Unilateral Hearing Loss in Youth: Development of Candidate Items for a Condition-Specific Validated Instrument

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Abstract

Objective. This study interviewed youth with unilateral hearing loss, utilizing their responses to generate candidate items for a condition-specific patient-reported instrument.

Study Design. Mixed methods, cross-sectional.


Participants and Methods. Youth with unilateral hearing loss and normal hearing in the contralateral ear were identified and recruited for participation through query of an audometric database and through hearing loss clinics. Interviews with the youth were qualitatively analyzed to identify common themes and generate items related to functional impact. A multi-institutional expert panel reviewed items with prespecified item selection criteria. Participants rated items for impact on daily life. For preliminary criterion validity assessment, statistical analyses explored correlations between functional scores and type and severity of hearing loss.

Results. Thirty-nine youth aged 9 to 18 years with unilateral hearing loss participated; 31% used a hearing device. Fifteen youth participated in interviews; thematic analysis, item crafting, and expert panel item review resulted in 41 items. Twenty-six youth responded to the items, reporting low functional scores in the domains of sound localization, ear positioning, and noise environment. They reported better levels of function in carrying out group conversations, focusing on schoolwork, and feeling safe during activities. Multivariate linear regression found that youth scored 0.4 points (or approximately 8%) lower on the functional impact scale with every 20–dB HL increase in pure tone average in the abnormal ear.

Conclusion. Youth with unilateral hearing loss report functional impact, particularly related to sound localization, ear positioning, and noise environment; therefore, they may benefit from a condition-specific functional assessment instrument.

Keywords

pediatric hearing loss, unilateral hearing loss, outcomes research, functional assessment, hearing rehabilitation

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Unilateral hearing loss (UHL) is estimated to affect at least 1 out of every 2000 newborns and between 1% and 5% of school-age children and adolescents. Individuals with UHL were found to experience difficulty with sound localization and understanding speech in noisy environments, but questions remain regarding the degree of functional impact and the role of hearing rehabilitation. Children with UHL typically communicate with spoken language, participate in general education classrooms, and may perform well in certain environments; therefore, teachers, caregivers, and health care providers may not be aware of the impact of hearing loss on children’s daily lives. A recent meta-analysis evaluating impact

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of pediatric hearing loss on quality of life found a clinically and statistically significant disparity in responses of children with UHL and children with normal hearing on the social and school domains of the PedsQL, one of the most commonly used generic quality-of-life instruments.\(^4\)

There is growing interest in the role of hearing rehabilitation for children with UHL, but there is no consensus regarding the best method for management.\(^5,6\) Some studies suggested that hearing devices provide improvement on audiometric testing measures, such as speech perception and speech recognition in noise\(^7\) and, potentially, quality of life.\(^8\) However, such studies are often small, and there is persistent uncertainty in terms of how youth with UHL can optimally benefit from amplification. A major limitation in attempts to evaluate the effectiveness of hearing devices is the lack of consensus regarding outcomes that should be measured. Audiometric testing results do not fully capture a patient’s experience with a hearing device on a day-to-day basis, and testing methods for symmetric, bilateral hearing loss may not apply well to individuals with unilateral deficit.

For these reasons, a functional assessment instrument for youth with UHL may provide useful information for hearing health providers. This study used face-to-face interviews and functional impact rating scores to better understand the impact of UHL as perceived by youth themselves. It is envisioned as a preliminary step in producing a validated condition-specific functional assessment instrument for youth with UHL based on self-report.

**Methods**

**Identification and Recruitment of Study Participants**

Institutional Review Board approval for this study was obtained from Seattle Children’s Hospital. The hospital’s audiometric database was queried to identify all children who had an audiogram between January 2007 and July 2014 and who had abnormal hearing in 1 ear, as defined by air conduction 4-frequency (500, 1000, 2000, and 4000 Hz) pure tone average (PTA) \(\geq 30\) dB HL, and normal hearing in the contralateral ear, defined as PTA <30 dB HL.

Following identification, electronic medical records were reviewed to ensure that youth met the following inclusion/exclusion criteria for participation:

- School-age (5-19 years)
- Presence of permanent UHL of any type (conductive, sensorineural, or mixed)
- Absence of medical condition that could affect physical or social functioning
- Performance within 2 grade levels of expectation based on age
- Able to complete interviews/questionnaires in English

Otolaryngology and audiology clinical records were reviewed. Permanent hearing loss was determined by evaluating the clinical history, etiology, and persistence of hearing loss across clinical encounters. Once it was determined that youth met criteria for participation, families were contacted by phone or in clinic to offer the youth the opportunity to participate—first for face-to-face interviews and then functional impact rating. Per Institutional Review Board protocol, informed consent was obtained from parents and youth who were 18 or 19 years old; assent was obtained for youth <18 years old.

