



Endoscopic anterior cricoid split and balloon dilation in pediatric subglottic stenosis

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ABSTRACT

Objectives: To analyze the outcome of a new endoscopic approach for the treatment of pediatric subglottic stenosis.

Study design: Case series.

Setting: Tertiary care center.

Material and methods: Eighteen pediatric cases of grade II to IV subglottic stenosis (8 congenital and 10 acquired) consecutively treated at our institutions by Endoscopic Anterior Cricoid Split (EACS) and balloon dilation between 2006 and 2010. Treatment protocol encompassed systematic postoperative laryngeal stenting (7 days of intubation or 1 month of Montgomery T-tube in previously tracheotomized patients) and endoscopic controls with possible additional balloon dilation every 15 days for at least 2 months.

Results: Patients' ages ranged from 1 to 101 months. Postoperative follow-up ranged from 4 to 45 months (median value \pm SD: 15.3 ± 11.9). The mean duration of the endoscopic procedure was 35.2 ± 13.2 min. The number of days spent in PICU during the perioperative period varied between 2 and 15. Four patients (22.2%) needed one and 14 patients (77.7%) required several (from 4 to 7) additional balloon dilations during the postoperative endoscopic controls. No incident was observed during or immediately after EACS. Treatment was efficient in 83% of cases ($n = 15$), with no residual respiratory symptoms and grade 0 to 1 SGS at the end of follow-up.

Conclusion: EACS is a safe and efficient technique to treat pediatric subglottic stenosis, regardless of their grade and length, provided to associate it with postoperative laryngeal stenting and regular endoscopic follow-up with possible additional balloon dilations. In our teams, it has become the first line treatment for most grades II to IV SGS. Its indications can be extended to congenital stenosis with cartilaginous involvement and to long-lasting acquired stenosis with firm fibrosis.

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1. Introduction

Since the 1980s, the classical management of symptomatic SGS consists of controlling associated risk factors and performing laryngotracheal surgery via an external approach. Three main surgical techniques have been proposed: cricoid split, single- or two-stage laryngotracheal reconstruction, and partial cricotracheal resection. These open airway procedures represented a great improvement in the management of pediatric laryngeal stenosis. They proved to be effective in about 90% of cases, limiting the need for long-term tracheotomy and its associated morbidities such as cannula obstruction or inadvertent decannulation. Some of them, such as cricoid split [1] or partial cricotracheal resection [2], can be

successfully proposed during infancy. However, they often require several surgical procedures, prolonged intensive care admission, prolonged intubation, or tracheotomy, and they have the potential for serious complications [2–5]. Their vocal outcome is often not very satisfactory [6] and they create unappealing cervical scars. In order to reduce or to avoid some of these drawbacks, several techniques of endoscopic treatments have been developed during recent years. In the present study, we present a series of 18 cases of congenital or acquired SGS treated with anterior cricoid split (EACS) associated with balloon dilation(s) and postoperative laryngeal stenting.

2. Material and methods

2.1. Population

Eighteen pediatric cases of SGS (8 congenital and 10 acquired) consecutively treated at our institution by EACS and balloon

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Table 1
Patients' clinical characteristics and endoscopic findings.

Patient number	Body weight (kg)	Age (months)	Type of stenosis	SGS grade ^a	Associated laryngeal anomalies	GERD ^b (type of treatment)	Airway stenting before surgery
1	1.8	9	Acquired	III	None	Yes (PPI)	Tracheostomy
2	4.3	6	Congenital	II	None	Yes (PPI)	Endotracheal tube
3	14.0	20	Congenital	III	None	Yes (PPI)	Tracheostomy
4	8.0	9	Acquired	II	Subglottic hemangioma	Yes (Nissen)	Tracheostomy
5	4.1	8	Congenital	III	None	Yes (Nissen)	Endotracheal tube
6	12.0	20	Acquired	III	None	No	None
7	5.4	8	Acquired	II	None	No	None
8	8.1	6	Acquired	IV	None	Yes (PPI)	Tracheostomy
9	4.0	1	Acquired	IV	None	Yes (Nissen)	Tracheostomy
10	22.0	101	Acquired	III	None	No	None
11	8.1	8	Congenital	II	None	No	None
12	7	4	Congenital	III	None	No	Tracheostomy
13	12	27	Acquired	III	Ectopic gastric and pancreatic tissue in the left aryepiglottic fold and false vocal fold	Yes (Nissen)	Tracheostomy
14	7	6	Congenital	III	None	Yes (PPI)	Tracheostomy
15	8	32	Congenital	III	None	Yes (Nissen)	Tracheostomy
16	4	5.3	Acquired	II	None	No	None
17	16	48	Acquired	II	Anterior glottis web	Yes (PPI)	None
18	5	6	Congenital	II	Anterior glottis web	Yes (Nissen)	None

