SLIDE LARYNGOTRACHEOPLASTY: A NEW SOLUTION FOR CONGENITAL SUBGLOTTIC STENOSIS IN NEONATES AND INFANTS

PhD Thesis

Eszter Erdélyi M.D.

Department of Oto-Rhino-Laryngology, Head and Neck Surgery

University of Szeged

University of Szeged,

Clinical Medical Sciences Doctoral School

PhD Program:

Clinical and Experimental Research for Reconstructive and Organ-sparing Surgery

Program director: Prof. Lajos Kemény M.D.

Supervisor: Ádám Bach M.D.

Szeged

2022

PUBLICATIONS RELATED TO THE PhD THESIS

I. Rovó L, **Erdélyi E**, Tóbiás Z, Gál P, Sztanó B, Sandu K, Bach Á.
Slide laryngotracheoplasty for congenital subglottic stenosis in newborns and infants
Laryngoscope 2020;130:199-205.
Impact factor: 2,650

II. Bach Á, **Erdélyi E,** Sztanó B, Tóbiás Z, Rovó L.
Innovative surgical solutions for laryngeal stenoses in newborns and infants
[Laryngealis szűkületek innovatív sebészi megoldásai újszülött- és csecsemőkorban]
Orvosi Hetilap 2021;162:2100-2106.
Impact factor: 0,540

III. **Erdélyi E**, Bach Á, Sztanó B, Gál P, Sandu K, Bereczki C, Rovó L.
Long-term Results of Slide Laryngotracheoplasty for Congenital Subglottic Stenosis in Newborns and Infants
International Journal of Pediatric Otorhinolaryngology 2022 May; 158:111192 DOI: 10.1016/j.ijporl.2022.111192
Impact factor: 1,675

CITABLE ABSTRACTS

I. **Erdélyi E**, Bach Á, Tóbiás Z, Szegesdi I, Gál P, Sztanó B, Rovó L. **Slide laryngotracheoplasty for subglottic stenosis in neonates and infants** Congress of the European Society of Pediatric Otorhinolaryngology (ESPO), June 2-5, 2018, Stockholm, Sweden

II. Bach Á, **Erdélyi E**, Tóbiás Z, Szegesdi I, Gál P, Sztanó B.
Slide laryngotracheoplasty in neonates and infants for subglottic stenosis
4th Congress of European ORL- HNS, October 7-11, 2017, Barcelona, Spain

PUBLICATIONS NOT DIRECTLY RELATED TO THE PhD THESIS

I. Madani S, Bach Á, Matievics V, **Erdélyi E**, Sztanó B, Szegesdi I, Castellanos PF, Rovó L.

A new solution for neonatal bilateral vocal cord paralysis: Endoscopic arytenoid abduction lateropexy

Laryngoscope 2017;127:1608-1614.

Impact factor: 2,442

II. Sztanó B, Bach Á, Matievics V, **Erdélyi E**, Rovó L.

Endoscopic arytenoid abduction lateropexy for the treatment of bilateral vocal chord paralysis in early childhood: Case report

American Journal of Otolaryngology and Head and Neck Surgery 2018;1(3), article 1011.

III. Sztanó B, Bach Á, Matievics V, **Erdélyi E**, Szegesdi I, Rovó L.

Endoscopic arytenoid abduction lateropexy for the treatment of neonatal bilateral vocal cord paralysis - Long-term results

International Journal of Pediatric Otorhinolaryngology 2019;119:147-150.

Impact factor: 1,290

CONTENT

ABBREVIATIONS

BiPAP – biphasic positive airway pressure

Ch – Charier

CO_2 – carbon-dioxide

CPAP – continuous positive airway pressure

CSGS – congenital subglottic stenosis

CT – computer tomography

CTR – cricotracheal resection

ECTR – extended cricotracheal resection

ETT – endotracheal tube

HNR – harmonics to noise ratio

LTR – laryngotracheoplasty

MRI – magnetic resonance imaging

NG tube – nasogastric tube

PVN – per vias naturales

PCTR – partial cricotracheal resection

PICU – perinatal intensive care unit

QOL – quality of life

RDS - respiratory distress syndrome

RSV – respiratory syncytial virus

SC – sectio caesarea

SGS – subglottic stenosis

SLTP – slide laryngotracheoplasty

TIVA – total intravenous anesthesia

UDP – ultra dream pulse

1. INTRODUCTION

The adequate treatment of subglottic stenosis (SGS) is a major challenge of laryngology even nowadays. After laryngomalacia and vocal fold palsy, SGS is the third most common congenital laryngeal malformation. However, it is the laryngeal anomaly most commonly necessitating tracheostomy in newborns and infants [1,2]. Low-grade congenital subglottic stenosis (CSGS) may improve with growth, but in severe cases, no spontaneous airway improvement is expected over time; thus, selecting the optimal surgical treatment strategy is paramount [3].

The ultimate goal of management is to ensure an adequate airway with the preservation of voicing and swallowing. Despite the well-defined endpoints (e.g., avoidance of a tracheotomy, definitive decannulation, socially acceptable voice quality, safe swallowing without aspiration), the surgical treatment of CSGS remains heterogeneous and depends almost as much on the experience of the airway team as on the child's condition [4]. Definitive airway surgery is usually preceded by tracheostomy despite its many well-known physical and psychosocial adverse effects [5-8].

In consideration of the special anatomy and vulnerable tissues of the pediatric airway, the potentially hidden comorbidities, and the associated congenital malformations, choosing the appropriate surgical method is crucial at this early age [9-12]. In cases of high-grade SGS, endoscopic procedures are not suggested [13-15]. Laryngotracheal reconstruction (LTR), cricotracheal resection (CTR), and extended cricotracheal resection (ECTR) have been used for decades, and they have proven to be favorable solutions with good long-term results. These complex open neck surgeries require major tissue resection, stent implantation, or airway expansion by rib cartilage grafts [12,16]. To reduce the possible complications and maintain airway stability extended resection of the cartilaginous framework should be limited to allow

tension-free anastomosis, and if graft implantation is necessary, well vascularized, easily available, regional tissue is desirable. Moreover, a definitive (one-step) surgical solution is preferable to perform as soon as possible in carefully selected patients.

Over the past three decades, the Department of Oto-Rhino-Laryngology, Head and Neck Surgery, University of Szeged has become a regional center for patients with upper airway stenosis. The intensive and exceptional relationship with the partner institutions made it possible to routinely apply and improve the well-known airway reconstruction techniques not only in adults but in pediatric patients too. The culmination of this cooperation is obviously the introduction of different innovative surgical techniques in the most fragile subpopulations of newborns and infants.

I have been working in the Department of Oto-Rhino-Laryngology, Head and Neck Surgery, University of Szeged since 2015. In the first year of my residency, I joined the upper airway stenosis workgroup under the supervision of Professor Dr. László Rovó. Over the past 6 years, I took part in several operations of different types of airway stenoses in adult and pediatric patients too. I have also presented our related results at several Hungarian and international conferences as well. During my daily work, one of my main tasks is the evaluation of the preoperative and postoperative status of children with upper airway stenosis. I am also responsible for liaising with the parents and our colleagues in the Pediatric department. Soon, I would like to continue my career as a pediatric otolaryngologist.

2. CONGENITAL SUBGLOTTIC STENOSIS

2.1. Pathogenesis

In a full-term neonate, SGS is defined as a lumen ≤4 mm in diameter at the level of the cricoid, whereas it is defined as a lumen <3 mm in diameter in preterm infants [12]. Congenital SGS is attributed to the incomplete recanalization of the laryngeal lumen during the 10^{th} week of gestation [17]. It is frequently associated with other congenital head and neck lesions and syndromes (e.g., CHARGE syndrome, Down syndrome, 22q11 deletion) [18]. Failures at different stages of recanalization of the epithelial lamina lead to various degrees of SGS [19]. This entity is closely related to laryngeal webs and atresia, which also result from a laryngeal recanalization failure [20]. This accounts for the frequent cartilaginous subglottic component seen in extensive glottic webs and the complete obliteration of the atretic larynx [21]. The most frequent forms of cartilaginous CSGS are composed of a thick anterior lamina and a generalized thickening of the cricoid ring or an elliptical cricoid [2]. CSGS amounts to 5% of all subglottic stenoses, but its true incidence is difficult to assess as many cases are aggravated by an emergency endotracheal intubation leading to the so-called acquired on congenital or mixed SGS [9-12].

2.2. Symptoms

Airflow resistance is inversely proportional to the radius to the fourth power (r^4) [22]. Accordingly, a 50% reduction of the radius causes a 16-fold increase in airflow resistance. This significantly increased resistance imposes a huge physical burden on newborns and infants equally. A cartilaginous CSGS causes biphasic stridor with a more prominent inspiratory phase. Depending on the degree of the CSGS, typical signs of respiratory distress, such as severe obstructive dyspnea with suprasternal or chest retractions appear right after delivery.

Nevertheless, even a 50% - 70% luminal diameter restriction can often remain asymptomatic for weeks. However, infants with this airway status may experience recurrent episodes of croup with a barking cough, primarily when the infection is associated with mucosal edema [23].

2.3. Diagnostics and classification

The prenatal diagnosis of subglottic stenosis is not routinely possible. The perinatal events largely determine the diagnostic and subsequent therapeutic interventions. Indisputably, endoscopic assessment is the golden standard for the evaluation of the compromised airway in infants and children [Figure 1].

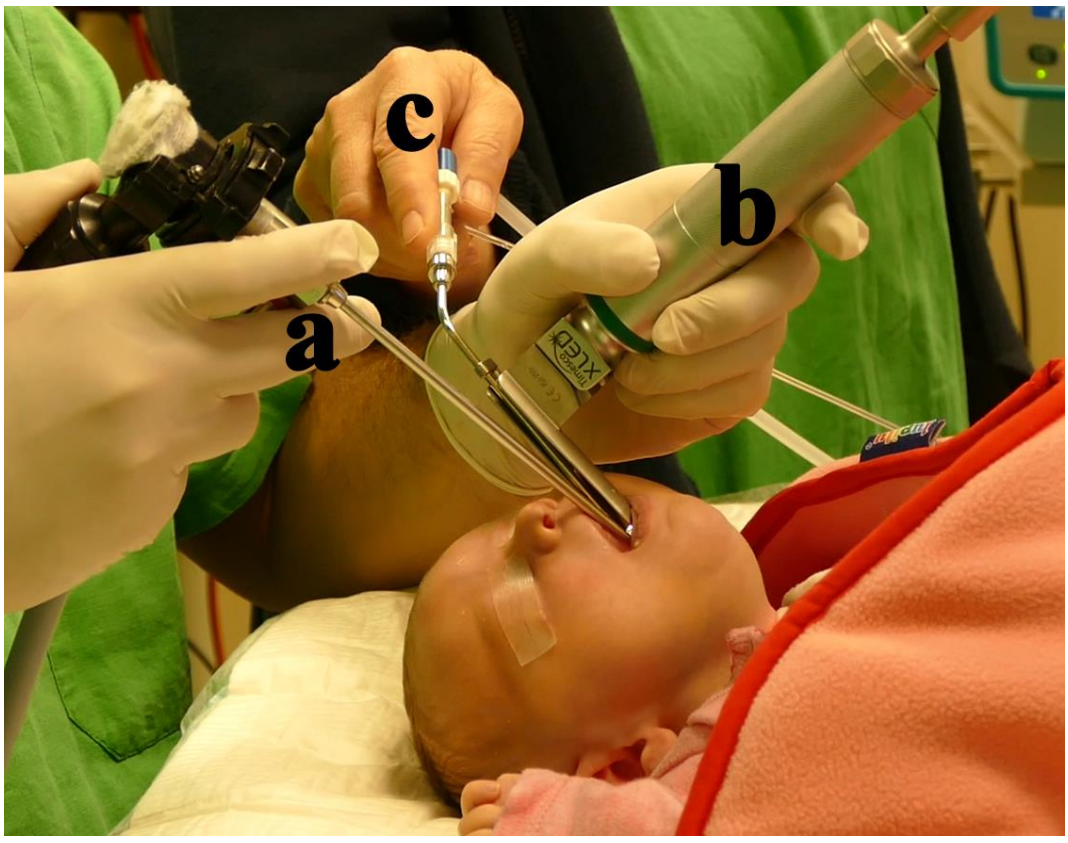

Figure 1: Direct laryngo-tracheoscopy - under general anesthesia via total intravenous anesthesia and supraglottic jet ventilation - of a 3-month-old child due to mild dyspnea and inspiratory stridor. a) 30° rigid endoscope; b) Miller laryngoscope; c) jet pipe

During laryngo-tracheoscopy, the exact location, craniocaudal extension, grade, and histological nature (cartilaginous/soft tissue) of a SGS can be appropriately assessed. However, the endoscopic appearance of SGS may be disproportionate concerning the clinical presentation as infants are remarkably tolerant to airway compromise [5]. During the endoscopic examination, other congenital disorders of the laryngotracheal airway can be explored as well. Imaging procedures are not required in terms of CSGS but might be useful in cases of suspected hidden comorbidities and associated congenital malformations. In this age group, magnetic resonance imaging (MRI) is preferred.

Since the symptoms and the applicable treatment modalities are mostly determined by the severity of stenosis, precise evaluation of the airway obstruction is crucial. The Cotton-Myer classification is a simple, effective, and internationally applied grading system. Originally it was used to predict lumen surface reduction in case of endotracheal tube application, it was then extended to describe both pediatric and adult subglottic and/or tracheal stenosis.

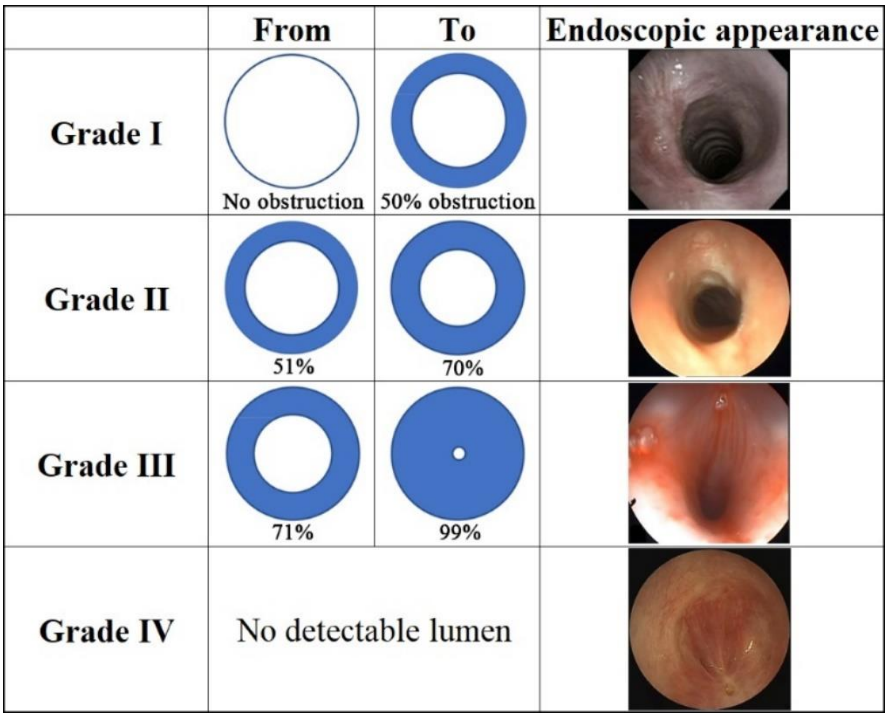

Figure 2: Cotton-Myer classification of subglottic stenosis

The classification consists of IV grades: Grade I: 0 to 50% decrease in lumen surface; Grade II: 51 to 70% decrease; Grade III: 71 to 99% decrease, and Grade IV–no evidence of detectable lumen [24], [Figure 2]. In order to describe the complex anatomy of this particular area, laryngotracheal stenosis classifications have proliferated in the literature in the last few decades. McCaffrey et al. and Lano et al. proposed classifications to describe the extension of the stenosis among the most commonly involved subsites: glottis, subglottis and trachea [25,26]. Meanwhile, the classic Cotton-Myer grading system was also extended, incorporating three additional parameters: comorbidities, glottic involvement and the association of both [27].

2.4. Treatment

2.4.1. Perinatal care

Depending on the degree of the CSGS, signs of respiratory distress at delivery (such as severe dyspnea with suprasternal or chest retractions) warrant prompt and proper airway management to ensure adequate ventilation and oxygenation of the newborn. Routine oropharyngeal/nasopharyngeal suctioning at birth has been the standard of care for newborns for decades. In the mildest cases, an oxygen hood might be a sufficient solution. In severe cases nasal high flow or nasal continuous positive airway pressure (CPAP) or biphasic positive airway pressure (BiPAP) is necessary. When non-invasive positive pressure ventilation fails, urgent endotracheal intubation or tracheotomy cannot be avoided.

2.4.2. Definitive treatment of congenital subglottic stenosis

Generally, children with low-grade (grade I or grade II) CSGS do not require surgical intervention. As affected children grow, spontaneous airway improvement can typically be

expected [28]. Even mild grade III stenosis can often remain asymptomatic for weeks. However, infants with this airway status may experience recurrent airway symptoms, primarily when the infection is associated with mucosal edema [23]. Physiological development and growth are significantly impaired in cases of high-grade CSGS; thus, a 'watch and wait' policy is not endorsed. When definitive extubation repeatedly fails, airway widening surgical intervention is necessary. For this purpose, tracheostomy is the most commonly performed primary intervention. As a life-saving procedure, the merits of tracheostomy are indisputable, however, it can negatively affect voicing, swallowing, and the quality of life (QOL) [29]. Bacterial colonization and dynamic collapse of the trachea are also real threats [30-32]. A definitive surgical solution that can be performed in one step could prevent many consequential problems such as accidental decannulation or airway obstruction, chronic airway infection, poor voice quality, negative effects on speech development, tracheomalacia, need for multiple procedures, and high medical and nursing costs.

The number of surgical options has significantly increased over the past 50 years. Still, the proper management of CSGS remains both challenging and complex [33]. Endoscopic, minimally invasive solutions (e.g., balloon dilatation, excision or incision of scar tissue with cold steel or CO_2 laser, intralesional or topical adjuvant therapy such as mitomycin-C application or steroid injection) are optimal for patients with isolated low-grade (grade I–II) SGS without a history of previous treatment failure [34]. However, these techniques can also increase the incidence of urgent/unplanned airway interventions and potentially delay a definitive solution [35,36]. Moreover, the chance of success with endoscopic, minimally invasive techniques decreases with the worsening of the initial grade of subglottic stenosis [13-15].

