

## Review Article

# Listening-Related Fatigue in Children With Unilateral Hearing Loss

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**Purpose:** Listening-related fatigue is an understudied construct that may contribute to the auditory, educational, and psychosocial problems experienced by children with unilateral hearing loss (UHL). Herein, we present an overview of listening-related fatigue in school-age children with hearing loss (CHL), with a focus on children with UHL.

**Method:** Following a review of research examining listening-related fatigue in adults and CHL, we present preliminary findings exploring the effects of unilateral and bilateral hearing loss on listening-related fatigue in children. For these exploratory analyses, we used data collected from our ongoing work developing and validating a tool, the Vanderbilt Fatigue Scale, for measuring listening-related fatigue in children. Presently, we are assessing 3 versions of the fatigue scale—child self-report, parent proxy, and teacher proxy. Using these scales, data have been collected from more than 900 participants. Data from children with unilateral and bilateral hearing loss and for children with no hearing loss are compared with adult Vanderbilt Fatigue Scale data.

**Results:** Results of our literature review and exploratory analyses suggest that adults and CHL are at increased risk for listening-related fatigue. Importantly, this increased risk was similar in magnitude regardless of whether the loss was unilateral or bilateral. Subjective ratings, based on child self-report and parent proxy report, were consistent, suggesting that children with unilateral and bilateral hearing loss experienced greater listening-related fatigue than children with no hearing loss. In contrast, results based on teacher proxy report were not sensitive to the effects of hearing loss.

**Conclusions:** Children with UHL are at increased risk for listening-related fatigue, and the magnitude of fatigue is similar to that experienced by children with bilateral hearing loss. Problems of listening-related fatigue in school-age CHL may be better identified by CHL themselves and their parents than by teachers and specialists working with the children.

Unilateral hearing loss (UHL) in children is a common condition. Although the estimated prevalence of congenital sensorineural hearing loss (HL; > 40 dB HL) at birth is only approximately two per 1,000 and 30%–40% exhibit UHL (Kral, Hubka, Heid, & Tillein, 2013; van Wieringen, Boudewyns, Sangen, Wouters, & Desloovere, 2019), the prevalence of UHL appears to increase with age. For example, the prevalence of UHL in school-age children (Grades 3, 6, and 9) is 3% or approximately 1,380,000 children in the United States (Bess, Dodd-Murphy, & Parker, 1998; Centers for Disease Control and

Prevention [CDC], 2005; Tharpe & Sladen, 2008). The leading causes of UHL in children are congenital cytomegalovirus followed by congenital inner ear malformation (e.g., enlarged vestibular aqueduct), bacterial/viral meningitis, viral/bacterial mumps, and Chiari malformation (Porter, Bess, & Tharpe, 2016; Tharpe & Sladen, 2008; van Wieringen et al., 2019). Although definitions of UHL may vary across studies, complicating comparisons, proceedings from the 2005 National Workshop on Mild and Unilateral Hearing Loss suggest UHL exists when the average pure-tone air-conduction threshold at 0.5, 1, and 2 kHz is  $\geq 20$  dB HL or when pure-tone air-conduction thresholds are > 25 dB HL at two or more frequencies above 2 kHz in the affected ear, coupled with an average pure-tone air-conduction threshold in the good ear of  $\leq 15$  dB HL (CDC, 2005).

Research findings related to the psychoeducational outcomes of children with UHL are mixed. Nonetheless, numerous studies have reported that school-age children with UHL experience a variety of auditory, educational, and psychosocial problems (Bess, 1982; Bess et al., 1998; Bess & Tharpe, 1984; Fitzpatrick et al., 2019; Lewis et al.,

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2016; Porter et al., 2016; Reeder, Cadieux, & Firszt, 2015). Compared to children without HL, children with UHL exhibit more difficulty understanding speech in noisy conditions (Bess, Tharpe, & Gibler, 1986; Hartvig Jensen, Johansen, & Borre, 1989; Lewis et al., 2016), localizing sounds in the horizontal plane (Bess et al., 1986; Humes, Allen, & Bess, 1980; Reeder et al., 2015), developing age-appropriate language and cognitive skills (Fitzpatrick et al., 2019; Halliday, Tuomainen, & Rosen, 2017; Klee & Davis-Dansky, 1986), and maintaining satisfactory performance in school (Bess et al., 1998; Bess & Tharpe, 1984; Brookhouser, Worthington, & Kelly, 1991; Lieu, 2004, 2013; Lieu, Tye-Murray, & Fu, 2012; Oyler, Oyler, & Matkin, 1988). In addition, some children with UHL have low self-esteem, anxiety, strained peer relations, and decreased social support (Bess et al., 1998). Even when the UHL is identified early, children may still experience some of the above-referenced communicative and educational problems (Fitzpatrick et al., 2019; Yoshinaga-Itano, Johnson, Carpenter, & Brown, 2008).

Although many studies have shown that children with UHL experience a vast array of academic and communicative problems, such findings are not universal (Hallmo, Møller, Lind, & Tønning, 2009; Keller & Bundy, 1980). These conflicting results may be due to a lack of statistical power (Keller & Bundy, 1980) and/or a failure to include standardized test measures or a control group (Hallmo et al., 2009); however, contemporary research clearly demonstrates that some children with UHL have an increased risk for communication and educational difficulties (Fitzpatrick et al., 2019; Porter et al., 2016).

The mechanisms responsible for the communicative and psychoeducational problems experienced by children with UHL are not well understood; however, a primary factor frequently referenced in the literature is related to the importance of binaural hearing in communication. It is well documented that binaural listening offers distinct advantages over monaural listening, especially in noisy conditions. Factors such as binaural summation, head shadow, and binaural release from masking (squench effect) provide listeners without HL the unique ability to identify and localize sound sources (Blauret, 1997). When children have UHL, however, these binaural cues are diminished, thus producing ripple effects that may impact areas such as education, speech recognition in noise, and language and cognitive skills. Other factors that might also contribute to the problems of children with UHL include changes in auditory and brain structures as a result of auditory deprivation, age of identification/intervention, degree of HL in the impaired ear, and our inability to identify “early on” those children at risk for academic and communicative difficulties (Fitzpatrick et al., 2019; Kumpik & King, 2019; Tharpe, 2008; van Wieringen et al., 2018).

Although research is limited, emerging evidence suggests that listening-related effort and fatigue may also be an important contributor to some of the difficulties experienced by children with UHL. Recent investigations have shown that children, some with even mild, bilateral HL, are at increased

risk of recurrent listening-related fatigue and its negative consequences (Bess, Gustafson, & Hornsby, 2014; Bess et al., 2016; Bess & Hornsby, 2014a; Hornsby, Werfel, Camarata, & Bess, 2014; Hornsby et al., 2017; McGarrigle, Gustafson, Hornsby, & Bess, 2019). Given these findings and the underlying deficits in binaural processing resulting from UHL, it is reasonable to suspect that children with UHL might also experience fatigue difficulties, especially in the school setting.

