

An Unusual Cause of Cough: Hamman's Syndrome

Mehmet Unlu¹, Pinar Cimen², Emre Oner³

¹Specialist, Department of Pulmonology, Luleburgaz State Hospital, Kırklareli, Turkey, ²Associate Professor, Department of Pulmonology, Izmir Training and Research Hospital for Thoracic Medicine and Surgery, Izmir, Turkey, ³Senior Resident, Department of Emergency Medicine, Ankara University Medical Faculty, Ankara, Turkey

Abstract

Spontaneous pneumomediastinum or Hamman's syndrome is a rare entity without any predisposing factors. Besides its rarity, it is generally presented with non-specific respiratory symptoms (such as an only cough) and may be misdiagnosed or underdiagnosed. A 16-year-old female presented to the emergency department of our hospital with progressive non-productive cough over the last 2 weeks. She had been prescribed non-specific antibiotics with the diagnosis of upper respiratory tract infection and no improvement occurred in 10 days. Further clinical evaluation of the patient revealed a case of Hamman's syndrome. This syndrome should always be kept in mind especially in young patients presenting with non-specific respiratory symptoms, and if there is a doubt further evaluation should be performed.

Key words: Diagnosis, Pneumomediastinum, Spontaneous, Syndrome, Treatment

INTRODUCTION

Pneumomediastinum is the presence of air within the confines of mediastinal structures which originates from the alveolar space or conducting airways.¹ This entity was first described by Laennec in 1819.² However, first reported case of spontaneous pneumomediastinum belongs to Hamman and the disease was later named in his honor (Hamman's syndrome).³ Hamman's syndrome is a rare medical condition without any apparent predisposing factor or disease. It is triggered by coughing, vomiting, intense exertion, and Valsalva maneuver, all of which cause sudden increase in the intra alveolar pressure without trauma.⁴ It may be misdiagnosed because of its uncommon presentation and non-specific symptoms such as only cough, as seen in this case.

Hereby, we report a case of Hamman's syndrome misdiagnosed as upper respiratory tract infection.

CASE REPORT

A 16-year-old female presented to the emergency department of our hospital with a progressive non-productive cough over the last 2 weeks. She was a non-smoker, and there was no past history of a chronic disease. She had never consumed illicit drugs and had no allergies. She had visited an otorhinolaryngologist 10 days ago and was prescribed non-specific antibiotics with the diagnosis of acute pharyngitis.

The patient presented with a respiratory rate of 14 breaths/min, heart rate of 70 beats/min, and blood pressure of 120/70 mmHg. The 12-lead electrocardiogram was without any rhythm abnormalities. Chest auscultation revealed the presence normal lung sounds. No crepitus detected on both sides of the neck. There was no sign of an obvious upper respiratory tract infection. Hemogram and results of blood chemistry were within normal limits. Erythrocyte sedimentation rate was 8 mm/h and serum level of C-reactive protein was 0.1 mg/dl. Plasma level of D-dimer calculated by enzyme-linked immunosorbent assay was <500 µg/L and Well's score for pulmonary thromboembolism was 0.

Her chest radiograph demonstrated a linear air shadow along the left upper border of trachea (Figure 1). She gave no history of trauma. Thus, she underwent computerized

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Corresponding Author: Mehmet Unlu, Lüleburgaz Devlet Hastanesi 2. Bina Göğüs Hastalıkları Polikliniği, Lüleburgaz, Kırklareli, Türkiye.
 Phone: +0090-507-6507616. E-mail: lidokain21@hotmail.com

tomography (CT) of the thorax with the prediagnosis of Hamman's syndrome and CT revealed the presence of air trapping in the mediastinum (Figure 2). There was no sign of a concomitant pneumothorax or subcutaneous emphysema.

The patient was hospitalized, and nasal oxygen therapy was administered during the bed rest. She was discharged on the 5th day of follow-up as her complaints disappeared. Last contact with her was a polyclinic visit for control 3 months after discharge, and she was free of all previous symptoms.

DISCUSSION

The pathophysiology of this syndrome was described by Macklin and Macklin based on the results of an animal study.⁵ According to their explanation, following the terminal alveolar rupture (primary pathology), alveolar air passes through the perivascular interstitial

tissue towards the hilum. Then, it reaches mediastinum and is being trapped among the mediastinal structures. Besides, pneumomediastinum may be complicated with subcutaneous emphysema or pneumothorax in 40-100% of the cases, if intrathoracic air leaks into the adjacent soft tissues.⁶

Hamman's syndrome occurs more frequent in young men. Although the exact incidence of this disease is still subject of debate, it is estimated to be present in approximately 1/30,000 emergency department referrals.⁷

Chest and neck pain, dyspnea, hypotension, dysphagia, subcutaneous emphysema, and cough are the common features of Hamman's syndrome. Chest pain is usually retrosternal and may radiate to the neck or into the back. In almost all cases, physical examination reveals no abnormality. Palpable crepitus is only can be detected in patients complicated with subcutaneous emphysema, so it may be absent in half of the patients.⁶ Precordial crunching sound synchronous with heart beat is the characteristic of this syndrome and called as Hamman's sign. However, again it is present approximately only in half of the cases. In this case, non-productive cough was the only symptom of the patient. There was no palpable crepitus and Hamman's sign was absent.

