

Paediatric Subglottic Mucocele: Early Intervention and Recovery

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How to cite this article:

Shriya Bhattarai, PS Suji, Abhishek Bhardwaj *et al.* Paediatric Subglottic Mucocele: Early Intervention and Recovery. RFP J ENT Allied Sci 2023;8(2):51-54.

ABSTRACT

Subglottic mucocele is a rare entity in the pediatric population, often associated with intubation-related complications. We present a case of a 1.5-year-old male child with a history of repeated hospitalizations for respiratory distress, ultimately diagnosed with subglottic mucocele. Despite being born at full term without comorbidities, the patient required mechanical ventilation shortly after birth. Imaging revealed an ill-defined lesion in the subglottic region, confirmed histologically as a mucocele. Following initial airway compromise during excision, emergency tracheostomy was performed, followed by successful removal of the mass via coblation. Postoperative follow-up showed significant improvement in respiratory symptoms, and the patient was successfully decannulated. This case highlights the importance of considering subglottic mucocele as a differential diagnosis in pediatric airway obstruction, necessitating prompt diagnosis and appropriate surgical management. We discuss the etiology, differential diagnosis, and surgical techniques for managing subglottic lesions in pediatric patients, emphasizing the significance of a multidisciplinary approach for optimal outcomes.

Keywords: Subglottis; Mucocele; Coblation; Stridor; Airway.

INTRODUCTION

Subglottic mucocele is a rare but potentially life-threatening condition in pediatric patients, often arising as a consequence of endotracheal intubation.¹ Despite its infrequency, the

management of subglottic mucocele demands a prompt and comprehensive approach to prevent airway compromise and ensure favourable outcomes.² In our case, early intervention played a pivotal role in expediting the patient's recovery. Prompt recognition of the subglottic mucocele and its potential for airway compromise led to immediate surgical intervention. Despite encountering a challenging intraoperative airway event necessitating emergency tracheostomy, decisive action allowed for continued surgical excision and subsequent resolution of the lesion. Additionally, close postoperative monitoring enabled timely adjustments in the management plan, facilitating successful decannulation and ensuring a smoother transition to respiratory autonomy. This emphasis on early recognition and intervention underscores the critical role of proactive management strategies in mitigating complications and promoting optimal patient outcomes in pediatric airway disorders.

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Received on: 07.08.2024

Accepted on: 10.09.2024



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CASE PRESENTATION

A 1.5-year-old male child presented to the paediatrics emergency in our tertiary care centre with a history of shortness of breath and noisy breathing for the past three months. The child had a history of mechanical ventilation 2 hours after birth following feeding and required mechanical ventilation for 48 hours. Till 1.5 years of age, the child had a history of repeated hospital admissions for breathing difficulty and received treatment for pneumonia. History revealed that he had a term delivery, developmental milestones as per age and

no other history of comorbidities. On examination, the child had a normal BMI (body mass index) and he had occasional biphasic stridor with suprasternal retractions.

Investigations

A contrast-enhanced computed tomography from the base of the skull to the diaphragm revealed an ill-defined, polypoidal, non-enhancing, hypodense lesion, likely granulation tissue or inflammatory aetiology in subglottis (mainly on the right), maximum thickness 6.5mm, narrowest diameter 4mm (TR), 14mm (AP) at the level of C5 (Fig. 1).

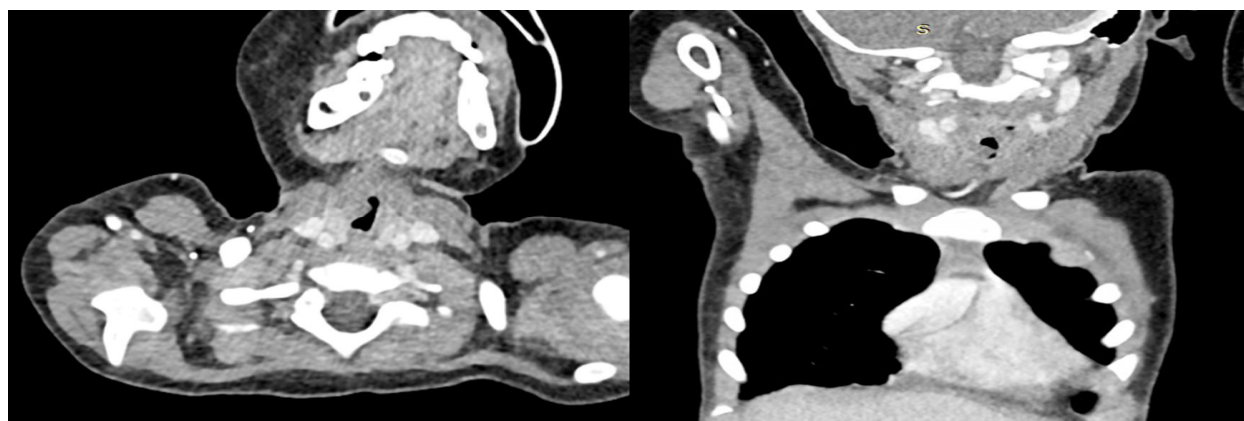


Fig. 1: Axial and coronal CECT neck showing soft tissue narrowing airway in subglottis

Treatment

Direct Laryngoscopy under Monitored anaesthesia noticed a smooth pinkish mass in the subglottis arising from the posterolateral wall of the right subglottic region. (Fig. 2A) Intraoperative sudden fall in SpO_2 necessitated emergency tracheostomy. The patient was kept on ventilator for 6 days and after stabilization on the eighth day after the first procedure, he was taken up under GA via

tracheostomy tube (4.5mm). Mass was excised with the help of a coblator. The histopathology report came out to be mucocele. The child was discharged after advising tracheostomy tube care. Twenty-five days later, the child was re-admitted, and a direct laryngoscopic assessment was done, which showed no recurrence and healthy, healed mucosa was seen in the postoperative bed. Decannulation has been done uneventfully (Fig. 2B).

