10.5005/jp-journals-10023-1028
ORIGINAL ARTICLE

Outcomes in Management of Pediatric Laryngotracheal Stenosis: Our Experience

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ABSTRACT

Background: Pediatric laryngotracheal stenosis is a difficult entity encountered in clinical practice which manifests with a myriad of etiologies and presentations. Management of pediatric laryngotracheal stenosis is a surgical challenge which requires meticulous preoperative evaluation and planning.

Aim of study: This is a retrospective study of the efficacy of different surgical techniques involved in restoring the airway patency in pediatric laryngotracheal stenosis performed at our institution over 4 years.

Materials and methods: Twenty-four pediatric patients underwent treatment for laryngotracheal stenosis over a 4-year period (June 2006-May 2010). Various surgical modalities, such as anterior cricoid split, costal cartilage interposition, stenting, laser procedures, etc. were used. Hood's stent, Montgomery T-tube, silastic swiss roll, indwelling nitinol tracheal stent were used to stent the airway after resection of the stenotic segment.

Results: Nine patients achieved successful decannulation with single-stage procedure and 14 patients underwent multiple procedures. Of 24 patients, decannulation was possible in 18 patients (75%), one patient had no indication for tracheostomy and five children (20%) have a persisting tracheostomy including two who were lost to follow-up.

Conclusion: It is vital to outline a rational approach to the management of laryngotracheal stenosis in children based on the site and severity of the stenotic segment. No single approach is ideal and often several procedures may be required before decannulation can be achieved.

Limitations of study: Patients were followed-up for 1 year. A long-term follow-up is preferable.

Keywords: Pediatric laryngotracheal stenosis, Hood's stent, Montgomery T-tube, Swiss roll.

How to cite this article: Shekhar A, Natarajan K, Sampath R, Kameswaran M, Murali S. Outcomes in Management of Pediatric Laryngotracheal Stenosis: Our Experience. Int J Phonosurg Laryngol 2012;2(1):14-19.

Source of support: Nil

Conflict of interest: None declared

INTRODUCTION

Laryngotracheal stenosis is a cicatricial narrowing of the laryngotracheal framework. Congenital stenosis occurs as a result of failure of recanalization. Subglottis is the most common site of stenosis in congenital cases. Increasing survival of patients in intensive care units with prolonged intubation has led to a dramatic increase in the incidence of laryngotracheal stenosis, often following premature birth. Management of pediatric laryngotracheal stenosis, whether acquired or congenital, remains a challenge. In infants and children, laryngotracheal stenosis may be associated with other congenital anomalies, which needs to be excluded. Selection of the appropriate surgical repair depends on the location of the stenotic segment, severity, duration and etiology of functional impairment.

STUDY METHODS

Twenty-four pediatric patients with laryngotracheal stenosis of varied etiology presented to Madras ENT Research Foundation between June 2006 and May 2010 and a rational treatment plan was formulated based on the site, extent and grading of stenosis. Various surgical modalities, such as anterior cricoid split, costal cartilage interposition, stenting, laser procedures, etc. were used. Hood's stent, Montgomery T-tube, silastic swiss roll and indwelling nitinol tracheal stent were used to stent the airway after resection of the stenotic segment. The stent was placed for an average period of 3 to 6 months and the airway was then reassessed. Majority of the patients were successfully decannulated, although several patients needed multiple procedures. The average follow-up period after surgery was 1 year. Follow-up assessment was based on clinical and endoscopic evaluation.

RESULTS (TABLES 1 TO 5)

Table 1: The various sites of stenosis that were encountered

Site of stenosis	*Number of patients (%)*
Supraglottic	1 (4.17)
Glottic	2 (8.33)
Glottic + subglottic	3 (12.5)
Subglottic	3 (12.5)
Subglottic + upper tracheal	4 (16.67)
Upper tracheal	11 (45.83)

Table 2: The largest group of patients were those suffering from postintubational stenosis

Etiology	*Number of patients (%)*
Congenital	5 (20.83)
Postintubation	12 (50.0)
Infection	1 (4.16)
Iatrogenic	2 (8.33)
Chemical injury (acid)	1 (4.16)
Neoplasia	1 (4.16)
Road traffic accident	2 (8.33)

Table 3: In our series, there were 10 patients who underwent single-stage procedure with nine achieving successful decannulation, while one is still on tracheostomy tube