No patient had any previous open air laryngotracheal surgery before undergoing EACS.

^a Grading according to Cotton and Myer's classification.

^b GERD: gastro-esophageal reflux disease; PPI: proton pump inhibitors.

dilation between 2006 and 2009 were analyzed. Patients' clinical characteristics and endoscopic findings are detailed in Table 1. Based on endoscopic assessment, stenosis were graded according to the Cotton–Myer classification [7].

2.2. Inclusion criteria

We performed EACS only if the following conditions were present:

- Grade II, grade III or grade IV SGS;
- No sign of systemic infection;
- Satisfactory pulmonary function;
- GERD absent or correctly controlled by specific medical or surgical treatment;
- Absence of associated neurological pathology or swallowing disorder;
- No history of failed surgical attempts.

2.3. Preoperative assessment

Preoperative evaluation always began with UNA VALUTAZIONE CLINICA FINALIZZATA ALLA VALUTAZIONE DELLA QUALITÀ DELLA VOCE PRIMA DELL'INTERVENTO, CUI SEGUIVA UNA VALUTAZIONE ENDOSCOPICA DELLA FUNZIONALITÀ LARINGEA. Dynamic assessment was performed with a fiberoptic endoscope (Karl Storz GmbH & Co. KG, Tuttlingen, Germany) under local anesthesia. Then, a complete airway assessment was done under general anesthesia using rigid telescopes and bronchoscopes (KARL STORZ). The diagnosis of congenital stenosis was based on the absence of risk factors for acquired stenosis and of severe laryngeal inflammation. When the stenosis was too narrow to go through it with an endoscope, a CT-imaging of the airway was done ($n = 2$; patients 8 and 9). In case of laryngeal or esophageal inflammation, a 24ph-esophageal probe was performed. If it was positive, medical therapy with proton pump inhibitors was prescribed for at least 1 month. If the latter treatment failed, patients underwent Nissen fundoplication before EACS.

2.4. Description of the procedure and postoperative management

EACS was performed under general anesthesia obtained by delivering inhalation anesthetics through a nasopharyngeal tube ($n = 9$) or through a tracheotomy cannula if the child was tracheostomized before the procedure ($n = 9$). Patients with a nasopharyngeal tube breathed spontaneously, and the airflow delivered through it equaled three to four times the tidal volume. This anesthetic technique permitted the surgical field to be free of any ventilation material. After spraying the vocal folds with 2% Lidocaine (3–5 mg/kg), the subglottic region was exposed by suspension microlaryngoscopy. Visualization was improved by retracting the false vocal folds with a Lindholm laryngeal distending forceps (#8654B, KARL STORZ GmbH & Co., Tuttlingen, Germany). A vertical midline anterior incision was made with a cold knife (#8655, KARL STORZ) through the cricoid anterior ring. In case of long or high grade stenosis, this incision was extended superiorly to the infraglottic of the thyroid cartilage, and inferiorly AI PRIMI 2 ANELLI TRACHEALI tracheal rings. Both the cartilage and its external perichondrium were incised (Fig. 1), so that the incision margins could be laterally displaced a few millimeters away from each other in a stable way by balloon dilatation. This dilatation was performed using an angioplasty balloon (CORDIS CORPORATION, Bridgewater, NJ, USA, or Boston Scientific – France, Nanterre, France) (Fig. 2). The diameter of the inflated balloon was approximately 2 mm larger than the normal size of the subglottic lumen at the age of the patient. The laryngeal dilatation was maintained until patient's oxygen saturation dropped under 90%. At the end of the procedure, in non-tracheotomized patients, a nasotracheal intubation was performed using a tube with a half-size larger diameter than the one usually used at the age of the patient. Patients were kept under sedation and mechanical ventilation until extubation at day 7. In previously tracheotomized children, the tracheotomy cannula was replaced by a Montgomery T-tube in order to stent the subglottic lumen. The T-tube was removed at day 30, except in two preterm patients (patients 1 and 13) in which it was maintained during 60 days. Post-operative endoscopic controls were systematically performed at the time of extubation or at the time of Montgomery tube removal, then once every 2 weeks during 2 months. Additional endoscopies were

