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ORIGINAL ARTICLE

Post-intubation Subglottic Stenosis in Children. Diagnosis, Treatment and Prevention of Moderate and Severe Stenosis☆

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KEYWORDS
Subglottic stenosis;
Intubation;
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Abstract
Introduction and objectives: Subglottic stenosis is one of the most common causes of upper airway obstruction. Almost 90% of them result from endotracheal intubation. Therapy depends on the degree of stenosis, among other factors. Therapeutic approaches range from watchful waiting, in mild stenosis, to complex surgery for severe cases. We report our experience on the surgical management of post-intubation subglottic stenosis in children, emphasising the need for recognition and prevention of predisposing factors of post-intubation stenosis.
Methods: We retrospectively evaluated 71 patients with moderate to severe post-intubation subglottic stenosis, operated in the Respiratory Endoscopy Service in a period of eight years. The clinical variables analysed were age at surgery, degree of stenosis, surgical technique, complications and outcome.
Results: In 84.5% of patients, only 1 surgical approach was required to achieve decannulation. Three surgical techniques were implemented as therapy: laryngotracheal reconstruction, partial cricotracheal resection and anterior cricoid split. Decannulation was achieved in 70 cases. In 71.8%, ventilation, swallowing and voice qualities were good; 23.9% presented dysphonia; and 2.8% presented a mild respiratory distress. One patient died.
Conclusion: In patients with subglottic stenosis, selection of the most accurate treatment is the key to success, reducing the number of surgeries and preventing complications.

PALABRAS CLAVE
Estenosis subglótica;
Intubación;
Niños;

Estenosis subglótica postintubación en niños. Diagnóstico, tratamiento y prevención de las estenosis moderadas y severas

Resumen
Introducción y objetivos: La estenosis subglótica es una de las causas más frecuentes de obstrucción laríngea. El 90% resultan de la intubación endotraqueal. La conducta terapéutica

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Tratamiento quirúrgico;
Prevención

dependerá entre otros factores del grado de estenosis que se presente. Variará desde la conducta expectante en los grados leves hasta la cirugía compleja en los graves. Presentamos nuestra experiencia en el manejo quirúrgico de la estenosis subglótica postintubación en niños, y enfatizamos la necesidad de reconocimiento y prevención de los factores predisponentes de la estenosis por intubación.
Método: Se evaluaron en forma retrospectiva 71 pacientes con estenosis subglótica moderada a severa postintubación, operados en el Servicio de Endoscopia Respiratoria durante un periodo de 8 años. Las variables analizadas fueron la edad al momento quirúrgico, el grado de la estenosis, la técnica quirúrgica utilizada, las complicaciones y los resultados.
Resultados: El 84,5% de los pacientes requirió un solo tratamiento quirúrgico para lograr la decanulación. Se implementaron 3 técnicas quirúrgicas: reconstrucción laringotraqueal, resección cricotraqueal parcial y división anterior del cricoides. Se logró la decanulación en 70 casos, presentando buena ventilación, deglución y voz el 71,8%, disfonía el 23,9% y dificultad respiratoria leve el 2,8%. Un paciente falleció.
Conclusión: Frente a un paciente con estenosis subglótica, seleccionar el tratamiento apropiado es la llave del éxito, disminuye el número de cirugías y previene las complicaciones.

Introduction

Subglottic stenosis (SGS) is a decrease in the width of the subglottis. The principal symptoms are respiratory distress, biphasic stridor and chest retraction of variable seriousness. It can be accompanied by weak crying or dysphonia. Although it can be congenital, 90% of the SGS cases are acquired, with intubation and mechanical respiratory assistance being the main cause.[1–8] This condition should be considered for any child that presents respiratory distress and has a history of intubation, instrumentation or airway trauma.

The incidence of post-intubation stenosis ranges from 0.9% to 3%.[1] The first sign that stenosis is developing can be extubation failure, the need to reintubate with a smaller endotracheal tube or persistent dysphonia after extubation.

