Annals of Otology, Rhinology & Laryngology 118(8):581-586.

Comparison of Pediatric Voice Handicap Index Scores With Perceptual Voice Analysis in Patients Following Airway Reconstruction

Alessandro de Alarcon, MD; Susan Baker Brehm, PhD; Lisa N. Kelchner, PhD; Jareen Meinzen-Derr, PhD; Janet Middendorf, MA; Barbara Weinrich, PhD

Objectives: We performed a retrospective review to compare a subjective parental proxy–derived voice handicap survey to an observer-derived method of measuring voice perturbation in children who have undergone airway reconstruction. The main outcome measures were the Pediatric Voice Handicap Index (pVHI) total score and the Overall Severity score on the Consensus Auditory-Perceptual Evaluation of Voice (CAPE-V).

Methods: The percent Overall Severity CAPE-V score (score divided by 100) and the percent pVHI score (score divided by 92) were calculated. A Wilcoxon signed rank test was used to compare CAPE-V scores with the pVHI total scores. The relationship between the pVHI scores and the CAPE-V scores was investigated with a Spearman correlation. Subgroup analysis was performed to determine the relationship of surgery type to CAPE-V and pVHI scores.

Results: Fifty subjects with a history of airway surgery who were evaluated between 2005 and 2008 were identified. Forty-two of the 50 subjects had complete data for review. Their median age was 7.1 years (range, 3.3 to 17.9 years). Their pVHI total scores had a median of 30 (range, 1 to 80). Their Overall Severity CAPE-V scores had a median of 50.5 (range, 0 to 98). Their median CAPE-V percent was higher than their median pVHI percent (50.5% versus 32.6%; $p = 0.0003$). A weak correlation was found between the Overall Severity CAPE-V score and the pVHI total score (rho = 0.41; $p = 0.0003$). There was a trend toward higher Overall Severity CAPE-V scores in patients who underwent cricotracheal resection. The total number of airway surgeries was significantly correlated with the Overall Severity CAPE-V score (rho = 0.6; $p < 0.0001$) but not with the pVHI score.

Conclusions: Children who undergo airway reconstruction often have a resulting voice disturbance that can affect their lives in multiple dimensions. The results of this study revealed a weak-to-fair correlation between the parent-reported pVHI total score and expert ratings of voice quality using the CAPE-V. In this patient population, both of these tools provided important information regarding the relationship of the severity of voice disturbance to its handicapping effects.

Key Words: CAPE-V, cricotracheal resection, dysphonia, laryngotracheoplasty, Pediatric Voice Handicap Index, quality of life, subglottic stenosis, voice.

INTRODUCTION

Children who undergo laryngotracheal reconstruction represent a unique subset of patients with voice disorders. The spectrum of voice disorders encountered in this patient population ranges from mild dysphonia to some of the most severe voice disorders that can be encountered by clinicians.[1] Historically, the main outcome measure of success for airway reconstruction has been decannulation; however, several authors have sought to understand the postoperative impact of airway reconstruction on the voice.[1-10] Furthermore, many of these authors have begun the important groundwork of trying to systematically describe voice outcomes in children who have undergone airway reconstruction.

The assessment of these children involves a team approach to evaluation and management that includes collaboration between speech-language pathologists and pediatric otolaryngologists. The evaluation process involves the use of endoscopic imaging tools (videostroboscopy) to evaluate the pediatric larynx and gain information about form, function, and pathology. Aerodynamic and acoustic analyses, as well as expert perceptual ratings of

From the Center for Pediatric Voice Disorders, Division of Pediatric Otolaryngology (all authors), the Aerodigestive Sleep Center (de Alarcon), the Department of Speech Pathology (Brehm, Kelchner, Middendorf, Weinrich), and the Division of Biostatistics and Epidemiology (Meinzen-Derr), Cincinnati Children's Hospital Medical Center, and the Department of Communication Sciences and Disorders, University of Cincinnati (Kelchner), Cincinnati, and the Department of Speech Pathology and Audiology, Miami University, Oxford (Brehm, Weinrich), Ohio.

