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Case Report

IDIOPATHIC PNEUMOMEDIASTINUM – CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT

Idiopathic pneumomediastinum is a form of spontaneous pneumomediastinum without predisposing factors and precipitating factors. **The purpose** of this publication is to present a rare case of this pathological condition.

Case presentation: A 21year-old female with spontaneous pneumomediastinum was revealed. Medical history, physical examination, imaging and interventional diagnosis couldn't determine any predisposing and precipitating factors for pneumomediatinum: case of idiopathic pneumomediastinum with unusual widespread subcutaneous emphysema.

Conclusion

In conclusion, this publication refers to one relatively rare condition in the thoracic pathology – idiopathic pneumomediastinum. Being a case of idiopathic pneumomediastinum, there was an unusual clinical presentation of widespread subcutaneous emphysema. Despite widespread subcutaneous emphysema, the presenting case confirms the good prognosis of idiopathic pneumomediastinum.

Key words: spontaneous pneumomediastinum, idiopathic pneumomediastinum, Hamman's syndrome.

INTRODUCTION

Spontaneous pneumomediastinum (SPM) is a rare condition, first described by Hamman in 1939 (1-4). It was defined as non-traumatic presence of free air in the mediastinum in a patient with unknown underlying pulmonary disease. However, it should be noted the wider using of the term spontaneous pneumomediastinum. Nowadays, it is generally accepted that SPM can occur with underlying pulmonary disease, being a predisposing factors e.g. asthma, interstitial and other lung diseases, tobacco, inhaled drug use, corticosteroids, inhalation of irritants (2, 5, 6). While predisposing factors favor SPM, there are so called precipitating factors which are direct causes of this pathological condition - triggers. The precipitating factors cause sudden increase of intra-alveolar pressure with alveolar rupture. The most common precipitating factors for SPM are: emesis, cough, defecation, intense physical exercise, labor, and Valsalva maneuvers (3, 7, 8).

The term idiopathic pneumomediastinum should be reserved only for those cases of spontaneous pneumomediastinum without predisposing and precipitating factors. The cases of idiopathic pneumomediastinum are very rare with limited number of publications in the literature. The purpose of this publication is to present a rare case of idiopathic pneumomediastinum with widespread subcutaneous emphysema. Based on the presenting case, the review of the literature is made.

CASE PRESENTATION

A 21year-old female with no prior medical history was presented to the emergency department of the University Hospital "St. George" – Plovdiv. The reason for her visit was one-day history of progressive swelling of her upper chest, neck, face and arms, chest pain and tightness in throat.

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The woman was afebrile, with normal hemodynamic parameters and showed no signs of respiratory distress. Physical examination revealed subcutaneous emphysema (palpable diffuse crepitus) of the face, neck, chest, shoulders, upper part of the arms and upper part of the abdomen.

An erect postero-anterior chest x-ray was obtained and revealed clear lungs and pneumomediastinum, based upon the presence of air dissecting along the mediastinum **(Figure 1)**. After woman's admission at the Department of Special Surgery, high resolution computerized tomography (CT) scan of the neck, chest and abdomen was performed. Pneumomediastinum with extensive soft tissue emphysema was established **(Figure 2A, B, C, D).**