Patients with grade III–IV SGS or multilevel airway stenosis mostly require open surgical procedures [13]. Although these interventions carry a higher risk of morbidity, this is

balanced by the fact that these treatments provide a definitive solution. In addition, we cannot ignore the fact that the first operation provides the best chance for a successful airway intervention [37]. Therefore, the avoidance of multiple, inappropriately selected, potentially futile (endoscopic) airway surgeries is essential. Laryngotracheal reconstruction (LTR) and cricotracheal resection (CTR) are the prevailing procedures globally [38-42]. In pediatric patients, CTR provides an overall decannulation rate exceeding 80% with a highly variable reoperation rate of 4%–41% (depending on the patient's comorbidities and grade of stenosis). Meanwhile, 22%–45% of patients require reoperation after LTR [10,16,40,42-47]. Significant manipulation of the laryngotracheal framework is inevitable using these techniques. However, by reducing the amount of resected tissue and minimizing potential graft problems, the success rate could be theoretically increased.

3. AIMS OF THE THESIS

1. Introducing a novel single-stage procedure without tracheostomy and stenting for high-grade congenital subglottic stenosis in neonates and infants to provide an adequate airway with the preservation of voicing and swallowing.
2. Evaluation of the functional results of the surgery by objective and subjective tests.
3. Evaluation of the physical development of the patients.
4. Assessing long-term reliability of the procedure performed in this age of rapid development of the laryngeal structures.

4. MATERIALS AND METHODS

4.1. Patients

Between January 2012 and May 2018 slide laryngotracheoplasty was performed in seven consecutive patients on the 10th, 14th, 68th, 105th, 92nd, 130th, and 120th days of life, respectively. Two patients (patients #1 and #2) had been admitted to the perinatal intensive care unit (PICU) immediately after birth, and two patients (patients #2 and #5) had been intubated due to severe stridor and inspiratory dyspnea. One infant (patient #3) required temporarily continuous positive airway pressure (CPAP) ventilation. One infant (patient #7) required tracheostomy and gastrotube in the fifth week after birth. In three infants (patients #3, #4, and #5) the severe inspiratory stridor occurred after an upper airway infection. Discontinuance of oxygen support (patients #3 and #4) and definitive extubation (patient #5) was not possible in these cases. Endoscopic evaluation of the airway and reconstructive airway surgery was performed on the fifth day of intubation in patient #5 and 3 days after the onset of the dyspnea in patients #3 and #4. Pregnancy-related information and patient data are reported in Table 1. Preoperative high resolution computed tomography (CT) or magnetic resonance imaging (MRI) scan was performed in all cases. The preoperative investigations included dynamic and rigid airway endoscopy along with neonatological and neurological evaluations. Concentric cartilaginous, Cotton-Myer grade III CSGS was detected with a direct endoscopic examination in six of seven patients (patients #2–7), and patient #1 had Cotton-Myer grade II SGS. In one case (patient #4), CSGS was associated with Cohen grade III laryngeal web [48].

Patient/sex	Birth	Gestational age [week]	Apgar score	Weight at birth	Oxygen supplementation before surgery	Grade of stenosis	Feeding before surgery	Other symptoms	Surgery [day of life]
1/m	PVN	37	8,9,9	1980g	oxygen hood	grade II	per os	-	10th
2/m	SC	40	7,9,9	3490g	intubation	grade III	NG tube	-	14th
3/f	PVN	38	8,10,10	3450g	high flow oxygen	grade III	per os	RSV infection, tachycardia	68th
4/f	SC	37	7,8,9	2370g	oxygen hood	grade III	per os	Tetralogy of Fallot, laryngeal web, DiGeorge syndrome	105th
5/f	PVN	36	_	3300g	intubation	grade III	NG tube	RSV infection, laryngitis subglottica	92nd
6/m	SC	34	7,8,8	1850g	oxygen hood	grade III	per os	laryngomalacia, RDS	130th
7/f	PVN	40	2,6,8	2390g	tracheostomy	grade III	gastro tube	DiGeorge syndrome	120th

Table 1: Pregnancy information and patient data.

f = female; m = male; PVN: per vias naturales; SC: sectio caesarea; NG tube: nasogastric feeding tube; RDS: respiratory distress syndrome; RSV: respiratory syncytial virus

4.2. Surgical technique

4.2.1. Endoscopic evaluation

The procedure began with the direct endoscopic examination of the upper and lower airways to assess the craniocaudal extension of the stenosis and determine its (cartilaginous/soft tissue) nature. Laryngo-tracheoscopy with a rigid 0° and 30° endoscope was performed under general anesthesia via total intravenous anesthesia (TIVA) and supraglottic jet ventilation with readiness for possible intubation. Jet ventilation was performed by using the Moonsune III device (Acutronic Medical Systems, Hirzel, Switzerland) with the following parameters: inhalation time = 20%, frequency = 130/min, pause pressure = 10 cmH_2O, and peak inspiratory pressure (PIP) 12 cmH_2O. The patient with tracheostomy was induced through the cannula.

4.2.2. Resection of the stenotic airway

After the diagnostic laryngomicroscopy orotracheal intubation was performed. The surgery began with a horizontal collar incision made at the level of the cricoid cartilage. The strap muscles were divided and the laryngotracheal complex was bluntly explored from the upper edge of the thyroid cartilage to the superior mediastinum, protecting the recurrent laryngeal nerves and the great vessels [Figure 3].

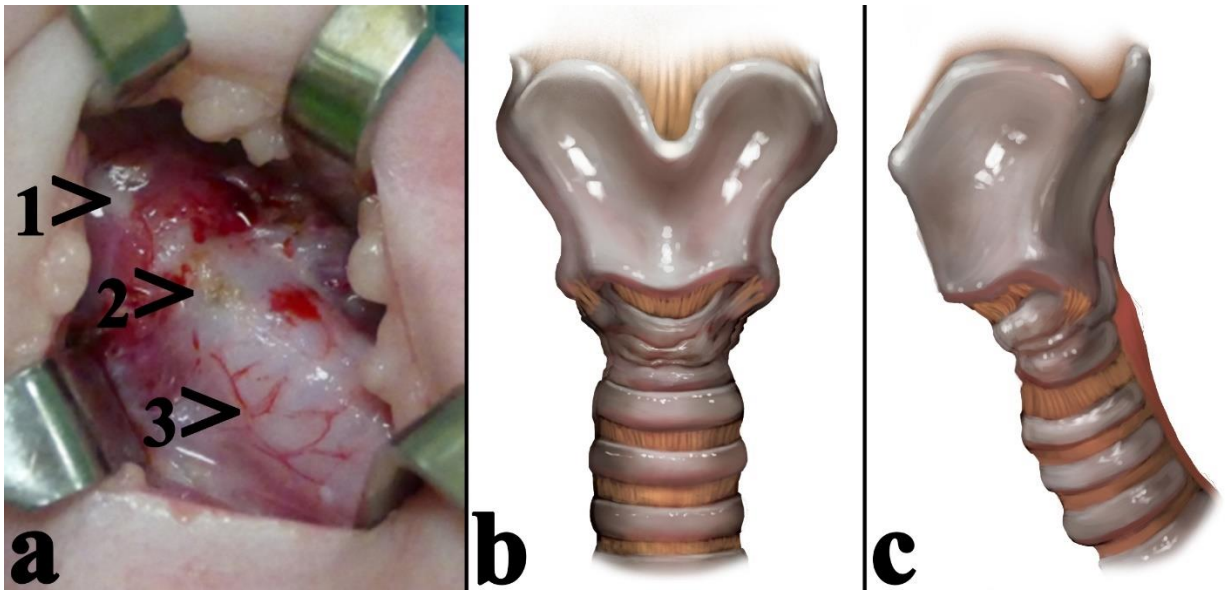

Figure 3: The explored laryngotracheal complex with a hypoplastic cricoid cartilage.

a) Intraoperative picture; 1: thyroid cartilage; 2: cricoid cartilage; 3: tracheal cartilages

b,c) schematic drawings; anterior and lateral view

The thyroid isthmus was divided in the midline and the cricothyroid muscles were left untouched. The cricotracheal ligament was dissected and a partial midline anterior laryngofissure was performed dividing the cricoid and the thyroid cartilage until the level of the anterior commissure. The cricotracheal junction was dissected circumferentially and divided carefully avoiding injury to the esophagus [Figure 4 a]. Cross-field ventilation was carried out using a second set of sterile anesthesia tubings. After visualization of the laryngeal lumen a posterior cricoid incision was performed with preservation of the posterior perichondrium, the posterior cricoarytenoid, and pharyngeal constrictor muscles integrity.

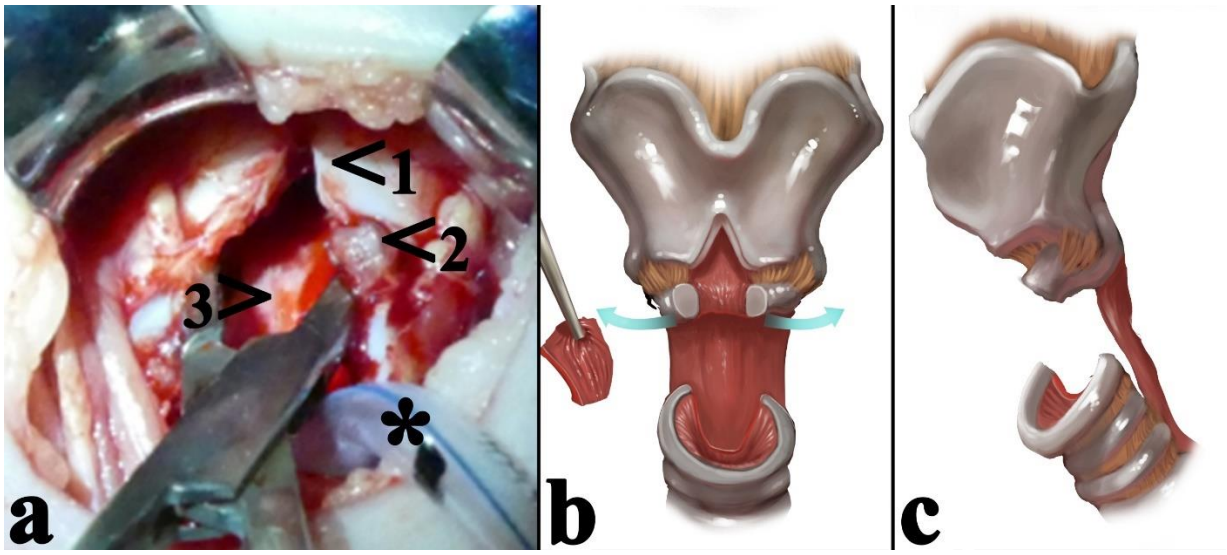

Figure 4: Partial anterior laryngofissure.

a) Intraoperative picture; 1: incised thyroid cartilage; 2: cricoid cartilage; 3: posterior wall of the laryngotracheal complex; *endotracheal tube

b,c) schematic drawings, anterior and lateral view

The distal trachea was mobilized until the anterior cartilage rings could be easily pulled up to the level of the anterior commissure. The membranous part was resected till the level of the second tracheal cartilage [Figure 4 b,c; 5].

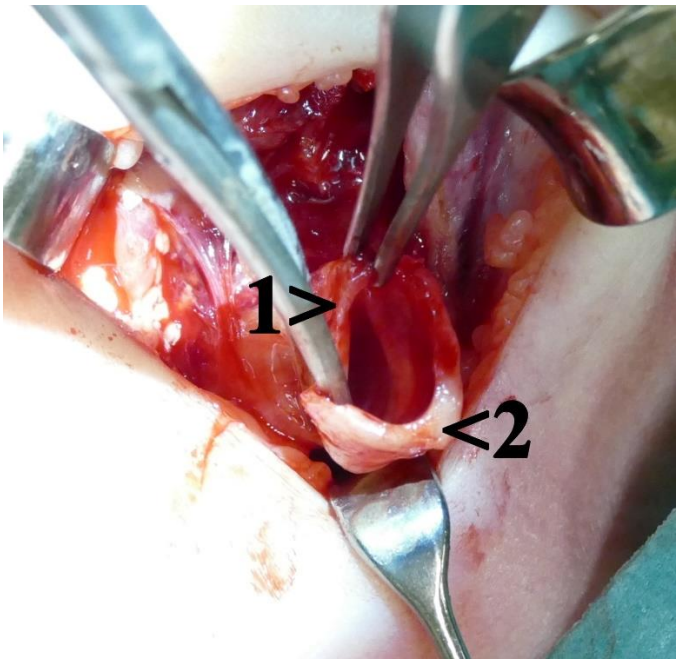

Figure 5: Mobilized trachea.

1: resection of the membranous part; 2: anterior arch of the tracheal ring

4.2.3. Surgical variation in case of laryngeal web

The membranous component of an associated grade III laryngeal web was divided in the midline using Ultra Dream Pulse (UDP) CO_2 laser (DS-40U, Daeshin Enterprise, Seoul, Korea) with 20 ms repeat time, 90 μs pulse duration, 315 W peak power, 0,16 mm spot diameter. The cartilaginous subglottic stenosis was untouched by the laser.

4.2.4. Laryngotracheal anastomosis

An anastomosis was created between the trachea, the anterior cricoid and the midline incised thyroid cartilage using 2.0 or 3.0 PDS suture. The anastomosis started in the posterior midline. Two double-armed continuous locked sutures were placed clockwise and counter-clockwise [Figure 6,7].

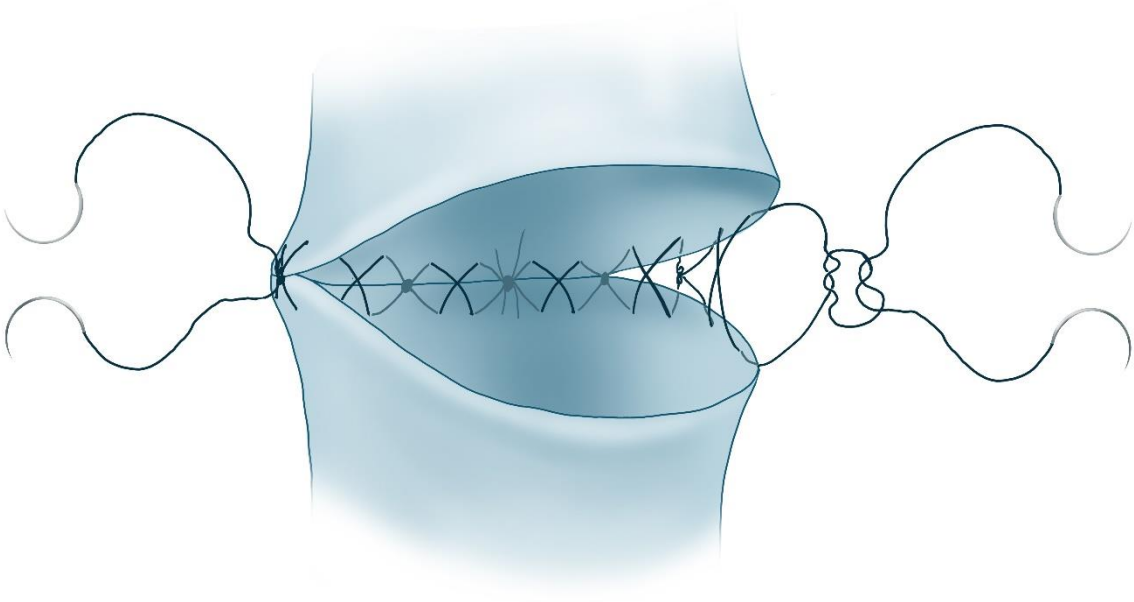

Figure 6: Schematic picture of the anastomosis.

Two double-armed continuous locked sutures are running clockwise and counterclockwise.

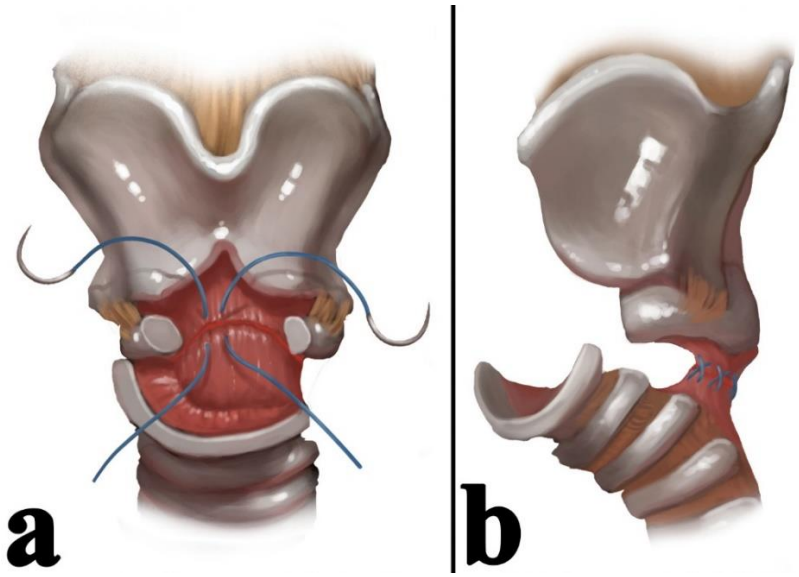

Figure 7: An anastomosis between the trachea, cricoid and thyroid cartilage is created. First, the posterior wall is reconstructed. Schematic drawings, anterior (a) and lateral (b) view

After the posterior wall was reconstructed the previously passed ETT was descended into the trachea, then the sutures of the lateral and the anterior wall were completed [Figure 8].

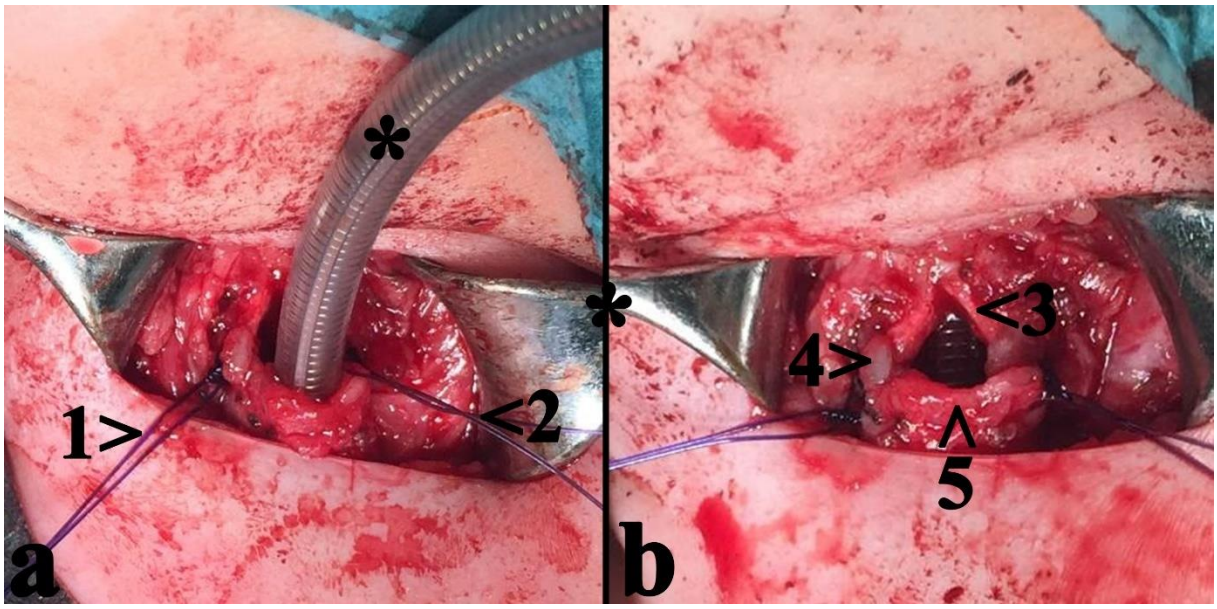

Figure 8: After the reconstruction of the posterior wall, the ETT passed transorally into the trachea to provide access to the anterior wall of the anastomosis. a) ETT in the tracheostomy b) the repositioned ETT does not cover the surgical field; counter-clockwise (1) and clockwise (2) running threads; 3: thyroid cartilage; 4: cricoid cartilage; 5: 1st tracheal cartilage; *: endotracheal tube

Finally, the threads arriving from the opposite directions were knotted in the anterior midline—with the knots lying outside the airway. Thus, a continuous suture ring was created [Figure 9]. The pre-laryngeal muscles and the skin were sutured in two layers, and a 10 Ch drain (Redax - Redon) was inserted for 2 to 3 days.

