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A Case of Thoracic Endometriosis Syndrome Presenting with Recurrent Catamenial Pneumothorax

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Female, 30
Final Diagnosis: Thoracic endometriosis syndrome
Symptoms: Abdominal pain
Medication: —
Clinical Procedure: Videothoracoscopic pleurodesis
Specialty: Pulmonology

Objective: Rare co-existance of disease or pathology

Background: Catamenial pneumothorax (CP) is a spontaneous pneumothorax commonly associated with menstrual periods. Endometrial tissues most commonly involve the pelvic region. However, after the pelvis, the lungs are most frequently involved. Thoracic endometriosis should always be suspected in young women presenting with CP.

Case Report: A 30-year-old woman with history of endometriosis presented with chief complaint of umbilical pain. A computerized tomography (CT) scan of the abdomen and pelvis was performed, which showed an incidental finding of a large right-sided pneumothorax. Chest X-ray imaging showed 50% pneumothorax. A right-sided chest tube was placed, and after the procedure, a chest X-ray image showed expansion of the right lung. The patient was readmitted for elective resection of an umbilical mass and was again incidentally found to have a recurrent pneumothorax on the right side. She underwent videothoracoscopic pleurodesis with pathology, establishing the diagnosis of catamenial pneumothorax.

Conclusions: Thoracic endometriosis resulting in catamenial pneumothorax should be suspected in young women of child-bearing age. Treatment options still under debate include endoscopic resection and videothoracoscopic pleurodesis followed by gonadotrophin-releasing hormone (GnRH) therapy to reduce the rate of postoperative recurrence.

MeSH Keywords: Endometriosis • Pleurodesis • Pneumothorax

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Background

Catamenial pneumothorax (CP) is a spontaneous pneumothorax that occurs in conjunction with menstrual periods, usually within 72 h after the onset of menstruation [1]. Although it is rare, it is considered a common cause of recurrent spontaneous pneumothorax in women of child-bearing age [1]. Catamenial pneumothorax is the most common form of thoracic endometriosis syndrome, which can also present clinically as catamenial hematothorax, catamenial hemoptysis, and endometrial lung nodules [1,2]. Maurer et al. first described pneumothorax associated with menses [3]. Thoracic endometriosis should always be suspected in reproductive-age woman presenting with chest pain from spontaneous pneumothorax, and a high level of clinical suspicion has to be maintained since symptoms and signs of catamenial pneumothorax are non-specific [4]. Even though the differential diagnoses can include LAM (lymphangioleiomyomatosis), the clinical, histologic, and radiographic findings can easily distinguish these 2 entities. Several studies suggested that video-assisted thoracoscopic VATS is preferable to open thoracotomy because the former is minimally invasive, has shorter postoperative hospital stay [5,6], and is associated with less postoperative pain, leading to shorter recovery time [6,7]. The aim of our case presentation is to correlate the clinical features of pneumothorax with endometriosis syndrome, diagnosis, and treatment options.

Case Report

A 30-year-old woman with past medical history of endometriosis status after laparoscopic ovarian cystectomy in 2009 and fulguration of endometriosis in 2012 presented to the hospital for umbilical pain in September 2015. Abdominal pain was located in the lower abdomen and pain was associated with menstrual periods. The patient denied any chest pain, shortness of breath, or recent chest trauma. On physical examination, she was not in acute distress, and had stable vital signs. There was no tracheal deviation, and breath sounds were clear to auscultation on the left but absent on the right. Chest X-ray was done in concern for decrease breath sounds, which showed 50% pneumothorax (Figure 1A). Computerized tomography of the abdomen and pelvis was performed, which showed an incidental finding of a large right-sided pneumothorax (Figure 1B). A right-sided chest tube was placed by surgery and a post-procedure chest X-ray showed re-expansion of the right lung. Finally, the patient was discharged, but was readmitted for elective resection of the umbilical mass and was again incidentally found to have a recurrent pneumothorax on the right side. She underwent videothoracoscopic pleurodesis, and the diagnosis of catamenial pneumothorax was confirmed by a biopsy taken from a solitary diaphragmatic nodule, as well as by pathology depiction of endometriosis on hematoxylin and eosin (HE) staining (Figure 2). She was again admitted in March 2016 for retrosternal sharp chest pain, with CT chest again showing a right-sided mild-to-moderate