For the diagnosis of Hamman's syndrome, all other secondary causes of pneumomediastinum should be ruled out. These causes include penetrating or blunt trauma to the chest, forceful vomiting (Boerhaave's syndrome), medical procedures such as bronchoscopy and esophagoscopy, esophageal and tracheobronchial rupture, and dental procedures. Besides, some studies reported usage of cocaine and marijuana, and the presence of asthma (usage of bronchodilators) as the secondary causes of pneumomediastinum.^{6,8} All these secondary reasons were excluded in this case.

The high degree of suspicion is very important for the establishment of the diagnosis.⁹ There is no consensus on the investigation of this disease. Some authors point to the chest radiography (combination of posteroanterior and lateral graphs) as being sufficient in nearly all cases and CT is recommended only in doubtful cases.^{3,10} However, it should be remembered that chest radiography may be normal on admission and CT is the gold standard in detecting mediastinal air. CT is also accurate in diagnosing tracheobronchial and esophageal ruptures. Electrocardiography may demonstrate non-specific ST segment changes, reduced voltage, and axis deviations in some cases.¹¹ In this case, posteroanterior chest radiography revealed the presence of air shadow suggesting

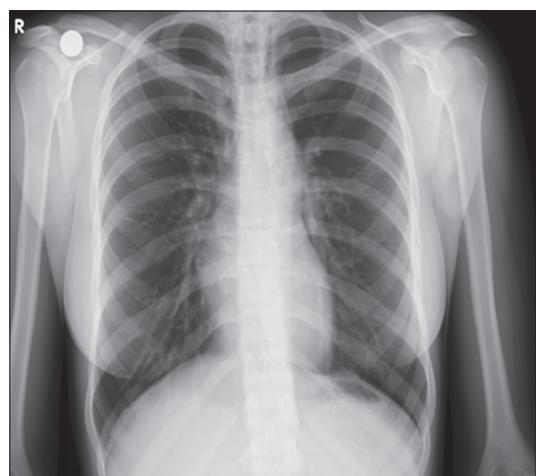


Figure 1: Chest radiograph of the patient on admission

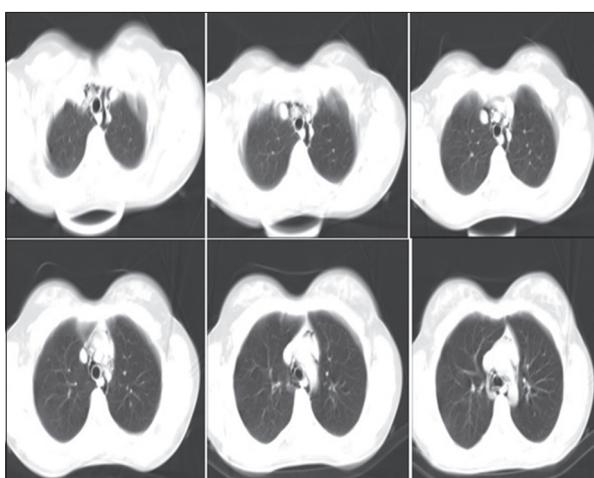


Figure 2: Thorax computerized tomography images of the patient on admission

pneumomediastinum and CT was ordered for the correction of the pre-diagnosis.

The treatment of this syndrome includes bed rest, analgesics if needed and oxygen administration. It is usually benign and non-recurrent. The patient should be hospitalized for a minimum of 24 h to prevent potential complications.⁸ In most cases, spontaneous pneumomediastinum resolves within several days, as seen in this case. Administration of antibiotics is only recommended in cases presented with signs of an infection or mediastinitis. However, there is also a life-threatening condition called as malignant pneumomediastinum which is characterized by the presence of excess air in the mediastinum. In such cases, subcutaneous aspiration and incisions may be required to evacuate mediastinal air, and if subcutaneous aspiration is not sufficient cervical mediastinotomy should be considered.¹²

CONCLUSION

Hamman's syndrome should always be kept in mind especially in young patients presenting with non-specific respiratory symptoms and if there is a suspicion further radiological evaluation should be performed.

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