

Cervical Lung Herniation Complicating a Case of Acute Asphyxial Asthma in a Child

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Abstract: The abrupt onset of respiratory failure secondary to asthma, known as acute asphyxial asthma (AAA) in adults, is uncommonly reported in children. Here, we report a case of a child with the acute onset of respiratory failure consistent with AAA complicated by the finding of a neck mass during resuscitation. This 11-year-old boy with a history of asthma initially presented in respiratory failure with altered mental status after the complaint of difficulty in breathing minutes before collapsing at home. Initially, his respiratory failure was thought to be secondary to status asthmaticus, and treatment was initiated accordingly. However, a neck mass noted during the resuscitation was cause for concern, and other etiologies for his respiratory failure were considered, including an airway obstructing neck mass. After pediatric surgery and anesthesia consultation for intubation and possible tracheostomy placement, general anesthesia was induced in the operating room with an inhaled anesthetic, with prompt resolution of the bronchospasm and decompression of the neck mass. Review of the imaging and clinical course ultimately yielded a diagnosis of cervical lung herniation as the etiology of his neck mass. We report this case of AAA and cervical lung herniation and a review of the literature of these 2 uncommon phenomena in children.

Key Words: asthma, acute asphyxial asthma, cervical lung herniation

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CASE

An 11-year old boy with a history of chronic persistent asthma was brought to our emergency department (ED) by ambulance in extremis after the acute onset of severe respiratory compromise at home, where paramedics found him in respiratory failure with altered mental status. His sitter called for help when he collapsed minutes after complaining of trouble breathing. There was no report of recent cough, wheeze, or shortness of breath above baseline symptoms or of a choking episode before the acute event. There was no history of recent fever or viral illness, or recent asthma exacerbation. His history was significant for numerous asthma admissions before age 8 years, but none in the preceding year. He had received 5 courses of oral steroids as an outpatient in the last 12 months; he had no history intubation or stays in the intensive care unit. His mother reported he has been compliant with taking his daily controller medications (fluticasone/salmeterol and beclomethasone dipropionate), but had exertional and nighttime symptoms daily and used albuterol at least once a day at baseline. His mother also related that the family just adopted a cat, to which our patient was allergic.

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He was treated by paramedics with intramuscular epinephrine and supplemental oxygen without improvement.

Upon arrival to the ED, our patient was obtunded and had severe distress with agonal respirations at a rate of 5 breaths per minute. He displayed significant work of breathing with supraclavicular, intercostal, and subcostal retractions. There was minimal air exchange throughout auscultation with faint wheezes over the bibasilar lung fields. He was afebrile, tachycardic to 122 beats per minute and hypertensive at 208/126 mm Hg. His extremities were warm and well perfused. There was no evidence of trauma.

Initial resuscitative efforts attempted to relieve critical bronchospasm and severely limited air exchange. The patient was given intramuscular epinephrine, continuous nebulized albuterol with ipratropium, intravenous magnesium sulfate, and subcutaneous terbutaline. Supplemental oxygen and positive pressure ventilation continued without interruption. Despite these interventions, he had no improvement in respiratory effort, oxygenation, or ventilation.

The patient was positioned upright to facilitate air exchange, revealing an anterior right-sided neck mass. It quickly enlarged over the course of the resuscitation and seemed more prominent during inspiration. It was soft, nonindurated, well circumscribed, and did not cross the midline. The trachea remained midline. A lateral neck radiograph demonstrated a lucency that projected over the trachea and esophagus and a thickened epiglottis (Fig. 1).



FIGURE 1. Lateral neck radiograph demonstrating lung protrusion into the neck.

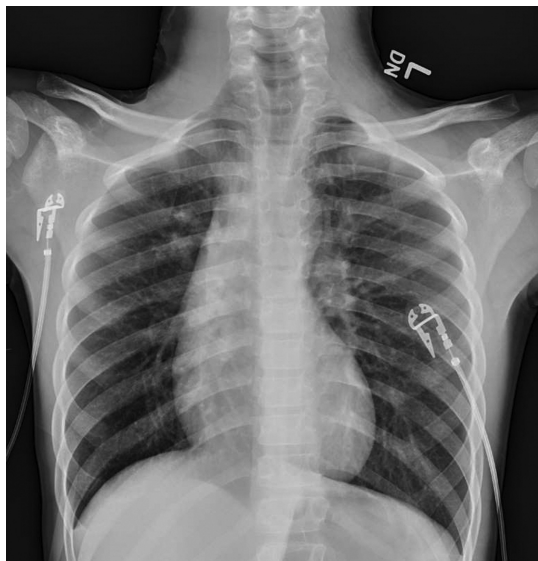


FIGURE 2. Chest radiograph demonstrating right cervical lung herniation.

A chest radiograph also revealed a lower right neck lucency (Fig. 2). Anesthesia, critical care, and general surgery were called to coordinate critical airway management, and the decision to intubate in the operating room was made given the unclear etiology and extent of his airway compromise.

Upon induction with the inhaled anesthetic, sevoflurane, the neck mass promptly decompressed (Fig. 3) relieving airway obstruction and air entry improved from diminished bronchospasm and air trapping. Laryngoscopy and bronchoscopy offered direct visualization of a normal appearing epiglottis, and the airway was clear of any foreign body. The airway was secured with oral intubation without complication. Computed tomography (CT) of the neck immediately after this procedure revealed no abnormality. It was felt that the resolved mass represented a severely hyperinflated right lung herniating beyond the pleural cavity.

