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Implementation of the European Laryngological Society classification for pediatric benign laryngotracheal stenosis: a multicentric study

Ivana Fiz[1] · Philippe Monnier[2] · Jan Constantin Koelmel[1] · Diana Di Dio[1] · Michele Torre[3] · Francesco Fiz[4,5] · Francesco Missale[6] · Cesare Piazza[6] · Giorgio Peretti[7] · Christian Sittel[1]

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Abstract

Purpose The European Laryngological Society (ELS) has published a revised classification for benign laryngotracheal stenosis (LTS), based on their degree, longitudinal extension, and associated comorbidities. We retrospectively applied this classification to pediatric patients treated in four referral centers to assess its reliability in predicting surgical outcomes.

Methods We included 191 pediatric LTS patients treated by segmental resection, restaged according to the degree of stenosis (I–IV according to Myer–Cotton grading system), number of subsites involved ("a" to "d" for 1–4 subsites among supraglottis, glottis, subglottis and trachea), and presence of systemic comorbidity ("+" sign). We analyzed the ability of this scoring system in predicting the rates of decannulation and complications, as well as the number of re-treatments.

Results The mean decannulation rate was 88%; a higher rate was observed in patients without comorbidities (95.7% vs. 78.1%, $p < 0.001$), with two or fewer vs. three or four subsites involved (89% vs. 72%, $p < 0.01$), and in those with an ELS score of IIIa+ or less vs. patients with IIIb or more (96% vs. 82%, $p < 0.001$). Surgical complications were not dependent on the degree of stenosis, but rather on the number of affected subsites ($p < 0.05$), as well as on the presence of associated comorbidities (RR 7.5, $p < 0.01$). The number of re-treatments was dependent on length of resection ($p < 0.05$), stage according to the revised ELS classification ($p < 0.001$), and presence of surgical complications (RR 17, $p < 0.001$).

Conclusions The revised ELS classification system is easy to apply in everyday practice and offers a sound contribution in the decision-making process.

Keywords Pediatric laryngotracheal stenosis · Laryngeal stenosis classification · ELS score · Reconstructive airway surgery · Cricotracheal resection

Introduction

Benign laryngotracheal stenosis (LTS) in children is considered to be one of the most common causes of chronic upper airway obstruction [1]. Approximately 90% of cases of acquired subglottic stenosis in infants and children are due to endotracheal intubation, with a reported incidence of 1–10% per procedure [2]. Caustic ingestion, foreign bodies, and iatrogenic origin due to laryngeal surgery may also result in LTS [2]. Finally, the incidence of congenital

✉ Ivana Fiz
ivana.fiz.orl@gmail.com

Philippe Monnier
monnier.phido@gmail.com

1 Department of Otorhinolaryngology Head and Neck Surgery, Katharinenhospital, Kriegsbergstrasse 60, 70174 Stuttgart, Germany

2 Airway Unit, Service of Otorhinolaryngology, Lausanne University Hospital, Lausanne, Switzerland

3 Department of Pediatric Surgery, G. Gaslini Children's Hospital, Genoa, Italy

4 Nuclear Medicine Unit, Department of Radiology, University of Tuebingen, Hoppe-Seyler-Strasse 3, 72076 Tübingen, Germany

5 Department of Internal Medicine, University of Genoa, Genoa, Italy

6 Department of Otorhinolaryngology, Maxillofacial and Thyroid Surgery, Fondazione IRCCS, National Cancer Institute of Milan, University of Milan, Milan, Italy

7 Department of Otorhinolaryngology, Head and Neck Surgery, IRCCS Ospedale Policlinico San Martino, University of Genoa, Largo R. Benzi 10, 16132 Genoa, Italy

subglottic stenosis is suspected to be around 5%. However, congenital alterations can be a contributing factor in acquired LTS, causing so-called acquired-on-congenital (AOC) stenosis [2].

In the last decades, a number of LTS classification systems were progressively introduced [2–14]. Cotton mentioned one of the first grading systems for pediatric stenoses, based on the percentage of airway obstruction [3]. Ten years later, Myer and Cotton modified it and proposed a new system consisting of four LTS grades related to the percentage of airway obstruction, determined using different sizes of endotracheal tubes [11]. As endotracheal tubes were widely available, and the system was easily reproducible, this classification has until now remained the most widely used in everyday practice. A further classification model, based solely on the number of involved subsites, has also been proposed by Lano and Netterville [12]. However, similar to the Myer–Cotton staging system, this and similar classifications are not able to accurately predict the likelihood of decannulation [12, 13].

