Case Report

Fixed extrathoracic obstruction masquerading as poorly controlled asthma

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ABSTRACT

We present the misdiagnosis of asthma in a 27-year-old female with a subsequent diagnosis of fixed extrathoracic obstruction supported by pulmonary function testing in the outpatient asthma clinic. Subsequent otolaryngology evaluation demonstrated arytenoid hooding and suspected upper tracheal narrowing on nasolaryngoscopy. Intraoperative airway evaluation confirmed advanced subglottic stenosis (SGS), which required surgical incision and balloon dilation. Tracheostomy decannulation as a toddler was the suspected etiology of acquired SGS.

Key words: Asthma, Misdiagnosis, Obstruction, Pulmonary function testing

Subglottic stenosis (SGS) is abnormal tracheal narrowing below the vocal cords, of which the majority are acquired from inflammation and scarring from prior intubation or surgical airways [1]. Acquired SGS is exceedingly uncommon in the pediatric population and is estimated to occur in 0.06% of patients [2]. This rare upper airway condition presents early on as wheezing, dyspnea on exertion, and cough, which are more commonly observed in lower airway obstruction [3,4]. Because its presentation can masquerade as "difficult-to-treat asthma" unresponsive to inhaler treatment, a diagnosis of SGS is delayed, a mean time of 2 years after symptom onset [5-7]. Differentiating SGS from more common pathologies makes outpatient recognition and timely intervention important in diagnostic considerations.

We present how the misdiagnosis of asthma was revealed through pulmonary function testing (PFT) to be a long-overlooked case of SGS and appropriately engaged interdisciplinary management with eventual surgical correction.

CASE PRESENTATION

A 27-year-old female was referred by her primary doctor to the outpatient asthma clinic for worsening wheezing and dyspnea, presumed due to uncontrolled asthma without response to an increased dose of her daily maintenance inhaler (fluticasone furoate/vilanterol). She had a history of developmental delay, prematurity (24 weeks), and tracheostomy, decannulated at age 2. Her family history was negative for asthma or known respiratory disease. She was diagnosed with asthma as a teenager and received limited relief from inhalers. Longstanding respiratory symptoms, which had worsened over the past year, were localized to the upper chest and throat and particularly exacerbated while supine and during exertion. Rescue inhalers provided immediate relief that lasted minutes, inconsistent with the expected pharmacologic response to short-acting beta-agonists.

Vital signs and intake data were unremarkable: blood pressure 124/76, pulse 74, peripheral capillary oxygen saturation of 100%, body mass index 25.39 kg/m^2. Noisy breathing, including a low-pitched biphasic stridor, was notable throughout her visit.

Prior chest X-ray had been unremarkable. PFT conducted in the asthma clinic was abnormal and consistent with fixed extrathoracic obstruction, with percent predicted as follows: forced expiratory volume in 1 second (FEV1) 54.7%, forced vital capacity (FVC) 55.2%, FEV1/FVC 99% (Fig. 1). Otolaryngology was engaged to investigate the suspicion of a structural cause of extrathoracic obstruction. Further chart review revealed that the patient was shown 12 years earlier to have a small area estimated at 15% of circumferential tracheal stenosis on suspension micro direct laryngoscopy (SMDL) after presenting to the emergency department with biphasic stridor. No surgical intervention was warranted

Access this article online	
Received - 11 July 2025 Initial Review - 04 August 2025 Accepted - 04 September 2025	Quick Response code
DOI: 10.32677/ijcr.v11i10.7736	

