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Original Research Article

Diagnosis and treatment of pediatric subglottic stenosis: evaluation of our treatment protocol

Sanajeet Kumar Singh, Roohie Singh*, Sunil Goyal, Ravi Roy, D. K. Gupta, Rajeev Chugh

Department of ENT and HNS, Army Hospital Research and Referral, New Delhi, India

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***Correspondence:**
Dr. Roohie Singh,
E-mail: roohiesingh@yahoo.in

ABSTRACT

Background: The aim was to describe our experience in management of pediatric subglottic stenosis and formulate a management scheme based on our experience.
Methods: A prospective study and follow-up was conducted in children diagnosed with subglottic stenosis (SGS) (both congenital and acquired) from January 2016 to January 2019 (36 months). We have proposed further subcategorization of each grade of SGS into mild and severe depending on absence or presence of the described "hard signs" respectively.
Results: A total of 28 patients with SGS were identified. 8 had congenital while 20 had acquired SGS. There were 16 cases of grade II, 7 cases of grade III and 5 cases of grade I stenosis. None had grade 4 stenosis. Most common surgical intervention was microlaryngoscopy and bronchoscopy which had both diagnostic and therapeutic role (in conjunction with other intervention). Other interventions were balloon dilatation, combined laser and balloon dilatation, anterior cricoid split, laryngotracheal reconstruction and Partial cricotracheal resection. As of July 2019, overall recovery rate was 89.2% and decannulation rate was 81.2%. One patient succumbed to death due to because unrelated to SGS. Two patients are still under follow-up.
Conclusions: Number of procedures increases with increasing severity of stenosis. Further subcategorization of SGS as mild or severe guides in selecting endoscopic or open surgical management scheme. The type of procedure for each patient needs to be tailored according to requirement.

Keywords: Evaluation, Pediatric, Subglottic stenosis, Treatment

INTRODUCTION

Subglottic stenosis (SGS) is narrowing of endolarynx that occurs below level of glottis till first tracheal ring. It is more common in children because it is the narrowest region in airway of children.[1] Congenital SGS occurs due to defective canalization of cricoid cartilage and/ or conus elasticus which results in thick, elliptical and small cricoid with or without excessive mucosal soft tissue. It is third most common congenital anomaly of larynx. Acquired SGS is most common acquired laryngeal abnormality usually due to intubation during neonatal period.[2]

The nature and severity of stenosis in children and associated comorbidities dictate the approach towards it.[3] The decision may be difficult in borderline cases for example; a circumferential Grade 2 or tough fibrotic stenosis can be as severe as a grade 3 stenosis. Also, the success rate might be less in patients with associated syndromes or comorbidities. We intend to study the epidemiology of pediatric SGS and evaluate our technique in diagnosis and management of such cases. There are no fixed management guidelines to treat such life threatening condition and this study will further help us to manage such cases effectively. The aim is to gain maximum recovery in terms of functional airway by

minimum intervention procedures with less complications in minimum recovery time.

METHODS

This prospective study was conducted in a tertiary care hospital (Army Hospital, Research and Referral, New Delhi) over a period of 36 months (January 2016-January 2019). Ethical clearance was taken from the Ethical committee of the hospital. The data collected were age, gender, severity of SGS, clinical presentation, comorbidities, history of intubation or tracheostomy, type of diagnostic and surgical interventions, findings of endoscopy pre and post intervention, complications and outcomes. Patients till age of 12 years were considered. Those who presented with symptoms prior to any intubation or other airway insults were taken as congenital SGS cases. Patients who had clinical symptoms after history of intubation or other airway trauma were assumed to be cases of acquired SGS. All cases underwent microlaryngoscopy and bronchoscopy to evaluate the severity of stenosis and to monitor progressive outcomes after therapeutic intervention. Diameter of SGS was estimated by using appropriate sized endotracheal tubes.

The SGS was graded as per Myer-Cotton system.[4] In addition to this, severity of each grade was assessed by us which was multifactorial. Presence of 'hard signs" like involvement of 3 or more walls intraluminally, length of stenosis >1 cm, a stenosis which is endoscopically non-distensible, failed multiple (>2) procedures, associated co-morbidities that can affect wound healing and duration >6 months was considered to be 'severe' in a particular grade. E.g. grade II with presence of any of the hard signs was considered as "severe grade II" while in absence of hard signs was considered as "mild grade II".