Presented at the meeting of the American Broncho-Esophagological Association, Orlando, Florida, May 1-2, 2008.

Correspondence: Alessandro de Alarcon, MD, Division of Pediatric Otolaryngology, Cincinnati Children's Hospital Medical Center, 3333 Burnet Ave, MLC 2018, Cincinnati, OH 45229-3039.

voice, using tools such as the Consensus Auditory Perceptual Evaluation–Voice (CAPE-V), help characterize the voice disorder. The impact of a voice disorder on the child can be assessed, in part, via a health-related quality-of-life instrument or a handicapping index, such as the Pediatric Voice Handicap Index (pVHI), the Pediatric Voice Outcome Survey, or the Pediatric Voice-Related Quality-of-Life survey.[2,11-14] The pVHI is the only survey with published data regarding children who have undergone airway reconstruction.[2]

It has been our experience that parents of children with severe voice disorders following airway surgery do not consistently report a severe handicapping effect. To date, the relationship between expert ratings of severity of voice disturbance and parent (or patient) reports of handicapping effects of the voice disorder are not well known.[15] Given the potential for extreme anatomic and physiologic changes following pediatric airway reconstruction, as well as possible lifelong voice effects, a fuller understanding of the relationship of these perceptual ratings is imperative.

The purpose of the current study was to examine the relationship between expert perceptual ratings of voice in children who had undergone airway surgery and their parent's proxy report of voice handicapping effects.

MATERIALS AND METHODS

The records of 50 patients with a history of airway reconstruction were identified from a cohort of more than 300 patients with voice disorders evaluated at the Center for Pediatric Voice Disorders (a tertiary referral center) between 2005 and 2008. Forty-two subjects had complete data for all analyses. Demographic data, operative notes, pVHI scores, and CAPE-V scores were obtained. Children were excluded if there was no prior history of airway reconstruction or if the pVHI and CAPE-V were not performed on the same date. Children were subdivided into the following groups for secondary analyses: 1) glottic procedures, 2) complete laryngofissure, 3) posterior graft laryngotracheoplasty, and 4) cricotracheal resection.

pVHI. The pVHI is a 23-item parental proxy voice-related quality-of-life instrument that has previously been validated for use in the pediatric population.[2] The overall score is a subset of three scores that include functional, physical, and emotional domains. The questionnaire was administered to the parents of all study participants before their clinical voice assessment.

CAPE-V. The CAPE-V is a perceptual rating scale used by voice clinicians to rate their patients' voice quality. Developed after the 2002 consensus conference convened by the American Speech-Language-Hearing Association's Special Interest Division 3, Voice and Voice Disorders, the CAPE-V uses a 100-mm visual analog scale to assess the voice parameters of Overall Severity, Loudness, Breathiness, Pitch, Roughness, and Strain. Patients are asked to repeat or read a standard set of sentences and sustain vowels. In this study, all voice samples were collected in a soundproof booth and consensus ratings were made by 2 experienced voice clinicians with 17 years of experience.

Statistical Analysis. Continuous data were reported as medians with ranges. Because the CAPE-V is scored on a scale between 0 and 100 and the pVHI is scored on a scale between 0 and 92, the scores were normalized as percentages of overall total score for comparisons (CAPE-V actual score divided by 100; pVHI actual score divided by 92). The Wilcoxon signed rank test was used to statistically compare Overall Severity CAPE-V scores with pVHI total scores. The relationship between the pVHI scores and the CAPE-V scores was investigated with a Spearman correlation coefficient. A secondary subgroup analysis with the Kruskal-Wallis test was performed to determine the impact of particular surgical interventions on the CAPE-V and pVHI relationships. All analyses were performed with SAS version 9.1 (SAS Institute, Inc, Cary, North Carolina).

This study was performed in accordance with the policies of the Institutional Review Board at Cincinnati Children's Hospital Medical Center and the Health Insurance Portability and Accountability Act.