**Interviews and Thematic Analysis**

An interview guide was developed through discussion among multiple hearing health providers. All interviews were conducted face-to-face, audio recorded, and transcribed via a professional service. This process was overseen by a pediatric otolaryngologist (K.C.Y.S.) and an outcomes scientist with the Seattle Quality of Life Group (T.C.E.), both of whom previously published their methods on functional and quality-of-life assessment among youth with hearing loss.\(^9\)

Once interviews were completed, de-identified transcripts were excerpted and coded to identify functional domains, similar to previously published protocols.\(^10\) Codes were reconciled among the reviewers to ensure consistent application. Once coded, transcript excerpts were then further examined to develop specific items related to the functional impact of UHL via the language expressed by the youth. Redundant themes were removed. Once all potential items of interest were developed, they were reviewed by youth with UHL and hearing health experts from multiple academic institutions.

Items were removed or retained according to the following prespecified item selection criteria: (1) there must be an association between the content of the item and UHL; (2) wording of the item would apply to most youth with UHL; (3) characteristics of the item must be able to change over time; and (4) intervention may influence the characteristics of the item. Through several iterations, the list of draft items was refined and revised by investigator consensus.

**Functional Impact Rating**

After item development, youth with UHL were then recruited by phone and through the clinic for functional impact rating. Youth who had previously completed interviews were also eligible to complete the item-rating exercise. The youth were asked to rate how frequently each item impacted their daily functioning with a 5-point Likert scale (0 = never, 1 = rarely, 2 = sometimes, 3 = most of the time, 4 = always). Questions were worded in such a way that a higher score indicated lower perceived burden of impact. For example, “I am able to hear my teacher well from any seat in the classroom.” The youth were also asked to prioritize how important each item felt to them when carrying out daily activities per a 3-point scale (1 = very important, 2 = somewhat important, 3 = not important).

To be included in the study, youth had to be able to read and provide functional impact ratings independently at time of encounter. Survey data were collected and managed with
were used. All analyses were performed with Stata 13.1. A P value of .05 or less was considered statistically significant. Two-sided tests in stepwise modeling but ultimately not included as potential covariates in the final regression model controlling for type of hearing loss (conductive, sensorineural, or mixed) was used to further assess the relationship between functional impact scores and hearing loss severity. Other demographic characteristics, such as age and sex, were explored as potential covariates in stepwise modeling but ultimately not included as they did not influence the final results. For all tests, P < .05 was considered statistically significant. Two-sided P values were used. All analyses were performed with Stata 13.1 (Stata, College Station, Texas).

**Results**

Fifteen youth with UHL participated in concept elicitation interviews. Participant ages ranged from 11 to 17 years at the time of interview, and 7 (47%) were using a hearing device (Table 1). Eight (53%) youth had single-sided deafness, defined as unaidable hearing in the affected ear. One (7%) had unilateral conductive hearing loss related to aural atresia. The youth interviewees discussed common concerns related to functional impact, including problems with sound localization (93%), importance of head/body positioning when listening (87%), and challenges with classroom learning (80%).

Thematic analysis identified transcript excerpts related to functional status. Figure 1 provides a flowchart for item selection based on the content of these excerpts. There were 299 excerpts coded as relating to physical functioning. After elimination of redundant concepts and application of item selection criteria, 20 physical function items were retained. Similarly, 443 excerpts were initially identified for social functioning, and 21 were retained by expert consensus. We were able to include 4 teenagers with UHL in our expert panel to help us qualitatively discuss which items seemed to resonate most strongly with their experience.

The 41 items were then thematically grouped into 8 functional domains: sound localization, ear position, noise environment, attention/focus, safety, relationships, group communication, and technology usage (see Table 2 for sample list). For the complete list of draft items, see the supplemental appendix in the online version of the article.

Functional impact ratings were obtained from 26 youth with UHL. The average age of the participants was 13.6 years (range, 9-18 years). Two youth who had completed interviews also completed functional impact ratings.