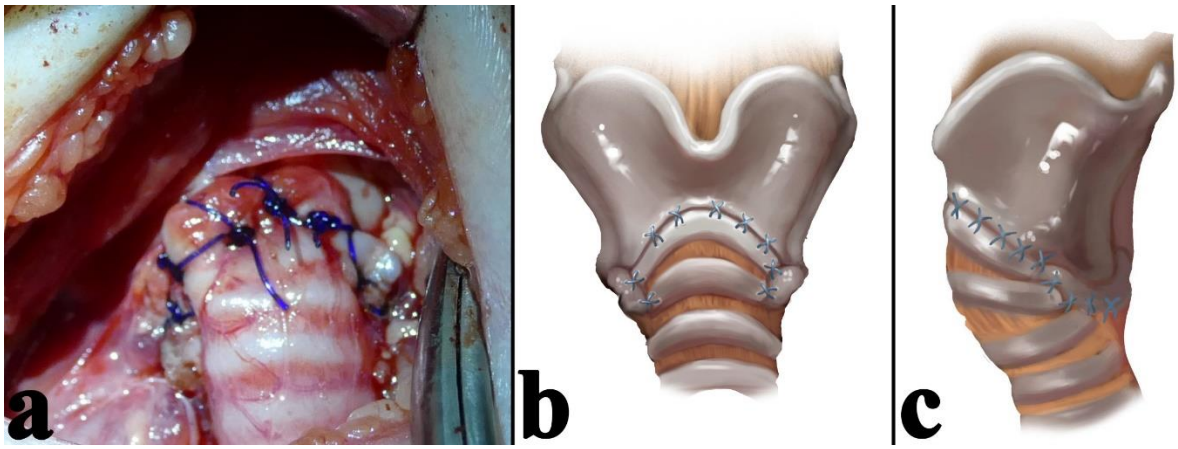

Figure 9: Intraroperative picture and schematic drawing of the reconstructed anterior (a,b) and lateral wall (c) of the anastomosis.

4.2.5. Postoperative care

The patient was transferred to a pediatric/neonatal intensive care unit. Parenteral antibiotic (amoxicillin-clavulanic acid 25 mg/5 mg/kg for 12 hours or depending on the bacteriologic aspirate) was administered for 7 days. On the day of extubation (3rd through 10th day), the patient was given a steroid (methylprednisolone, 4 mg/kg) bolus. Nasogastric tube feeding was continued for a few days after extubation and oral feeding started progressively.

4.3. Functional evaluation and follow-up

The functional results of the intervention were evaluated with the efficacious help of the parents. Length growth, body weight gain, breathing or swallowing problems were systematically recorded. Breathing, swallowing, voice, and overall satisfaction was assessed using a quality of life (QOL) questionnaire [16]. The following items were rated by the parents by using the scales as indicated: dyspnea (grade 1 ‘absent’ to grade 5 ‘at rest’); noisy breathing (grade 1 ‘absent’ to grade 5 ‘very noisy breathing even at rest’); coughing (grade 1 ‘absent’ to grade 4 ‘frequent episodes’); dysphonia (grade 1 ‘normal voice’ to grade 4 ‘aphonia’); dysphagia (grade 1 ‘absent’ to grade 3 ‘nasogastric tube feeding’); and global satisfaction (grade 1 ‘totally satisfied’ to grade 4 ‘totally unsatisfied’). In all parameters lower grades meant better conditions. Voice analysis was performed 36 months after SLTP according to our previously published protocol based on the guidelines elaborated by the Committee on Phoniatrics of the European Laryngological Society [49]. The voice samples were recorded with a high sensitivity (40 Hz – 6 kHz) condenser head microphone (ATM75; Audio Technica, Machida, Tokyo, Japan) at a sampling frequency of 96 kHz. Shimmer (%), jitter (%), fundamental frequency, and the harmonics-to-noise ratio were analyzed using Praat 5.3.2.9. software [www.praat.org]. Endoscopic examinations under general anesthesia were strongly recommended in the first postoperative year. Thereafter, direct endoscopy was performed only if surgery-related airway symptoms occurred. Follow-up intervals were 125, 118, 88, 68, 66, 50, and 48 months for the patients, respectively.

5. RESULTS

There were no major peri- and postoperative complications. The postoperative timeline of vents is presented in Table 2.

Patient/ sex	Extubation [postoperative day]	Removal of NG tube [postoperative day]	Complications and further treatment	Follow-up period [month]
1/m	7^{th}	7^{th}	laser MLS	125
2/m	5^{th}	7^{th}	-	118
3/f	10^{th}	12^{th}	laser MLS; reintubation due to peumonia	88
4/f	4^{th}	10^{th}	-	68
5/f	3^{rd}	7^{th}	-	66
6/m	7^{th}	7^{th}	reintubation due to peumonia	50
7/f	5^{th}	*2^{nd} year	-	48

**Table 2: Events of the postoperative period. ***removal of the gastrotube; MLS: microlaryngosurgery

Extubation was considered safe on the 7^{th}, 5^{th}, 10^{th}, 4^{th}, 3^{rd}, 7^{th}, and 5^{th} postoperative days, for each of the seven patients, respectively. All patients were able to tolerate a progressive oral diet without the use of any thickener within 2 to 3 days post-extubation except one child, who was fed through gastro tube from the fifth week after birth (patient #7). In the case of patients #3 and #6, postoperative reintubation was necessary because of their RSV infection, for 4 and 2 days respectively. Repeated endoscopic airway surgery (UltraPulse CO_2 laser vaporization) was required in patients #1 and #3 in the second postoperative month because of the excessive formation of granulation tissue. In the other patients, only planned control endoscopies were performed (two or three times in the first postoperative year). No patient required open revision surgery. All children had stable and adequate airway during follow-up [Figure 10]. Significant restenosis was not observed in any of the patients, moreover, the subglottic airway has become wider than the physiological one after SLTP [Figure 11]. According to the parents' judgment,

breathing was normal in all patients. The children did not require an exemption from physical education. QOL scores are presented in Table 3.

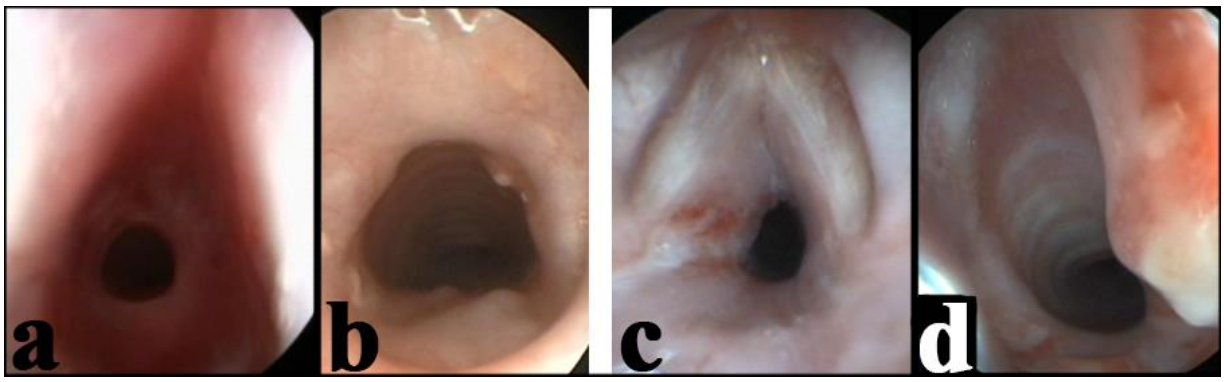

Figure 10: Preoperative and 3rd-month postoperative endoscopic pictures of patients #2 (a,b) and #3 (c,d) with Cotton–Myer grade III subglottic stenosis

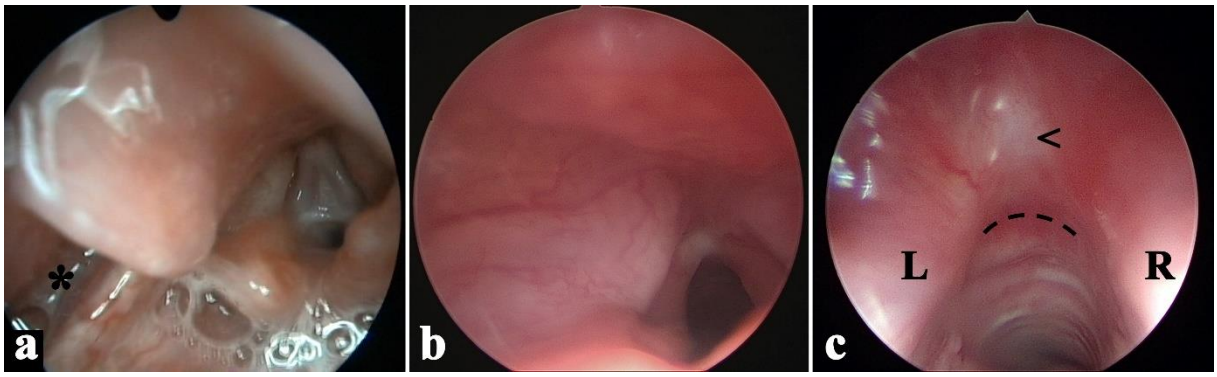

Figure 11. Direct endoscopic pictures of patient #4. a) Preoperative picture: Cotton–Myer grade III subglottic stenosis and associated Cohen grade III laryngeal web; *: nasogastric feeding tube. b) Adequate glottic airway in the 45th postoperative month. Direct endoscopy is not obligatory in the late postoperative follow-up. Laryngo-tracheoscopy was performed prior to powered intracapsular tonsillotomy and adenoidectomy for obstructive sleep apnea. c) Wide subglottic without signs of restenosis. <, minimal mucosal scar in the anterior commissure; dashed line: upper rim of the interposited trachea flap, L, left vocal fold; R, right vocal fold.

Patient/sex	Dyspnea	Noisy breathing	Coughing	Dysphonia	Dysphagia	Overall satisfaction	∑QOL
1/m	1	1	1	2	1	1	7
2/m	1	1	2	1	1	1	7
3/f	1	1	1	1	1	1	6
4/f	1	1	1	3	1	3	10
5/f	1	1	1	2	1	1	7
6/m	1	1	1	2	1	1	6
7/f	1	1	1	1	1	1	6
Range	1-5	1-5	1-4	1-4	1-3	1-4	6-25

Table 3: Subscores and total score of the quality of life questionnaire.

Patient #4 experiences learning difficulties and delayed speech development. She can create all the vowels and some consonants (e.g., k, m, n, t). In parallel, her vocabulary is extremely low, she creates only short sentences and nursery rhymes. Her 'Wechsler Preschool and Primary Scale of Intelligence Fourth Edition' score is also low and the 'MacArthur-Bates Communicative Development Inventory' shows the skill level of a 2-year-old child, despite being 5 years old. The remaining children had a social life in line with their biological age. In case of patient 4, weak voice was detected because of a 2-mm blunting at the anterior commissure. Acoustic parameters are presented in Table 4. Figures 11 and 12 present the weight-for-age and length-for-age percentiles of the patients.

Patient/sex	Shimmer [%]	Jitter [%]	HNR [dB]	Fundamental frequency [Hz]
1/m	1,73	0,36	21,54	240,0
2/m	2,76	0,87	23,24	199,9
3/f	1,25	0,25	26,78	189,8
4/f	1,87	0,77	21,11	245,9
5/f	2,29	0,3	24,45	320,4
6/m	3,05	0,66	21,11	190,1
7/f	2,54	0,79	20,3	233,7
Physiological values	<3,81	<1,04	>20	

Table 4: Acoustic parameters in the 36th postoperative month.

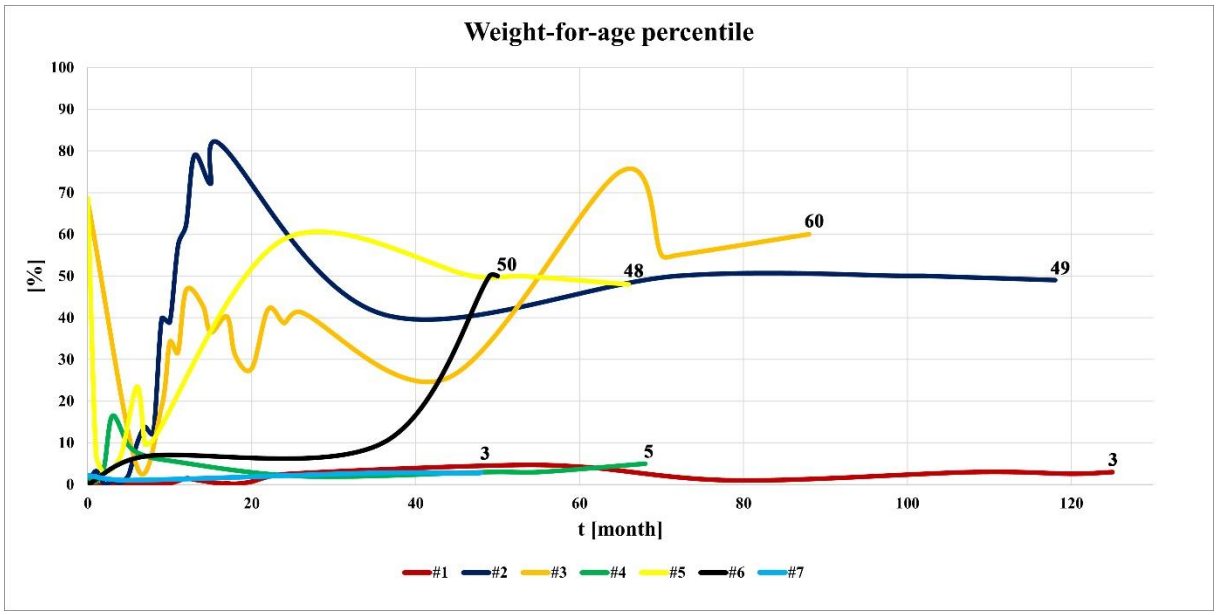

Figure 11. Weight-for-age percentile of the patients.

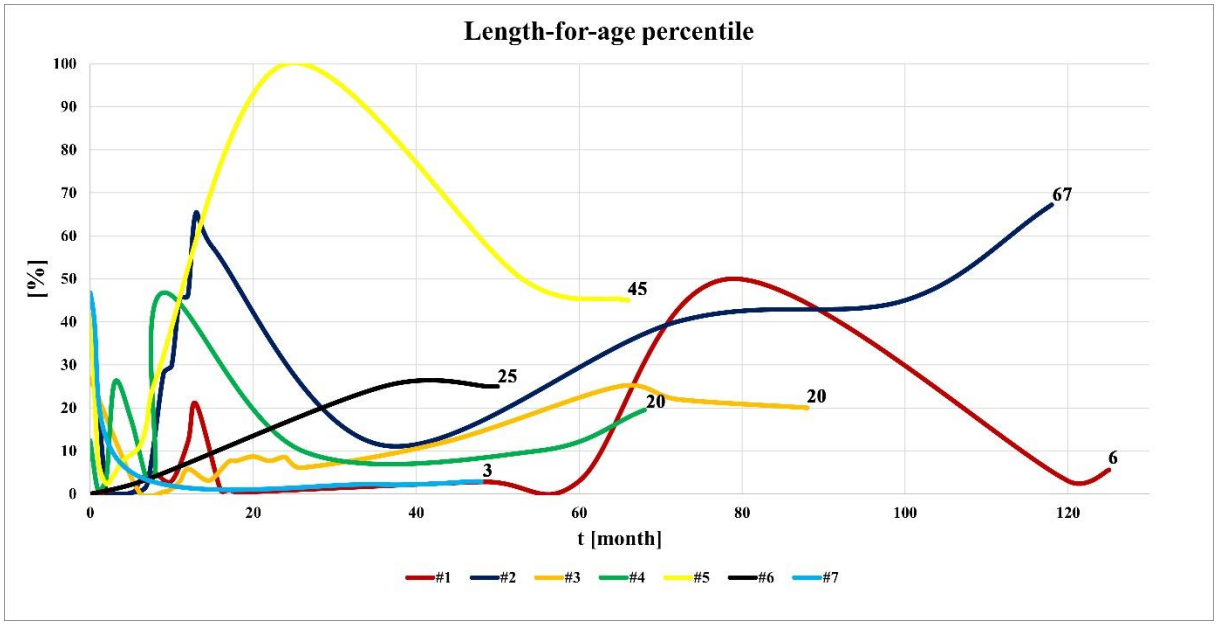

Figure 12: Length-for-age percentile of the patients.

6. DISCUSSION

After SLTP, the reconstructed subglottic area is wider than the physiological one, even without extended resection or rib cartilage grafting. This ‘reserve capacity’ ensures a patient airway even in the event of mild restenosis. The cricothyroid complex together with the interposed trachea flap provides a well-vascularized stable ring. The cartilages in these young children are flexible and are well stabilized with a double-armed continuous knotted suture. Sacrifice of the segmental tracheoesophageal arteries is necessary at the site of resection because of mobilization of the trachea and cropping of its membranous wall. However, the lateral longitudinal anastomoses and transverse intercartilaginous arteries can be preserved, potentially allowing a complication-free, quick recovery. Therefore, resorption of the local tracheal graft is modest, and the late postoperative result becomes easier to predict. In the absence of rib cartilage grafting, donor site complications can also be excluded. Furthermore, the trachea is covered by respiratory mucosa, which prevents granulations, subsequent restenosis, and adhesion of airway secretions through physiological mucociliary clearance [9,11,50]. By reducing the extent of tissue resection and omitting the dissection of the cricod cartilage, the cricothyroid muscle, the chance of recurrent laryngeal nerve injury is lower, which leads to optimal voicing and swallowing function. During the surgery, the anterior commissure remains untouched, which has an obvious positive impact on the postoperative voice quality. In the case of a severe grade subglottic stenosis, if the manipulation of the anterior commissure is unavoidable, a second-stage laryngeal framework surgery (anterior commissure retrodisplacement) might be necessary to ensure socially acceptable voice quality. However, decent voice analysis is cumbersome for infants, the objective voice parameters supported the parents’ satisfied opinion on voice quality. The children’s voices were more than socially acceptable. As demonstrated in Table 4, the assessed objective voice parameters were in the physiological ranges in all patients.