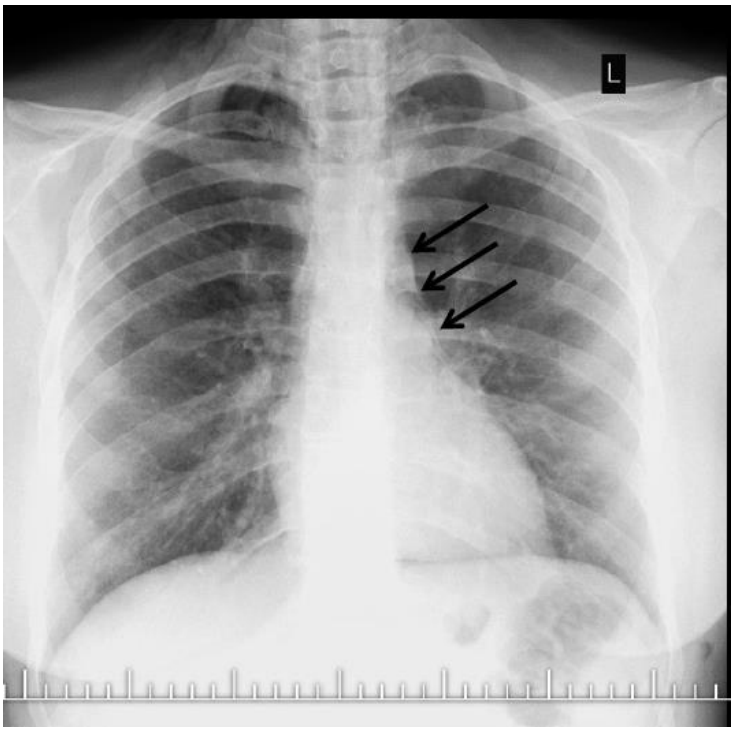

Figure 1. Anteroposterior chest radiograph. The lungs are clear and there is air in the mediastinum (double line around the cardiac silhouette pointed by arrows).

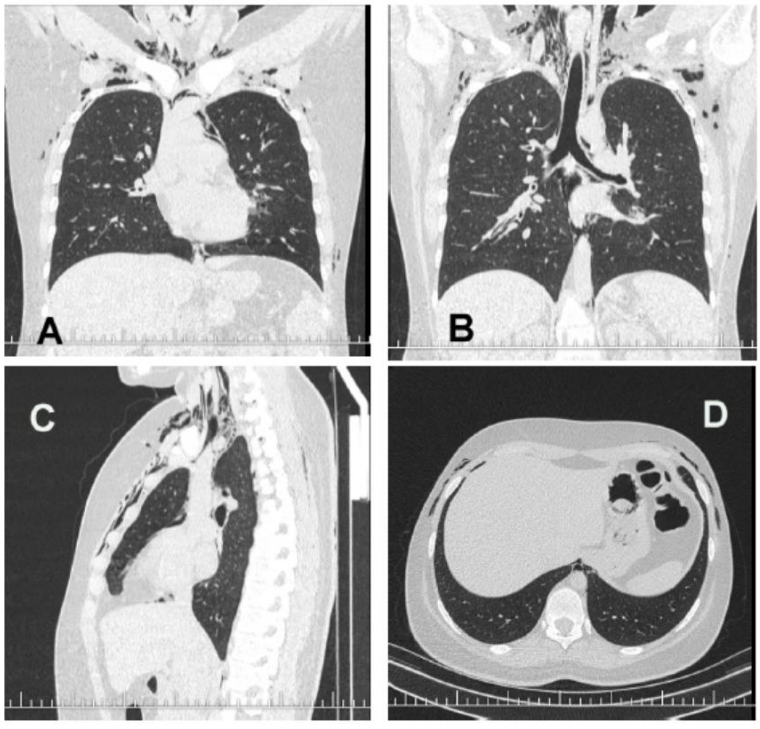

Figure 2. A, B, C, D. Computed tomography scan of neck, chest and abdomen. Pneumomediastinum and soft tissue emphysema are presented.

Upper digestive contrast study with oral gastrografin was carried out and no extravasation of the contrast was reviewed **(Figure 3).** Bronchoscopy was undertaken with normal finding.

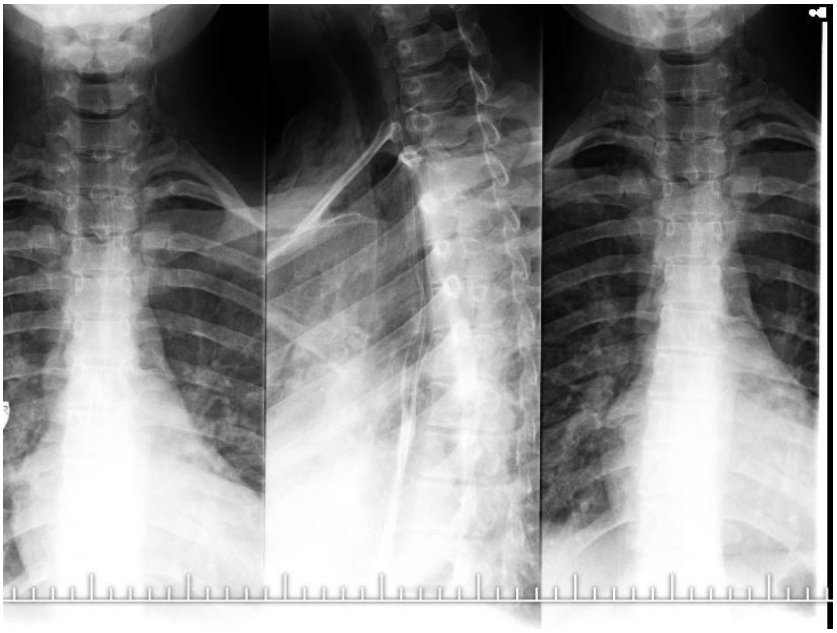

Figure 3. Upper digestive contrast study: normal esophagogram.