RESULTS

Fifty participants were identified who had been evaluated in the Center for Pediatric Voice Disorders from 2005 to 2008. For analysis purposes, we excluded 8 subjects who did not have complete data for evaluation. The median age of the subjects at the time of the assessment was 7.1 years (range, 3.3 to 17.9 years). The median pVHI total score (which is on a scale from 0 to 92) was 30 (range, 1 to 80), and the median overall CAPE-V score (which is on a scale from scale 0 to 100) was 50.5 (range, 0 to 98). The median CAPE-V percent for the study population was significantly higher than the pVHI percent (50.5% versus 32.6%; $p = 0.0003$). The median ratings and ranges for the pVHI scores and the Overall Severity CAPE-V and CAPE-V subset scores are summarized in Table 1.

TABLE 1. CAPE-V AND pVHI SCORES FOLLOWING AIRWAY RECONSTRUCTION

	Median	*Range*
CAPE-V		
Overall Severity	50.5	0-98
Loudness	21.5	0-97
Breathiness	17.5	0-98
Pitch	11	0-98
Roughness	30.5	0-98
Strain	13.5	0-85
pVHI		
Total	30	1-80
Emotional	5	0-20
Functional	12	0-27
Physical	13.5	1-34
Talkativeness	5.5	2-7

CAPE-V — Consensus Auditory-Perceptual Evaluation of Voice; pVHI — Pediatric Voice Handicap Index.

Comparison of CAPE-V and pVHI Scores. The Overall Severity CAPE-V score was positively correlated with the pVHI total, the pVHI emotional domain score, and the pVHI functional domain score. Table 2 describes the correlations between the CAPE-V and pVHI domains. Talkativeness on the pVHI was not correlated with any of the CAPE-V scores, nor was it correlated with any of the pVHI domain scores. The total severity rating on the pVHI was only correlated to Breathiness on the CAPE-V (rho = 0.43; p = 0.003)

Subgroup Analysis. To examine the influence of the numbers and types of operations on subsequent voice quality, we grouped data for the participants into the following non–mutually exclusive surgical categories: 1) glottic procedures (10 procedures), 2) complete laryngofissure (10 procedures), 3) posterior graft laryngotracheoplasty (22 procedures), and 4) cricotracheal resection (8 procedures). Participants were identified as having a glottic procedure if any one or more of the following procedures were performed: open vocal fold lateralization, endoscopic vocal fold lateralization, or carbon dioxide laser cordotomy. Many (14 or 33%) of the children in the study fit into multiple categories for subgroup analysis and therefore were not analyzed. Eight children did not have any of the four procedures. Among the 10 children who had glottic procedures, 5 had only glottic procedures. All children who had the complete laryngofissure procedure had other procedures. Ten children had only a posterior graft laryngotracheoplasty procedure, and 5 children had only a cricotracheal resection procedure.

In order to understand the differences in scores among the different procedures, we compared median domain pVHI and CAPE-V scores among mutually exclusive groups (only one procedure or no procedure). Children who underwent cricotracheal resection had higher scores than did the other groups regarding Overall Severity CAPE-V percent and several CAPE-V domains (Table 3). However, because of the relatively small sample size within each group and the necessary multiple comparison adjustments, none of the post hoc group comparisons were statistically significant at the $\alpha = 0.05$ level. The number of total open airway procedures was significantly correlated with the Overall Severity CAPE-V score, the Loudness score (too soft), and the Breathiness score (too breathy; Table 4). The number of total open airway procedures was not correlated to any of the pVHI scores.

TABLE 2. CORRELATIONS BETWEEN CAPE-V DOMAINS AND pVHI DOMAINS

	pVHI	*pVHI Domain*		
CAPE-V Domain	*Total*	*Emotional*	*Functional*	*Physical*
Overall Severity				
rho	0.39*	0.53*	0.45*	0.17
p	0.01*	0.0004*	0.003*	0.29
Loudness				
rho	0.10	0.21	0.17	–0.02
p	0.52	0.19	0.29	0.88
Breathiness				
rho	0.27	0.41*	0.20	0.16
p	0.08	0.007*	0.21	0.30
Pitch				
rho	0.20	0.25	0.36*	0.02
p	0.23	0.13	0.03*	0.91
Roughness				
rho	0.25	0.29	0.32*	0.07
p	0.12	0.06	0.04*	0.68
Strain				
rho	0.25	0.34*	0.27	0.07
p	0.11	0.03*	0.09	0.65

rho — Spearman rho correlation coefficient.
*Statistically significant ($p < 0.05$) and corresponding rho value.