Since the integrity and the innervation of the posterior commissure remains intact, the possibility of aspiration is significantly lower. The patients' bodyweight gain and length growth were satisfactory according to growth charts, and the parents were pleased with the children's postoperative QOL in general. One child (patient #4) with Di George syndrome has experienced learning difficulties and delayed speech development. During at least 3 years of observation, the anastomosis was stable and growing dynamically with the patient.

If the general health status is appropriate and no critical comorbidity is present, SLTP is an addition to the surgical armamentarium for treating selected cases of CSGS. Adequate patient selection is crucial. The decision concerning whether to perform SLTP must be based on the overall health conditions of the infant in contrast to the severity of SGS itself. In children with certain craniofacial anomalies, neuromuscular disorder, high risk for aspiration, or low pulmonary function, decannulation may be counterproductive.

The drawbacks of this study include its small sample size and single-center nature. Therefore, further studies are required to explore the potential and limitations of this novel surgical technique.

7. CONCLUSION AND NEW RESULTS

Slide laryngotracheoplasty is an excellent single-stage procedure without tracheostomy and stenting for the reconstruction of high-grade congenital subglottic stenosis in neonates and infants to provide an adequate airway.

Swallowing function and voice production are not disrupted by the procedure, which ensure the possibility of physiological development despite the severe congenital airway anomaly.

Despite the rapid development, the newly formed laryngotracheal structure remains stable over the course of time.

8. ACKNOWLEDGEMENT

Without help, support, and encouragement from several persons, I would never have been able to complete this work.

First of all, I would like to express my gratitude and thanks to my supervisor, Dr. Ádám Bach for encouraging and supporting my work from the very beginning. I would also like to thank Prof. Dr. László Rovó. As the Head of the Department, he introduced me to clinical work and provided me with the opportunity to begin my scientific career as well. I am deeply indebted to my colleagues, the members of our upper airway stenosis workgroup, and especially to Dr. Balázs Sztanó for his cooperative work. Thanks to my colleagues at the Department of Oto-Rhino-Laryngology, Head and Neck Surgery, and the Department of Pediatrics and Pediatric Health Center. My thoughts and gratitude also go toward my family, husband, and friends whose continuous support, encouragement, belief, and patience filled me with energy and made everything worthwhile.

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10. APPENDIX

Quality of life' kérdőív felső légúti szűkületes betegek számára

Név: **Dg.:**

- Nehézlégzés
    1. Nincs
    2. Nehéz munkavégzés esetén
    3. Enyhe fizikai megerőltetés esetén is
    4. Nyugalomban
    5. Tracheotomia

- Hangos légzés
    1. Nincs, a légzés hangtalan
    2. Nehéz munkavégzés eseten hangossá váló légzés
    3. Enyhe fizikai megerőltetés esetén is hangos
    4. Nyugalomban
    5. Tracheotomia

- Köhögés
    1. Nincs köhögés
    2. Enyhe köhögés
    3. Erős köhögés
    4. Tracheotomia

- Hangképzési panasz
    1. Normális
    2. Kis fokú beszédzavar (hangos környezetben nehezebben érthető)
    3. Súlyos beszédzavar
    4. Aphonia

- Nyelési panasz
    1. Normális
    2. Kisebb nehézségek, időnkénti félrenyelés
    3. Súlyos nyelészavar

- Elégedettség az állapottal
    1. Teljesen elégedett
    2. Nagyrészt elégedett
    3. Kissé elégedetlen
    4. Elégedetlen

I.

The Laryngoscope

Slide Laryngotracheoplasty for Congenital Subglottic Stenosis in Newborns and Infants

László Rovó, MD, PhD; Eszter Erdélyi, MD; Zoltán Tóbiás, MD; Péter Gál, MD; Ilona Szegesdi, MD; Balázs Sztanó, MD, PhD; Kishore Sandu, MD; Ádám Bach, MD, PhD

Objectives: Subglottic stenosis is the most common laryngeal anomaly necessitating tracheostomy in early childhood. Crico- and laryngotracheal resection and laryngotracheal reconstruction—usually with autologous cartilage graft implantation—are the most effective treatments. These surgical techniques are obviously challenging in neonatal age and infancy. However, a reconstructive surgery performed at early age may prevent the sequel of complications.

Methods: The authors present their novel surgical method for congenital subglottic stenosis. Seven infants had inspiratory stridor; two of them had to be intubated and one required tracheostomy. Laryngotracheoscopy, CT or MRI revealed subglottic stenosis: Cotton-Myer grade II in one, and grade III in six cases. Slide laryngotracheoplasty was performed before 5 months of age (10–130 days), with a follow-up period of average 36 months (4–80 months). Phoniatry and quality of life questionnaire were used for evaluation of postoperative results.

Results: Slide laryngotracheoplasty in the neonatal age made the temporary tracheostomy unnecessary. All babies remained intubated for 3 to 10 days with an uncuffed tracheal tube. After extubation, no dyspnea or swallowing disorder occurred. A subjective quality of life questionnaire, laryngotracheoscopy, clinical growth charts showed satisfactory functional results.

Conclusions: Single-stage slide laryngotracheoplasty might be a favorable solution for subglottic stenosis, even in early childhood. In one step, the airway can be maintained without stenting and tracheostomy.

Key Words: Subglottic stenosis, congenital stridor, slide laryngotracheoplasty, neonatal.

Level of Evidence: 4

***Laryngoscope*, 00:1–7, 2019**

INTRODUCTION

Congenital subglottic stenosis (SGS) is the third most common congenital laryngeal anomaly after laryngomalacia and vocal fold paralysis, however this is the most common laryngeal anomaly necessitating tracheostomy in children less than 1 year of age.[1,2] Congenital SGS amounts to 5% of the laryngotracheal stenosis, but its true incidence is difficult to assess due to the high incidence of intubation in patients with preexisting narrow subglottic airway or other significant comorbidities.[3–6] The prenatal diagnosis of subglottic stenosis is not routinely possible. Severe dyspnea at birth requires urgent intubation followed by tracheotomy to allow adequate ventilation and oxygenation of the newborn. This then starts a difficult cycle of tracheostomy-related physical and psychosocial problems for the child and his or her family.[7–10]

Laryngotracheal stenosis has been a major challenge for surgeons due to the complex laryngeal structure and functions. Choosing the optimal surgical intervention is extremely difficult at this age due to the special anatomy of the pediatric airways, the vulnerable tissues and the potential associated congenital malformations. In order to prevent the late sequel and complications in such patients, a definitive (one-step) surgical solution is preferably performed as soon as possible in carefully selected patients. In the case of high grade congenital subglottic stenosis endoscopic interventions are not recommended.[11,12] According to the literature, nowadays laryngotracheal reconstruction (LTR), partial cricotracheal (PCTR), or extended cricotracheal resection (ECTR) are the most recommended interventions for severe grades of glotto-subglottic stenosis (GSGS). However, these complex procedures require significant tissue resection, airway expansion by rib cartilage grafts, and stent implantation.[5,11–14] To maintain the airway stability extended resection of the cartilaginous framework should be limited to allow tension-free anastomosis, and if graft implantation is necessary, well-vascularized, easily available, regional tissue is desirable. We describe here an innovative surgical technique for congenital SGS that has given good objective and subjective results.

MATERIALS AND METHODS

Patients

Between January 2012 and May 2018 slide laryngotracheoplasty was performed in seven consecutive patients on the 10th, 14th, 68th, 105th, 92nd, 130th, and 120th days of life, respectively. Two patients (patients 1 and 2) had been admitted to the perinatal

From the Department of Otorhinolaryngology and Head & Neck Surgery (L.R., E.E., Z.T., B.S., A.B.), the Department of Pediatrics and Pediatric Health Center (P.G.), the Department of Anesthesiology and Intensive Therapy (I.S.), University of Szeged, Szeged, Hungary; and the Department of Otorhinolaryngology, Head and Neck Surgery (K.S.), University Hospital of Lausanne, Lausanne, Switzerland.

Editor's Note: This Manuscript was accepted for publication on June 27, 2019.

The authors have no funding, financial relationships, or conflicts of interest to disclose.

Send correspondence to Eszter Erdélyi, Tisza Lajos krt. 111., 6725, Szeged, Hungary; E-mail: eszter.erdelyi@med.u-szeged.hu

DOI: 10.1002/lary.28192

TABLE I.
Pregnancy Information and Patient Data.

Patient/sex	Perinatal data	Age at laryngo-tracheoplasty (days)	Preoperative airway support	Grade of SGS [Cotton-Myer gr./ %]	Comorbidities
1/m	37 week 1980 g Apgar 8-9-9 p.v.n.	10	Oxygen mask	II 60–70%	—
2/m	40 week 3490 g Apgar 7-9-9 s.c.	14	Intubation	III 70–80%	—
3/f	38 week, 3450 g Apgar 8-10-10 p.v.n.	68	High frequency airway support	III 70–80%	Supraventricular tachycardia
4/f	37 week 2370 g Apgar 7-8-9 s.c.	105	Oxygen mask	III 70–80%	Laryngeal web, Tetralogy of Fallot, DiGeorge syndrome
5/f	36 week 3300 g Apgar: 7-8-9 p.v.n.	92	Intubation	III 80–85%	—
6/m	34 week 1850 g Apgar: 7-8-8 s.c.	130	Oxygen mask	III 70–75%	Laryngomalacia, RDS
7/f	40 week 2390 g Apgar: 2-6-8 p.v.n.	120	Tracheostomy	III 80–85%	DiGeorge syndrome

p.v.n. = per vias naturales; RDS = respiratory distress syndrome; s.c. = sectio caesarea; SGS = subglottic stenosis.

intensive care unit (PICU) immediately after birth, and two patients (patients 2 and 5) had been intubated due to severe stridor and inspiratory dyspnea. One infant (patient 3) required temporarily continuous positive airway pressure (CPAP) ventilation. One infant (patient 7) required tracheostomy and nasogastric feeding tube in the fifth week after birth. In three infants (patients 3, 4, and 5) the severe inspiratory stridor occurred after an upper airway infection. Discontinuance of oxygen support (patients 3 and 4) and definitive extubation (patient 5) was not possible in these cases. Endoscopic evaluation of the airway and reconstructive airway surgery was performed on the fifth day of intubation in patient 5 and 3 days after the onset of the dyspnea in patients 3 and 4. Pregnancy-related

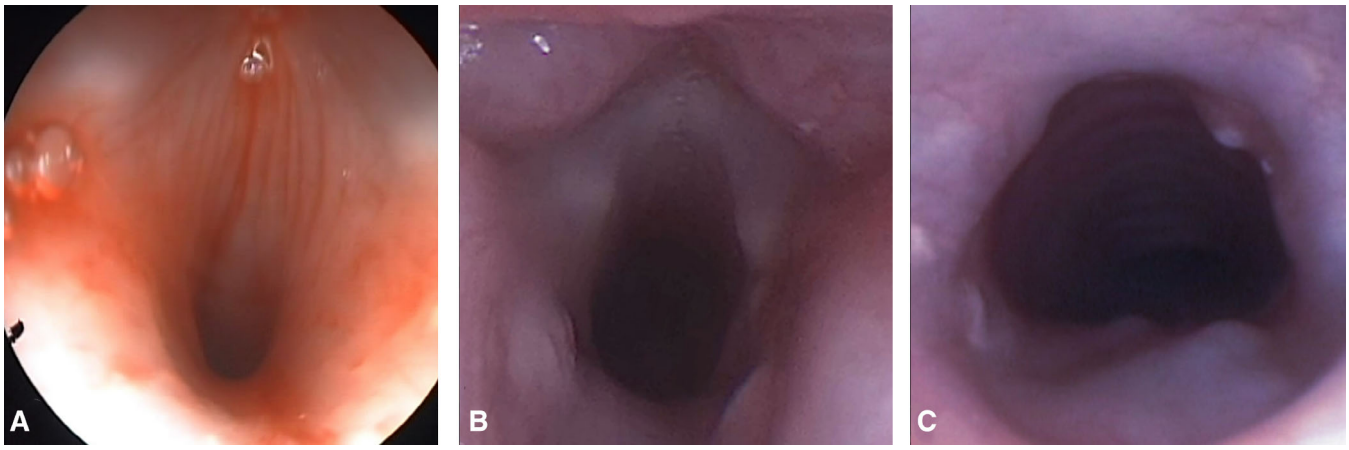

Fig. 1. Direct laryngoscopy of patient 2
(A) Preoperative laryngeal status (Cotton-Myer grade III)
(B) Postoperative picture of the glottis
(C) Postoperative picture of the glottis (13th postoperative month). [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

information and patient data is reported in Table I. Preoperative high resolution computed tomography (CT) or magnetic resonance imaging (MRI) scan was performed in all cases. The preoperative investigations included dynamic and rigid airway endoscopy along with neonatological and neurological evaluations. Concentric cartilaginous, Cotton-Myer grade III SGS was detected with direct endoscopic examination in six of seven patients (patients 2–7), and patient 1 had Cotton-Myer grade II SGS (Fig. 1).[7] In one case (patient 4), SGS was associated with Cohen grade III laryngeal web.[7]

Surgical Technique

The procedure began with direct endoscopic examination of the upper and lower airways to assess the craniocaudal extension of the stenosis and determine its (cartilaginous/soft-tissue) nature. Laryngo-tracheo-bronchoscopy with a rigid 0° and 30° endoscope was performed under general anesthesia via total intravenous anesthesia (TIVA) and high-frequency supraglottic jet ventilation with readiness for a possible intubation. Jet ventilation was performed by using the Moonsune III device (Acutronic Medical Systems, Hirzel, Switzerland) with the following parameters: inhalation time = 20%, frequency = 130/min, pause pressure = 10 cmH_2O, and peak inspiratory pressure (PIP) 12 cmH_2O. The patient with tracheostomy was induced through the cannula and an additional age-appropriate endotracheal tube (ETT) was passed orally and left in the laryngeal inlet. After the diagnostic laryngomicroscopy orotracheal intubation was performed.

The surgery began with a horizontal collar incision made at the level of the cricoid cartilage. The strap muscles were divided and the laryngotracheal complex was bluntly explored from the upper edge of the thyroid cartilage to the superior mediastinum, protecting the recurrent laryngeal nerves and the great vessels (Fig. 2A, 2B). The thyroid isthmus was divided in the midline and the cricothyroid muscles were left untouched. First, superior laryngeal release was performed: along the upper rim of the thyroid cartilage, the thyrohyoid membrane was incised until the superior thyroid horns were reached. The cricotracheal ligament was dissected and partial midline anterior laryngofissure was performed dividing the cricoid and the thyroid cartilage until the level of the anterior commissure (Figs. 2C, 2D, and 3A). The cricotracheal junction was dissected circumferentially and divided carefully avoiding injury to the oesophagus. Cross-field ventilation was carried out using a second set of sterile anesthesia tubings. After visualization of the laryngeal lumen a posterior cricoid incision was performed with preservation of the posterior perichondrium, the posterior cricoarytenoid and pharyngeal constrictor muscles integrity. The distal trachea was mobilized until the anterior cartilage rings could be easily pulled up to the level of the anterior commissure. The membranous part was resected till the level of the second tracheal cartilage (Figs. 2D and 3B).

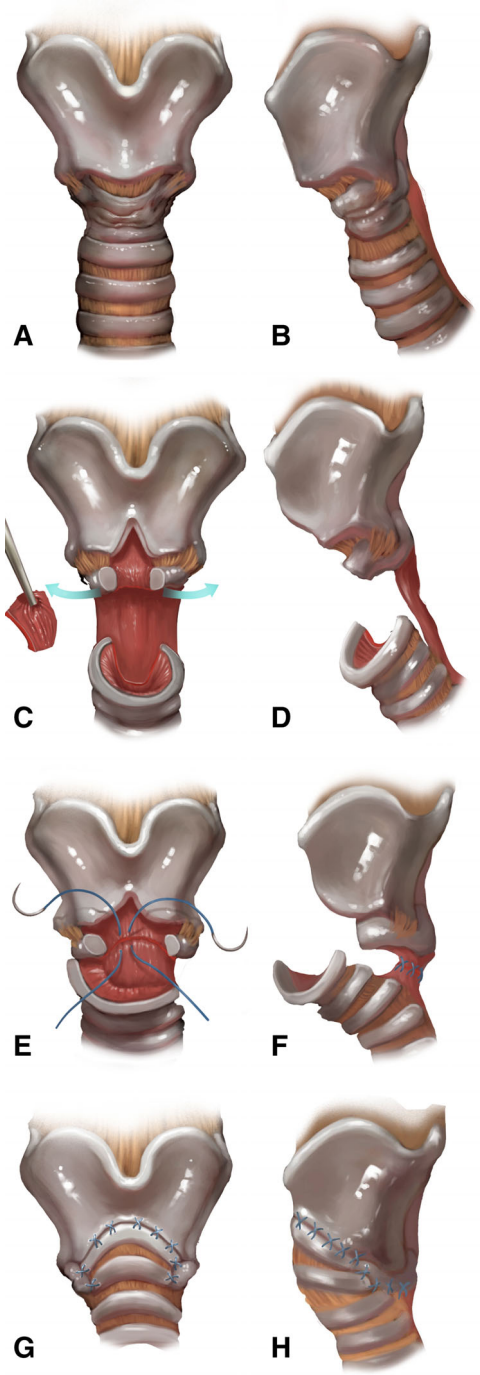

Fig. 2. The steps of slide laryngotracheoplasty (schematic drawings, anterior and lateral view)
(A,B) Subglottic stenosis, hypoplastic cricoid cartilage
(C,D) Partial anterior laryngofissure with the preservation of the anterior commissure of the vocal folds, membranous part of the trachea is partially resected till the level of the second tracheal ring
(E,F) An anastomosis between the trachea, cricoid and thyroid is created. First the posterior wall is reconstructed
(G,H) The reconstructed anterior and lateral wall. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

Surgical Variation in Case of Laryngeal Web

The membranous component of an associated grade III laryngeal web was divided in the midline using Ultra Dream Pulse (UDP) CO_2 laser (DS-40U, Daeshin Enterprise, Seoul, Korea) with 20 ms repeat time, 90 μs pulse duration, 315 W peak power, 0.16 mm spot diameter. The cartilaginous subglottic stenosis was untouched by the laser.