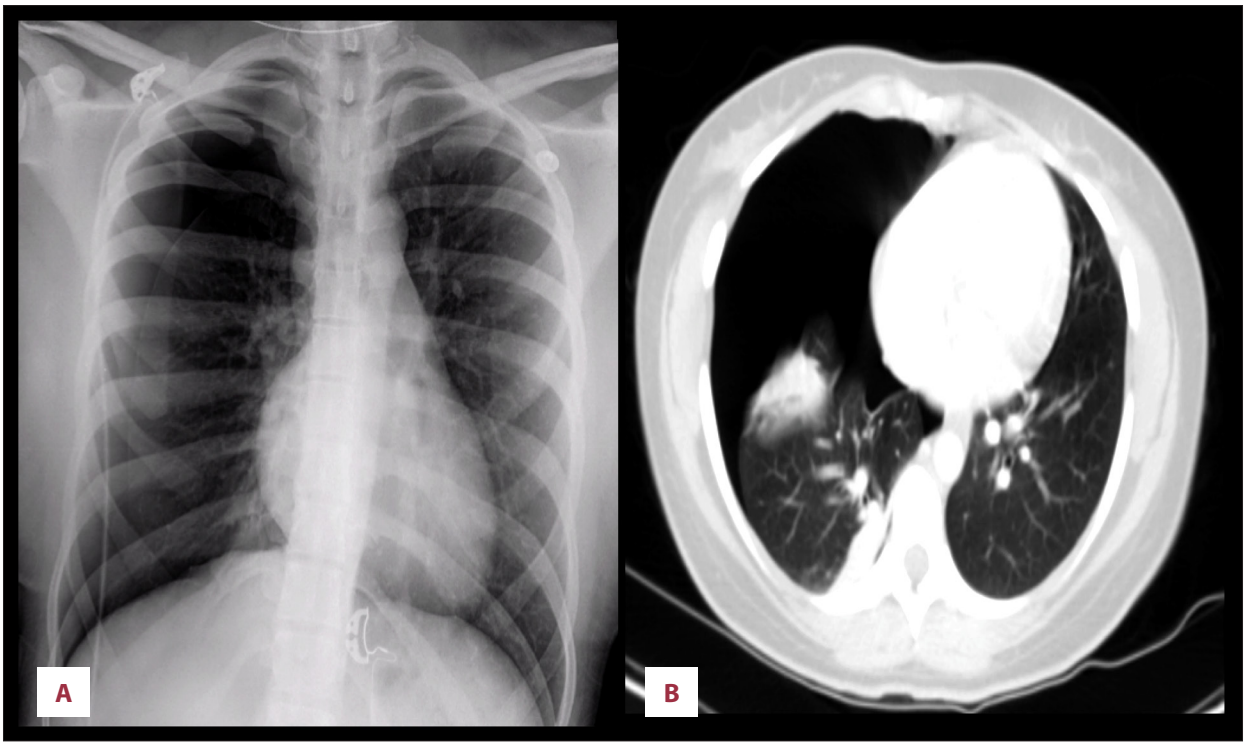

Figure 1. (**A**) Chest X-ray and (**B**) computerized tomography demonstrating 50% pneumothorax.

