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Effect of Supraglottoplasty on congenital tracheomalacia

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ABSTRACT

Purpose: Evaluation of the effect of supraglottoplasty on co-existing tracheomalacia in pediatric patients with congenital laryngotracheomalacia to establish the venturi effect in vivo.
Methods: A prospective interventional study was conducted in a tertiary care hospital from 2020 to 2024. All consecutive pediatric patients undergoing supraglottoplasty for congenital laryngomalacia, with co-existing tracheomalacia on pre-operative bronchoscopic assessment were included and were assessed for change in severity of tracheomalacia by bronchoscopy and clinical parameters post-surgery as a comparison to the pre-operative period.
Results: Twenty-eight patients including sixteen boys and twelve girls aged 1–30 months underwent supraglottoplasty. Statistically significant reduction in tracheal collapse was noted in all twenty-eight patients post-surgery on bronchoscopic evaluation (mean reduction by 41.45 ± 9.72 %). Clinically significant improvement was seen in terms of severity of stridor, frequency of hospitalization, apparent life-threatening events, z score for weight for age and parental perception of resolution of symptoms of their ward.
Conclusion: Supraglottoplasty for correction of laryngomalacia results in significant improvement in co-existing tracheomalacia. Associated medical comorbidities were not found to affect the positive outcome. Supraglottoplasty being a simple surgery with insignificant complication rate and very high success rate may be considered as the first line of surgical intervention in severely symptomatic pediatric patients with laryngotracheomalacia.

1. Introduction

Laryngomalacia (LM) is the most common laryngeal anomaly in infants with an estimated incidence of 1 in 2100–2600 general population [1]. In LM, the supraglottic structures collapse into the airway during the inspiratory phase of respiration which causes obstruction. The condition is associated with co-existing tracheomalacia (TM) in 30 % of cases [2]. These patients with laryngotracheomalacia (LTM) often present with stridor, which may be accompanied by poor feeding, failure to gain weight, apparent life-threatening events (ALTE) of apnea or cyanosis necessitating hospitalization. The condition, like LM, is usually self-resolving, and can be managed conservatively in most of the patients.

Endoscopic supraglottoplasty (SGP), a simple and effective procedure, is currently the mainstay surgical procedure for cases with severe LM. Unlike LM, intervention for severe TM include continuous positive airway pressure (CPAP), tracheostomy, stenting or aortopexy etc. which have comparatively higher rates of complications and have a morbid impact on mental and social health of the patient and caregivers. Considering the high morbidity of these procedures, currently surgical management is reserved for severe cases.

Symptomatic patients with severe TM (more than 50 % dynamic collapse of the tracheal lumen) with LM (of any severity) are considered for CPAP or surgical options for TM which are invariably invasive, morbid and technically demanding procedures. In some studies, on SGP in patients with severe LM, children with co-existing TM have been

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reported to improve after SGP [3,4]. However, the severity of pre-operative tracheomalacia and the post-SGP effect on the TM has not been analyzed in those case reports. The present study aims to evaluate the effect of supraglottoplasty on tracheomalacia in patients with coexisting laryngomalacia.

