CASE REPORTS

An Unusual Case of Primary Spontaneous Tension Pneumothorax in a Jamaican Female

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ABSTRACT

Spontaneous pneumothorax is a well-recognized entity with a classical presentation of acute onset chest pain and shortness of breath. It may be complicated by the development of a tension pneumothorax or a haemopneumothorax. We report an interesting case of a spontaneous tension haemopneumothorax which presented atypically and was diagnosed on computed tomography (CT) scan of the chest. The clinical and pathophysiological characteristics and treatment of this unusual entity is discussed.

Keywords: Jamaican female, spontaneous pneumothorax

Un Caso Inusual de Neumotórax Espontáneo Primario por Tensión en una Mujer de Jamaica

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RESUMEN

El neumotórax espontáneo es una conocida entidad patológica cuya manifestación clínica clásica consiste en un dolor de pecho agudo inicial y dificultad para respirar. Se puede complicar con el desarrollo de un neumotórax por tensión o un hemoneumotórax. Reportamos como caso interesante un hemoneumotórax espontáneo por tensión, el cual se presentó de forma atípica, y fue diagnosticado mediante escaneo con tomografía computarizada (CT) del tórax. Se discuten las características clínicas y fisiopatológicas, así como el tratamiento de esta entidad patológica inusual.

Palabras claves: Neumotórax espontáneo, mujer Jamaicana

West Indian Med J 2014; 63 (3): 274

INTRODUCTION

A pneumothorax is defined as "air in the pleural cavity" (1). It can arise spontaneously or after blunt or penetrating chest trauma. A spontaneous pneumothorax is classified as primary or secondary. A primary spontaneous pneumothorax occurs in a previously healthy patient with no lung disease while a secondary spontaneous pneumothorax is associated with parenchymal lung disease such as emphysema or pulmonary fibrosis (2). The reported incidence of primary spontaneous pneumothorax is 18 to 28 per 100 000 per year in men and 1.2 to six per 100 000 per year in women (1). Mortality is greater in men than in women.

A tension pneumothorax is usually associated with trauma or mechanical ventilation and the incidence of spontaneous development is rare. A spontaneous tension pneumothorax may complicate one to three per cent of unrecognized pneumothoraces (3). This feared complication typically presents with obvious respiratory distress and signs of cardiovascular instability requiring emergency needle decompression and tube thoracostomy.

Presented is an interesting case of a young female who presented to the Emergency Room with right-sided chest pain

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 DOI: 10.7727/wimj.2013.301

and was found to have a spontaneous right tension pneumothorax diagnosed on radiological imaging. The clinical presentation, risk factors and pathophysiology are discussed.

CASE REPORT

A 21-year old female presented to the Emergency Department (ED) of the University Hospital of the West Indies (UHWI) with a one-day history of chest pain. She had no known medical illnesses and was well until the evening prior to presentation when she developed sudden onset right-sided chest pain with chest tightness post-coitus. This was described as sticking in nature, with worsening on deep inspiration and movement and relieved by leaning forward. There was an associated dry cough but no haemoptysis. There was no report of trauma, shortness of breath or palpitations. She had no significant past medical or surgical history, but reported a two-year history of smoking one to two cigarettes per day. The patient reported heavy menses with no associated dysmenorrhoea or menstrual irregularity. There was no personal or family history of bleeding diathesis. The patient had experienced these symptoms on two occasions in the past (once nine months previously and another episode one year before that). She visited a family doctor on those occasions. No abnormality was found and her symptoms resolved.

Examination revealed a young female, awake and alert but in moderate painful distress. Her respiratory rate was 24 breaths per minute with an oxygen saturation of 100%. Her pulse was 74/minute, blood pressure 118/72 mmHg and temperature 35.8 °C. The examining physician noted tenderness along the right posterolateral chest wall along the eighth and tenth ribs. Breath sounds and percussion note were documented as normal. An electrocardiogram (ECG) revealed sinus rhythm with a normal axis and an S1Q3T3 pattern. Pulmonary embolism was considered and a computed tomography pulmonary angiogram (CTPA) requested. This revealed a right-sided pneumothorax with contralateral deviation of the mediastinum (Figure). The radiological impression was that of a right tension pneumothorax. The patient was immediately returned to the ED. Re-examination of the chest by a senior physician revealed markedly decreased air entry on the right side with mild tracheal deviation. A right thoracotomy tube was immediately placed under sterile conditions. She received parenteral analgesia and was referred to the cardiothoracic service.

