Prognostic Factors in the Management of Pediatric Subglottic Stenosis

Sarah Debs, BA[1]; Aasif A. Kazi, MD, PharmD[1]; Dustin Bastaich, BS[2]; Leroy Thacker, PhD[2]; Rajanya S. Petersson, MD, MS[1,3]

[1]Dept. of Otolaryngology – Head and Neck Surgery, Virginia Commonwealth University; Richmond, VA; [2]Department of Biostatistics, Virginia Commonwealth University, Richmond, VA; [3]Dept. of Otolaryngology – Head and Neck Surgery, Children's Hospital of Richmond at VCU

ABSTRACT

Objective: We report outcomes of conservative and invasive interventions in the management of pediatric subglottic stenosis (SGS).

Methods: A retrospective chart review of all pediatric patients with SGS, treated by a single surgeon, at a tertiary academic medical center from 2012 to 2020 was conducted. Variables recorded included patient demographics, initial grade of stenosis, gestational age, length of intubation, comorbidities as well as total number of interventions.

Results: A total of 47 patients were included in the study, of which 51% (24) were female. Laryngotracheal reconstruction (LTR) was performed in 48.9% (23) of patients. Fifteen patients did not have tracheostomy. Decannulation was achieved in 25 of 32 tracheostomized patients. When the patients were stratified based on LTR, there was a significant difference in gestational age (28.7 ± 5.36 vs 33.2 ± 6.13), initial grade of stenosis (2.3 ± 0.82 vs 1.6 ± 0.88), and total number of interventions (5.7 ± 2.8 vs 2.3 ± 1.5) between the two groups (p<0.05). There was no difference, however, in the length of intubation. Of the comorbidities recorded, only bronchopulmonary dysplasia was found to have a significant impact on the outcome.

Conclusion: SGS is a difficult condition to treat, often requiring multiple interventions including LTR. We propose an algorithm that may assist in the treatment of SGS patients with certain comorbidities to minimize interventions and maximize outcomes.

INTRODUCTION

- Subglottic stenosis (SGS) is narrowing of the airway that can occur between the level of the true vocal folds to the lower border of the cricoid cartilage (Figures 1and 2).

- Ninety percent of cases are acquired, typically from endotracheal intubation, and the remainder are congenital.

- Risk factors associated with SGS include premature gestational age, length of intubation, and endotracheal tube size.

- Associated comorbidities are acid reflux, pulmonary hypertension, bronchopulmonary dysplasia, chromosomal abnormalities, and chronic lung disease.

- Presentation can range from asymptomatic to, most commonly, biphasic stridor with increased work of breathing ± cough.

Figure 1. Myer-Cotton SGS Grading Scale

Grade	From	To
I	No Obstruction	50% Obstruction
II	51%	70%
III	71%	99%
IV	No detectable lumen	