Here, we offer a review on listening-related fatigue in children with UHL. Given that research specific to UHL does not exist, we first provide a focused review of the literature related to subjective fatigue in adults with HL and children with bilateral HL. Additionally, we explore existing evidence related to fatigue in adults with UHL and present some preliminary findings on listening-related fatigue in children with UHL using validation data from our ongoing work developing the Vanderbilt Fatigue Scale (VFS).

### *Fatigue, Effort, and HL*

A comprehensive examination of fatigue is beyond the scope of this review article; readers interested in an in-depth coverage of this subject are referred to other overview papers (Bess & Hornsby, 2014a; Hornsby, Naylor, & Bess, 2016; McGarrigle et al., 2014). Briefly, fatigue is commonplace in our society. It is a complex construct with physical and mental/cognitive dimensions, and it is often characterized by feelings of weariness, tiredness, a general lack of energy, and/or a reduced desire to continue on with a task. When fatigue is severe and recurrent, these subjective effects may also be accompanied by fatigue-related decrements in physical and/or mental performance.

While many factors modulate its magnitude, fatigue—both mental and physical—is a common consequence of sustained high levels of effort (Hockey, 2013). Here, we focus on fatigue resulting from mental, rather than physical, effort as it is most relevant to the problems of children with hearing loss (CHL). Our specific focus is on listening effort, a type of mental effort that has been described as the allocation of mental resources to listen, process, and understand speech and other auditory stimuli (Downs, 1982; McGarrigle et al., 2019; Pichora-Fuller et al., 2016).

Children in classroom settings have many different demands placed on them that require significant mental effort for successful completion. For example, mental effort is required to complete a written assignment, read a schoolbook, or listen to the teacher and other children in the classroom. The magnitude of listening effort required in these situations can depend on many factors, such as the student's cognitive and attention capabilities and the classroom acoustics (McGarrigle et al., 2019). Importantly, to compensate for their listening difficulties, children and adults with HL must increase their mental effort compared to persons without HL when attempting to detect, process, and respond to auditory stimuli, such as speech (Hicks & Tharpe, 2002; McGarrigle et al., 2014, 2019). This recurrent need for high levels of listening effort puts individuals with HL

at risk for development of listening-related fatigue and its negative consequences.

For most children, complaints of mild transient fatigue are normal, and such fatigue typically resolves readily with a short rest or breaks. Hence, the impact of transient fatigue on academic performance and quality of life is expected to be minimal. For some children, however, their fatigue can be more severe and recurrent and elicited by routine daily activities, such as self-care; completing difficult, but typical, classroom assignments; or relevant to CHL, listening in noisy conditions (Bess & Hornsby, 2014b). This type of fatigue is known to impose adverse consequences on both adults and children. For example, research suggests that fatigue experienced by working adults, with or without HL, can negatively impact work performance and overall life quality (Deluca, 2005; Hetu, Riverin, Lalande, Getty, & St-Cyr, 1988; Kramer, Kapteyn, & Houtgast, 2006; Morata et al., 2005).

Likewise, research examining children with chronic health conditions (CHCs; e.g., cancer, cerebral palsy, rheumatic diseases, and chronic fatigue syndrome) suggests that this type of fatigue can impose adverse effects. For instance, compared to nonfatigued children, those suffering from fatigue are more likely to have increased school absences, reduced academic performance, and a reduced ability to engage in normal daily activities, all of which negatively affect quality of life (Gaba & Howard, 2002; Hockenberry-Eaton et al., 1999; Ravid, Afek, Suraiya, Shahar, & Pillar, 2009). Importantly, individuals with additional handicapping conditions, as are commonly found in CHL, are especially vulnerable to fatigue and its negative consequences (Hardy & Studenski, 2010). The significance of the fatigue experienced by CHL is highlighted when we compare their fatigue to that of children with other CHCs. The overall fatigue of CHL is similar to, or significantly greater than, that of children with most other CHCs known to affect fatigue (e.g., cancer, diabetes, rheumatoid arthritis, and inflammatory bowel disease; Hornsby et al., 2017).

Summarizing to this point, fatigue is a common phenomenon in our society, and the consequences of fatigue can be significant. Mounting evidence suggests that adults and CHL are at increased risk for listening effort and fatigue. Because of their hearing difficulties, a greater level of effort is required in order to listen attentively and understand speech in noisy conditions. The additional attention, concentration, and listening effort required to overcome auditory deficits associated with HL results in increased reports of listening-related fatigue. Moreover, as the listening effort and fatigue of CHL increases throughout the day, their impact on important cognitive abilities (e.g., listening, memorizing, attending) for processing/decoding speech and learning may also increase (Bess et al., 2014; Hornsby et al., 2016). The section below highlights some of the evidence related to fatigue in adults and CHL.

### ***HL and Listening-Related Fatigue in Adults***

Studies examining fatigue in individuals with HL are, in general, scant. Most of those that have been conducted

have concentrated on fatigue in the adult population (Alhanbali, Dawes, Lloyd, & Munro, 2017; Hetu et al., 1988; Hornsby & Kipp, 2016; Nachtegaal et al., 2009). Many of these studies focused on quality of life in the workplace using broad interviews and general surveys, as opposed to validated, fatigue-specific measures. However, interview and survey results suggested that adults with HL experienced more stress, a common antecedent of fatigue (Hockey, 1983) in the workplace, than their peers without HL, and such stress and fatigue negatively impacted work performance (Hasson, Theorell, Wallén, Leineweber, & Canlon, 2011; Kramer et al., 2006; Morata et al., 2005; Nachtegaal et al., 2009).

Hornsby and Kipp (2016) were one of the earliest to investigate fatigue and vigor<sup>1</sup> using a validated, fatigue-specific measure. They used the Profile of Mood States (McNair, Lorr, & Droppleman, 1971) to assess fatigue and vigor in a large sample of adults with mild-to-profound loss who presented to an audiology clinic seeking help for hearing difficulties. Their results showed that, compared to normative data, adults seeking help for hearing difficulties were more than twice as likely to experience severe fatigue and more than four times as likely to report severe deficits in energy.<sup>2</sup> Interestingly, regression analyses revealed no association between degree of HL and subjective ratings of fatigue or vigor. That is, adults with a mild HL or a profound loss were equally susceptible to increased fatigue and vigor deficits.

