

# A case of young woman with recurrent right pleural effusion

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## ABSTRACT

Endometriosis is usually found in women of child-bearing age. A case is presented of massive right-sided pleural effusion caused by endometriosis. The final diagnosis was made by thoracoscopic pleural biopsy. Physicians should be aware of this potentially treatable cause of pleural effusion having excluded other possibilities such as malignancy and tuberculosis.

**KEY WORDS:** Catamania hemothorax, pleural endometriosis, recurrent pleural effusion

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## INTRODUCTION

Endometriosis is a pathological condition characterized by the growth of functioning endometrial tissue outside the uterine cavity or myometrium. Endometriosis of the lung and the diaphragm is rare. Patients may present with symptoms such as shortness of breath, chest pain, and shoulder pain or they may be asymptomatic. We here by present a case of a very young woman who presented to us with right-sided recurrent hemorrhagic pleural effusion.

## CASE REPORT

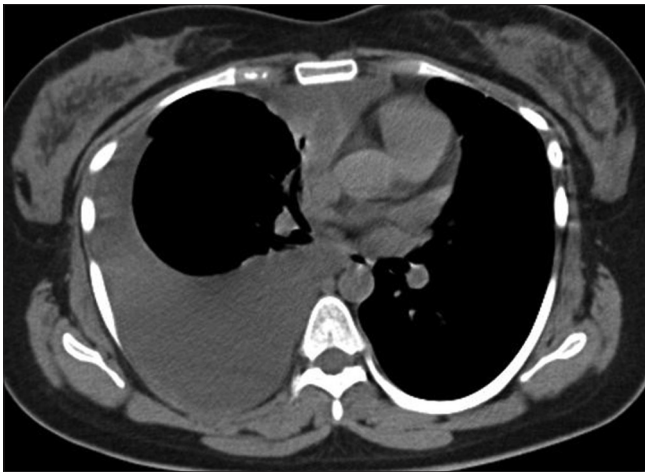
A 23-year-old woman homemaker from Southern part of India, was admitted to our hospital with history of shortness of breath, cough with mucoid expectoration and right-sided chest pain of 6-months duration. She was treated at outside hospital with oral and subsequent intravenous antibiotics with no symptomatic relief. She denied any history of hemoptysis, joint pain, rashes, fever, loss of weight or appetite. She denied any history of atopy, allergy, asthma, or pulmonary tuberculosis. Her past medical record showed that she was evaluated at outside place and was found to have right-sided pleural effusion. Pleural tapping was done which revealed fluid

to be hemorrhagic and lymphocytic exudative in nature with normal ADA. Ultrasound abdomen was showing bilateral polycystic ovarian disease, bilateral pleural effusion and mild ascites. She was sent to our center for further evaluation and management. On examination she was found to be comfortable at rest with HR: 90/min, RR: 20/min and oxygen saturation on breathing room air was 98%. Respiratory system examination findings were consistent with mild to moderate pleural effusion. No abnormality was detected in any other system. The tuberculin test (100 T.U.) was negative. Her blood investigations were following: Hb 11.5 gm/dl, total white blood cell counts 7.2 ku/ml, neutrophil 73%, eosinophil 1.8%. Her liver function test, renal function test and serum electrolytes were within normal limits. A chest radiograph showed right-sided moderate pleural effusion. 2D ECHO was normal. Contrast enhanced and HRCT of chest was ordered [Figure 1]. Medical thoracoscopy was done.

## QUESTION

Q: What is the diagnosis?

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**Figure 1:** Computed tomography picture showing right-sided moderate to massive pleural effusion



**Figure 2:** Thoracoscopic view of endometrial implants on the diaphragmatic pleura



**Figure 3:** Thoracoscopic view of endometrial implants in the costal and parietal pleural



**Figure 4:** Vascular and cheese-like areas interspersed with hemorrhagic spots on parietal pleural surface

## ANSWER

A: Pleural Endometriosis.

### Interpretation of radiological and thoracoscopic images

Chest roentgenogram shows right-sided massive pleural effusion with minimal left pleural effusion.