While in the ED, approximately 350 milliliters of blood was drained from the thoracostomy tube. The patient remained haemodynamically stable and was subsequently admitted to the surgical ward. In the ensuing 24 hours of admission, 1750 ml of blood was drained *via* the thoracostomy tube. Her haemoglobin count dropped from 10.6 grams per decilitre (g/dL) to 8.2 g/dL. A prolonged prothrombin time of 20 seconds (control 14 seconds) was also noted. Packed red cells and fresh frozen plasma were ordered for the patient. Vitamin K 10 mg and tranexamic acid one gram were administered parenterally. Emergency posterolateral thoracotomy and mechanical pleurodesis were performed. Five hundred millilitres of clotted blood was found in the pleural space. The right lung parenchyma, pleura and diaphragm were normal in appearance. A coagulopathy was entertained but subsequently excluded. Thoracostomy tube drainage remained high and a chest X-ray done on day three of admission (day two post surgery) showed a large right haemothorax. Her haemoglobin dropped further from 6.5 g/dL to 4.4 g/dL, prompting exploration of the right haemothorax. Seven hundred milliliters of blood and clots and a collapsed right lung with few apical bullae was observed. A small (less than 0.5 cm) tan lesion noted on the dome of the right hemi-diaphragm was biopsied. Histopathologic examination subsequently revealed it to be an organized clot. No endometriotic deposits were identified.

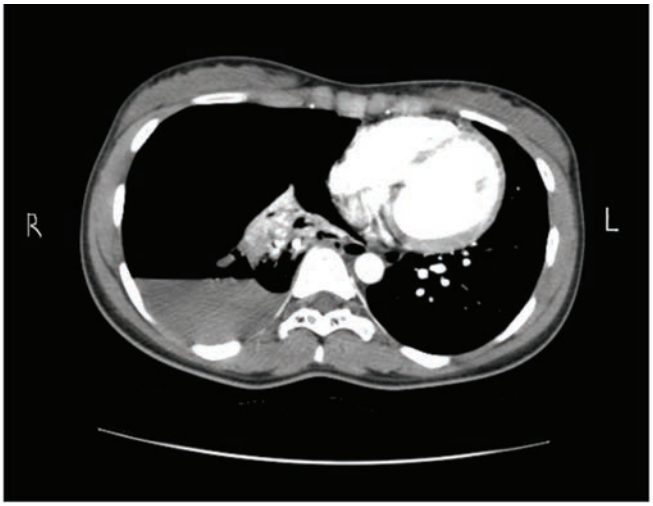

Figure: Computed tomography pulmonary angiogram showing a large right-sided pneumothorax with contralateral deviation of the mediastinum. There was no evidence of pulmonary embolism. (Image courtesy of the Radiology Department, University Hospital of the West Indies)

Subsequently, the patient's chest tube drainage became minimal. A chest X-ray done on day eight of admission after removal of the thoracostomy tube showed complete re-expansion of the right lung. Her haemoglobin rose to 13.3 g/dL post transfusion of three units of packed red blood cells and her prothrombin time normalized. She had received four unit of fresh frozen plasma. The patient remained stable and was discharged three days post re-exploratory surgery.

DISCUSSION

Primary spontaneous pneumothorax (PSP) occurs in patients with no underlying lung disease. In 80−90%, subpleural bullae are found on computed tomography (CT) imaging or at surgical exploration (1, 2). An increase in pleural porosity secondary to inflammation is another proposed mechanism by which PSP may occur (2). Factors which may be related to the occurrence of blebs, bullae and pleural porosity include distal airway inflammation, distal bronchial tree anomaly,

connective tissue disorders, local ischaemia and malnutrition (2). The disease is mainly seen in healthy, tall, thin, young males who smoke (1). The index case was a healthy, thin, young female who admitted to a two-year smoking history. She remained at home for several hours before seeking medical care. Forty-six per cent of patients with PSP wait more than two days before seeing a physician (4). Indeed 10% are asymptomatic (4). Recurrence rates range from 16−52% with 10 years of follow-up (5). Most recur within 6−12 months of first episode (5). The index case reported having similar symptoms on two previous occasions which resolved. It is quite likely that she had previous small pneumothoraces which resolved spontaneously. Previous episodes of PSP would have likely resulted in adhesions and placed her at risk for the haemothorax which developed during this admission. The adhesions may also explain why, despite developing tension pneumothorax, the patient remained haemodynamically stable. It is quite likely that by tethering the lung, the adhesions limited the degree of tracheal deviation, preventing significant mediastinal shift and large vessel kinking.