2. Material and methods

A prospective observational cohort study was commenced in a tertiary care hospital from July 2020 in the department of Otorhinolaryngology and Pediatrics following approval from the Institute Ethics Committee (IECPG-344). The aim of the study was to assess the outcomes of supraglottoplasty in patients with laryngomalacia with severe symptoms. Patients presenting with the symptoms of airway malacia were assessed carefully by a team of otolaryngologists and pediatricians. Awake fiber optic laryngoscopy to evaluate airway pathologies was performed and the type as well as severity of laryngomalacia as per Groningen Laryngomalacia Classification system was noted down [5]. Patients with mild or resolving symptoms were kept under close follow-up with regular monitoring for weight gain or worsening of symptoms. Those with moderate to severe symptoms, atypical features and those fulfilling below mentioned clinical features for surgical intervention underwent dynamic flexible bronchoscopy under sedation in the department of Pediatrics for complete airway assessment. Additionally, in patients with suspected aspiration, recurrent lower respiratory tract infections, Fiberoptic Endoscopic Evaluation of Swallowing (FEES) was done. Bronchoalveolar lavage sampling during bronchoscopy, videofluoroscopic swallowing study, gastroesophageal reflux scan, and chest imaging were considered as required on individual case-need basis. Based on our preliminary results, a single arm prospective intervention study was carried out from June 2023 to June 2024. (CTRI/20223/05/067,196). Patients were assessed for recruitment under the study with the inclusion criteria as follows: 1) Age group of 1 month–5 years undergoing supraglottoplasty; (2) Endoscopic confirmation of co-existing laryngomalacia and tracheomalacia with or without bronchomalacia; (3) More than 50 % collapse in the trachea during expiration/inspiration on dynamic flexible bronchoscopy; (4) The clinical indications for supraglottoplasty included presence of at least two of the following clinical features-failure to thrive, three or more hospitalizations for airway malacia related complications, severe respiratory distress necessitating endotracheal intubation, episodes of bradycardia/cyanosis/apnea due to airway malacia, oxygen saturation less than 90 % at room air without oxygen or CPAP support, previously tracheostomized for airway malacia with failed attempts at decannulation, presence of pulmonary artery hypertension secondary to airway malacia. Patients with airway narrowing secondary to extrinsic compression were excluded from study. Additionally, patients with severe cardiac pathology not compatible with life, poor pulmonary reserve and coexisting severe neurological comorbidities were considered for upfront tracheostomy.

All enrolled children underwent supraglottoplasty according to the type of LM after informed written parental consent. Type 1 SGP included reduction of redundant arytenoid mucosa. Type 2 SGP included releasing aryepiglottic folds maintaining integrity of pharyngoepiglottic folds while Type 3 SGP included epiglottopexy. Postoperatively, all children were kept intubated for 48–72 h and were closely monitored following extubation in ICU. For incomplete response, repeat bronchoscopy was performed and revision SGP or tracheostomy was considered. All patients were actively followed up and repeat bronchoscopy was performed at 4–6 weeks duration post operative to document severity of residual airway malacia.

The outcomes which were specifically compared with the pre-operative values to analyze the effect of supraglottoplasty are change in severity of stridor, TM severity (mean of the maximum percentage of collapse in tracheal lumen as seen in the bronchoscopic video and reported by two different observers), need for hospitalization due to LTM related events, frequency of apparent life threatening episodes (bradycardia/apnoea/cyanosis), weight gain (z score for weight for age), and parental perception of resolution of symptoms of their ward.

3. Results

A total of 118 patients with airway malacia were screened and 28 patients with synchronous airway malacia with severe symptoms fulfilling the inclusion criteria were included in this study. Of these 28 patients, 18 underwent surgery as part of observational study, and 10 patients were recruited as part of single arm interventional study. Patients with laryngomalacia with severe symptoms who underwent supraglottoplasty from 2020 to 23 but without significant tracheomalacia were excluded. Among the recruited patients, 16 (57 %) were boys and 9 (32 %) were preterm born. Majority of patients (90 %) were symptomatic by one month of age with stridor being the most frequent symptom (71 %). Twelve (44 %) patients had a history of respiratory distress at birth requiring oxygen support and eight of them required transient CPAP support (Table 1).

Supraglottoplasty was performed in 28 patients, using cold instruments for type 2 SGP and co-ablator for type 1 and 3 SGP. CO_2 laser was used in only tracheostomized patients due to unavailability of adequate size laser compatible endotracheal tubes. Type 2 LM was present in all the patients necessitating type 2 SGP in all the patients. Additionally Type 1 and type 3 SGP was performed in 53 % and 21 % of patients respectively. One patient required all three types of SGP in a single sitting. Two patients developed post intubation subglottic stenosis and required additional surgery for dilatation of stenosis. Revision SGP was performed in three patients (Table 2).

All the patients have been followed up till October 2024 (except one who succumbed to non-airway related pathology after follow-up for 4 weeks post-operatively). The median duration of follow-up is 27 months (interquartile range 4–47).