DISCUSSION

Voice disturbance following airway reconstruction has been described previously by numerous authors.[1-10] Many of the early studies relied on the voice outcome analysis of a small group of children, making it difficult to draw firm conclusions about this complex population. Clary et al[8] studied the largest series to date, which included 50 subjects; however, the evaluation did not include the use of a validated health-related quality-of-life instrument to assess the impact of voice disturbance on activities of daily living. In this study, we sought to describe the relationship between the pVHI and CAPE-V scores in a group of children who had undergone airway

TABLE 3. IMPACT OF PROCEDURES ON pVHI AND CAPE-V DOMAIN SCORES

	Glottic Procedure (N = 5)	*Posterior Graft Laryngotracheoplasty (N = 10)*	*Cricotracheal Resection (N = 5)*	*Other Airway Procedures (N = 8)*	*p*
Percent pVHI	26 (15-43)	38 (1-55)	33 (23-49)	38 (9-87)	0.57
Percent CAPE-V	37 (11-58)	33 (5-98)	73 (47-77)	39 (15-98)	0.14
pVHI Emotional	4 (3-4)	8 (0-15)	8 (2-14)	5.5 (2-19)	0.57
pVHI Functional	11 (7-15)	10 (0-20)	12 (10-16)	13.5 (4-27)	0.65
pVHI Physical	9 (4-21)	15.5 (1-24)	14 (7-18)	19.5 (234)	0.41
Talkativeness	6 (5-7)	5 (3-7)	5 (4-6)	6.4 (4-7)	0.2
CAPE-V Loudness	12 (1-43)	19.5 (0-65)	37 (32-71)	10.5 (1-97)	0.24
CAPE-V Breathiness	11 (0-38)	15.5 (0-85)	32 (8-77)	6 (1-98)	0.39
CAPE-V Pitch	11 (1-65)	1 (0-76)	33.5 (11-77)	9 (2-45)	0.16
CAPE-V Roughness	27 (11-43)	15 (4-97)	73 (40-77)	12 (0-50)	0.059
CAPE-V Strain	27 (12-39)	16 (0-85)	24 (8-40)	4 (1-42)	0.19

Fourteen subjects were excluded from analysis for non–mutually exclusive procedures. Data are medians, with range in parentheses.

reconstruction. At this time, we know of only one other study that has sought to evaluate the relationship between voice-related quality-of-life measures and clinician-based scales of voice disturbance.[15]

The results from this investigation revealed only a fair correlation between the severity of the child's voice disorder and the scores reported on the pVHI. The lack of a clear, strong relationship in this patient population warrants discussion. Children who have undergone airway reconstruction represent a unique subset of patients. Most have been medically fragile from birth and have undergone numerous hospitalizations involving complex medical-surgical procedures. Parents have been focused on the child's survival and well-being. Their assessment of the impact of the child's voice outcome may be overshadowed by larger medical concerns, such as airway patency or issues related to overall development. We suspect that the focus on the child's voice quality changes during childhood. Voice quality may become more important after other, more threatening medical issues resolve and the focus is shifted to social and/or educational concerns. Clearly, the long-term impact of voice on quality of life in this patient population is not well understood.

TABLE 4. CORRELATION COEFFICIENTS FOR pVHI AND CAPE-V SCORES WITH NUMBER OF TOTAL AIRWAY PROCEDURES

	rho	*p*
CAPE-V		
Overall Severity	0.60*	<0.0001*
Loudness	0.40*	0.009*
Breathiness	0.34*	0.03*
Pitch	0.28	0.09
Roughness	0.24	0.13
Strain	0.27	0.09
pVHI		
Total	0.19	0.23
Emotional	0.25	0.10
Functional	0.18	0.25
Physical	0.14	0.37

*Statistically significant ($p < 0.05$) and corresponding rho value.

