Case Report

Young woman with recurrent spontaneous pneumothorax: a case report

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Abstract
Spontaneous pneumothorax associated with menstruation is a rare entity named catamenial pneumothorax. A 36-year-old woman presented with recurrent right-sided pneumothorax requiring repeated chest tube insertions. She underwent video-assisted thoracoscopy (VATS). Histopathology was suggestive of thoracic endometriosis. When spontaneous pneumothorax occurs in ovulating women in relation to menstruation catamenial pneumothorax should be suspected. Correct diagnosis will lead to effective treatment and prevention of recurrences.

Keywords: Recurrent spontaneous pneumothorax; Catamenial pneumothorax

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Introduction
Spontaneous pneumothorax associated with menstruation is called catamenial pneumothorax. We report a case of a young woman with recurrent catamenial pneumothorax treated with video-assisted thoracoscopic surgery (VATS). Recurrent nature of the condition causes significant morbidity. Diagnosis is important because treatment is curative.

Case presentation
A 36-year-old mother of one child presented to the Teaching Hospital Anuradhapura with difficulty in breathing and right sided pleurisy for three days. Symptoms began a day before her menstruation. She denied any history of trauma or fever. She had undergone elective cesarean section eight years ago and was not on any form of contraception. She had been treated for an episode of right-sided spontaneous pneumothorax with intercostal tube drainage in a surgical ward four weeks ago.

On examination she was breathless with respiratory rate of 35 cycles per minute. Pneumothorax in right hemithorax was confirmed by chest radiography. Baseline investigations were normal including inflammatory markers. An intercostal drainage tube was inserted. Her first episode of pneumothorax had occurred on the first day of her previous menstrual cycle.

The recurrent nature and temporal relationship with menstruation suggested that she had catamenial pneumothorax. While waiting for further investigations she developed a third episode of right-sided pneumothorax a month later. These three episodes occurred during three consecutive menstrual periods.

Contrast CT scan of chest done during the third episode of the pneumothorax revealed a right-sided pneumothorax with segmental collapse and consolidation of right lower lobe. Fibreoptic bronchoscopy was normal.

Patient was referred to a cardiothoracic surgeon for VATS. During VATS her right lung was found to be attached to the chest wall at the previous intercostal tube scar with no sign of endometriosis, pleural blebs or diaphragmatic abnormalities. She underwent parietal pleurectomy and pleurodesis with uneventful recovery.

Histopathology of parietal pleural tissue revealed large areas of fresh haemorrhage, thick walled blood vessels, fibrosis and oedema with a reactive mesothelial lining. Although there were no endometrial type glands or stroma found, endometriosis was the most likely pathological diagnosis.

Patient had no recurrence of pneumothorax nine months after surgery despite regular menstruation.

Discussion
Recurrent spontaneous pneumothorax during menstruation is rare and raises suspicion of catamenial pneumothorax. Pneumothorax occurs between the day before and 72 hours after the onset of menstruation. It is rare during the inter-menstrual period. All three episodes occurred during the first day of menstruation in this patient. Risk factors for catamenial pneumothorax include personal or family history of endometriosis, subfertility and previous pelvic surgery or procedures (1, 2). This patient had previous uncomplicated caesarian section. Caesarian section is abdominal but the uterus shrinks to the pelvis a few days after surgery.

Chest and periscapular pain during the menstruation may precede catamenial pneumothorax by months or years. Pain in the chest or periscapular region, mild to moderate dyspnoea and cough are the presenting symptoms of catamenial pneumothorax. Commonly it is right sided (in this patient too) but it may be on the left or rarely bilateral (1, 2, 3, 4).

Catamenial pneumothorax can be related or not related to thoracic endometriosis. Thoracic endometriosis is present in at least half of the cases of catamenial pneumothorax. Pneumothorax is the commonest presentation of thoracic endometriosis. Diagnosed pelvic endometriosis at presentation is rare (1, 3, 5). This patient did not have any clinical features of pelvic endometriosis. Although histology did not reveal endometrial tissue it was highly suggestive.

Proposed theories for aetipathogenesis of catamenial pneumothorax include alveolar rupture due to high levels of prostaglandin F2 during menstruation, porous diaphragmatic syndrome and cyclical erosion by endometrial implants in visceral pleura, lung and diaphragm causing entry of air into pleural cavity (1, 2, 3). Chest radiograph and computed tomography may not show findings other than right-side pneumothorax. Gross abnormalities such as endometrial tissue in pleura, parenchyma or diaphragm, defective diaphragm, bullae, blebs and scarring of pleura may be detected during VATS. Bronchoscopy may reveal bleeding inside the bronchus in cases with bronchial endometrial deposits. Increased serum level of cancer antigen 125 may be found in endometriosis related cases (2, 3, 4, 5).

Preferred treatment in catamenial pneumothorax is VATS with procedures such as pleurodesis, pleurectomy and diaphragmatic repair to prevent recurrences. In addition to cure, VATS enables tissue sampling and histological confirmation of thoracic endometriosis. Open thoracotomy may be needed occasionally. Suppression of menstruation with gonadotrophin-releasing hormone analogue may be used to prevent recurrences especially immediately after surgery (2, 4). This patient refused suppression of menstruation due to personal reasons but did not have any recurrence nine months after.

In conclusion, catamenial pneumothorax should be considered when evaluating menstruating women with spontaneous pneumothorax. Thoracic or periscapular pain during menstrual period is an early symptom. Patient must undergo VATS or open surgery to find the characteristic lesions and curative treatment.

Consent
Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal.
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