Recurrent Unilateral Hemorrhagic Pleural Effusion: A Rare Manifestation of Thoracic Endometriosis Syndrome

Abstract

Unilateral recurrent pleural effusions are commonly encountered in critical care practice. Relevant clinical history, physical examination, radiology, and diagnostic thoracentesis usually identify the cause of pleural effusion in most cases. Thoracoscopy or video-assisted thoracic surgery may be required in selective cases. We report a case of 32-year-old female with recurrent unilateral hemorrhagic pleural effusion that was the presenting feature of thoracic endometriosis syndrome.

Keywords: *Catamenial hemothorax, hemorrhagic pleural effusion, thoracic endometriosis*

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Introduction

Isolated recurrent unilateral pleural effusion is a common diagnostic dilemma. It can be the presenting feature of tuberculosis (TB) in endemic regions, trauma, local (pleural or pulmonary) malignancy, or metastasis. Endometriosis though common in females of reproductive age, rarely presents as thoracic endometriosis syndrome (TES), of which catamenial hemothorax is an unusual manifestation. This multidisciplinary case highlights that (1) although catamenial hemothorax is a differential diagnosis of unilateral pleural effusion in females of reproductive age, investigations should proceed in a standard way to rule out common causes first. (2) The need for a therapeutic procedure as thoracocentesis for massive pleural effusion may outweigh the need for diagnostic investigations as a pleural biopsy for suspected TB. (3) Failure of resolution of symptoms despite treatment should prompt search along the diagnostic ladder.

Case Report

A 32-year-old married, nulliparous female, residing in an urban slum area, presented to the emergency department with complaints of progressive breathlessness, right-sided chest pain, and dizziness. Her previous medical records stated a right-sided moderate pleural effusion that was drained, with similar symptoms 8 months back. The pleural fluid was hemorrhagic with adenosine deaminase (ADA) levels of 59 IU/L (ADA > 70 IU/L: tubercular pleural effusion, 40–70: suspected tubercular pleural effusion). Mantoux test with 10 tuberculin units PPD (purified protein derivative) produced a 12-mm induration. Her sputum and pleural fluid were negative for acid-fast bacilli (AFB). She had received Category III DOTS (directly observed treatment, short course) antitubercular treatment (ATT) for 6 months, as per revised national tuberculosis control program for tubercular pleural effusion, but had to undergo multiple pleural taps (five times in last 8 months) even after commencement and completion of ATT. Currently, she had no history of fever, weight loss, cough with expectoration, hemoptysis, easy bruising, and trauma/fall. Routine laboratory investigations revealed: Hb: 7.8 mg/dL, with normal total leucocyte, platelet count, and coagulation profile. A bedside chest X-ray [Figure 1a] showed opaque right hemithorax without a mediastinal shift. She maintained a saturation of 91% on facemask with oxygen @ 6 L/min and was promptly shifted for computed tomography (CT) of the chest. CT chest [Figure 2] revealed massive pleural effusion with the underlying collapse of the right lung without any pulmonary mass/nodules. A therapeutic thoracocentesis with the insertion of 28F intercostal drain (ICD) was done [Figure 1b] and 1 L of hemorrhagic fluid was drained in the intensive care

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unit (ICU). Pleural fluid was hemorrhagic on physical examination, with 65% lymphocytes and plenty of red blood cells on smear. Pleural fluid was again negative for AFB, and malignant cells, gram stain, and pleural fluid culture did not reveal any growth. Pleural fluid protein/serum protein was 0.55, and pleural lactate dehydrogenase (LDH)/serum LDH was 0.68 signifying an exudative effusion. Pleural fluid ADA levels were 56 IU/L. She tested negative for the human immunodeficiency virus. Her echocardiography revealed good right and left ventricular function. Contrast-enhanced CT chest and abdomen additionally revealed bilateral tubo-ovarian masses with bulky uterus and cervix. As the pleural effusion resolved, the ICD was removed on day 5 with good reexpansion of the right lung and the patient was shifted to ward. Two days later, the patient again complained of right-sided chest pain, coinciding with the third day of menstruation. A repeat Chest X-ray again showed a moderate right-sided effusion [Figure 1c]. CT chest had already ruled out any lung pathology, pulmonary embolism/infarction, trauma, lymphadenopathy, or pleural nodules/thickening. With the differential diagnosis of recurrent tubercular effusion, pleural endometriosis, and malignancy, a right-sided rigid thoracoscopy and pleural biopsy were planned on the next day. Thoracoscopy revealed multiple minute cherry red/greyish spots over the parietal and diaphragmatic pleura with active ooze. Pleural biopsy of the lesions was done and histopathology confirmed the diagnosis of pleural endometriosis [Figure 3a]; immunostains for estrogen receptors were positive in the glands and surrounding spindle cells [Figure 3b]. Patient retrospectively confirmed episodic breathlessness and chest pain coinciding with menstruation. A diagnostic laparoscopy later confirmed pelvic endometriosis. She was commenced on leuprolide acetate 3.75 mg monthly intramuscular injection with no recurrence at 6 months follow-up.

