Catamenial Pneumothorax: Still a Rare Syndrome.

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SUMMARY

A case of catamenial pneumothorax; a rare type of spontaneous pneumothorax is reported in a 30 year old nursing officer. She presented with recurrent right pneumothorax coinciding with her menstrual period. There was no history of cough, fever, smoking or chest trauma and she was not a known asthmatic.

Investigation confirmed the right pneumothorax, and closed tube thoracostomy drainage and chemical pleurodesis added to the retreatment resulted in cure.

KEYWORDS: *Catamenia, Pneumothorax, Endometriosis, Thoracostomy, Menstruation, Pleurodesis.*

INTRODUCTION

Pneumothorax is the presence of air within the pleural space. Pneumothorax may be spontaneous, or due to traumatic, iatrogenic, or disease related event[1]. A primary spontaneous pneumothorax occurs without any known cause or evidence of diffuse pulmonary disease. It results from rupture of small subpleural air cysts (blebs). Catamenial pneumothorax occurs due to escape of air from alveoli during shedding of the abnormal endometrium present in the superficial part of the lung (endometriosis). This occurs pari pasu with menstrual period, being influenced by the same hormonal mileu. Neonatal pneumothorax may result from rupture of congenital cystic lesion (examples: congenital lung cyst, congenital cystic adenomatoid malformation, congenital lobar emphysema and bullous emphysema), neonatal staphylococcal pneumonia with pneumatosis and alveolar rupture from vigorous ventilation.

A pneumothorax compresses lung tissue and reduces pulmonary compliance, ventilatory volumes, and diffusing capacity. These pathophysiologic consequences depend primarily on the size of pneumothorax and condition of underlying lung[2].

Treatment options for primary and secondary pneumothorax are similar. Treatment with tube thoracostomy drainage alone has a high recurrence rate. Effective treatment should include chemical or surgical pleurodesis in combination with complete lung re-expansion and effective sealing of air leaks, in addition to treatment of underlying lung disease.

Table 1: classification of pneumothorax[1]

1. Spontaneous
   a. Primary
   b. Secondary
      i. Chronic obstructive pulmonary disease (COPD)
      ii Bullous disease
      iii Cystic fibrosis
      iv Pneumocystis related cysts
      v. Idiopathic pulmonary fibrosis
      vi. Pulmonary embolism
   c. Catamenial
   d. Neonatal

2. Traumatic
   a. Penetrating
   b. Blunt
3. Iatrogenic
   a. Mechanical ventilation
   b. Thoracentesis
   c. Lung biopsy
   d. Venous catheterization
   e. Post surgical
4. Other
   a. Oesophageal perforation

CASE REPORT

We report a 30 year old unmarried nursing officer from Kano, Nigeria. She presented in our emergency unit with two-week history of shortness of breath and right sided sharp and pleuritic chest pain. There was no history of trauma, cough, fever, and contact with a chronically coughing patient, cigarette smoking or previous history of chronic lung disease. There were no symptoms of cardiac decompensation and she was not a known asthmatic. She was previously in good health. The onset of symptoms coincided with her menstrual period.

On examination she was an acutely ill-looking young lady who was dyspnoeic at rest, not cyanosed and no significant peripheral adenopathy or pedal oedema. She was tachypnoeic with the trachea shifted to left, reduced chest wall excursion and expansion on the right, no compression tenderness, diminished tactile fremitus, hyper- resonant percussion note and diminished intensity vesicular breath sound over the right lung zones. Other systems examination findings were essentially normal. The clinical diagnosis of

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right spontaneous pneumothorax was confirmed by urgent chest radiogram which showed 66% right pneumothorax, with mediastinal displacement to contra-lateral side, and no apparent cause (appendix I).

She was admitted and emergency right closed thoracostomy drainage done and connected to underwater seal drainage bottle. This was complemented with chest physiotherapy and by the second day, there had been complete re-expansion of the lung, chest tube removed on the third day and check radiogram on the fourth day before discharge showed full lung expansion of apparently normal lungs with no hilar or parenchyma infiltrates and air cysts (appendix II). Full blood count, erythrocyte sedimentation rate, and Mantoux test results were all normal.