### Medical thoracoscopy showed

Multiple soft cyst-like nodules on diaphragmatic and visceral pleural surface along with islands of vascular and cheese-like areas interspersed with hemorrhagic spots on parietal pleural surface [Figures 2-4]. Pleural biopsy showed fibrocollagenous tissue lined by hyperplastic mesothelium and cleft like spaces along with dilated congested vessels, siderophages, hemorrhage and giant cells. Nodules of ovoid cells around vessels in a myxoid matrix (probable endometrial stromal cells) were seen beneath the pleural lines. Immunohistochemical studies for CD10 showed positivity in cytoplasm and ER and PR showed nuclear positivity in the cells within the nodules

which was suggestive of stromal endometriosis.

### Final diagnosis

Pleural endometriosis with right-sided catmanial hemothorax.

### Treatment and clinical course

She was treated with intercostal pleural drainage with good post procedure clearance. She was discharged with an advice to review with gynecologist. However, there was re-accumulation of fluid on right side which required surgical management in mode of de-cortication. Post de-cortication she responded well and there was no re-accumulation of fluid on right side. She has been on regular follow-up there after with significant relief of symptoms.

## DISCUSSION

Endometriosis is an extra-uterine growth of the functioning endometrial tissue. Thoracic endometriosis is classified as either pleural or parenchymal.<sup>[1]</sup> Thoracic endometriosis

syndrome (TES) refers to the varying clinical and/or radiological manifestations associated with the growth of endometrial glands and stroma in the pleura, lung parenchyma, airways and or diaphragm.<sup>[2]</sup> TES includes four well-recognized clinical entities, namely catamenial pneumothorax (CP), catamenial hemothorax (CHt), catamenial hemoptysis (CH) and lung nodules (LN).<sup>[2]</sup> In two-thirds of cases, women had a history of sterility or pelvic disease. The most frequent presentation of TES is catamenial pneumothorax accounting for approximately 80% of cases.<sup>[3]</sup> Catamenial hemothorax is an infrequent manifestation of TES, occurring in 14% of the cases of this syndrome.<sup>[3]</sup> The right side is involved in almost all the cases, also seen in our patient.<sup>[4]</sup> The most common presenting symptoms are nonspecific and include cough, chest pain, and dyspnea.<sup>[5]</sup> Diagnosis of thoracic endometriosis weighs heavily on clinical suspicion.<sup>[6]</sup> Chest radiograph, CT, MRI, thoracentesis, and bronchoscopy have been deemed useful in evaluating thoracic endometriosis. However, video-assisted thoracoscopic surgery (VATS) remains the gold standard for both definitive diagnosis and surgical treatment.<sup>[6,7]</sup> Despite the advances of video-assisted surgical techniques, open surgery remains an important procedure in the treatment of thoracic endometriosis. Standard thoracotomy continues to be performed in cases of previous unsuccessful surgery.<sup>[3,8]</sup> The repair of possible diaphragmatic holes and the resection of endometrial implants are easily accomplished, if it is possible to resect all involved areas.<sup>[3]</sup> Lesions close to the phrenic nerve insertion or its main division branches are best treated by limited resection (if possible) and repair.<sup>[9]</sup> Sometimes it is necessary to do laparoscopic resection of upper abdominal lesions especially if the under surface of the diaphragm is involved may be indicated. Owing to the fear of leaving occult defects, Bagan *et al.*<sup>[8]</sup> proposed the insertion of a polyglactin mesh to cover the tendinous part of the diaphragm. Proposed therapy for endometriosis also includes hormonal suppression or ovarian ablation by surgery or radiation therapy.<sup>[2]</sup> Most experts consider total hysterectomy and bilateral salpingo-oophorectomy the definitive treatment because hormonal therapy appears to provide only temporary improvement.<sup>[2,10]</sup> However, it

was not appropriate for our patient considering her age and her wish to have child.

## CONCLUSIONS

Catamenial hemothorax most commonly occurs in nulliparous women in their 3<sup>rd</sup>-4<sup>th</sup> decade of life. Pelvic and peritoneal endometriosis presents with recurrent abdominal pain associated with the onset of menses as seen in our patient. Surgical repair of possible diaphragmatic holes and the resection of endometrial implants cure the hemothorax.

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