The patient remained intubated in the pediatric intensive care unit while his status asthmaticus was managed with steroids and β agonists. A CT scan of the head, completed to evaluate for hypoxic ischemic injury, was concerning for increased intracranial pressure and early cerebral edema. He was treated with hyperventilation and hypertonic saline. Repeat head CT showed no intracranial injury and resolved cerebral edema. Head magnetic resonance imaging confirmed no evidence of ischemia. Respiratory support and therapy were weaned, and he was extubated and discharged home without disability 5 days after presentation.

DISCUSSION

Although asthma is a very common condition in childhood, affecting 9.5% of children in the United States, life-threatening episodes of asthma resulting in respiratory failure and intubation are not common.¹ When present, respiratory failure most typically occurs via a pattern of gradually worsening symptoms and decline of respiratory function over days to weeks.² Less commonly, respiratory failure can occur abruptly, within 6 hours of the onset of symptoms, a phenomenon recognized in adults as acute asphyxial asthma (AAA). Acute asphyxial asthma attacks, unlike slowly progressing asthma exacerbations, tend to have few identifiable triggers and respond more rapidly to asthma therapies.³ In AAA, it is hypothesized that respiratory failure is secondary to intense bronchospasm as opposed to the bronchial inflammation

and mucous plugging seen in more slowly progressive asthma.⁴ Asthma exacerbation with an abrupt progression to respiratory failure, AAA, has only rarely been reported in children.³⁻⁸ One retrospective review by Maffei et al reported 13 cases over 11 years of rapid onset respiratory failure from asthma in children consistent with AAA in patients admitted to their pediatric intensive care unit requiring intubation and mechanical ventilation for status asthmaticus. Similar to what has been reported in adults, they describe patients with an abrupt onset of respiratory failure (requiring intubation within 30 minutes of ED arrival) and rapid response to intubation and asthma therapies. In our patient, the first complaint of difficulty in breathing was minutes before the onset of respiratory failure and his subsequent collapse at home. Additionally, he improved rapidly after the resolution of his bronchospasm with induction anesthesia. This clinical course was consistent with courses of AAA as described in the adult literature and in Maffei's study.

This patient's course was complicated by the finding of a neck mass during his resuscitation which raised the concern for an airway obstructing lesion as the cause of his respiratory failure. The differential diagnosis for neck masses is broad and includes inflammatory, congenital, and neoplastic causes with congenital and inflammatory lesions being the most common.^{9,10} The differential for an intermittent neck mass, expanding with positive pressure ventilation is narrower and includes lesions, such as a pharyngeal pouch, laryngocoele, vascular dilatation, posttraumatic hematoma, and cervical lung herniation (CLH).¹¹ We speculate that our patient's neck mass fit this description because it was initially observed only after continued positive pressure ventilation in the setting of bronchospasm and promptly resolved after induction anesthesia and bronchospasm relief. It was felt that given the progression of symptoms and imaging results, the mass was most consistent with CLH.

Lung herniation is uncommon and defined as protrusion of the pulmonary parenchyma outside of the boundaries of the thoracic cage.¹² This rare disorder was first described by Roland in 1499 and less than 300 cases have been reported to date.¹³ In 1847, lung hernias were classified by Morel and Lavelle according to location and cause. Location classifications include thoracic (herniation through the intercostal space), cervical (through the



FIGURE 3. Resolution of the right cervical lung herniation on the first post-intubation chest radiograph.

apex of the thorax between the sternocleidomastoid and scalene anterior muscles), and diaphragmatic.¹⁴ Etiologic classifications include congenital and acquired, with acquired cases being further subdivided into traumatic, spontaneous, and pathologic causes. Among acquired cases, traumatic causes predominate, accounting for more than 50% of cases (penetrating wounds, fractured ribs, or postsurgical).¹⁵

As described in the case presented here, CLH is typically prevented by the parietal pleura and Sibson's fascia.¹⁶ Herniation occurs when there is either an increase in laxity or absence of 1 or both of these structures allowing protrusion of the pulmonary tissue through the thoracic inlet.¹² CLH typically presents as an intermittent unilateral or bilateral neck mass and is often noted to be transient, worsening with increases in intrathoracic pressure and resolving spontaneously with diminution of that pressure. It is often apparent during coughing, crying, blowing, or Valsalva.¹⁷ Typically, it is asymptomatic but may present with hoarseness, chronic cough, or stridor due to compression of the trachea.¹² Rarely, the lung apices may be tender, but incarceration of the lung tissue has only rarely been reported. The diagnosis can be made by oblique or lateral radiographs of the involved area (neck or chest film).¹⁷ Valsalva at the time of radiograph may be helpful for intermittent lesions; fluoroscopy during a period of crying may be helpful in infants and young children unable to cooperate with Valsalva.¹⁶ In our patient, air trapping associated with his asthma and exacerbated by positive pressure ventilation lead to a persistent neck mass during the period of evaluation which was easily discernible on chest and airway radiographs.

Treatment of CLH is typically not necessary as lesions resolve with reduction in intrathoracic pressure, and complications are rare.¹⁷ Indications for surgical intervention include cosmesis, symptoms of local compression of adjacent structures (eg, airway obstruction or dysphagia), and incarceration. In our patient, the mass resolved with the reduction of intrathoracic pressure after induction of general anesthesia, and no surgical intervention was necessary.

In summary, we have presented a case of AAA and concomitant CLH. This report brings attention to the presence of the phenomenon of rapid onset respiratory failure, or AAA, that occurs in a small subset of asthmatic patients. Additionally, it brings attention to the rare disorder of CLH, which should be considered on the differential diagnoses for an intermittent neck mass that worsens with positive pressure ventilation.

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