At first follow-up visit in our surgical outpatient clinic two weeks after hospital discharge, which coincided with her last menstrual period, she complained of a new onset of gradually worsening shortness of breath and right sided chest pain. There were no other symptoms. Re-evaluation and chest radiograph confirmed recurrent right pneumothorax (30%) probably of catamenial type (appendix III). She was re-admitted for emergency right closed tube thoracostomy drainage, and chest physiotherapy. Following complete re-expansion of the lung on the second day (appendix IV), chemical pleurodesis using 1000mg of tetracycline was done before chest tube removal on the fifth day. She was discharged home on the sixth day of re-admission and has remained free of recurrence six months after re-treatment.

DISCUSSION

[v]Catamenial pneumothorax is recurrent spontaneous pneumothorax occurring within 72 hours of onset of menstruation. It is a rare syndrome with prevalence of 1-5% among menstruating women and is attributed to intra-thoracic endometriosis. However concomitant pelvic endometriosis is present in only 61% of patients[3]. Our reported patient did not have symptoms suggesting peritoneal endometriosis. The mean age at presentation varies between 30 and 37 years with a range of 19 to 54 years. The index patient was 30 years old.

The most common symptoms are chest pain and dyspnoea. It occurs predominantly in the right pleura, as in our reported patient. Three mechanisms of causation of catamenial pneumothorax have been proposed; metastatic, hormonal and anatomic theories. The metastatic theory of Lillington (1972)[4] suggests migration of endometrial tissue via peritoneal cavity through transdiaphragmatic lymphatic channels or diaphragmatic fenestrations, or haematogenously into the pleural space. Because, these channels and fenestrations are more common in right hemidiaphragm, therefore catamenial pneumothorax occurs predominantly in right pleural space[4].

The hormonal theory proposed by Rossi and Goplerud[5] in 1974 suggests that high serum level of prostaglandin F_2 at ovulation may lead to vasospasm and associated ischaemia in the lungs with alveolar rupture and pneumothorax. However this cannot explain the preponderance of right sided involvement. Also neither is there clinical features of bronchospasm nor non-steroidal anti-inflammatory medications capable of preventing recurrence of catamenial pneumothorax in respective reported series.

The anatomic theory is based on influx of air into the pleural space from the peritoneal cavity via diaphragmatic fenestrations[6]. The loss of cervical mucus plug during menstrual cycle allows influx of air from atmosphere into peritoneal cavity. The same diaphragmatic fenestrations are implicated in the pathogenesis of the predominantly right sided pleural effusion in patients with hepatic hydrothorax and Meig's syndrome[6]. Also concomitant pneumoperitoneum is found in some patients with catamenial pneumothorax although our reported patient did not have this. However induced pneumoperitoneum during laparoscopy does not frequently lead to pneumothorax and catamenial pneumothorax has occurred after hysterectomy[2,3].

Alternatively, it has been postulated that endometrial tissue may be deposited in the chest cavity during embryonic development[1]. Monthly shedding of such tissue results in pneumothorax.

Our reported patient, was clinically diagnosed as having catamenial pneumothorax because she falls into the age range of catamenial pneumothorax, her pathology was on right side, was recurrent, coincided with her menstrual cycle and no other cause of the pneumothorax was found. In addition to chest radiogram, thoracoscopy for evaluation of the lung, the pleura and the right hemi-diaphragm should be undertaken where facility is available, for discounting natural pneumothorax due to bulla, observation of diaphragmatic defective pores or fenestrations and biopsy of allopatric endometriosis in the pleura[7].

However, this was not done for our reported patient because of unavailability of thoracoscope.

Treatment of our patient focused on surgical therapy which included drainage of the pneumothorax for complete lung re-expansion and induction of chemical pleurodesis using 1.0g of tetracycline powder instillation into the pleural space. Other efficacious pleurodesants include bleomycin and talc powder. Other acceptable modalities of treatment include surgical closure of diaphragmatic fenestrations where present and surgical pleurodesis, and hormonal therapy. The surgical therapy can be done through video-assisted thoracoscopic surgery (VATS)[8] or open thoracotomy.

