Comparison of Pediatric Voice Outcome Survey, Reflux Symptom Index, Reflux Finding Score, and Esophageal Biopsy Results

Jeffrey P. Simons, MD; Clark A. Rosen, MD; Margaretha L. Casselbrant, MD, PhD; David H. Chi, MD; Barry M. Schaitkin, MD; Elaine N. Rubinstein, PhD; David L. Mandell, MD

Objective: To examine correlations between the Pediatric Voice Outcome Survey (PVOS) score, the Reflux Symptom Index (RSI) score, the Reflux Finding Score (RFS), and esophageal biopsy findings in children undergoing upper aerodigestive tract endoscopy.


Setting: Tertiary care children’s hospital.

Patients: The study included 36 children with the primary problem of dysphonia (n=28) or cough (n=8) who underwent endoscopy.

Interventions: The PVOS and the RSI were administered to the patient’s parents before surgery. The patients underwent laryngotracheobronchoscopy and esophageal biopsy. Four raters independently assigned an RFS to the laryngeal photographs.

Main Outcome Measures: The assessment included (1) PVOS scores, RSI scores, and RFSs; (2) internal consistency of PVOS and RSI scores; (3) RFS intrarater and interrater reliability; and (4) correlations between PVOS score, RSI score, RFS, and esophageal biopsy findings.

Results: The mean (SD) age of the patients was 7.5 (2.6) years; the mean (SD) PVOS score, 71.9 (21.4); and the mean (SD) RSI score, 16.2 (9.1). The PVOS and the RSI scores demonstrated good internal consistency (Cronbach’s α=0.79 and 0.78, respectively). The RFS exhibited good intrarater reliability (r=0.66-0.98) and moderate interrater reliability (r=0.32-0.70). The PVOS and RSI instruments displayed significant correlation (r=−0.30; P=.04). There were no other significant correlations between RFSs, esophageal biopsy results, PVOS scores, or RSI scores (P>.05).

Conclusions: The RSI may be a useful parent-proxy instrument in addition to the PVOS for pediatric voice patients. The RFS is reliable in children, but its validity could not be demonstrated in this patient population.

The main purpose of this study was to examine correlations between PVOS scores, RSI scores, RFSs, and esophageal biopsy results in children undergoing upper aerodigestive tract endoscopy. Other goals were to attempt to validate the RSI as a proxy survey for children and to examine the reliability and validity of the RFS in children.

The Reflux Finding Score (RFS) is an 8-item clinical severity scale that is based on findings during laryngoscopy (Figure 3). The total RFS ranges from 0 (no abnormal findings) to 26 (worst possible score). Like the RSI, the RFS has also been shown to display excellent psychometric properties in an adult population with dysphonia as well as those with chronic, refractory cough in this study because it is generally accepted that both of these symptoms can be extraesophageal manifestations of GER. The PVOS and the RSI were administered preoperatively to the confirmed diagnosis of LPR.6 However, currently there is no validated grading scale for endoscopic laryngeal abnormalities.

At the time that the endoscopies were performed, it was the general practice of the senior author (D.L.M.) to perform operative endoscopy and esophageal biopsy on most children with hoarseness. As such, the group of dysphonic patients reviewed in this study does happen to be consecutive. The degree of dysphonia was determined primarily based on parental history and a brief office examination, but formal perceptual rating with a speech-language pathologist using an accepted scale was not included as one of the elements of the analysis.

Operative endoscopy was not considered an initial step in the workup of pediatric chronic cough but was eventually performed in cases with negative responses to some or all of the following diagnosis and management options: allergy testing and treatment, asthma workup and treatment, chest radiography, empirical antireflux medication, immunodeficiency workup, and adenoidectomy. We included children with dysphonia as well as those with chronic, refractory cough in this study because it is generally accepted that both of these symptoms can be extraesophageal manifestations of GER.

The PVOS and the RSI were administered preoperatively to all the children’s caregivers on the day of surgery. Four blinded raters independently assigned RFSs to the endoscopic laryngeal photographs. The reviewers consisted of 2 pediatric otolaryngologists (raters A and D [D.H.C. and M.L.C.]), 1 adult laryngologist (rater B [C.A.R.]), and 1 adult general oto-

METHODS

This study is a retrospective medical record review, including pediatric voice quality-of-life and LPR surveys. The protocol was reviewed by the institutional review board of the Children’s Hospital of Pittsburgh, Pittsburgh, Pennsylvania, and approved by expedited review, in compliance with Health Insurance Portability and Accountability Act guidelines. Thirty-six children with the primary chief complaint of dysphonia (n=28) or chronic cough (n=8) underwent upper aerodigestive tract endoscopy with standardized photodocumentation and esophageal biopsies performed by 1 pediatric otolaryngologist (D.L.M.) at the hospital (Figure 4 and Figure 5). Exclusion criteria included tracheotomy, previous intubation, premature birth, previous laryngeal surgery, and known laryngeal abnormalities.

Within the last month, how did the following problems affect your child?