Management of severe SGS in children is complex. A wide variety of surgical techniques have been developed. Treatment for each patient should be individualised and various endoscopic or surgical techniques are often required to obtain successful results.[9]

We present a review of the results obtained in patients with post-intubation SGS operated on with techniques of expansion and of airway resection. In addition, we emphasise the need to recognise and prevent the factors that predispose to intubation stenosis.

Method

We carried out a retrospective assessment on 71 patients with moderate to severe post-intubation SGS, operated on in the Respiratory Endoscopy Service during an 8-year period (December 2002–December 2010).

We considered as moderate to severe SGS those cases that were grade II-b–IV-d according to the Myer-Cotton classification system modified by Monnier.[1]

The variables analysed were age at time of surgery, degree of stenosis, surgical technique used, complication and outcome.

The patients received an in-depth assessment prior to surgery, with images and endoscopies using local anaesthesia (with flexible fibre optics) to evaluate the status of the supraglottic structures (movement of vocal folds and arytenoids), and using general anaesthesia (with rigid instruments) to evaluate the characteristics of the lesion (grade, type, site, and extension of the stenosis).

In the postoperative period, patients were advised antireflux treatment (lansoprazole), antibiotics (amoxicillin), analgesics (ibuprofen) and feeding by nasogastric tube until oral tolerance was tested. In patients operated on that did not receive a prosthesis, periodic endoscopies were performed, from 2 to 4 in a 2-month period, to evaluate the site of the graft, size of the airway and formation of granulation tissue.

Results

There were 40 men and 31 women with post-intubation SGS. The period of intubation ranged from 4 to 150 days (median: 30 days). There were 5 patients that also presented congenital SGS.

According to the Cotton-Myer classification modified by Monnier[1] (Fig. 1), 1 patient presented grade II (1b) SGS, 57 patients showed grade III (13a, 41b, and 3d) and the remaining 13 patients were grade IV (1a, 7b, 1c, and 4d).

Comorbidities were present in 56 patients (78.9%). The most frequent associated conditions were gastroesophageal reflux (18 cases), bronchopulmonary dysplasia (10 cases), cardiopathy (7 cases) and genetic syndromes (5 cases).

At the time of surgery, 62 patients (87.3%) were tracheotomized. In 35 patients endoscopic procedures were performed (dilatations, CO_2 laser) before the open surgery. Mean age at the time of surgery was 4.5 years, with the median being 1.8 years (range: 1 month–17 years). Three surgical techniques were utilised (Table 1):

- Laryngotracheal reconstruction (LTR): 55 patients (77.5%) (Fig. 2).
- Partial cricotracheal resection (CTR): 10 patients (14.1%).
- Anterior cricoid split: 6 patients (8.4%).

Average hospital stay was 11 days, which was prolonged in patients that had complications.

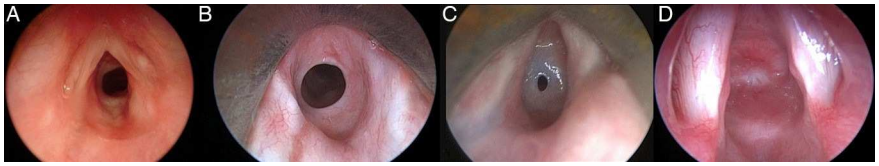

Figure 1 Cotton-Myer classification. (A) Grade I: obstruction of up to 50% of the laryngeal lumen. (B) Grade II: up to 70%. (C) Grade III: up to 99%. (D) Grade IV: without perceptible lumen. Monnier adds to the percentage of obstruction: (a) if the stenosis is isolated, (b) if it is associated with comorbidities, (c) if the glottis is involved, or (d) both.

