Pediatric Exercise Stress Laryngoscopy following Laryngotracheoplasty: A Comparative Review

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Abstract

Objective. Exercise-induced airway obstruction in pediatric patients is a unique phenomenon with multiple potential etiologies. An accurate diagnosis can be challenging to establish in pediatric patients because they are frequently asymptomatic at rest. Exercise stress laryngoscopy (ESL) is a modality by which pediatric patients can be evaluated under physiologic conditions that produce their symptoms. The purpose of this study was to demonstrate (1) the diagnostic effectiveness of pediatric ESL and (2) the ability of ESL to guide treatment for “normal” and post-airway reconstruction patients with exercise intolerance.

Study Design. Case series with chart review.


Subjects and Methods. Patients undergoing ESL for exercise intolerance were reviewed. Demographics, surgical history, examination findings, and management recommendations were extracted.

Results. Thirty-seven patients (average age, 13.5 years; range, 5-21 years) were included. There were 14 male and 23 female patients. Airway abnormalities became evident in 56% of patients. Of these, 24% had focal supraglottic collapse, 43% had evidence of paradoxical vocal fold motion, 24% had combined supraglottic and glottic dysfunction, and 9% had distal airway abnormalities. Overall, 18 patients had changes in management after ESL. Twelve patients in this review had histories of laryngotracheoplasty with equivocal findings on operative bronchoscopy. Of these patients, 10 (83%) received focal diagnoses after ESL.

Conclusion. ESL is a contemporary modality by which complex patients with undiagnosed exercise intolerance can be effectively evaluated. ESL can be an important tool used to guide treatment in pediatric patients with exercise-induced dyspnea after airway reconstruction.

Keywords

exercise laryngoscopy, exercise-induced dyspnea, laryngotracheal reconstruction

Introduction

Exercise-induced dyspnea with or without stridor can be a challenging diagnosis with a multitude of etiologies. Historically, exercise intolerance secondary to laryngeal pathology is often misdiagnosed as reactive airway disease, and unnecessary treatment may be administered.1 These patients often undergo micro-laryngoscopy in conjunction with rigid and flexible bronchoscopy under various levels of sedation in the operating room for diagnostic purposes. Although operative endoscopy may result in an appropriate diagnosis under some circumstances, patients with exercise-induced symptoms frequently have normal findings in the operating room, resulting in a challenging diagnostic dilemma.

Over the past decade, flexible laryngoscopy during exercise has increasingly been used to evaluate patients with exercise-induced dyspnea or suspected laryngeal dysfunction during exertion.1-4 Exercise stress laryngoscopy (ESL) provides the practitioner with a unique opportunity to evaluate the larynx during exercise, better reflecting the physiologic conditions that are associated with dyspnea or stridor. At our institution, such patients are frequently referred with airway symptoms during exercise that may be diagnostically
elusive or do not respond to treatment. A subset of patients with dyspnea on exertion have undergone laryngotracheal surgery before presentation, confounding an already difficult diagnosis. Endoscopy in the operating room is an established component of postoperative surveillance after laryngotracheoplasty; however, ESL may be a beneficial component of the diagnostic algorithm, as it allows a precise airway evaluation during exertion in complex postoperative patients. The purposes of this investigation were to review our history in performing ESL over the past 2 decades and to identify the benefits and shortcomings of ESL in our operative and nonoperative patient populations. An exploratory analysis of differences in the pathologic processes leading to symptoms in patients with and without histories of prior laryngotracheoplasty (LTP) is presented.

Methods
This investigation was approved by the Cincinnati Children’s Hospital Medical Center Institutional Review Board. Patients aged ≤21 years who had undergone ESL for exercise-induced dyspnea or stridor (between January 1996 and January 2013) at this institution were included in this review. Age criteria were chosen on the basis of the US Department of Health and Human Services Food and Drug Administration definition of a pediatric patient. Patients with incomplete records or studies in which endoscopy was not performed were excluded.

Patient Demographics and History
Standard demographic data were extracted from patient records, including age at the time of ESL, race, and gender. Additional patient information included history of LTP, postprocedural diagnosis, and subsequent management after ESL. Patients without complete documentation or imaging data were excluded.

