

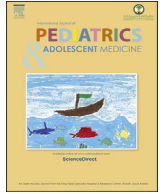
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International Journal of Pediatrics and Adolescent Medicine

journal homepage: <http://www.elsevier.com/locate/ijpam>

Original research article

Tertiary center experience with primary endoscopic laryngoplasty in pediatric acquired subglottic stenosis and literature review

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ARTICLE INFO

Article history:

Received 14 May 2016

Received in revised form

10 September 2016

Accepted 22 November 2016

Available online xxx

Keywords:

Acquired subglottic stenosis

Laryngoplasty

Pediatric

ABSTRACT

Background and objectives: To share our experience with primary endoscopic laryngoplasty in pediatric acquired subglottic stenosis and critically review the previously published studies.

Setting: Tertiary Referral Centers, King Abdulaziz Medical City, Riyadh, Saudi Arabia.

Patients and methods: A retrospective case series study was conducted, where the case notes of all pediatric patients who underwent endoscopic management as a primary surgical intervention for acquired subglottic stenosis (SGS) from 2004 to 2014 were reviewed. All patients who underwent surgical correction with primary open laryngoplasty for congenital subglottic stenosis had been excluded.

Results: A total of 60 patients with a workable diagnosis of subglottic stenosis were reviewed. Forty-five patients were included in the study and 15 patients were excluded because they underwent open laryngoplasty as a primary treatment modality for congenital subglottic stenosis. The majority of the patients were males 29 (64%), with 16 (36%) females. The main presentation was stridor and intercostal recession. Thirty-nine (86%) patients had subglottic stenosis due to prolonged intubation; 5 (11%) patients were idiopathic and one patient (3%) had inflammatory reasons. The site of stenosis was isolated SGS in 41 while 4 patients had glottic-subglottic stenosis (GSGS). In terms of the grade of stenosis: 13 patients had grade I; 23 had grade II and 9 had grade III. The character of stenosis was soft except in 5 patients with hard (mature) scar. The number of dilatation procedures ranged from 1 to 6 with a mean of 2. The endoscopic management was considered to be successful when the patient is completely asymptomatic after the follow up period of one year. No major complications were recorded among the patients. Thirty-seven (82.3%) patients had a benign course post endoscopic intervention without complications and 8 (17.7%) underwent a secondary open surgical management due to re-stenosis (P value < 0.01).

Conclusion: Our study showed that primary endoscopic management was successful in 82.3% of cases of acquired subglottic stenosis including those with high grade stenosis and long segment of more than 12 mm in terms of the craniocaudal length. CO₂ laser was an important tool to convert mature hard stenotic segment into a soft one. The latter yielded to the lateral pressure created by balloon dilatation better.

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1. Introduction

Laryngotracheal stenosis in children can be congenital or

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Peer review under responsibility of King Faisal Specialist Hospital & Research Centre (General Organization), Saudi Arabia.

<http://dx.doi.org/10.1016/j.ijpam.2016.11.001>

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acquired. The subglottis is the most common site of airway narrowing in children [1]. In congenital cases, malformation of the cricoid cartilage is characteristic of the stenosis. Prolonged intubation is the most commonly identified risk factor for acquired subglottic stenosis (SGS), accounting for 90% of cases [2]. However, the likely pathogenesis of SGS begins with subglottic mucosal pressure necrosis secondary to endotracheal intubation, followed by mucosal ulceration, perichondritis, and mature scar tissue

formation. Gastroesophageal reflux and exacerbates this process and has been demonstrated as a risk factor for failed airway reconstructive surgery [3,4]. The incidence of SGS has decreased from 24% in 1960 to 1–2% in the year 2000 [5]. This case reduction has largely been the result of advances in airway management and intubation guidelines [6].

In 1994, SGS was staged by Myer–Cotton into 4 stages. Grade I lesions have less than 50% obstruction, Grade II lesions have 51%–70% obstruction, Grade III lesions have 71%–99% obstruction, and Grade IV lesions have no detectable lumen or complete stenosis [7]. A variety of options for managing SGS exist, including observation, pharmacotherapy, dilatation, laser, single or multi-stage open surgical reconstructions, and tracheostomy [8,9]. SGS management remains a challenge for pediatric otolaryngologists. Endoscopic techniques for repairing airway stenosis have been employed since 1870 [10], but they had decreased by the 1970s in favor of open laryngotracheal reconstruction (LTR) [11]. With the advent of balloon angioplasty catheters for cardiac procedures, dilatation procedures were expanded to other body systems, including pediatric-acquired airway stenosis [12]. Endoscopic management has recently gained popularity, but guidelines regarding post-operative observation and repeat dilations have not yet been established, and its use remains uncommon for mature and severe stenosis [13,14].