All the patients had preoperative TM with reduction in the luminal area by a minimum of 60 % at the narrowest cross section. The tracheal collapse was anteroposterior type, more prominent during end expiratory-inspiratory phase and involving complete tracheal length including both intra-thoracic and extra-thoracic trachea in all the patients. On bronchoscopic re-evaluation, the postoperative tracheal collapse was reduced to 50 % or less in all the patients. The differences between the pre- and post-SGP tracheal collapse ranged between 65 % and 30 % (Fig. 1). Mean preoperative vs postoperative tracheal collapse was 74.4 $\pm$ 11.93 % and 32.3 $\pm$ 8.67 % respectively with mean reduction of 41.45 $\pm$ 9.722 % which was statistically significant (p value $<$ 0.001). Interestingly, the response in TM was not limited to only in cases with severe laryngomalacia, and was also seen in cases with mild LM with disproportionately severe TM (Fig. 2).

Resolution of acute life-threatening episodes occurred in all 14 patients including those ten, where episodes were severe enough necessitating intervention in the form of either intubation or bag and mask ventilation. Intervention reduced the frequency of hospitalizations due to LTM related causes as only three patients needed hospitalization against 20 during the follow-up period. Two of these patients had persistent aspiration and one was admitted for an episode of LRTI.

Resolution of stridor was seen in all the patients, which was complete in 80 % of the cases, and partial in the remaining 20 % of the cases. 24 parents reported complete resolution of symptoms (Table 3). Out of the four cases whose parents perceived the response to surgical intervention as partial, one had incomplete response to SGP requiring tracheostomy, another had persistent wheeze due to reactive airway and two had persistent aspiration. Positive effect of surgical intervention was seen on weight gain, as the mean weight for age (z score) significantly increased from preoperative value of -2.52 to -2.07 post operatively (p value 0.002) (Fig. 3).

Table 1
Detailed symptomatology of all recruited patients.

Sr. no	Age at symptom onset	Term/ Preterm	Complaints	Co-existing pathologies
1.	birth	Preterm (35 weeks)	Recurrent apnea requiring intubation, stridor	Retrognathia, glossoptosis, microcephaly
2.	4 weeks	Term	Tracheostomized for stridor following extubation after cardiothoracic surgery, failed decannulation attempts	ACHD (4 mm PDA/ moderate VSD) post ligation
3.	4 weeks	Preterm (34 weeks)	Recurrent LRTI, aspiration	None
4.	Day 2	Preterm (36 weeks)	persistent CPAP requirement, recurrent LRTI	Microcephaly
5.	6 months	Term	Cyanotic episodes, recurrent LRTI	Reactive airway disease
6.	2 months	Term	Tracheostomized for stridor, failed decannulation attempts	Resolved subglottic hemangioma
7.	5 weeks	Term	Stridor, apneic episodes, cyanosis	Retrognathia, glossoptosis
8.	Day 2	Term	Stridor, apneic episodes	None
9.	Birth	Post term	Stridor, apneic episodes	None
10.	Birth	Preterm (36 weeks)	Stridor, aspiration, recurrent LRTI, apneic episodes	Complex seizure
11.	5 months	Term	Stridor cyanotic episodes, 1 episode of LRTI	none
12.	18 months	Term	Stridor, OSA	None
13.	birth	Pre term (36 weeks)	Persistent respiratory distress with apnea and cyanosis	Noonan syndrome
14.	Birth	Term	Persistent CPAP requirement	None
15.	birth	Preterm (26 weeks)	Stridor, persistent CPAP requirement, respiratory distress requiring multiple intubation	Grade 3 bronchopulmonary dysplasia
16.	Birth	Preterm (28 weeks)	Persistent CPAP requirement	None
17.	4 weeks	Term	Stridor, recurrent LRTI, aspiration	None
18.	birth	Term	Aspiration, recurrent LRTI	None
19	birth	term	Recurrent LRTI	microcephaly/ megaesophagus with esophageal motility disorder/ hypervitaminosis D
20	birth	preterm	Persistent CPAP requirement	Bronchopulmonary dysplasia
21	4 weeks	term	Persistent LRTI	none
22	2 weeks	term	Recurrent LRTI	none
23	birth	preterm	Tracheostomised for stridor, failed decannulation	Severe HIE
24	4 weeks	term	Recurrent LRTI, tracheostomised - failed decannulation	Grade 1 subglottic stenosis
25	birth	term	Recurrent apnea and cyanosis	none
26	2 weeks	term	Recurrent apnea and cyanosis	none
27	2 weeks	term	Severe respiratory distress requiring CPAP support	none
28	birth	term	Recurrent LRTI	none

