100
90
80
70
60
50
40
Score (percentage)
PEACH in quiet
PEACH in noise
Unilateral
Mild bilateral
Hearing


Unilateral
Mean for normal hearing group
PEACH score – noise (percentage)
100
90
80
70
60
50
40
30
20
10
0
70 121 79 99 67 85 46 83 116 101 69 56 39 18 91 87 59 45 84 1 92 7 120 68 111 62 102 57 100
Child number
Mild bilateral
Mean for normal hearing group
PEACH score – noise (percentage)
100
90
80
70
60
50
40
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20
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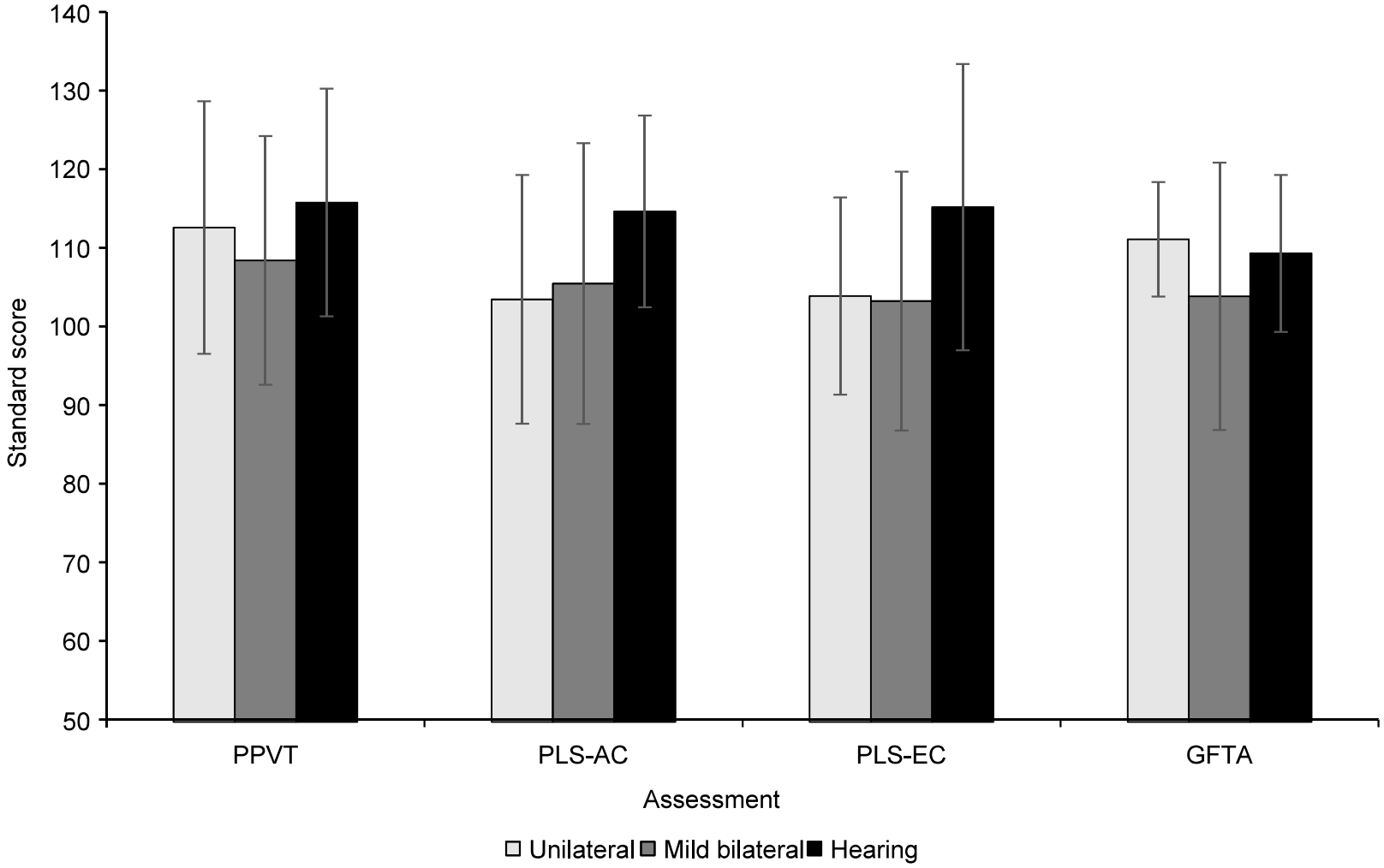
0
73 93 81 2 55 71 98 77 110 89 107 74 25 75 12 82 72 94 95 64 106 63 80 112
Child number