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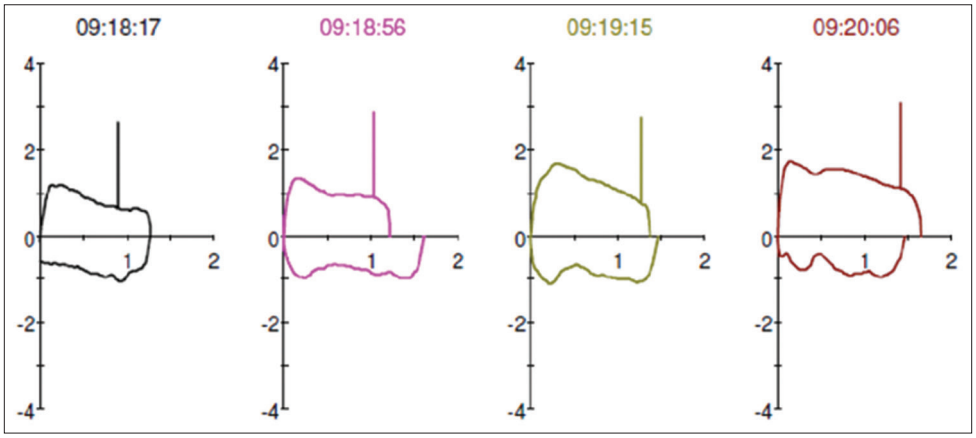

Figure 1: Pulmonary function testing of fixed extrathoracic obstruction. Flattening of inspiratory and expiratory flow-volume loops was reproduced on serial measurements, consistent with fixed extrathoracic obstruction

at the time. On her current referral visit, significant abnormalities were demonstrated on transnasal fiberoptic laryngoscopy, including arytenoid hooding such that the glottis could be visualized and suspected SGS. Of note, further discussion with the patient's mother revealed the patient had experienced coughing and nasal regurgitation during eating and drinking. Subsequent modified barium swallow showed normal oropharyngeal swallowing function without aspiration or laryngeal penetration. She was counseled on mealtime habits and maintaining a neutral position rather than leaning forward, which was thought to contribute to nasal regurgitation.

Arrangements were made to proceed with intraoperative airway evaluation and possible surgical intervention. Repeat SMDL demonstrated stenosis of the proximal subglottis and an A-frame deformity in the distal subglottis, resulting in 70% and 40% airway narrowing of the native tracheal diameter, respectively (Fig. 2a). No concerning lesions were found in the larynx or subglottis. Radial incisions were made with a carbon dioxide laser, and tracheal dilation was performed with a controlled radial expansion balloon (Fig. 2b). The patient tolerated the procedure well, with a much-improved tracheal airway after the procedure.

DISCUSSION

Wheezing and dyspnea are common features of asthma, which improve with appropriate treatments. However, uncontrolled respiratory symptoms or atypical inhaler response should raise suspicion for alternate explanations, especially in the setting of risk factors for anatomical airway complications. When the differential of SGS is raised, clinicians should consider acquired, non-acquired, or mixed etiologies. Acquired etiologies include subglottic hemangioma, congenital vocal cord palsy, and laryngotracheomalacia [1]. Non-acquired diagnoses include respiratory papillomatosis due to human papillomavirus, laryngeal or tracheal malignancy, or foreign body aspiration [1]. The initial workup to investigate cardiopulmonary abnormalities is often benign. Chest radiography alone will likely be normal, as was seen in our patient [8].

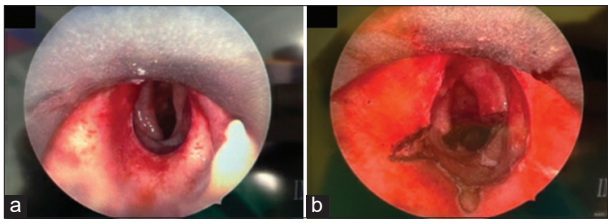

Figure 2: Intraoperative tracheal visualization. (a) Pre-operative view showing stenosis of the immediate subglottis, resulting in narrowing of the airway to 70% of its native diameter, and an A-frame deformity approximately 3 cm below the glottis, resulting in narrowing of the airway to 40% of its native diameter. (b) Post-operative view showing radial incisions made at 7 and 9 o'clock in the proximal subglottic scar band and wedge-shaped incisions at 3 and 9 o'clock in the more distal A-frame stenotic segment

Despite the rarity of SGS, the risks of delayed diagnosis are high and include progressive respiratory failure or tracheal malignancy [7]. Over one-third of SGS patients are misinterpreted to have asthma when initially presenting with respiratory symptoms [6]. In our case, tracheostomy decannulation as a toddler was the suspected etiology of acquired SGS. Attentiveness to risk factors for compromised airways and patient response to inhalers is important to discern the true etiology of chronic respiratory symptoms.

