International Journal of Medical Science and Clinical Invention 6(9): 4562-4564, 2019
DOI:10.18535/ijmsci/v6i9.02 **ICV 2016: 77.2**
e-ISSN:2348-991X, p-ISSN: 2454-9576

Case Report

Case report: catamenial pneumothorax

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Abstract:
Primary spontaneous pneumothorax is defined as the presence of air in the pleural space with no precipitating cause and is four times less likely to occur in women than in men[1,2]. Common causes for spontaneous pneumothorax in females include: interstitial pneumonia, primary lung cancer and lung metastasis, and asthma[3]. An uncommon cause for secondary spontaneous pneumothorax is catamenial pneumothorax, which is associated with thoracic endometriosis. The word "catamenial" is derived from the greek word "katamenios" meaning monthly recurrence.[4] Catamenial pneumothorax refers to recurrent spontaneous pneumothorax during menstruation in the absence of concomitant respiratory disease[5].

Introduction:

Catamenial pneumothorax, defined as a spontaneous pneumothorax that occurs within 72 hours before or after the onset of menstruation, is rare and accounts for only 3-6% of female pneumothorax[3,6]. It was first identified in 1972 and it affects women in their reproductive years and most commonly has a right-sided predisposition[7]. In this article, we report the unusual presentation of right-sided catamenial pneumothorax.

Case report:

A 27-year-old female, with a past history of endometriosis and an ex-smoker, presented to a secondary hospital with recurring complaints of right-sided chest pain and shortness of breath, which occurred during her menstruation over a 6-months duration.

During her first admission to the hospital, she was haemodynamically stable with an oxygen saturation of 99% in room air. A chest roentgenology was performed and it revealed a moderate sized spontaneous pneumothorax. She had an uneventful recovery, following pigtail drain insertion under local anesthetic and incentive spirometry exercises.

She was followed up in outpatient clinic where her outpatient CT scan depicted three confluent nodular densities measuring 7mm in total in the lateral basal segment of her right lower lung with otherwise normal-looking lung appearance (Image 1A). 5 months later, she developed her second unprovoked spontaneous pneumothorax, at the start of her menstrual cycle, which self-resolved.

She represented to the hospital six months later with a similar presentation and radiographic diagnosis of right spontaneous pneumothorax on CXR (Image 1B). This was managed with a chest drain insertion. An MRI scan showed small areas of enhancement over the right diaphragmatic pleura with areas of low intensity in pleural fluid in T2-weighted sequence confirming our suspicion of diaphragmatic endometriosis and associated blood products (Image 1C, D). Following discussion with a cardiothoracic surgeon at a tertiary centre, she underwent a video-assisted thoracoscopic surgery and talc pleurodesis. She recovered well and was discharged 5 days later with no complications or recurrence in the ensuing 2 years of follow-up.

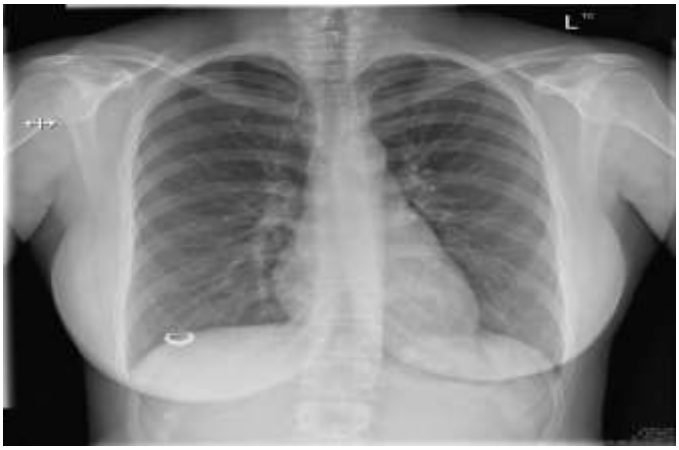


Image 1A: CXR - Right sided pneumothorax 2.68cm from the apex of lung.

