

Pleural and Abdominal Endometriosis: A Rare Case Report and Review of Literature

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Abstract

Introduction: Thoracic endometriosis syndrome (TES) is the presence of endometrial tissue in the lung parenchyma or in the pleural cavity. Thoracic endometriosis syndrome consists of four distinct clinical entities: catamenial pneumothorax, catamenial hemothorax, hemoptysis, and pulmonary nodules. Thoracic endometriosis syndrome is a rare and complex condition, and diagnosis is often delayed or missed by clinicians, which can result in recurrent hospitalizations and other complications.

Current treatments include hormone therapy and, where warranted, surgical intervention. We report the case of a 33-year-old woman with endometriosis of the lung and abdomen causing catamenial hemothorax, pneumothorax and ascites.

Method: We are reporting one case of the endometriosis of the thorax which is a rare form of extrapelvic endometriosis. This patient is typically present with catamenial pneumothorax, hemothorax and ascites. Adequate clinical history coupled with HRCT help in early diagnosis and appropriate management of thoracic endometriosis.

Conclusion: TES is a rare and complex condition, resulting in delayed or missed diagnosis by clinicians. To prevent such problem and to apply appropriate treatment, a high index of suspicion is essential in any woman of reproductive age or receiving hormone replacement therapy who is experiencing cyclical chest pain, dyspnea, and/or hemoptysis. Hormone therapy is a suitable first-line treatment because it is less invasive and can preserve fertility. However, surgical intervention is available for women for whom medical therapy fails or who have a high load of disease.

Keywords: Endometriosis; Lung; Pleura; Abdomen; High Resolution Computed Tomography

Background

Although usually confined to the pelvis, endometriosis is also known to occur in the extra pelvic organs or tissues which are encountered in this case in pleural cavity and in the abdomen.

Symptomatology of extra pelvic endometriosis is always coordinated to the menstrual cycle, this is not directly apparent in all patients and the diagnosis is notoriously difficult [1].

Efforts to estimate the prevalence of extra pelvic endometriosis are hampered by great variety of symptoms, signs, location and ambiguous diagnosis.

Confirmation of thoracic and abdominal endometriosis require a combination of clinical symptoms, imaging findings and post-operative histopathological assessment [2].

Introduction

Thoracic endometriosis syndrome (TES) is the presence of endometrial tissue in the lung parenchyma or in the pleural cavity. Thoracic endometriosis syndrome consists of four distinct clinical entities: catamenial pneumothorax, catamenial hemothorax, hemoptysis, and pulmonary nodules. Thoracic endometriosis syndrome is a rare and complex condition, and diagnosis is often delayed or missed by clinicians, which can result in recurrent hospitalizations and other complications.

Case Report

In this study, we present a 33-year-old woman who presented to the emergency department with one-week history of right-sided cramping abdominal pain with non-bloody, non-bilious emesis, shortness of breath and chest pain. She reported regular menstrual cycles with no previous history of any medical illnesses. Her family history was unremarkable.

On examination, the patient was pale and was thin built. She had tachycardia with diffuse abdominal tenderness and distension. Blood tests revealed hemoglobin level of 9.5 g/dL. Peripheral smear examination revealed microcytic hypochromic anemia. The patient was referred to radiological examination.

She is referred to us for pelvi-abdominal ultrasound (PAUS) and chest x-ray. PAUS revealed a well-defined right ovarian cystic lesion, measuring 4.0 x 4.5 cm, suggestive of endometriotic cyst. There was associated ascites. On chest x-ray, there was total opacification of right hemithorax with mediastinal shift, representing massive right-sided pleural effusion (Figure 1).



Figure 1: Chest X-Ray showing total opacification of the right hemithorax denoting pleural effusion with contralateral mediastinal shift.

The differential diagnosis at that time could be endometriosis, Meigs' Syndrome.

Then, the patient underwent a diagnostic laparoscopy, right chest tube placement (Figure 2) and ascitic fluid analysis.

