Recurrent Subglottic Stenosis in a 16-month-old Male in the Setting of Influenza A, Intubation, and Honey Consumption

Allison T. Jakiel, DO 1, Krisdaniel Berreta, DO 2, Sami Rishmawi, MD 3, 4, Hanna S. Sahhar, MD 5, 4

1. Pediatric Intensive Care Unit, Edward Via College of Osteopathic Medicine–Virginia, Blacksburg, USA. 2. Pediatric Intensive Care Unit, Edward Via College of Osteopathic Medicine–Carolinas, Spartanburg, USA. 3. Pediatric Medicine, Edward Via College of Osteopathic Medicine–Carolinas, Spartanburg, USA. 4. Pediatric Intensive Care Unit, Spartanburg Regional Healthcare System, Spartanburg, USA.

Corresponding author: Allison T. Jakiel, DO, aburns@vcom.edu

Abstract

Recurrent episodes of subglottic stenosis are rare in literature and the etiologic causes are misunderstood, but can be congenital, idiopathic, or iatrogenic in nature. Complications of intubation can result in subsequent inflammation and reactive processes. This case involves a 16-month-old male who suffered from a recurrent episode of subglottic stenosis in the setting of croup, influenza, and honey consumption. He had presented to the emergency department in respiratory distress after ingesting a home remedy of onion juice and honey. He had been discharged one day prior from the pediatric intensive care unit after four days of intubation and a seven-day hospital course with evidence of croup on imaging. He was readmitted and subglottic edema and narrowing was confirmed via endoscopy, which prompted antibiotic treatment and close monitoring. After three days of monitoring and re-evaluation by bronchoscopy, the patient’s condition began to improve, and no intubation was necessary. It is unclear of the cause of recurrent subglottic stenosis due to the patient’s clinical picture being clouded by a potential allergic reaction to honey versus an inflammatory reactive process post-intubation from the previous admission days prior. This case emphasizes the need for further research on prevalence and etiology of recurrent subglottic stenosis and a deeper understanding of how to optimize diagnosis and treatment.

Keywords: pediatric intensive care unit (PICU), post-intubation complications, post intubation tracheal stenosis, honey consumption, recurrent tracheal stenosis, pediatric influenza a, subglottic stenosis

Introduction

This Case Report was previously displayed as a poster at the American College of Osteopathic Pediatricians (ACOP) 2023 Spring Conference on April 29, 2023 in Louisville, KY.

Subglottic stenosis is the narrowing of the area between the vocal folds and the inferior border of the cricoid cartilage. It can be congenital, acquired, or idiopathic, which is often misdiagnosed as asthma [1]. Symptoms include biphasic stridor, dyspnea, and intercostal, suprasternal, and abdominal retractions [2]. Honey is often used to reduce asthma-related symptoms and in this case it was used as a therapeutic means; however, the etiology of subglottic stenosis is not fully understood [3]. Subglottic stenosis could be acquired post-intubation, infection, inflammation, tumors, ingestions, and trauma [4].

In cases of post-intubation, pressure necrosis can occur due to the endotracheal tubing and can lead to mucosal edema and ulceration [5]. This inflammation causes increased mucus production, predisposition to bacterial overgrowth, type II collagen thickening, and damaged ciliate epithelium. The growth of scar tissue, mucosal sloughing, and granulation ultimately results in intraluminal narrowing of the upper airway and subglottic stenosis. Delayed stenosis occurs when this process prolongs [6]. Over 24-36 hours after extubation, reactive edema, mucosal damage and obstruction, retractions, and stridor can be evident. It will typically worsen before improving, warranting close monitoring of patients. After 72 hours, if stridor persists or new onset, the airway is severely affected and prompts endoscopic evaluation and intervention [7].

In 2013, a prospective study indicated an incidence of 11.38% of subglottic stenosis in 123 children post-intubation [8]. Subglottic stenosis can be evaluated via bronchoscopy and imaging. Subglottic stenosis is typically managed through endoscopy with either removal of obstructing granulation tissue, steroid injection to reduce the inflammation, and balloon dilation to widen the airway [9]. Placement of tracheotomy or laryngotracheal reconstruction may also be considered in more severe cases where the cartilage is compromised or there is congenital subglottic stenosis [7].

The goal of this case report is to highlight a unique presentation of recurrent subglottic stenosis in the
setting of Influenza A infection, croup, intubation, and honey ingestion resulting in subsequent immune reaction.

Case Presentation
A 16-month-old African American male presented to the Emergency Department (ED) with a primary concern of lip edema after consumption of a honey and onion juice home remedy. This was the patient’s second occurrence of immune reaction following honey consumption. The patient had been discharged the day prior from the pediatric intensive care unit (PICU) after a seven-day hospital course and four days of intubation due to subglottic stenosis and diffuse tracheal narrowing (Figure 1, 2, 3, 4) secondary to croup in the setting of croup secondary to influenza A. According to his mother, he had been improving overall but began to have trouble breathing and a barky cough. He had recently been prescribed albuterol from an urgent care, who had diagnosed the patient with Influenza the week prior after presenting due to lip swelling and respiratory distress. On examination, the patient was febrile at 100.3 F, tachypneic at a rate of 35, heart rate of 162, audible stridor, intercostal and supravacular retraction, and abdominal breathing.