In 2012, Charlotte Hautefort et al. [15] distinguished between primary and secondary SGS treatments with balloon dilatation. In primary treatment, balloon dilatation was the only SGS treatment modality following open surgery, provided that open surgery was performed one year or more prior to the intervention (in the form of balloon dilatation). For secondary treatment with balloon dilatation, the balloon dilatation would follow open surgery, provided that the open surgery was performed within one year of the balloon dilatation. Likewise, a distinction was made between acute (soft) versus mature (hard) acquired SGS. In both cases, the goal of endoscopic management is to mechanically interrupt the process of scar formation in the subglottis [6].

Our study focused on the outcomes of endoscopic management as a primary modality for acquired SGS treatment, using a CO₂ laser to convert the mature hard stenosis into soft stenosis. We also took this opportunity to critically review the literature of the subject.

2. Patients and methods

Ethical approval of the study was provided by the King Abdullah International Medical Research Center (KAIMRC).

2.1. Patients

This retrospective case series included 45 pediatric patients (ranging in age from 1 month to 14 years) who underwent endoscopic intervention as a primary modality of treatment for acquired subglottic stenosis in a tertiary referral center (King Abdulaziz Medical City, Riyadh, Saudi Arabia) from April 2004 until June 2014. All children who had a workable diagnosis of acquired subglottic stenosis and who underwent at least one endoscopic intervention as primary modality of treatment without previous primary open laryngotracheoplasty were included in the study.

Children who were diagnosed with cartilaginous congenital subglottic stenosis were excluded. Charts were reviewed for the number of primary surgeons, age, gender, history of prematurity, significant medical comorbidities, descriptions of subglottic stenosis, types and timing of interventions for subglottic stenosis and airway status at the last available follow-up visit. The diagnosis and description of subglottic stenosis was performed by the pediatric otolaryngologists in the operating room at the time of the direct

laryngobronchoscopy (DLB). Information on the characteristics of the subglottic stenosis was taken from the surgeon's description in the surgical notes. A total of 3 pediatric otolaryngologists (1 consultant, 2 fellows) participated in the surgeries, and the consultant was always there to confirm the description. The Myer–Cotton grading system was used to standardize all descriptions of the degree of the subglottic stenosis. The follow-up lasted for one year after the last intervention.

2.2. Statistical analysis

All collected data were analyzed with SPSS version 17 using the mean \pm SD for the quantitative variables and frequencies and percent for the qualitative variables. All statistical analyses of the qualitative data before and after endoscopic treatment of SGS were performed using McNemar's test. Differences were considered to be significant when the *P* value was $>.05$ and highly significant when the *P* value was $>.01$.

2.3. Surgical procedure

DLB was performed using a 2.7 or 4 mm 0° Karl Storz telescope with the patient under general anesthesia, and the spontaneous ventilation technique was used. A complete evaluation of the stenosis (i.e., describing the grade, site, craniocaudal extension and characteristic of stenosis as soft or rigid) was performed at the first diagnostic session. When the stenosis was initially too tight or too sinuous, a complete description could not be achieved. To complete our examination, we dilated with the Savary-Gilliard® dilator to pass a 2.7 mm scope. If the stenosis was hard and mature, as found in 5 patients, the hard part had to be excised and converted into soft using a CO₂ laser in the ultrapulse mode (125 mjoules/cm² and 10 Hz repetition rate). This setting minimized overheating of the tissue and, subsequently, the recurrence of fibrous tissue (Fig. 1A).

The soft stenotic tissue was then dilated using the Balloon Dilation System, which was composed of a high pressure, non-compliant balloon catheter with an integrated stylet and optimized for airway anatomy, INSPIRA AIR™ (Acclarent Inc., CA, U.S.A.). These components are translucent to facilitate balloon positioning at the level of the stricture. The balloon was inflated using an inflation/deflation handle mounted with a syringe and gauge assembly (ACCLARENT® Balloon Inflation Device) designed to monitor and maintain pressure for 60 s (time-induced local ischemia) or until the patient's oxygen saturation level dropped below 92%. The balloon size was selected according to the ideal subglottic diameter for the patient's age. This procedure was performed 2 or 3 times during each session (Fig. 1B). The number of dilatations per procedure never exceeded three. There was no specific protocol concerning the diameter of the balloon, which was adapted according to each individual by the endoscopist in relation to the patient's age, as well as the degree and etiology of stenosis.

After the balloon dilatation, a local injection of 1–2 ml (Kenalog-40®) (triamcinolone topical steroid) diluted in 1 ml of normal saline infiltrated into the surrounding tissue to decrease the healing process and scar formation (Fig. 1C). The area was then cooled and cleaned with a saline-soaked pledget, and any extra debris was removed (Fig. 1D). In all cases (i.e., hard and soft scarring), the children were not intubated after dilatation; instead, they were monitored in the pediatric intensive care unit (PICU) for 24–48 h using a non-invasive ventilating system when required. The second procedure was performed after a minimum interval of 2–3 weeks in all cases. The follow-up endoscopy included a second balloon dilatation, as necessary. All patients received systemic steroids (prednisolone, 1–2 mg/kg/d) and proton pump inhibitors (esomeprazole, 2 mg/kg/day) for 3–10 days. Treatment was considered to

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