1. Hoarseness or a problem with your child’s voice
2. Throat clearing
3. Excess throat mucus or postnasal drip
4. Difficulty swallowing food, liquids, or pills
5. Coughing after eating or after lying down
6. Breathing difficulties or choking episodes
7. Troublesome or annoying cough
8. Sensation of something sticking in the throat or a lump in the throat
9. Heartburn, chest pain, indigestion, or stomach acid coming up

Figure 1. Pediatric Voice Outcome Survey. Score range, 0 (poor voice-related quality of life) to 100 (no voice problems).

Figure 2. Reflux Symptom Index. Score range, 0 to 45. A score of 0 indicates no problem; a score of 5 or more, a severe problem.

Figure 3. Reflux Finding Score. Score range, 0 (no abnormal findings) to 100 (worst possible score).

The following items ask about activities that your child might do in a given day.

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

The following items ask about activities that your child might do in a given day.

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
   • Good
   • Slightly
   • Moderately
   • Quite a bit
   • Extremely

4. 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

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Subglottic edema (pseudosulcus vocalis)
0 = Absent
2 = Present

Ventricular obliteration
0 = None
2 = Partial
4 = Complete

Erythema/hyperemia
0 = None
2 = Arytenoids only
4 = Diffuse

Diffuse laryngeal edema
0 = None
1 = Mild
2 = Moderate
3 = Severe
4 = Obstructing

Granuloma/granulation
0 = Absent
2 = Present

Thick endolaryngeal mucus
0 = Absent
2 = Present

Table 1. Laryngeal Abnormalities

<table>
<thead>
<tr>
<th>Abnormality</th>
<th>0 = Absent</th>
<th>1 = Mild</th>
<th>2 = Moderate</th>
<th>3 = Severe</th>
<th>4 = Obstructing</th>
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Table 2. Laryngeal Abnormalities

- Excellent
- Good
- Adequate
- Poor or inadequate
- My child has no voice

- Limited a lot
- Limited a little
- Not limited at all

- Not at all
- Good
- Slightly
- Moderately
- Quite a bit
- Extremely

- Not at all
- A little bit
- Moderately
- Quite a bit
- Extremely

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gologist (rater C [B.M.S.]). Fourteen duplicate subjects (39% of the total subject population) were randomly selected and scattered throughout the set of endoscopic images to assess intrarater reliability. The addition of the duplicated subject images led to a convenient, round number of 50 sets of endoscopic images to be rated by each reviewer.

The main outcome measures included (1) PVOS scores, RSI scores, and RFSs; (2) PVOS and RSI internal consistency; (3) RFS intrarater and interrater reliability; and (4) correlations between PVOS scores, RSI scores, RFSs, and esophageal biopsy results. The internal consistency of the PVOS and the RSI scores were calculated with the Cronbach’s coefficient. The intrarater and interrater reliability of the RFS were determined using Pearson correlation coefficients. Correlations between PVOS scores, RSI scores, RFSs, and esophageal biopsy results were also calculated with Pearson correlation coefficients. The statistical significance of these correlations was determined using the t test. For all statistical tests, significance was defined as P < .05.

RESULTS

The mean (SD) patient age was 7.5 (2.6) years (age range, 3.0-14.1 years). Fifty-three percent of the patients were female and 47% were male. The mean (SD) PVOS score was 71.9 (21.4). The PVOS demonstrated good internal consistency (Cronbach’s α = 0.79). The mean (SD) RSI score was 16.2 (9.1). The RSI also demonstrated good internal consistency (Cronbach’s α = 0.78). The overall mean (SD) RFS score was 5.6 (3.0). The 2 pediatric otolaryngologists (raters A and D) had RFSs of 5.8 (4.4) and 5.2 (4.2), respectively. The adult laryngologist (rater B) had an RFS of 4.3 (2.8), and the general otolaryngologist (rater C) had an RFS of 7.4 (3.8) (Figure 6). There was no statistically significant difference in RFS scores between the pediatric otolaryngologists and the other otolaryngologists (P = .50).

The intrarater reliability was very high for the 2 pediatric otolaryngologists (r = 0.98 for both). The intrarater reliability was also good for the adult laryngologist (r = 0.66) and the general otolaryngologist (r = 0.70). The RFS intrarater reliability correlation coefficients ranged from 0.32 to 0.70 (moderate intrarater reliability). For the 2 pediatric otolaryngologists, the intrarater reliability correlation coefficient was 0.67. For the 2 other otolaryngologists, the intrarater reliability correlation coefficient was 0.42. The overall intraclass correlation coefficient for all 4 reviewers was 0.52.

There was a statistically significant correlation between PVOS and RSI scores (r = −0.30; P < .05; Figure 7).
However, there were no significant correlations between RFS and PVOS scores, RFS and RSI scores, RFSs and esophageal biopsy results, RSI scores and esophageal biopsy results, or PVOS scores and esophageal biopsy results. Age also was not correlated with PVOS scores, RSI scores, or RFSs.