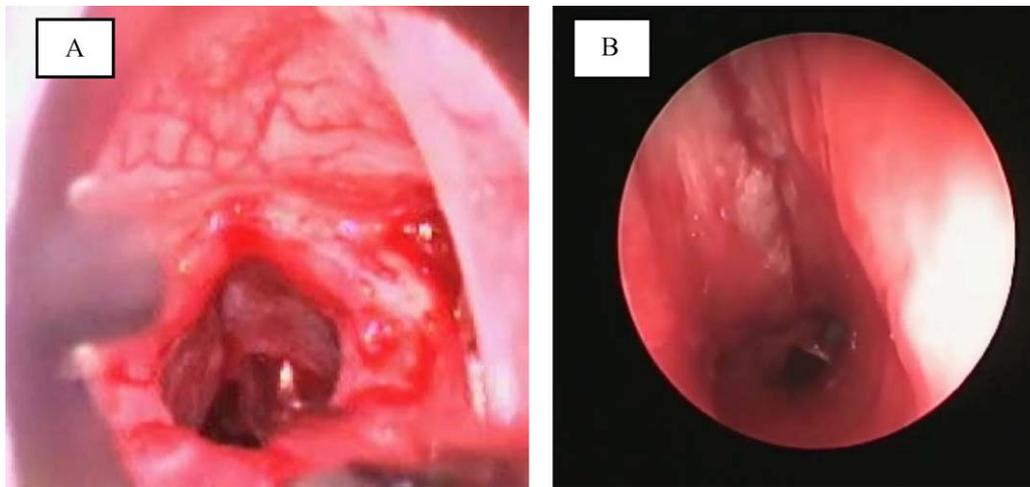


Fig. 1. Endoscopic view of the section of the subglottic lumen during the anterior midline section of the laryngeal cartilages (A) and just after this section (B).

performed as a function of patients' clinical evolution. During these endoscopic sessions, balloon dilatations were performed in case of persistent narrowing of the subglottic lumen.

In the present study, successful treatment was defined as an open stable adequate airway usually consisting of a grade 0 or 1 SGS, without clinical respiratory compromise.

3. Results

Patients' follow-up ranged from 4 to 45 months (median value \pm SD: 15.3 ± 11.9). Their clinical characteristics and endoscopic findings are detailed in Table 1. Their age ranged from 1 to 101 months (median value \pm SD: 18.1 ± 23.9) and their body weight from 1.8 to 22 kg (median value \pm SD: 8.37 ± 5.1). The 10 acquired cases resulted from intubation trauma. Gastroesophageal reflux was detected by 24h-pH probe in 12 patients and 6 of them had severe reflux resistant to medical treatment necessitating a Nissen fundoplication. At the time of EACS, nine patients were tracheotomized. In four cases, there were some associated laryngeal comorbidities (Table 1). Patient 4 had a subglottic hemangioma previously removed by endoscopic diode laser resection (CERALAS Bare fiber BFF, CERAMOPTEC GmbH, Bonn, Germany). Iatrogenic symptomatic grade II SGS developed within a few months after this resection. Patient 13 initially presented with a mass implanted in the left aryepiglottic fold, false vocal cord and hypopharynx and which consisted of ectopic gastric and pancreatic tissue. This benign tumor was endoscopically removed with diode laser. During the postoperative period, both a SGS and a short (about 5 mm in length) but straight upper esophageal stenosis were observed. The SGS was treated with

EACS and balloon dilation whereas the upper esophageal stenosis required surgical resection of a 25 mm long segment of upper esophagus. Patients 17 and 18 had both a SGS and an anterior glottic web. The latter was treated during the EASC procedure with cold instruments.