With the introduction of cricotracheal resection (CTR) as a surgical treatment of LTS, mere evaluation of stenosis grade alone was no more considered a reliable predictor of decannulation rates, especially if scar tissue extends to the glottis or if patient had severe associated comorbidities. It was then hypothesized that concentrating on one single characteristic of the stenosis at a time (be it its degree or cranio-caudal extent) could not accurately depict the patients' status and thus the success of intervention. Consequently, in the last decade, a new generation of classifications was introduced to integrate important predictors of surgical outcomes in a single system [13–16]. This decision was grounded on the evidence that not just the grade of stenosis, but also its longitudinal extension and patient comorbidities, play a crucial role in predicting outcomes of airway reconstructive surgery [13, 14, 16, 17]. Furthermore, children affected by benign LTS often suffer from other ailments, including airway, cardiovascular or esophageal comorbidities, which can significantly affect surgical outcomes [16, 17].

In light of this, the European Laryngological Society (ELS) published a consensus paper in 2015 that offered a new classification system of benign LTS in adults and children [17]. It included a routine five-step endoscopic evaluation of patients, to assess the grade of stenosis (Cotton–Myer grade I–IV), number of subsites involved ("a–d" for one to four of supraglottic, glottic, subglottic, and tracheal segments involved), and an additional plus sign ("+") for patients with severe airway or medical comorbidities.

Herein, we tested the ability of this revised classification system in predicting surgical outcomes in a pediatric population affected by benign LTS and treated by tracheal resection (TR) or CTR in four referral centers.

Materials and methods

Patient population

We retrospectively collected clinical data on 191 pediatric patients (mean age 4.9 ± 4.2 years; range 0.09–14 years) affected by congenital, acquired or AOC benign LTS, and treated by TR or CTR at 4 Departments of Otorhinolaryngology: Lausanne University Hospital, Switzerland (141 patients), Katharinenhospital in Stuttgart, Germany (33 patients), Pediatric Hospital "Istituto Giannina Gaslini" in Genoa, Italy (10 patients), and University Hospital of Brescia, Italy (7 patients) between March 1978 and July 2017 (Table 1). Procedures were performed by six different surgeons. Expansive procedures, such as LTR, were not included in the present study. Legal guardians signed a written informed consent form, which was reviewed and approved by the respective local Ethics Committees and included the utilization of anonymized patients' data for research purposes. As the study dealt with the off-line

Table 1 Patients' characteristics

	Number (%)
Gender	
Male	115 (60.2)
Female	76 (39.8)
Age	
< 12 months	32 (16.8)
> 12 months	159 (83.2)
Etiology	
Congenital	35 (18.3)
Acquired	130 (68.1)
Acquired on congenital	26 (13.6)
Comorbidities	
Airway	87 (45.5)
Other medical	92 (48.2)
Preoperative tracheotomy	168 (88)
Previous treatment	71 (37.2)
Stenosis grade (Myer–Cotton)	
Grade II	13 (6.8)
Grade III	125 (65.4)
Grade IV	53 (27.8)
ELS score	
< IIIa	78 (40.8)
> IIIb+	113 (59.2)
Type of performed CTR	
Type A	8 (4.2)
Type B	31 (16.2)
Type C	113 (59.2)
Type D	39 (20.4)

retrospective analysis of anonymized data, specific approval from the Institutional Review Boards was waived in all cases.

Preoperative work-up

The preoperative work-up has been evolving in each center during the prolonged period of time comprised in our study. However, endoscopic examination including detailed airway assessment was always performed. This age-dependent step comprised awake or asleep transnasal fiberoptic laryngoscopy (TNFL) under general anesthesia in spontaneous respiration for dynamic assessment of vocal fold mobility. Further on, direct laryngotracheoscopy in spontaneous ventilation using 0° and 30° rigid telescopes (Karl Storz, Tuttlingen, Germany) was performed to examine passive vocal fold mobility, the site and cranio-caudal extension of the stenosis, as well as detection of any secondary LTS (laryngomalacia, tracheomalacia, extrinsic tracheal compression or presence of tracheotomy-related lesions). Microlaryngoscopy was obtained with different laryngoscopes such as the Benjamin and Benjamin-Parsons Pediatric Laryngoscope (Karl Storz, Tuttlingen, Germany) to precisely evaluate and measure the extension, consistency, and grade of stenosis.