Figure 2. Subglottic stenosis. A) Grade 1. B) Grade 2. C) Grade 3.
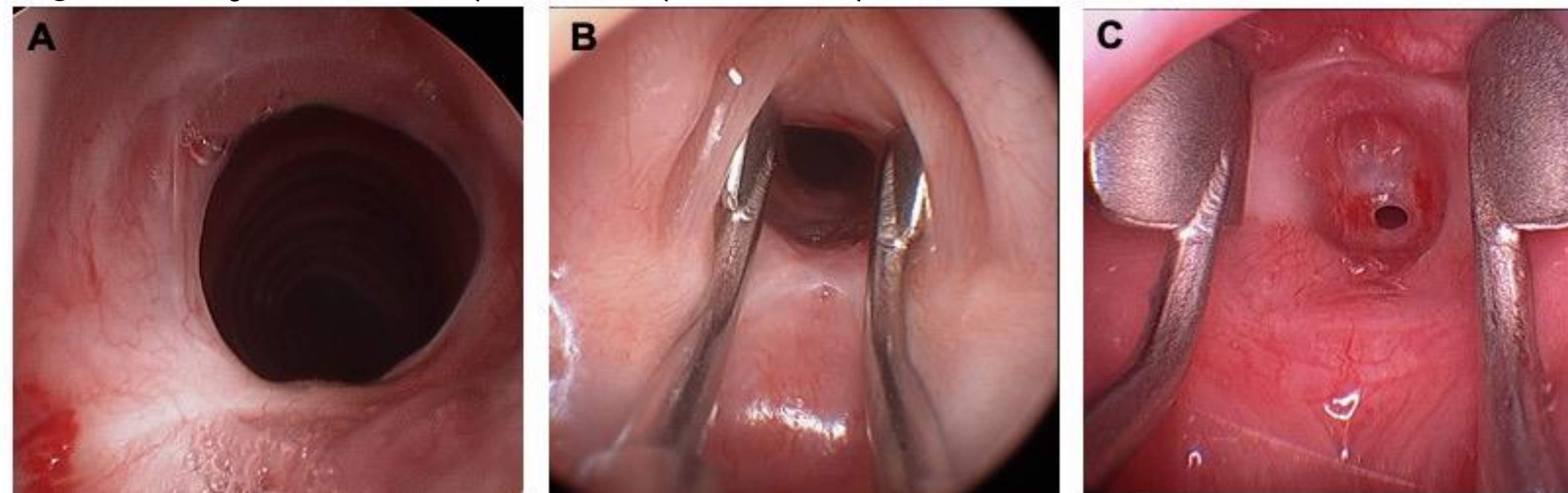
METHODS AND MATERIALS

- We conducted a retrospective chart review of all pediatric patients (<18 years old) with SGS who received management at our tertiary academic medical center from 2012 to 2020.

- Variables recorded included patient demographics, initial grade of stenosis, gestational age, length of intubation, comorbidities as well as total number of interventions.

- After staging with direct laryngoscopy, treatment modalities included balloon dilation, steroid injection, lysis of scar band with sickle knife, laser excision, or laryngotracheal reconstruction (LTR).

RESULTS

Patient Characteristics (Table 1)

- Forty-seven pediatric patients were treated for SGS and 23 (48.9%) had an LTR.

- The average intubation length was 61.5 days ± 57.2 days (range 0-180 days).

- The average initial grade of stenosis was 3.93 ± 2.81.

LTR vs. Non-LTR Patients (Table 2)

- Premature patients were almost 5 times more likely to have an LTR when compared to full-term patients (OR = 4.75, 95% CI = 1.24, 18.2). For each additional week of gestational age, the odds of LTR decreased by 12% (OR = 0.88, 95% CI = 0.79, 0.97).

- LTR patients were significantly different in the average initial grade of stenosis when compared to non-LTR patients (2.3 vs 1.6 respectively, p < 0.05).

- LTR is nearly 10 times more likely in a patient with an initial SGS grade of 3 compared to 1 and 6 times more likely than an initial SGS grade of 2.

- Decannulation rate was significantly different with 87% in the LTR group vs. 21% in the non-LTR group (p < 0.001).

- Length of intubation was not associated with higher rate of LTR.

Table 1. Patient Characteristics

Characteristics, n (%)	Overall	Laryngotracheal Reconstruction (N= 23)	No Reconstruction (N= 24)	p-value
Preemie (<37 weeks)				0.0184
Yes	31 (66.0%)	19 (82.6%)	12 (50.0%)	
No	16 (34.0%)	4 (17.4%)	12 (50.0%)	
Decannulation				<.0001
Yes	25 (53.2%)	20 (87.0%)	5 (20.8%)	
No	7 (14.9%)	1 (4.4%)	6 (25.0%)	
Never got a trach	15 (31.9%)	2 (8.7%)	13 (54.2%)	
Bronchopulmonary Dysplasia				0.0499
Yes	28 (59.6%)	17 (73.9%)	11 (45.8%)	
No	19 (40.4%)	6 (26.1%)	13 (54.2%)	
Comorbidities				0.4130
Yes	41 (87.2%)	21 (91.3%)	20 (83.3%)	
No	6 (12.8%)	2 (8.7%)	4 (16.7%)	
Initial Grade of Stenosis				0.0077
1	21 (44.7%)	5 (21.7%)	16 (66.7%)	
2	8 (17.0%)	6 (26.1%)	2 (8.3%)	
3	18 (38.3%)	12 (52.2%)	6 (25.0%)	
Gestational Age (Weeks) [b, c, d]	31.0 (6.14)	28.7 (5.36)	33.2 (6.13)	0.0102
Intubation Length (Days) [a, b, d]	61.5 (57.19)	78.9 (53.82)	48.4 (57.22)	0.0875
Number of Interventions [b, c]	3.9 (2.81)	5.7 (2.82)	2.3 (1.45)	<.0001

