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CASE REPORT

A Child with Hamman's Syndrome - A Case Report

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Abstract

Objective: To report a rare case of spontaneous pneumomediastinum that occurred in childhood. **Case Report:** Male patient, eight years old, with cough, dyspnea, and goring, with no improvement in asthma treatment, venous corticoid base and inhaled bronchodilator. Patient underwent extensive investigation with imaging tests that suggested and confirmed a diagnosis of pneumomediastinum, excluding external causes - such as trauma or chest surgery - that could be clearly related to the observed characteristics. The patient had spontaneous involution with supportive treatment. **Conclusion:** Diagnostic confirmation syndrome by image, usually of benign evolution, with spontaneous resolution and only need of supportive treatment.

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INTRODUCTION

In most cases, spontaneous pneumomediastinum is a benign, self-limiting condition. It affects mostly young male individuals and is diagnosed in 1/30,000 hospital admissions^{3,4}. The pathophysiology of spontaneous pneumomediastinum has been described as a rupture of terminal alveoli due to increased intra-alveolar pressure causing air to migrate into the interstitial and peribronchial space, hilum, and mediastinum. The penetration of air into the mediastinum during breathing to attain pressure equilibrium is described via the Macklin effect⁵.

Hamman's syndrome is characterized by the occurrence of spontaneous pneumomediastinum unrelated to trauma or surgery, potentially preceded by intense exertion, coughing, vomiting, or drug inhalation. It is a rare and potentially underdiagnosed condition that involves mostly young men¹.

It may cause coughing, dysphonia, chest pain, dyspnea, and stridor. It usually develops in a benign fashion and requires only support therapy with analgesia and resting. Computed tomography is the gold standard diagnostic method².

CASE REPORT

A brown 8-year-old boy with residence in Macaé, Rio de Janeiro, was brought to an emergency unit with dyspnea and coughing lasting for three days. He was in moderate respiratory distress and presented with subcostal and suprasternal notch retractions. Lung auscultation revealed universally audible vesicular breath sounds and wheezing. The patient was diagnosed with asthma and prescribed three courses of nebulized short-acting bronchodilator with oxygen therapy during treatment intervals and intravenous hydrocortisone. At a later examination, the patient still had vesicular breath sounds without adventitious sounds. However, he was still tachypneic. A chest X-ray ordered subsequently did not show alterations and the patient was discharged on a prescription of prednisolone, nebulized bronchodilator, and anticholinergic medication. Since the symptoms did not improve, the patient was back at the emergency unit later on the same day and was hospitalized.

The patient comes from a complex family and social background, which involves physical and sexual abuse. His mother is a drug user who speaks openly about the subject in the presence of her children. His brother was diagnosed with esophagitis of a psychosomatic etiology. The patient's record of diseases includes untreated asthma.

The symptoms appeared suddenly. Neither the patient nor his guardian was able to determine the event that might have triggered the onset of symptoms. The patient was hospitalized for severe asthma based on the observation of symptoms of coughing and dyspnea.

Physical examination revealed vesicular breath sounds with mild wheezing and an oxygen saturation level of 96%. A physician at the emergency unit noted the patient presented

with sighing dyspnea. His prescription included no food, intravenous hydration, methylprednisolone 1mg/kg/dose, a nebulized short-acting bronchodilator, and continuous oxygen therapy with a mask. He was kept on continuous monitoring and tested for venous blood gas level. The patient presented with agitation and dyspnea inconsistent with auscultation findings and reported having a globus sensation in the pharynx when he was examined three hours after the initiation of treatment. His blood gas level was measured again and his prescription was changed to include the alternating use of nebulized adrenaline and short-acting bronchodilator.

After two days in the outpatient clinic, the patient's symptoms worsened as he showed signs of respiratory effort and suprasternal notch retractions, rhonchi as he inhaled, lung auscultation without wheezing, and a globus sensation in the pharynx despite the administration of asthma treatment. Assessment by an otolaryngologist was ordered on account of the rhonchi and respiratory effort. CT scans of the neck and chest showed the patient had pneumomediastinum and paratracheal subcutaneous emphysema extending along the neck (Figure 1).



Figure 1. Chest CT scan showing free air in the mediastinum on January 21, 2019.

A pediatric surgeon examined the patient and prescribed a conservative approach and referral to the intensive care unit (ICU). The patient spent seven days in the ICU, two of which on a ventilator. He underwent an upper gastrointestinal endoscopy and a bronchoscopy, in which anomalies were not found. He was referred to the outpatient clinic after a second CT scan showed spontaneous resolution of the pneumomediastinum, although the patient still complained of a globus sensation in the pharynx and had bouts of dyspnea. He did not present oxygen saturation decreases or wheezing in auscultation. A second bronchoscopy showed no alterations.

During his stay at the hospital, a pediatrics resident following the patient noted that his bouts of dyspnea and respiratory effort were similar to those observed in individuals with anxiety disorder. The psychiatry team was called in and the patient was prescribed fluoxetine and psychology care. His symptoms improved partially and he was later discharged and scheduled for follow-up appointments.

DISCUSSION

The main causes of spontaneous pneumomediastinum are intense physical exertion, labor, high intensity coughing, vomiting, asthma, deep dives, and inhalation of narcotics². The patient described in this case report had asthma.

The most frequently used imaging methods are X-rays and chest CT scans, with the latter ranking as the gold standard method for individuals with Hamman's syndrome². Chest X-rays are generally used in patient screening. However, our patient's X-ray images did not show alterations, a reminder of the fact that other imaging methods might be needed. Chest CT scans should be performed whenever X-ray images are normal or inconclusive, since it may help identify the extent of air dissemination and rule out other associated lesions⁶.

In the case described in this report, CT scans revealed the presence of pneumomediastinum and subcutaneous emphysema. Endoscopy, bronchoscopy, and esophageal imaging are required if dysphagia, vomiting, previous trauma, pleural effusion or digestive tract disease are found, which was not the case for our patient^{2,6}. Our patient underwent upper gastrointestinal endoscopy and bronchoscopy in attempt to find alterations that might explain his symptoms.

In regard to treatment, most studies recommend the adoption of a conservative approach, with resting and analgesia when needed, in light of the benign development of the condition^{4,7}. Most patients progress well, with complete air resorption, as seen in our case^{5,7}.

Spontaneous pneumomediastinum or Hamman's syndrome is a rare condition with a favorable prognosis, known for its benign and self-limiting progression. A diagnosis may be achieved by exclusion, although it is not always properly conducted. Therefore, we must be mindful of the signs of this condition. In addition, clinical history, physical examination, and imaging must be considered in the production of a diagnosis⁷.

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