Laryngotracheal Anastomosis

An anastomosis was created between the trachea, the anterior cricoid and the midline incised thyroid cartilage using 2.0 or 3.0 PDS suture (Fig. 2E, 2F). The anastomosis started in the posterior midline. Two double armed continuous locked sutures were placed clockwise and counter-clockwise (Fig. 4). After the posterior wall was reconstructed the previously passed ETT was descended into the trachea, then the sutures of the lateral and the anterior wall were completed. Finally, the threads arriving

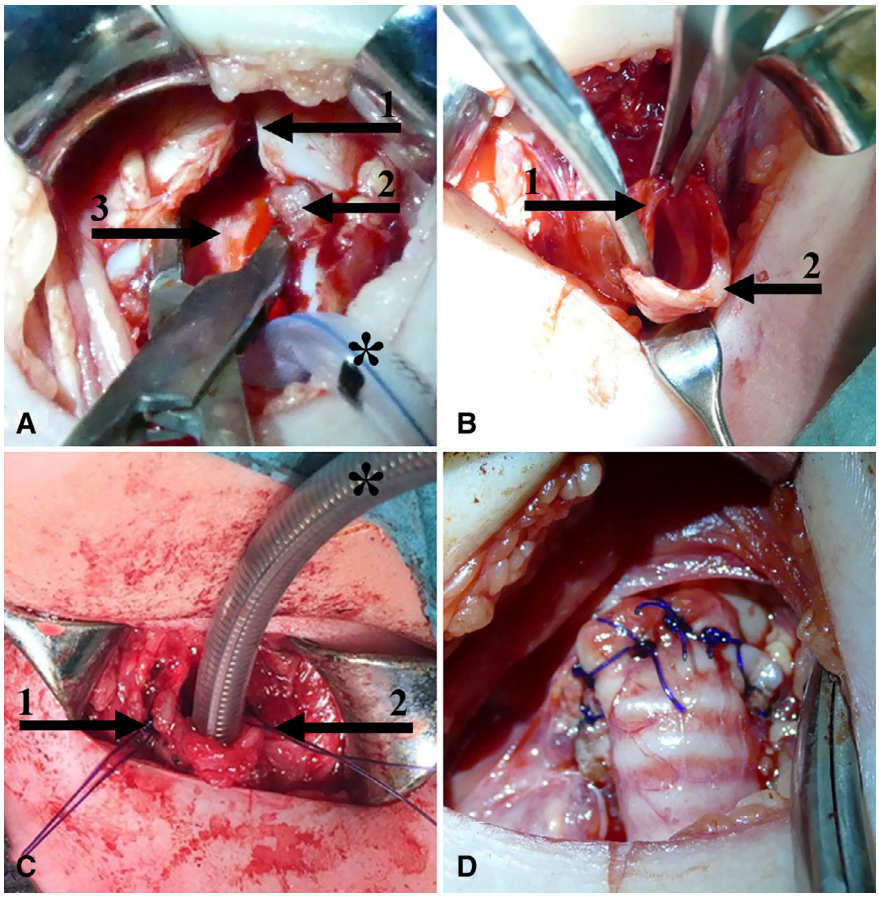

Fig. 3. Slide laryngotracheoplasty (intraoperative photos, patient 3)
(A) Partial anterior laryngofissure. Arrow 1: incised thyroid cartilage; arrow 2: cricoid cartilage, arrow 3: posterior wall of the laryngotracheal complex; *endotracheal tube
(B) Mobilized trachea Arrow 1: resection of the membranous part; arrow 2: anterior arch of the tracheal ring
(C) The posterior wall is reconstructed Arrows 1 and 2: shows the clockwise ant counter-clockwise running threads
(D) The reconstructed anterior wall. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

from the opposite directions were knotted in the anterior midline—with the knots lying outside the airway. Thus, a continuous suture ring was created (Fig. 2G, 2H, and 3C, 3D). The pre-laryngeal muscles and the skin were sutured in two layers, a 10 Ch drain (Redax - Redon) was inserted for 2 to 3 days.

Postoperative Care

The patient was transferred to a pediatric/neonatal intensive care unit. Parenteral antibiotic (amoxicillin-clavulanic acid 25 mg/5 mg/kg for 12 hours or depending on the bacteriologic aspirate) was administered for 7 days. On the day of extubation (third through 10th day), the patient was given a steroid (methylprednisolone, 4 mg/kg) bolus. Nasogastric tube feeding was continued for few days after extubation and oral feeding started progressively.

Functional Evaluation and Follow-Up

The functional outcomes of the surgery in terms of breathing, voice, swallowing, and overall satisfaction were evaluated by a quality of life (QOL) questionnaire.[15] The following items were rated by the parents of the patients using the scales as indicated: dyspnea (0 = absent to grade 4 at rest); noisy breathing (grade 0 = absent to grade 3 = very noisy breathing even at rest); coughing (grade 0 = absent to grade 2 = frequent episodes); dysphonia (grade 0 = normal voice to grade 3 = aphonia); dysphagia (grade 0 = absent to grade 3 = nasogastric tube feeding); and global satisfaction (grade 0 = outstanding to grade 4 = unsatisfied). Follow-up period included regular dynamic and rigid endoscopic examinations under general anesthesia. The parents systematically registered body weight gain, length growth, swallowing or breathing difficulty. The voice samples were recorded with a high

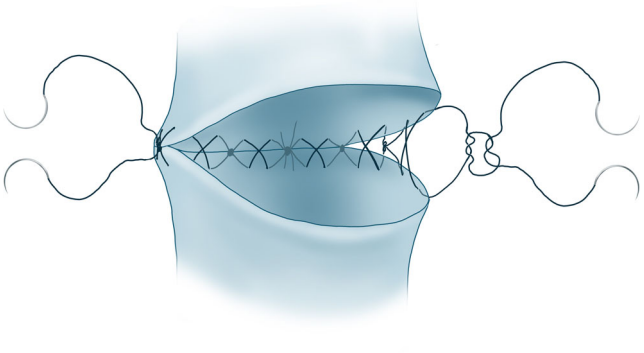

Fig. 4. Schematic picture of the anastomosis. Two double armed continuous locked sutures are running clockwise and counter-clockwise. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

TABLE II.
Events of the Postoperative Period.

Name/sex	Extubation (days)	Complications and further treatment	Follow-up period (mo)
1/m	7	mild restenosis—laser	80
2/m	5	–	73
3/f	10	Pneumonia that required reintubation (4 days), mild restenosis—laser	43
4/f	4	–	24
5/f	3	–	21
6/m	7	Pneumonia, reintubation (2 days)	4
7/f	5	–	5

sensitivity (40–6 kHz) condenser head microphone (ATM75; Audio Technica, Machida, Tokyo, Japan) at sampling frequency of 96 kHz (24 bit US analyzed by Praat 5.3.2.9., Amsterdam, Netherlands). The following acoustic parameters were recorded in this study: mean pitch (F_0), jitter, shimmer, and harmonics-to-noise ratio (HNR). Follow-up intervals were 80, 73, 43, 24, 21, 4, and 5 months for the patients, respectively.

RESULTS

There were no major peri- and postoperative complications. The postoperative timeline of events is presented in Table II. Extubation was considered safe on the 7th, 5th, 10th, 4th, 3rd, 7th, and 5th postoperative days, for each of the seven patients, respectively. In the case of patients 3 and 6, postoperative reintubation was necessary because of their RSV infection. All patients were able to tolerate progressive oral diet within 2 to 3 days post-extubation except one child, who was fed through nasogastric tube from the fifth week after birth (patient 7). The weight-for-age and length-for-age percentiles are depicted in Figure 5. The acoustic parameters, QOL scores are shown in Table III. Compared to the preoperative values, the QOL scores improved significantly. Based on the parents' observations, the breathing was normal in all cases. Gurgling and cooing were similar to their siblings according to the parents. In cases of two infants (patients 1 and 3) minor grade restenosis was detected. No patient required revision surgery. In case of patient 4, weak voice was detected because of a 2-mm blunting at the anterior commissure.

DISCUSSION

Congenital SGS is typically cartilaginous and often associated with abnormal forms of the cricoid (small hypoplastic cartilage) or may be associated with a dense laryngeal web. Minimally invasive endoscopic surgery has high potential for restenosis and hence open airway reclaiming operations are preferred in such pathologies.[16,17] The choice between LTR and PCTR predominantly depends on the severity and length of the subglottic stenosis. LTR with an anterior graft alone is used as a single-stage operation for

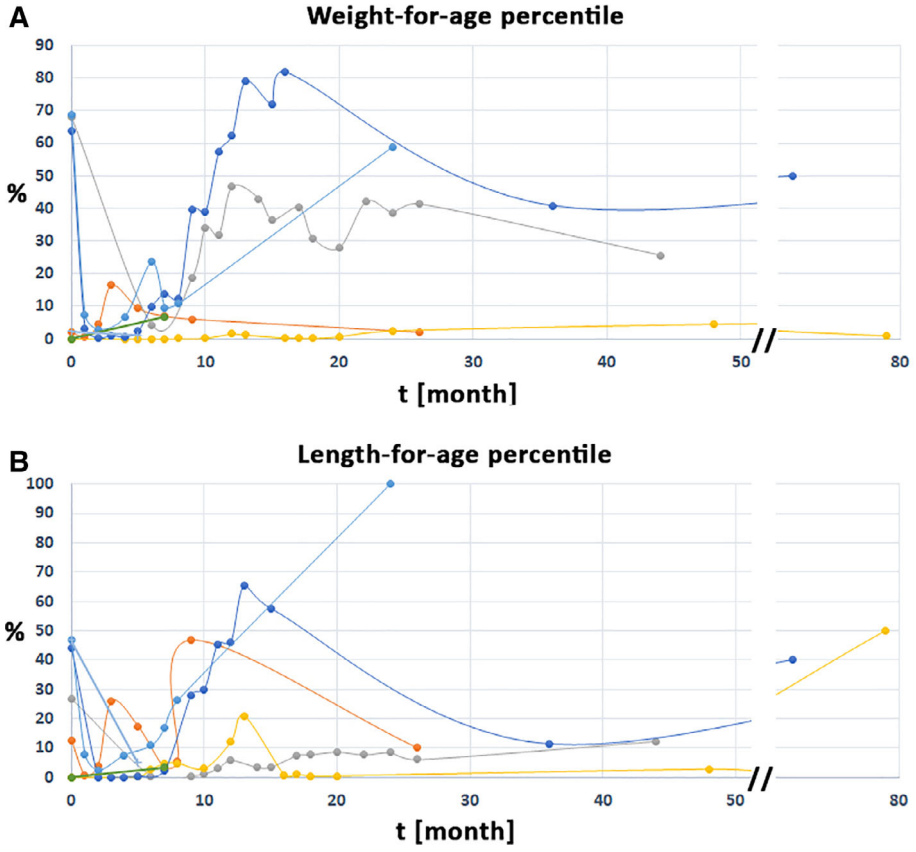

Fig. 5. Weight-for-age and length-for-age percentile. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

TABLE III.
Functional Outcomes of the Surgery.

Patient/sex	Jitter (%)	Shimmer (%)	HNR (dB)	Mean Pitch (Hz)	QoL
1/m	0.8	9.9	16.2	278	7
2/m	3.3	15.9	8.6	161	7
3/f	1.2	16.1	14.3	151	6
4/f	No data obtained				11
5/f	0.4	4.8	19.1	280	7
6/m	1.1	8.7	19.4	434	10
7/f	0.4	2.9	20.3	471	10
Physiological values	<1.04	<3.81	>20	–	Min = 6; max = 25

HNR = harmonic-to-noise ratio; QOL = quality of life score.

the management of grade II stenosis. Grade III stenosis is likely to require both anterior and posterior grafts and can be performed in one or two stages. However, over the last decade, PCTR has emerged as a superior alternative to LTR for the treatment of grade III and IV SGSs.[7,18–20] The technique of PCTR involves resection of the anterior cricoid arch, thinning and flattening of the posterior cricoid plate and performing an anastomosis between the thyroid cartilage and the trachea. The extended variant of CTR is advocated for severe SGS with additional posterior or trans-glottic stenoses, and additionally requires a full laryngofissure, posterior cricoid split and expansion using a rib cartilage graft and a temporary laryngeal stent.[12]

Due to potential comorbidities, associated anatomical malformations, and complex requirements of the treatment, tracheostomy is still needed in cases of a severe grade congenital SGS.[1,2] It severely affects the life quality of the patient and the parents as well. The complexity of the neonatal anatomy and anesthesia (small lung reserve capacity, high oxygen requirements, risk of hypo- and hyperthermia, associated comorbidities) make the surgical options even more limited in this age group. The optimal surgical intervention should be both quick, safe, and provide an immediate and adequate airway, acceptable voice quality and good swallowing function.

Because of the above-mentioned difficulties, a definitive surgical solution is usually delayed for years, although an early tracheostomy is often unavoidable.[21–24] The most common surgical solutions (CTR/ECTR, PCTR, LTR) are recommended in general from the age of 1 year.[4,11,14,15,21–27] The youngest PCTR patient described was a 1-month-old infant without any comorbidity.[22] However, even though the overall decannulation rate after CTR is over 80% - the rate of reoperation is between 4-41%, depending on the grade of the stenosis and the comorbidities. The rate of reoperation is between 22-45% after LTR.[4,11,13–15,18,19,22,25,28]

All of the presented cases included high grade, cartilaginous, concentric stenoses. Accordingly, no endoscopic interventions, but external approach was preferred. In case of slide laryngotracheoplasty the subglottic airway is augmented with a good vascularized normal-size local trachea flap. This way, no donor site reaction or graft absorption occurs, and the technique provides a wide airway at the level of the subglottis and the proximal trachea, because of the partial resection of the posterior tracheal wall and the flexibility of the intercartilaginous ligaments of the trachea. Using the slide tracheoplasty principle, we were able to make the trachea slide on a narrow cricoid and thus create a wider airway. The cartilages in these young children are flexible and are well stabilized with a double-armed continuous knotted suture. In all our patients, the reconstructed airway has remained age-appropriate, static, and undistorted.

During the surgery, the anterior commissure remains untouched, which has an obvious positive impact on the postoperative voice quality. In the case of a severe grade subglottic stenosis, if the manipulation of the anterior commissure is unavoidable, a second stage laryngeal framework surgery (anterior commissure retrodisplacement) might be necessary to ensure a socially acceptable voice quality. However, decent voice analysis is obviously cumbersome for infants, the objective voice parameters supported the patients' satisfied opinion on voice quality.

The good functional results are facilitated by the preservation of the entire cricoid cartilage, the recurrent nerves and the cricothyroid muscles. Since the integrity and the innervation of the posterior commissure (respiratory glottis) remains intact, the possibility of aspiration is significantly lower. Congenital SGS is not caused by scar tissue, but rather by an incomplete recanalization—hence the respiratory mucosa is intact. Needless to say that in airway surgery if the intact respiratory mucosa is preserved, the chances of the restenosis are lower.[3,5,21] None of our patients had any postoperative feeding difficulties. Asymptomatic, grade I restenosis was observed in two cases, which could be effectively treated by single laser resection. During the further follow-up no significant restenosis was found, and the growth of the larynx seemed to be unimpaired. The surgical method presented here has the advantage of reduced risk of anastomotic dehiscence and recurrent nerve damage that could be encountered in the conventional CTR technique.

CONCLUSION

According to our preliminary results, the single-stage slide laryngotracheoplasty might be a favorable solution for subglottic stenosis, even in early childhood. In one step, the airway can be maintained without stenting and tracheostomy. Voice production and optimal swallowing function are well preserved. Follow-up long-

term outcomes and additional patients need to be studied to further validate this procedure.

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II.

Laryngealis szűkületek innovatív sebészi megoldásai újszülött- és csecsemőkorban

Bach Ádám dr. ▪ Erdélyi Eszter dr. ▪ Sztanó Balázs dr. Tóbiás Zoltán dr. ▪ Rovó László dr.

Szegedi Tudományegyetem, Általános Orvostudományi Kar, Fül-Orr-Gégészeti és Fej-Nyaksebészeti Klinika, Szeged

A veleszületett légúti szűkületek gyakran kombináltan jelentkeznek, és más szervrendszert is érintő kísérőbetegségekkel, illetve malformációkkal is társulhatnak. Figyelembe véve ezeket a tényezőket, illetve a csecsemőkori légút speciális anatómiáját és sérülékeny szöveteit, a felső légúti szűkületek sebészi kezelése újszülött- és csecsemőkorban igen nagy kihívást jelentő feladat, melynek célja a mihamarabbi definitív, stabil légút biztosítása a hangképzés és a nyelési funkció megőrzésével. A laryngomalacia, a hangszalagbénulás és a subglotticus stenosis együttesen a gége veleszületett rendellenességeinek megközelítőleg 90%-áért felelős. A szerzők erre a három kórképre fókuszálva egy-egy eset kapcsán bemutatják a Szegedi Tudományegyetem Fül-Orr-Gégészeti és Fej-Nyaksebészeti Klinikáján működő légútsebészeti munkacsoport által rutinszerűen alkalmazott innovatív sebészi módszereket. A bemutatott sebészeti megoldások egy lépésben, tracheostoma, sztentelés és graft beültetése nélkül azonnali stabil légutat biztosítanak jó hangminőséggel és nyelési funkcióval a supraglottis, a glottis és a subglottis dinamikus és statikus szűkületei esetén egyaránt.
Orv Hetil. 2021; 162(52): 2100–2106.

Kulcsszavak: hangszalagbénulás, laryngomalacia, subglotticus stenosis, 'slide' laryngotracheoplastica, endoszkópos arytenoid abdukciós lateropexia

Innovative surgical solutions for laryngeal stenoses in newborns and infants

Congenital airway stenoses occur frequently in combinations or may be associated with comorbidities and malformations affecting other organ systems. Considering these factors as well as the special anatomy and vulnerable tissues of the pediatric airway, surgical treatment in neonates and infants is an extremely challenging task. The ultimate goal of the management is to ensure a definitive and adequate airway as soon as possible with the preservation of voice and swallowing. Laryngomalacia, vocal cord palsy and subglottic stenosis together account for approximately 90% of congenital laryngeal disorders. Focusing on these three diseases, the authors – the airway surgery working group at the Department of Otolaryngology and Head and Neck Surgery, University of Szeged, Hungary – present their routinely applied innovative surgical strategies in connection with three cases. The presented 'one-step' surgical solutions provide immediate stable airway with good voice quality and swallowing function without tracheostomy, stenting, or graft implantation for both dynamic and static stenoses of the supraglottis, glottis, and subglottis.

