Surgical Intervention for Exercise-Induced Laryngeal Obstruction

John-Helge Heimdal, MD, PhD[a,b,*], Robert Maat, MD, PhD[c], Leif Nordang, MD, PhD[d]

KEYWORDS

• Exercise • Laryngeal obstruction • Surgery • Laryngoplasty • Laryngomalacia

KEY POINTS

- The larynx quickly adapts to increased airflow during activity by inspiratory abduction of the aryepiglottic folds and vocal folds, which increases laryngeal aperture and decreases airflow resistance.
- Respiratory distress during strenuous exercise may be due to malfunction of this adaptive mechanism causing airflow obstruction in the larynx in patients with exercise-induced laryngeal obstruction (EILO).
- Laryngeal obstruction caused by inward rotation of aryepiglottic folds (supraglottic) shows similar findings as laryngomalacia in infants.
- Supraglottic laryngeal obstruction can be treated successfully with operative techniques that are also used in infants with laryngomalacia.
- Key elements in surgical treatment of EILO are presented including selection criteria for surgery, procedural details, outcomes, and risk factors.

Video content accompanies this article at http://www.immunology.theclinics.com.

INTRODUCTION

During exercise, ventilation increases substantially. High airflow presumably causes notable negative pressures at narrow portions of the airways, including the larynx.

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[a] Department of Surgery, Haukeland University Hospital, Bergen University, Bergen, Norway; [b] Department of Otorhinolaryngology, Head and Neck Surgery, Haukeland University Hospital, Bergen University, Bergen, Norway; [c] Department of Otolaryngology, Röpcke-Zweers Hospital, Jan Weitkamplaan 4A, Hardenberg 7772 SE, The Netherlands; [d] Department of Surgical Sciences, Otorhinolaryngology, and Head and Neck Surgery, Uppsala University, Akademiska sjukhuset ing 78-79, Uppsala 75185, Sweden

* Corresponding author. Kirurgisk klinikk, Helse Bergen, Haukelandsveien 22, Bergen 5021, Norway.

E-mail address: john.heimdal@helse-bergen.no

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The larynx quickly adapts to the ventilatory demands of exercise through abduction of the glottic as well as supraglottic structures. Exercise-induced laryngeal obstruction (EILO) is a condition in which the larynx fails to remain fully patent during exercise because of dysfunction at a glottic or supraglottic level, and is characterized by typical symptoms of respiratory distress, dyspnea, stridor, and wheezing.[1]

Smith and colleagues described the condition in which EILO appeared largely isolated to the supraglottic structures. At the time of publication, the authors named their finding exercise-induced laryngomalacia (now known as EILO-supraglottic type), because they observed exertional laryngeal motions that appeared similar to those seen in infants with congenital laryngomalacia (CLM).[2,3] CLM, which is anatomically characterized by supraglottic laryngeal collapse, is the most common cause of stridor in infants and children, and similar conditions can occur throughout later childhood and adulthood.[4–6] Following initial reports of the condition, larger series were published, indicating that EILO-supraglottic type may be more common than previously recognized.[7] Patients with CLM that persists into childhood and adulthood may experience symptoms at rest[8,9] or during exercise alone.[5] There are reports indicating that CLM may increase the risk for EILO later in life, although a definite relationship between CLM and EILO has not yet been verified.[10]

Experience from the surgical treatment of CLM in infants has now been applied with clinical success in both children and youths with laryngomalacia, including some with EILO-supraglottic type.[2] Initial published case reports have been followed by larger series of patients treated surgically for EILO-supraglottic type.[11–13]

This article summarizes the surgical experience of the treatment of EILO-supraglottic type. It will describe the proposed indications for surgery, technical surgical considerations, outcomes, and complications. In areas not clearly guided by literature, surgical experience of the authors will be included.