The duration of the EACS procedure and characteristics of the postoperative management are detailed in Table 2. The mean duration of the endoscopic procedure was 35.2 ± 13.2 min. No incident was observed during or immediately after EACS. In 83.3% of patients (15/18), the endotracheal or the Montgomery tubes were removed according to the planned protocol. The number of days spent in PICU varied between 2 and 15 (median value \pm SD: 7.72 ± 4.19).

During the postoperative endoscopic procedures, four patients (22.2%) needed one and 14 patients (77.7%) required several balloon dilations (from 4 to 7) (Table 3). In two patients, topical applications of mitomycin-C were performed according to the protocol described in 2001 by Hartnick et al. [8] (mitomycin concentration: 0.4 mg/ml; application duration: 2 min).



Fig. 2. Balloon dilation of the subglottic stenosis.

Table 2
Duration of the EACS procedure and postoperative management.

Patient number	Procedure duration (min)	Duration of hospitalization in PICU ^a /total (days)	Duration of postoperative intubation or tracheostomy (days)	Number of laryngeal endoscopic dilatations during the postoperative endoscopic controls
1	35	2/9	60	1
2	27	10/14	7	6
3	20	2/7	30	4
4	40	2/7	30	1
5	22	8/13	7	7
6	20	9/13	7	4
7	25	8/11	7	4
8	38	14/20	7	4
9	35	15/25	7	4
10	55	9/15	7	4
11	38	7/12	7	0
12	25	7/10	7	0
13	55	3/15	60	2
14	65	15/20	12	3
15	45	3/10	30	2
16	20	8/10	7	1
17	30	9/12	7	1
18	40	8/10	7	3

^a PICU: pediatric intensive care unit.

Table 3
Clinical outcome.

Patient number	Follow-up duration (months)	Respiratory outcome ^a	Voice quality ^b	General clinical conditions
1	39	Good	Normal	Good
2	27	Good	Normal	Good
3	42	Good	Normal	Good
4	24	Obstructive supraglottic granulomas impeding decannulation	Hoarse	Patient deceased 18 months after EACS
5	22	Residual stenosis requiring reconstruction laryngotracheoplasty Patient still tracheotomized	Soft Voice	Good
6	14	Good	Normal	Good
7	13	Good	Normal	Good
8	10	Good	Hoarse	Good
9	10	Good	Hoarse	Good
10	7	Good	Normal	Good
11	12	Residual stenosis requiring reconstruction laryngotracheoplasty Patient still tracheotomized	Hoarse	GERD
12	2.5	Good	Normal	Good
13	17	Good	Normal	Good
14	4	Good	Normal	Good
15	13	Good	Normal	Good
16	5	Good	Normal	Good
17	7	Good	Normal	Good
18	4	Good	Normal	Good

^a Definition of good respiratory outcome: no dyspnea, and no residual laryngeal stenosis during the last endoscopic control.

^b The voice was assessed clinically and classified as normal, soft or hoarse. In patient 4, the voice quality was tested after brief obstruction of the tracheotomy cannula.

Three EACS failures (17%) were observed (patients 4, 5, and 11). In patient 4, the supraglottic region was obstructed by granulomas in the area of contact between the laryngeal mucosa and the upper extremity of the Montgomery T-tube. These granulomas were probably caused by the associated GERD, and maybe also by the laryngeal inflammation induced by the previous laser resection of a subglottic hemangioma. This child was found dead by her parents at home in the morning 18 months after the EACS procedure. The cause of decease remains unknown. The possibility of a cannula plugging cannot be discarded.