Keywords: endoscopic arytenoid abduction lateropexy, laryngomalacia, slide laryngotracheoplasty, subglottic stenosis, vocal cord palsy

Bach Á, Erdélyi E, Sztanó B, Tóbiás Z, Rovó L. [Innovative surgical solutions for laryngeal stenoses in newborns and infants]. Orv Hetil. 2021; 162(52): 2100–2106.

(Beérkezett: 2021. május 6.; elfogadva: 2021. június 11.)

Rövidítések

EAAL = endoszkópos arytaenoid abdukciós lateropexia; ETGI = (endolaryngeal thread guide instrument) endolaryngealis fonalvezető eszköz; SLtp = 'slide ' laryngotracheoplastica

A laryngotrachealis komplexum csecsemő- és gyermekkori szűkületei számos olyan veleszületett és szerzett kórképet foglalnak magukban, melyek potenciális veszélyeik miatt gyors és precíz diagnosztikát, illetve személy-

re szabott, lehetőleg egy lépésben történő sebészi megoldást követelnek meg [1, 2]. A légút fejlődési rendellenességei ritkák, pontos gyakoriságuk nem ismert, incidenciájuk 2–10/100 000 élveszülés intervallumra tehető [3]. A szerzett esetek száma az utóbbi évtizedekben növekvő tendenciát mutat a hosszan tartó intubáció elterjedése és a kis súlyú új- és koraszülöttek túlélési esélyeinek növekedése miatt. A szóban forgó kórképek az enyhe inspiratorikus stridortól kezdve az akut légzési elégtelenségig széles skálán mozgó panaszokat okozhatnak [4]. Habár a főként az endoszkópos vizsgálatokra támaszkodó diagnosztikus lépések és kritériumok nemzetközi szinten is jól meghatározottak, a laryngotrachealis stenosisok menedzsmentjéből a mai napig hiányoznak a jól definiált sebészeti protokollok. A legtöbb ellátóhelyen a kezelési stratégia legalább annyira függ a légúti team tapasztalataitól, mint a csecsemő állapotától [5].

A régió szűkületeinek sebészi megoldása igen nagy kihívást jelent. Végső cél a definitív, stabil légút biztosítása, a hangképzés és a nyelési funkció megőrzésével. A veleszületett légúti szűkületek gyakran kombináltan jelentkeznek, és más szervrendszert érintő kísérőbetegségekkel, illetve malformációkkal is társulhatnak [6]. Figyelembe véve ezeket a tényezőket, illetve a csecsemőkori légút speciális anatómiáját és sérülékeny szöveteit, a megfelelő sebészi beavatkozás megválasztása létfontosságú ebben a fiatal korosztályban. A csecsemők gyors fejlődési üteme miatt a laryngealis struktúrák posztoperatív stabilitása szintén kiemelt fontossággal bír a hosszú távú eredmények szempontjából.

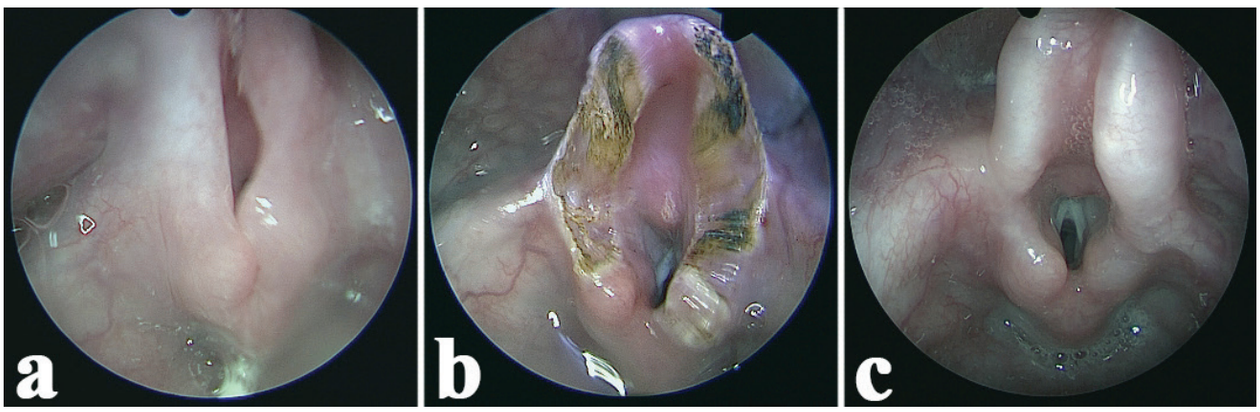
Az elmúlt évtizedekben a légútsebészet komoly fejlődésen ment keresztül az új, innovatív műtéti technikák elterjedésével és az egyre fejlődő anesztéziai lehetőségek által [4]. A legtöbb esetben azonban még napjainkban is a sürgősségi, elsődleges légútbiztosítási módszer a tracheotomia, annak minden komplikációjával, pszichés és pszichoszociális nehézségével együtt [7–9]. A laryngomalacia, a hangszalagbénulás és a subglotticus stenosis együttesen a gége veleszületett rendellenességeinek megközelítőleg 90%-áért felelős [10]. Jelen munkánk célja, hogy egy-egy eset kapcsán illusztráljuk a Szegedi Tudományegyetem Fül-Orr-Gégészeti és Fej-Nyaksebészeti Klinikáján működő légútsebészeti munkacsoport által rutinszerűen alkalmazott innovatív sebészi koncepciókat.

Első eset

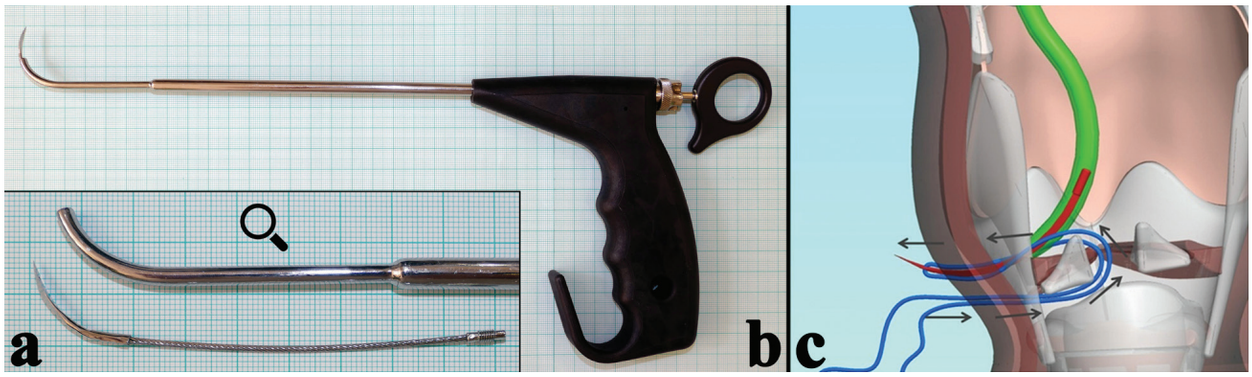
A két hónapos csecsemő esetében alváskor jelentkező, testhelyzettől függő intenzitású horkolás és jelentős táplálási nehezítettség miatt indult kivizsgálás. A légúti status felmérése során ómega alakú epiglottist észleltünk, illetve belégzéskor az aryepiglotticus redők kifejezett obstrukciót okozó collapsusát tapasztaltuk (II-es típusú laryngomalacia). Ennek megfelelően aryepiglottoplastica mellett döntöttünk, amelyet intratrachealis narkózisban, ultrapulzációs CO_2-lézerrel végeztünk el *(1. ábra)*. A mindössze 15 perces beavatkozás során az ómega alakú epiglottis széli részének vaporizációja és az aryepiglotticus redők ékreszekciója történt. A műtétet követően a csecsemőt intubált állapotban a gyermek-intenzívosztályra adtuk át, ahol 2 órás obszervációt követően extubálást végeztek. A műtétet követő napon intenzív osztályos megfigyelés már nem volt indokolt. Eseménytelen obszervációt követően a csecsemőt a 4. posztoperatív napon emittálták. Stridora ekkorra már lényegében megszűnt, ezzel párhuzamosan a csecsemő *per os* táplálhatósága is rendeződött.

Második eset

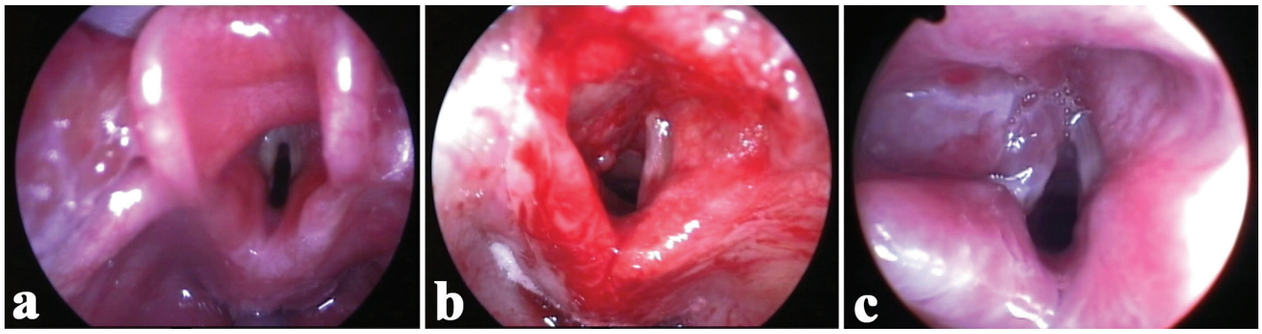
A 37. gestatiós hétre született újszülött esetében közvetlenül a születést követően inspiratorikus stridort észleltek. *Per os* táplálása nagymértékben akadályozott volt, emellett folyamatos oxigénszupplementációs igénye is fennállt. Az 5 napos korában végzett direkt endoszkópia során kétoldali gégefélbénulást észleltünk, paramedián



1. ábra II-es típusú laryngomalacia műtéti megoldása: CO_2-lézeres aryepiglottoplastica két hónapos csecsemő esetén (endoszkópos képek). a) Az aryepiglotticus redők jelentős collapsusa látható belégzéskor. A gégebemenet nehezen hozható látótérbe. b) Közvetlenül az epiglottis széli részének vaporizációja és az aryepiglotticus redők ékreszekciója után a hangszalagok láthatóvá válnak. A gégebemenet feltágul. c) 1 hónappal a műtét után a nyálkahártya teljes regenerációja látható. Belégzéskor dinamikus obstrukció nem észlelhető



2. ábra A csecsemők számára kialakított fonalvezető eszköz (endolaryngeal thread guide instrument, ETGI) és az endoszkópos arytaenoid abdukciós lateropexia (EAAL) sémás ábrája. a) Csecsemők számára kialakított tűvezető szár és tű (2,5-szeres nagyítás). b) Csecsemők számára kialakított endolaryngealis fonalvezető eszköz (tűvezető szár és a markolat összeszerelve). c) A processus vocalis körül kialakított kettős öltés a kannaporcot maximálisan abdukált helyzetben rögzíti (sémás ábra, a gége hátsó nézetből)



3. ábra Bal oldali endoszkópos arytaenoid abdukciós lateropexia kétoldali hangszalagbénulásban szenvedő ötnapos újszülött esetén (endoszkópos képek). a) Endoszkópos vizsgálat során a hangszalagok sem abdukciós, sem addukciós mozgást nem mutatnak. A hangrés szűk. b) Bal oldali endoszkópos arytaenoid abdukciós lateropexia intraoperatív képe. c) Két héttel a beavatkozást követően kellően tág hangrés látható. A bal gégefél lateralizált helyzetben

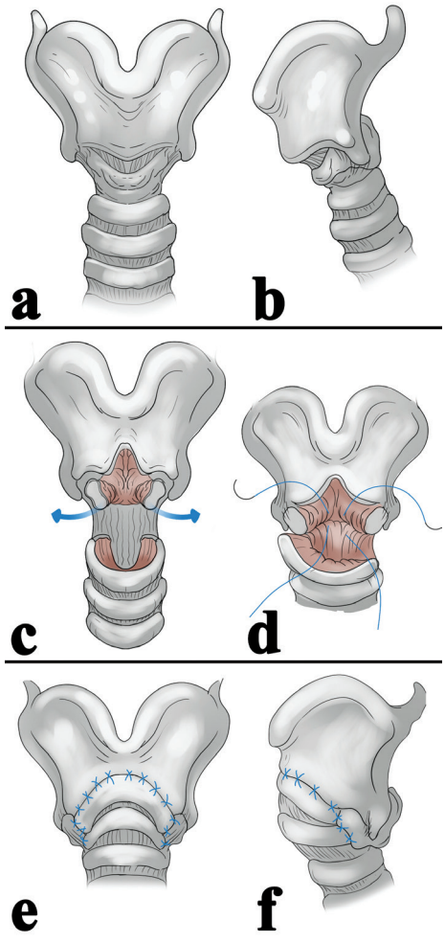
állású hangszalagokkal. 'Jet' narkózisban, szteroid- és antibiotikumvédelemben bal oldali endoszkópos arytaenoid abdukciós lateropexiát (EAAL) végeztünk, melynek során dupla, nem felszívódó fonallal a processus vocalist megkerülve a kannaporcot és ennek következtében a hangszalagot egy speciális fonalvezető eszköz (endolaryngeal thread guide instrument; ETGI; Mega Kft., Szeged) segítségével fiziológiás, maximálisan abdukált helyzetben rögzítettük *(2. és 3. ábra)* [11–13]. A gyermeket intubálva az újszülött-intenzívosztályra adtuk át. 5 napos intubációt követően szteroidbolus adása után az újszülöttet műtéti körülmények között – készen állva az esetleges azonnali légútbiztosításra – sikeresen extubáltuk. Ezt követően a stridor és az oxigénszupplementációs igény megszűnt. Az extubálást követő 3. napon a nasogastricus tápszonda is eltávolításra került. *Per os* táplálása során aspiráció jelei nem mutatkoztak.

Harmadik eset

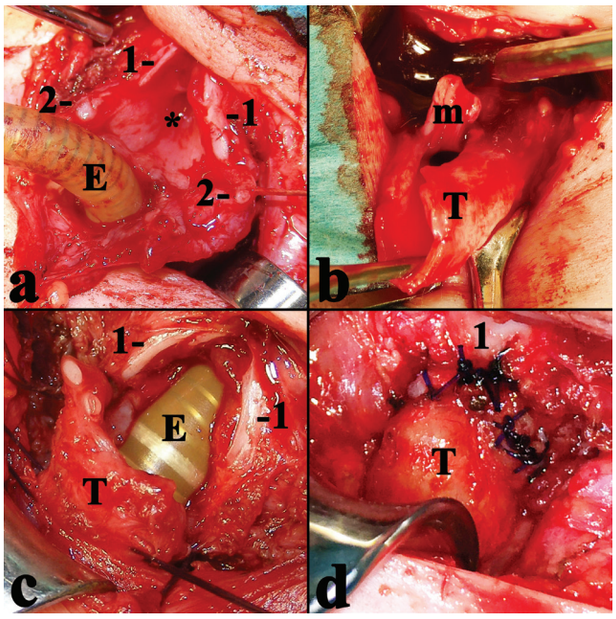
A közvetlenül a születése után intubált, 10 napos újszülött esetében direkt endoszkópia során körkörös, porcos, Cotton–Myer grade III subglotticus stenosis igazolódott, így külső feltárásból történő légúttágító műtét mellett döntöttünk [4]. 'Slide' laryngotracheoplastica (SLtp) során – bőséges laryngotrachealis mobilizációt követően – részleges elülső laryngofissiót és hátsó laminotomiát végzünk, majd a gyűrűporc ívét és részben a pajzsporcot is a trachea elülső falának interpozicionálásával tágítottuk fel *(4. és 5. ábra)* [14]. Az anasztomózis épségének érdekében a gyermek 7 napig intubálva maradt. A 8. posztoperatív napon történt extubálást követően a gyermek légzése akadálytalanná vált, ismételt légúttágító beavatkozás már nem volt indokolt.

Megbeszélés

Az áramlási ellenállás fordítottan arányos a légút sugarának negyedik hatványával [15]. Ennek megfelelően a légút sugarának felére csökkenése az ellenállás tizenhatszoros emelkedésével jár. Ez a szignifikánsan megnövekedett rezisztencia hatalmas terhet jelent az újszülöttekre, és jelentős mértékben gátolja a csecsemők fizikális és pszichés fejlődését. Ennek tükrében a hosszan tartó szoros obszerváció, illetve a „watch and wait policy" nem javasolt. Amennyiben a noninvazív pozitív nyomású lélegeztetés nem elégséges, intubáció, hosszabb távon pedig sebészeti beavatkozás szükséges. Mint urgens,



4. ábra A 'slide' laryngotracheoplastica sémás ábrája (elülső és oldalsó nézet). a) és b) Subglotticus stenosis és hypoplasiás gyűrűporc (preoperatív ábra). c) Parciális elülső laryngofissio az elülső commissura megkímélésével. A trachea membranosus fala az anasztomózis megfelelő illeszkedése érdekében a második porcig részben reszekált. d) Anasztomózis kialakítása a trachea a gyűrű- és a pajzsporc között. Elsőként a hátsó falat rekonstruáljuk. e) és f) A rekonstruált laryngotrachealis komplexum elülső és oldalsó fala



5. ábra A 'slide' laryngotracheoplastica tíznapos újszülött esetén (intraoperatív képek). a) Parciális elülső laryngofissio a hangszalagok szintjéig
1 = a pajzsporc lemezei a középvonalban szétválasztva; 2 = a gyűrűporc íve a középvonalban szétválasztva; E = endotrachealis tubus; *gégelumen
b) A mobilizált trachea (T) membranosus részét (m) a 2–3. tracheaporcig reszekáljuk a lehető legnagyobb lumen elérése érdekében. (Az endotrachealis tubus ideiglenesen eltávolítva.)
c) A hátsó fali varratsor elkészülte után transoralis intubációval folytatjuk a műtétet.
1 = a pajzsporc lemezei; E = endotrachealis tubus; T = trachea
d) Kétoldalról haladva kialakítjuk a pajzs-, a gyűrűporc és a trachea közötti anasztomózist az elülső falon is.
1 = a pajzsporc egyesített lemezei; T = trachea

életmentő beavatkozás, a tracheotomia érdemei vitathatatlanok, a műtét azonban együtt jár a hangminőség, a nyelési funkció és az életminőség drasztikus romlásával [16]. A definitív, egy lépésben végezhető sebészi megoldások megelőzhetik a következményes problémákat: a magas egészségügyi költségeket, a többszörös beavatkozásokat, a tracheamalacia kialakulását, a tracheostoma beszédfejlődésre káros hatásait, a rossz hangminőséget, az esetleges véletlen dekanülációt, a légút elzáródását és a krónikus légúti infekciók kialakulását [17–19].