LITERATURE REVIEW

Systematic searches in Medline databases, available through PubMed, have revealed descriptions of surgical treatment to a total of 64 EILO patients through the end of 2016 (**Table 1**). In recent years, the number of reports of successful treatment of supraglottic laryngeal collapse has increased substantially. Children and adults are included in these reports.

Table 1
Previous reports of surgically treated exercise-induced laryngeal obstruction

Authors, Year	Number	Examination
Smith et al,[2] 1995	1	Exercise and laryngoscopy
Bent et al,[14] 1996	2	Exercise and laryngoscopy
Björnsdóttir et al,[15] 2000	2	Exercise and laryngoscopy
Chemery et al,[20] 2002	1	Exercise simultaneous laryngoscopy
Mandell et al,[16] 2003	1	Laryngoscopy and spirometry
Richter et al,[6] 2008	3	Exercise laryngoscopy
Maat et al,[7] 2011	23	Exercise simultaneous laryngoscopy
Norlander et al,[12] 2015	14	Exercise simultaneous laryngoscopy
Mehlum et al,[13] 2016	17	Exercise simultaneous laryngoscopy
Total number	64	

Initial Case Reports

Smith and colleagues published the first paper describing supraglottoplasty in a patient with EILO-supraglottic type in 1995. Removal of the corniculate cartilages by laser resulted in improvement in aerobic endurance as measured by unspecified parameters in physical fitness testing.[2] Bent and colleagues[14] published a combined report that included the patient in the Smith report, as well as a second patient, and concluded a positive outcome in response to carbon dioxide laser supraglottoplasty featuring removal of the corniculate cartilages, as this second patient was significantly improved. In 2000, Björnsdóttir and colleagues described treatment with surgical removal of superfluous mucosa at the rim of the aryepiglottic folds and superfluous mucosa on the tuberculum corniculatum and tuberculum cuneiforme in 2 patients with EILO-supraglottic type.[15] A positive outcome was concluded based on unspecified resolution of symptoms that persisted for 3 years. In 2003, Mandell and Arjmand presented a case of surgical treatment of EILO-supraglottic type in a 10-year-old patient via staged sequential unilateral procedures featuring a releasing incision in each of the aryepiglottic folds and excision of the corniculate cartilages with surgical scissors.[16] A positive response was concluded based on a change in the inspiratory portion of the resting flow-volume loops and self-reported "increased exercise tolerance." Gessler and co-workers described a 27 year-old female with a condition that they described as an adult type of laryngomalacia characterized by inspiratory stridor.[8] This specific patient described symptom onset during exercise followed by symptom persistence for days at rest (a presentation highly atypical for EILO-glottic type),[17] which did not respond to reflux therapy or corticosteroids. She was successfully treated (based on visual description of postoperative laryngoscopy during deep inspiration) with carbon dioxide laser excision of redundant arytenoid mucosa as well as the corniculate and cuneiform cartilages.

SUPRAGLOTTOPLASTY AS A TREATMENT FOR EXERCISE-INDUCED LARYNGEAL OBSTRUCTION-SUPRAGLOTTIC TYPE: TECHNICAL CONSIDERATIONS

Supraglottoplasty is performed under general anesthesia by suspension laryngoscopy.[11] The laryngoscope is introduced to the vallecula and suspended in a position that exposes the aryepiglottic folds or epiglottis. The operating microscope improves visualization of the larynx. Technically the operation can be performed either by using surgical scissors or carbon dioxide laser.

The general aim of supraglottoplasty for EILO is to reduce supraglottic collapse with the rationale that this will improve inspiratory airflow through the larynx during exercise. In this procedure, surgeons increase the diameter of the laryngeal inlet (via elongation of the aryepiglottic folds, excision of redundant tissue, and possible rotation of the epiglottis toward the tongue base) and attempt to increase the structural integrity of the supraglottic structures. Specific anatomic decisions can be guided by findings on preoperative continuous laryngoscopy during exercise testing.