Procedure-Specific Data
Patients were evaluated for the presence or absence of visible airway dysfunction that (1) was observed to manifest during exercise and (2) resulted in subjective shortness of breath with or without stridor. Patients were categorized as having the visible pathologic process occurring at the level of the supraglottis, the glottis, combined (dysfunction of the supraglottis and glottis), or distal to the level of the vocal cords. Definitions of each category are as follows:

**Supraglottic:** Arytenoid, epiglottic, aryepiglottic, or false vocal fold prolapse during inspiration that occurs in the setting of symptoms but without dysfunction at the level of the glottis. Mild medialization of the cuneiform cartilages on inspiration has been described as a normal variant during stress laryngoscopy and was not considered an abnormal finding.

**Glottic:** Isolated adduction of the true vocal folds during inspiration without dysfunction at the level of the supraglottis.

**Combined:** Supraglottic collapse on inspiration with subsequent true vocal fold adduction (primary supraglottic collapse) or true vocal fold adduction with subsequent supraglottic collapse (secondary supraglottic collapse).

**Distal:** Tracheal collapse or malacia.

Standard ESL Procedure
The procedure is explained to the patient and parents, who then familiarize themselves with the equipment. The patient is placed on the treadmill and connected to a blood pressure cuff and a 12-lead electrocardiograph. A pulse oximeter and spirometer are also used. A nasopharyngoscope is placed as described later. The patient first walks slowly in the absence of an incline to become comfortable with the feel of the attached equipment. The treadmill speed and incline are increased until the patient becomes symptomatic or is determined to be at maximum exercise capacity.

Endoscopy. If necessary, the nose is topically anesthetized with a small quantity of nasal decongestant and viscous lidocaine. The endoscopist is positioned at the head of the bed and may choose to stand on a platform. A distal-chip flexible nasopharyngoscope is then inserted and manually suspended over the larynx. If indicated, distal endoscopy can be performed beyond the level of the glottis after the judicious application of topical anesthetic to the larynx. To reduce fatigue, an armrest that is secured to the treadmill may be used. Constant adjustment of the laryngoscope may be necessary to compensate for patient motion. The examination is recorded and archived.

Statistical Analysis and Comparison of Data
Data were collected in Excel (Microsoft Corporation, Redmond, Washington) and analyzed using Stata/SE 13 (StataCorp LP, College Station, Texas). Statistical comparison of proportions was performed using z tests for proportions with $\alpha = 0.05$. Power and sample size calculations were not performed, because analysis was exploratory and because adequate data do not yet exist to allow the determination of an appropriate effect size for the comparison of patients with and those without LTP undergoing ESL.

Results
Demographic Data and Patient History
A total of 37 patients were included in this review. There were more female patients, with a mean patient age of 13.5 years (range, 5-21 years). The majority of patients were Caucasian. Twelve patients had histories of LTP.

Endoscopic Findings
Specific airway abnormalities was recognized during ESL in 21 patients (56%). Of these, 5 patients (24%) had focal supraglottic collapse that became evident only on exertion (Figure 1). Nine patients (43%) had evidence of isolated
glottic dysfunction, 5 patients (24%) had combined dysfunction, and 2 patients (9%) had distal airway abnormalities (Figure 2). Focal airway abnormalities resulting in changes in management occurred in 18 of the 21 patients (86%). Of the 3 patients (14%) for whom treatment was not changed, 2 had distal collapse and 1 had a diagnosis of glottic dysfunction confirmed but was already receiving appropriate management. Patient demographics, history, and endoscopic findings are summarized in Table 1.

History of Airway Surgery

Prior LTP. Twelve patients in this study had histories of LTP, constituting 32% of the overall study sample. Ten of these 12 patients (83%) had focal abnormalities identified on ESL, constituting 48% of all patients with abnormal results.

No history of LTP. The remaining 25 patients in this study had no histories of LTP, constituting 68% of the overall study sample. Eleven of these 25 patients (44%) had focal abnormalities identified on ESL, constituting 52% of all patients with abnormal results.

When comparing patients with and without histories of LTP, significantly greater proportions of patients with airway abnormalities (z = –2.26, \( P = .02 \)), isolated supraglottic dysfunction (z = –2.44, \( P = .01 \)), and distal dysfunction (z = –2.10, \( P = .03 \)) were present in the population of patients with histories of LTP. There were no statistically significant differences between the LTP and non-LTP groups in the proportion of patients with isolated glottic dysfunction, or combined dysfunction, who had changes in management (\( P = .45 \), \( P = .69 \), and \( P = .67 \), respectively).