Since 2015, as suggested by the ELS publication [17], all patients underwent asleep TNFL under general anesthesia in spontaneous respiration for systematic assessment of the upper airway and detection of narrowing related to obstructive sleep apnea (OSA) as well as adenoid, tonsillar, or base of tongue hyperplasia. Accordingly, information relative to comorbidities was also gathered and a checklist compiled. In case of treatment performed before 2015, retrospective restaging of LTS according to the new ELS classification proposal was performed, integrating the grade (I–IV according to the Myer–Cotton grading system), and number of subsites involved (“a”–“d” for 1–4 subsites among supraglottis, glottis, subglottis, and trachea), as well as information about the presence (“+” sign) of important airway (OSA-related narrowing, secondary LTS, malacia) or medical comorbidities (respiratory, cardiovascular, neurological, swallowing disorders, symptomatic gastroesophageal reflux or syndromic anomalies) from clinical charts, endoscopic pictures, and videos (Fig. 1).

Operative technique

All patients underwent segmental resection of the stenosis and subsequent airway anastomosis, which was noted according to the proposed classification [18]. Eight patients (4.2%) underwent a type A resection (i.e., TR with removal of tracheal rings only, with subsequent crico-tracheal or tracheo-tracheal anastomosis). Type B resection (i.e., CTR with removal of first tracheal rings plus anterior arch of the cricoid and subsequent thyro-crico-tracheal anastomosis) was performed in 31 (16.2%) patients. Furthermore, 113 (59.2%) patients underwent type C resection (i.e., CTR with removal of the anterior cricoid arch and posterior subglottic mucosa covering the cricoid plate with subsequent thyro-crico-tracheal anastomosis just below the crico-arytenoid joints), and 39 (20.4%) underwent type-D resection (i.e., extended CTR with removal of the anterior cricoid arch plus posterior cricoid split followed by its expansion with costal cartilage graft and subsequent thyro-crico-tracheal anastomosis).

TR or CTR was performed as single- or double-staged surgical procedures. At the end of the single-staged procedure, patients were primarily extubated and monitored in the pediatric Intensive Care Unit (ICU).

Statistical analysis

The overall ELS grade of LTS, previous treatments, grade/extension of stenosis, airway comorbidities, age, and

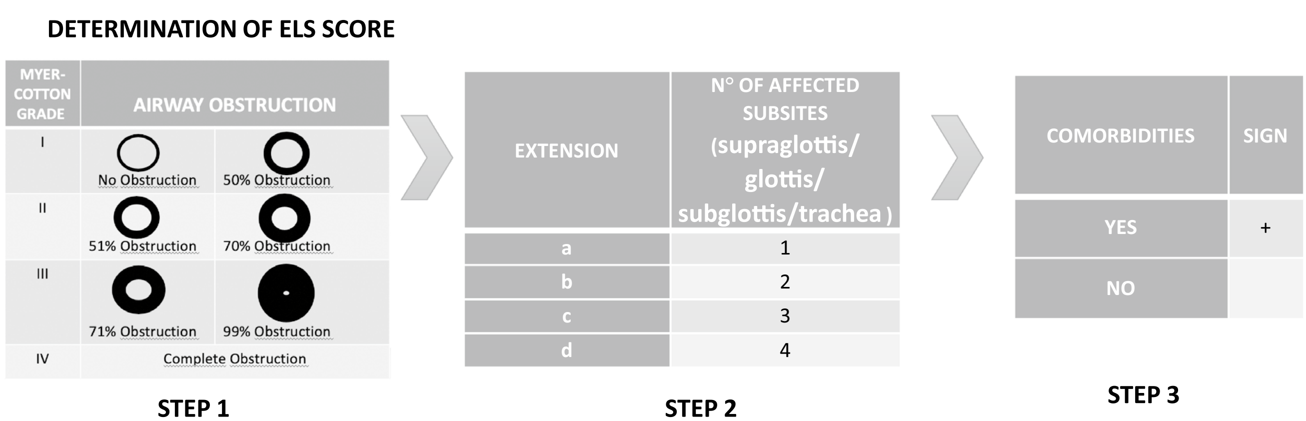


Fig. 1 Determination of ELS score. The ELS score consists of three components: 1, Myer–Cotton grade of airway obstruction, marked from I to IV; 2, number of affected airway subsites, marked from “a” to “d”, and 3, presence of comorbidities, marked with a “+”

