Validation of a Pediatric Voice Quality-of-Life Instrument

The Pediatric Voice Outcome Survey

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Objective: To validate a parent proxy instrument to study the voice-related quality of life (VR-QOL) for the pediatric population.

Methods: The voice outcome survey (VOS) was administered to 108 caregivers of children aged 2 to 18 years, who either had a tracheotomy or had achieved surgical decannulation. The VOS was altered so that each item was addressed to the parent proxy as opposed to the child. A scoring paradigm was developed to report the scores on a range of 0 to 100. Structural properties of the instrument examined include validity, reliability, and principal component analysis. The VOS was then revised to suit the results of this analysis.

Results: The mean ± SD (range) score for the pediatric VOS was 49.8 ± 27.1 (0-100). Reliability analysis and principal component analysis supported the reduction of 1 item from the final pediatric VOS. The revised instrument demonstrated an overall Cronbach α value of .86. Substratification by age revealed robust Cronbach α values from ages 2 to 18 years. Construct validity analysis also supported statistical significance (P = .004). Principal component analysis of the revised instrument supports its internal structure and design.

Conclusions: The pediatric VOS is a simple, short, health status instrument designed to measure the VR-QOL in the pediatric population. Cross-sectional analysis suggests that children and adolescents with tracheotomies have a poorer VR-QOL than do those who have achieved decannulation.

MATERIALS AND METHODS

Parents and caregivers of children and adolescents who have undergone tracheotomy were contacted via an active Web site that provides information, counsel, and support to such families or by direct mailing to families whose children had received a tracheotomy at the Cincinnati Children’s Hospital Medical Center, Cincinnati, Ohio. The Web site chosen (http://www.tracheotomy.com) is specifically designed for families in which a child has received a tracheotomy. Currently, several hundred families access this Web site as a source of information. The pediatric voice outcome survey (VOS) was administered after obtaining permission for its use from its senior author (R. E. Gliklich, MD). The VOS was altered to allow for parent-proxy administration. The items were therefore addressed to the health care proxy in lieu of addressing the children or adolescents themselves. Prior to the beginning of the study, it was reviewed by the institutional review board of the Cincinnati Children’s Hospital Medical Center and received approval. An introductory paragraph was placed on the first page of the Web site for purposes of informed consent and a link was established for the parent/primary caregiver to receive the instrument itself. Then the parent/primary caregiver either completed the instrument online and e-mailed the completed instrument to the primary investigator (C.J.H.) or downloaded the instrument for completion and then sent the form directly to the primary investigator. The completed forms were entered into a Microsoft Access (Microsoft, Seattle, Wash) database, and analysis was performed by linking the database to the SAS 8.1 software package (SAS Institute Inc, Cary, NC).

RESULTS

Overall, 108 caregivers of children or adolescents requiring tracheotomies or who had required tracheotomies in the past and have been decannulated participated in the study and completed the instrument. Missing data were handled by assigning “0” values as missed entry values. Overall, there was less than a 0.001% incidence of missing data points. The population characteristics for the respondents and their children are delineated in Table 1.

A mean score was calculated using all the items included in the VOS. Three items required score reversal to remain consistent with a scale whereby low scores indicate a “poor” HR-QOL. The overall score was then fitted to a scale ranging from 0 to 100.

The instrument’s reliability was assessed by means of calculating Cronbach α values (this allows for the assessment of item-total correlations to measure the impact of systematically deleted variables within the central construct). A coefficient of α greater than .65 was interpreted to be robust (α > .55 was deemed acceptable). Principal component and factor analysis was then performed to evaluate the internal structure and the design of the instrument with reference to its application to the pediatric population. While reliability analysis and factor analysis supported the overall structure, they also suggested that 1 particular item (relating to swallowing) within the instrument

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>No. (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Respondents who are mother of the child</td>
<td>95 (88)</td>
</tr>
<tr>
<td>Male children and adolescents</td>
<td>64 (59)</td>
</tr>
<tr>
<td>Female children and adolescents</td>
<td>44 (41)</td>
</tr>
<tr>
<td>Age, y</td>
<td></td>
</tr>
<tr>
<td>2-5</td>
<td>68 (63)</td>
</tr>
<tr>
<td>6-10</td>
<td>24 (22)</td>
</tr>
<tr>
<td>11-18</td>
<td>18 (15)</td>
</tr>
</tbody>
</table>