Discussion

ESL has been performed at our institution for nearly 2 decades and has been previously described in the literature as a validated method of identifying exercise-induced laryngeal obstruction.\(^7\) Several findings of this investigation are in keeping with previous reports. First, ESL appears to be a straightforward and effective method of evaluating exercise-induced dyspnea in pediatric patients. Second, we have been able to provide some preliminary insight into the subtle differences that may be expected when performing ESL in patients after airway reconstruction. To our knowledge, this report represents one of the first reviews of ESL in pediatric patients with exercise-induced dyspnea after LTP.

Effectiveness

Exercise laryngoscopy has been previously described as an effective tool in diagnosing laryngeal dysfunction during exercise and is recognized as a tolerable procedure in young patients.\(^1,2,4\) Our results appear to echo previous findings detailing the success of exercise laryngoscopy; ESL revealed previously undiagnosed findings that resulted in changes in management in the majority of patients with abnormalities identified. At our institution, patients with paradoxical vocal fold mobility are referred to a speech-language pathologist for evaluation and treatment. All patients with paradoxical vocal fold mobility in our review followed this protocol. Patients with supraglottic laryngeal pathology were managed with focused supraglottoplasty, intended to treat the specific site of obstruction identified on endoscopy. All patients had subjective improvements in symptoms after intervention; however, because the focus of this review was primarily diagnostic, objective outcomes data pertaining to treatment are not reported.

Some of the procedures commonly performed in our study population before ESL include microlaryngoscopy as well as both rigid and flexible bronchoscopy. The limitations of laryngoscopy and bronchoscopy in patients with...
exercise-induced dyspnea arise secondary to sedation requirements, bronchoscopic techniques, and the influences of airway instrumentation on airway dynamics. Patients undergoing laryngoscopy and rigid endoscopy require a deeper plane of anesthesia than those undergoing flexible bronchoscopy and have the potential for airway stenting by the laryngoscope and rigid bronchoscope. However, neither rigid nor flexible bronchoscopy, when performed under anesthesia, will routinely or reliably provide an airway evaluation that is equal to that achieved in a non-sedated patient who is actively exercising in the presence of symptoms (Figure 1).

Patient Tolerance

On the basis of our experience in performing ESL, we feel that it is tolerable in appropriately selected pediatric patients. Although some authors have reported discouraging experiences with patient cooperation or tolerance during pediatric ESL, the largest studies to date suggest otherwise.2,8 In a 2009 study conducted at Haukeland University Hospital in Norway, a 91% success rate was achieved in performing ESL on 166 young patients with exercise-induced dyspnea. The average patient age in that study was approximately 16 years.2 The average age of our patient population was 13.5 years, and several patients were <10 years of age. Thus, the success rate of ESL largely parallels that of resting transnasal laryngoscopy in the clinic setting. Specifically, maintaining a calming and explanatory demeanor, developing effective patient and parent rapport, and the judicious use of topical anesthesia when needed all contribute to the success of ESL. Nevertheless, patient tolerance, although acceptable, will certainly vary among patients on the basis of age, medical history, and environment. On the basis of anecdotal evidence at our institution, we share the opinion of Halvorsen et al9 that this procedure can be readily performed in most young patients.4

Laryngeal Abnormalities Identified with ESL

Several focal diagnoses were identified in this review. Glottic dysfunction was the most frequent isolated abnormality observed, followed in equal parts by supraglottic collapse and combined dysfunction. The specific glottic abnormality identified was paradoxical vocal fold motion manifesting during exertion, occurring in 24% of the study sample. Paradoxical vocal fold dysfunction is an established phenomenon, known to have a female predilection and to produce symptoms such as cough, dyspnea, and extrathoracic airway obstruction.10,11 Although these characteristics are consistent with our results, the prevalence of this diagnosis in our cohort falls slightly below that previously reported. In a recent study of 88 athletes evaluated for exercise-induced dyspnea using exercise laryngoscopy in the United Kingdom, exercise-induced laryngeal obstruction was identified in 35% of patients.11 In a 2009 report by Tervonen et al12, reproducible dyspnea during exercise was determined to be due to vocal cord dysfunction in 60% of patients. Supraglottic collapse was considered a component of exercise induced vocal cord dysfunction in that study and was used to differentiate laryngeal dysfunction from exercise-induced asthma. One explanation for our slightly lower prevalence compared with prior studies may be our definition of glottic dysfunction. Vocal fold dysfunction during exercise was specifically defined as the inappropriate adduction of the vocal folds with exertion as an isolated finding. By broadening this definition, and when evaluating the entire study sample, our findings are consistent with those of other studies.10-12