Type of surgery	*No. of patients (%)*	*Site*	*Decannulation*
ACS, CCI, SR	1 (4.17)	Glottic	Yes
ACS, CCI, Montgomery T-Tube	3 (12.5)	Upper tracheal	Yes
		Upper tracheal	Yes
		Subglottic	Yes
Laser and Montgomery T-Tube	4 (16.67)	Upper tracheal	Yes
		Upper tracheal	Yes
		Upper tracheal	Yes
		Subglottic + upper tracheal	No
Hood's stent	1 (4.17)	Glottis	Yes
Serial dilatation	1 (4.17)	Suprastomal	Yes

ACS: Anterior cricoid split; CCI: Costal cartilage interposition; SR: Swiss roll insertion

Table 4: Fourteen patients underwent multiple procedures out of which nine were eventually decannulated, four had persisting tracheostomy including two who were lost to follow-up. One patient with supraglottic stenosis did not require a tracheostomy

Sn	*Site*	*Primary procedure*	*Subsequent procedures*						*Outcome*
1	Supra	L	L	L	L	L	L		No preop trach
2	G + SG	ACS, CCI, SR	L	D	L				De
3	G + SG	Hood stent	SR	L	SR	SR	D	ACS, CCI,	De
4	G + SG	L, ACS, CCI	L	L	L	L		SR	De
5	SG	ACS, CCI, SR	D	T					Trach (no follow-up)
6	SG	ACS, CCI, SR	L	D					De
7	SG + T	ACS, CCI, SR	SR	D	SR				De
8	SG + T	L,T-t	ACS, CCI, SR	D	D	L	D	D	Trach
9	SG + T	L,T-t	T-t						De
10	T	L,T-t	D	T-t					De
11	T	L,T-t	TS	D	L,D	D	Trach	L	Trach
12	T	L,T-t	T-t	D	D				De
13	T	L,T-t	D	T-t	D				Trach (no follow-up)
14	T	L,T-t	L	L					De

Supra: Supraglottis; G: Glottis; SG: Subglottis; T: Tracheal; L: Laser; D: Dilatation; ACS: Anterior cricoid split; CCI: Costal cartilage interposition; SR: Swiss roll; T-t: Montgomery T-tube; TS: Tracheal stent; Trach: Tracheostomy; De: Decannulation

DISCUSSION

Congenital laryngeal stenosis is differentiated from acquired stenosis by the absence of history of instrumentation or trauma (typically from endotracheal intubation).[1] Laryngotracheal stenosis may be associated with tracheoesophageal fistula, esophageal atresia, urinary tract abnormalities, limb defects, encephalocele, horseshoe kidney and low set ears. Children with trisomy 21 have high incidence of laryngotracheal stenosis.[2,3] Since, the advent of prolonged mechanical ventilation in cases of premature neonates with immature pulmonary development, the incidence of acquired stenosis is higher than the congenital form. Other acquired causes of stenosis can be post tracheostomy, blunt or penetrating trauma, laryngeal surgery or rarely a laryngeal tumor. Among all the acquired cases, postintubational and post-tracheostomy stenosis accounts for the bulk of the cases with the former having slightly higher incidence than post-tracheostomy. Laryngotracheal stenosis continues to occur in approximately 1% of pediatric patients after intubation.[4]

Table 5: The final outcome of the patients

Outcome	*Number of patients (%)*
No preop tracheostomy	1 (4.17%)
Decannulation	18 (75%)
Persisting tracheostomy	5 (20.83%) including 2 (8.33%) lost to follow-up

The most common supraglottic anomaly is a supraglottic web and most are managed by laserization (Fig. 1).

Glottic webs always should be considered in children with a congenital history of hoarseness and recurrent croup presenting before 6 months of age. Cohen[5] classified them into four groups depending upon the lumen occupied by the web. Type I: occupying up to 35% of glottis, type II: 35 to 50% of glottis, type III: 50 to 75% of glottis and type IV: more than 75% of glottis (Fig. 2).