Alhanbali et al. (2017) used a different validated fatigue-specific measure, the Fatigue Assessment Scale (Michielsen, De Vries, Van Heck, Van de Vijver, & Sijsma, 2004), to replicate and expand on the findings of Hornsby and Kipp. They compared fatigue ratings from three different groups of adults with HL (UHL:  $n = 50$ ; bilateral hearing aid users:  $n = 50$ ; and cochlear implant users:  $n = 50$ ) to ratings from an age-matched control group without HL ( $n = 50$ ). All HL groups reported significantly more subjective fatigue than the age-matched control group. Importantly, fatigue ratings were similar for all three HL groups, regardless of whether they wore bilateral hearing aids, used a cochlear implant, or, relevant to this review article, had only UHL. Thus, research in adults suggests that HL of any kind (e.g., bilateral or unilateral losses, cochlear implant users or hearing aid users) may increase risk for fatigue. However, additional work is needed to ensure other factors, such as sample bias, are not driving this finding (see Hornsby & Kipp, 2016; Alhanbali et al., 2017, for discussions).

<sup>1</sup>Vigor, like fatigue, is a mood state. It is associated with a general feeling of being energetic, alert, and full of energy or pep. Although fatigue and vigor are often strongly and negatively correlated, validation studies suggest they are independent constructs, rather than a single bipolar factor (McNair & Heuchert, 2010; Stein, Jacobsen, Blanchard, & Thors, 2004; Stein, Martin, Hann, & Jacobsen, 1998).

<sup>2</sup>Severe fatigue and vigor deficits were defined as ratings that were 1.5 *SDs* or more above (high fatigue) or below (vigor deficit) the Profile of Mood States normative means.

## ***HL and Listening-Related Fatigue in Children***

In the past, much of what we knew about fatigue in CHL was based on anecdotal reports and pilot studies. In recent years, however, we have learned much more about fatigue in pediatric HL. Similar to adults, there is a growing body of research showing that sustained listening demands experienced by CHL put them at increased risk for listening-related fatigue and its adverse consequences. The studies to follow focus on a review of subjective fatigue in children with bilateral HL and children with UHL.

### **Bilateral HL**

One of the earliest studies to explore fatigue in CHL was Bess, Dodd-Murphy, and Parker (1998). These investigators conducted a clinical-based study on children with minimal HL—minimal HL included three categories of HL (unilateral sensorineural HL, minimal bilateral sensorineural HL, and high-frequency sensorineural HL; for definitions, see CDC, 2005). One component of this study was an analysis of functional health status in school-age children (Grades 6 and 9) using the COOP Adolescent Chart Method (COOP; Nelson et al., 1987). The COOP charts measure functional status on a core set of functional dimensions—physical, emotional, and social. Children with minimal sensorineural HL exhibited more dysfunction than normal controls on many of the subtests—especially stress and energy, two constructs closely associated with fatigue.

In a pilot study, Hornsby et al. (2014) assessed fatigue in a small group ( $n = 10$ ) of school-age CHL and an age-matched group ( $n = 10$ ) of children without HL using a generic, validated subjective measure of fatigue, the Pediatric Quality of Life Inventory Multidimensional Fatigue Scale (PedsQL-MFS; Varni, Burwinkle, Katz, Meeske, & Dickinson, 2002; Varni, Burwinkle, & Szer, 2004). The PedsQL-MFS is a self-report scale designed to assess three different fatigue domains (cognitive, sleep/rest, and general) and has been commonly used to assess fatigue in children with a variety of CHCs. Hornsby and coworkers found that a small group of school-age CHL reported more fatigue than an age-matched control group of children without HL across all three domains—although the differences between groups were not significant for the cognitive domain. The severity of the fatigue in CHL, when compared to existing work in the literature, was equivalent to or greater than fatigue ratings reported by children with other CHCs.

In a follow-up to the aforementioned pilot study, Hornsby et al. (2017) collected PedsQL-MFS data on a much larger population of school-age CHL ( $n = 60$ ) with mild to moderately severe hearing losses. Moreover, in addition to obtaining data from the children, parent proxy reports were also obtained. These data were compared to reports from a control group ( $n = 43$ ) of children without HL and their parent proxies. Consistent with outcomes from the pilot study, results showed that CHL experienced significantly more overall fatigue and cognitive fatigue than children without HL. Also, consistent with results from the adult population (Hornsby & Kipp, 2016), no

association between degree of HL and fatigue ratings was observed, suggesting children with even mild hearing losses were as equally susceptible to fatigue as children with more moderately severe losses. In addition, highlighting the potential importance of increased fatigue in CHL, significant associations between language ability and ratings of cognitive fatigue were observed in that study. Werfel and Hendricks (2016) reported a similar finding in children with profound HL who used cochlear implants. These authors found that children with cochlear implants (Grades 3–6,  $N = 19$ ) reported PedsQL-MFS ratings similar to those of the CHL from the study of Hornsby et al. (2014). Importantly, they also found that the increased fatigue ratings were associated with poorer language and literacy skills in their CHL. Whether the increased fatigue experienced by CHL leads to language and literacy deficits or vice versa remains an unknown, but important, research question.

### **UHL**

Although information on fatigue in children with UHL is limited, pilot studies and reports from parents, clinicians, and teachers offer indirect evidence that children with UHL could be at increased risk for fatigue, such as their peers with bilateral HL. For example, in the 1998 study by Bess and coworkers described earlier, COOP results (particularly on the stress and energy subtests) showed that CHL experienced more dysfunction. Although the study did not parse the scores for each HL group, it is noteworthy that more than half of the participants that completed the COOP had UHL. Additional support can be found in the study by Hornsby et al. (2014), in which the PedsQL-MFS was administered to a small diverse group of CHL ( $n = 10$ ) and an age-matched control group. Two of the CHL in that study had UHL, and their fatigue ratings were among the most severe of any child participant.

Recently, our ongoing research on listening-related fatigue has led us to hold interviews and focus groups that included parents of children with UHL and their teachers and clinicians to discuss issues around listening-related fatigue. Reports from these stakeholders have been insightful. For example, a parent of a child with UHL stated, “My daughter is exhausted most days after school or when she has to listen for a long time.” An educational audiologist offered these comments: “Our kids with UHL loss are similar to children with mild to moderate hearing losses—they require auditory breaks throughout the day and struggle more academically than one would expect given their hearing loss.” Comments such as these from parents and professionals offer additional support for the premise that children with UHL may become fatigued following sustained listening demands in school—and such fatigue may impact on school performance.