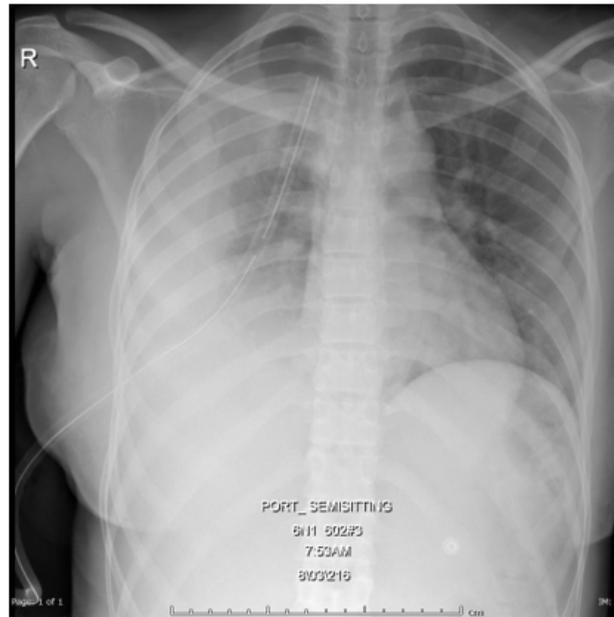


Figure 2: Chest X-Ray after insertion of right-sided chest tube, with slight improvement of the pleural effusion.

Laparoscopy revealed pelvic adhesion with hemorrhagic free fluid and chest tube drained hemorrhagic effusion. Ascitic fluid analysis revealed numerous hemosiderin laden macrophages and blood suggestive of endometriosis and negative for malignancy. The tube was removed on the third day. Her diet was slowly advanced and she was discharged in good condition six days later.

Follow up CT scan of chest after one month showed evidence of pulmonary embolism (Figure 3A) post thoracoscopy with left common iliac vein thrombosis and right-sided pneumothorax (Figure 3B). She received anticoagulant for 8 months.

One month later, the patient underwent CT chest and MRI of chest, abdomen and pelvis, that revealed mild right-sided hydropneumothorax (Figure 3C-3D) with findings consistent with endometriosis with no evidence of malignancy. MRI findings are uterine endometriosis, bilateral ovarian endometriotic cysts, endometriosis of the pelvis including uterorectal, uterovesical interfaces, peritoneum, abdominal wall (Figure 4A-4C), surgical scar with picture suggestive of pleural endometriosis as well as associated pelvi-abdominal hemorrhagic ascites (Figure 4D).

Mirena® loop was inserted to stop bleeding.

Later on, the patient was on hormonal treatment; to disrupt the normal ovarian estrogen secretion, followed by surgical excision of ectopic pleural endometrial tissue. Afterwards, she was discharged and followed-up in the outpatient clinic and remained asymptomatic.