resection length (calculated in cm) were put in relation with the mean decannulation rate, time to decannulation, onset of postoperative complications, and number of additional treatments. Differences in distribution of categorical data between groups were tested by Chi square or Fisher's exact tests as appropriate. Interactions between independent variables and a dependent one was tested with a univariate model. Post-hoc multiple comparison for observed means was carried out with Bonferroni's method. Kaplan–Meier analysis was used to determine the time to successful decannulation. The difference between groups was tested with the Log Rank test.

The SPSS statistical program (SPSS, v. 24, IBM, Armonk NY, USA) was used for analysis. A $p < 0.05$ was considered statistically significant.

Results

Decannulation rate after TR/CTR

A single-staged procedure was performed in 106 patients (55.5%), granting immediate postoperative extubation. The remaining 85 children (44.5%) underwent a double-staged procedure, requiring a tracheal cannula in the immediate postoperative period. The operation-specific decannulation rate was 76%, as 145 patients did not require any additional treatment to achieve definitive decannulation. However, the final decannulation rate was 88%, as 168 patients had no tracheal cannula at their last follow-up.

The decannulation rate was significantly higher ($p < 0.01$) among patients with AOC stenosis (96%) vs. both congenital (86%) and post-traumatic (87%) forms (Fig. 2a). Furthermore, a significantly higher decannulation rate was observed among patients without airway comorbidities (97% vs. 78%, $p < 0.001$), and stenosis limited to 1–2 subsites vs. those with 3–4 subsites involved (90% vs. 72%, $p < 0.05$), as well as in subjects with an ELS score < IIIa vs. those with a score ≥ IIIb (97% vs. 90%, $p = 0.0017$) (Fig. 2b; Table 2).

Time to decannulation

The mean interval between operation and definitive decannulation showed a significant variability (mean 143.6 ± 243.4 days; range 1–1275 days). Univariate analysis showed that a higher ELS score, onset of surgical complications, as well as congenital LTS etiology were correlated with prolonged decannulation time (Table 2). Kaplan–Meier analysis showed that patients with postoperative complications, more severe Myer–Cotton grade of stenosis, and higher number of affected subsites needed a significantly longer time to achieve decannulation (Fig. 3a–c).

Postoperative complications

The mean overall complication rate was 24.1%, as 46 patients experienced minor/major surgical or medical postoperative events. The most common surgical complication was laryngeal edema requiring treatment, which was observed among 17 (8.9%) patients, followed by anastomotic dehiscence in 12 (6.3%), restenosis in 8 (4.2%), and obstructing granulation tissue necessitating additional treatment in 7 (3.7%) patients. Unilateral vocal fold palsy and tracheal necrosis occurred in one patient (0.5%) each (Fig. 3d). Respiratory insufficiency (defined as the pathophysiologic condition in which the respiratory system fails in one or both of its gas exchange functions: oxygenation and carbon dioxide elimination) occurred as a major medical complication in eight patients (4.2%) requiring an additional ventilation support such as C-PAP or temporary intubation in the ICU. Among all children, higher rates of surgical complications were observed in stenoses extending to 3–4 subsites compared to those involving 1–2 subsites (35.3% vs. 18.4%, $p < 0.05$), as well as in patients with airway comorbidities vs. those without (26.7% vs. 14.3%, $p < 0.01$) (Fig. 2b). Similarly, a greater incidence was seen in those whose ELS score was IIIb or higher (34% vs. 20%, $p < 0.001$) (Table 2).

Additional treatment

Seventy-four (38.7%) patients required at least one additional surgical procedure (whether endoscopic or open) to maintain a patent airway (mean 2.3 ± 1.7 procedures; range 1–10). Forty-nine (66.2%) patients underwent at least one endoscopic revision (mean 2.8 ± 1.9; range 1–8). These procedures were performed as plain balloon (MaxForce esophageal balloon; Boston Scientific, Marlborough, MA, USA) dilatations ($n = 65$) or in combination with topical injection of triamcinolone acetonide ($n = 19$) or mitomycin C at 2 mg/ml for 2 min ($n = 9$). Endoscopic removal of granulation tissue was required in 30 cases. Endoscopic CO2 laser surgery with unilateral partial arytenoidectomy was performed in three patients, supraglottoplasty in 2, and unilateral posterior cordotomy in one.