### **A Preliminary Examination of Listening-Related Fatigue in CHL as Reported by the Child, Parent, and Teacher**

Given the scarcity of information related to fatigue in children with UHL, we use self-report data in this section to conduct a preliminary exploration of listening-related



fatigue in children with unilateral and bilateral HL. The self-report data were collected as part of our ongoing work developing a scale to assess listening-related fatigue in children. Some of the information reported here is based on a conference poster (Hornsby, Davis, Cho, & Bess, 2018).

### The VFS: A Tool for Measuring Listening-Related Fatigue

Although nearing completion, the VFS is still being developed, and as such, we only briefly describe here the development process of the scales to date. Currently, we are conducting validity studies on three versions of the VFS—a child, a parent, and a teacher version. To develop these scales, we used information from interviews/focus groups (separate groups of children, parents, and teachers participated) to create a large pool of potential test items (157–212 items depending on the target respondent). Using best practices and an iterative process, including factor analyses, classical test theory, and item response theory (IRT), we evaluated item quality to reduce the total number of items down to eight to 12, depending on the scale. Factor analyses revealed a unidimensional structure for the child and teacher scales. However, the parent scale was best fit as a two-factor model consisting of a physical factor and a social-emotional-cognitive factor. IRT analyses confirmed all scale items were high quality (i.e., all items had high information and discriminability). The parent version contains 12 items, the teacher version has eight items, and the child version consists of 10 items. All instruments use a 5-point Likert scale to assess the frequency (1 = *never* to 5 = *almost always*) of fatigue-related complaints. Examples of items included in all three versions of the VFS and an adult version (VFS-A) are shown in Table 1. All scales allow for IRT scoring and using summed Likert-scale scores. In this review article, scale results will be described using IRT, rather than summed scores. An IRT analysis and scoring approach has several advantages compared to more traditional scale assessment methods (see Hambleton & Swaminathan, 2013, for a review). Importantly, an IRT analysis method takes the sensitivity of each test item into account in the scoring process, providing a more precise estimate of the underlying latent construct being measured—in our case, listening-related fatigue (Embretson & Reise, 2000). IRT scores obtained using the VFS are standardized scores (like *z* scores)

that show the severity of listening-related fatigue for a given individual. Thus, an IRT score of 0 reflects a score equal to the mean score across all scale respondents (i.e., across all HL and no HL groups). In contrast, an IRT score of –3 and +3 reflects very low and very high ratings of listening-related fatigue, respectively.

### VFS Data Collection and Participants

Potential participants were recruited in person following routine audiological appointments at our clinic as well as via social media and other online recruitment portals. As a result, data collection was completed using online versions of the VFS and in person using hard copies of the scales. Parents and teachers completed the scales themselves. Children also completed the scale themselves when possible, although for younger children (< 10 years old) or any child who had questions or needed assistance, the parent (or research assistant if present) was available to read the scale items aloud to the child and answer any questions. Data for all three versions of the VFS were collected, stored, and managed using Research Electronic Data Capture (Harris et al., 2009). Research Electronic Data Capture is a secure, web-based software platform designed to support data capture for research studies. All procedures were reviewed and approved by Vanderbilt’s Institutional Review Board, and all participants provided informed consent or assent prior to starting any study procedure.

Participant responses were obtained from (a) 399 parents of children (aged 6–17 years) with (*n* = 263) and without (*n* = 136) HL, (b) 137 children (aged 10–17 years) with (*n* = 98) and without (*n* = 39) HL, and (c) 363 teachers of students (aged 6–17 years) with (*n* = 289) and without (*n* = 70) HL. Not all parents of the 137 child respondents completed a parent version of the scale. Likewise, some children of parents who completed the scale did not complete a child version of the scale. In this review article, we do not investigate parent proxy and child agreement or agreement between teacher responses and parent or child. Those topics will be explored in future works. The child’s HL was self-reported by the parent and teacher as unilateral or bilateral. Degree of loss was self-reported by the child’s parent as mild/slight, moderate, severe, or profound based on their perceived speech understanding (World Health Organization, 2019). A detailed demographic description of the participants is provided in Table 2.

**Table 1.** Examples of questionnaire items from the Vanderbilt Fatigue Scale (VFS) for listening-related fatigue in children.

VFS-A: Adult items	VFS-C: Child items
<ul style="list-style-type: none"> <li>I feel worn out from everyday listening.</li> <li>It takes a lot of energy to listen and understand.</li> </ul>	<ul style="list-style-type: none"> <li>After school, I’m so tired I don’t want to talk to anyone.</li> <li>Listening at school wears me out.</li> </ul>
VFS-P: Parent items	VFS-T: Teacher items
<ul style="list-style-type: none"> <li>It is hard for my child to concentrate for a long time.</li> <li>My child is completely worn out after listening for a long time.</li> </ul>	<ul style="list-style-type: none"> <li>When the student gets tired from listening, he or she “checks out.”</li> <li>The student appears worn out after working hard to listen all day.</li> </ul>

*Note.* The three versions of the VFS for children include a child self-report (VFS-C), a parent proxy report (VFS-P), and a teacher proxy report (VFS-T). Samples from the adult version of the Vanderbilt Fatigue Scale (VFS-A; Hornsby et al., 2018) are provided as a comparison. Responses are on a 5-point Likert scale from *never* to *almost always*.

**Table 2.** Participant characteristics for respondents completing the various versions of the Vanderbilt Fatigue Scale for children.

Variable	Parent respondents	Child respondents	Teacher respondents
No. of participants	399	137	363
Male/female/NR	209/189/1	73/64/0	186/169/8
Age (years)	10.7 (3.4)	13.6 (1.9)	10.4 (3.3)
HL/no HL/NR	263/136/0	98/39/0	289/70/4
UHL/BHL	64/199	28/70	40/249

Note. Gender, mean age (1 SD), and hearing status of children being reported on are provided for all three participant groups. NR = not reported; HL = hearing loss; UHL = unilateral hearing loss; BHL = bilateral hearing loss.