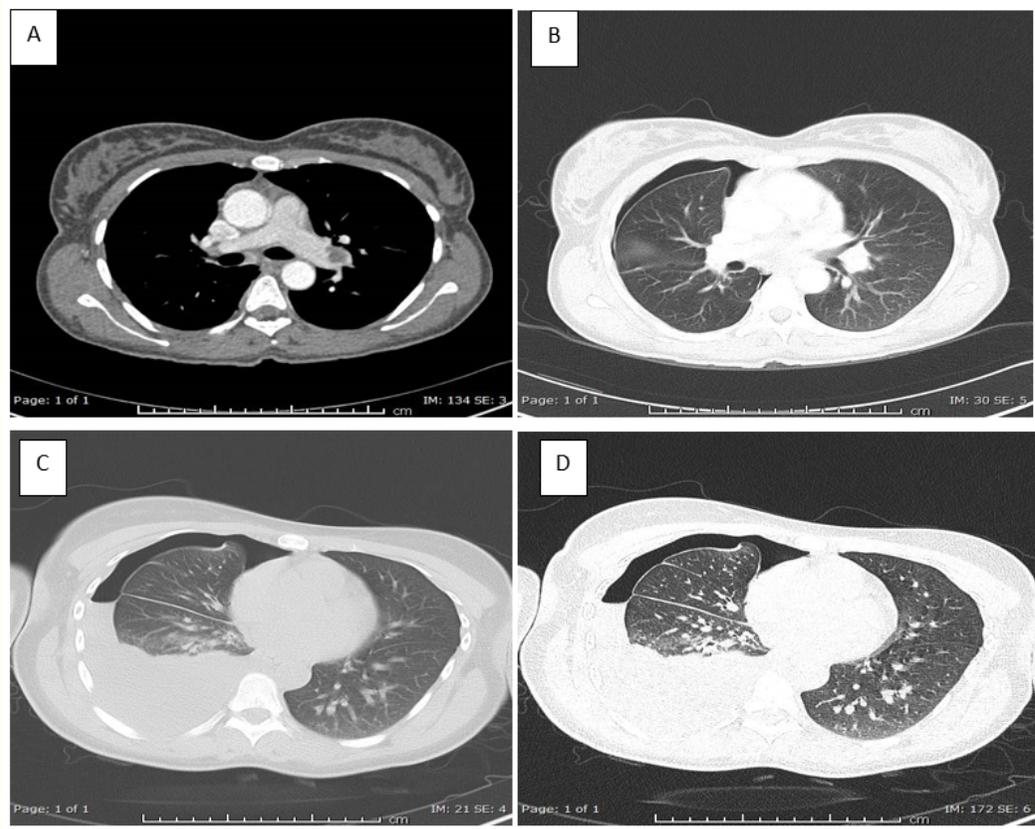


Figure 3A-3D: CT Pulmonary Angiogram. (A) Filling defect seen in the left main pulmonary artery, denoting left pulmonary Embolism. (B) Lung window showing evidence of right-sided pneumothorax. (C-D) Lung window showing the right-sided hydropneumothorax.

Discussion

Endometriosis predominantly affects women of reproductive age group and is estimated to affect approximately 5 - 10% of the female population. Pelvic pain and infertility are the most common symptoms. Pelvic pain usually occurs during or just before menstruation and lessens after menstruation. The most accepted explanation for endometriosis is metastatic implantation theory. This theory states that there is retrograde transport of endometrial tissue into the peritoneal cavity. Thorax is a rare site of endometriosis. It can present in various manifestations, the most common of them is catamenial pneumothorax (which is defined as spontaneous and recurrent pneumothorax occurring within 72 hours from the onset of menstruation). Other common manifestations include catamenial hemoptysis, catamenial hemothorax, lung nodules, isolated catamenial chest pain and catamenial pneumomediastinum [3].

Thoracic endometriosis is a rare manifestation of endometriosis. There are two forms of thoracic endometriosis; the pleural and the pulmonary form. The pleural form presents as catamenial pneumothorax, catamenial hemothorax, catamenial pneumomediastinum and