Abbreviations: ACHD, acynotic congenital heart disease; VSD, ventricular septal defect; CPAP Continuous positive airway pressure; OSA obstructive sleep apnea. PDA- Patent Ductus Arteriosus, HIE- Hypoxic Ischemic Encephalopathy, LRTI- Lower Respiratory Tract Infcction.

Table 2
Table depicting age at surgery, type of surgery and other coexisting airway pathology.

Case	Age (in months)	Intervention	Synchronous airway pathology
1.	5	Type 2 + 3 SGP	Severe PM, BM, glossoptosis
2.	24	Type 2 + 3 SGP[a]	Left TVC palsy; BM
3.	5	Type 2 + 1 SGP	Grade 1 cleft. BM
4.	2	Type 2 SGP	Grade 1 cleft, BM
5.	11	Type 2 SGP	BM
6.	24	Type 2 SGP[a] supra stomal ledge removal	Suprastomal ledge
7.	2	Type 2 + 3 SGP	Glossoptosis
8.	1	1. Type 2 SGP 2. Type 1 SGP	Grade 1 laryngeal cleft
9.	2	Type 2 + 1 SGP	–
10	3	Type 2 + 3 SGP	–
11	5	1 Type 2 + 3 SGP 2 Laryngeal cleft repair	Grade 1 laryngeal cleft
12	24	Type 2 SGP retropharyngeal abscess drainage	Chronic retropharyngeal abscess
13	1.5	Type 2 + 1 SGP	Grade 1 laryngeal cleft
14	1	Type 2 + 1 SGP	BM
15	1	Type 2 + 1 SGP	BM
16	2	Type 3 + 2 +1 SGP	–
17	15	Type 2 + 1 SGP	–
18	5	Cleft repair, Type 2 SGP	Grade 2 laryngeal cleft
19	6	Type 2 + 1 SGP	
20	2	Type 2 + 1 SGP	
21	3	Type 2 + 1 SGP	BM
22	6	Type 2 + 1 SGP	
23	1	Type 2 + 1 SGP	
24	3	Type 2 + 1 SGP + balloon dilation of subglottic stenosis	GRADE 1 Subglottic stenosis
25	3	Type 2 + 1 SGP Revision type 1	
26	6	Type 2 + 1 SGP	
27	3	Type 2 + 3 SGP	BM
28	2	Type 2 SGP + cleft repair	Grade 2 laryngeal cleft

Type 1 SGP- Arytenoid reduction; type 2 SGP- Aryepiglottoplasty; type 3 SGP- Epiglottopexy.
Abbreviations: BM, bronchomalacia; TVC, true vocal cords; PM, pharyngomalacia.
[a] CO_2 LASER assisted.

4. Discussion

This study describes the positive effect of relieving upper airway obstruction on downstream airway collapse in the form of supra-glottoplasty on tracheomalacia in twenty-eight patients. Clinically significant improvement was seen in all 28 patients in terms of severity of stridor, frequency of hospitalization, apparent life-threatening events, z score for weight for age and parental perception of resolution of symptoms of their ward.