Primary spontaneous tension pneumothorax is a rare complication of PSP. These patients typically present in extremis with obvious signs of respiratory distress and cardiovascular compromise due to impaired venous return and decreased cardiac output as a result of mediastinal shift (1). Examination may reveal decreased breath sounds, decreased chest wall expansion, hyper-resonance, decreased tactile vocal fremitus, displaced apex beat and tracheal deviation (2). In patients with pneumothoraces that occupy less than 15% to 20% of the hemithorax, these signs may be absent (2). A spontaneous tension pneumothorax may not present with classical signs such as haemodynamic instability (4, 6), as was evident in this patient. One case report indicated that a 19-year old male who was diagnosed with a first episode of primary spontaneous tension pneumothorax on chest X-ray was tachypnoeic and tachycardic but remained haemodynamically stable (7). Holloway and Harris reported on four cases of spontaneous pneumothorax. In three of the four patients, the diagnosis of tension pneumothorax was made radiologically. Two of these four patients were clinically stable with no tracheal deviation. In one of those four cases, the treating physician elicited the signs of spontaneous pneumothorax and tracheal deviation but did not believe his assessment because the patient looked so well (8). Simpson and colleagues commented on nine patients with PSP who were diagnosed with tension pneumothorax on radiological imaging but had no classic clinical features of a tension pneumothorax (9).

Spontaneous haemopneumothorax occurs when greater than 400 ml of blood accumulate in the pleural cavity in association with a spontaneous pneumothorax (10). Aberrant blood vessels which grow from the chest wall through adhesion bands into pleural lesions are thought to be torn when lung collapse occurs (2). Bleeding blood vessels may also arise from the surface of ruptured bullae (2).

In a review of 18 cases, the most frequent lateralizing sign of a tension pneumothorax was decreased air entry (1). A more thorough clinical examination should be done to assess for the presence of mediastinal shift which may be evidenced by tracheal deviation, displaced apex beat and resonance over the sternum (8). Distended neck veins and reversible Horner's syndrome may also be apparent in patients with tension pneumothorax (11). The index patient had an S1Q3T3 pattern noted on her ECG. This was previously reported in an 18-year old male with a right-sided PSP and reversed with treatment (12). This case highlights the importance of history-taking and proper clinical examination and demonstrates that a tension pneumothorax does not always present in extremis.

There are rare female-specific causes of spontaneous pneumothorax, namely catamenial pneumothorax and pneumothorax with pulmonary hamartoangiomyomatosis (13). Catamenial pneumothorax, defined as "a recurrent pneumothorax occurring within 72 hours from the onset of menstruation", is the most common manifestation of thoracic endometriosis (13, 14). It is usually unilateral and right-sided (14, 15). Patients with catamenial pneumothorax often have a history of pelvic endometriosis or infertility (14). There have been reports of cases occurring during or shortly after sexual intercourse (14). In Jamaica, two to six per cent of cases of spontaneous pneumothorax in females in the reproductive age group are associated with pleural endometriosis (16). Hamartoangiomyomatosis is said to occur predominantly in young to middle-aged women and has an overall low incidence (13).

Surgical strategies for the management of PSP include open thoracotomy and pleurectomy or video-assisted thoracoscopic surgery (VATS) with pleurectomy and pleural abrasion (1, 2). An open approach offers lower recurrence rates while a less invasive approach with VATS has the advantages of lower morbidity, less postoperative pain, improved pulmonary function and decreased length of hospital stay (2). Open thoracotomy is the procedure of choice in patients with active bleeding and haemodynamic instability (17) and was chosen in the index case.

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