Patient 5 was not tracheotomized prior to EACS and had no associated laryngeal comorbidities. He had a severe GERD treated by Nissen fundoplication. This treatment failed with persistent hypotonic inferior esophageal sphincter and GERD after surgery. This might have contributed to EACS failure.

In patient 11, failure was probably due to the fact that after the EACS procedure, he was followed in another pediatric center and was not planned for any postoperative endoscopic control. In this patient, the SGS eventually became more severe than before the EACS, requiring an open surgical procedure (Table 3).

4. Discussion

As stated in the introduction, the classical treatment of SGS is open surgery. Endoscopic techniques, in comparison, offer fewer operative risks, a shorter hospital stay, and possibly less emotional and financial burden on the family [9,10].

The two main endoscopic techniques described so far are laser resection and laryngeal dilation. According to Monnier et al. [9], laser resection should be reserved for grade I to mild grade III SGS with a cranio-caudal extension of less than 15 mm and in the absence of loss of cartilage support (Table 4). It should not be repeated in case of recurrence to the initial grade of stenosis. Triglia et al. [11] advised laser resection only for grade I stenosis less than 5 mm in length.

Laryngeal dilation can be performed with rigid dilators or with inflatable balloons. In a recent Brazilian study including 64 cases, dilatation with rigid Chevalier Jackson dilators was used in grades I to III stenosis with a success yield of 100% [12]. However, repeated procedures were necessary, especially for high grade stenosis. For instance, the mean number of dilations required to treat grade III

stenosis was 12 (Table 4). As compared to the use of rigid dilators, the main advantages of balloon dilatation are the absence of longitudinal shearing forces, and the possibility to control both the balloon diameter and the dilation pressure. In a small series of 10 SGS treated with this technique, the success rate was 100% for the two grade II stenosis and 62.5% for the eight grade III stenosis [13] (Table 4). According to the authors, balloon laryngoplasty should be avoided in cases in which the SGS is long standing, congenital or has cartilaginous involvement.

The benefit of combining several endoscopic techniques has been described previously. Thus, in a series of 29 pediatric cases of SGS, Bakthavachalam and McClay used various combinations of rigid manual dilation, carbon dioxide laser and microdebridement [10]. Their percentage of success ranged from 100% (3/3) for grade I stenosis to 76% (9/17) for grade III stenosis. However, the authors state in their discussion that these good results might be attributable to the fact that they mainly reserved endoscopic treatment for low grade, short-segment (less than 1.5 cm) stenosis as well as thin, membranous, granular and spiral ones.

As compared to all previously described endoscopic procedures, the new technique of EACS has several potential advantages:

- By utilizing a full depth incision of the subglottic laryngeal cartilages and first tracheal rings, the indications can be extended to congenital stenosis with cartilaginous involvement and to long-lasting acquired stenosis with firm fibrosis;
- The anterior opening of the larynx and of the first tracheal rings also enables to obtain major enlargement of the subglottic lumen, so that EACS can be used in high-grade stenosis (Fig. 3);
- As compared to laser resection, the use of cold instruments precludes thermal damage.

The comparison between the clinical outcomes obtained with EACS and with other endoscopic techniques suggests that EACS is as safe and probably more efficient in high-grade and in congenital forms of SGS (Table 4). Its efficacy is probably not far from that obtained with open procedures such as anterior cricoid split or laryngotracheal reconstruction with cartilaginous grafts [14–19].

The major drawback of EACS is the necessity of post-operative intubation or 1-month indwelling with a Montgomery T-tube, and

Table 4
Outcome of endoscopic treatment of pediatric subglottic stenosis. Comparison between literature data and the present series.

