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The outcomes of endoscopic management in young children with subglottic stenosis

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ABSTRACT

Subglottic stenosis (SGS) is a common cause of obstructed airway in children, and the treatment of pediatric SGS, especially congenital SGS, remains a challenge for the otolaryngologist.
Objective: To analyze the outcomes of endoscopic management in young children with SGS.
Methods: We performed a retrospective review of treatment with endoscopic balloon dilation (EBD) or EBD combined with endoscopic anterior cricoid split (EACS) for young SGS children, from December 2008 to December 2015. The ages of patients ranged from 2 days to 12 years, median age was 5 months. The grade of them ranged from II to IV.
Results: For acute acquired SGS, 19 cases received EBD alone and the other 3 cases received EBD and EACS, the success rate was about 95.5%; For chronic acquired SGS, EBD and EACS was performed in 6 patients with a success rate of 66.7%; For congenital SGS, EBD and EACS was performed in 28 patients with a success rate of 85.7%. Overall, the success rate of endoscopic management in 56 young children was about 87.5%. Besides, No procedure-related complications were observed in any patients.
Conclusions: Endoscopic surgical technique offers a safe and effective approach for treatment of young children with SGS, especially in congenital SGS.

1. Introduction

In children, subglottic stenosis (SGS) is one of the most common causes of an obstructed airway, and is classified as either congenital or acquired [1]. Congenital SGS was first reported in the 1950s [2], some researches then found more cases in the past 10 years, due to the development of endoscopic technology and perinatal medicine. Although rare, it is still the third most common congenital laryngeal anomaly, after laryngomalacia and vocal cord paralysis [3]. For acquired SGS, it is the most common acquired anomaly of the larynx, and accounts for 95% of all cases, with 90% due to long-term or prior intubation, the incidence rate of acquired SGS is increased by mechanical ventilation, which is widely used in neonatal intensive care units (NICU) [4].

The treatment of pediatric SGS, especially congenital SGS, remains a challenge for the otolaryngologist. Defective canalization of the cricoid cartilage and/or conus elasticus results in a small, elliptical, thickened cricoid and/or excessive submucosal tissue [5]. Over the last 40 years, many procedures have been used in the treatment of SGS, including endoscopic management and open surgeries, such as endoscope balloon dilation (EBD) [6], laryngotracheoplasty [7], anterior cricoid split (ACS) [8], laryngotracheal reconstruction [9], cricotracheal resection [10], and others.

Increasingly, EBD is being proven as a valuable therapeutic option worldwide. A systematic review and meta-analysis demonstrated that EBD is an inviting, noninvasive option to manage pediatric SGS, that could preclude the need for tracheostomy and/or laryngotracheal reconstruction, it is successful in most patients over short-term follow-up and complication rates are low. Still, limitations are present, repeated procedures are necessary and increasing severity of SGS increases the likelihood of the treatment's failure [4]. In short, EBD is not recommended in cases where SGS is long-standing, congenital, or has cartilaginous involvement. In recent years, combining several endoscopic techniques has proven to be an effective treatment. For example, Lorenzo Mirabile et al. used endoscopic anterior cricoid split (EACS) and EBD in the treatment of 18 children, ranging from ages 1–101 months and grade II to IV. Of these: 8 children were diagnosed with congenital SGS while the remaining were acquired SGS, the postoperative

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follow-up ranged from 4 to 45 months, and treatment was effective in 83% of cases, with no residual respiratory symptoms and a grade of 0–1 SGS upon conclusion of the follow-up [11].

Open surgeries such as laryngotracheoplasty, laryngotracheal reconstruction, and cricotracheal resection are both effective and feasible, with good decannulation rates in patients with severe SGS [12,13]. However, complications, especially in vocal function and voice, cannot be completely avoided in open surgery [14], and the resulting damage is significantly worse than in endoscopic procedures. Although several studies suggest the feasibility of open surgery in children as young as newborns [15], in China, most open surgery is performed during the preschool years. As a result, a long-term tracheostomy has not to be avoided, high cost of the nursing is also a problem for the patient's family. Long-term tracheostomy can lead to tracheomalacia and stenosis at the tracheostomy site, tracheocutaneous fistula and so on, furthermore, tracheostomy tube obstruction can leads to sudden death, which cannot be ignored by pediatric otolaryngologist.

This study retrospectively reviews the endoscopic management of 56 children with SGS. This study's purpose is to assess the effectiveness of EBD alone and EBD combined with EACS as methods of endoscopic management in the treatment of varying grades of SGS in children.