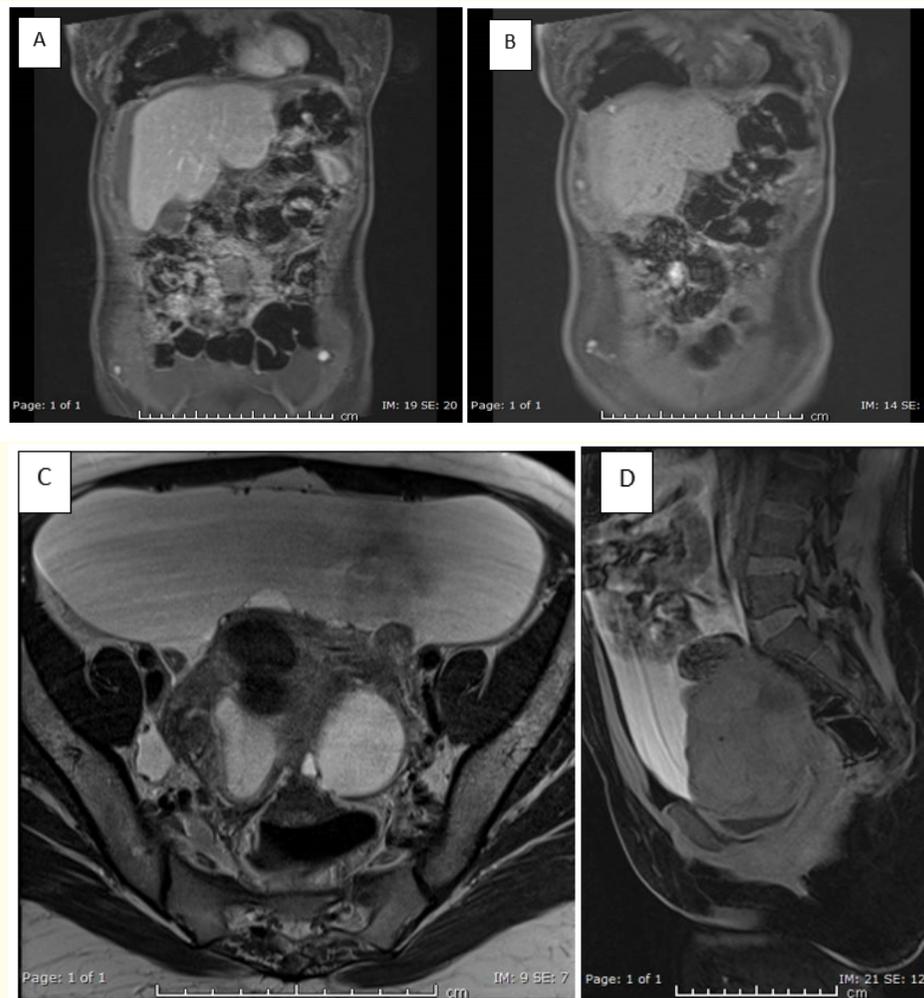


Figure 4A-4D: MRI Abdomen and Pelvis. (A-B) Coronal T1WI showing multiple foci of high signal intensity anterior to the liver and at both lower lateral abdominal wall regions. (C-D) Axial T2 and sagittal T1 WI showing kissing ovaries with bilateral ovarian endometriotic cysts and pelvi-abdominal hemorrhagic ascites.

chest pain. The pulmonary form presents as catamenial hemoptysis and pulmonary nodules. The four most common manifestations include catamenial pneumothorax, catamenial hemothorax, catamenial hemoptysis and pulmonary nodules. Catamenial pneumothorax is the most common manifestation of thoracic endometriosis (73%). Catamenial hemothorax is less common (14%), followed by catamenial hemoptysis (7%) and lung nodules (6%). The most common symptom of thoracic endometriosis is chest pain [4].

Pathogenesis of thoracic endometriosis is debated on two theories: microembolization theory and peritoneal-pleural migration. Microembolization theory states that endometrial tissue can be transported through the lymphatics or vascular channels into the lung parenchyma. Peritoneal-pleural migration theory states that endometrial tissue reaches the pleura through the peritoneum via defects

in the diaphragm [5]. Endometriosis of the lung is a clinically serious form of the disease, which requires careful differential diagnosis, as in the present case. Menstruation related hemoptysis is not obviously present in all patients, and accurate diagnosis of thoracic endometriosis is always difficult to make [6].

The diagnosis of catamenial pneumothorax (CP) should be suspected when the recurrence of pneumothorax coincides with the menstrual period. Other less common findings include hemoptysis and hemothorax [7].