The remaining 25 (33.8%) patients underwent an additional open procedure to achieve decannulation. Revision surgery in the form of CTR (type B and C resections) or extended CTR (type D resection) was performed in ten cases due to anastomotic dehiscence and in five due to restenosis. Additional surgery for restenosis in the form of LTR with anterior and posterior costal cartilage graft (ACCG + PCCG) was performed in four patients, while three underwent LTR with PCCG and two LTR with ACCG. One patient required a tracheoplasty with ACCG to achieve definitive decannulation.

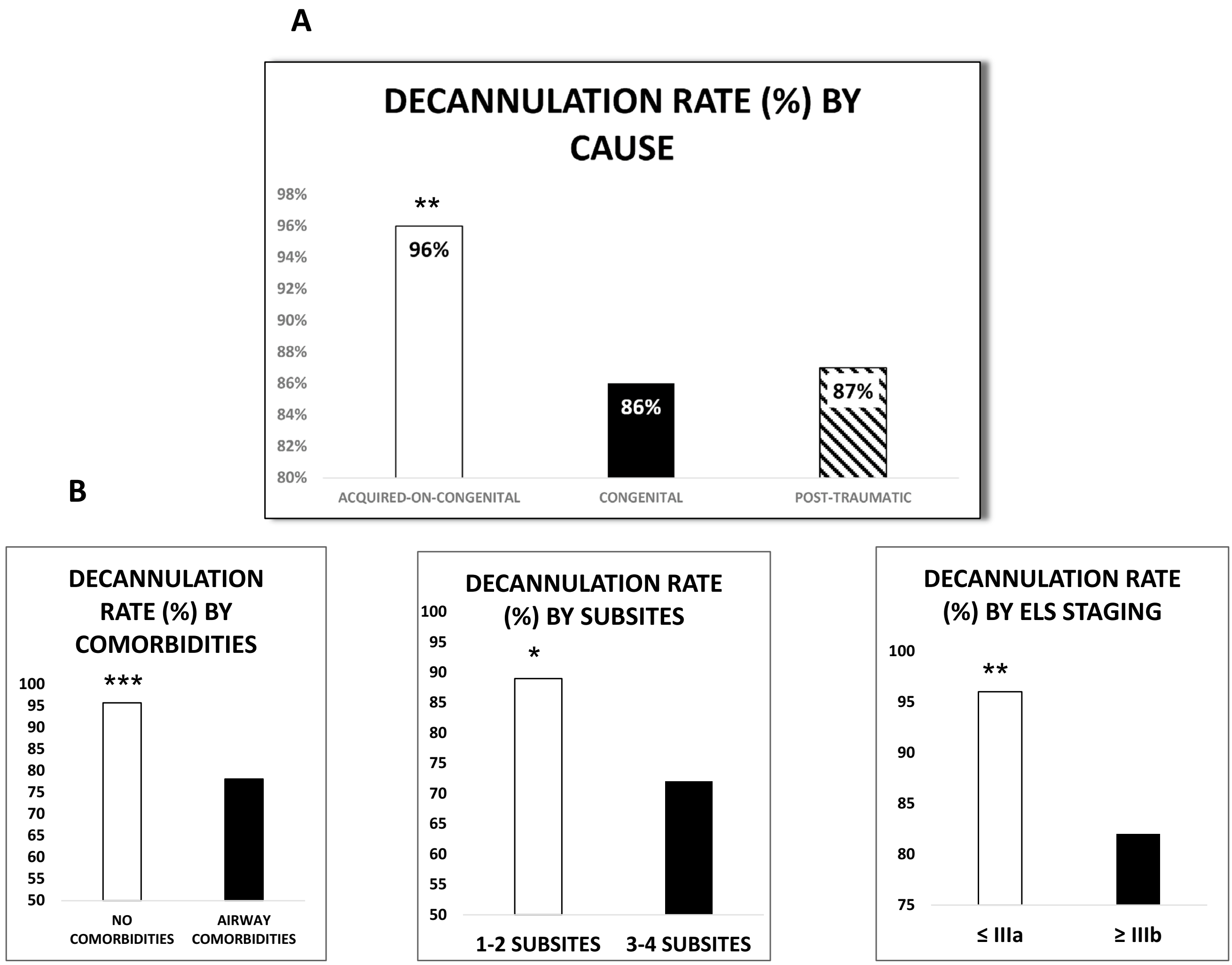


Fig. 2 Decannulation rate stratified by cause of stenosis (**a**). The highest decannulation rate was observed among acquired congenital stenosis, followed by purely iatrogenic forms. The lowest rate was observed among congenital stenosis (86%) vs. both post-traumatic (87%) and acquired-on congenital (96%). Decannulation rate stratified by comorbidities, number of subsites involved, and ELS score (**b**). The decannulation rate was higher among patients without comorbidities, with 1–2 subsites involved, and IIIa+ or lower ELS score (***$p<0.001$; **$p<0.01$; *$p<0.05$)

At univariate analysis, the number of additional operations needed after primary treatment was predicted by the ELS score, as well as by the length of resection and the onset of surgical complications ($p<0.001$) (Table 2).

Previous treatment