### VFS Initial Analyses and Results

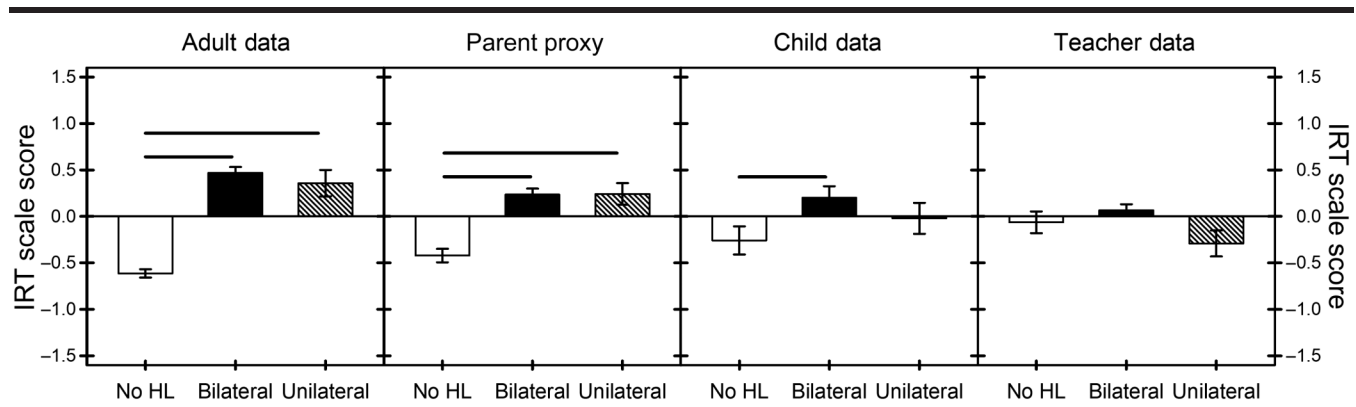
It is important to note that the data used in the analyses described below were not collected for the purpose of examining the effects of unilateral or bilateral HL on listening-related fatigue. Rather, these data were collected to assist in developing a valid, reliable scale for assessing listening-related fatigue. Thus, the analyses described below are exploratory in nature. Nonetheless, given the paucity of data related to fatigue in CHL, we felt such analyses would be of interest to clinicians and could provide useful information to guide future research in this area. We used a single-factor analysis of variance (ANOVA) to compare IRT ratings of listening-related fatigue in children with self-reported bilateral and UHL to a control group of children with no HL. Separate ANOVAs were conducted for each respondent group—parent, teacher, and children. In addition, for the two-factor structure for the parent scale, an “overall” fatigue score was created by averaging the IRT scores of each factor. Separate analyses were conducted on each factor score and on the overall fatigue score. The statistical findings were the same for each analysis method, so only the results for overall (average) fatigue are reported for the parent data.

Figure 1 shows the mean IRT scaled scores for those children with (bilateral and unilateral) and without HL as

reported by each respondent group (parents, children, and teachers). The mean IRT score for a group of adults with HL, based on results using an adult version of our listening-related fatigue scale, the VFS-A, is also provided as a comparison (Hornsby et al., 2018).

Among parent respondents, there was a significant main effect of group ( $F = 24.5, p < .001$ ). Post hoc testing, using a series of Bonferroni-adjusted  $t$  tests, revealed significantly higher IRT scores (higher fatigue) for both HL groups compared to the control group without HL ( $t = -6.6, p_{adj} < .001$  and  $t = -4.9, p_{adj} < .001$  for the bilateral and UHL groups, respectively). Interestingly, parent ratings for children with unilateral and bilateral HL were quite similar in magnitude and not significantly different ( $t = 0.019, p_{adj} > .05$ ). A significant main effect of group was also observed for the child ratings ( $F = 3.27, p < .05$ ). As with the parent data, post hoc testing revealed that the bilateral group reported significantly higher levels of fatigue than the control participants ( $t = -2.5, p_{adj} < .05$ ). However, the fatigue scores for children with UHL fell between those of the bilateral HL group and the control group of children without HL. Listening-related fatigue scores for children with UHL were not significantly different than either the control group ( $t = -0.9, p_{adj} > .05$ ) or the children with bilateral HL ( $t = -1.36, p_{adj} > .05$ ).

**Figure 1.** Mean item response theory (IRT) scale scores of respondents with self-reported unilateral, bilateral, and no hearing loss (HL) obtained using three versions of the Vanderbilt Fatigue Scale (VFS) for assessing listening-related fatigue in children (VFS-C, VFS-P, VFS-T). The mean scale scores for adults were obtained using the Vanderbilt Fatigue Scale for Adults (Hornsby et al., 2018) and are provided for comparison. Solid lines over the bars reflect significant differences between groups.



Finally, an analysis of teacher ratings also revealed a significant main effect of group ( $F = 3.52, p < .05$ ). However, post hoc testing using a series of Bonferroni-adjusted  $t$  tests revealed no significant differences between any of the groups (all  $p_{adj}$  values  $> .05$ ). The main effect appeared to be driven by the difference in the teachers' fatigue ratings for children with bilateral and UHL, not differences between CHL and the control group. As seen in Figure 1, the mean fatigue ratings from teachers for children with UHL were lower than that of the children without HL (control group) and the children with bilateral HL. We discuss this unexpected finding in more detail later in the review article.

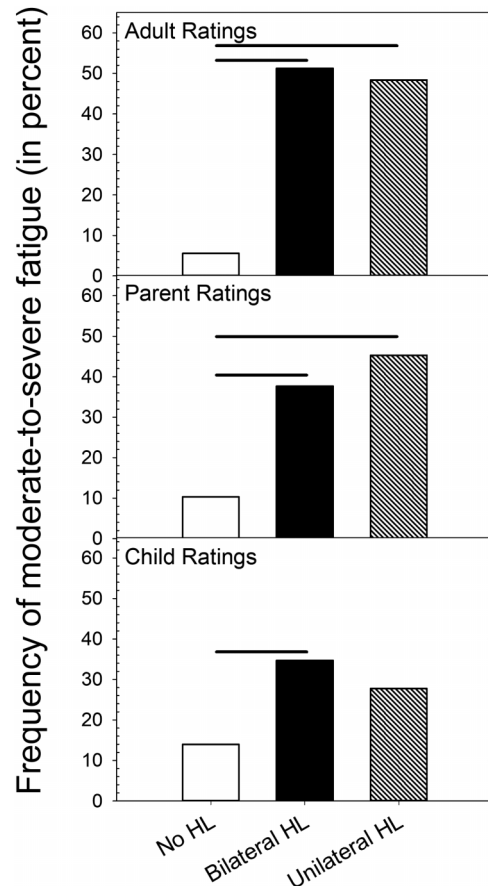
Additionally, recall that Hornsby and Kipp (2016) found that adults seeking help for HL were much more likely to report severe fatigue and vigor deficits than expected based on normative data. To explore the issue of susceptibility to more severe fatigue in children, we used an IRT score of  $> 0.5$  as a cut-point to operationally define "moderate-to-severe" listening-related fatigue. We then compared the prevalence of moderate-to-severe fatigue in children with and without HL across respondent groups. To do so, we calculated the percentage of individuals with and without HL in each respondent group who reported moderate-to-severe listening-related fatigue. Although the use of 0.5 as a cut-point is arbitrary, for our purposes, it seems a reasonable estimate because only about 31% of the population would be expected to report greater fatigue problems.