Gastroesophageal reflux was taken care during perioperative period. Management of SGS was either by endoscopic or open surgical method. Endoscopic intervention included balloon dilatation or combined laser ablation and balloon dilatation. Open surgical intervention included anterior cricoid split, laryngotracheal reconstruction (LTR) and partial cricotracheal resection (PCTR). Mitomycin was locally applied in all case to prevent formation of granulation or to treat granulation due to previous insult or intervention.

The follow up period ranged from 6 months to 12 months. The outcome was measured as improvement of symptoms or improved endoscopy findings in patients who were not having tracheostomy prior to surgical interventions. While in case of tracheostomised or intubated individuals, decannulation or extubation was considered as good outcomes respectively.

RESULTS

We diagnosed 28 children with SGS after doing Diagnostic microlaryngoscopy and bronchoscopy that were symptomatic and required surgical intervention. 17 were females and 11 were males.8 had congenital stenosis and 20 had acquired stenosis.16 had Myer-Cotton grade II SGS while 7 had grade III and 5 had grade I stenosis (Table 1). Various causes for intubation in patients of acquired SGS are depicted in Figure 1.

Table 1: Grades of subglottic stenosis with number of patients.

Grade of SGS	Congenital SGS	Acquired SGS	Total
I	2	3	5
II	5	11	16
III	1	6	7

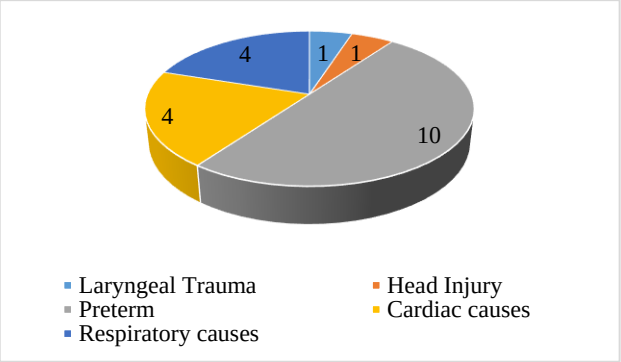

Figure 1: Causes of Intubation in acquired SGS.

The presentation was varied. In congenital cases of SGS; 2 had recurrent croup, 4 had stridor and 2 had breathlessness. Among the acquired cases 6 were already on tracheostomy and failed decannulation; 11 had failed extubation while 3 had difficulty in breathing on presentation. The age of presentation with symptoms ranged from 01 month to 12 yrs.

Tracheostomy was required in 3 congenital SGS cases and 7 acquired SGS cases prior to any intervention aimed for treatment.

Table 2: Types of final therapeutic interventions in each grades of subglottic stenosis which resulted in good outcomes.

Final intervention	Grade I	Grade II	Grade III
Balloon dilatation	3	8	-
Laser and balloon dilatation	2	4 (+1 died)	-
Anterior cricoid split	-	3	1
LTR	-	-	3 (+2 under follow-up)
PCTR	-	-	1

All grade I and most grade II (75%) were treated endoscopically. Few grade II (18.7%) and all grade III required open surgical intervention. Overall recovery rate was 89.2%. Three patients could not be decannulated, two are under follow-up and one died due to cause unrelated to SGS or its intervention (cause was congestive cardiac failure) (Table 2).

An evaluation of efficacy of management procedures according to severity of each grade of stenosis was done. Patients with mild and severe grade I stenosis as well as mild grade II stenosis showed adequate recovery after endoscopic treatment. Severe grade II and all grade III patients were treated efficiently by open surgical intervention.

DISCUSSION

Advancement in neonatology has resulted in prolonged endotracheal intubation resulting in increased incidence of acquired SGS.[5] In our study; a predominance of acquired SGS was observed (71.4%). The true incidence is difficult to determine as many cases might have had congenital SGS but got intubated and had been then considered as acquired SGS. Hence, true acquired SGS might have been over calculated and true Congenital SGS might had been under calculated. Myer-Cotton Grading system was used to scale the severity of SGS.[4]

Management may be either endoscopic or open surgical. Majority of grade I stenosis may be managed conservatively.[6] In our study, all grade I and most grade II were treated with good outcomes endoscopically i.e. five out of five (100%) and twelve out of sixteen (75%) respectively. Severe grade II (18.7%) and all grade III required open surgical intervention (Table 2).

