International Journal of Pediatrics and Adolescent Medicine 4 (2017) 33–37

HOSTED BY

Contents lists available at ScienceDirect

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

Jaber Alshammari [a], Arwa A. Alkhunaizi [b,*], Abdullah S. Arafat [c]

[a] King Saud bin Abdulaziz University for Health Sciences (KASU-HS) – Riyadh, Saudi Arabia, King Abdulaziz Medical City (KAMC) – Riyadh, Saudi Arabia, P.O.Box 22490, Riyadh 11426, Saudi Arabia
[b] King Abdulaziz Medical City (KAMC) – Riyadh, Saudi Arabia, P.O.Box 22490, Riyadh 11426, Saudi Arabia
[c] King Saud bin Abdulaziz University for Health Sciences (KASU-HS) – Riyadh, Saudi Arabia, P.O.Box 3660, Riyadh 11481, Saudi Arabia

ARTICLE INFO

Article history:
Received 14 May 2016
Received in revised form
10 September 2016
Accepted 22 November 2016
Available online 10 February 2017

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. CO_2 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.

1. Introduction

Laryngotracheal stenosis in children can be congenital or 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

* Corresponding author.
E-mail addresses: Jabber.orl@gmail.com (J. Alshammari), Arwa.alkhunaizi@hotmail.com (A.A. Alkhunaizi), Arafat.as@outlook.sa (A.S. Arafat).

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

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 postoperative 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 CO_2 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 ± 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 CO_2 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 AIRTM (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 scaring), 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 dilation, 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

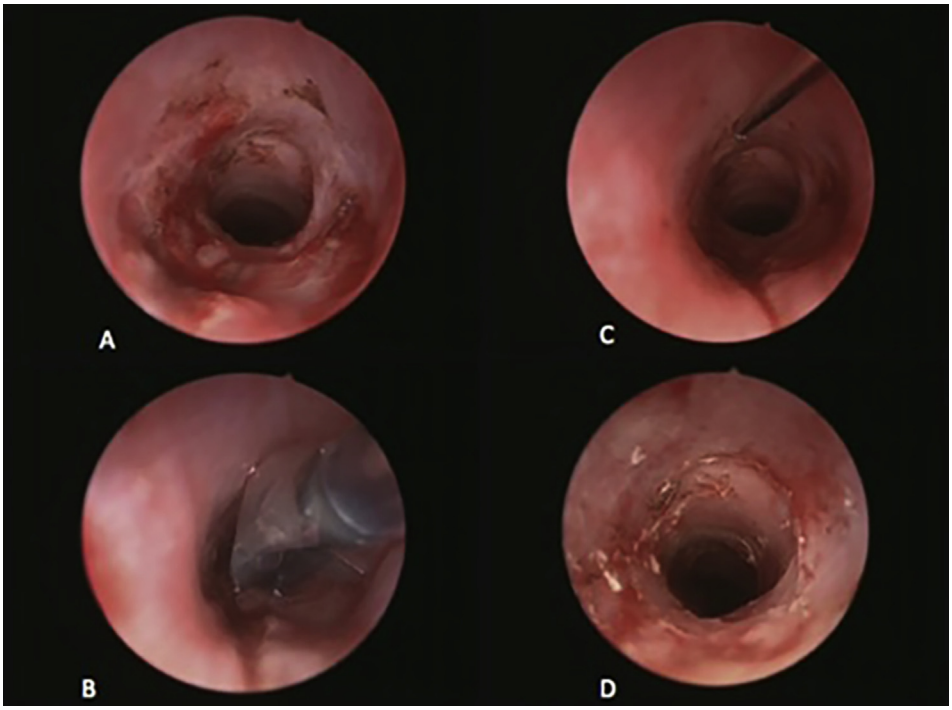

Figure 1. The surgical steps for endoscopic management of SGS. A) After complete excision of the scar using a CO_2 laser; B) a balloon catheter inside the stenotic part; C) a local steroid injection in the surrounding tissue; and D) the final image after one therapeutic session.

be successful if a noticeable improvement in the scar thickness, length or texture was observed and necessitated further sessions until complete healing was achieved (Fig. 2). After complete resolution of the stenosis, the patients were followed with diagnostic sessions every 3–6 months for one year.