2. Methods

2.1. Subjects and staging of SGS

A retrospective evaluation was conducted of endoscopic management procedures performed on 56 children with SGS by the Otolaryngology Department of Children's Hospital at Fudan University from December 2008 to December 2015. Of these 56 children, 32 cases were boys and 24 cases were girls, middle age was 5 months with a range from 2 days to 12 years, all of them received either intubation or tracheotomy due to retractions, dyspnea, or carbon dioxide retention. Children with a maxillofacial deformity, gastroesophageal reflux, a respiratory tract malformation, or a neuromuscular disorder were excluded from this study. All subjects of this study signed informed consent forms. The following data was reviewed from the medical records of patients: age, stage of SGS at the time of initial diagnosis, presence of tracheotomy, number of dilatation procedures, comorbidity, outcome, success rate and so on.

The etiology of SGS determines its classification as either congenital or acquired. Acquired SGS can be further classified as either acute acquired SGS or chronic acquired SGS. Acute acquired SGS is defined as the stage of diagnosis and treatment up to 30 days after extubation or tracheotomy as a result of failed extubation; Chronic SGS is defined as the stage of diagnosis and treatment more than 30 days of extubation or tracheotomy as a result of failed extubation. Of these 56 children, 28 cases were diagnosed as congenital SGS, and the others were diagnosed as acquired SGS.

The Myer-Cotton grade was used to estimate the grade of stenosis according to the following table [16]: grade I, less than 50% obstruction; grade II, 51%–70% obstruction; grade III, 71%–99% obstruction; grade IV, no detectable lumen or complete stenosis. Of these children, 21, 33 and 2 cases were grade II, grade III and grade IV, respectively. The detail clinical characteristic of 56 children were shown in Table 1.

2.2. Endoscopic procedures (shown in Fig. 1)

2.2.1. EBD

The diameter and length of balloon varied according to the age of the patient and the size of the airway. 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, and the length varied from 20 mm to 30 mm. The balloon was inflated to rated burst pressure and maintained for 2 min, and most children received 3 dilations in intervals of approximately 2 min, except in some special cases where the time and the number of inflations varied according to the size of airway, pulmonary reserve, and/or perception of glottis and/or supraglottic edema secondary to the balloon. Mean number of balloon dilatation procedures per child was 2.6, with a range from 1 to 8.

2.2.2. EACS

Utilizing a laryngoscope, a vertical midline anterior incision was made with a cold scalpel through the cricoid anterior ring.

2.2.3. Acquired SGS

BD was the first course of treatment for patients with acquired SGS. First, under an endoscope, the precise radial incisions were made on the stenosis. Next, balloon dilations were performed. If this failed, the diameter of trachea cannot be expanded to normal size, the EACS was used to incise scar tissue instead and the balloon dilations were retried, after operation, the tracheal intubation was maintained for two weeks in order to stent the subglottic lumen, while at the same time, tracheotomy at single-stage operation was closed.

2.2.4. Congenital SGS

EACS was performed under laryngoscopy, followed by EBD. After the procedure, the tracheal intubation was maintained two weeks [17], at the same time, tracheotomy at single-stage operation was closed. If the child received a laryngeal stent before the procedure, it can be keeped two months. Next, the tracheotomy was closed at single-stage operation, and the laryngeal stent was taken out.

2.3. Postoperative management

All patients received anti-reflux medication, analgesics, and intravenous antibiotics for 7–14 days. Next, laryngoscopy was performed on each patient, and if no dyspnea and stenosis was present, the tracheal intubation can be removed, with the exception of patients with the laryngeal stent, who received fiberbronchoscopy after the laryngeal stent was removed after two months.

2.4. Evaluation of outcome

Successful treatment was defined as an open, stable, and adequate airway, usually consisting of grade 0 or 1 SGS using endoscopic evaluation, without clinical respiratory compromise, hence, the patient being able to maintain their airway without tracheostomy.

2.5. Follow-up examination

The follow-up time ranged from 0.5 to 7 years. Laryngoscopy was performed every two weeks after removal of the intubation or laryngeal stent. If the Subglottic tissue was still narrow, EBD should be repeated. Once normal results were achieved two times, the patients were advised to receive a fiberbronchoscopy test between 24 and 30 weeks after the last time check.

3. Results

In acquired acute SGS group, all patients received single-stage