

CASE REPORT



Subglottic Stenosis in a 3-Year-Old Child Following Prolonged Intubation - A Case Report

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Abstract: Subglottic stenosis is a serious complication of prolonged intubation in pediatric patients and Over 90% of acquired cases are iatrogenically caused and result from endotracheal intubation. Subglottic stenosis is an unanticipated problem that needs to be diagnosed and treated immediately. The majority of pediatric subglottic stenosis occurrences are mild to moderate. This case highlights the challenges and outcomes associated with this condition. Here, we present a 3-year-old child presented with high fever and scrotal pain with swelling, diagnosed as scrotal cellulitis. Laboratory results showed anemia, leukocytosis, and metabolic acidosis, and the child's condition deteriorated due to respiratory distress, requiring intubation and PICU admission. Following a scrotal abscess drainage, the child was discharged but returned with severe respiratory distress, cyanosis, and subglottic stenosis, confirmed by laryngobronchoscopy and imaging. After failed extubation attempts and further complications, a tracheostomy and endoscopic subglottic dilation were performed. The child was successfully decannulated after eight days, with near-complete recovery except for a slight change in voice tone. The case highlights the challenges in managing complex respiratory complications in pediatric patients. This case underscores the importance of monitoring for subglottic stenosis in children who have undergone prolonged intubation and demonstrates the efficacy of endoscopic dilatation in managing this complication.

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1. INTRODUCTION

Tracheal subglottic stenosis is one of the two types of congenital or acquired airway obstructions that affect newborns and children. The narrowest portion of the airway in infants and children is the subglottis of the anatomical area of the trachea, which is located next to the cricoid cartilage and directly below the surface of the voice cords. This region's inflexibility makes subglottic stenosis potentially fatal because it reduces the airway's diameter in this area. Furthermore, 90% of patients with acquired subglottic stenosis undergo iatrogenic endotracheal intubation [1]. Based on either retrospective or prospective evaluations, children's subglottic stenosis prevalence varies between 0.6% and 11.38% [2, 3].

Especially in children, the most common risk factors for subglottic stenosis are extended intubation, incorrect endotracheal tube size selection, insufficient sedation and analgesia, repeated intubation, and nursing errors such as forceful suctioning. Other risk factors for subglottic stenosis include underlying respiratory illnesses, gastroesophageal reflux disease (GERD), premature birth, brain damage, congenital

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airway disorders, cerebral palsy, Down syndrome, prolonged seizures, viral infection leading to intubation, and inadequate perfusion of the airway mucosa due to hypotension, anemia, sepsis, obesity, diabetes, and shock [4-10].

Stridor during inspiration is the initial indication of subglottic stenosis. In addition, if blockage worsens, biphasic stridor ensues. The prevalence rate of stridor, a common complication in children following extubation, stands at 44%. It is particularly concerning when it is severe, late, or progressive, and requires prompt evaluation and intervention [11]. Laryngobronchoscopy is considered the most reliable method for diagnosing subglottic stenosis. Medical treatment or bronchoscopic intervention is necessary for mild to moderate cases. Furthermore, cases classified as moderate to severe require the use of open surgery and tracheostomy [4].

2. CASE PRESENTATION

A 3-year-old child presented to the emergency department with a high fever and scrotal pain accompanied by swelling, persisting for 5 days. A scrotal ultrasound was performed, revealing scrotal cellulitis, and laboratory investigations showed anemia, marked leukocytosis, and metabolic acidosis. The child's condition deteriorated due to respiratory distress, prompting transfer to the pediatric intensive care unit (PICU).

In the PICU, the child was intubated, stabilized, and pediatric surgery was consulted for drainage of a scrotal abscess. The child remained intubated for a month but was eventually extubated and discharged home after spending an additional 5 days in the pediatric hospital ward.

