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CASE REPORTS

Apical Lung Herniation Associated with Continuous Positive Airway Pressure in a 4-Year-Old Girl

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We report a case of apical lung herniation through the superior thoracic aperture of an obese child using nocturnal CPAP. Lung herniation has been described in association with congenital thoracic abnormalities and elevated intra-thoracic pressure, such as trauma. This patient was hospitalized with community acquired pneumonia and required nocturnal CPAP for treatment of concurrent obstructive sleep apnea. Her lung hernia was discovered incidentally on routine follow-up chest radiography and resolved with cessation of CPAP treatment. Lung herniation in association with the use of continuous positive airway pressure (CPAP) has not been previously described.

Keywords: pulmonary hernia, lung hernia, obstructive sleep apnea, continuous positive airway pressure, sleep-disordered breathing

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INTRODUCTION

Pulmonary herniation is an extension of the lung and pleura beyond their native positions in the thoracic cavity. Pulmonary hernias have been described through the diaphragm, intercostal spaces, and into the cervical space.[1] A hernia involving the cervical space has also been described as apical in the literature. All pulmonary hernias described have been associated with either congenital anomalies and/or elevated intrathoracic pressure, such as trauma or Valsalva-type maneuvers.[2] Most hernias are dynamic and are present only when intrathoracic pressure is elevated. To our knowledge, pulmonary herniation associated with continuous positive airway pressure (CPAP) has not been described. Here, we report a case of cervical lung herniation in a 4-year-old girl associated with continuous positive airway pressure.

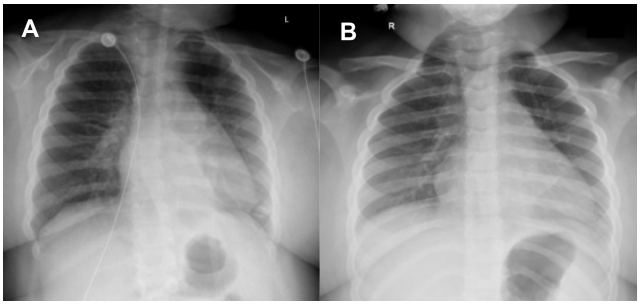
REPORT OF CASE

A 4-year-old morbidly obese girl with a body mass index of 49 and obstructive sleep apnea (apnea-hypopnea index: 28.4 events per hour) was admitted with productive cough, post-tussive retching, fever, shortness of breath, and hypoxia. Prior to this admission, she was hospitalized under observation after tonsillectomy and adenoidectomy. Postoperatively, she continued to require nocturnal CPAP, at 8 cm H_2O, for residual symptomatic airway obstruction, but was not compliant. A chest x-ray on this admission was significant for low lung volumes, atelectasis, and a retrocardiac infiltrate concerning for pneumonia (**Figure 1A**). She was treated for community-acquired pneumonia with amoxicillin, supplemental oxygen, and nocturnal CPAP at 8 cm H_2O. She defervesced and was gradually weaned from supplemental oxygen, but continued to have a productive cough. A follow-up chest x-ray, after four days of consistent CPAP use, detected a right apical pulmonary herniation extending to the superior border of C6 (**Figure 1B**). The patient was asymptomatic at this time except for improving productive cough, occasional post-tussive retching, and mild subclavicular tenderness. No cervical asymmetry, crepitation, or abnormal breath sounds were appreciated at this time. CPAP was discontinued and a repeat chest x-ray the following day showed resolution of the hernia, though she continued to have coughing and post-tussive retching. She was discharged the following day on nocturnal supplemental oxygen, with plans to reattempt CPAP at lower pressures guided by polysomnography. Of note, 6 weeks prior to the current admission, the patient had been hospitalized for pneumonia, at which time she required bilevel positive airway pressure support at 14/8 cm H_2O without any evidence of lung hernia. Two months after the current admission, she was admitted for an orthopedic procedure, which required ventilator support during anesthesia with a mean airway pressure support of 13 cm H_2O with no evidence of recurrence of the hernia on chest x-ray.

DISCUSSION

Apical lung hernias are uncommon. Few more than 50 cases of apical lung hernias have been described in children.[2] The majority are found incidentally, have a benign clinical course, and resolve without intervention. Surgical management is best described in traumatic hernias and has been reserved for lung tissue at risk for strangulation, incarceration, or puncture.[3] In this case, CPAP was discontinued because it posed a theoretical risk of worsening herniation, pneumothorax, and/or

Figure 1



(A) Chest x-ray taken on admission; the lungs are in their native position in the thorax. **(B)** Chest x-ray taken on day four of admission. Right apical lung hernia is seen extending through the superior thoracic aperture to the level of C6.

strangulation. The hernia resolved after the discontinuation of CPAP, which is not surprising as most apical lung hernias are dynamic and reduce after resuming physiologic tidal pressures. To our knowledge, this is the only reported case of apical lung hernia associated with continuous positive airway pressure. The hernia described in this case is best explained by injury to Sibson's fascia from inflamed pulmonary parenchyma, coughing, and retching. This injury allowed the pulmonary tissue to herniate under duress of intermittent Valsalva-type maneuvers and sustained elevation in intrathoracic pressure from CPAP support. Lung hernias in association with Valsalva-type maneuvers alone are well described in the literature.[2] However, we believe the CPAP played a pivotal role in the formation of this hernia, because the x-rays before and after CPAP did not show herniation, despite continued coughing and retching.

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