Supraglottic and combined laryngeal dysfunction each occurred in 24% of the patients with abnormal findings on ESL. Supraglottic dysfunction is a known phenomenon in patients with and without histories of LTP.12,13 In the previously cited study by Roksun et al,2 exercise laryngoscopy demonstrated laryngeal abnormalities in 75% of the 151-patient study population. Findings in their study included supraglottic (arytenoid) medialization, with many patients demonstrating subsequent vocal fold medialization. The authors differentiated these findings from supraglottic collapse in isolation and secondary supraglottic collapse preceded by vocal fold medialization. In our sample, when considering all laryngeal dysfunction, including isolated paradoxical vocal fold motion, abnormalities were identified on ESL in 51% of patients. However, when considering

### Table 1. Patient Demographics, Endoscopic Findings, and Surgical History.

<table>
<thead>
<tr>
<th>Variable</th>
<th>All Patients (n = 37)</th>
<th>Patients with Histories of LTP (n = 12)</th>
<th>Patients with No Histories of LTP (n = 25)</th>
<th>P value, LTP vs No LTP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, y, mean ± SD (range)</td>
<td>13.5 ± 3.6 (5-21)</td>
<td>12.9 ± 4.1 (6-19)</td>
<td>13.7 ± 3.4 (5-21)</td>
<td>.53</td>
</tr>
<tr>
<td>Female gender</td>
<td>23 (62%)</td>
<td>5 (42%)</td>
<td>18 (72%)</td>
<td>.08</td>
</tr>
<tr>
<td>New airway abnormality diagnosed</td>
<td>21 (57%)</td>
<td>10 (83%)</td>
<td>11 (44%)</td>
<td>.02</td>
</tr>
<tr>
<td>Supraglottic dysfunction</td>
<td>5 (14%)</td>
<td>4 (33%)</td>
<td>1 (4%)</td>
<td>.01</td>
</tr>
<tr>
<td>Glottic dysfunction</td>
<td>9 (24%)</td>
<td>2 (17%)</td>
<td>7 (28%)</td>
<td>.45</td>
</tr>
<tr>
<td>Combined dysfunction</td>
<td>5 (14%)</td>
<td>2 (17%)</td>
<td>3 (12%)</td>
<td>.69</td>
</tr>
<tr>
<td>Distal airway abnormality</td>
<td>2 (5%)</td>
<td>2 (17%)</td>
<td>0 (0%)</td>
<td>.03</td>
</tr>
<tr>
<td>Change in management as a result of ESL</td>
<td>19 (51%)</td>
<td>8 (67%)</td>
<td>10 (40%)</td>
<td>.67</td>
</tr>
</tbody>
</table>

Abbreviations: ESL, exercise stress laryngoscopy; LTP, laryngotraceheoplasty.
only those patients with supraglottic or combined dysfunction, only 27% of the entire study sample was affected. The discrepancy may be a result of the inclusion criteria for this investigation, which evaluated all patients undergoing ESL with exercise-induced dyspnea from any referring source and was thus broad in comparison with the aforementioned study. In contrast, the investigation by Røksund et al included patients recruited from the pulmonary medicine department with histories of exercise-induced dyspnea, with symptoms of exercise-induced laryngeal obstruction, and without evidence of exercise-induced asthma on exercise provocation testing. This may have selected for patients with exercise-induced laryngeal dysfunction by virtue of study inclusion criteria and the experience of the referring source, thereby increasing the incidence of positive findings on stress laryngoscopy.