A total of 71 patients (37.2%) underwent previous treatment(s) before undergoing the TR/CTR procedure (mean 5.2 ± 7.7 procedures; range 1–32). Among these, 39 (54.9%) children underwent endoscopic procedures, while 32 (45.1%) had open-neck surgery. Lower decannulation rate was observed among patients who underwent any form of previous treatment vs. those who underwent CTR as primary treatment (85% vs. 93%, $p<0.01$).

Discussion

The present study represents, to the best of our knowledge, the first evaluation of the applicability, reproducibility, and prognostic impact of the ELS classification within the pediatric population affected by LTS. The reasons underlying the development of this new scoring system lay in the evolving surgical landscape of the pediatric population. Specifically, with the introduction of CTR as a routine therapeutic approach for LTS (which comprised the complete excision of stenotic segment rather than its pure expansion, as practiced by LTR), it became clear that the Myer–Cotton classification alone (based merely on the grade of stenosis) was unable to adequately predict CTR surgical outcomes in terms of decannulation rate [16, 17, 19]. In fact, this classification

Table 2 Predictive value of studied variables on end points

	< IIIb	≥ IIIb	*p*
ELS score			
Decannulation rate (%)	97%	90%	< 0.001
Mean time to decannulation (days)	52	162	< 0.001
Onset of surgical complications	34%	20%	< 0.001
Mean number of additional treatments	1.9	2.4	< 0.001

	Acute-on chronic	Congenital	Post-traumatic	*p*
Etiology				
Decannulation rate (%)	96%	86%	87%	< 0.01
Mean time to decannulation (days)	112	160	94	0.001
Onset of surgical complications	18%	26%	20%	NS
Mean number of additional treatments	2.3	2.7	2.2	NS