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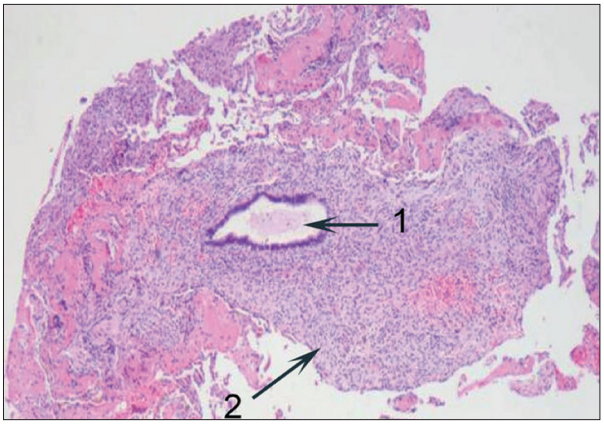

Figure 2. Depiction of endometriosis (HE) 1 and 2.

right pneumothorax. However, before any intervention could be done, she patient left the hospital against medical advice.

Discussion

Catamenial pneumothorax (CP) is a rare clinical entity of thoracic endometriosis syndrome characterized by collapsed lung, which occurs within 72 h of menstruation onset. Nearly all catamenial pneumothorax occurs on the right side, as pleural lesions are almost exclusively right-sided [8]. A complex pathogenesis involving multiple pathways for thoracic endometriosis syndrome has been described, which also varies based on individual patient presentation. The pathogenesis of catamenial pneumothorax is thought to result from the focal pleural implants migrating during leakage of air from the peritoneal cavity, which subsequently break down during menstruation or swell in response to hormone changes. In contrast, catamenial hydrothorax results from the trans-coelomic spread of pelvic uterine or extrauterine endometrial tissue elements in the pleural space [9]. Diagnosis is easily missed or delayed, causing recurrent symptoms, hospitalizations, and other life-threatening complications [10]. In a patient with thoracic endometriosis syndrome, clinical suspicion and recognition of the catamenial pneumothorax is essential for early diagnosis. Haga et al. reported that catamenial pneumothorax is distinguished from spontaneous pneumothorax by 4 clinical factors: right-sided pneumothorax, history of pelvic endometriosis, age ≥31 years, and a no smoking history. These 4 clinical presentations have high predictive value for diagnosing thoracic endometriosis syndrome-related pneumothorax over spontaneous pneumothorax [11]. Our patient met all the clinical factors mentioned by Haga et al. except for age. Diagnostic imaging is based on high-resolution computed tomography (HRCT) and, preferably, magnetic resonance imaging (MRI), since it is able to detect the blood products in the endometrial deposits [12]. In our case, based on the solitary diaphragmatic localization of endometriosis, we preferred to limit surgery to videothoracoscopic pleurodesis. The primary management of catamenial pneumothorax is treating the acute symptom, which is aspirating pleural air, and thoracotomy tube placement. Secondary management involves the prevention of recurrent episodes with surgery and/or hormonal ovarian suppression. Hormonal treatment after surgery provides successful outcome [13,14]. The hormonal suppression treatment is considered to be more effective for catamenial pneumothorax [15]. Surgery is the treatment of choice because the rate of recurrence is lower as compared to medical treatment alone [16]. Surgical options include VATS, blebectomy of visible blebs, eliminating the pleural space via chemical talc pleurodesis, or pleurectomy [17,18], and closure of diaphragmatic defect, which allows passage of endometrial tissue and air into the thorax, leading to recurrent catamenial pneumothorax. The recurrence rate of catamenial pneumothorax is higher in patients treated only with pleurodesis compared with closure of diaphragmatic defects [19]. Surgery or medical management alone lead to suboptimal response rates. The best treatment approach is surgery immediately followed by adjuvant hormonal suppression [16]. Ideally, direct visualization of the endometrial implants with thorascopic evaluation would give confirmation; however, in our case there were no macroscopic findings at surgery.

Conclusions

We described a case of recurrent catamenial pneumothorax, which is the most common presentation of thoracic endometriosis syndrome. Although a rare presentation, there are limited cases reported in the literature. Catamenial pneumothorax should be considered in a young patient with chest pain. We discussed the diagnostic and treatment options, which are still debated, but best results are achieved by videothoracoscopic pleurodesis combined with hormonal therapy.

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