The hearing loss characteristics and functional impact scores of the youth who provided functional impact ratings are available in Table 3. There was a statistically significant difference in average PTA between the youth with conductive hearing loss (53 dB HL, SD = 13), and those with sensorineural or mixed hearing loss (87 dB HL, SD = 17; P < .001). The average total functional impact score was very similar across all types of hearing loss, at 2.7. Subgroup functional domain scores were also quite similar, with the exception of the relationships domain, in which youth with aural atresia scored significantly worse than the other youth (mean score, 2.3 vs 3; P = .03). In addition, 2 youth with congenital single-sided deafness completed the instrument. They reported average sound localization scores of only 1.7, in contrast to 2.5 for the remainder of the youth.

We also compared functional scores based on whether a youth was using a hearing device at time of instrument completion. Five youth were using hearing devices at the time of participation, and they reported a higher average total score as compared with youth not using a hearing device.

### Table 1. Demographic Characteristics of Youth with UHL.

<table>
<thead>
<tr>
<th>Method of Participation</th>
<th>n</th>
<th>Age, Mean (Range), y</th>
<th>SSD, %</th>
<th>PTA, Mean (SD)</th>
<th>Hearing Device, %</th>
<th>Congenital HL, %</th>
<th>Atresia, %</th>
<th>Male, %</th>
<th>White Non-Hispanic, %</th>
</tr>
</thead>
<tbody>
<tr>
<td>Interviews</td>
<td>15</td>
<td>13.4 (11-17)</td>
<td>53</td>
<td>74 (30)</td>
<td>47</td>
<td>40</td>
<td>7</td>
<td>60</td>
<td>80</td>
</tr>
<tr>
<td>Functional impact ratings</td>
<td>26</td>
<td>13.6 (9-18)</td>
<td>15</td>
<td>62 (20)</td>
<td>19</td>
<td>58</td>
<td>46</td>
<td>46</td>
<td>54</td>
</tr>
<tr>
<td>Total</td>
<td>39</td>
<td>13.5 (9-18)</td>
<td>31</td>
<td>68 (26)</td>
<td>31</td>
<td>54</td>
<td>33</td>
<td>54</td>
<td>67</td>
</tr>
</tbody>
</table>

Abbreviations: HL, hearing loss; PTA, pure tone average; SSD, single-sided deafness.

*Two youth participated in interviews and functional impact ratings.

REDCap (Research Electronic Data Capture), a secure web-based application designed to support data capture for research.\(^1\)**

**Analysis**

Total average functional impact score was calculated by averaging the responses to items on the 5-point scale. Items were also thematically grouped by functional domain, and average functional impact scores were calculated for each functional domain. In a preliminary assessment of construct validity, we evaluated scores within a domain that would seem most likely to be impacted by UHL—sound localization—and statistically compared it to the other functional domains with 1-way analysis of variance.

Because audiometric testing is considered the gold standard for hearing loss assessment, we performed a preliminary assessment of criterion validity by comparing total average functional impact scores and PTA. The Pearson correlation coefficient was calculated to assess the strength of association between functional impact score and 4-frequency PTA (500, 1000, 2000, and 4000 Hz) as well as word recognition scores. Finally, a multivariate linear regression model controlling for type of hearing loss (conductive, sensorineural, or mixed) was used to further explore the relationship between functional impact scores and severity of hearing loss. Other demographic characteristics, such as age and sex, were explored as potential covariates in stepwise modeling but ultimately not included as they did not influence the final results. For all tests, \( P < .05 \) was considered statistically significant. Two-sided \( P \) values were used. All analyses were performed with Stata 13.1 (Stata, College Station, Texas).
(3 vs 2.6), although the difference was not statistically significant ($P = .2$). The 5 youth with hearing devices also had higher average scores in every functional domain, but again the differences did not meet statistical significance.

Impact scores were then compared across the functional domains. The lowest functional scores were reported in the domains of sound localization (2.4) and ear positioning (2.3). To determine if there were significant differences among scores in different functional domains, we compared the scores of the other domains against the mean sound localization impact score using 1-way analysis of variance modeling (see Table 4). Using this analysis, we found functional burden associated with sound localization, ear positioning, noise environment (2.5), and stigma (2.8) to be similar.

To assess criterion validity, we then explored whether there was an association between functional impact score and hearing loss severity as measured by 4-frequency PTA. Figure 2 is a scatterplot of total average functional impact score by 4-frequency PTA of the abnormal ear. We evaluated this relationship statistically by calculating a Pearson correlation coefficient of 0.35, consistent with moderate correlation; however, this uncontrolled analysis did not reach significance ($P = .08$). Nineteen youth also had word recognition score results available. We calculated a weak correlation between total average functional impact score and word recognition score (Pearson coefficient = 0.21); however, this correlation was not significant ($P = .4$).