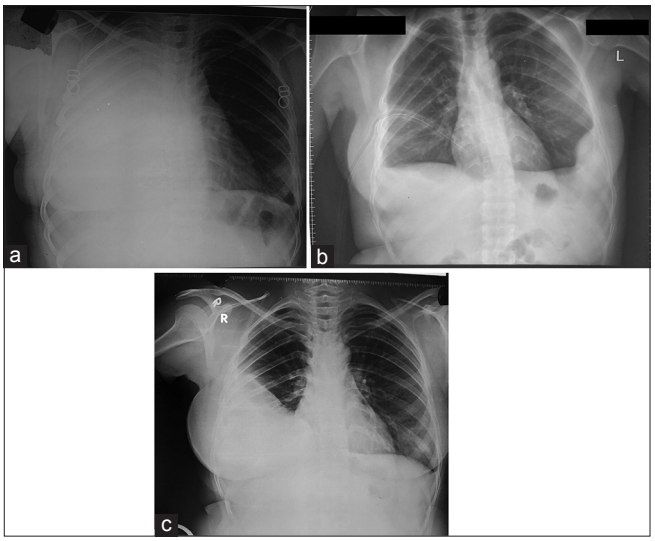

Figure 1: (a) CXR (PA) view showing opaque right hemothorax without a mediastinal shift. (b) CXR (PA) view showing resolution of right-sided pleural effusion after thoracocentesis with 28F ICD *in situ*. (c) CXR (PA) view showing recurrent right-sided moderate pleural effusion after ICD removal

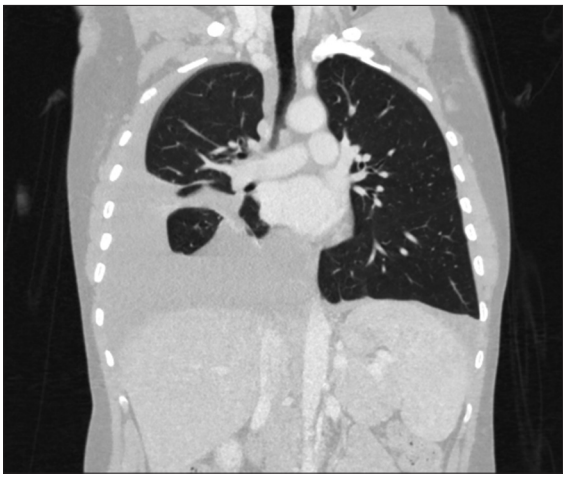

Figure 2: CT scan chest showing massive right-sided pleural effusion with the associated collapse of the right lung without any pulmonary pathology

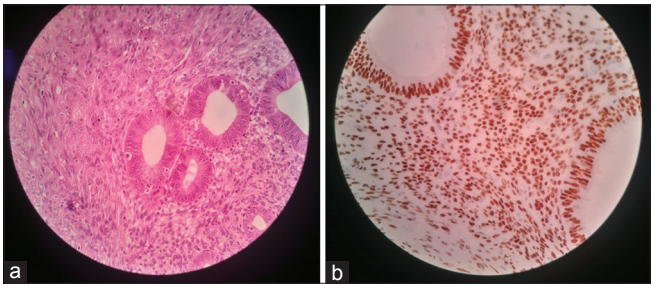

Figure 3: (a) Hematoxylin and eosin-stained section of endometriotic tissue shows endometrial glands lined by benign columnar cells with basally placed oval nuclei having bland chromatin pattern. The surrounding stroma is composed of spindle cells with elongated nuclei suggestive of endometriotic tissue. (b) Immunostains for estrogen receptors are positive in the glands and surrounding spindle cells

Discussion

Endometriosis is a common condition affecting 15% of women in the reproductive age group, wherein endometrial tissue is present outside the uterine cavity, usually in pelvis or peritoneum.[1] TES is the presence of endometrial tissue in pleura, airway, or lungs. Many theories exist for the etiology of TES, one is lymphatic or hematogenous embolization from the uterus, the second is the coelomic metaplasia theory, and the third proposes retrograde menstruation.[2,3] TES consists of four distinct clinical entities: catamenial pneumothorax (73%), catamenial hemothorax (CH 14%), catamenial hemoptysis (7%), and pulmonary nodules (6%).[4] As in this study, CH usually manifests as right-sided effusion, although both bilateral[5] and left-sided CH have been described.[6] Symptoms of CH typically include acute onset of chest pain, cough, and breathlessness, and radiology usually reveals pleural effusion that is a common presentation with pulmonary pathologies of which tuberculous pleural effusion is most common especially in endemic areas, followed by neoplastic, synpneumonic effusions and effusions secondary to

congestive cardiac failure.[7,8] CT essentially rules out other pulmonary diseases.[9] Magnetic resonance imaging though superior to chest CT in detecting TES[10] was not performed due to initial alternate diagnosis. VATS may be used as a gold standard for direct visualization of endometrial lesions in the thorax and tissue biopsy.[11] The management options for TES could be medical or surgical therapy depending on the need to balance symptomatic cure and fertility. Medical treatment is focused on the suppression of ovarian estrogen secretion, utilizing danazol and gonadotropin-releasing hormone (GnRH) agonist[12,13] as leuprolide, which was started in this study. When hormonal therapy fails, VATS or standard thoracotomy can be performed.[10] Total hysterectomy with bilateral salpingo-oophorectomy results in permanent infertility and does not address extrapelvic endometriosis.[4,6,14] Chemical pleurodesis may be required for recurrent pleural effusions.[6]

Learning points

1. This study guides caregivers to be aware of catamenial hemothorax as an unusual cause of unilateral, recurrent, hemorrhagic pleural effusion, in females of the reproductive age group that could have been suspected earlier by a meticulous gynecological history taking
2. Although VATS is the diagnostic modality of choice for TES standard diagnostic workup must always be done to exclude more probable causes first
3. Therapeutic needs (thoracocentesis) may precede diagnostic workup (pleural biopsy) for symptomatic relief
4. Lack of symptomatic relief/radiological resolution despite treatment should prompt a search for alternative etiology.

Although a diagnostic dilemma initially, a multidisciplinary approach involving critical care, gynecology, radiology, and pathology helped in putting together the pieces of a jigsaw puzzle, in this study.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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