


A Case of Hemothorax as Manifestation of Thoracic Endometrial Syndrome

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Abstract

Thoracic endometriosis is a rare progression of a mostly benign disease of ectopic endometrial activity involving the pleura and lung. This is a case of a young female who presented with progressive shortness of breath and was found to have significant anemia. Further investigations showed a massive right-sided pleural effusion and ascites. Subsequent thoracentesis and pelvic diagnostic laparoscopy showed a hemorrhagic pleural effusion and ascites, along with dense pelvic adhesions. Pathology was consistent with endometriosis. Patient improved on leuprolide acetate and norethindrone. This case illustrates an important consideration in the differential of a reproductive-age female with new onset shortness of breath and anemia.

Keywords

pulmonary critical care, radiology / imaging

Introduction

Endometriosis usually involves the pelvic organs and surrounding peritoneum. Thoracic endometriosis syndrome (TES) is a rare extra-pelvic involvement of the pleura or lung. It should be considered in a reproductive-age female presenting with anemia, shortness of breath, and pleural effusion. Management involves medical and surgical interventions to suppress endometrial activity.

Case Presentation

A 21-year-old female with no significant past medical history presented to the emergency room with progressive abdominal distension and bloating for 9 months. She also had right-sided upper abdominal cramps. In the last month, she also started having progressive shortness of breath on exertion. Her menstruation had been regular, but she had noticed new cramps associated with vomiting and rectal pain during her menstruation. She also experienced progressive shortness of breath with exertion in the month prior to arrival. She was afebrile, blood pressure 113/91, heart rate 106, and oxygen saturation 97% on room air. Her physical examination was consistent with decreased breath sounds and dullness to percussion on the right side of the lung, along with distended abdomen with shifting dullness. She had a hemoglobin of 5.4 g/dL with a hematocrit of 20.8%. A chest x-ray showed massive right pleural effusion with only a small amount of residual aeration of the right lung apex and a normal left lung. Computed tomography (CT) of the chest, abdomen, and pelvis demonstrated massive right pleural

effusion with a mediastinal shift along with a moderate amount of abdominal and pelvic ascites (Figure 1). She was given 2 units of packed red blood cells and underwent thoracentesis with the removal of 1250 mL of serosanguinous fluid which was consistent with an exudate as per light's criteria. The total cell count of pleural fluid was 788/mm³, and 64% were histiocytes, with plenty of red blood cells (RBCs). Her cytology showed hemorrhagic fibrinous material and reactive mesothelial hyperplasia suggestive of endometriosis. She also developed an iatrogenic pneumothorax for which she briefly had a chest tube placement. Subsequent diagnostic laparoscopy revealed dense adhesions in her pelvis, and the pathology was consistent with endometriosis. She received leuprolide acetate and norethindrone and is scheduled to follow-up as an outpatient with OB/Gyn.

Discussion

Endometriosis is defined as the presence of endometrial-like glands and stroma outside the uterus.¹ Pelvic endometriosis

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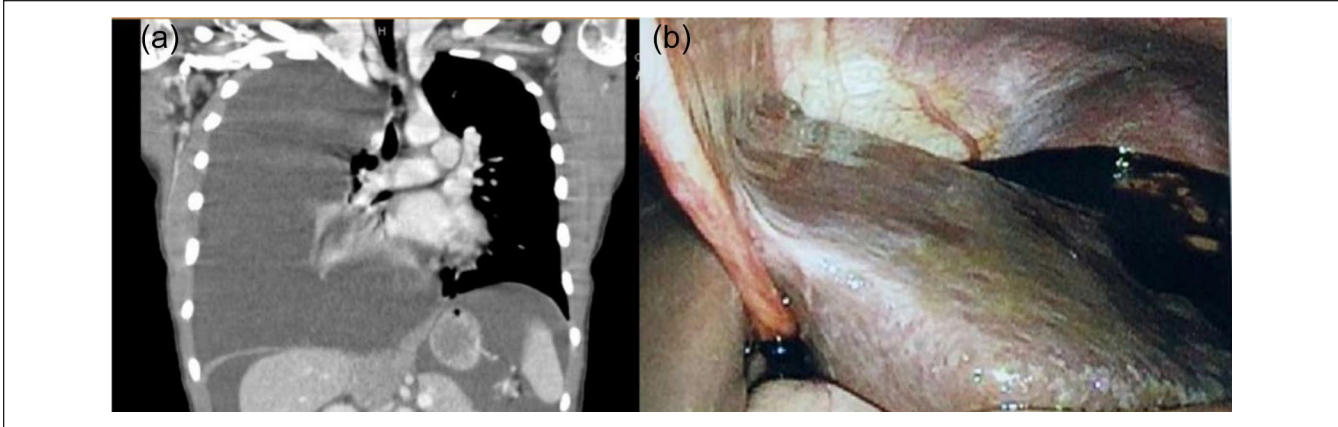


Figure 1. (a) Computed tomography of the chest showing a large right pleural effusion with leftward mediastinal shift. (b) Intra-abdominal laparoscopic view of the liver with surrounding hemorrhagic ascites.

affects 5% to 15% of reproductive-age women.² A rare progression of endometriosis is TES, and it is also the most frequent form of extra abdominopelvic endometriosis.^{1,3} TES presents around the age of 34.2 ± 6.9 years, while pelvic endometriosis symptoms precede this by 5 to 7 years.⁴ Of women diagnosed with TES, 50% to 84% have associated pelvic lesions; however, the percentage of women with pelvic disease prior to a TES diagnosis is largely unknown.⁴ In TES, functional endometrial tissue is found in the pleura, lung parenchyma, and airways, and may include clinical presentations such as catamenial pneumothorax, catamenial hemothorax, catamenial hemoptysis, and lung nodules.⁵ The pathophysiology of TES is likely multifactorial; however, the most prominent theory is retrograde menstruation which suggests that endometrial cells move in a retrograde fashion through the fallopian tubes and into the peritoneal cavity where they implant on various surfaces.¹ Patients typically develop symptoms 24 to 48 hours after the onset of menstruation, with chest pain being the most common symptom, followed by dyspnea and hemoptysis.⁵ An updated review of 110 TES cases also found that pneumothorax was the most common clinical presentation (72%), while hemothorax occurred in 12% of cases and were mostly right sided.⁶ Diagnosis is based on clinical grounds; however, imaging such as chest X-ray and chest CT scans are useful.⁵ CT scan is the first-line imaging method since it can rule out other diagnoses and map the lesions if surgery is required.³ Video laparoscopy (VL) is the gold standard for the diagnosis of diaphragmatic endometriosis, and video-assisted thoracoscopic surgery (VATS) is the gold standard for thoracic endometriosis.¹ TES involves medical and surgical management. Medical treatment involves suppression of the ectopic endometrium. Gonadotropin-releasing hormone (GnRH) agonists, such as leuprolide, may be useful perioperatively.^{5,7} Although new GnRH antagonists can also be administered, add-back treatment should be added to this therapy to reduce menopausal symptoms.^{1,8} Furthermore,

VATS allows for surgical management which can involve fulguration of lesions, sharp dissection, wedge resection, and even lobectomy.¹ In this case, the patient underwent diagnostic laparoscopy with simultaneous VATS, and was placed on elagolix and leuprolide, along with norethindrone. Currently her symptoms remain controlled with leuprolide and norethindrone, and has continued to follow with thoracic surgery with no further pleural or parenchymal lung disease.

Author Note

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Declaration of Conflicting Interests

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Ethical Approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed Consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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