Of the 3 patients with diagnoses made on ESL but for whom treatment was not changed, 2 had distal collapse and 1 had a diagnosis of glottic dysfunction confirmed. The patient with glottic dysfunction had a preliminary diagnosis of paradoxical vocal fold dysfunction and had begun appropriate intervention before ESL. The 2 patients with distal collapse did not have specific treatment implemented; however, focal origins of the patients’ symptoms were identified. Although changes in management did not occur for these patients, the diagnosis established using ESL is not trivial. Informing a patient of the origin of his or her symptoms is an empowering process and can be a critical step in reducing symptoms. Not only does it alleviate the anxiety of an unknown diagnosis, it obviates the need for additional diagnostic studies and eliminates the administration of inappropriate treatment.

History of LTP

Patients with histories of laryngotracheal surgery present practitioners with an increased level of complexity when performing an evaluation of the airway. Many patients have an abnormal appearance to the airway after 1 or several laryngotracheal operations; therefore, the onus is on the physician to determine which abnormalities are contributing to a patient’s symptoms. Additionally, patients with histories of LTP may have altered structural integrity of a segment of the airway despite a normal appearance at rest. This area may become unstable only during specific stimuli such as exercise. For this reason, ESL may have even more utility in patients with exercise-induced dyspnea after LTP.

When comparing patients with histories of LTP with those patients without, several findings of this study warrant mention. With respect to the distribution of patients, the population of individuals in this study with histories of LTP comprised one third of the overall study population but nearly one half of those with abnormalities identified on endoscopy. Compared with patients without histories of LTP, those who had previously undergone LTP were more likely to have abnormalities identified using ESL. In addition, the proportions of patients with supraglottic collapse and distal dysfunction were also significantly greater in the LTP population. These findings may be important for several reasons. Patients with histories of LTP often have comorbidities such as swallowing dysfunction or intermittent aspiration that are related to their previous surgery. As a result, it is important establish a definitive diagnosis before any intervention that could increase the risk for aspiration or potentiate swallowing dysfunction. Likewise, it is also important to specifically identify and treat only the structures leading to a patient’s dyspnea on exertion.

Study Strengths and Limitations

In this report, we were able to share our experience using ESL in a complex population of pediatric patients with exercise-induced dyspnea. Our results provide valuable information for practitioners attempting to narrow the differential diagnosis in patients with exercise-induced dyspnea and provide preliminary insight into some of the potential differences expected for a patient with a history of airway reconstruction.

Despite our findings, several limitations merit mention. First, the relatively small study population limits the power of our findings, and any statistical analysis must be considered preliminary. Nevertheless, our goal to provide early insight into a population of patients with an uncommon diagnosis was achieved by producing what can be considered hypothesis-generating results.

Second, the long time period over which this review was performed leads to inherent variability over time in individual practitioners, practice and referral patterns, and ESL technique. This shortcoming is a consequence of our retrospective study design and remains an important consideration for the reader.

Third, our comparison of patients with and without LTP was exploratory and designed for hypothesis generation rather than hypothesis testing.

Finally, although ESL is readily performed on many young patients, some individuals may not have been included in this review because of comorbidities or the inability to tolerate the procedure despite meeting the inclusion criteria otherwise. Because patients who did not tolerate the examination were not consistently recorded over time, our ability to objectively define our rate of success with ESL is restricted. Nevertheless, anecdotal experience suggests that we are able to perform ESL on the vast majority of patients who are selected to undergo the evaluation.

Conclusions

ESL can be effectively used to evaluate select pediatric patients with exercise intolerance. This procedure may be particularly useful in establishing a diagnosis and guiding the treatment of patients with exercise-induced dyspnea after laryngotracheal surgery. After LTP, patients with exercise intolerance may be more likely to have airway abnormalities diagnosed on ESL, supraglottic collapse, and distal airway collapse.
Author Contributions

Douglas R. Sidell, study design, patient evaluation, data collection, data review and analysis, manuscript preparation; Karthik Balakrishnan, data review and analysis, manuscript preparation; Catherine K. Hart, patient evaluation, data analysis, manuscript preparation; J. Paul Willging, study design, patient evaluation, data analysis, manuscript preparation; Sandra K. Knecht, study design, patient evaluation, data collection, data review; Alessandro de Alarcon, study design, patient evaluation, data review and analysis, manuscript preparation.

Disclosures

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