HRCT is the modality of choice for localization of endometrial deposits in the lung and pleura. Although HRCT findings are nonspecific in the form of ground glass attenuation with or without nodules; in the presence of characteristic history and clinical examination findings HRCT is considered diagnostic of pulmonary endometriosis. Pleural lesions are almost exclusively right-sided, whereas lung lesions have no such predilection. The possible cause for this right sided predilection could be explained from the fact that the lymphatic drainage does not take place evenly over the whole diaphragmatic surface but is more extensive on the right side. So, the embolized endometrial tissue is more likely to be carried to the right than the left. HRCT can also act as a guide for needle aspiration of the lesion if bronchoscopy turns out negative. Pelvic ultrasound is the first and foremost investigation done in case of suspected endometriosis. In thoracic endometriosis it assumes even more importance due to the fact that pulmonary endometriosis usually coexists with pelvic endometriosis. The cause of this high association between pelvic and thoracic endometriosis is that the most likely pathogenesis of thoracic endometriosis is thought microembolization from an existing pelvic endometriosis. So, it is mandatory to look for endometriosis in the pelvis in all suspected cases of thoracic endometriosis [8].

MRI is considered superior to CT in diagnosis of endometriosis due to the presence of blood products in the endometrial deposits. Shading is a characteristic finding of pelvic endometriotic cysts and is defined as a centrally or peripherally located low-intensity area in the hyperintense cyst on T2 weighted images. On MRI thoracic lesions are hyperintense on T2-weighted spin-echo images that increases in size at the time of menstruation and shows pronounced uptake of intravenous MRI contrast agent as compared with that in the intermenstrual period [9,10].

Patients with lung endometriosis usually undergo either surgery or hormonal treatments. However, no large-scale randomized trial has been conducted and the optimal treatment regimen remains controversial [11].

There is a limited role for bronchoscopy in diagnosis because most pathologic features are located in the peripheral lung. However, Wang, *et al.* [12] found that bronchoscopy was useful if performed within 1 to 2 days of the onset of menses. Shiota, *et al.* [13] were able to make the diagnosis of pulmonary endometriosis by cytologic examination of bronchial washings, although earlier studies indicated that the diagnostic yield of bronchial brushings or washing is very low. Additionally, bronchoscopy may play a role in the localization of a bleeding lobe or segment of the lung [14].

Management of pulmonary endometriosis can be either medical or surgical. Medical therapy is the first line management. Here the aim is to disrupt the normal ovarian estrogen secretion. This is done with the help of oral contraceptives and Danazol. Standard mode of treatment is use of oral contraceptives for initial therapy and Danazol in case of recurrence [15].

Surgical lines of management include localization of the ectopic endometrial implant in the lung or pleura and their removal. The proper treatment for catamenial hemoptysis is a limited pulmonary resection with the removal of all ectopic endometrial tissue. This can be accomplished with a lung sparing segmentectomy or wedge resection. Chemical pleurodesis is effective in preventing catamenial pneumothorax and hemothorax.

Video-assisted thoracoscopic surgery allows direct visualization of the lung and diaphragmatic surfaces, and descriptions of the findings include perforations on the surface of the diaphragm, brown and violet endometrial deposits, and larger masses [16].

Conclusion

TES is a rare, complex condition and clinically serious form of the disease which require careful differential diagnosis. The diagnosis of TES is usually delayed or missed by clinicians. To keep away from such problem and to apply appropriate treatment, a high index of suspicion is essential in any woman of reproductive age or receiving hormone replacement therapy who is experiencing cyclical chest pain, dyspnea, and/or hemoptysis. Hormone therapy is a suitable first-line treatment because it is less invasive and can preserve fertility. However, surgical intervention is available for women whom medical therapy fails or who have a high load of disease.

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Statement of Ethics

The authors have no ethical conflicts to disclose. The case report was conducted ethically in accordance with the guideline of the Medical Research Center (MRC) of Hamad Medical Corporation (HMC), and after obtaining their approval. Informed consent was provided by the patient to publish the case.

Disclosure Statement

The authors declare that they have no relevant financial interests.

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Authors Contributions

Dr. Reda Yousef and Dr. Amr Elmahdy did the literature review, wrote the radiology manuscript. Dr. Ahmed Sabry, Dr. Ahmed Elgebal, Dr. Ashwini, Dr. Samah Kohla and Dr. Amal Alobaidly reviewed the manuscript.

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