Seven days post-discharge, the child returned with complaints of shortness of breath and cough. On examination, he exhibited severe chest indrawing and low oxygen saturation (<90%). His vital signs included a temperature of 37°C, a respiratory rate of 50 breaths per minute indicative of tachypnea, and a pulse rate of 140 beats per minute, consistent with tachycardia. He was administered supplementary oxygen; however, 12 hours after admission, he developed worsening respiratory distress and dyspnea. While initial pharmacotherapy provided temporary relief, he soon experienced severe respiratory distress again, accompanied by cyanosis.

Upon transfer to the pediatric critical care unit, his vital signs were as follows: Glasgow Coma Scale (GCS) of 8, partial pressure of carbon dioxide (PCO₂) at 75 mmHg, pulse rate of 60 beats per minute, and oxygen saturation (SpO₂) of 80% despite being on high-flow oxygen therapy via a face mask. Rapid sequence intubation (RSI) was immediately performed.

Attempts to extubate were unsuccessful due to persistent signs of obstruction, including chest indrawing, secretions, and inadequate ventilation. The child was initially intubated with a 4.0-sized endotracheal tube (ETT). Following a failed extubation attempt the next day, he required reintubation. Due to difficulty passing the previous ETT size, reintubation was achieved with a guidewire and a 3.5-sized ETT.

Subsequently, the child underwent a tracheostomy, and subglottic stenosis was diagnosed. Once his respiratory and hemodynamic status was stabilized, a diagnostic laryngobronchoscopy was performed by an otolaryngologist, which revealed severe subglottic stenosis (grade 3-4), located 2 cm below the vocal cords.

Further imaging, including CT scans of the neck and chest with intravenous contrast, confirmed the presence of subglottic stenosis and vocal cord secretions. The patient was referred to a specialized Ear, Nose, and Throat (ENT) clinic for management of tracheal stenosis.

Diagnostic laryngobronchoscopy showed near-complete tracheal stenosis (grades 3-4), located 1.5-2 cm below the vocal cords. The child underwent a lower-level retracheostomy and endoscopic subglottic dilation using dilators of 7 and 9 mm to widen the airway. A Foley catheter, inflated with 2 cc of water, was left in the dilated area for 8 days.

After eight days, the tracheostomy was removed, and the child made a near-complete recovery, with only a slight alteration in voice tone that did not cause any further breathing difficulties. He was monitored in the ICU and gradually transitioned to normal respiratory support (Figs. 1 and 2).



Figs. (1 and 2). Axial CT scan of the neck with intravenous contrast showing significant narrowing of the tracheal lumen, indicative of severe subglottic stenosis. The narrowing is observed approximately 2 cm below the vocal cords. Soft tissue swelling and irregularities can be noted around the stenotic segment.

2.1. Intervention

- **Procedure:** Endoscopic dilatation of the subglottic stenosis was performed. The procedure involved inserting a balloon or other dilating instruments into the narrowed area to widen the airway.
- **Post-Procedure Management:** The child was monitored in the ICU and gradually transitioned to normal respiratory support.

2.2. Outcome

- **Immediate Post-Procedure:** Significant improvement in airway patency and reduction in stridor.
- **Follow-Up:** At 6 months, the child was stable with no recurrence of stenosis and normal respiratory function. (Figs. 3 and 4)



Figs. (3 and 4). Endoscopic view of the trachea showing severe subglottic stenosis. The image depicts significant narrowing of the airway lumen, with evident concentric fibrotic changes and thickening of the subglottic tissue, consistent with grade 3-4 stenosis. The lumen is markedly reduced, contributing to the patient's respiratory distress.

3. DISCUSSION

The most typical differential diagnosis for stridor in children is subglottic stenosis after prolonged intubation. Despite the high morbidity and mortality rates in children, additional attention is required to be made to prevent subglottic stenosis.

Patients themselves can be attributed to predisposing factors for post-intubation stenosis, including congenital airway narrowing, preterm, gastric reflux, keloid development, and systemic variables that lead to reduced blood flow in the mucosa, such as hypotension, anemia, sepsis, and shock. Extrinsic variables refer to external circumstances. These factors include the size or rigidity of the ETT (endotracheal tube), the method of intubation (traumatic, multiple attempts, or long-term intubation) [12], and the quality of nursing care (insufficient sedation, excessive tube movement, and repeated and traumatic aspirations) [13].