Felső légúti szűkület gyanúja estén a direkt endoszkópos vizsgálat a diagnosztika legfőbb módszere, amely kisgyermekek esetén dominálóan általános érzéstelenítésben végezhető. Amennyiben a gyermek anamnézise, illetve tünetei szignifikáns légúti stenosis gyanúját vetik fel, intézetünkben rutinszerűen propofol indukálta narkózisban, spontán légzés és oxigén adása mellett flexibilis, majd merev eszközzel direkt laryngotracheoscopiát végzünk. 2014 és 2019 között 75 kisgyermeknél történt meg a fenti módszerrel a légúti status felmérése. Eredményeinket az *1. táblázat* foglalja össze.

Amennyiben a csecsemő nem szenved mentális retardációban és semmilyen más jelentős kísérő betegségben (főként pulmonalis, cardialis vagy neurológiai abnormalitástól mentes), a légúti szűkületeket célszerű egy lépésben, tracheostoma kialakítása nélkül megoldani. Kritikus fontosságú, hogy a hosszú távon sikeres légúti beavatkozás legnagyobb esélye az elsődleges sebészeti beavatkozás idején van [20]. Ennek megfelelően a többszöri, nem megfelelően kiválasztott légúti műtétek elkerülése létfontosságú. A minimálisan invazív endoszkópos beavatkozások előnyben részesítendők, amennyiben azokkal definitív, stabil légút biztosítható. Az endoszkópos beavatkozások sikertelensége esetén azonban azok ismétlése helyett külső feltárásból végzett műtét mérlegelése javasolt, még annak tudatában is, hogy ezek a beavatkozások szignifikánsan nagyobb terheléssel és morbiditással járnak.

A leggyakoribb, dyspnoét okozó congenitalis betegség a laryngomalacia, mely a supraglotticus struktúrák dinamikus és passzív összeesésével járó szerkezeti és

1. táblázat Felső légúti szűkület gyanúja miatt végzett direkt laryngotracheoscopia eredményei, illetve az alkalmazott műtéti terápia. További 17 gyermeknél nem észleltünk műtéti beavatkozást igénylő légúti szűkületet

Diagnózis	Laryngomalacia	Kétoldali hangszalagbénulás	Subglotticus/trachea stenosis	Laryngealis web
Esetszám	21	21	15	1
Beavatkozás	CO_2-lézeres supraglottoplastica	Féloldali EAAL	CTR, tracheareszekció, SLtp	CO_2-lézeres reszekció

CTR = cricotrachealis reszekció; EAAL = endoszkópos arytaenoid abdukciós lateropexia; SLtp = 'slide' laryngotracheoplastica

funkcionális elváltozás [21–24]. Az esetek megközelítőleg 15%-ában a laryngomalacia súlyos dyspnoét, obstruktív alvási apnoét és a fizikális fejlődést szignifikánsan befolyásoló táplálási nehezítettséget okoz, melyek miatt elkerülhetetlen a sebészi beavatkozás. A nemzetközileg elfogadott megoldás napjainkban az endoszkópos supraglottoplastica, melynek alapjait az 1980-as években fektették le. A műtét elvégzéséhez a mai napig több típusú eszköz használatos [25]. A „hideg" mikrosebészeti műszerekkel történő beavatkozás és a CO_2-lézer használata egyaránt elfogadott, ezek mellett ritkábban alkalmazott módszer a microdebrider is. A lézer használatát 1985-ben *Seid és mtsai* vezették be a megrövidült aryepiglotticus redők reszekciójára [26]. Hazánkban 2002-ben *Katona Gábor és mtsai* elsőként végeztek hideg-eszközös supraglottoplasticát csecsemőn [27]. A lézer vitathatatlan előnye, hogy a jól kontrollált vágás mellett alkalmas vérzéscsillapításra is. A technika ellenzői ezzel szemben a potenciális hőkárosodás veszélyeire (fokozott ödémaképződés, fájdalom, dysphagia, hegesedés) hívják fel a figyelmet. Ezek a hátrányok azonban kiküszöbölhetők, amennyiben a folyamatos energiájú CO_2-lézer helyett ultrapulzációs lézert használunk, melynek energiaprofilja eltér a hagyományos CO_2-lézerétől. Az ultrapulzációs módra jellemző, hogy az energiát a lézer háromszögimpulzusokban adja le, így az impulzusok közötti időben a szöveteknek van idejük lehűlni. Ebből adódóan kisebb az oldalhő mértéke, csökken a szöveti károsodás, minimalizálható az ödéma, így a gyógyulás időtartama is lerövidül [28]. A morfológiai variánsoknak megfelelően a műtéti beavatkozások a következők lehetnek [29]: Enyhe esetben (I. típus: az arytaenoid, valamint a corniculataporcok anterocaudalis collapsusa) az aryepiglotticus redőt a cartilago corniculatumtól indulva az epiglottis széléig haladva vaporizáljuk. A II. típus esetében (a rövid aryepiglotticus redők prolapsusa) az ómega alakú epiglottis széli részét vaporalizáljuk, és az aryepigloticus redőkön ékreszekciót végzünk *(1. ábra)*. Súlyos esetben (III. típus: az epiglottis posterocaudalis suctiója) a laza epiglottison a lingualis felszínen és a nyelvgyöki tonsillán is nagy hegesedést okozó sebzést ejtünk, és az epiglottist a nyelvgyökhöz öltjük. A beavatkozást követően a gyermekek 1–2 órán keresztül intubálva maradnak. Az intenzív osztályt 24 órán belül elhagyhatják [28].

A hangszalagbénulás a gége második leggyakoribb fejlődési rendellenessége [30]. A féloldali esetek újszülött- és csecsemőkorban túlnyomó többségükben nem igényelnek kezelést, természetüknél fogva gyakran diagnosztizálatlanok maradnak [31]. Kétoldali hangszalagbénulás esetén azonban a következményes súlyos fokú dyspnoe azonnali beavatkozást követel [32]. Az irodalomban számos, külső feltárásból (arytaenoidectomia, arytaenoidopexia, arytaenoidectomia a hangszalagok öltéssel történő lateralizálásával, a hátsó commissura tágítása porcgraft beültetésével) és endoszkópos úton (CO_2-lézeres arytaenoidectomia, lézeres posterior cordotomia, hangszalag-lateralizáció) végzett hangréstágító műtét ismert [33]. Ezek általában a glottis struktúráinak változó mértékű reszekciójával járnak. A laryngomalacia sebészetéhez képest az alkalmazott sebészi megoldások heterogenitása szembeötlő. Az újszülöttkori hangszalagbénulások kb. 60%-a részben vagy teljesen reverzibilis [34]. A bénulás észlelésekor azonban a prognózis nem jósolható meg, az esetleges regeneráció heteket, hónapokat vehet igénybe. Ennek megfelelően célszerű minimálisan invazív, a gégestruktúrákban irreverzibilis változásokat nem okozó megoldást választani a dyspnoe megszüntetésére. Az optimális sebészi beavatkozás reverzibilis, kis műtéti terheléssel jár, gyors, azonnal megfelelő tágasságú légutat biztosít a hangképzési és a nyelési funkció megőrzésével. Ezeket az elvárásokat a bemutatott EAAL maradéktalanul teljesíti. Kiemelendő, hogy a hangszalagok megfelelő abdukciós mozgásainak visszatérésekor a lateralizáló öltések egyszerűen eltávolíthatók. Az EAAL a hangszalagok pseudoparesisekor (mechanikai fixációjakor), a hátsó commissurahegesedés esetén is jól alkalmazható technika a kannaporcok ideiglenes rögzítésére. A cricoarytaenoid ízületet fixáló hegek oldását követően a mobilizált kannaporcok kétoldali lateralizációjával megelőzhető a hegek ismételt kialakulása [35, 36]. Amint a hátsó commissura területén a reepithelisatio lezajlott, a lateralizáló öltések ugyancsak eltávolíthatók.

Subglotticus stenosisról beszélünk, amennyiben a subglottis átmérője újszülött esetében kevesebb mint 4 mm, koraszülött esetében kevesebb mint 3 mm [4]. A laryngomalacia és a hangszalagbénulás után ez a gége harmadik leggyakoribb fejlődési rendellenessége, azonban ez a kórkép a csecsemőkori tracheotomia leggyako-

ribb oka [37, 38]. A minimálisan invazív endoszkópos technikák (hidegeszközzel vagy lézerrel történő hegkimetszés, ballonos dilatáció, lokális szteroid- és/vagy mitomicininjekció) csak kis fokú szűkületek esetén adnak definitív megoldást. Emellett a fenti beavatkozások növelik az esetleges urgens légúti beavatkozások incidenciáját, és késleltethetik a probléma végleges megoldását [39, 40]. Mindemellett a minimálisan invazív technikák eredményessége fordítottan arányos a subglotticus stenosis súlyossági fokával [41–43]. Amennyiben porcos vagy kombinált laryngotrachealis stenosist észlelünk, vagy a szűkült szakasz 1 cm-nél hosszabb, illetve a trachea membranosus fala is érintett, a restenosis magas valószínűsége miatt endoszkópos beavatkozások nem javasoltak. A laryngotrachealis rekonstrukció és a cricotrachealis szegment reszekciója évtizedek óta alkalmazott, jól bevált műtéti módszerek megbízható, hosszú távú eredményekkel. Ezek a komplex beavatkozások azonban nagy méretű reszekciót, bordaporcgrafttal történő légúti tágítást, sztentbehelyezést és tracheotomiát igényelnek [44, 45]. Ezeket a hátrányokat az intézetünkben kialakított SLtp képes áthidalni. A 'slide' tracheaplastica elvének módosításával a fiziológiásnál tágabb subglotticus tér, illetve jó vérellátású, stabil szerkezetű laryngotrachealis anasztomózis hozható létre. Az anasztomózis szintjében ép nyálkahártyával borított és porctámasztékkal rendelkező stabil varratsor alakítható ki, mely azonnal megfelelően tág, stabil légúti keresztmetszetet biztosít, lerövidíti a sebgyógyulást, csökkenti a granulációképződés lehetőségét, és szükségtelenné teszi az ideiglenes tracheostomát és a hosszú ideig tartó sztentelést [14]. A műtéttel elérhető, fiziológiásnál tágabb subglotticus tér esetleges enyhe fokú restenosis esetén is megfelelő tágasságú légutat biztosít.

Következtetés

A bemutatott sebészi módszerek egy lépésben, tracheostoma, sztentelés és graft beültetése nélkül azonnali stabil légutat biztosítanak jó hangminőséggel és nyelési funkcióval a supraglottis, a glottis és a subglottis dinamikus és statikus szűkületei esetén egyaránt. Alkalmazásukkal elérhető, hogy a ritka, ám az életminőségre igen súlyos hatást gyakorló felső légúti szűkületekben szenvedő gyermekek teljes életet éljenek, illetve fizikális és pszichés fejlődésük a kortársaikkal azonos ütemű legyen. A veszélyeztetett légút menedzsmentje komplex feladat, amely csak a legmagasabb szintű személyi és technikai feltételek együttes teljesülése mellett lehet sikeres, ezért minden, légúti szűkületben szenvedő gyermek kezelése erre kialakított centrumban kell hogy történjen!

Anyagi támogatás: A közlemény megírása, illetve a kapcsolódó kutatómunka anyagi támogatásban nem részesült.

Szerzői munkamegosztás: B. Á.: A kézirat megszövegezése, irodalmi áttekintés, műtétek elvégzése. E. E.: Irodalmi áttekintés, műtétek dokumentálása. Sz. B.: Szakmai véleményezés és tanácsadás, műtétek elvégzése. T. Z.: Irodalmi áttekintés, műtétek dokumentálása, az illusztrációk elkészítése. R. L.: Szakmai véleményezés és tanácsadás, műtétek elvégzése és kifejlesztése. A cikk végleges változatát valamennyi szerző elolvasta és jóváhagyta.

Érdekeltségek: A szerzőknek nincsenek érdekeltségeik.

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(Bach Ádám dr.,
Szeged, Tisza Lajos krt. 111., 6725
e-mail: bach.adam@med.u-szeged.hu)

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International Journal of Pediatric Otorhinolaryngology 158 (2022) 111192

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Long-term results of slide laryngotracheoplasty for congenital subglottic stenosis in newborns and infants

Eszter Erdélyi [a], Ádám Bach [a,*], Balázs Sztanó [a], Péter Gál [b], Kishore Sandu [c], Csaba Bereczki [b], László Rovó [a]

[a] Department of Otorhinolaryngology - Head and Neck Surgery, Faculty of Medicine, University of Szeged, Tisza Lajos körút 111, Szeged, 6725, Hungary
[b] Department of Pediatrics and Pediatric Health Center, Faculty of Medicine, University of Szeged, Korányi fasor 14-15, Szeged, 6720, Hungary
[c] Department of Otorhinolaryngology, Head and Neck Surgery, University Hospital of Lausanne, Rue du Bugnon 46, Lausanne, 1011, Switzerland

ARTICLE INFO

Keywords:
Congenital subglottic stenosis
Long-term follow-up
Open surgery
Slide laryngotracheoplasty

ABSTRACT

Objectives: Slide laryngotracheoplasty is an effective, single-step procedure without tracheostomy and stenting for treating high-grade congenital subglottic stenosis in neonates and infants. Long-term outcomes were evaluated to assess the reliability of the procedure performed in this age of rapid development of the laryngeal structures.
Methods: We report five children who underwent slide laryngotracheoplasty before the age of 4 months, each with >3 years follow-up. Increases of length and bodyweight were systematically assessed. Breathing, swallowing, voice, and overall satisfaction was assessed by a quality of life questionnaire. Voice quality was objectively evaluated by measuring shimmer, jitter, fundamental frequency, and the harmonics-to-noise ratio.
Results: All patients had a stable and adequate airway during follow-up without any additional open airway surgery. The patients' voices were physiological, and the intervention had no negative impact on speech development. Swallowing function was optimally retained, and the patients' bodyweight gain and length were satisfactory. During at least 3 years of observation, the anastomosis remained stable and grew dynamically with the patient.
Conclusions: Slide laryngotracheoplasty (as a single-step procedure) provides an adequate airway without tracheostomy, grafting, or stenting with good long-term functional results in selected neonates and infants with congenital subglottic stenosis.

1. Introduction

The adequate treatment of subglottic stenosis (SgS) is a major challenge of laryngology even nowadays. After laryngomalacia and vocal fold palsy, SgS is the third most common congenital laryngeal malformation. However, it is the laryngeal anomaly most commonly necessitating tracheostomy in newborns and infants [1,2]. Low-grade congenital SgS may improve with growth, but in severe cases, no spontaneous airway improvement is expected over time; thus, selecting the optimal surgical treatment strategy is paramount [3]. The ultimate goal of management is to ensure an adequate airway with the preservation of voicing and swallowing. Despite the well-defined endpoints (e.g. avoidance of a tracheotomy, definitive decannulation, socially acceptable voice quality, safe swallowing without aspiration), the surgical treatment of congenital SgS remains heterogeneous and depends almost as much on the experience of the airway team as on the child's condition [4]. Definitive airway surgery is usually preceded by tracheostomy despite its many well-known physical and psychosocial adverse effects [5–8]. In consideration of the special anatomy and vulnerable tissues of the pediatric airway, the potentially hidden comorbidities, and the associated congenital malformations, choosing the appropriate surgical method is crucial at this early age [9–12]. In cases of high-grade SgS, endoscopic procedures are not suggested [13–15]. Laryngotracheal reconstruction (LTR), cricotracheal resection (CTR), and extended cricotracheal resection have been used for decades, and they have proven to be favorable solutions with good long-term results. These complex open neck surgeries require major tissue resection, stent implantation, or airway expansion by rib cartilage grafts [16,17].

Our recent publication demonstrated, that slide laryngotracheoplasty (SLtp) is an effective single-step intervention, which does not require stenting for high-grade congenital SgS to widen the subglottic airway and avoid tracheotomy in neonates and infants [18].

* Corresponding author.
E-mail address: bach.adam@med.u-szeged.hu (Á. Bach).

https://doi.org/10.1016/j.ijporl.2022.111192
Received 19 October 2021; Received in revised form 22 May 2022; Accepted 24 May 2022
Available online 27 May 2022

Table 1
Pregnancy information and patient data.

Patient/ sex	Birth	Gestational age [week]	Apgar score	Weight at birth	Oxigen supplementation before surgery	Grade of stenosis	Feeding before surgery	Other symptomes	Surgery [day of life]
1/m	p.v. n.	37	8,9,9	1980g	oxigen hood	grade II	per os	–	10th
2/m	s.c.	40	7,9,9	3490g	intubation	grade III	NG tube	–	14th
3/f	p.v. n.	38	8,10,10	3450g	high flow oxigen	grade III	per os	RSV infection, tachycardia	68th
4/f	s.c.	37	7,8,9	2370g	oxigen hood	grade III	per os	Tetralogy of Fallot, laryngeal web, DiGeorge syndrome	105th
5/f	p.v. n.	36	_	3300g	intubation	grade III	NG tube	RSV infection, laryngitis subglottica	92nd

f = female; m = male; p.v.n. = per vias naturales; s.c. = sectio caesarea; NG tube: nasogastric feeding tube; RSV = respiratory syncytial virus.