Elongation of the aryepiglottic folds is most often achieved by incisions in the upper rim of the aryepiglottic folds close to the lateral rim of the epiglottis, anteriorly to the corniculate tubercles. The incisions extend inferiorly to the superior border of the ventricular folds[11] (**Fig. 1**, Video 1). Reduction of excess tissue on the aryepiglottic folds, fixation of the epiglottis, or reduction of its size can be included in the operative procedure. Maat and colleagues[11] incised the aryepiglottic folds bilaterally close to the epiglottis and created a circular mucosa incision at the top of the corniculate tubercle to remove redundant mucosa.

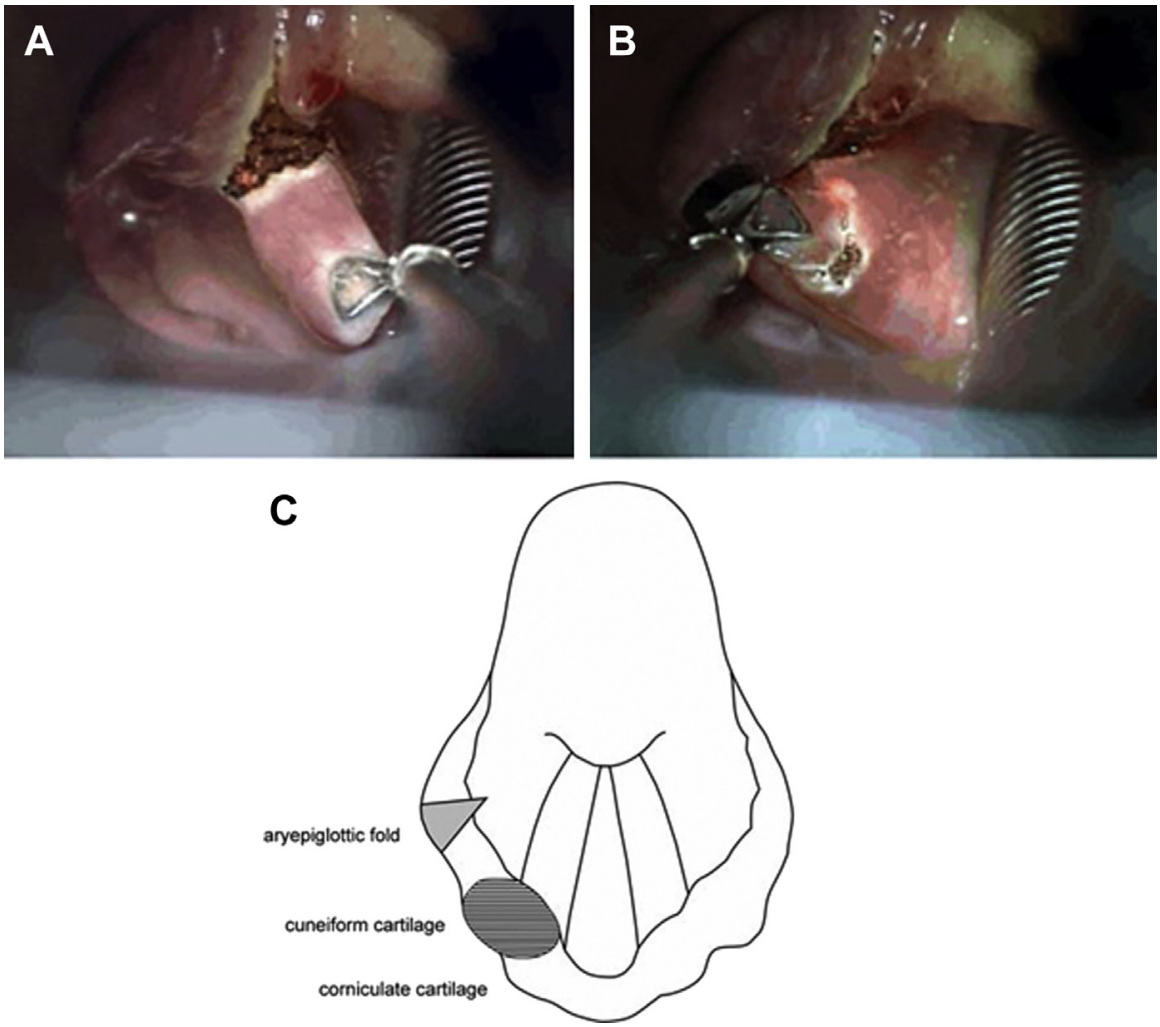

Fig. 1. Supraglottoplasty. (*A*) A CO_2 laser is used to make an incision in the aryepiglottic fold and (*B*) to remove the top of the cuneiforme (tubercle) cartilage and surrounding tissue. (*C*) Schematic drawing of the supraglottoplasty. (*From* Maat RC, Roksund OD, Olofsson J, et al. Surgical treatment of exercise-induced laryngeal dysfunction. Eur Arch Otorhinolaryngol 2007;264(4):403; with permission.)

SURGICAL INDICATIONS

At the current time, the indication for surgery in patients with refractory EILO is based solely on expert opinion. Maat and colleagues[7] proposed surgically treating patients with symptomatic EILO limited to the supraglottis. It is important that symptoms that are reported during CLE are similar in quality and quantity to field symptoms, because patients who do not fully reproduce symptoms in hospital settings may struggle with EILO that intermittently affects the glottis (which is likely unresponsive to surgical treatment).

It is the authors' opinion that surgery should be limited to patients with moderate or severe obstruction based on visual assessment and standardized scoring of CLE videos, because mild obstruction (slight inward rotation of the apex of the corniculate cartilage) may be a normal finding.[18] The authors recommend limiting surgery to those experiencing notable changes in quality of life attributable to the upper airway obstruction as defined by the patient. Finally, the authors recommend limiting surgery to patients who are fully informed of the potential risks and complications of this surgery and its irreversible nature. At the current time, given the series of limitations, surgery is likely to be limited to a few selected cases, with severe, refractory symptoms attributable to visualized supraglottic obstruction.

PUBLISHED OUTCOMES