The effect of SGP on LM has been well established in the literature. Several retrospective studies, have correlated the presence of

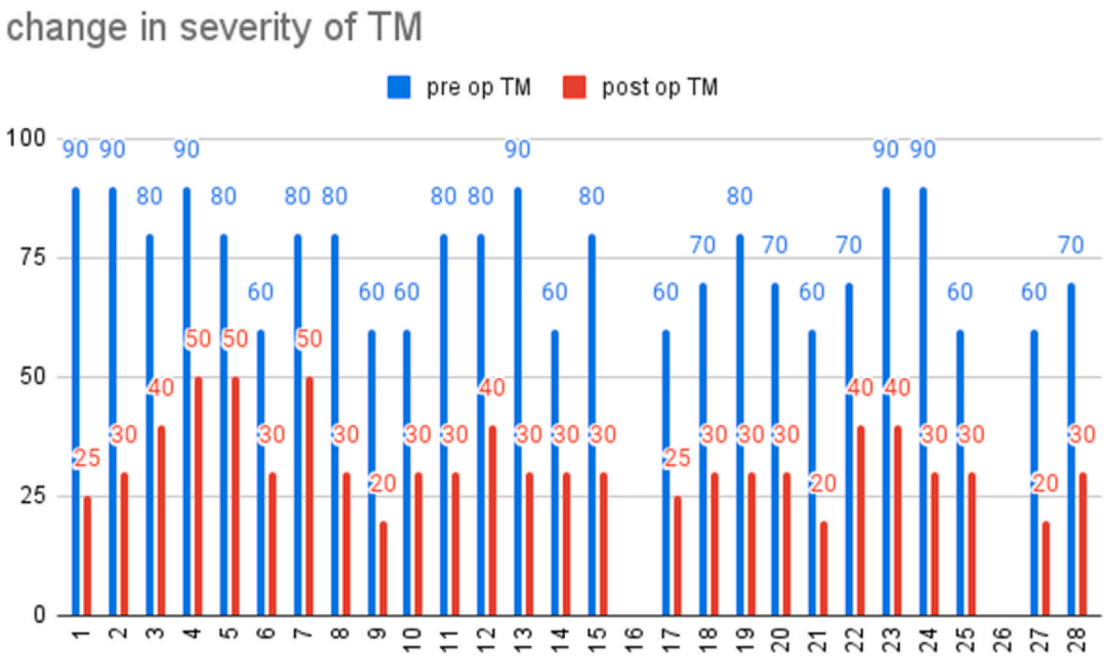

Fig. 1. Bar graphic representation of change in severity of tracheal collapse, pre-operatively and 4-6-weeks post intervention (maximum percentage of collapse in the area of cross section).
Post operative bronchoscopy could not be done for 2 patients - case 16 and 26.