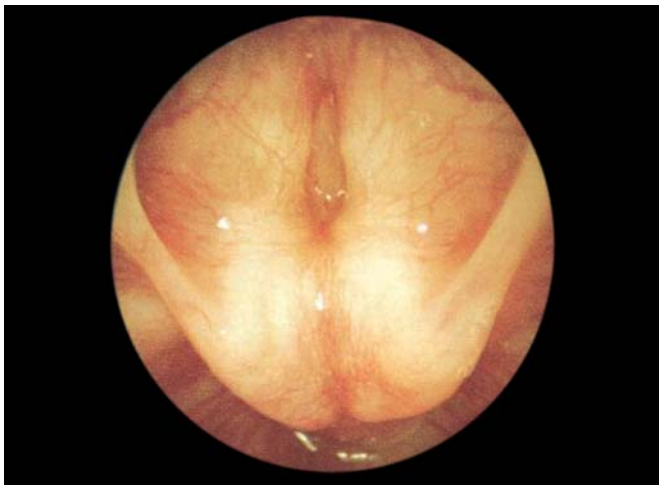


Fig. 1: Supraglottic stenosis

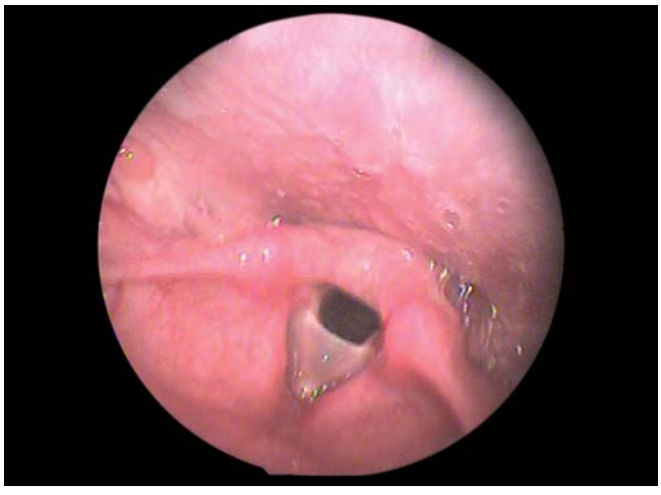


Fig. 2: Laryngeal web

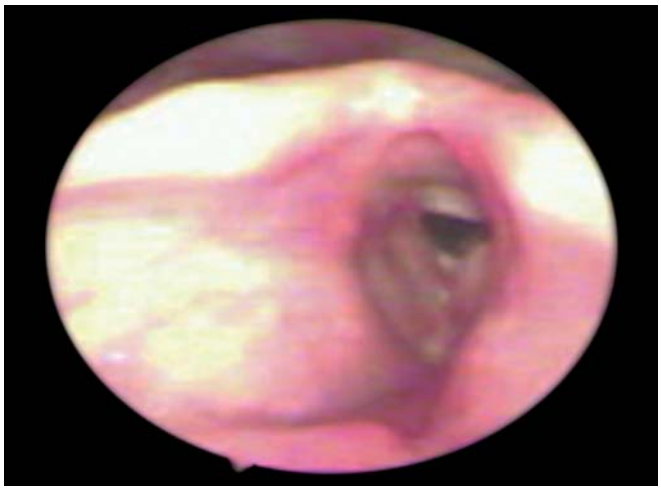


Fig. 3: Subglottic stenosis

Acquired glottic stenosis may result from trauma such as caustic injury, blunt or penetrating injury or may result due to repeated use of lasers, laryngeal surgery, chronic inflammation, infection or laryngeal neoplasm, e.g. papilloma.

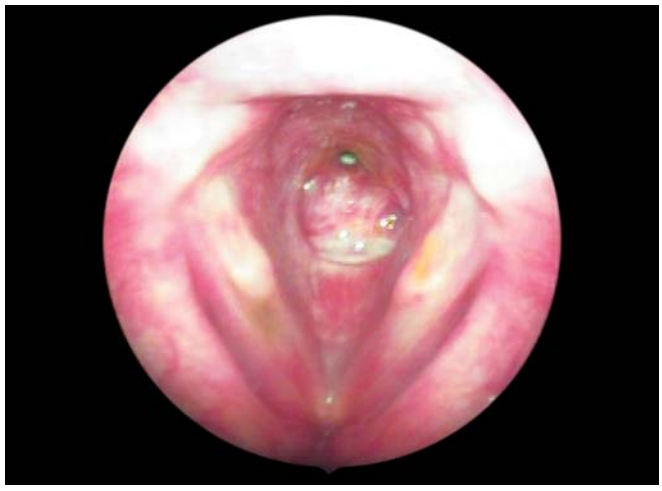


Fig. 4: Tracheal stenosis

Subglottic stenosis is the third most common cause of congenital laryngeal anomaly after laryngomalacia and vocal fold paralysis. A subglottic diameter of 4 mm or less in a full term infant or 3 mm or less in a premature infant is considered narrow and consistent with a diagnosis of subglottic stenosis (Fig. 3).[2,6]

Anand et al (1992) and Gady Har et al[7] (1993) found endotracheal intubation and emergency tracheostomy as the commonest causes for tracheal stenosis. Intubation beyond 7 to 10 days increases the risk of stenosis (Fig. 4).