Having no predisposing and precipitating factors this case was accepted as idiopathic pneumomediastinum. The woman was managed by conservative approach – rest, analgesia and observation. Almost complete resolution of pneumomediastinum and subcutaneous emphysema was established and she was discharged in a good health at the 4th day of her hospitalization.

DISCUSSION

Spontaneous pneumomediastinum (Hamman's syndrome) is a self-limited, benign condition that affects young patients with ages ranging from 17 and 25 years. The incidence rate is extremely low, with the condition being observed in approximately 1:30,000 hospital admissions (2, 9, 10).

Originally, spontaneous pneumomediastinum is a nontraumatic presence of air in the mediastinum with unknown underlying pulmonary disease. There is some controversy as to whether pneumomediastinum associated with preexisting lung disease should be considered spontaneous pneumomediastinum. Yet, in the medical literature, such cases are not excluded. It is accepted that SPM can occur in patients with underlying pulmonary disease that serves as predisposing condition. Such predisposing conditions for development of SPM are: bronchial asthma (the most common), lung emphysema, lung fibrosis, tobacco, inhaled drugs, etc.

With or without pre-existing medical conditions, the pathophysiologic process of SPM is the same e.g. sudden increase in intra-alveolar pressure initiated by triggers (precipitating factors). Increased intra-alveolar pressure causes alveolar ruptures, air dissection along broncho-vascular sheaths and spreading of this pulmonary interstitial emphysema into the mediastinum. However, there are cases of spontaneous pneumomediastinum with neither predisposing nor precipitating factors which are very rare. For these cases the term idiopathic pneumomediastinum is reserved. The case that is described presents exactly that form of pneumediastinum - idiopathic.

The clinical diagnosis of SPM is based on the symptom triad of chest pain, dyspnea, and subcutaneous emphysema. Subcutaneous emphysema may be either discreet or evident inducing crepitus or facial swelling and its presence varies from 40 to 100%, in different reports. In the presented case the subcutaneous emphysema was widespread that is unusual for the form of idiopathic pneumomediastinum.

Being a benign and self-limited condition some authors consider that not all of the patients with SPM should be hospitalized, particularly those with idiopathic. However, the opposite consideration for the admission to hospital for at least 24 hours to avoid any potential complications exists. According to the protocol of my department all patients with SPM are hospitalized and procedure was performed in the presenting case.

Once pneumomediastinum is suspected, the diagnosis can be confirmed by chest radiograph (3, 5, 11, 12). If the chest radiograph is diagnostic of pneumomediastinum and there is no risk of perforated intrathoracic viscus, no further investigations are required. When chest roentgenogram finding is inconclusive, chest CT scan appears to be necessary to establish a correct diagnosis and to exclude any underlying pulmonary disease. Other supplementary examinations include: esophagogram, which is indicated to rule out esophageal perforation, and bronchoscopy, which can help evaluate the tracheobronchial tree. Widespread subcutaneous emphysema and no pre-existing medical conditions and precipitating factor of SPM explain why chest CT scan and other supplementary examinations were used in the presenting case.

The spontaneous pneumomediastinum is a benign condition and its management is conservative. There are three main approaches to the treatment of SPM: rest, oxygen therapy, and analgesia (2, 5, 6, 11, 12). Patients respond well to this treatment: clinical manifestations resolve and radiographic signs of the condition diminish, that was confirmed by the presenting case. Antibiotics are generally used only if there is a respiratory infection (as a coexisting pathology and underlying trigger of SPM).

CONCLUSION

In conclusion, this publication refers to one relatively rare condition in the thoracic pathology – spontaneous pneumomediastinum. The presenting case is unusual due to two circumstances. At first, there were not determined any predisposing and precipitating factors for the pneumomediastinum - a rare case of idiopathic pneumomediastinum. On the second, being a case of idiopathic pneumomediastinum, there was an unusual clinical presentation of widespread subcutaneous emphysema. Finally, despite widespread subcutaneous emphysema, the presenting case confirms the good prognosis of idiopathic pneumomediastinum.

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