There were interesting relationships noted between subscores of the CAPE-V and pVHI that warrant attention. The strength correlation was strongest between the Overall Severity CAPE-V scores and the pVHI total and subset scores for functional and emotional domains. However, the physical subscore of the pVHI did not correlate with the Overall Severity CAPE-V score. Of the parameters assessed on the CAPE-V, Loudness did not correlate with any facet of the pVHI.

Similarly, the physical domain of the pVHI did not correlate with any aspect of the CAPE-V assessment. These discrepancies may be due to several factors. The physical domain of the pVHI may not be measured in the perceptual analysis. Much of the physical component questions relate to breath support and its effect on voice production. In addition, physical ratings can often be difficult to measure via proxy measures (eg, pVHI), and expert ratings of physical domains can likewise be unreliable (eg, CAPE-V Strain).[15]

The CAPE-V Loudness score may not correlate well with the pVHI score because of the fact that there are only a few questions within the pVHI that could be associated with loudness measures in the perceptual analysis. In this cohort, Loudness (eg, too soft) may not be perceived by parental proxy as a significant detriment to voice. Further analysis with a larger cohort and question-specific correlations may lead to better understanding of these re-

lationships.

Examination of the relationship between the severity of postsurgical voice disturbance and surgical factors revealed important trends. The subgroup analysis demonstrated that there are certain aspects of surgery that may be associated with greater voice disturbance as assessed by the CAPE-V. It was noted that children who had a history of cricotracheal resection tended to have more voice disturbance as measured by the CAPE-V than did those who had other procedures. Cricotracheal resection has the potential to significantly alter the anatomy and function of the laryngeal complex and thereby ultimately affect the voice outcome. A cricotracheal resection can result in injury to the cricothyroid muscle, significant anatomic changes to the subglottis, and obliteration of the cricothyroid space. These anatomic changes theoretically should result in problems with the ability to modulate pitch and vocal fold tension.

Interestingly, in our cohort, glottic procedures and posterior graft laryngotracheoplasty did not appear to be associated with higher Overall Severity CAPE-V scores or a higher parameter score, such as the CAPE-V Breathiness score. This finding was unexpected. It is possible that for patients who underwent glottic procedures (eg, patients with both subglottic stenosis and vocal fold paralysis), the overall voice may not have been affected, since many of the adjunctive procedures affect the non-vocalizing portion of the vocal fold. Posterior graft laryngotracheoplasty did not seem to be associated with CAPE-V scores in a significant manner, perhaps because of a change in our technique at Cincinnati Children's Hospital Medical Center. Since 1999, posterior graft laryngotracheoplasty has been performed in the majority of cases without the need for a complete laryngofissure to gain access to the posterior cricoid plate. Many of the children in the current cohort had complex laryngotracheal stenosis, and many underwent multiple airway procedures. These confounding aspects of the current cohort may make it difficult to draw conclusions regarding posterior graft laryngotracheoplasty.

There are several potential weaknesses to the current study. The cohort involves a very complex subset of patients who have undergone multiple and varied procedures on the airway that can affect voice. This is partly reflected in the subset analysis performed on the current cohort (eg, posterior graft laryngotracheoplasty, cricotracheal resection). We suspect that there are aspects to specific airway procedures that have a greater impact on postoperative voice outcomes and that the degree and type of airway stenosis can affect the ultimate voice outcome. Future studies to identify the impact that specific procedures have on the airway may elucidate more about the relationships with voice outcomes. Despite these weaknesses, the current study is an important advance in understanding the relationship between patient-derived, subjective voice quality and clinicians' perceptual assessments of voice disturbance (ie, the CAPE-V) in children who have undergone airway reconstruction.