The Montgomery T-tube (MTT) was used in 59 patients, 1 patient received a short-term suprastomal stent (mould) and an endotracheal tube (ETT) was used in 11 patients. As for time of prosthesis permanence, the ETT remained in place for 7 to 10 days, the mould for 3 months and the MTT for 12 months in 39 patients (66.1%), for 6 to 8 months in 8 patients (13.6%) (8 cases: LTR with graft), and for 16 months in the remaining 12 patients (20.3%) (5 cases of LTR with graft, 6 cases of LTR with lateral release and 1 case of LTR with posterior cricoid split).

Oral feeding was started in the patient with the mould at 48 h; in the patients with ETT at 24 h or 48 h after extubation; and in the patients with MTT, solids were given on the fifth day in 62.7% of the cases and liquids between 7 and 10 days in 71.8%.

Complications were presented in 32 patients (45.1%). The most frequent was granuloma of the upper free border of the MTT (16 cases). Other complications were as follows: accidental removal of the MTT (2 cases), cervical fistula (6 cases) and dehiscence of part of the suture of the graft to the laryngotracheal lumen (1 case). Two patients presented complications secondary to the extraction of the costal graft: costal haematoma and pleural effusion. Among the complications associated with the admission to therapy, we observed transitory quadriparesis associated with the drugs (1 case), reintubations from accidental extubation (2 cases) and failures in extubation (3 times, 2 cases).

Only a single surgical treatment to achieve decannulation was needed in 84.5% of the patients (60 cases). However, 21.6% of them (13 cases) required posterior endoscopic treatment. Eleven patients (15.5%) required a second surgical treatment and 2 patients (2.8%), a third (Table 2).

Post-treatment follow-up time was 2 years. Decannulation was achieved in 70 cases, with 71.8% (51 cases) presenting good ventilation, swallowing and phonation; 23.9% (17 cases) presenting dysphonia and 2.8% (2 cases), slight respiratory distress. One patient died a month after surgery (anterior cricoid split) from a non-related cause (Table 3).

Table 1 Surgical Technique Used.

LTR with MTT	49p
Anterior cartilage graft	
Concha	1p
Costal	2p
Thyroid alar	18p
Posterior cartilage graft	
Costal	3p
Thyroid alar	1p
Lateral release	20p
Posterior split	4p
LTR with ETT	5p
Anterior cartilage graft	
Costal	3p
Thyroid alar	2p
LTR with mould	1p
Anterior costal cartilage graft+posterior split	1p
CTR with MTT	10p
Anterior division of the cricoid	6p

CTR: partial cricotracheal resection; ETT: endotracheal tube; LTR: laryngotracheal reconstruction; MTT: Montgomery T-tube; p: patients.

Discussion

Laryngeal stenosis is one of the most frequent causes of airway obstruction in children. Likewise, it is one of the most common indications for tracheotomy in babies less than a year old. The main cause is intubation.

Factors that predispose to post-intubation stenosis can be related to the patients themselves (congenital narrowing of the airway, prematurity, gastroesophageal reflux, keloid formation and systemic factors causing hypoperfusion of the mucosa, such as hypotension, anaemia, sepsis, and shock).

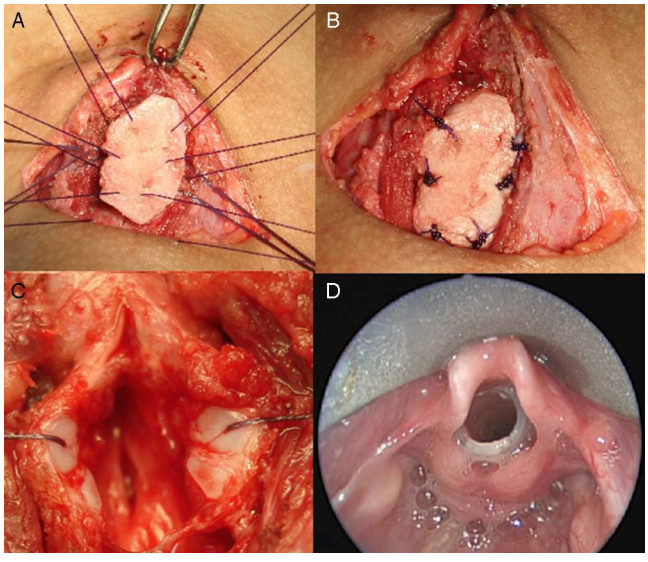

Figure 2 Intraoperative images. (A) Placement of the costal graft. (B) Anterior costal graft in position. (C) Posterior cricoid split. (D) T-tube in position.