Reference	Technique	Number of patients	Mean age (range)	Stenosis severity	Type of stenosis	Mean follow-up	Mean number of procedure	Restenosing Requiring tracheotomy or open surgery	Complications
Triglia et al. [11]	Laser (CO ₂ , KTP)	25	6 years	I: 80% II: 4% III: 8% IV: 8%	Not specified	Not specified	1.5	4%	Not specified
Monnier et al. [9] ^a	Radial CO ₂ laser incision with or without dilatation with Savary-Gilliard dilators	100	22 years (6 months–73 years)	I: 15% II: 44% III: 41%	Acquired 85% Congenital 7% Mixed 8%	18 months	48%: n = 1 47% n = 2 5% n = 3–5	13% of patients still tracheotomized at the end of the study	0
Maksoud-Filho et al. [12]	Dilatation with Chevalier Jackson Dilators	64	18.9 months (1 month–7 years)	I: 70% II: 19% III: 11%	Acquired SGS	Not specified	From 6.3 (grade I) to 11.9 (grade III)	0	0
Bakthavachalam and McClay [10]	Manual rigid dilation; CO ₂ laser: microdebridement	29	26.6 months (0–168 months)	I: 10% II: 31% III: 59%	Not specified	Not specified	From 1.3 (grade I) to 3.5 (grade III)	12% grade II 24% grade III	Not specified
Durden and Sobol [13]	Balloon dilatation	10	4.8 months (2–12)	II: 20% III: 80%	Acquired SGS	3.5 months	1.4	30%	0
Present series	Endoscopic Anterior Cricoid SPLIT	11	17.8 months (1–101 months)	II: 36.4% III: 45.4% IV: 18.2%	Acquired and congenital SGS	16.4 months	3.9	18.2%	0

^a This series encompassed 45% of adult patients.

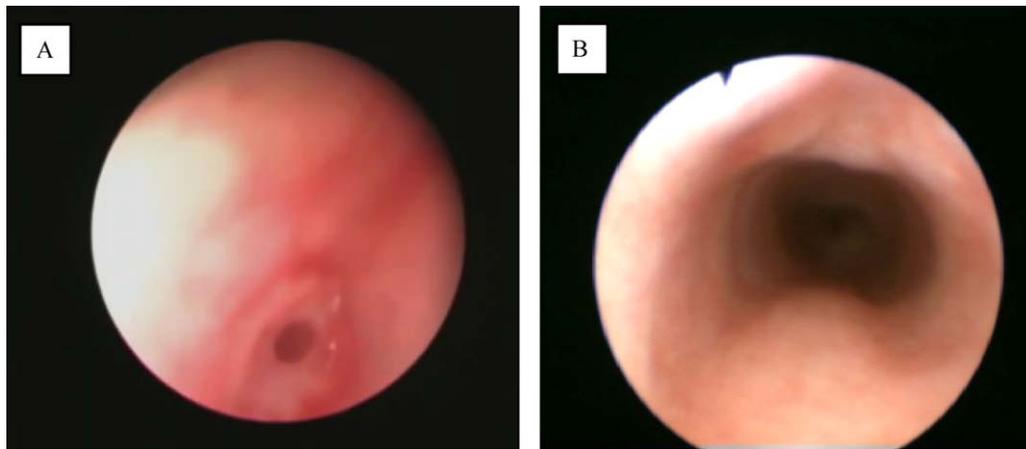


Fig. 3. Endoscopic view of the laryngeal lumen before (A) and three months (B) after EACS in patient 6.

of several planned postoperative endoscopic controls with balloon dilations. Thus, in short, low-grade and mainly membranous stenosis, a more straight forward approach such as simple balloon dilations or laser treatment, might be preferable.

5. Conclusion

This study suggests that EACS is probably a safe and efficient treatment of SGS. In our team, it has become the first line treatment for most grades II to IV SGS, given that the glottic and supraglottic structures are free of any associated morphological or dynamic anomaly, and that there is no uncontrolled GERD-induced inflammation of the larynx. Our remaining indications for open air procedures are the following cases: (1) laryngeal stenosis that concern both the glottic and subglottic levels; (2) failures of EACS; (3) impossibility to correctly expose the larynx under suspension laryngoscopy. Further prospective studies encompassing higher numbers of patients are required to confirm these results and to better specify the indications of this technique.

Conflict of interest

None.

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