Uncontrolled reports show a general trend indicating that surgery has a beneficial impact on EILO-related symptoms (Table 2). Maat and colleagues found that EILO patients reported fewer symptoms at 2- to 5-year follow-up than similar, conservatively treated patients using unvalidated symptom measures.[7] Supraglottoplasty seemed to have a lasting effect on EILO-related symptoms, and surgically treated patients were more physically active than nonsurgically treated patients with EILO at the time of follow-up. Mehlum and colleagues reported symptom improvement in surgically-treated patients in their follow-up study of 17 patients in 2016, using changes in visual analog scales of symptoms as a measure in an uncontrolled population.[13] Norlander and colleagues[12] reported improved symptom scores as rated by VAS and improved activity levels at the end of the follow-up period in surgically treated patients when compared with conservatively treated controls with EILO. Based on experience from these 48 patients treated with supraglottoplasty for EILO, the have preliminary data that suggest that the procedure is safe with effects lasting for several years.

Overall, although these reports are encouraging, overall assessment of surgical efficacy must be guarded. Analysis of the comparison studies is clearly confounded by selection bias, which likely leads to enrollment of highly motivated patients in groups that received surgery. Publication bias and reporting bias could theoretically enrich the literature with reports of successful cases. For this reason, the authors recommend surgery be reserved for highly selected cases with a frank and open discussion about the risks undertaken with any individual considering this approach to EILO treatment.

COMPLICATIONS

Analysis of medical literature on surgical complications in aggregate has revealed that supraglottoplasty is a safe procedure in neonates and children with CLM, as aspiration and airway stenosis are rare.[19] To the authors' knowledge no serious complications of supraglottoplasty in patients with EILO-supraglottic type have been reported. The 64 patients included in reports have to date not experienced serious adverse effects according to authors. However, long-term data with respect to complications related to aspiration are needed. The number of published procedures so far is limited and may underestimate rare complications. The authors recommend that surgeons publish complications or create surgical registries in order to assess rare procedural complications.