A multivariate linear regression model controlling for type of hearing loss (conductive, mixed or sensorineural) identified a statistically significant association between PTA and functional score, with a regression coefficient of –0.02 ($P = .008$; 95% CI −0.03 to −0.006). In other words, when controlling for type of hearing loss, a youth would be expected to score 0.4 points (or approximately 8%) lower on the functional impact scale with every 20–dB HL increase in PTA in the abnormal ear. The $R^2$ for our model was calculated at 0.26, which is the percentage of variation explained by our regression model out of the total variation.

We evaluated the correlation between type of hearing loss and PTA and found a high correlation (Pearson correlation coefficient = 0.7); however, additional analysis did not identify multicollinearity. Because of potentially important differences in perception between individuals with conductive and sensorineural hearing loss (SNHL), type of hearing loss was included as a covariable in our model. Age and sex

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**Table 2. Sample List of Draft Items for Functional Assessment of Youth with Unilateral Hearing Loss.**

<table>
<thead>
<tr>
<th>Functional Domain</th>
<th>Items, n</th>
<th>Example</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sound localization</td>
<td>3</td>
<td>I can tell where a sound is coming from just by listening.</td>
</tr>
<tr>
<td>Ear position</td>
<td>6</td>
<td>People are able to get my attention even if they speak toward my deaf ear.</td>
</tr>
<tr>
<td>Noise environment</td>
<td>6</td>
<td>In a loud place, I am able to speak easily with others.</td>
</tr>
<tr>
<td>Relationships</td>
<td>5</td>
<td>I feel comfortable talking to my friends about my hearing loss.</td>
</tr>
<tr>
<td>Attention/focus</td>
<td>5</td>
<td>I am able to pay attention in class and do my schoolwork.</td>
</tr>
<tr>
<td>Technology usage</td>
<td>4</td>
<td>I am able to listen to TV at the same volume as my family members and friends.</td>
</tr>
<tr>
<td>Group communication</td>
<td>7</td>
<td>I am easily able to talk to multiple friends at the same time.</td>
</tr>
<tr>
<td>Safety</td>
<td>5</td>
<td>I am able to hear cars coming toward me when I cross the street.</td>
</tr>
</tbody>
</table>
were also explored as covariables without a significant change in results.

A similar multivariate regression model evaluating the association between functional score and degree of hearing loss (mild, moderate, moderately severe, severe, profound) calculated a similar regression coefficient (–0.02) but did not meet statistical significance ($P = .08; 95\% \text{ CI, } –0.04 \text{ to } 0.2$).

Finally, we evaluated the prioritization survey results, in which the youth were asked to rate how important the items felt to them in achieving what they wanted to do every day. Prioritization scores were tallied for the 23 youth who completed the exercise. The 3 items with the highest prioritization scores were as follows (rated as “very important”): “I am able to hear my teacher well from any seat in the classroom” (n = 21), “People are able to get my attention even if they speak toward my deaf ear” (n = 17), and “When people speak toward my deaf ear, I understand what they say” (n = 16). Figure 3 is a weighted plot depicting the youth’s responses to these 3 questions in 5-point Likert scale format.

**Discussion**

When compared with normal-hearing peers, children with UHL have higher rates of grade failure,$^{12,13}$ poorer speech
and language outcomes and possibly lower IQ scores. Youth with sensory abnormalities such as UHL may be at a disadvantage when engaging in learning. According to the principle of social learning theory, children learn by adopting what they hear and observe from adults and older peers in their environment. Children with hearing loss, including UHL, may have more limited access to this information. However, it is important to also consider the importance of social situation, educational environment, and individual resiliency when determining the impact of UHL on a child’s functional experience. The presence of UHL does not dictate a particular outcome for a child. We must also consider the etiology of the hearing loss, other medical conditions associated with it, and the overall hearing health environment to determine how well a youth will adapt to having UHL.