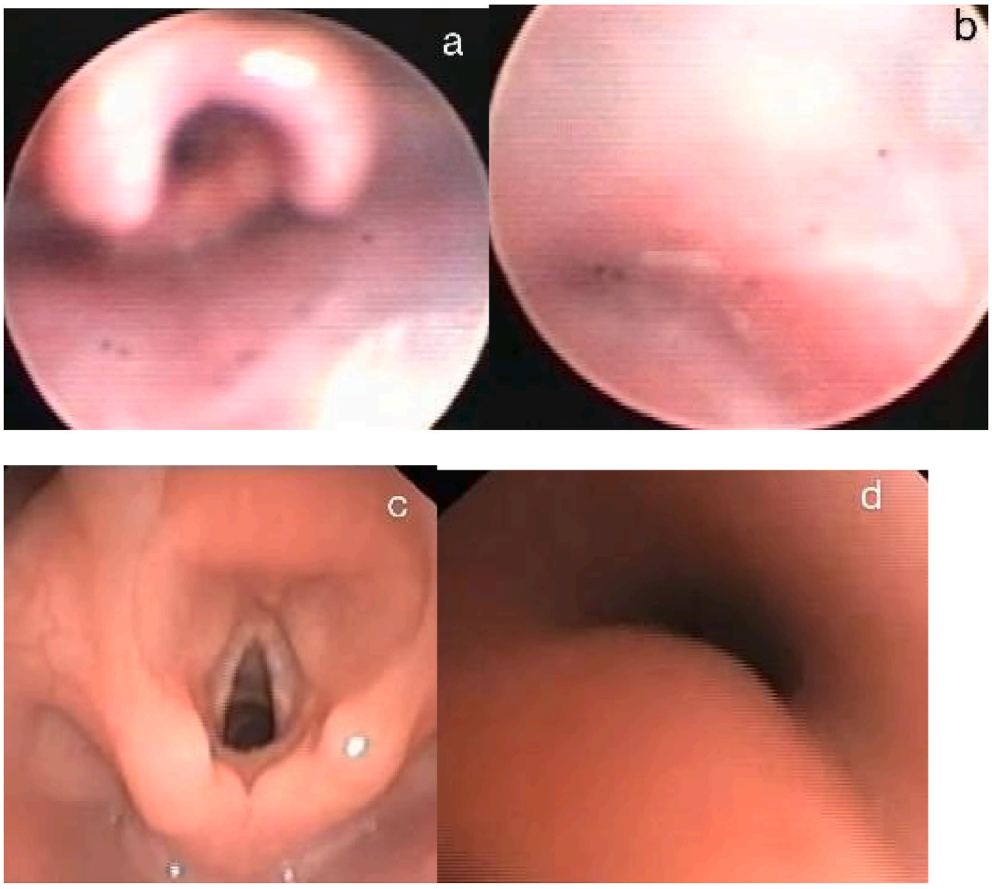

Fig. 2. Preoperative and postoperative laryngotracheomalacia (case no.5)– a-Pre-operative mild type 2 laryngomalacia, b-severe tracheomalacia (80 % collapse), c-Postoperative resolved laryngomalacia, d-improved tracheomalacia (50 % collapse).

Table 3
Comparison of stridor characteristics and parental perception pre and post intervention.

S. no	Preoperative stridor	Postoperative stridor	Parental perception of symptomatic relief
1.	Biphasic	Inspiratory[a]	Partial
2.	Inspiratory[b]	No	Complete
3.	No	No	Complete
4.	Biphasic	No	Complete
5.	Biphasic wheeze[c]	Biphasic wheeze	Partial
6.	No	No[d]	Complete
7.	Inspiratory	No	Complete
8.	Inspiratory	Inspiratory	Not available
9.	Inspiratory	None	Complete
10.	Inspiratory	None	Partial
11.	inspiratory	None	Complete
12	Inspiratory	None	Complete
13	None	None	Complete
14.	None	None	Complete
15	None	None	Complete
16	Inspiratory	None	Complete
17.	Inspiratory	None	Partial
18.	None	None	Complete
19	Biphasic	None	Complete
20	inspiratory	None	complete
21	none	None	complete
22	inspiratory	None	complete
23	inspiratory	None	complete
24	inspiratory	None	complete
25	inspiratory	None	complete
26	inspiratory	None	complete
27	inspiratory	None	complete
28	inspiratory	None	complete

[a] Required tracheostomy i/v/o CO_2 retention.
[b] During failed trial for decannulation.
[c] Associated reactive airway disease.
[d] Biphasic stridor after 1 week post operatively due to subglottic stenosis, stridor resolved post stenosis release.

synchronous airway malacia with low success of SGP for LM [6–8]. However, Dickson et al., in their retrospective review, found 3 patients with LTM with successful outcome post SGP [9]. Presence of tracheomalacia has been considered an indicator for more morbid procedures like aortopexy, tracheal stenting and tracheostomy [10–13]. We hypothesized that relieving obstruction at the laryngeal level would improve dynamic narrowing of the trachea based on Bernoulli's principle. Since instability in trachea is nonlinear (snap through instability) where a small change in pressure can result in collapse of trachea, widening of larynx through SGP in patients with LTM can result in decrease in airflow velocity thus increasing intraluminal pressure downstream [14]. Also, as the breathing and respiratory effort improves after SGP, an ease of expiratory effort leads to reduced expiratory collapse. This is the first prospectively designed study documenting the effect of SGP on tracheomalacia. Our hypothesis was proved by the successful resolution of symptoms and avoidance of tracheostomy in 96.4 % of cases.