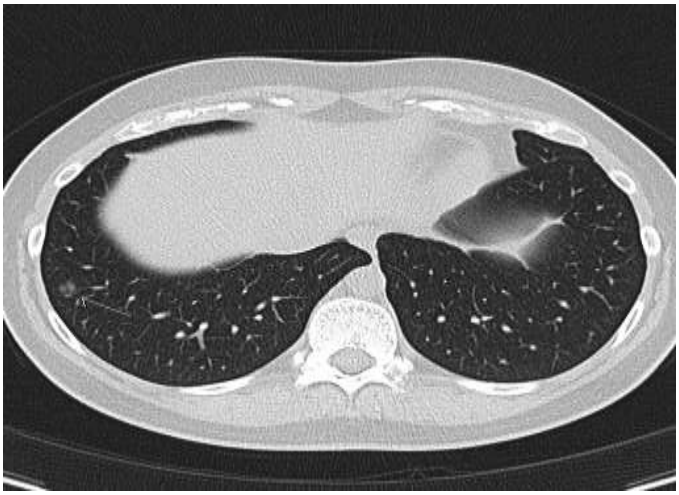
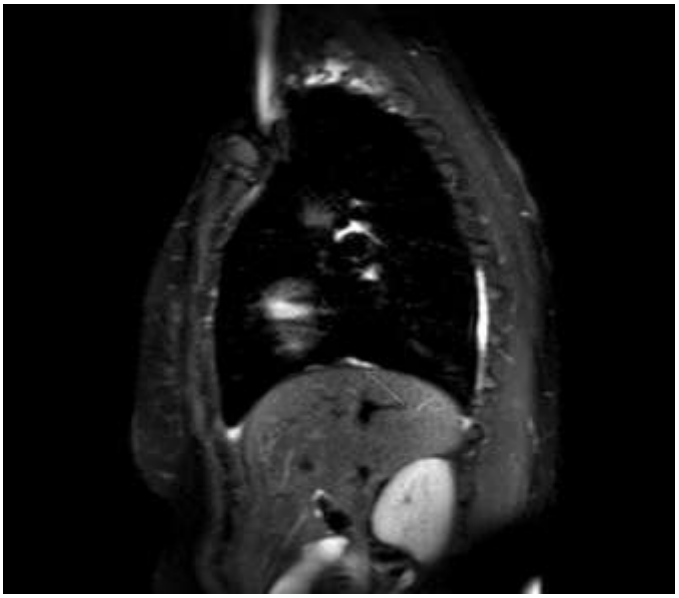


Image 1B: CT Scan - Three small confluent nodular densities visualised in the lateral basal segment of right lung lower lobe together measuring about 7mm. Normal appearance of rest of lung parenchyma otherwise. No pleural effusion, pleural mass lesion, mediastinal lymphadenopathy or mass seen.



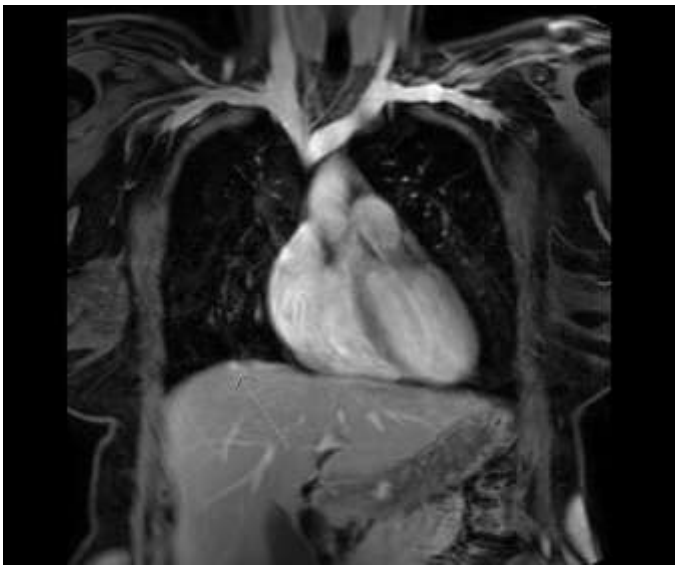


Image 1C & D: MRI Scan (C: Sagittal view, D: Coronal view) - There are small areas of enhancement seen over the right diaphragmatic pleura. Areas of low signal intensity in the pleural fluid is seen bilaterally in T2W sequence which may represent blood products or is less likely pulsation artefacts. No mediastinal mass lesion visualised.