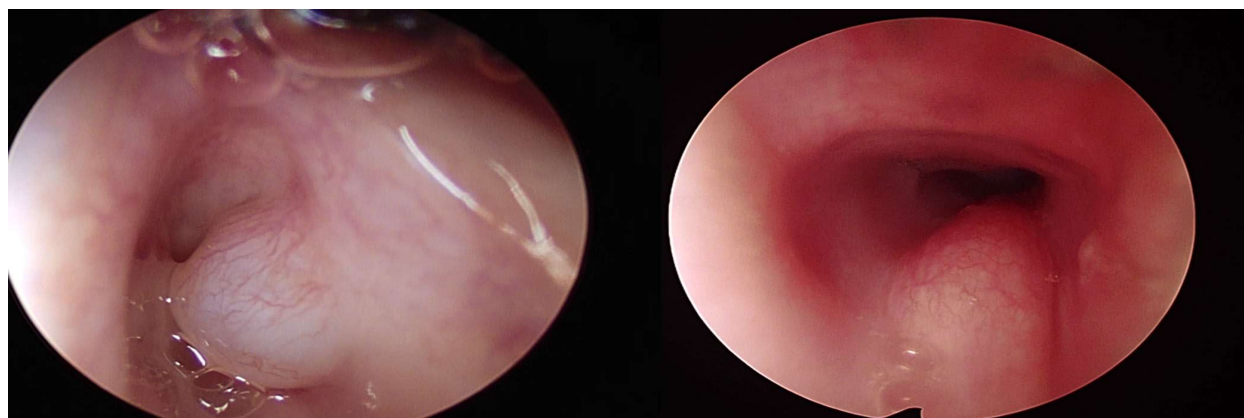


Fig. 2: a) Endoscopic picture of subglottic mass

b) 1 month post excision picture

Table 1: Differentiating features of different subglottic masses

Cause	Differentiating feature	Treatment
Subglottic haemangioma	Biphasic stridor at 2-3 months of age, asymmetric narrowing of trachea on imaging	Open excision, endoscopic laser excision or microdebrider, propranolol. ⁵
Subglottic cyst	History of endotracheal intubation, prematurity, cystic nature-air/fluid content on imaging	Endoscopic marsupialization with laser/cold instruments. ⁹
Respiratory papillomatosis	History of anogenital warts in mother, 'cauliflower-like' exophytic growth lesions, solid/cavitated pulmonary nodules in cases of lung involvement	Excision with microdebrider, laser or coblator, interferon alpha, bevacizumab. ⁶

Outcome and follow-up

After discharge, the child remained clinically stable. His noisy breathing subsided. The child was followed up every two weeks post-excision, and no signs of respiratory distress. He gained weight from 11kg(post-operative period) to 16kg (1 month postoperative).

DISCUSSION

In literature, most subglottic cysts are related with prematurity due to requirement of intubation. Our report, with an average term of 38 weeks of amenorrhea at birth, is inconsistent with the literature. Obstructive lesions of the pediatric airway can occur at any level, from the nose and nasopharynx through the pharynx and supraglottis to the glottis, subglottis, trachea, and lower airway structures. At any level, the lesions can be congenital or acquired; expansile, dynamic, or static; and partially to progressively to entirely obstructive. The subglottis is particularly injury-prone in neonatal and pediatric populations because it is a significant point of contact for the life-saving measures of endotracheal intubation.¹

There appears to be a clear emergence of subglottic mucus retention cysts as a complication following intubation in pediatric population. The presence of the endotracheal tube causes erosion, inflammation, and subsequent scarring in the subglottic region, leading to the obstruction of submucous gland ducts and the formation of cysts.³ The subglottic submucosa has a more significant proportion of glandular soft tissue than the rest of the airway, making it the narrowest part of the neonatal airway as opposed to the glottis in the adult. The clinical importance of subglottic cysts depends on the degree of airway compromise, as small and solitary cysts may be an incidental finding on endoscopy, and large or multiple cysts may cause catastrophic airway obstruction. Subglottic cysts should be differentiated from hemangioma, stenosis, and

respiratory papillomatosis, which are more common causes of paediatric stridor and rare subglottic lesions such as thyroglossal cysts and ectopic thymic cysts.⁴ The different causes of subglottic masses and the differentiating features are given in Table 1.

Contingency plans should be delineated while planning for excision, including a plan for emergency tracheostomy in case of difficult ventilation or intubation. Excision of the cysts will often be sufficient to correct the obstructed airway; however, the recurrence rate is as high as 40%.⁶ Endoscopic removal allows a clear view of the lesion, which aids the excision of cyst content, de-obstructing the airway and causing no damage to the underlying mucosa. This approach could explain the low recurrence rate observed compared to other modalities, such as CO2 laser.^{3,6} The use of microdebrider has been reported to allow better control of the excision of the cysts. Steehler et al. also reported promising results of the post-marsupialization application of Mitomycin-C in a recent study. It could prevent recurrence and reduce post-marsupialization scarring.⁷

CONCLUSION

- Timely diagnosis and surgical intervention are paramount in managing pediatric subglottic mucocele to prevent airway compromise and minimize morbidity.
- Early recognition of subglottic lesions, coupled with decisive surgical excision and vigilant postoperative monitoring, facilitates successful resolution of symptoms and avoidance of long-term complications.
- Multidisciplinary collaboration among otolaryngologists, paediatricians, anesthesiologists, and other healthcare professionals is essential for optimizing outcomes in pediatric patients with subglottic mucocele.

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