Failure rates of 4–70 % have been noted; and prematurity, presence of cardiac or neurological comorbidity, associated aspiration, GERD, synchronous airway lesions such as tracheomalacia, congenital abnormality, and micro/retrognathia have been described as predictors of negative outcomes for SGP [15–17]. In our series ten patients had hypoxic ischemic encephalopathy, two patients had cardiac comorbidity, all had associated tracheomalacia, nine patients had severe bronchomalacia, three had micrognathia with glossoptosis, and two were extremely preterm (born <28 weeks). Complete response was seen in all but one out of 28 patients amounting to a failure rate of 3.6 %. There were multiple factors for incomplete response in this case like prematurity, hypoxic ischemic encephalopathy, Pierre Robin sequence obstructing the upper airway, severe pharyngomalacia, severe GERD requiring feeding jejunostomy, and associated severe bronchomalacia. Revision surgery was performed for three patients, two patients were taken up for revision SGP for persistent LM while it was done as a precautionary measure for the other during dilation of subglottic stenosis. We could successfully decannulate 4 patients who were previously tracheostomized due to severe LTM.

According to the most accepted hypotheses, LM and TM are due to abnormal sensorimotor integration which resolves spontaneously by 12–18 months of age [18]. However, in our series, we encountered two patients with persistent severe LM with TM till the age of 24 months. In one of these cases, diversion of airflow through the tracheostomy might be postulated as the factor preventing resolution of LM. The former of these two, had left vocal fold paralysis consequent to cardiothoracic surgery and had TM which was primarily on the left side in the peristomal trachea. It is possible that loss of trachealis muscle tone secondary to recurrent laryngeal nerve injury was an aggravating or

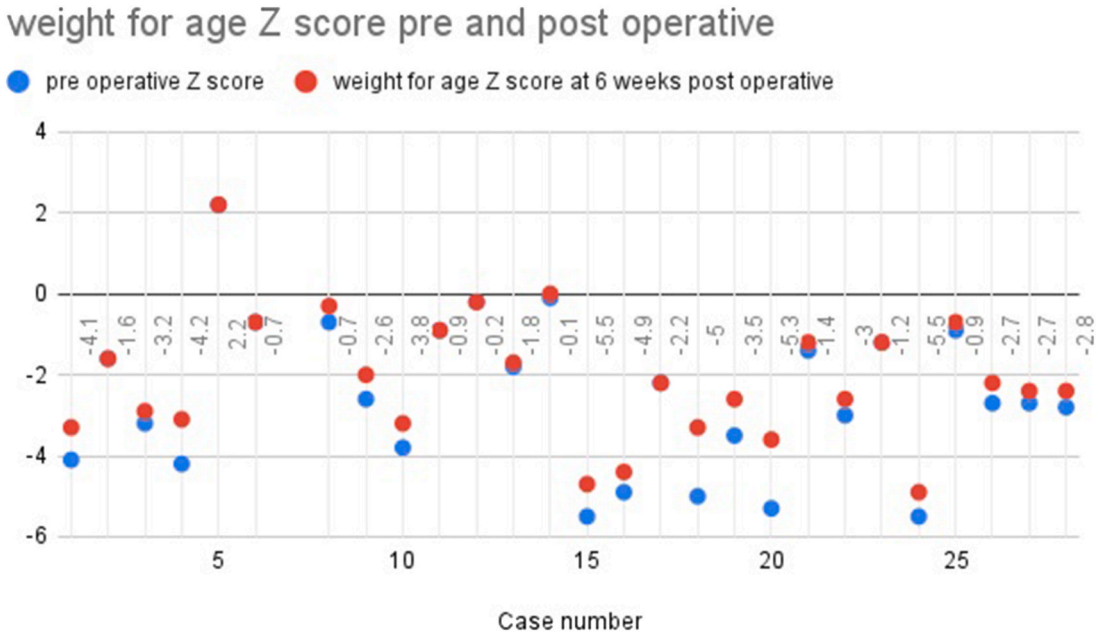

Fig. 3. Chart depicting the pre and post operative weight for age (Z score) for all patietnts.

perhaps an inciting factor, while for the other, tracheal collapse could be secondary to downstream negative pressure due to retropharyngeal abscess.