3. Results

The case notes of 60 children (ranging in age from 1 month to 14 years) with a workable diagnosis of SGS were reviewed. Fifteen patients were excluded because they underwent open surgery as a

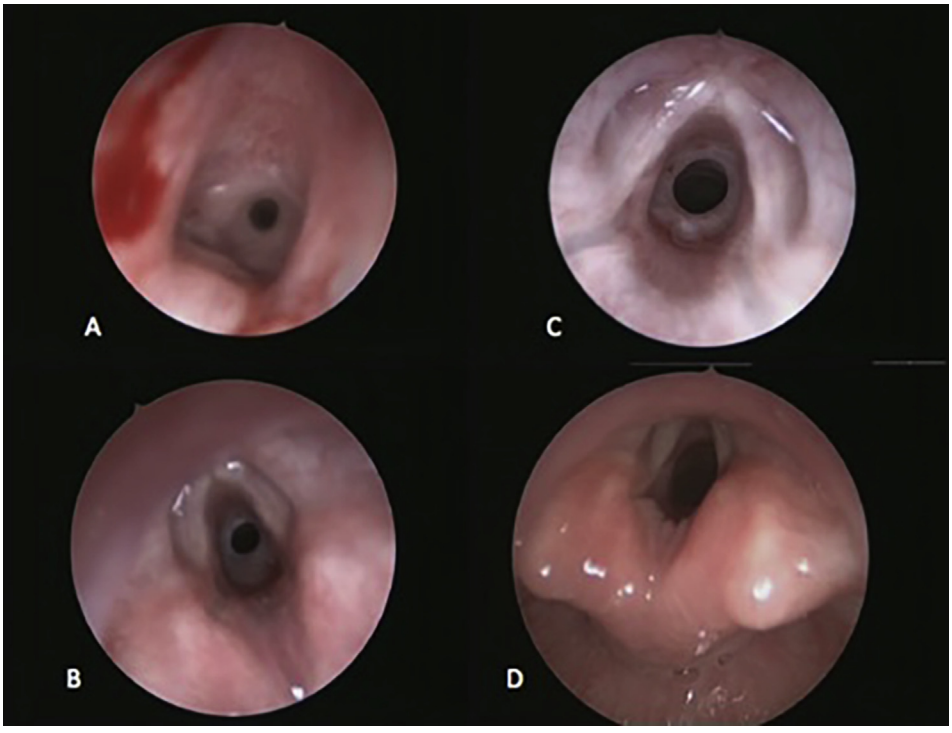

Figure 2. In the same patient, the gradual response of SGS to four endoscopic management sessions.

Table 1
Demographic and clinical criteria of the 45 SGS patients.

Criterion	
Age range	1 month to 15 yrs
Sex	
Male	29 (64.4%)
Female	16 (35.6%)
Site of stenosis	
Isolated subglottic stenosis (SGS)	41 (91.1%)
Glottic and subglottic stenosis (GSGS)	4 (8.9%)
Etiology	
Prolonged intubation	39 (86.7%)
Idiopathic	5 (11.1%)
Inflammatory	1 (2.2%)
Type	
Soft	40 (88.9%)
Hard	5 (1.1%)
Tracheostomy	
No	34 (75.6%)
Yes	11 (24.4%)
Grade of stenosis	
Grade I	13 (28.9%)
Grade II	23 (51.1%)
Grade III	9 (2.0%)
Grade IV	0 (0.0%)

primary treatment modality for congenital subglottic stenosis. Table 1 shows the breakdown of the included patients by demographics, etiology, previous tracheostomy, and the size, grade and type of stenosis. The majority of the patients were males 29 (64%), with 16 (36%) females, and they ranged in age from 1 month to 14 years. The main presentations were stridor and intercostal recession. Eleven patients had received a tracheostomy before our intervention. In terms of the etiology, 39 patients (86.7%) had SGS due to prolonged intubation; 5 patients (11.1%) were idiopathic, and one patient (2.2%) had inflammatory causes. As a risk factor, the duration of intubation ranged from 1 to 12 weeks. Gastroesophageal reflux disease (GERD) was confirmed in 4 patients. Other associated medical comorbidities were prematurity (15 pts), cardiac anomalies (14 pts), head injury (3 pts) and DiGeorge syndrome (1 pt). The stenosis site was limited to the subglottic region in 41 patients (91.1%), while 4 patients (8.9%) had glottic-subglottic stenosis (GSGS). The glottic stenosis in these latter patients was in the form of soft fibrous band occupying the posterior glottis, and it was not in continuation with the subglottic narrowing. The severity of stenosis was grade I in 13 patients (28.9%), grade II in 23 patients (51.1%), and grade III in 9 patients (20.0%). None of our patients had grade IV stenosis. The character of stenosis was soft in all patients, except in 5 patients with hard mature scaring. The craniocaudal extension was ranging from thin membranous tissue (2 mm) to (12 mm) and a mean of (7.6). All patients were included in 62 balloon treatment sessions and underwent a total of 132 balloon dilatation procedures. The number of dilatation sessions per patient ranged from 1 to 6, with a mean of 2. The first balloon size ranged from 4 to 10, and size 6 was used most frequently. The last balloon size ranged from 6 to 14, and the most common size was 6.