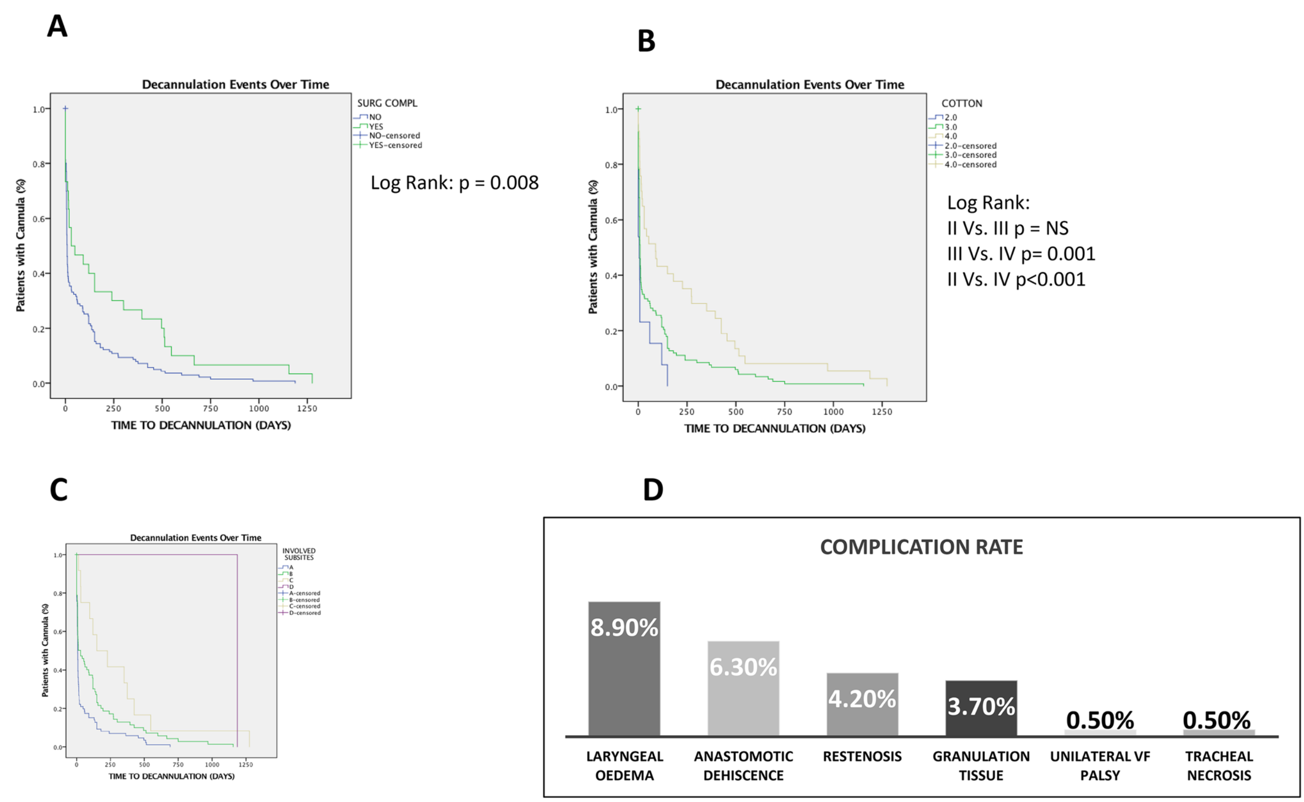
	Yes	No	*p*
Surgical complications			
Decannulation rate (%)	87%	91%	NS
Mean time to decannulation (days)	194	92	< 0.01
Mean number of additional treatments	2.3	1.6	< 0.001

NS non significant

does not take into account cranio-caudal extension of the stenotic process and the presence of comorbidities, which are independent predictors of decannulation. Accordingly, patients with cranial extension of stenosis from the subglottis to the glottis, as well as those with severe comorbidities, showed lower decannulation rates and longer times to achieve it compared to those with the same Myer–Cotton grade but without glottic extension or comorbidities [16]. Moreover, glottic extension implicated the need to perform extended CTR (type D resection) and, consequently, prolonged stenting, which in turn caused delayed decannulation [16, 19, 20]. It is of note that the glottic extension scenario does not preclude good functional outcomes and a high decannulation rate. However, it obliges the surgeon to properly counsel parents in terms of prognosis and the possibility of a prolonged postoperative course after CTR [16]. Moreover, in the last 2 decades, different authors have underlined that other factors than Myer–Cotton grade of stenosis can impact decannulation [4–6, 13, 14, 16, 19, 20].

This motivated airway surgeons to create a new classification system for benign LTS. Such a staging system should be easily applied in everyday practice and be reliable in predicting surgical outcomes [17]. Our results show that the proposed ELS score was able to predict the decannulation rate, which is the most relevant success outcome of reconstructive airway surgery for LTS (Table 2). Specifically, an ELS score IIIb or higher (indicating relevant stenosis associated with long cranio-caudal extension) was associated with lower decannulation rates. Furthermore, individual parts of the new scoring system, such as the presence of airway comorbidities and stenosis extension to 3–4 subsites, was related to significantly lower decannulation rate as well (Table 2). However, a combination of different components in the ELS score can simplify the classification process. These findings can enable the surgeon to foretell the possibility of decannulation in each patient, based on preoperative evaluation and patient characteristics.

This information, therefore, might affect the choice of treatment in each individual case as well as the decision of whether tracheotomy should be closed primarily or staged. Thus, it represents an important tool in preoperative parent counseling. Moreover, our results indicate that parents of previously treated patients should be advised of the lower probability of successful decannulation. Actually, the lower decannulation rate following salvage CTR compared to the primary procedure was observed in our patient population and concurs with literature data [5, 21, 22]. This observation could be explained by the fact that repeated treatments induce an exuberant wound healing process requiring further revision surgeries, which could eventually lead to a higher risk of restenosis [22]. Furthermore, a longer time in achieving decannulation is to be expected in children with a higher ELS score, as already reported in the literature [16, 19, 20]. On the other hand, the authors concur in observing that patient age per se did not affect the success of this type of surgery [21]. There was no statistically significant difference in decannulation rate, onset of complications, or number of additional treatments between children younger vs. older than 1 year.