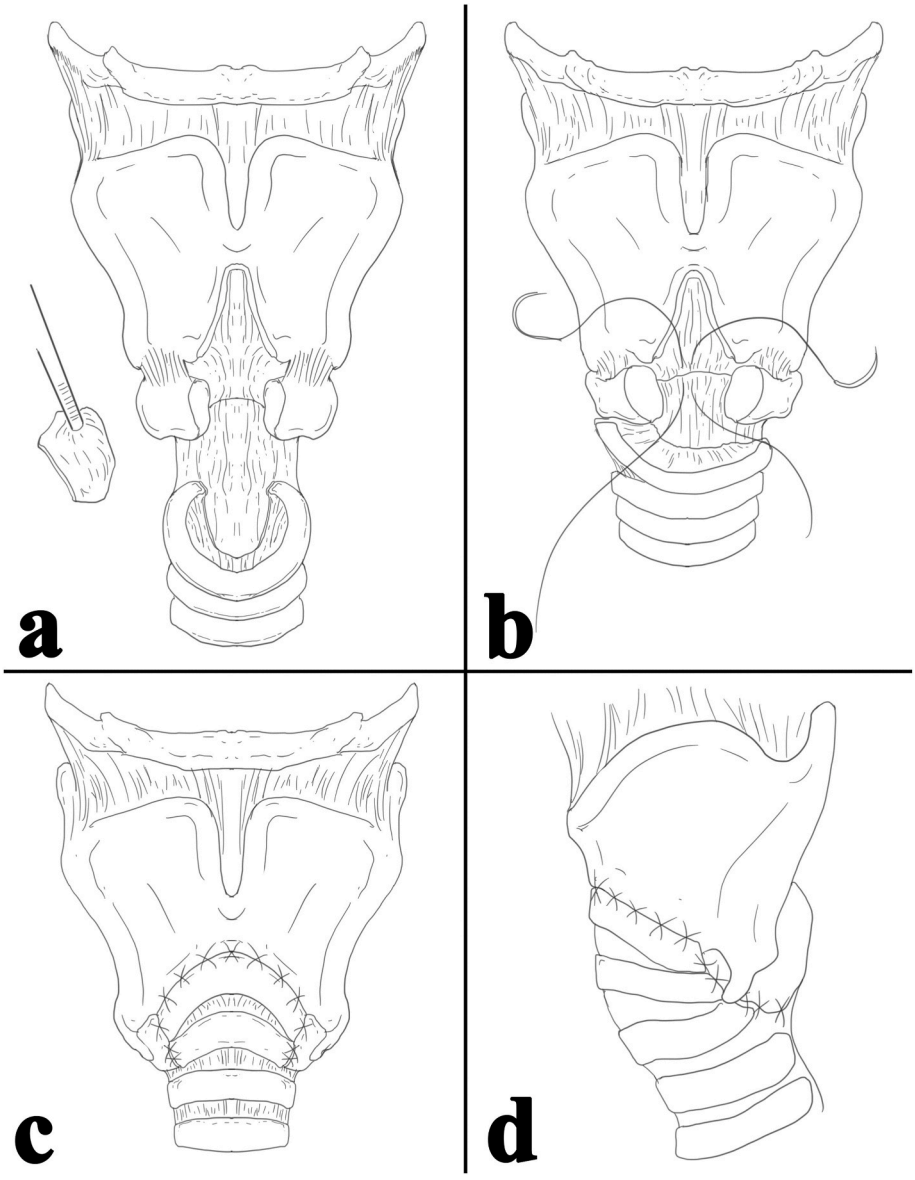

Fig. 1. Slide laryngotracheoplasty (schematic drawing). a) Partial midline anterior laryngofissure with preservation of the anterior commissure. The membranous wall of the trachea is sufficiently cropped. b) A tension-free anastomosis was created starting from the posterior wall. c–d) The reconstructed anterior and lateral wall.

Optimal swallowing function and voice production are sustained with this procedure. Because of the rapid development of infants, the long-term results and stability of the anastomosis in the fast-growing larynges at this young age are real concerns. With the primary outcome measure being a permanent solution of dyspnea and with secondary outcomes focusing on growth, voicing functions, and swallowing, we report five children with >3 years of follow-up after SLtp performed for congenital SgS.

2. Materials and methods

2.1. Patients

SLtp was performed in seven consecutive patients aged 10, 14, 68, 105, 92, 130, and 120 days old, respectively. In this study, the first five patients with more than 3 years of postoperative follow-up were included. Inspiratory dyspnea and stridor occurred immediately after birth in two cases (patients #1,2). These newborns had been admitted to the neonatal intensive care unit. Patients #3,4,5 became symptomatic after an upper airway infection. Patients #2 and #5 had been intubated, whereas the remaining patients required non-invasive oxygen support (oxygen mask in patients #1 and 4 and continuous positive airway pressure ventilation in patient #3), [Table 1].

Direct endoscopic examination verified Cotton–Myer grade III SgS in four (patients #2–5) and Cotton–Myer grade II SgS in one patient (patient #1) [5]. In this case the subglottic stenosis was cartilaginous, and the cricoid cartilage had an atypical shape. Therefore, we opted for external surgery. In patient #4, SgS was associated with Cohen grade III laryngeal web, as well as tetralogy of Fallot and DiGeorge syndrome.

2.2. Surgical technique

The patients underwent SLtp as described in our previous publication [18]. Therefore, only a summary is reported in this study. After exploration of the laryngotracheal complex, the cricotracheal junction was dissected circumferentially, and partial midline anterior laryngofissure was performed by dividing the cricoid and thyroid cartilage until the level of the anterior commissure. Thereafter, a posterior midline cricoid incision was made with preservation of the integrity of the posterior perichondrium, posterior cricoarytenoid, and pharyngeal constrictor muscles. The tracheal trunk was bluntly dissected until the anterior arch of the upper tracheal cartilages could be pulled up to the level of the anterior commissure tension-free. The posterior (membranous) wall of the trachea was sufficiently cropped to fit the posterior subglottic mucosa, and the airway was expanded with the interposed trachea flap via an anastomosis created between the anterior cricoid, midline-incised thyroid cartilage, and trachea [Fig. 1.a]. The suturing commenced at the posterior midline [Fig. 1.b]. Two double-armed continuous locked sutures were placed clockwise and counter-clockwise. After reconstruction of the posterior wall, the previously passed nasal endotracheal tube was descended into the trachea, and then the suturing of the lateral and the anterior wall was completed. Finally, the threads arriving from the opposite directions were knotted in the anterior midline with the knots lying outside the airway. Thus, a continuous suture ring was created [Fig. 1c,d].

In the patient with an associated laryngeal web, the membranous component of the web was vaporized using an UltraPulse CO_2 laser prior to the open neck procedure.

2.3. Postoperative care

All patients were assigned to the neonatal/pediatric intensive care unit. Intravenous antibiotics (amoxicillin/clavulanic acid 25 mg/5 mg/kg for 12 h or a different protocol depending on the bacteriologic aspirate) were administered for 7 days. On the day of extubation (days 3–10), a steroid bolus (methylprednisolone, 4 mg/kg) was administered.

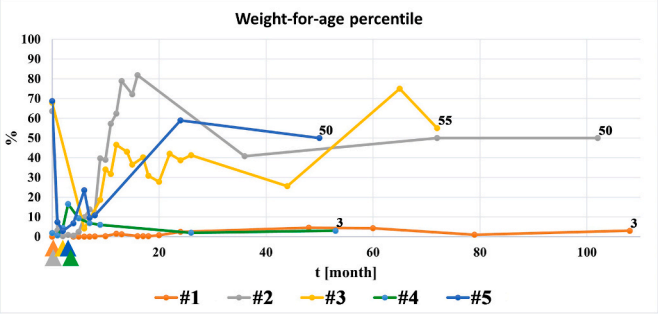

Fig. 2. Weight-for-age percentile of the patients. The date of the surgery is marked by triangles.

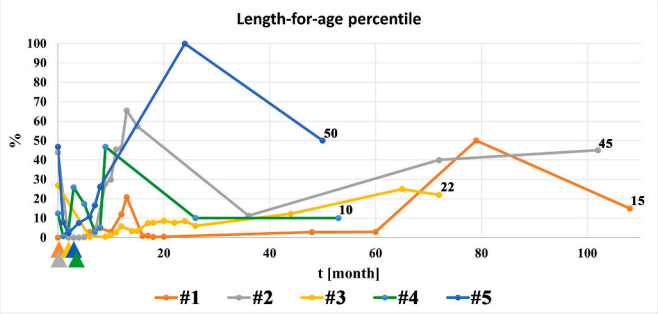

Fig. 3. Length-for-age percentile of the patients. The date of the surgery is marked by triangles.

The nasogastric feeding tube was removed a few days after extubation, and oral feeding started progressively.

2.4. Assessment of functional outcomes

The long-term functional results of the intervention were evaluated with the efficacious help of the parents. Length growth, bodyweight gain, and breathing or swallowing problems were systematically recorded. Breathing, swallowing, voice, and overall satisfaction was assessed using a quality of life (QOL) questionnaire [17]. The following items were rated by the parents by using the scales as indicated: dyspnea (grade 1 'absent' to grade 5 'at rest'); noisy breathing (grade 1 'absent' to grade 5 'very noisy breathing even at rest'); coughing (grade 1 'absent' to grade 4 'frequent episodes'); dysphonia (grade 1 'normal voice' to grade 4 'aphonia'); dysphagia (grade 1 'absent' to grade 3 'nasogastric tube feeding'); and global satisfaction (grade 1 'totally satisfied' to grade 4 'totally unsatisfied'). In all parameters lower grades meant better condition. Voice analysis was performed 36 months after SLtp according to our previously published protocol based on the guidelines elaborated by the Committee on Phoniatrics of the European Laryngological Society [19]. Shimmer (%), jitter (%), fundamental frequency, and the harmonics-to-noise ratio were analyzed using Praat 6.1.09 software [www.praat.org]. Endoscopic examinations under general anesthesia were strongly recommended in the first postoperative year. Thereafter, direct endoscopy was performed only if surgery-related airway symptoms occurred. The follow-up intervals for patients #1–5 were 108, 102, 70, 49, and 47 months, respectively.

Ethical approval for this study was obtained from the Institutional Ethics Committee of the University of Szeged (registration number 162/2019-SZTE).

3. Results

Repeated endoscopic airway surgery (UltraPulse CO_2 laser

Table 2
Postoperative care and functional results of the surgery.

Patient/sex	Removal of NG tube [postoperative day]	Shimmer [%]	Jitter [%]	HNR [dB]	Fundamental frequency [Hz]	Follow-up period [month]
1/m	9th	1.73	0.36	21.54	240.0.	108
2/m	7th	2.76	0.87	23.24	199.9	102
3/f	12th	1.25	0.25	26.78	189.8	70
4/f	10th	1.87	0.77	21.11	245.9	49
5/f	7th	2.29	0.3	24.45	320.4	47
Physiological values		<3.81	<1.04	>20		

f = female; m = male; NG tube: nasogastric feeding tube; HNR: harmonics-to-noise ratio.

Table 3
Subscores and total score of the quality of life questionnaire.

Patient/sex	Dyspnea	Noisy breathing	Coughing	Dysphonia	Dysphagia	Overall satisfaction	∑QOL
1/m	1	1	1	2	1	1	7
2/m	1	1	2	1	1	1	7
3/f	1	1	1	1	1	1	6
4/f	1	1	1	3	1	3	10
5/f	1	1	1	2	1	1	7
Range	1–5	1–5	1–4	1–4	1–3	1–4	6–25

f = female; m = male; QOL = quality of life.

vaporization) was required in patients #1 and #3 in the second postoperative month because of the excessive formation of granulation tissue. In the other patients, only planned control endoscopies were performed (two or three times in the first postoperative year). All children had stable and adequate airway during follow-up. Significant restenosis was not observed in any of the patients, moreover, the subglottic airway has become wider than the physiological one after SLtp. According to the parents' judgment, breathing was normal in all patients. The children did not require an exemption from physical education. After the removal of the NG tube, the patients retuned to oral feeding on a normal diet without the use of any thickener [Table 2]. Figs. 2 and 3 present the weight-for-age and length-for-age percentiles of the patients. Patient #4 experiences learning difficulties and delayed speech development. She can create all the vowels and some consonants (e.g. k, m, n, t). In parallel, her vocabulary is extremely low, she creates only short sentences and nursery rhymes. Her 'Wechsler Preschool and Primary Scale of Intelligence Fourth Edition' score is also low and the 'MacArthur-Bates Communicative Development Inventory' shows the skill level of a 2-year-old child, despite being 5 years old. The remaining children had a social life in line with their biological age. QOL scores and acoustic parameters are presented in Tables 2 and 3.

4. Discussion

Congenital SgS is attributed to the incomplete recanalization of the laryngeal lumen during gestation. It is frequently associated with other congenital head and neck lesions and syndromes (e.g., CHARGE syndrome, Down syndrome, 22q11 deletion) [20]. In a full-term neonate, SgS is defined as a lumen ≤4 mm in diameter at the level of the cricoid, whereas it is defined as a lumen <3 mm in diameter in preterm infants [21]. Airflow resistance is inversely proportional to the radius to the fourth power (r^4) [22]. Accordingly, a 50% reduction of the radius causes a 16-fold increase in airflow resistance. This significantly increased resistance imposes a huge physical burden on newborns and infants equally.

Generally, children with low-grade (grade I or grade II) SgS do not require surgical intervention. As affected children grow, spontaneous airway improvement can typically be expected [23]. Even mild grade III stenosis can often remain asymptomatic for weeks. However, infants with this airway status may experience recurrent airway symptoms, primarily when the infection is associated with mucosal edema [24]. Physiological development and growth are significantly impaired in cases of high-grade SgS; thus, a 'watch and wait' policy is not endorsed. When non-invasive positive pressure ventilation fails, urgent surgical intervention is necessary. For this purpose, tracheostomy is the most commonly performed primary intervention. As a life-saving intervention, the merits of tracheostomy are indisputable, however, it can negatively affect voicing, swallowing, and QOL [25]. Bacterial colonization and dynamic collapse of the trachea are also real threats [26–28]. A definitive surgical solution that can be performed in one-step could prevent many consequential problems such as accidental decannulation or airway obstruction, chronic airway infection, poor voice quality, negative effects on speech development, tracheomalacia, a need for multiple procedures, and high medical and nursing costs.

The number of surgical options has significantly increased over the past 50 years. Still, the proper management of SgS remains both challenging and complex [29]. Endoscopic, minimally invasive solutions (e.g., balloon dilatation, excision or incision of scar tissue with cold steel or CO_2 laser, intralesional or topical adjuvant therapy such as mitomycin C application or steroid injection) are optimal for patients with isolated low-grade (grade I–II) SgS without a history of previous treatment failure [30]. However, these techniques can also increase the incidence of urgent/unplanned airway interventions and potentially delay a definitive solution [31,32]. The chance of success with endoscopic, minimally invasive techniques decreases with the worsening of the initial grade of subglottic stenosis [13–15].

Patients with grade III–IV SgS or multilevel airway stenosis mostly require open surgical procedures [13]. Although these interventions carry a higher risk of morbidity, this is balanced by the fact that these treatments provide a definitive solution. In addition, we cannot ignore the fact that the first operation provides the best chance for a successful airway intervention [33]. Therefore, the avoidance of multiple, inappropriately selected, potentially futile (endoscopic) airway surgeries is essential. Laryngotracheal reconstruction and cricotracheal resection are the prevailing procedures globally [34–38]. In pediatric patients, CTR provides an overall decannulation rate exceeding 80% with a highly variable reoperation rate of 4%–41% (depending on the patient's comorbidities and grade of stenosis). Meanwhile, 22%–45% of patients require reoperation after LTR [10,17,36,38–43]. Significant manipulation of the laryngotracheal framework is inevitable using these techniques. However, by reducing the amount of resected tissue and minimizing potential graft problems, the success rate could be theoretically increased.

After SLtp, the reconstructed subglottic area was wider than the physiological one, even without extended resection or rib cartilage grafting. This 'reserve capacity' ensures a patent airway even in the

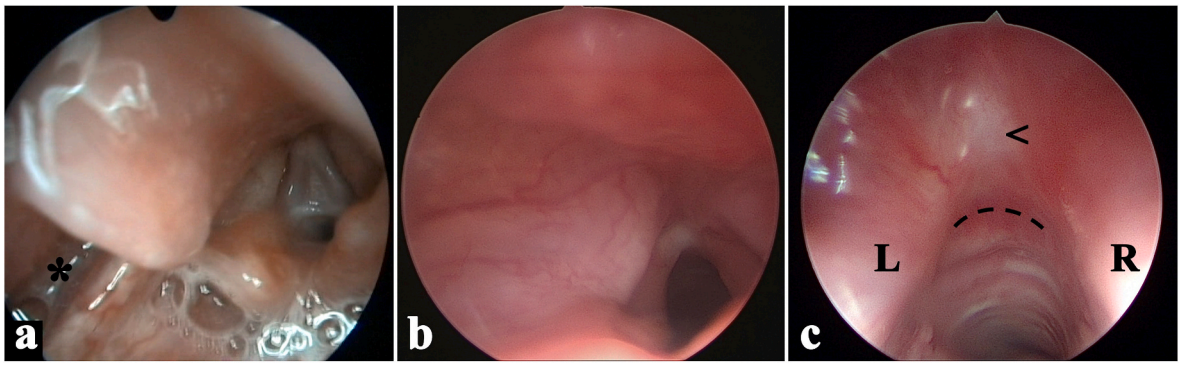

Fig. 4. Direct endoscopic pictures of patient #4. a) Preoperative picture: Cotton–Myer grade III subglottic stenosis and associated Cohen grade III laryngeal web; *: nasogastric feeding tube. b) Adequate glottic airway in the 45th postoperative month. Direct endoscopy is not obligatory in the late postoperative follow-up. Laryngotracheoscopy was performed prior to powered intracapsular tonsillotomy and adenoidectomy for obstructive sleep apnea. c) Wide subglottic without signs of restenosis.<, minimal mucosal scar in the anterior commissure; dashed line, upper rim of the interposited trachea flap, L, left vocal fold; R, right vocal fold.

event of mild restenosis. The cricothyroid complex together with the interposed trachea flap provides a well-vascularized stable ring. Sacrifice of the segmental tracheoesophageal arteries is necessary at the site of resection because of mobilization of the trachea and cropping of its membranous wall. However, the lateral longitudinal anastomoses and transverse intercartilaginous arteries can be preserved, potentially allowing a complication-free, quick recovery. Therefore, resorption of the local tracheal graft is modest, and the late postoperative result becomes easier to predict. In the absence of rib cartilage grafting, donor site complications can also be excluded. Furthermore, the trachea is covered by respiratory mucosa, which prevents granulations, subsequent restenosis, and adhesion of airway secretions through physiological mucociliary clearance. By reducing the extent of tissue resection and omitting the dissection of the cricothyroid muscle, the chance of recurrent laryngeal nerve injury is lower, which leads to optimal voicing and swallowing function. The children's voices were more than socially acceptable. As demonstrated in [Table 2], the assessed objective voice parameters were in the physiological ranges in all cases. The patients' bodyweight gain and length growth were satisfactory according to growth charts, and the parents were pleased with the children's postoperative QOL in general. One child (patient #4) with Di George syndrome has experienced learning difficulties and delayed speech development. During at least 3 years of observation, the anastomosis was stable and growing dynamically with the patient [Fig. 4].

If the general health status is appropriate and no critical comorbidity is present, SLtp is an addition to the surgical armamentarium for treating selected cases of congenital SgS. Adequate patient selection is crucial. The decision concerning whether to perform SLtp must be based on the overall health conditions of the infant in contrast to the severity of SgS itself. In children with certain craniofacial anomalies, neuromuscular disorder, high risk for aspiration, or low pulmonary function, decannulation may be counterproductive.

The drawbacks of this study include its small sample size and single-center nature. Therefore, further studies are required to explore the potentials and limitations of this novel surgical technique.

5. Conclusion

SLtp is an excellent surgical option for reconstruction in selected cases of SgS in a single-stage procedure without tracheostomy and stenting. Swallowing function and voice production are not disrupted by the procedure, and the newly formed laryngotracheal structure remains stable, ensuring the possibility of physiological development despite the severe congenital airway anomaly.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethics approval

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with this guideline. Ethical approval for this study was obtained from the Institutional Ethics Committee of the University of Szeged (registration number 162/2019-SZTE).

Written informed consent was obtained from the parents/legal representatives of the participants.

Consent for publication

Each author listed on the manuscript has seen and approved the submission of this version of the manuscript and takes full responsibility for the manuscript.

Declaration of competing interest

The authors have no conflicts of interest to disclose.

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