The main variables that lead to post-intubation subglottic stenosis (SGS) are traumatic intubation and the pressure exerted by the endotracheal tube (ETT). When the endotracheal tube (ETT) exerts pressure that exceeds the capillary perfusion requirement, ischemia develops, leading to edema, necrosis, and ulcers. Subsequently, during the repair process, the formation of granulation tissue may result in a reduction of the airway passage and cause blockage. We regard intubation lasting more than 4 weeks as elevating the risk of stenosis due to superinfection; however, damage can occur within just 48 hours [14].

Treatment for children with subglottic stenosis has advanced significantly, but managing the condition is still complicated with controversy. Some of the treatments that can be used are medication, tracheostomy, balloon endoscopic dilatation, endoscopic anterior cricoid split with balloon dilatation, endoscopic posterior cricoid split with cartilage graft, laryngotracheal reconstruction (LTR), and partial cricotracheal resection (CTR) [15]. Medication, such as systemic or inhaled corticosteroids and racemic epinephrine, is used to treat the majority of mild cases [16].

Systematic review studies have shown that endoscopic balloon dilatation has been successful in approximately two-thirds of cases with low to moderate severity. However, as the severity of stenosis and intubation period increased, the failure rate increased [17,18].

A retrospective study found that 84.5% of patients with moderate to severe post-intubation subglottic stenosis achieved decannulation with just one surgical procedure. We treated the patients using either laryngotracheal reconstruction, partial cricotracheal resection, or anterior cricoid split methods [19,20].

CONCLUSION

This case demonstrates that while subglottic stenosis grade 4-5 can be a serious complication of prolonged intubation, timely and effective management with endoscopic dilatation can lead to favorable outcomes.

RECOMMENDATIONS

Monitoring: Vigilance in monitoring for subglottic stenosis in children following prolonged intubation.

Treatment: Endoscopic dilatation as an effective treatment modality.

AUTHORS' CONTRIBUTIONS

The author confirms sole responsibility for the following: study conception and design, data collection, analysis and interpretation of results, and manuscript preparation.

CONSENT FOR PUBLICATION

We have obtained written informed consent from the patient's parent for the publication of this case report and any accompanying images. The consent form has outlined that all personal identifiers, including but not limited to names, initials, and hospital numbers, will not be disclosed in this publication to ensure the confidentiality of the patient.

CONFLICT OF INTEREST

None.

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REFERENCES

- [1] Marston AP, White DR. Subglottic stenosis. *Clin Perinatol.* 2018; 45(4):787-804. [DOI:10.1016/j.clp.2018.07.013] [PMID]
- [2] Rodríguez H, Cuestas G, Botto H, Cocciaglia A, Nieto M, Zanetta A. Post-intubation subglottic stenosis in children. Diagnosis, treatment and prevention of moderate and severe stenosis. *Acta Otorrinolaringol.* 2013; 64(5):339-44. [DOI:10.1016/j.otoeng.2013.10.005] [PMID]
- [3] Schweiger C, Marostica PJ, Smith MM, Manica D, Carvalho PR, Kuhl G. Incidence of post-intubation subglottic stenosis in children: Prospective study. *J Laryngol Otol.* 2013; 127(4):399- 403. [DOI:10.1017/S002221511300025X] [PMID]