Table 2 Post-treatment Laryngeal Lumen and Need for Second and Third Surgical Treatment.

Surgical technique	Laryngeal lumen after surgery			Second surgery	Third surgery
	Normal	SGS 1	SGS 2		
LTR with MTT					
Graft	22p (88%)	3p (12%)	0	3p (12%)	2p (8%)
Lateral release	17p (85%)	2p (10%)	1p (5%)	1p (5%)	0
Posterior split	2p (50%)	2p (50%)	0	1p (25%)	0
LTR with ETT	2p (40%)	3p (60%)	0	3p (60%)	0
LTR with mould	0	1p (100%)	0	0	0
CTR with MTT	7p (70%)	3p (30%)	0	1p (10%)	0
Anterior cricoid split	4p (66%)	2p (33%)	0	2p (33%)	0

CTR: cricotracheal resection; ETT: endotracheal tube; LTR: laryngotracheal reconstruction; MTT: Montgomery T-tube; p: patient; SGS 1: subglottic stenosis Cotton grade I; SGS 2: subglottic stenosis Cotton grade II.

Table 3 Final Results.

Surgical technique	Good ventilation, swallowing and voice	Dysphonia	Respiratory distress	Death
LTR with MTT				
Graft	16p (64%)	9p (36%)	0	0
Lateral release	17p (85%)	1p (5%)	2p (10%)	0
Posterior split	2p (50%)	2p (50%)	0	0
LTR with ETT	4p (80%)	1p (20%)	0	0
LTR with mould	0	1p (100%)	0	0
CTR with MTT	8p (80%)	2p (20%)	0	0
Anterior cricoid split	4p (66%)	1p (17%)	0	1p (17%)

CTR: cricotracheal resection; ETT: endotracheal tube; LTR: laryngotracheal reconstruction; MTT: Montgomery T-tube; p: patient.

The factors can also be extrinsic: related to the ETT (large size or excessively hard), to the intubation (traumatic, multiple, and prolonged[10]) and to nursing care (insufficient sedation, excessive tube mobilisation and repeated and traumatic aspirations).[1,11]

Traumatic intubation and the pressure induced by the ETT are the principal factors that contribute to post-intubation SGS.[1] When the pressure from the ETT exceeds the pressure of capillary perfusion, it causes ischaemia, followed by oedema, necrosis and ulcers. Later on, in the repair process, the granulation tissue can lead to a decrease in the airway lumen and to obstruction.

Longer than 4 weeks of intubation is considered to increase the risk of stenosis from superinfection, but damage is possible even within 48 h.[1]

The locations most affected by ETT are the vocal processes of the arytenoid cartilages, as well as the lateral and posterior areas of the cricoid cartilage. Preventing ETT with a balloon in children younger than 8 years old or its low-pressure use has notably lowered the incidence of post-intubation tracheal stenosis.[1]

Extubation failure or persistent dysphonia 3 days later than such failure requires endoscopic evaluation to prevent post-intubation stenosis.[1] Acute lesions due to intubation should be treated promptly, even if tracheotomy is inevitable to ensure the airway.[1] Without treatment, these lesions can develop into laryngeal scar sequelae that affect quality of life, causing dysphonia or dyspnoea (Fig. 3).

The category SGS 3b (SGS+comorbidities) corresponded to 57.7% of our patients. The most frequent comorbidity was gastroesophageal reflux.