Results for adults, parents (IRT score averaged over Factors 1 and 2), and children are shown in Figure 2. The teacher data are not plotted as, similar to our mean results, they showed no significant difference between groups. A series of chi-square analyses (see Table 3) showed that moderate-to-severe listening-related fatigue is much more common in adults and children with unilateral or bilateral HL when compared to those without HL. More specifically, adults with bilateral and UHL were significantly (approximately nine times) more likely to report moderate-to-severe fatigue than adults without HL. A similar significant finding, although smaller in magnitude, was observed for the parent proxy respondents. Parents of children with bilateral or UHL were approximately four times more likely to report their children experienced moderate-to-severe fatigue than parents of children without HL. Finally, children with bilateral and UHL were approximately 2.5 and 2.0 times more likely, respectively, to report moderate-to-severe fatigue than children without HL—although the difference for the children with UHL did not reach statistical significance. Importantly, there were no significant differences in relative risk for moderate-to-severe fatigue between children with unilateral and bilateral HL (see Table 3).

## Discussion

A primary goal of this review article was to introduce the construct of listening-related fatigue and highlight its importance for CHL, with a particular focus on children

**Figure 2.** Percentage of children with unilateral and bilateral hearing loss (HL) exhibiting moderate-to-severe fatigue (item response theory scores  $> 0.5$ ) as compared to a group of children without HL. Solid lines over the bars reflect significant differences between groups.



with UHL. To do so, we reviewed and discussed existing work in this area. Our review revealed that systematic research on fatigue in children with UHL is scant. The most relevant information comes from studies examining listening-related fatigue in children and adults with bilateral HL. While limited in scope, some research examining listening-related fatigue in adults with UHL and general fatigue in children with other CHCs also provided relevant information.

Given the limited work in this area, we also included preliminary analyses of self-reported listening-related fatigue in CHL, including UHL. Data for these analyses were collected as part of a series of studies to develop and validate a suite of scales for assessing listening-related fatigue (the VFS) in children. Considering the available literature and our self-report fatigue data leads us to conclude that children with unilateral and bilateral HL are at increased risk for listening-related fatigue. Moreover, the magnitude of fatigue observed in children with UHL appears to be similar to that experienced by children with bilateral HL—again highlighting the significant impact of UHL in children. This review article is the first to address the topic of subjective

**Table 3.** Chi-square analyses of prevalence of “moderate-to-severe” listening-related fatigue among our hearing loss and no hearing loss groups as well as between respondents with bilateral and unilateral hearing loss.

Variable	$\chi^2$	<i>p</i>	Prevalence ratio
<b>Adults with no HL vs. bilateral HL</b>	<b>106.1</b>	<b>&lt; .001</b>	<b>9.2 [5.1, 16.6]</b>
<b>Adults with no HL vs. unilateral HL</b>	<b>48.8</b>	<b>&lt; .001</b>	<b>8.7 [4.4, 17.2]</b>
Adults with bilateral vs. unilateral HL	0.1	.76	0.9 [0.6, 1.4]
<b>CNHL vs. CBHL</b>	<b>5.9</b>	<b>&lt; .05</b>	<b>2.5 [1.1, 5.6]</b>
CNHL vs. CUHL	2.3	.13	2.0 [0.8, 5.0]
CBHL vs. CUHL	0.5	.47	0.8 [0.4, 1.5]
<b>Parents of CNHL vs. parents of CBHL</b>	<b>32.1</b>	<b>&lt; .001</b>	<b>3.8 [2.2, 6.4]</b>
<b>Parents of CNHL vs. parents of CUHL</b>	<b>31.1</b>	<b>&lt; .001</b>	<b>4.4 [2.5, 7.9]</b>
Parents of CBHL vs. parents of CUHL	1.2	.28	1.2 [0.9, 1.7]

*Note.* Responses were obtained from adults, children, and their parents as proxy reporters. Values in [ ] show confidence intervals around the prevalence ratio. Bolded values show significant differences. HL = hearing loss; CNHL = children with no hearing loss; CBHL = children with bilateral hearing loss; CUHL = children with unilateral hearing loss.

fatigue in children with UHL. Clearly, additional work examining this complex, multifaceted construct and its relationship to HL is needed.

One important area of inquiry is to better understand why our current teacher data were not sensitive to the effects of HL on listening-related fatigue in school-age children (see Figure 1). This discrepancy could simply suggest that teachers are not sufficiently aware of the signs and symptoms of listening-related fatigue in CHL. Such an explanation, however, is inconsistent with anecdotal reports and comments made during our focus groups with teachers, educators, and support staff who work with CHL. In these discussions, reports of the substantial difficulties with listening-related fatigue among CHL were common—hence, the underlying reason for the discrepancy remains unclear.

Although focus group comments suggest otherwise, our reduced ability to detect the effects of HL in our teacher data could reflect a limited insight of this group into the fatigue of their students. Recall that teachers must provide proxy ratings of a child’s fatigue. As such, their ability to do so may be limited by how much they interact with the child during the day. Unlike parents, the teacher may only see the child for a limited period of time each day, potentially limiting their insight into the fatigue of a child. If this were the case, we might expect a repeated-measures analysis, in which the same teacher rates the fatigue of a child with and without HL, would be better suited to detecting between-groups differences should they exist.

Unfortunately, only two teachers provided VFS ratings for a child with and without HL as part of the validation data collection described above. Both teachers reported on children with bilateral HL, and those outcomes were mixed with one teacher reporting greater fatigue for the child with HL and the other reporting the opposite. However, we were able to test this hypothesis using data from an earlier version of our scale that was obtained from a subset of teachers who provided ratings for both children with and without HL ( $n = 21$ ). This early version of the scale had a total of 60 items. Although the scale included the 10 items used in our current teacher version of the VFS, IRT scores

used in this analysis were based on responses from all 60 items. In contrast to our initial analysis, a single-factor ANOVA on this small subset of data (comparing a given teachers IRT score for a child with and without HL) revealed a significant main effect of group ( $F = 10.7$ ,  $p < .05$ ). The mean IRT scores provided by teachers for the CHL ( $-0.10$ ) were significantly higher than that of the children without HL ( $-0.71$ ). In fact, the teacher IRT scores for CHL were higher in 16 of the 21 cases.

The result above is encouraging and suggests that teachers may be sensitive to the problems of listening-related fatigue in CHL, at least when comparing children with and without HL within their own classroom. However, additional research is clearly needed. For example, it is possible that the format of the current scale may not be optimal for teacher use. Scale modifications, such as specifically instructing respondents to first consider how they might respond if they were rating a typically developing child prior to completing the scale for an at-risk child (e.g., one with HL), may enhance the scale’s ability to identify those at most risk.