140
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Standard score
PPVT
PLS-AC
PLS-EC
GFTA
Assessment
Unilateral
Mild bilateral
Hearing

Table 1. Characteristics of children with unilateral hearing loss, mild bilateral hearing loss and hearing children

Characteristics	Unilateral HL	Mild Bilateral HL	Hearing	P-value
N	38	31	51	
All Groups				
Sex (% Male)	23 (60.5%)	13 (41.9%)	24 (47.1%)	0.26
Ethnicity				
Canadian / Canadian-Other	28 (73.7%)	20 (64.5%)	45 (88.2%)	0.04
Other	10 (26.3%)	11 (35.5%)	6 (11.8%)	
Maternal education, # years, mean (SD)	17.2 (3.4)	17.7 (3.6)	17.7 (1.9)	0.67
Income[1]				
Up to 80K	14 (37.8%)	12 (40.0%)	10 (20.0%)	0.10
Greater than 80K	23 (62.2%)	18 (60.0%)	40 (80.0%)	
Age enrolment, months,median (IQR)	14.2 (9.0, 26.4)	19.3 (10.8, 28.1)	5.7 (3.5, 13.5)	<0.001
Age assessment, months, median (IQR)	47.8 (38.8, 48.5)	48.1 (47.2, 49.3)	47.8 (47.3, 48.8)	0.79
Hearing Loss Groups				
Screening status				
Screened	35 (92.1%)	28 (90.3%)		0.65
Not screened or unknown status	3 (7.9%)	3 (9.7%)		
Age at diagnosis, months, median (IQR)	3.4 (2.0, 5.5)	3.6 (2.7, 5.9)		0.45
Onset of hearing loss, n (%)				
Congenital	35 (92.1%)	24 (77.4%)		0.10
Early onset (< 6 months)	0	4 (12.9%)		
Late onset (> 6 months)	2 (5.3%)	1 (3.2%)		
Unknown	1 (2.6%)	2 (6.5%)		
Type of hearing loss, n (%)				
Sensorineural	24 (63.2%)	28 (90.3%)		0.01
Conductive	14 (36.8%)	3 (9.7%)		
Etiology known				
ENT malformations	13 (34.2%)	2 (6.5%)		
Hereditary/genetic	2 (5.3%)	6 (19.4%)		
Syndromes	1 (2.6%)	2 (6.5%)		
Neonatal intensive care unit	2 (5.3%)	0		
Cytomegalovirus	1 (2.6%)	1 (3.2%)		
Etiology unknown	19 (50.0%)	20 (64.5%)		
Degree of hearing loss at diagnosis (impaired/worse ear)				
High frequency[2]	0	5 (16.1%)		
Mild (20-40 dB HL)	3 (7.9%)	19 (61.3%)		
Moderate (41-55 dB HL)	7 (18.4%)	4 (12.9%)		
Moderately severe (56-70 dB HL)	13 (34.2%)	3 (9.7%)		
Severe (71-90 dB HL)	11 (28.9%)	0		
Profound (>90 dB HL)	4 (10.5%)	0		
Age rec amplif, months, median (IQR) [3]	6.7 (4.6, 30.0)	4.9 (3.2, 14.9)		0.21
Age fitting amplif, mos, median, (IQR)[4]	12.2 (7.2, 29.9)	10.5 (5.2, 21.7)		0.29

Key: amplif: amplification; IQR: Interquartile range; ENT: Ear Nose Throat (anomalies included atresia and microtia); HL: hearing loss; rec: recommendation; SD: standard deviation

[1] Not reported by 3 families [1]

[2] defined as ≥25 dB HL at ≥2 frequencies above 2 kHz

[3] Age was available for 27 of 30 children with UHL and for 27 or 29 with mild bilateral loss who received amplification recommendations.
[4] Age of fitting was available for 21 of 26 children with UHL and for 25 of 27 with mild bilateral loss loss An additional 6 had amplification recommended but did not acquire it (4 unilateral and 2 mild bilateral).