- [4] Rutter M, Kuo IC. Predicting and managing the development of subglottic stenosis following intubation in children. *J Pediatr.* 2020; 96(1):1-3. [DOI:10.1016/j.jpmed.2019.04.001] [PMID]
- [5] Wei JL, Bond J. Management and prevention of endotracheal intubation injury in neonates. *Curr Opin Otolaryngol Head Neck Surg.* 2011; 19(6):474-7. [DOI:10.1097/MOO.0b013e32834c7b5c] [PMID]
- [6] Poetker DM, Ettema SL, Blumin JH, Toohill RJ, Merati AL. Association of airway abnormalities and risk factors in 37 subglottic stenosis patients. *Otolaryngol Head Neck Surg.* 2006; 135(3):434-7. [DOI:10.1016/j.otohns.2006.04.013] [PMID]
- [7] Schweiger C, Manica D, Pereira DRR, Carvalho PRA, Piva JP, Kuhl G, *et al.* Undersedation is a risk factor for the development of subglottic stenosis in intubated children. *J Pediatr.* 2017; 93(4):351-5. [DOI:10.1016/j.jpmedp.2017.02.004] [PMID]
- [8] Manica D, Schweiger C, Maróstica PJC, Kuhl G, Carvalho PRA. Association between length of intubation and subglottic stenosis in children. *Laryngoscope.* 2013; 123(4):1049-54. [DOI:10.1002/lary.23771] [PMID]
- [9] Jefferson ND, Cohen AP, Rutter MJ. Subglottic stenosis. *Semin Pediatr Surg.* 2016; 25(3):138-43. [DOI:10.1053/j.sempedsurg.2016.02.006] [PMID]
- [10] Nicolli EA, Carey RM, Farquhar D, Haft S, Alfonso KP, Mirza N. Risk factors for adult acquired subglottic stenosis. *J Laryngol Otol.* 2017; 131(3):264-7. [DOI: 10.1017/S0022215116009798] [PMID]
- [11] Schweiger C, Eneas LV, Manica D, de Souza Saleh Netto C, Carvalho PRA, Piva JP, *et al.* Accuracy of stridor-based diagnosis of post-intubation subglottic stenosis in pediatric patients. *J Pediatr.* 2020; 96(1):39-45. [DOI:10.1016/j.jpmed.2018.08.004] [PMID]
- [12] Manica, Denise, *et al.* "Association between length of intubation and subglottic stenosis in children." *The Laryngoscope* 123.4 (2013): 1049-1054.
- [13] Zanetta A, Cuestas G, Rodríguez H, Tiscornia C. Nuevo stent laríngeo en el tratamiento de la estenosis subglótica en niños. *Acta Otorrinolaringol Esp.* 2012, ~ <http://dx.doi.org/10.1016/j.otorri.2012.05.008>.
- [14] Monnier P. Acquired post-intubation and tracheostomy-related stenoses. In: Monnier P, editor. *Pediatric airway surgery*. Lausanne, Switzerland: Springer eds; 2011. p. 183-98.
- [15] Marston AP, White DR. Subglottic stenosis. *Clin Perinatol.* 2018; 45(4):787-804. [DOI:10.1016/j.clp.2018.07.013] [PMID]
- [16] Sinha A, Jayashree M, Singhi S. Aerosolized L-epinephrine vs budesonide for post extubation stridor: A randomized controlled trial. *Indian Pediatr.* 2010; 47(4):317-22. [DOI:10.1007/s13312-010-0060-z] [PMID]
- [17] Lang M, Brietzke SE. A systematic review and meta-analysis of endoscopic balloon dilation of pediatric subglottic stenosis. *Otolaryngol Head Neck Surg.* 2014; 150(2):174-9. [DOI:10.1177/0194599813510867] [PMID]
- [18] Romero Manteola EJ, Patiño González C, Ravetta P, Defago V, Tessi C. Dilation with rigid dilators as primary treatment of subglottic stenosis in pediatrics. *Pulmonology.* 2020; S2531-0437(20)30046-5. [DOI:10.1016/j.pulmoe.2020.03.007] [PMID]
- [19] Bitar MA, Al Barazi R, Barakeh R. Airway reconstruction: Review of an approach to the advanced-stage laryngotracheal stenosis. *Braz J Otorhinolaryngol.* 2017; 83(3):299-312. [DOI:10.1016/j.bjorl.2016.03.012] [PMID]
- [20] Watson, D. C., *et al.* (2011). "Endoscopic management of pediatric subglottic stenosis: A review." *International Journal of Pediatric Otorhinolaryngology*, 75(8), 989-995.