Table 1. Population Characteristics

1. In general, how would you say your child’s speaking voice is?
   - Excellent
   - Good
   - Adequate
   - Poor or inadequate
   - My child has no voice

2. To what extent does your child’s voice limit his or her ability to be understood in a noisy area?
   - Limited a lot
   - Limited a little
   - Not limited at all

3. During the past 2 weeks, to what extent has your child’s voice interfered with his or her normal social activities or with his or her school?
   - Not at all
   - Slightly
   - Moderately
   - Quite a bit
   - Extremely

4. How often does your child have trouble with food or liquids “going down the wrong pipe” when he or she eats food or drinks liquid and begins to cough after eating or drinking?
   - All the time
   - Most of the time
   - Sometimes
   - Rarely
   - Never

5. Do you find your child “straining” when he or she speaks because of his or her voice problem?
   - Not at all
   - A little bit
   - Moderately
   - Quite a bit
   - Extremely

Initial pediatric voice outcome survey (VOS). Revised and final pediatric VOS does not include question 4.
did not fit as well as the other items within the overall schema (Figure). Accordingly, this item was deleted from the instrument, and the instrument was then reanalyzed for purposes of reliability and principal component analysis. The results of both the initial and final analyses are given in Table 2 (see the “Comment” section). The α values for the revised pediatric VOS were then evaluated to assess the reliability of the instrument for various subpopulations of children and adolescents according to their age (Table 3). The revised instrument with the 1 item deleted was then examined, and the overall mean ± SD (range) score was 49.8 ± 27.1 (0-100).

To assess the validity of the instrument, an evaluation of the instrument’s construct validity was performed (ie, the instrument’s ability to identify and demonstrate significant differences in subpopulations with regard to preconceived, a priori hypotheses). The initial hypothesis evaluated was that for 2 discrete populations of children and adolescents, those with tracheotomies (population 1) would experience vocal disturbances in a more significant and profound way than those whose tracheotomies had been removed (population 2). A sample size of 130 was chosen to be sufficiently robust to detect significant differences (P = .05), assuming medium effect size.23 Although the actual sample size was slightly smaller than that hypothesized, a 2-tailed t test performed to compare the mean scores (population 1, 39.4; population 2, 51.6) for these 2 populations demonstrated statistical significance (P = .004).

### Table 2. Cronbach α Values (Reliability) and Factor Analysis Values for Overall Instrument With (Initial Pediatric VOS) and Without (Revised Pediatric VOS) Item Regarding Swallowing*

<table>
<thead>
<tr>
<th>VOS Question</th>
<th>Cronbach α Value</th>
<th>Factor Analysis Value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Initial</td>
<td>Revised</td>
</tr>
<tr>
<td>1</td>
<td>.70</td>
<td>.67</td>
</tr>
<tr>
<td>2</td>
<td>.89</td>
<td>.74</td>
</tr>
<tr>
<td>3</td>
<td>.68</td>
<td>.68</td>
</tr>
<tr>
<td>4</td>
<td>.28</td>
<td>...</td>
</tr>
<tr>
<td>5</td>
<td>.54</td>
<td>.57</td>
</tr>
</tbody>
</table>

*VOS indicates voice outcome survey. Boldface indicates that the revised and final VOS does not include this question.

### Table 3. Cronbach α Values for Overall Instrument Documenting Reliability Analysis for the Pediatric VOS as Substratified by Age*

<table>
<thead>
<tr>
<th>Age, y</th>
<th>Cronbach α Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>2-18</td>
<td>.83</td>
</tr>
<tr>
<td>2-5</td>
<td>.86</td>
</tr>
<tr>
<td>6-10</td>
<td>.85</td>
</tr>
<tr>
<td>11-18</td>
<td>.69</td>
</tr>
</tbody>
</table>

*VOS indicates voice outcome survey.