[a] Missing: n=5; [b] Continuous variables presented as mean (SD); [c] Imputed Missing: n=5; [d] pooled variance was used for t-test; [e] Satterwaite approximation was used for t-test

Table 2. Univariate Odds Ratios of LTR vs Non-LTR Patients

Risk Factors	Odds Ratio	95 % Confidence Limits		P-Value
Gender (Male vs Female)	0.917	0.292	2.879	0.8816
Preemie (<37 weeks)	4.750	1.240	18.189	0.0229
Decannulation				0.0003
Yes vs No	24.000	2.329	247.368	
No trach vs No	0.923	0.069	12.280	
Bronchopulmonary Dysplasia (Y vs N)	3.348	0.979	11.444	0.0540
Comorbidities (Y vs N)	2.100	0.346	12.759	0.4204
Initial Grade of Stenosis				0.0118
2 vs 1	9.600	1.451	63.500	
3 vs 1	6.400	1.573	26.034	
Gestation Age (Weeks)	0.877	0.790	0.974	0.0140
Intubation Length (Days)	1.010	0.998	1.021	0.0908
Number of Interventions	2.316	1.443	3.718	0.0005

DISCUSSION

- Despite the multiple treatment methods available, SGS remains a challenging condition to treat.

- Balloon dilation typically cures only low-grade SGS; however, it can be useful to reduce the severity of SGS to prepare for a less complicated LTR.

- Length of intubation was not associated with higher rate of LTR likely due to advances in neonatal intensive care airway management to minimize airway trauma (smaller endotracheal tubes [ETT], low-profile or uncuffed ETT, premedication with sedatives prior to intubation, intubation with video laryngoscopy, maintaining cuff pressure <14 mmHg).

- Limitations of this study include no control group (given procedural nature of study group) and small sample size (single institution). Further prospective multi-center studies with longitudinal follow-up would be beneficial to affirm our findings.

- Future research directions include evaluating maternal health, socio-economic factors, and environmental exposures in pediatric SGS patients. Assessing these may elicit additional risks that deepen our understanding of external factors and their effects on respective SGS treatment outcomes.

CONCLUSIONS

- Our study suggests that patients with a gestational age < 33 weeks, an initial Myer-Cotton SGS grade of at least 3, or at least 3 failed conservative interventions were more likely to require LTR.

- We hope this work will guide personalized treatment strategies for SGS patients to minimize interventions and maximize outcomes.

Rajanya S. Petersson, MS, MD, FACS
Department of Otolaryngology - Head and Neck Surgery
Virginia Commonwealth University Health System
PO Box 980146
Email: rajanya.petersson@vcuhealth.org