Alternatively, factors other than the scale itself could also have influenced our outcomes. Recall that our teacher data were collected for the purpose of identifying high-quality scale items and validating a final version of the scale—not to empirically examine the effects of HL on self-reported fatigue. To this end, we specifically cast a wide recruitment net with a goal of including respondents with a wide range of fatigue ratings. It is possible that the sample of children rated by teachers had additional factors, other than HL, that may have affected their fatigue (e.g., some other disability)—potentially masking the effects of HL. To be certain, we need to develop a better understanding of the factors responsible for the apparent limited sensitivity of the teacher scale.

#### **Identification and Management of Fatigue in CHL**

Given that children with UHL and bilateral HL are at increased risk for listening-related fatigue, an increasingly



important role for pediatric and educational audiologists will be the identification and management of listening effort and fatigue in school-age children with UHL as well as those with bilateral HL. School-age CHL are often eligible for specialized instruction under the Individuals with Disabilities Education Act (Johnson & Seaton, 2012).

If a child does not qualify for an Individualized Education Program (IEP), they may be considered for a 504 plan under the Section 504 of the Rehabilitation Act of 1973, an antidiscrimination civil rights statute that requires that the needs of students with disabilities be met as adequately as the needs of children without disabilities (Zirkel, 2009). Both plans require education team members to holistically review a child's performance in the classroom and consider potential accommodations to provide equal communication access to classroom instruction.

As a part of specialized instruction, a team of professionals (educational audiologists, certified teachers of the hearing impaired, and speech-language pathologists) work together with CHL and their families to support academic success. They collaborate to determine whether the child should receive preferential seating, is meeting auditory goals in their IEP, and is appropriately utilizing hearing-assistive technology, if recommended. In addition, the team reviews the child's educational progress periodically and considers additional factors that may be impacting educational progress.

In such reviews, the concept of fatigue is frequently discussed and often considered in the educational programming of CHL; however, systematic identification and management protocols are not available. The most logical way to determine a child's risk for fatigue is for educational audiologists, speech-language pathologists, and teachers to look for symptoms and behaviors commonly associated with fatigue in children. Our results suggest that teachers and support staff may need to be trained to identify prominent characteristics of listening-related fatigue (see Fatigue Education section below). Such characteristics would include reports or behaviors indicative of tiredness/weariness, mood changes, sleepiness (especially in the morning), a decrease in stamina or energy, or a lack of desire to continue with a task. If fatigue is suspected for a given child, the audiologist or speech-language pathologist would administer a validated fatigue measure to the child and/or parent and teacher. This type of measure could also be incorporated into existing (or new) self-advocacy competency checklists to be used during IEP/504 meetings. Examples of fatigue scales for children include the PedsQL-MFS (Varni et al., 2002), the Childhood Fatigue Scale (Hockenberry et al., 2003), and the soon-to-be-available VFS, which specifically assesses listening-related fatigue. These standardized measures provide support for anecdotal evidence that a CHL may be struggling with fatigue in the classroom setting, an important factor when determining specialized services in the school setting. While this is a logical approach, additional research is needed to evaluate the sensitivity and benefits of such an approach to identification of listening-related fatigue in children.

## Suggestions for Fatigue Interventions

Although evidence-based intervention protocols are not presently available, some obvious common sense strategies seem appropriate for implementation in the school setting. Importantly, given the risk for fatigue appears to be similar for those with unilateral or bilateral HL, the suggested intervention/management strategies that appear below could be appropriate for all CHL. Many of these strategies were suggested and discussed during our focus groups with teachers and other school service providers.

## Classroom Strategies

The classroom is often a difficult listening environment for CHL, as the spaces do not meet recommended standards for noise levels and reverberation times (Grep & Easterbrooks, 2018; Knecht, Nelson, White, & Feth, 2002). Even when utilizing properly fitted amplification, CHL may still struggle in this setting. CHL may struggle to listen and comprehend in noisy settings and have difficulty localizing talkers during group discussions.

Our focus group discussions with teachers who work with CHL revealed that the educational team may consider several anecdotal interventions when a child with hearing loss demonstrates behaviors they believe to be fatigue related. For example, focus group participants noted that CHL tend to be "more zoned out" compared to typical hearing peers, especially "toward the end of the school day." Hence, it was not surprising to learn that many teachers and clinicians reported arranging their caseload schedules to accommodate CHL in the morning to reduce the possible effects of fatigue on a child's therapy sessions. To help CHL cope with fatigue, teachers reported interventions, such as allowing the child to remove their hearing-assistive technology for short listening breaks or scheduling consistent "movement breaks," where the child could move and stretch, into the routine of the classroom.

Most CHL are provided with flexible, preferential seating (i.e., seating that allows for the best access to both visual and auditory information) in the classroom setting. For children with UHL, preferential seating may need to be reviewed based on the activity of the classroom; however, the normal hearing ear should typically be faced toward the speaker of interest.

## Listening Breaks

To our knowledge, there is no research specifically focused on using listening or rest breaks to reduce fatigue in school-age CHL. Despite this absence, discussions among our focus group participants suggest it is a common strategy of clinicians and teachers who work with CHL. While research on rest breaks in CHL is nonexistent, there is a large related literature from studies of driving behavior, flight simulation, and workforce, suggesting rest breaks are an effective tool for reducing fatigue and its negative effects. In the workforce, for example, rest breaks are important for increasing performance, avoiding accidents, and promoting good health (Fritz, Lam, & Spreitzer, 2011). Many studies have reported that rest breaks, especially short rest breaks,

have many positive benefits, including decreases in fatigue, stress, and discomfort and increases in vitality and time spent on task (Arlinghaus et al., 2012; Henning, Jacques, Kissel, Sullivan, & Alteras-Webb, 1997; Tucker, 2003; Zacher, Brailsford, & Parker, 2014). For example, Zacher et al. (2014) studied the impact of microbreaks on fatigue and vitality in a group of working adults ( $N = 124$ ). Using a diary study design, they showed that micro breaks resulted in a decrease in fatigue and an increase in vitality.

Consistent with this related research, some teachers report that allowing a child a “listening break”—the opportunity to step away from the classroom for a few minutes to go to the restroom or rest in another teacher’s classroom—allows the child to come back to the activity refreshed and prepared for re-engagement. However, there is no consensus concerning the number of rest breaks or the duration of breaks that should be used. It is anticipated that the recovery time needed by a student may vary, based on the child’s age, additional disabilities, or other factors. Furthermore, there is no validated method for teachers and clinicians to know when a child requires intervention or how to systematically provide the intervention.