The field of outcomes research with regard to vocal disturbances has only recently begun to be explored. At present, most work has focused on the impact of vocal disturbances on adult populations with a variety of disorders before and after various treatments. Within the field of pediatric otolaryngology, there are also a host of disorders in which the impact of a child’s ability to vocalize and to interact with his or her peers and community are of notable importance. Examples of children and adolescents with disorders that coexist with impaired ability to vocalize include those with vocal cord paralysis, those with subglottic stenosis necessitating a tracheotomy, those with velopharyngeal insufficiency, or those with gastroesophageal reflux disease and vocal cord nodules. These examples represent only a small subset of pediatric conditions in which pathological vocal alterations may affect the child and his or her ability to interact and to develop appropriate communication skills in a meaningful way.

A formal algorithm for assessing the pediatric voice has yet to be developed. Any such examination of necessity includes a detailed history and physical (obtained according to age by the caregiver or child, or a combination of these two). Visualization of the airway is accomplished by flexible and rigid endoscopy as appropriate; imaging studies (computed tomography or magnetic resonance imaging) can be useful for initial evaluation of certain voice disorders such as unexplained vocal cord paralysis. Other studies that are used at various centers include stroboscopy, electroglossography, and ultrasonography. All of these procedures are technician dependent, and the interpretation of these examination findings varies inversely with the age of the child. Laboratory tests are helpful in a small discrete population in whom the history and physical examination findings are suggestive.

Functional assessment of the child’s ability to vocalize may provide invaluable supplementary information to the field of pediatric laryngology, which at present lacks well-accepted objective parameters. While many of the disorders that affect the pediatric airway require interventions that necessitate a balance between establishing a patent airway and preserving an adequate voice, the definition of what constitutes an adequate voice needs clarification. Moreover, as more focus is placed on integrating functional outcomes regarding vocal performance into comprehensive care regarding the pediatric airway, novel strategies may be devised to allow for both goals to be more readily accomplished. Finally, reporting of functional outcomes allows for an understandable objective criteria to assess outcomes regarding one institution’s results or multi-institutional results; such in-
formation would be invaluable for caregivers trying to make informed decisions regarding their children.

The present study draws on a defined population of children and adolescents and their caregivers to validate a instrument to study voice-related quality of life (VR-QOL). The result therefore is a validated instrument for children and adolescents with tracheotomies aged 2 to 18 years. The next step in creating a global pediatric VR-QOL instrument is to administer the instrument to otherwise healthy pediatric subjects for analysis purposes. With regard to this specific population, the VOS is a valid instrument, although reliability analysis suggests that the 1 item describing pediatric swallowing issues does not provide information that is unique and separate from the responses to the other 4 items. These reliability data were obtained post hoc, and all the items in the original VOS were used for initial application. Nevertheless, for this particular pediatric population, the modified pediatric VOS is statistically more streamlined in terms of congruent reliability. For other pediatric populations, the full VOS might be appropriate.

The pediatric VOS is cross-sectional by design, and a longitudinal application of this instrument is in progress to explore more fully the changes in VR-QOL over time on a patient-by-patient basis. To give more meaning to the scores generated by this instrument, further work needs to be done to establish normative data for purposes of cross-population comparisons. Simultaneous application of generic instruments measuring a child's overall HR-QOL and instruments measuring the caregiver's HR-QOL and measure of caregiver burden would further bolster the ease and breadth of interpretability of the pediatric VOS. Finally, for the disorders that affect the pediatric airway while the child is still relatively young, a parent proxy instrument remains valid. The pediatric VOS has been validated for children and adolescents aged 2 to 18 years, but most children in this study were younger than 5 years. Validation of a self-administered instrument for older children would appear warranted for those who are more cognizant of their situation.

In conclusion, the pediatric VOS represents a validated, brief instrument to assess the VR-QOL in the pediatric population. It has been shown to discriminate between subpopulations to document that children and adolescents with tracheotomies have a poorer VR-QOL than do those who have achieved decannulation.

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This study was presented at the annual meeting of the American Society for Pediatric Otolaryngology, Boca Raton, Fla, May 13-14, 2002, and has received first place for the Charles F. Ferguson Resident Research Award.

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REFERENCES