Endoscopic management was considered to be successful when the patient was completely asymptomatic, which was achieved in 37 patients (82.2%) (*P* value <.01) (Table 2). After a one-year follow-up period, 34 patients (75.6%) had no stenosis (Grade 0), 6 patients (13.3%) had grade I stenosis, and 5 patients (11.1%) had grade II stenosis (*P* value <.01). Three patients with grade I stenosis and five patients with grade II stenosis (8 pt, 17.7%) who had stridor upon exertion underwent secondary open surgical management after the failure of primary endoscopic management. The 11 patients (24.4%) with tracheostomy before our intervention underwent decannulation later during their treatment program. There were no clinically significant observed complications with our endoscopic management.

Table 2
Stridor and endoscopic demographic grading before and after the endoscopic treatment of SGS.

Variable	Pre-endoscopy	Post-endoscopy	*P* value
Stridor			
No	0 (0.0%)	37 (82.2%)	<.01
Yes	45 (100.0%)	8 (17.8%)	
Grade of stenosis			
Grade 0	0 (0.0%)	34 (75.6%)	<.01
Grade I	13 (28.9%)	6 (13.3%)	
Grade II	23 (51.1%)	5 (11.1)	
Grade III	9 (2.0%)	0 (0.0%)	
Grade IV	0 (0.0%)	0 (0.0%)	

4. Discussion

The first case of balloon laryngoplasty for SGS described in the literature was in 1985, when Axon et al. reported a case of a symptomatic four-year-old child who was diagnosed with grade II SGS after intubation. The authors used a balloon (6 mm in diameter and 15 mm long) inflated at 6 atm of pressure, leaving it in the subglottis until the child's hemoglobin saturation dropped to 80%. The child remained asymptomatic after 15 months of observation [12].

Balloon laryngoplasty seems to be more effective than the other methods of dilatation because the entire force employed is radial, toward the stenosis. The lack of shearing forces reduces subglottic trauma at the mucosa and in the deeper areas, thereby reducing the chances of re-stenosis [12]. Moreover, because of the small deflated balloon diameter, it can be passed through extremely narrow areas, without traumatizing the area [14]. One of the challenges encountered was the difficulty in assessing the craniocaudal length of grade III stenosis because the lumen is too narrow to accommodate a 2.7 mm endoscope. Therefore, we had to use a Savary dilator to widen the lumen enough to allow passage of the endoscope and enable us to view the distal end of stenosis. This process was even more challenging in our 5 patients with hard and long segment of stenosis (more than 12 mm long).

The risk of re-stenosis can be minimized by radial incision of the circumferential stenosis (by scalpel or laser) prior to dilation [6]. Our experience with the CO_2 laser is that it converts the mature stenotic segment into an immature one through complete excision of the hard stenotic scar. The latter will respond well to the lateral pressure created by balloon dilatation.

There is a wide range of previously reported success rates for resolution of symptoms and decannulation using endoscopic techniques. In the literature, rates range from 40% to 100%, likely due to variability in the types of stenosis, complexity of the patients, and application of the multiple endoscopic techniques [16]. Our study concurs with the previous studies in that balloon dilatation is successful in managing selective cases of SGS, namely, the acquired soft stenosis. Nonetheless, our study shows that hard and long stenosis segments of 12 mm or more can be successfully managed endoscopically as the primary treatment modality if we could convert the hard segment to a soft one using CO_2 laser for excision. The outcomes are determined by postoperative symptomatology, endoscopic grading of any residual SGS, complications, and the need for subsequent interventions to control the residual SGS [6].

Currently, no consensus exists regarding the specific indications for endoscopic versus open surgical management of SGS in children. The development of guidelines for the use of various

endoscopic procedures is complicated by the multiplicity of techniques and tools available for endoscopic procedures and variability in the characteristics of the stenosis from patient to patient [17]. Our method could be shortened because we performed serial endoscopic sessions, as previously described, if there was any response in the form of grading, extension or clinical picture. If no improvement was noticed after two consequent sessions, we moved to the open surgical technique.

We suggest a potential role for intraoperative local steroid injection. Although they are frequently used, there are few published definitive studies regarding the utility of such injections. The optimal timing, administration route, and dosing are still primarily anecdotal in the literature [13]. The application of topical steroids may contribute to the inhibition of restenosis following dilatation. Inflammation management with local and systemic steroids and gastroesophageal reflux management with proton pump inhibitor therapy must be part of the overall management scheme [6].

There are multiple inherent difficulties in interpreting retrospective studies, such as multiple primary surgeons, different endoscopic approaches (laser type, laser settings, balloon dilatation, Savary dilatation), variation in the application of adjuvant medical treatment (steroid local and oral), patients with a wide range of comorbidities and lack of uniformity among airway patients (e.g., age, weight, gender, ethnicity, stenosis diameter, length, histology, neurologic function, glottal status, and comorbidities). The factors that we encountered and that limited our study results make it difficult to design a controlled study comparing balloon dilatation with more conventional therapy. Despite these obstacles, this case series represents an important mechanism for sharing experiences [18].