### **Fatigue Education**

Although fatigue and its negative effects are pervasive, not all individuals living or working with CHL are aware of the potential side effects that sustained fatigue can have on an individual’s overall well-being. In addition, our interview data with CHL suggest that many (even middle and high schoolers) are unaware of their presentation of common fatigue symptoms as compared to their peers. In focus groups and interviews, some teenagers denied any fatigue-related problems, while their parents, who also participated in separate focus groups, reported significant difficulties with listening-related fatigue in their CHL. One child reported “never feeling worn out from listening” while her mother shared that this child often “came home from school, took off her implants,” and “laid down for 30 minutes or so with the lights off.” A young adult with HL shared that it was not “until the caseload of college” that she was able to look back and recognize how much listening-related fatigue affected her through her adolescent years.

These findings suggest that additional education about listening-related fatigue and its negative effects is required to inform and equip CHL, parents, and school personnel working with this population (i.e., teachers and therapists). Structured educational programs that target the identification of common symptoms associated with fatigue and provide guidelines for identification and management, including intervention strategies, are likely to improve outcomes for CHL. Additionally, school professionals, audiologists, and parents should work with CHL to help them identify symptoms associated with listening-related fatigue and self-advocate for their listening needs.

### **Hearing Devices and Assistive Technology**

CHL are typically prescribed personal amplification devices (i.e., hearing aids, cochlear implants, bone-anchored

devices) to provide access to auditory information. For children with UHL, these amplification options as well as a contralateral routing of signal may be recommended. Personal devices aim to improve audibility so that a child is better able to hear and communicate with those around him. Clinical audiologists often make the recommendation for “full-time use of amplification,” which, for a typical school-age child, would range from approximately 10 to 14 hr per day (Walker et al., 2013). Some clinicians and teachers believe that well-fitted amplification helps to reduce fatigue effects due to a long day of listening in the classroom. However, evidence supporting such an assumption is limited. Hornsby (2013) is the only study to date to examine this empirically, and participants in that study were adults. Participants were required to complete a cognitively demanding listening task for approximately 60 min without a break. Testing was completed with and without hearing aids. Results showed a small benefit of hearing aids for reducing listening effort and mental fatigue—defined as a decrement in cognitive processing speed over time. Specifically, when wearing hearing aids, participants were better able to maintain vigilant attention during the sustained listening task. However, subjective ratings of effort and fatigue were not significantly different in the unaided and aided conditions.

In addition, recent evidence from focus groups and interviews with adults suggests that extended device use itself can be fatiguing (Holman, Drummond, Hughes, & Naylor, 2019; Hughes, Hutchings, Rapport, McMahon, & Boisvert, 2018). Similar comments were made by some CHL during focus group and interviews conducted as part of our fatigue scale development project. The mixed findings related to benefits from device use may be due in part to the limitations of hearing aids and cochlear implants alone in noisy environments. Moreover, not all CHL wear their aids while attending school—24% of children (6.5–12.9 years old) with mild-to-moderate losses do not use their personal devices on a day-to-day basis (Gustafson, Davis, Hornsby, & Bess, 2015).

In addition to personal hearing aids and cochlear implants, access to important classroom information can be achieved by using a personal remote microphone (RM) system. RM systems effectively improves the signal-to-noise ratio and allows the child to hear the teacher’s voice more clearly in the presence of excessive classroom background noise (Wolfe, Morais, Schafer, Agrawal, & Koch, 2015). The benefits of RM technology for CHL are well recognized, and these systems are widely accepted in the educational setting (Anderson & Goldstein, 2004). In contrast, research examining the benefits of RM systems for reducing fatigue is limited. However, a recent study by Rance, Saunders, Carew, Johansson, and Tan (2014) suggests that RM systems may help to reduce stress associated with difficult listening conditions, thus potentially reducing fatigue. In addition, some professionals working with CHL in the school setting report observing a decrease in fatigue symptoms when the student uses an RM system in the classroom; however, systematic research in support of these accounts is lacking.

In summary, based on our interpretation of the existing literature and comments from focus group and interview participants, we believe the intervention options discussed here provide a reasonable starting point for parents and professionals concerned about listening-related fatigue in CHL. However, given the limited research in this area, the optimal intervention remains unknown and may well depend on the root cause of the fatigue.

## Conclusions and Future Directions

We have reviewed some of the pertinent literature on subjective fatigue in adults with unilateral and bilateral HL and children with bilateral HL. Also, we have described anecdotal and preliminary/pilot evidence of listening-related fatigue in children with UHL. Finally, we examined our own data from children with unilateral and bilateral HL that were collected as part of the development and validation of a listening-related fatigue measure—the VFS. The VFS is composed of the parent, child, and teacher versions. We used the three scales to assess listening-related fatigue in children with unilateral and bilateral HL and children without HL.

Overall, our findings suggest that listening-related fatigue in children can be reliably measured using these scales. Analyses of VFS data from children with and without HL reveal that children with UHL are at increased risk for listening-related fatigue and that their fatigue is similar to that of children with bilateral HL. Also, a significantly larger percentage of children with unilateral and bilateral HL reported moderate-to-severe fatigue, compared to children without HL. However, the substantial overlap in IRT scores of child respondents with and without HL suggests that, compared to adults and parent proxy report, children may be less able to identify and describe their own fatigue. In addition, in contrast to data from adults, parents, or children, data obtained using the teacher scale were not sensitive to the effects of HL.

It is important to emphasize here the need for caution in the interpretation of the data taken from our validation studies. Recall that these data were generated for the purpose of developing a fatigue scale using conventional methodology for health outcome measures—the data were not intended for investigating the impact of unilateral and bilateral HL on listening-related fatigue. Nevertheless, these data are useful for prefatory examination of subjective fatigue in children with UHL. Additional systematic research on fatigue effects in all CHL, however, is paramount.

An important area of future study is to develop a better understanding of subjective fatigue as a function of age, degree and type of HL, presence of disabilities, parent-child and teacher-child differences on our self-report scales, and the responsiveness of our scales to fatigue reduction. A need also exists for exploring the relationships between cognitive fatigue, language, and literacy skills in CHL (see Camarata, Werfel, Davis, Hornsby, & Bess, 2018). Finally, it is essential for us to explore evidence-based intervention strategies for reducing the debilitating effects of recurrent

fatigue in all CHL. To this end, more research is needed to determine the contributions provided by amplification devices (e.g., hearing aids, RM technology, cochlear implants) for reducing listening-related fatigue in CHL. The importance of an evidence-based and validated intervention to support our teachers, clinicians, and children with all forms of HL is clear.

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