CURRENT CHALLENGES AND AREAS FOR FUTURE RESEARCH REGARDING SURGICAL MANAGEMENT OF EXERCISE-INDUCED LARYNGEAL OBSTRUCTION

There are several important issues to consider regarding the surgical treatment of EILO-supraglottic type. First, there are many unanswered questions related to

Table 2
The studies from Table 1 including pre-and post-operative evaluations with visual analogue scale (VAS) or CLE-score

Author, Year	Number	CLE score[18] Pre vs Post	Number	VAS Pre vs Post
Maat et al,[7] 2011	19	3.0 v.s. 1.7	23	90/30
Norlander et al,[12] 2015	0	—	14	8.2/3.8
Mehlum et al,[13] 2016	11	4.2 v.s. 3.2	11	78/33

disease mechanism. Is this a purely anatomic condition primarily affected by airway size, tissue pliability, and airflow? Are other factors equally important, including tissue reactivity, extrinsic triggers, breathing mechanics, and cognitive-behavioral factors? Are EILO-glottic type and EILO-supraglottic type part of the same disease spectrum, or are they separate entities with separate mechanisms? Some have hypothesized that supraglottic obstruction may cause glottic obstruction based on observations that have occurred across time in specific CLE examinations.[1] No specific data regarding mechanism of EILO-supraglottic type exist at the current time. This poor understanding of disease mechanism extends to a poor understanding of normal muscular function in the larynx during high-intensity exercise. Such an understanding is critical, as it will guide the development of future behavioral interventions as well as identification of specific surgical targets within the procedure.

Second, the specific location of surgical intervention within treatment algorithms is unclear. In reality, the role of surgery likely varies across centers in part because the availability and effectiveness of nonsurgical interventions (including speech-language pathology, inspiratory muscle training, and behavioral health consultations) also varies across centers. In order to address this challenge, the authors recommend standardization and assessment of nonsurgical interventions used in the treatment of EILO-supraglottic type. Inherent in this recommendation is identifying the need to quantitatively define normal function and quantitatively assess airway obstruction and symptom burden. Such quantitative assessment tools will also be useful in the assessment of surgical effectiveness.

Third, there are many unanswered questions regarding specific details of surgical technique during supraglottoplasty for EILO-supraglottic type. Although a general understanding of normal laryngeal function exists, the effect of supraglottoplasty for EILO on this function may be complicated. Which specific maneuvers within the surgery have the highest likelihood of improving symptoms and airflow? Which specific have the highest likelihood of increasing risk of complications? Are there maneuvers that can improve function without dramatically altering structure?

Finally, there are important questions about surgical outcomes. With fewer than 100 cases reported in the literature, none with follow-up reported into later life, it is reasonable to view surgery for EILO-supraglottic type cautiously. What percentage of patients improve to their baseline status postoperatively? Are there predictors of response to surgical intervention? Are there predictors of nonresponse? What are the long-term benefits and complications seen?

SUMMARY

Anatomic features of EILO-supraglottic type observed during CLE that resembled those seen in infants with CLM led to the extension of surgical interventions from CLM to patients with EILO-supraglottic type. Case reports and case controlled series of surgical interventions for this condition feature data that provide the rationale for future study of supraglottoplasty for refractory EILO-supraglottic type. Many unanswered questions remain regarding disease mechanism, indications for surgery, surgical technique, and outcomes. For these reasons, at the current time, the authors recommend consideration of supraglottoplasty only in patients with high impact, refractory symptoms clearly attributable to visualized EILO-supraglottic type who are clearly informed about the limited literature in the area, permanent nature of the procedure, and lack of long-term outcomes data.

SUPPLEMENTARY DATA

Supplementary data related to this article can be found online at https://doi.org/10.1016/j.iac.2018.01.005.

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