One of the weaknesses of this study is the relatively small sample size (n = 36). There is also the possibility that different populations of children may have been lumped together in the analysis (ie, those with vocal nodules and those with LPR). A study involving a larger number of patients might have a greater likelihood of finding significant differences between PVOS scores, RSI scores, RFSs, and esophageal biopsy results among different subpopulations of children, should such differences exist. The study sample size was limited by the retrospective nature of the research, in which the decision to perform endoscopy was based on a case-by-case clinical basis and not as part of a preexisting research protocol. To address these issues, a larger-scale, prospective study would have to be designed with more strictly defined patient-selection parameters.

It should also be mentioned that the statistical analysis in our study does not rule out parental reporting bias. For example, the correlation between PVOS and RSI scores could potentially be the result, in part, of some parents having a tendency to overreport or underreport symptoms on any questionnaire they fill out. Another drawback to this study includes the lack of any formal perceptual rating of the children’s dysphonia using an accepted scale. The reason for the lack of formal perceptual evaluation of voice is that not all of the patients in our retrospective study were seen by a speech pathologist before surgery.

Many patients with LPR do not have GER disease, as the esophagus can tolerate a greater amount of refluxate from the stomach without leading to symptoms or disease as compared with the laryngopharynx. Dual pharyngeal and esophageal pH probe testing is likely required to evaluate reflux that is occurring at the level of the pharynx, but there is still no universally accepted criterion regarding the diagnosis of pathologic LPR, thereby enhancing the confusion associated with this disease process. The relative lack of normative data in children and the high frequency of “physiologic” regurgitation in normal healthy infants, along with other technical issues regarding probe placement and maintenance of hydration of the sensor, have hampered the ability of clinicians to accurately diagnose the presence of LPR in children, even more so than in adults. The relationship between reflux and pediatric voice disorders is still unclear. Because the assessment of pediatric LPR is confounded by a lack of normative data, as well as logistical and technical issues related to testing techniques, indirect methods of assessment such as reflux questionnaires and laryngeal grading instruments are desirable and deserving of further study.

In conclusion, the findings of our study demonstrate the feasibility of the RSI to serve as a parent-proxy instrument in addition to the PVOS for pediatric voice patients. However, more work is needed to validate the RSI for this population. The RFS was reliable in children, but its validity could not be demonstrated, as its scores did not correlate with those of the PVOS or the RSI or the esophageal biopsy results. There is a potential for further development and refinement of an objective pediatric laryngeal grading instrument.

To our knowledge, this is the first study to use the RSI as a parent-proxy instrument in children. We found that the scores on the PVOS (a validated pediatric voice quality-of-life instrument) and the RSI correlate with each other and display good internal consistency as parent-proxy instruments to measure voice-related quality of life and severity of LPR in children. The RSI therefore appears to have some clinical utility when used in a pediatric laryngology practice.

The RFSs in the children in our study were relatively low overall, with fewer abnormal findings (mean score, 5.6 of possible 45) when compared with adults with LPR (mean scores, 9.3 and 11.5), suggesting that there may be certain characteristics of the pediatric larynx that the adult grading scale may not accurately assess. For example, the presence of midmembranous vocal fold lesions is not included on the RFS. In our pediatric population, the RFS did demonstrate good intrarater reliability and moderate interrater reliability, similar to studies in adults, suggesting that such an instrument does have clinical potential, although pediatric-specific modifications may be required.

One goal of this study was to try to correlate laryngoscopic images (using the RFS) with measurement of voice quality of life (using the PVOS), assessment of LPR symptoms (using the RSI), and an objective measurement of GER (via esophageal biopsy). Some investigators believe that certain laryngeal findings are pathognomonic for reflux in children (eg, severe arytenoid edema, posterior glottic edema, and enlarged lingual tonsils). However, our group has previously failed to find any correlation between the endoscopic appearance of the pediatric larynx and the presence of esophagitis, thus highlighting the controversy regarding this topic.

In the present study, we found that in our pediatric population the RFS did not correlate with the PVOS scores, the RSI scores, or the esophageal biopsy results. In adults, the relationship between reflux symptoms and laryngeal findings also is unclear. One recent study has shown that in adults, extraesophageal reflux symptoms, Voice Handicap Index scores, Gastrointestinal Symptom Rating Scale scores, Short Form 36 scores, and videolaryngoscopic findings were not predictive of pathologic extraesophageal reflux as measured by pH probe testing. On the other hand, a recent study of adults with symptoms of LPR did show a strong, positive correlation between RFSS and RSI scores. One reason for the lack of correlation between RFSS and RSI scores in children could be that the manifestations of LPR in children, both with respect to symptoms and signs of laryngeal inflammation, may not be measured accurately by the instruments designed for adults.

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Author Contributions: Drs Mandell and Simons had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Simons, Rosen, Chi, and Mandell. Acquisition of data: Simons, Rosen, Chi, Schaitkin, and Mandell. Analysis and interpretation of data: Simons, Rosen, Casselbrant, Rubenstein, and Mandell. Drafting of the manuscript: Simons, Chi, Rubenstein, and Mandell. Critical revision of the manuscript for important intellectual content: Simons, Rosen, Casselbrant, Schaitkin, and Mandell. Statistical analysis: Rubenstein. Study supervision: Casselbrant and Mandell.

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REFERENCES