Three surgical techniques were used. The decision as to which technique to use was a result of the prior endoscopic

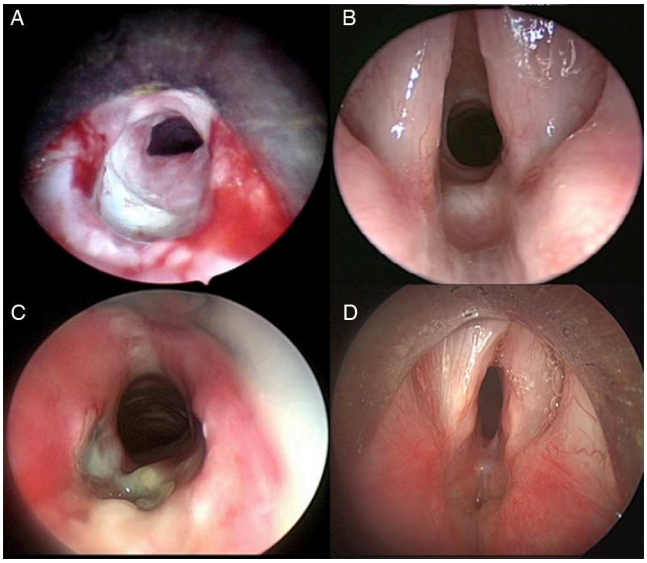

Figure 3 (A and C) Acute post-intubation lesions. (B and D) Laryngeal sequelae: (B) Predominantly posterior subglottic stenosis. (D) Synechia of posterior commissure.

assessment of the patient, combined with the experience of the surgeon, and secondly from the radiological images. The family context for patient contention, both in the immediate postoperative period and concurrence to the follow-ups, was also taken into consideration.

At the moment of surgery, 87.3% of patients were tracheotomized. The reasons for this therapeutic option were the wait for growth of the patient and of the laryngeal structures, and the wait for the ''cooling'' of the structures damaged by the period of intubation. Laryngeal surgery was performed subsequently.

In our cases, the technique most frequently used was LTR with MTT (69%), with a slight prevalence of grafts (25 cases) over lateral release. Cartilage proved to be an excellent graft material for LTR, given that its structure is similar to that which we were attempting to improve. The type of cartilage, as well as its shape and size, depended on the location and severity of SGS. The graft most placed was the thyroid alar graft, although in the last few years it was principally the costal graft because of its size and rigidity. In 1 patient we also used the auricular; although the treatment was a success, we noted the disadvantage of the structural weakness of the graft due to its wall thinness.

For 20 patients we chose to obtain an expansion of the laryngeal framework using lateral division at 3 and 9 o'clock of the cricoid, while for 4 patients we used a posterior split in the midline of the posterior lamina of the cricoid. The interposition of the graft is not always needed to obtain stable expansion, but its use improves re-epithelialization, minimises the development of granulation tissue and fibrosis, which leads to a reduction in the duration of the stent and better time for decannulation.[5] Of the 49 patients with LTR with MTT, the 8 patients that remained with MTT for 6–8 months had a graft, while the 58.3% who had MTT for 12–16 months did not.

There were no significant differences with respect to the diameter of the lumen to the MTT extraction when comparing the surgical techniques, although the grafts did require more treatments *a posteriori*. Two patients that continued with slight ventilatory difficulty correspond to the group treated with lateral release.

The endoluminal prosthesis collaborates with improved stability in the new reconstructed airway.[12–14] The ones most used are of silicone, with the most well known being the MTT.[15] An MTT was placed in 59 patients. It provokes little tissue reaction, it is relatively easy to place and remove and it has automatic fixation. The MTT does involve a risk of obstruction, especially when the internal diameter is small.[16] This risk is minimised with continuous plugging of the prosthesis and appropriate aspiration. World-wide literature recommends keeping the prosthesis for 2 to 8 weeks; in cases of multiple-step surgeries, severe stenosis or extensive stenosis, the recommendation is for up to a year.[17]

One patient received a short suprastomal stent, created from an MTT, closed in the proximal and distal ends with rounded silicone caps.[12] Among its advantages were early oral feeding without risk of aspiration, and short decannulation time. The rounded ends of the prosthesis prevented the formation of granulation tissue.[12]