5. Conclusion

Our study showed that endoscopic management was successful in 82.2% of cases of acquired subglottic stenosis, including those with long segment stenosis of 12 mm or more, in terms of the craniocaudal length. The CO_2 laser was an important tool to convert mature hard stenotic segments into soft segments. The latter yields better to the lateral pressure created by balloon dilatation.

Conflict of interest

The authors declare that they have no competing interests.

Funding

This research did not receive any specific grants from public, commercial, or non-profit funding agencies.

Ethical approval

This paper was reviewed and ethically approved by King Abdullah International Medical Research Center, Riyadh, Saudi Arabia.

Acknowledgments

The authors would like to thank all the residents and interns who participated in the study. Special thanks to the Otorhinolaryngology Department at King Abdulaziz Medical City and the Medical Education Department at King Saud bin Abdulaziz University for Health Sciences for their technical support.

References

[1] Hebra A, Powell DD, Smith CD, Othersen Jr HB. Balloon tracheoplasty in children: results of a 15-year experience. J Pediatr Surg 1991;26(8):957–61.
[2] Cotton RT, Evans JN. Laryngotracheal reconstruction in children. Five-year follow-up. Ann Otol Rhinol Laryngol 1981;90:516–20.
[3] Baker S, Kelchner L, Weinrich B, Lee L, Willging P, Cotton R, et al. Pediatriclaryngotrachealstenosisandairway reconstruction: a review of voice outcomes, assessment and treatment issues. J Voice 2006;20(4):631–41. 4.
[4] Halstead LA. Gastroesophageal reflux: a critical factor in pediatric subglottic stenosis. Otolaryngol Head Neck Surg 1999;120(5):683–8.
[5] Choi SS, Zalzal GH. Changing trends in neonatal subglottic stenosis. Otolaryngol Head Neck Surg 2000;122(1):61–3.
[6] Durden Fredrick, Sobol Steven E. Balloon laryngoplasty as a primary treatment for subglottic stenosis. Arch Otolaryngol Head Neck Surg 2007;133(8):772–5.
[7] Myer 3rd CM, O'Connor DM, Cotton RT. Proposed grading system for subglottic stenosis based on endotracheal tube sizes. Ann Otol Rhinol Laryngol 1994 Apr;103(4 Pt 1):319–23.
[8] Durden T, Sobol S. Balloon laryngoplasty as a primary treatment for subglottic stenosis. Arch Otolaryngol Head Neck Surg 2007;133(8):772–5.
[9] Hebra A, Powell D, Smith C, Othersen H. Balloon tracheoplasty in children: results of a 15-year experience. J Pediatr Surg 1991;26(8):957–61.
[10] Holinger PH, Kutnick SL, Schild JA, Holinger LD. Subglottic stenosis in infants and children. Ann Otol Rhinol Laryngol 1976;85:591–959.
[11] Fearon B, Ellis D. The management of long term airway problems in infants and children. Ann Otol 1971;80:669–77.
[12] Axon PR, Hartley C, Rothera MP. Endoscopic balloon dilation of subglottic stenosis. J Laryngol Otol 1995;109:876–9.
[13] Edmondson Natalie E, Bent John. Balloon dilation as an alternative to open repair of subglottic stenosis. Int J Pediatr Otorhinolaryngology 2010;74:1078–81.
[14] Lindhe Guarisco J, Yang Christina J. Balloon dilation in the management of severe airway stenosis in children and adolescents. J Pediatr Surg 2013;48:1676–81.
[15] Hautefort Charlotte, Teissier Natacha, Viala Paul, Van Den Abbeele Thierry. Balloon dilation laryngoplasty for subglottic stenosis in children: eight years' experience. Arch Otolaryngol Head Neck Surg 2012;138(3):235–40 [III: Serial intralesional steroid injection combined with].
[16] Shapshay SM, Simpson GT. Lasers in bronchology. Otolaryngol Clin North Am 1983;16:879–86.
[17] Quesnel Alicia M, Soo Lee Gi, Nuss Roger C, Volk Mark S, Jones Dwight T, Rahbar Reza. Minimally invasive endoscopic management of subglottic stenosis in children: success and failure. Int J Pediatr Otorhinolaryngology 2011;75:652–6.
[18] Bent, Shah John P, Nord Maulik B, Parikh Ryan, R Sanjay. Balloon dilation for recurrent stenosis after pediatric laryngotracheoplasty. Ann Otology, Rhinology Laryngology Sep 2010;119:9.