Most treatment decisions are based on the severity of a stenosis, anatomical characteristics of a stenosis, and the degree of inflammation rather than on etiology (congenital *vs* acquired). Various degrees of tracheal stenosis may occur in infants, causing mild-to-severe airway symptomatology. Each pediatric patient must undergo a comprehensive assessment to formulate an individualized treatment plan. Eliciting clinical history forms a very important base for management of laryngotracheal stenosis patients. It is complimented by a detailed and comprehensive clinical examination and investigation. Other associated anomalies must be excluded. Investigations are done to assess the site of stenosis, degree of stenosis, length of stenosis and to rule out concurrent pathologies. Radiologic evaluation plays a role in most patients. CT scans can provide anatomic information, such as the length of a stenosis; however, they cannot differentiate between the true lumen and overlying secretions, which introduce inaccuracies in the presence of blood, mucus and crusting. MRI scans have a slight edge over CT scans in showing the soft tissue details of the stenosis. The mainstay for evaluating laryngeal stenosis is direct laryngotracheoscopy (Fig. 5).

Laryngopharyngeal reflux exacerbates laryngeal stenosis and may compromise surgical repair leading to treatment failure. It is treated with H2-blockers and proton pump

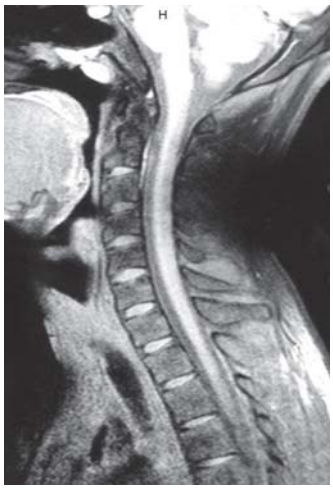


Fig. 5: Radioimaging: Glottic and subglottic stenosis

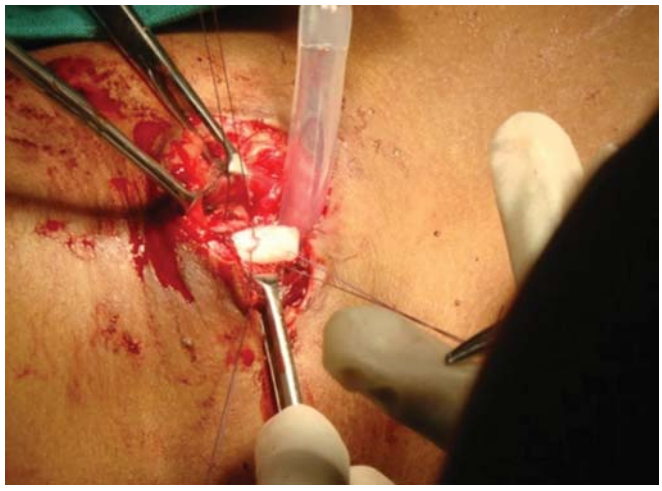


Fig. 6: Costal cartilage interposition and Montgomery T-tube insertion

inhibitors. Spirometry and pulmonary function tests are the other investigations required.