FIGURE 1: Posterior-Anterior Radiograph Showing Initial Subglottic Stenosis and Diffuse Tracheal Narrowing
FIGURE 2: Lateral Radiograph Showing Initial Subglottic Stenosis and Diffuse Tracheal Narrowing
FIGURE 3: Chest Radiograph for Initial PICU Admission
FIGURE 4: Chest Radiograph on Day of Discharge from Initial PICU Admission Showing Clinical Improvement of Stenosis

In the ED, he received 5 L/min of oxygen via nasal cannula. Given his recent PICU admission and intubation, the patient was admitted due to persistent respiratory distress and exacerbation. The patient was given Budesonide-Pulmicort 0.25mg/2mL nebulizer solution, dexamethasone IM 5mg, Benadryl IM 11 mg, Epinephrine IM 0.1 mg, Ractopamine 2.25% nebulizer solution, sodium chloride 0.9% bolus pre-mix. The patient was found to have wandering roving eye movements, hypoactivity from baseline, refusal to stand, spontaneous movement of all four extremities, decreased reflexes in the lower extremity without clonus. According to his mother, the patient had been able to bare weights and walk along objects earlier that day. Radiographs revealed subglottic stenosis and diffuse tracheal narrowing (Figure 5, 6).
FIGURE 5: Posterior-Anterior Radiograph Showing Recurrent Subglottic Stenosis and Diffuse Tracheal Narrowing at Second PICU Admission (One Day After Discharge from Initial PICU)
FIGURE 6: Lateral Radiograph Showing Recurrent Subglottic Stenosis and Diffuse Tracheal Narrowing on Second PICU Admission (One Day After Discharge from Initial PICU)

On day 1 of re-admission, ENT evaluated the patient at bedside via a diagnostic flexible laryngoscopy and found him to have significant subglottic edema and secretions with difficulty to discern if tracheitis was present. ENT recommended broad spectrum antibiotic coverage for possible tracheitis. The patient was treated with 200mg of ampicillin-sulbactam every 6 hours, a ciprodex nebulizer, and a dexamethasone taper. He was reevaluated by bedside bronchoscopy on day 2, which revealed an open but narrow subglottic airway, improvement in the number of secretions, continued deficit in ability to clear secretions, and vocal cord hypomobility. Due to neurological symptoms, a non-sedated MRI was attempted, but unsuccessful.

On day 3 of re-admission, the patient’s activity and mental status significantly improved. The patient cried for the first time since re-admission with improvement in alertness and activity. On day 4, the patient was then transferred to the general pediatric floor and continued to receive physical, occupational, and speech therapy. He made significant improvement and was able to move all extremities and normal strength returned. On day 5, the patient was able to safely discharge home. He was placed on a mechanical soft diet with thickened liquids and thin liquids for medications, a 5-day course of amoxicillin-clavulinate, and was given an EpiPen due to the concern for honey allergy and/or anaphylaxis. Additionally, he was referred to outpatient OT, PT, and ST for continual support in recovery and development. To date, the patient is recovering without any further known admission for recurrent subglottic stenosis.

Discussion
This case of recurrent subglottic stenosis was confounded by a subsequent reaction following honey consumption and multiple hospitalizations. The communication between both PICU teams to avoid intubation given the patient’s airway edema and structural anatomy proved to be critical. This effective communication is imperative in optimal patient care across multiple providers and healthcare facilities.

Upon readmission to the PICU, the differential was expanded to include botulism due to the patient’s decline of baseline motor skills, an abnormal neurological exam, and generalized hypotonia. Over the course of several days, patient improved without any specified treatment or anti-toxin, indicating delirium was likely present. In type I hypersensitivity reactions, exposure to an antigen causes an asymptomatic or minor reaction which then results in anaphylaxis or atopy with subsequent exposure [10]. This could explain why the patient’s first consumption of honey led to lip swelling but no significant change in respiratory status but with the second consumption, in the setting of respiratory compromise, resulted in an apparent and rapid change in respiratory status. While the patient lacked urticaria or hives, it is possible his dark complexion and lack of consideration for honey allergy contributed to non-observation of any mild skin
changes. It is also plausible it was coincidental timing of increased respiratory distress around the time of the honey consumption. This case further emphasizes the importance of strong medical history interview during an evaluation of a recently extubated pediatric patient.

As discussed, acquired subglottic stenosis post-extubation is seen in roughly 11.38% of children, which explains the necessity of why this patient was closely monitored following extubation; however, within 48 hours of post-intubation, this patient’s stridor was only present with agitation with no retractions and was improving. Between 48 and 72 hours, the stridor worsened, and a biphasic stridor was noted after he consumed honey at home which raises the concern for understanding the true etiologic cause. This case reveals the importance of closely monitoring post-intubation for further respiratory deterioration, retractions, and stridor to reduce readmission, reintubation, and clinical worsening.

Conclusions
In conclusion, this case report highlights a unique presentation of recurrent subglottic stenosis in a 16-month-old African American male in the setting of croup, influenza A, and honey consumption. The case emphasizes the clinical implications of continuity of care, effective communication between providers, and the need for additional research on the prevalence and causes of recurrent subglottic stenosis. This condition is rare in literature and prompts more investigation and acquisition of cases to understand its etiology which could benefit medical management and limit complications.

Additional Information
Disclosures
Human subjects: Consent was obtained or waived by all participants in this study. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

Acknowledgements
Allison T. Jakiel, DO and Krisdaniel Berreta, DO contributed equally to the work and should be considered co-first authors.

References
2. UC Irvine Medical Center. Subglottic stenosis. Subglottic Stenosis | UC Irvine Medical Center. Published February 15, 2023, 27:2023.