Flattening of inspiratory and expiratory flow-volume loops with limited reversibility after bronchodilators is classic for fixed extrathoracic obstruction [7,9,10]. Our patient demonstrated this characteristic shape, which was reproduced on each in-office trial, which helped guide our clinical decision-making. PFT is considered a favorable, non-invasive test to screen for airway obstruction in the outpatient setting, especially as stenosis rates are projected to rise after the increased need for intubation after the global COVID-19 pandemic [11]. PFT can be crucial in the outpatient setting and support prompt referral to confirm the suspicion of fixed extrathoracic obstruction. In our case, the misdiagnosis of asthma was first revealed through PFT.

Additional measurements can be collected in the office and have shown promise in distinguishing SGS from lower respiratory disorders, such as asthma and chronic obstructive pulmonary disease. The ratio of FEV1 to peak respiratory flow rate, also known as the Empey

index, has long been known to indicate upper airway obstruction at values >10 [12]. More recently, the high sensitivity and specificity of expiratory disproportion index (98%, 96%), or ratio of FEV1 to peak expiratory flow, as well as the dyspnea index (83%, 78%), a Likert-style questionnaire to assess SGS symptoms, suggest that these newer outpatient tools can provide strong evidence for further subspecialty workup [3]. While we did not collect these data in our workup, we acknowledge the utility of these measures, which could be available to those in the outpatient clinic or community practice.

Nevertheless, it is important to note that PFT can still be normal in SGS and not suggest underlying obstruction [13]. Coaching by an experienced respiratory therapist was used in our clinic, which can be particularly helpful in collecting accurate measurements in a young patient or one with intellectual disability, like our patient. Thus, it is necessary to be observant of risk factors that may raise suspicion for alternate explanations for refractory respiratory symptoms or atypical inhaler response, along with objective testing in the clinic.

Otolaryngology evaluation demonstrated arytenoid hooding and suspected upper tracheal narrowing on nasolaryngoscopy. Visualization through laryngotracheoscopy or bronchoscopy is the gold standard for definitive SGS diagnosis, which ultimately was used to evaluate our patient's condition [3,5,6]. However, scopes are not readily accessible to the community physician and often require subspecialty referral.

Intraoperative airway evaluation confirmed advanced SGS, which required surgical incision and balloon dilation. Overlapping symptoms with asthma, such as wheezing, dyspnea, and cough, typically become noticeable when SGS advances such that airway narrowing exceeds 50% or Myer-Cotton grade II [14].

CONCLUSION

In our case, it is likely that the patient's symptoms became more apparent and progressively worsened as her multi-level stenosis advanced to as high as 70%, as compared to her last SMDL of 15% over a decade prior. This patient demonstrates a rare and overlooked case of SGS, which mimicked uncontrolled asthma and resulted in delayed diagnosis of chronic respiratory symptoms. Fortunately, she was appropriately referred for interdisciplinary management with eventual surgical correction.

ACKNOWLEDGMENTS

We would like to thank the nurses and staff of the allergy and otolaryngology teams for the care of this patient.

AUTHOR'S CONTRIBUTION

Nikhil Crain - Acquisition of data or analysis and interpretation of data; drafting the article; final approval of the version to be published. Jennifer Thompson - concept and design of study; acquisition of data or analysis and interpretation of data; revising it critically for important intellectual content; final approval of the version to be published. Leslie Cristiano - Concept and design of study; acquisition of data or analysis and interpretation of data; revising it critically for important intellectual content; final approval of the version to be published.

CONSENT

All identifying information, including reports, studies, and images, has been anonymized to protect patient confidentiality and comply with ethical policies.

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Funding: Nil; Conflicts of interest: Nil.

How to cite this article: Crain NA, Thompson JL, Cristiano LM. Fixed extrathoracic obstruction masquerading as poorly controlled asthma. Indian J Case Reports. 2025; 11(10):496-498.