Of seven patients with co-existing laryngeal cleft, three had clinically significant aspiration. Aspiration was also noted in two patients without any co-existing laryngeal cleft. FEES study preoperatively showed posterior to anterior aspiration through inter-arytenoid cleft in the former three and lateral spillage over aryepiglottic folds in latter two patients. In the former group, aspiration responded to SGP by addressing negative pressure in one, while it was repaired for the other 2. In the latter group, in one; aspiration resolved spontaneously at 7 months of age (4 months post op) while the other patient had late onset aspiration (starting at 11 months of age) and resolved spontaneously at 2 years of age. One of the patients with grade 1 cleft had aspiration which became apparent post SGP, cleft was repaired at 3 months post operative. All three patients required long term nasogastric feeds till the resolution of aspiration. Ritcher et al., in their retrospective database, identified aspiration in 72 % of patients with LM, which resolved in 86 % of patients with SGP. Authors proposed the presence of GERD, cardiac disease and genetic or congenital anomaly to be predictors of negative outcome for improvement in aspiration following surgery [19]. In our experience, inter-arytenoid aspiration responded well to SGP while lateral spillage did not completely respond to surgery.

None of our patients had any postoperative complications related to the procedure itself. Thus, we found SGP as a safe procedure with significantly less morbidity and can be considered as the preferred intervention for correction of LTM. The procedure is effective irrespective of relative severity of LM and TM. Additionally, applying this hypothesis, we could avoid tracheostomy and its morbidities, decannulate and avoid the need for nasogastric feeding in 96 % of the patients.

The limitation of this study is the small sample size. Although failure to thrive and severe stridor are considered as indications for surgical intervention in LM, we followed the inclusion criteria stringently resulting in a small number of recruited patients during the study period. Another limitation is the inherent drawback of subjective evaluation of tracheal collapse on flexible dynamic bronchoscopic evaluation. We have tried addressing it by averaging the blinded evaluation of percentage reduction in tracheal cross section area by two independent observers.

Third limitation is lack of control group as laryngomalacia may improve spontaneously over the time. The surgery is indicated only in patients with severe symptoms which has been described in our stringent inclusion and exclusion criteria. Surgical intervention was strongly warranted in all the recruited cases and forming a control group was not possible in this study as it would have been considered unethical. However, if conducted in a very large number of such cases, patients whose parents deny surgical intervention might provide the scope for a control group. Additionally, symptomatic resolution in the immediate post-extubation period (which was 72 h to allow resolution of airway edema) cannot be attributed to spontaneous resolution of airway malacia.

5. Conclusion

Significant improvement in the degree of tracheomalacia despite presence of other airway and non-airway pathologies has been noted in our study. This result is very encouraging and indicates towards consideration of SGP as the possible first line intervention for patients of LTM with severe symptoms keeping other morbid and challenging procedures in reserve for unsuccessful cases and cases with co-existing severe neurologic or cardiopulmonary comorbidities affecting the life expectancy. Patients with mild laryngomalacia but disproportionately severe tracheomalacia also are likely to benefit from supraglottoplasty. Meticulous co-ordination between otolaryngologists and pediatricians is very crucial and stringent post-operative observation of the patients in ICU set up is vital to avoid unwanted events.

CRediT authorship contribution statement

Gaurav Goel: Writing – review & editing, Writing – original draft, Visualization, Methodology, Investigation, Funding acquisition, Conceptualization. **Prem Sagar:** Writing – review & editing, Supervision, Resources, Project administration, Methodology, Conceptualization. **Rajeev Kumar:** Writing – review & editing, Supervision, Conceptualization. **Kanaram Jat:** Writing – review & editing, Supervision, Resources, Conceptualization. **Sushil Kumar Kabra:** Writing – review & editing, Validation, Supervision, Resources, Conceptualization. **Vimi Rewari:** Writing – review & editing, Supervision, Conceptualization. **Rakesh Kumar:** Writing – review & editing, Supervision, Conceptualization. **Alok Thakar:** Writing – review & editing, Visualization, Validation, Supervision, Conceptualization.

Statements and declarations

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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