Discussion:

Endometriosis is a complex gynecological disease, which affects 5% of the female population[8]. It is defined as ectopic endometrium tissue reacting to hormonal and inflammatory factors in relation to the menstrual cycle[9]. Thoracic endometriosis syndrome (TES) is a rare manifestation of endometriosis and refers to the presence of endometrial tissue in the thoracic cavity[10]. It comprises of 4 manifestations: catamenial pneumothorax, catamenial haemothorax, catamenial haemoptysis and lung nodules[10].

Catamenial pneumothorax accounts for 72% of TES[11]. Its pathophysiology is hypothesized to be a combination of 3 theories: Seeding of endometrial stromal tissue in the diaphragm following dissolving cervical mucus and retrograde air flow during menstruation; Transvascular dissemination of endometrial tissue into the lung with cycling sloughing causing hyperinflation; Increased prostaglandin F2 production from the endometrium causes alveolar constriction and destruction resulting in pneumothorax[12,13].

The mean age at time of diagnosis is 35±7 years old[10]. It is commonly right-sided, however, there has been previous occurrence on the left or bilateral chest[12,14,15,16,17]. The diagnosis of catamenial pneumothorax can be a clinical, radiological, surgical or histological[14,15].

Most patients present with chest pain and dyspnoea[10,12,16,18]. The authors propose eliciting the clinical quartet of chest pain, dyspnoea, menstruation onset and a past history of endometriosis. On examination, the patient may be tachpnoeic with reduced oxygen saturation or haemodynamically stable, with a decreased chest expansion and reduced breath sound on the afflicted side[14,19,20].

The radiological workup includes: CXR, CT and MRI scans[15] (Table 1). In our patient, the CXR only showed pneumothorax. Her CT scan revealed confluent nodules on the right lung base. Her MRI scan displayed endometrial deposits and blood in the setting of menstruation.

Table1:Radiological workup of catamenial pneumothorax.

Imaging	Findings
CXR	Pneumothorax ± Ground-glass haze in lung base/pleural
CT scan	Pneumothorax and ground-glass nodular lesion in subpleural location
MRI scan	Pneumothorax and Ground-glass pleural lesion with blood which appears hyperintense on T2-weighted imaging during menstruation

Histopathological findings of catamenial pneumothorax may include: endometrial glands and stroma, haemosiderin-ladden macrophages, other inflammatory cells, pulmonary blebs, or fibrosis[21,22]. Although histological confirmation provides definitive evidence of thoracic endometriosis, endometrial tissue may be absent on histology due to technical difficulties in sampling and the avoidance of diaphragmatic perforation[22,23,24].

The approach to managing of catamenial pneumothorax is multi-faceted often involving a respiratory physician, thoracic surgeon, gynaecologist, radiologist and histopathologist[3,6,10,13,14]. In the first instance of spontaneous pneumothorax in clinically stable patients with small catamenial pneumothorax (<2-3cm on CXR), patients should be observed over ≥ 6 hours period, given high flow oxygen to aid resorption of pleural air, have a repeat CXR prior to discharge, as well as an outpatient appointment in 2-4 weeks[25]. Moderate-large catamenial pneumothorax requires needle aspiration or chest tube insertion and longer duration of hospital stay[15]. Catamenial pneumothorax can also be

managed with hormonal therapy, surgical intervention or a combination of both[10].

Hormonal therapy involving gonadotrophin-releasing hormone agonist or progesterone-based oral contraceptive pills work by inhibiting intrinsic gonadotrophic releasing hormone production, blocking oestrogen production, preventing endometrial cell proliferation and its downstream effects. They have been reported to be associated with 0% and 16.7% recurrence rates respectively[10].

Surgical interventions include: resection of lung blebs or endometrial blebs, pleurodesis, pleurectomy, and plication of small diaphragmatic defects[23]. Video-assisted bullectomy and talc pleurodesis is the recommended option and is associated with a low recurrence rate of 0-8%[13,18,27]. The consideration of surgical correction of diaphragmatic defects involving plication or resection with mesh repair is dependent on the size of defect. They are associated with recurrence rates of 40% and 32% respectively[13]. In patients with suspected high risk of recurrence, hormonal suppression should be considered[24,28].

Conclusion:

Catamenial pneumothorax is a clinical diagnosis, which should not be missed, in a young female with chest pain during her menstruation. The lack of good history and physical examination findings is a reason behind the under-reporting of these cases in the literature. The management approach to catamenial pneumothorax is multidisciplinary and should be contextualized to the patient.

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