CONCLUSIONS

Voice disturbance following airway reconstruction is a potentially important outcome measure. Evaluation of voice disturbance includes a variety of measures using videostroboscopy, aerodynamic and acoustic analyses, perceptual evaluations (eg, the CAPE-V), and voice-related quality of life (eg, the pVHI). In children who have undergone airway reconstruction, the median pVHI total score was 30 and the median Overall Severity CAPE-V score was 50.5. The Overall Severity CAPE-V score and the pVHI total score had a fair correlation in this patient population. Although there was not a clear, strong relationship between the CAPE-V scores and the pVHI scores, both of these tools provide important information regarding the relationship of the severity of voice disturbance to its handicapping effects.

REFERENCES

1. Baker S, Kelchner L, Weinrich B, et al. Pediatric laryngotracheal stenosis and airway reconstruction: a review of voice outcomes, assessment, and treatment issues. J Voice 2006;20: 631-41.

2. Zur KB, Cotton S, Kelchner L, Baker S, Weinrich B, Lee L. Pediatric Voice Handicap Index (pVHI): a new tool for evaluating pediatric dysphonia. Int J Pediatr Otorhinolaryngol 2007;71:77-82.

3. Smith ME, Marsh JH, Cotton RT, Myer CM III. Voice problems after pediatric laryngotracheal reconstruction: videolaryngostroboscopic, acoustic, and perceptual assessment. Int J Pediatr Otorhinolaryngol 1993;25:173-81.

4. Smith ME, Mortelliti AJ, Cotton RT, Myer CM III. Phonation and swallowing considerations in pediatric laryngotracheal reconstruction. Ann Otol Rhinol Laryngol 1992;101:731-8.

5. Krival K, Kelchner LN, Weinrich B, et al. Vibratory source, vocal quality and fundamental frequency following pediatric laryngotracheal reconstruction. Int J Pediatr Otorhinolaryngol 2007;71:1261-9.

6. Zalzal GH, Loomis SR, Derkay CS, Murray SL, Thomsen J. Vocal quality of decannulated children following laryngeal reconstruction. Laryngoscope 1991;101:425-9.

7. Zalzal GH, Loomis SR, Fischer M. Laryngeal reconstruction in children. Assessment of vocal quality. Arch Otolaryngol Head Neck Surg 1993;119:504-7.

8. Clary RA, Pengilly A, Bailey M, et al. Analysis of voice outcomes in pediatric patients following surgical procedures for laryngotracheal stenosis. Arch Otolaryngol Head Neck Surg 1996;122:1189-94.

9. MacArthur CJ, Kearns GH, Healy GB. Voice quality after laryngotracheal reconstruction. Arch Otolaryngol Head Neck Surg 1994;120:641-7.

10. Bailey CM, Clary RA, Pengilly A, Albert DM. Voice quality following laryngotracheal reconstruction. Int J Pediatr Otorhinolaryngol 1995;32(suppl):S93-S95.

11. Boseley ME, Cunningham MJ, Volk MS, Hartnick CJ. Validation of the Pediatric Voice-Related Quality-of-Life survey. Arch Otolaryngol Head Neck Surg 2006;132:717-20.

12. Hartnick CJ. Validation of a pediatric voice quality-of-life instrument: the pediatric voice outcome survey. Arch Otolaryngol Head Neck Surg 2002;128:919-22.

13. Hartnick CJ, Volk M, Cunningham M. Establishing normative voice-related quality of life scores within the pediatric otolaryngology population. Arch Otolaryngol Head Neck Surg 2003;129:1090-3.

14. Blumin JH, Keppel KL, Braun NM, Kerschner JE, Merati AL. The impact of gender and age on voice related quality of life in children: normative data. Int J Pediatr Otorhinolaryngol 2008;72:229-34.

15. Karnell MP, Melton SD, Childes JM, Coleman TC, Dailey SA, Hoffman HT. Reliability of clinician-based (GRBAS and CAPE-V) and patient-based (V-RQOL and IPVI) documentation of voice disorders. J Voice 2007;21:576-90.