REFERENCES

1. Jaillette E, Martin-Loeches I, Artigas A, Nseir S. Optimal care and design of the tracheal cuff in the critically ill patient. Ann Intensive Care 2014; 4:7.
2. Newth CJ, Rachman B, Patel N, Hammer J. The use of cuffed versus uncuffed endotracheal tubes in pediatric intensive care. J Pediatr 2004; 144:333-337.
3. Arianpour K, Forman SN, Karabon P, Thottam PJ. Pediatric acquired subglottic stenosis: Associated costs and comorbidities of 7,981 hospitalizations. Int J Pediatr Otorhinolaryngol 2019; 117:51-56.
4. Jefferson ND, Cohen AP, Rutter MJ. Subglottic stenosis. Semin Pediatr Surg 2016; 25:138-143.
5. Wentzel AM, Schweiger C, Manica D et al. Impact of balloon laryngoplasty on management of acute subglottic stenosis. Eur Arch Otorhinolaryngol 2018; 275:2325-2331.
6. Amin RS, Rutter MJ. Airway Disease and Management In Bronchopulmonary Dysplasia. Clin Perinatol 2015; 42:857-870.
7. Walner DL, Loewen MS, Kimura RE. Neonatal subglottic stenosis—incidence and trends. Laryngoscope 2001; 111:48-51.
8. Monica D, Schweiger C, Marostica PJ, Kuhl G, Carvalho PR. Association between length of intubation and subglottic stenosis in children. Laryngoscope 2013; 123:1049-1054.
9. Schweiger C, Manica D, Pereira DRRet al. Undersedation is a risk factor for the development of subglotticstenosis in intubated children. J Pediatr (Rio J) 2017; 93:351-355.
10. Franco RA, Jr. Laryngopharyngeal reflux. Allergy Asthma Proc 2006; 27:21-25.
11. Blum BY, McGowan FX, Jr. Chronic upper airway obstruction and cardiac dysfunction: anatomy, pathophysiology and anesthetic implications. Paediatr Anaesth 2004; 14:75-83.
12. Sacca R, Zur KB, Crowley TB, Zackai EH, Valverde KD, McDonald-McGinn DM. Association of airway abnormalities with 22q11.2 deletion syndrome. Int J Pediatr Otorhinolaryngol 2017; 96:11-14.
13. Zhang H, Zhang J, Zhao S. Airway damage of prematurity: The impact of prolonged intubation, ventilation, and chronic lung disease. Semin Fetal Neonatal Med 2016; 21:246-253.
14. Choi SS, Zalzal GH. Changing trends in neonatal subglottic stenosis. Otolaryngol Head Neck Surg 2000; 122:61-63.
15. Whigham AS, Howell R, Choi S, Pena M, Zalzal G, Preciado D. Outcomes of balloon dilation in pediatric subglottic stenosis. Ann Otol Rhinol Laryngol 2012; 121:442-448.
16. Alshammari J, Alkhunaizi AA, Arafat AS. Tertiary center experience with primary endoscopic laryngoplasty in pediatric acquired subglottic stenosis and literature review. Int J Pediatr Adolesc Med 2017; 4:33-37.
17. Myer CM, 3rd, O'Connor DM, Cotton RT. Proposed grading system for subglottic stenosis based on endotracheal tube sizes. Ann Otol Rhinol Laryngol 1994; 103:319-323.
18. Bent JP, Shah MB, Nord R, Parikh SR. Balloon dilation for recurrent stenosis after pediatric laryngotracheoplasty. Ann Otol Rhinol Laryngol 2010; 119:619-627.
19. Cohen MD, Weber TR, Rao CC. Balloon dilation of tracheal and bronchial stenosis. AJR Am J Roentgenol 1984; 142:477-478.
20. Maresh A, Preciado DA, O'Connell AP, Zalzal GH. A comparative analysis of open surgery vs endoscopic balloon dilation for pediatric subglottic stenosis. JAMA Otolaryngol Head Neck Surg 2014; 140:901-905.
21. Chen C, Ni WH, Tian TL, Xu ZM. The outcomes of endoscopic management in young children with subglottic stenosis. Int J Pediatr Otorhinolaryngol 2017; 99:141-145.
22. Park RS, Peyton JM, Kovatsis PG. Neonatal Airway Management. Clin Perinatol 2019; 46:745-763.
23. Sawyer T, Foglia E, Hatch LDet al. Improving neonatal intubation safety: A journey of a thousand miles. J Neonatal Perinatal Med 2017; 10:125-131.
24. https://www.frontiersin.org/files/Articles/491815/fped-07-00532-HTML/image_m/fped-07-00532-g001.jpg