In 5 patients a 1-step LTR was performed. This can be effective to avoid tracheotomy or long periods with a stent. Proper postoperative care is crucial. There should be an excellent intensive care unit. In the 1-step LTR, the nasotracheal tube collaborates with laryngeal and upper tracheal support.[14] Sedation prevents agitation and accidental extubation.[18] The 2-step LTR does not require care in therapy. In our patients, the technique of 1-step LTR provided less delay in decannulation, in spite of the fact that 60% of them required a second surgical treatment.

In 10 patients we performed CTR with MTT for 12 months, achieving final success (good ventilation, swallowing and voice production) of 80%. It is a safe, efficient procedure for the treatment of severe SGS with or without upper tracheal compromise. It is especially appropriate for managing serious SGS with concentric scarring in the lower subglottis, and as a rescue technique for the airway when LTR has failed. It glottic compromise exists, an extended CTR (with posterior graft) should be performed.[19,20]

In 6 patients an anterior cricoid split was performed. This is indicated in infants with unsuccessful attempts of extubation due to subglottic disease, to avoid tracheotomy. At present, the preference is to perform an anterior cartilage interposition at the same time.

All patients received indication of anti-reflux treatment during the period of time that they had the stent. Gastroesophageal reflux has been shown to have a negative effect on the laryngotracheal structures, which can lead to the failure of the surgical treatment.[4,5]

In our patients, 15.5% (11 cases) had to be operated on again to achieve decannulation with acceptable parameters of ventilation, swallowing and voice. Two patients required a third surgical treatment.

Failure in the airway reconstruction can be secondary to factors that are not under the surgeon's control; However, several of them can be avoided with an exact preoperative assessment and intraoperative reassessment of the stenosis.[21] In our series, the factors related with failure were preoperative: inadequate assessment of the multilevel compromise of the stenosis, combined with the presence of comorbidities (gastroesophageal reflux or glossoptosis); intraoperative: length of the stent or improper procedure chosen; and postoperative: keloid formation, tracheal collapse, inadequate follow-up or gastroesophageal reflux.

The most frequent complication was granuloma of the upper free border of the MTT, due to the brush of the impurities of its borders with the supraglottic laryngeal structures, needing to be changed in 16 patients. Later development after having found the correct placement site was similar to that of the other patients. In the 2 cases of accidental extraction of the MTT, the stent was replaced and this complication did not affect the result.

Dysphonia was the most frequent sequela. Extending the inter-arytenoid space by the posterior graft can affect voice results negatively, as complete closure of the vocal folds is prevented during phonation (the 4 patients with a posterior graft presented dysphonia). The vocal results of CTR (20% dysphonia) would probably be better than those of the LTR (25.4% dysphonia) for similar lesions.

The majority of the patients with MTT were fed 5 days after surgery, after the adaptation to the MTT that keeps the vocal folds in abduction. Ingestion of liquids had a longer delay due to the difficulty in oral and pharyngeal management. The mould permitted early oral feeding (at 48 h).

Decannulation was achieved in 70 cases. The principal cause of delay in decannulation was the presence of restenosis. One patient died 1 month after surgery (anterior cricoid split) from causes not related to the surgery. We excluded from this study 11 patients with post-intubation SGS operated on during this period because at the moment of analysis they still had an endoluminal prosthesis.

Conclusions

Prevention is fundamental. An ETT should never be inserted by brute force. The tube used should be of the smallest diameter needed to ventilate the patient, with appropriate sedation to stop tube movement from harming laryngeal structures. There should be energetic treatment of gastroesophageal reflux and of the factors that cause hypoperfusion, exacerbating the trauma of the tube.[1]

When there is a patient with SGS, choosing the appropriate treatment is the key to success. It decreases the number of operations and avoids complications.

Conflict of Interests

The authors have no conflicts of interests to declare.

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