Balloon dilatation of SGS is easy to perform as it can be passed through narrow spaces, effective due to radial force and has less chances to injure the mucosa and create granulation as a sequel of trauma.[7,8] Laser ablation with balloon dilatation can be performed in cases with circumferential fibrotic stenosis.[9] 2 cases of Grade I and 5 cases of grade II were managed in this way.

We used mitomycin in adjunct to all endoscopic treatment interventions since it inhibits fibroblast and scar formation. It was also used for reduction of granulation tissue. There are various studies that prove benefit of mitomycin.[10,11]

Anterior cricoid split involves giving an anterior incision on trachea from second ring up to cricoid and lower one third of thyroid ala. The child is left intubated for ten days with partially open neck wound to minimize subcutaneous emphysema.[2] In our study three neonates of grade II (75%) and one of grade III (25%) underwent this intervention with positive outcome.

Grade II and mild grade III require LTR with anterior cartilage graft while severe grade III require LTR with both anterior and posterior graft.[2] Five patients of grade III SGS underwent LTR with anterior cartilage graft or anterior and posterior cartilage graft. Three of them had granulation following the procedure which interfered with good outcome. After laser ablation of granulation, all three of them had positive outcome. Two patients are under follow-up and still remain cannulated. Hence recovery rate is 60% till date.

Severe grade III and grade IV stenosis and failure of LTR require PCTR. There are many studies which show absence of any laryngotracheal growth abnormality post PCTR. Hence the previous reluctance to this technique has been now overcome.[2] For PCTR, grading system is not predictor of success or failure, because the stenotic segment is completely resected. We did one PCTR in case of severe grade III SGS in which LTR failed proper outcome. After PCTR, three balloon dilatation at weekly intervals were done and then the child was successfully decannulated.

The most common complication was granulation post intervention. Other was dysphonia. Granulation was dealt with laser ablation and mitomycin application. For dysphonia, voice therapy was given.

Based on above experience, we have tried to develop a management scheme for Subglottic stenosis which forms the basis of our current practice. We categorized the cases into grade I, mild grade II, severe grade II, mild grade III, severe grade III and grade IV. A primary endoscopic procedure is considered useful in all grade I and mild grade II cases. In severe grade II, grade III and grade IV cases open surgical intervention is considered.

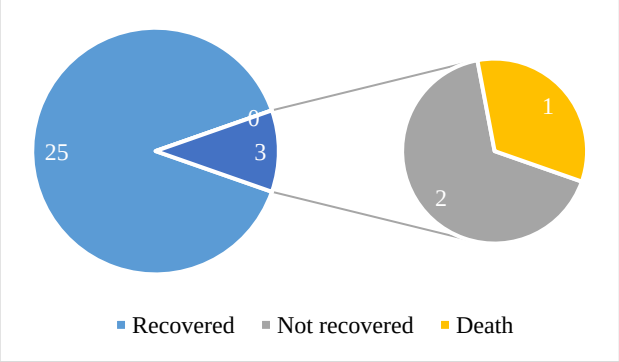

Figure 2: Overall recovery rate after final intervention.

Most authors give overall recovery rate and decannulation rate of about 70-90%.[6,8,12] Better outcomes are anticipated in less severe grades than in more severe grades.[2] In our study, overall recovery rate was 89.2% (twenty five patients out of twenty eight) and decannulation rate was 81.2% (thirteen out of sixteen patients) (Figure 2). More number of patients in less

severe grades of stenosis might be explained as cause for inflated decannulation and recovery rates in our study.

CONCLUSION

In the absence of strict management guidelines for management of subglottic stenosis, selecting the best treatment modality with optimal recovery and minimum complications can be challenging. Further sub-categorization of subglottic stenosis as mild or severe guides in selecting endoscopic or open surgical management scheme. Endoscopic management alone may be sufficient in majority of grades I and II SGS. The mean numbers of open surgical procedures increases with increasing severity of SGS. The stepping up of possible endoscopic intervention to open surgical intervention should be gradual and decision making should not be in haste. A combination of endoscopic and open reconstructive procedures should be planned as per requirement of each case to achieve appropriate functional outcomes.

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Ethical approval: The study was approved by the Institutional Ethics Committee

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