Perhaps because of these other factors, previous studies found that youth with hearing loss who have similar hearing thresholds do not necessarily report the same degree of impact on their quality of life. Our study lends support to the suggestion that objective audiometric testing and patient-reported assessments may provide different but complementary information in the management of UHL. Note that there are other well-established patient-reported outcomes instruments for the assessment of children with hearing loss, such as the HEAR-QL (Hearing Environments and Reflection on Quality of Life) and YQOL-DHH (Youth Quality of Life Instrument–Deaf and Hard of Hearing); however, these instruments were developed with research that included a significant proportion of youth with bilateral hearing loss. These studies were not carried out with the purpose of specifically evaluating youth with UHL. Further research may prove beneficial as we refine hearing rehabilitation protocols for this population.

Youth with different forms of UHL will likely have differing perceptions regarding the burden of impact. For this initial study, we aimed to include youth with a wide variety of types and severity of UHL so that a broad range of candidate items could be developed. For validation of the instrument, we would aim to evaluate differences among the youth, as there may be variability in functional impact based on age of youth and type of UHL.

Further research should be conducted to determine if there are differences in functional impact based on the etiology of UHL. For example, the 2 youth with congenital single-sided deafness who completed the instrument reported lower functional status for sound localization as compared with the other youth with UHL; this finding is consistent with previous studies that identified the importance of early bilateral auditory pathways for binaural fit with a behind-the-ear hearing aid. Youth with UHL seem to report variable degrees of benefit with hearing devices: While some enjoy using them, others report dissatisfaction with currently available technology. The 5 youth who were using hearing devices when they completed the instrument reported higher average scores in every functional domain, although the difference was not large enough to meet statistical significance with such a small sample size. A functional assessment instrument for youth with UHL may serve as a guide for treatment discussions between hearing health providers, including otolaryngologists, and families. Hearing health providers often find it difficult to know how strongly to recommend hearing rehabilitation for youth with UHL, and previous studies found this uncertainty to be a source of concern for parents. It is important to mention the limitations of this study. First, it is preliminary with a small sample size, which was limited to a single site for recruitment. While the group of 39 participants was well balanced in terms of type of hearing loss and demographic characteristics, there were important differences between the youth who participated in interviews and those who participated in functional impact ratings. In the future, we hope to recruit a larger sample of patients, including those at other institutions, to validate the functional assessment instrument.

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hearing.\textsuperscript{24} In addition, congenital cytomegalovirus (CMV) is known to be associated with congenital and progressive SNHL and developmental delays. Approximately 10% to 20% of children with asymptomatic congenital CMV develop SNHL, and of these, the majority experience unilateral loss.\textsuperscript{25,26} In our study, none of the children with SNHL who underwent blood spot testing returned positive for CMV, but future studies could compare the functional status between youth with CMV-related UHL and those with other forms of UHL. Finally, patients with unilateral SNHL also seem to be at greater risk for vestibular disorders.\textsuperscript{27} Among the youth who participated in our study, I had a diagnosis of enlarged vestibular aqueduct; 1 had history of labyrinthine hemorrhage; and I developed disabling disequilibrium at the time of initial hearing loss progression. At the time of participation in our study, none of the youth were undergoing treatment for vestibular problem, but balance disorders could contribute to cognitive loading and fatigue among children with UHL. In the future, we hope to investigate how other factors associated with UHL impact functional outcomes.

Conclusion

This study is a patient-centered examination of the functional impact of UHL among youth. Our analysis identified a moderate correlation between severity of hearing loss and greater perceived burden of disease, suggesting that change in hearing threshold does not fully explain variability in functional impact score. Youth with UHL reported the lowest functional scores in the domains of sound localizability and ear position, indicating that the functional problems perceived by youth with UHL may differ from youth with symmetric, bilateral hearing loss. Further study is needed to validate the utility of the items generated through this study for functional assessment of youth with UHL.

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Author Contributions

Patricia L. Purcell, study conception/design; data acquisition; analysis, drafting; final approval and agreement; Todd C. Edwards, study conception/design; analysis; manuscript revision; final approval and agreement; Meghan Wisneski, study conception/design; analysis; manuscript revision; final approval and agreement; Dylan K. Chan, study conception/design; data analysis; manuscript revision; final approval and agreement; Henry Ou, study conception/design; analysis; manuscript revision; final approval and agreement; David L. Horn, study conception/design; analysis; manuscript revision; final approval and agreement; Jonathan R. Skirko, study conception/design; analysis; manuscript revision; final approval and agreement; Kathleen C. Y. Sie, study conception/design; data acquisition and analysis; manuscript revision; final approval and agreement.

Disclosures

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Supplemental Material

Additional supporting information is available in the online version of the article.

References