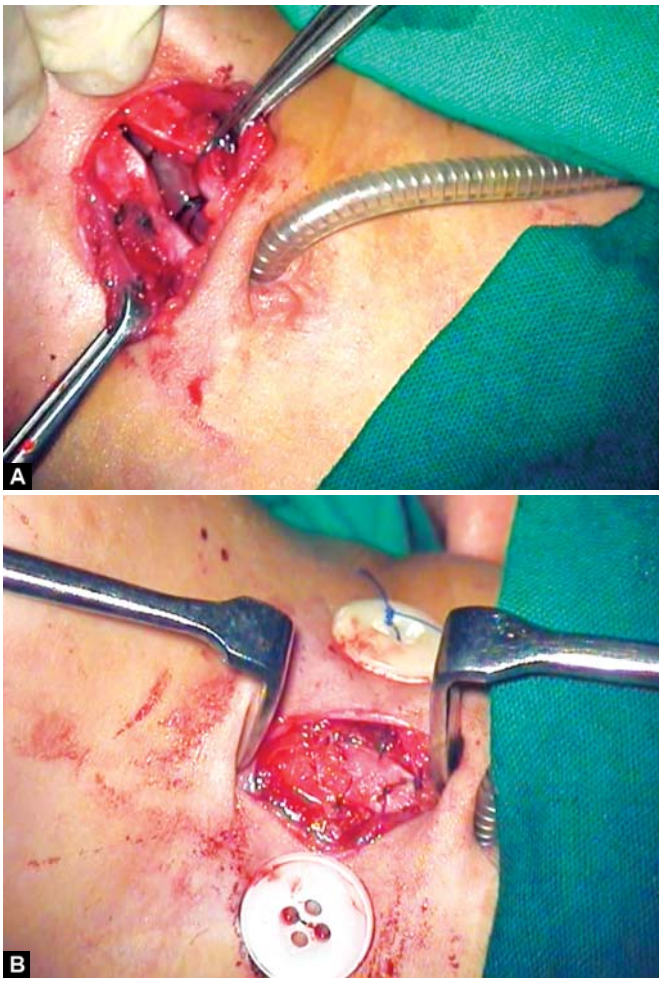
Surgery for laryngotracheal stenosis requires a rational approach. The site and extent of the stenosis must be ascertained prior to deciding on the surgical approach. External and endoscopic approaches have been described in the management of laryngotracheal stenosis. Supraglottic web can be very effectively treated by CO_2, KTP Nd:YAG laser, etc. Other modalities employed for treating supraglottic stenosis are—dilatation, laryngeal widening procedures. Small glottis webs not causing significant morbidity to the patient can be left alone. Webs occupying >50% of glottis need treatment because of potential airway compromise with a concurrent URTI and webs causing respiratory distress or significant aphonia. Glottic stenosis can be treated by - laserization of the stenosis, dilatation of the stenosed segments, repair by laryngofissure or by insertion of stents such as Hood's stent. McNaught glottic tantalum or Silastic Keel has proved their efficiency in dealing with anterior glottic stenosis, however, keels are reserved for adult age group. Thin posterior glottic stenosis can be treated by endoscopic laser excision. Other techniques that can be used are dilatation procedures, microtrap door flap technique or by laryngofissure and mucosal advancement flap. Severe scarring and arytenoid fixation can be treated by division of scar and posterior cricoid split with cartilage augmentation. Endoscopic laser resection is also employed as an adjunctive procedure to augment open repair. Treatment of subglottic stenosis depends upon the severity and length of stenosis and the patient's general medical condition, swallowing ability, age and weight. Nonoperative care is often sufficient in Myer-Cotton grade l stenosis as the larynx likely reaches adequate size with growth. Stenosis involving more than 50% lumen obstruction usually requires some sort of surgical intervention. External/combined external and endoscopic approaches are used. Laryngotracheal reconstruction with cartilage grafting with/without stenting with T-tube/silastic swiss roll insertion/Hood laryngeal stent is useful. Anterior laryngotracheal decompression (cricoid split) was used in newborns that have congenital subglottic stenosis. This procedure was described by Rethi in 1956 and modified by Cotton and Seid in 1980.[8] This procedure is usually combined with anterior cartilage graft usually as a single-stage procedure. Many autogenous cartilaginous grafts have been described and these include costal, auricular, nasal, septal, hyoid and thyroid ala. The success rate is slightly higher for this procedure as compared to cricoid split alone (88 *vs* 83%) with a lower complication rate (0 *vs* 9%) (Fig. 6).

Stern and Cotton evaluated the use of Montgomery T-tube in children from 2 weeks to 3 years and did not observe increased granulation tissue formation and concluded that the T-tube is safe to use in children.