Pairwise Comparisons

	MOD GRAD	A Chi-Square	A Sig.	B Chi-Square	B Sig.	C Chi-Square	C Sig.	D Chi-Square	D Sig.
Log Rank (Mantel-Cox)	A			7.774	.005	11.904	.001	4.954	.026
	B	7.774	.005			4.224	.040	4.682	.030
	C	11.904	.001	4.224	.040			.865	.352
	D	4.954	.026	4.682	.030	.865	.352		

Fig. 3 Decannulation events over time according to complications (**a**): patients with surgical complications presented longer time to decannulation. Log rank: $p = 0.008$. Decannulation events over time according to Myer–Cotton grade (**b**): patients with Grade II Myer–Cotton stenosis had significantly shorter time to decannulation vs. Grade IV patients (log rank: II vs. IV $p < 0.001$) as well as patients with Grade III stenosis vs. patients with Grade IV stenosis (log rank: III vs. IV $p = 0.001$). Decannulation events over time according to number of subsites involved (**c**): Extension of stenosis played an important role in determining time to decannulation. Complication rate (**d**): the most common surgical complication was laryngeal edema, followed by anastomotic dehiscence, restenosis, and granulation tissue formation, unilateral vocal fold palsy, and tracheal necrosis

The occurrence of complications was relatively common in the study population. Our analysis shows that the extent of stenosis and the presence of comorbidity are associated with the onset of such events (Table 2). In particular, longer stenosis (involving 3–4 subsites) indicates greater airway resections, which could ultimately lead to a higher occurrence of anastomotic dehiscence and complications [18]. Furthermore, if a child suffers from important respiratory or cardiovascular impairments, more surgical complications are to be expected. In our series, 40% of patients required multiple additional treatments before achieving patent airway without tracheotomy. This trend matches that described in the literature, especially regarding patients with associated comorbidities or glottic involvement [16, 21, 23]. Therefore, it is of great importance to explain this point thoroughly to parents, informing them that one in three children is likely to be retreated. Moreover, two-thirds of these patients are likely to have a minor complication, which is amenable of endoscopic correction, while one-third could experience a severe complication such as anastomotic dehiscence or restenosis, and therefore require an open revision. Herein, the ELS score had strong predictive value in helping to identify patients who will need more additional corrections before reaching a satisfactorily patent airway (Table 2). The same detected risk factors that led to the higher onset of complications, in fact, could lead to the need for a greater number of re-treatments.

Even though the immediate impression could be that of a complex scoring system, our experience showed that all the information needed is normally already contained in clinical charts and surgical reports, as is relevant for the planning and execution of the procedure. It was thus easy to insert these data and to accordingly classify each patient retrospectively. Nonetheless, this study is affected by some limitations. It is a retrospective study, analyzing patients treated at different institutions by different surgeons, through an extensive time span. When considering the low incidence of this pathologic condition, it follows that the learning curve of each surgeon is definitely long. For this reason, only a limited number of operators performed these procedures. These considerations call for a homogeneous classification system, so as to attain a uniform surgical approach, better communication and information sharing among different centers, and more proficient and standardized training of fellow colleagues. The precise definition of diagnostic criteria and constant communication among local investigators ensured that patients managed in these centers were analyzed as homogeneously as possible.

Future larger prospective studies are, however, needed to confirm the applicability of such a proposed scoring system by independent authors.

Conclusion

The recently proposed ELS score system for benign LTS integrates all relevant patient and stenosis-related characteristics, and is easily usable in everyday practice. It helps the airway surgeon in identifying patients who are suitable for a certain type of procedure and in recognizing those at a higher risk for complications, re-treatments, or prolonged time before definitive decannulation. With the ability to predict these crucial surgical outcomes, the present ELS scoring system represents a valuable tool in parental counseling and therapeutic planning.

Compliance with ethical standards

Conflict of interest All authors declare that they have no conflict of interest.

Ethical standards All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Human/animal rights statement This article does not contain any studies with animals performed by any of the authors.

Informed consent Informed consent was obtained from all individual participants included in the study.

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