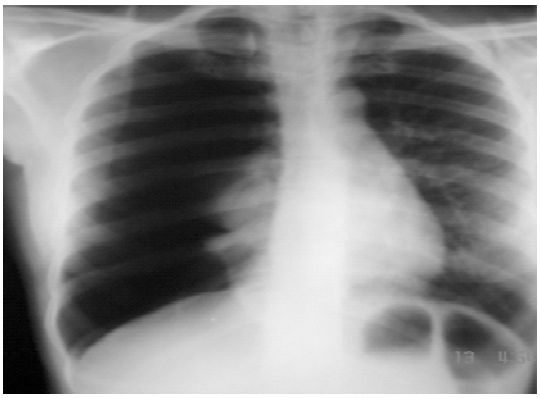
Hormonal therapy follows drug treatment for endometriosis, including testosterone derivatives, Gn-RH agonists and oral contraceptives[8]. This must also consider the patients desire for pregnancy. Hormonal treatment is associated with side effects, and recurrence of pneumothorax when discontinued, and therefore not a curative treatment method.

Our reported patient had surgical drainage of the pneumothorax and chemical pleurodesis which has apparently appeared to have cured her. She was not given hormonal therapy because she is desirous of pregnancy.

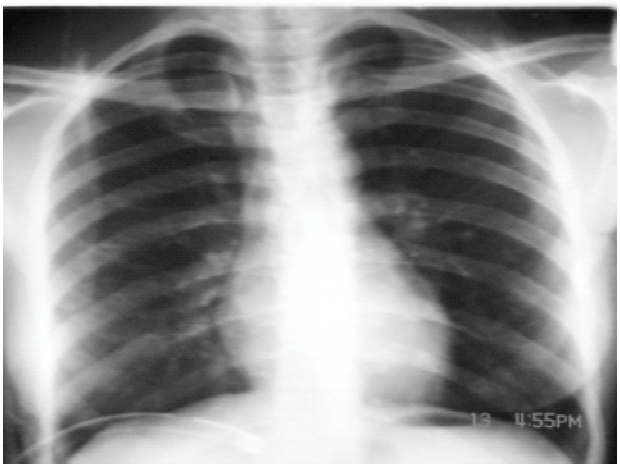
CONCLUSION

Catamenial pneumothorax should be highly suspected in any menstruating woman presenting with recurrent spontaneous pneumothorax especially on the right. And in such patient pelvic endometriosis and intra-thoracic endometriosis and diaphragmatic fenestrations should be looked for.

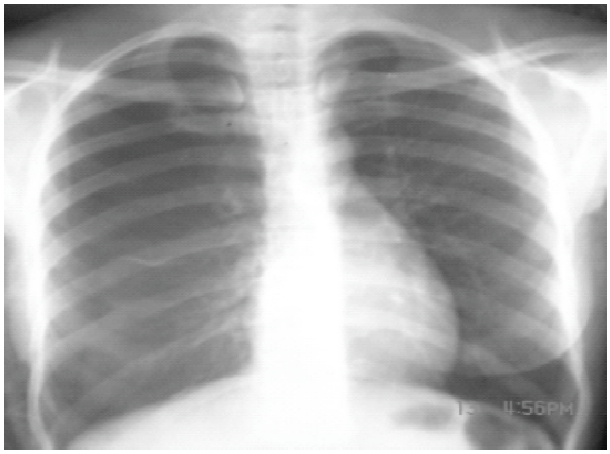
APPENDIX



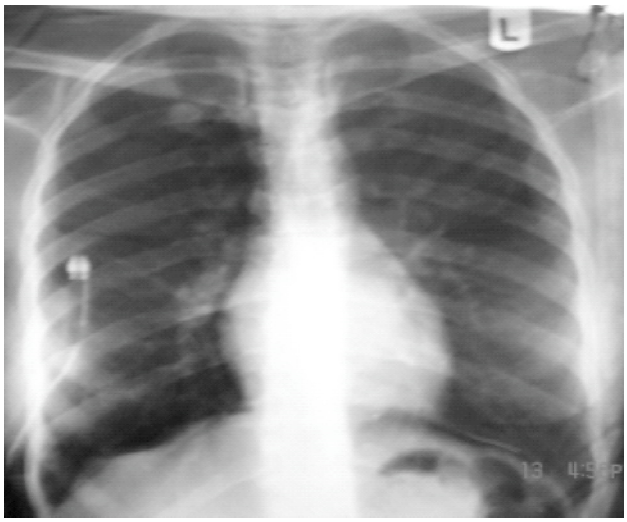
APPENDIX I: 1st presentation chest radiogram (66% Rt. pneumothorax)



APPENDIX II: Chest radiogram after initial chest tube drainage



APPENDIX III: 2nd presentation chest radiogram (30% recurrent Rt Pneumothorax



APPENDIX IV: Post – pleurodesis chestradiogram.

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