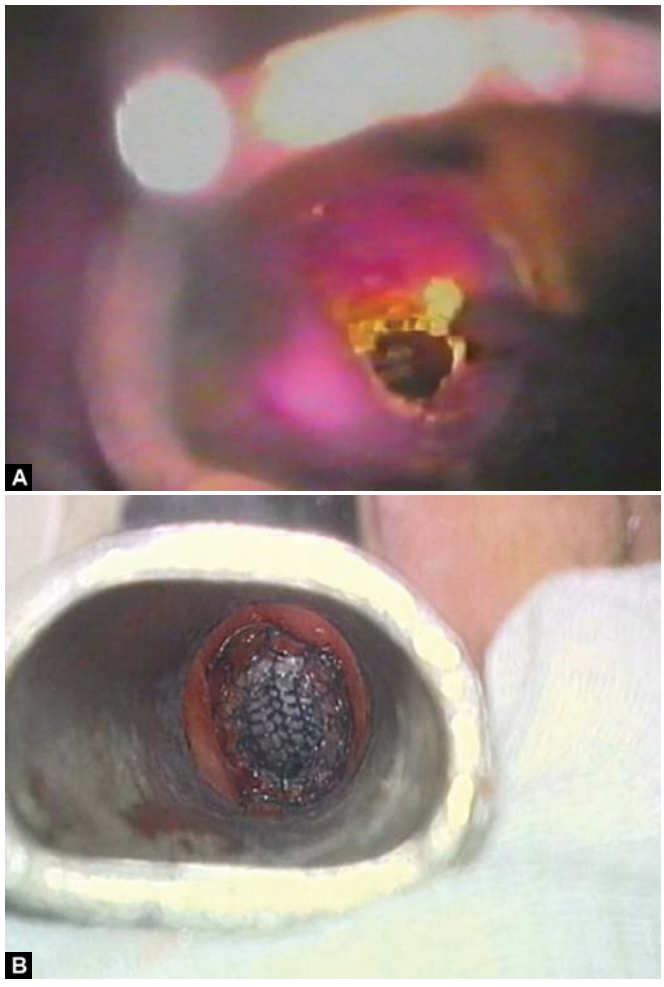
Silastic sheet can be rolled and used to stent the larynx (Figs 7A and B).

Hood laryngeal stents provide soft, solid, conforming support for use in glottic and subglottic stenosis. The stent is held in place by silicone suture buttons. They are indicated in older children. The stent should stay in place for 4 to 8 weeks or more depending on the extent of the injury and repair (Fig. 8).

Traditionally, tracheal stenosis has been managed with a variety of external surgical procedures—excision of the stenotic segment with reanastomosis, incision of the segment with reconstruction using a variety of local muscular or tracheal flaps or autogenous costal cartilage grafts or Montgomery T-tube insertion. Tracheal stenosis can also



Figs 7A and B: (A) Swiss roll insertion, (B) swiss roll secured in place with buttons



Figs 9A and B: (A) Laserization of tracheal stenosis, (B) tracheal stent *in situ*

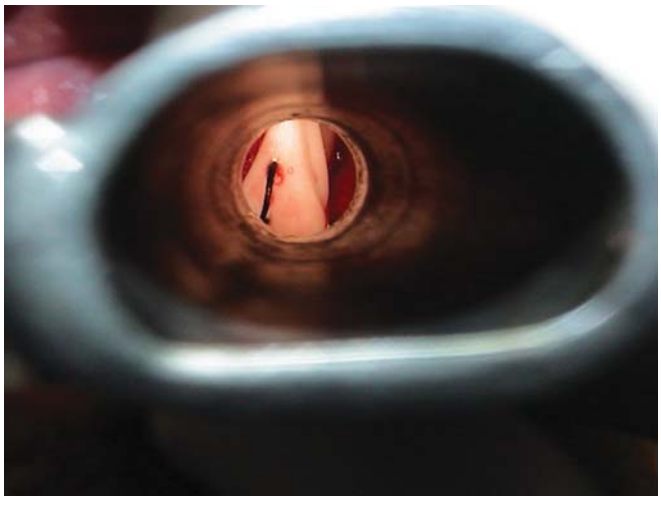


Fig. 8: Direct laryngoscopy to confirm position of Hood's stent

be managed endoscopically and lasers have proved to be useful in the management of tracheal stenosis. After laserization of the stenosed segment, self retaining indwelling nitinol tracheal stent can be used in older children (Figs 9A and B).

Revision Surgery for Pediatric Laryngotracheal Stenosis

Operative and perioperative risks that can lead to surgical failure[9] include graft necrosis, infection, steroids high-dose and long-term use, inadequate muscle coverage, dehiscence, hidden airway lesions—laryngomalacia, vocal cord dysfunction, tracheomalacia, gastroesophageal reflux disease and obstructive sleep apnea. It is advisable to wait for atleast 6 months before attempting another major open airway operation.

CONCLUSION

The management of pediatric laryngotracheal stenosis continues to pose challenges to the otolaryngologist. There are several treatment options for the management of pediatric laryngotracheal stenosis. The treatment must be individualized based on several factors, such as the site, severity and etiology of stenosis. No single procedure is appropriate for all situations. Meticulous surgery with minimal trauma is imperative. Although there may be a need

for several surgical procedures before the patient can be eventually decannulated, the overall outcomes are extremely gratifying. Advances in biomedical engineering, better adjuvant therapies and improved antigrowth factors may also aid in the prevention of restenosis and scarring after surgical repair.

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