

Thoracic endometriosis: a case report

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Radiology Case. 2012 Jan; 6(1):25-30 :: DOI: 10.3941/jrcr.v6i1.614

ABSTRACT

Thoracic endometriosis is a rare form of extrapelvic endometriosis. These patients typically present with catamenial pneumothorax or hemoptysis. Adequate clinical history coupled with HRCT helps in early diagnosis and appropriate management of thoracic endometriosis.

CASE REPORT

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A 34 yr old female patient was referred to us for ultrasound abdomen for excessive pain during menstruation. On further questioning the patient revealed a strange complaint that she used to cough up blood regularly on her days of menstruation since past 1 yr. We then called her spouse and confirmed her complaints were indeed true. She used to cough up approximately 2-3 tablespoons of blood every menstruation and had grown weak in the past 1 yr. The patient was sent back to the clinician for clinical examination with catamenial hemoptysis in mind.

On clinical examination the patient was pale and was thin build and nourished. Blood examination revealed her Hb: 9.5% (normal: 13-15%), TC-5,500 per mm³ (normal: 4,000-11,000), with predominant lymphocytes. Peripheral smear examination revealed microcytic hypochromic anaemia. The patient was then referred to radiological examination.

Grayscale Ultrasound study of the abdomen and pelvis was done. We found a well defined cystic lesion (figure-1) measuring 34 x32 mm in the right ovary. Cyst was partially filled with internal echoes and displayed fluid level. The left ovary was normal in size and echogenicity.

A provisional diagnosis of hemorrhagic cyst of right ovary was made and patient was subjected to high resolution CT study of the chest.

On HRCT of the chest, focal areas of ground glass attenuation (figure-2) were found in the posterior segment of

right upper lobe signifying pulmonary hemorrhage. Focal ground glass attenuation was found in the anterior segment of right upper lobe. A presumptive diagnosis of pulmonary endometriosis was done and the patient was subjected to bronchoscopic examination for confirmation. Bronchoscopic lavage was negative on first occasion, on having high clinical suspicion the lavage was repeated and found endometrial tissue.

A diagnosis of pulmonary endometriosis was made and patient was put up on oral contraceptives. Patient was then followed up every month and her hemoptysis did not recur in next 5 months.

DISCUSSION

Endometriosis predominantly affects women of reproductive age group and is estimated to affect approximately 5-10% of the female population (1). Pelvic pain and infertility are the most common symptoms (2). Pelvic pain usually occurs during or just before menstruation and lessens after menstruation. The most accepted explanation for endometriosis is metastatic implantation theory (3). 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 (4). Other common manifestations include catamenial haemoptysis, catamenial hemothorax, lung nodules, isolated catamenial chest pain and catamenial pneumomediastinum. Joseph and

Sahn (5) unified all these terms and renamed them thoracic endometriosis syndrome (TES).

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 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%) (5). The most common symptom of thoracic endometriosis is chest pain (6). 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 (7).

HRCT is the modality of choice for localization of endometrial deposits in the lung and pleura. Although HRCT findings are non specific in the form of ground glass attenuation with or without nodules (8); 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 (1). 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 (9). 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 (2). 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 (5). So it's mandatory to look for endometriosis in the pelvis in all suspected cases of thoracic endometriosis.

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 (10). 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 (11).

Tissue diagnosis of pulmonary endometriosis is secured with the help of bronchoscopic lavage or a CT guided aspiration or biopsy. Presence of endometrial tissue in the lavage or needle aspirate confirms the diagnosis (12).

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 (13).

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. (14).

TEACHING POINT

Thoracic endometriosis syndrome constitutes a group of disorders which includes catamenial pneumothorax, catamenial hemothorax, catamenial hemoptysis, lung nodules, isolated catamenial chest pain and catamenial pneumomediastinum. Clinical history of catamenial symptoms coupled with HRCT findings of ground glass attenuation is suggestive of pulmonary endometriosis. Pelvic ultrasound should be done in all cases as pelvic and pulmonary endometriosis coexists.

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FIGURES

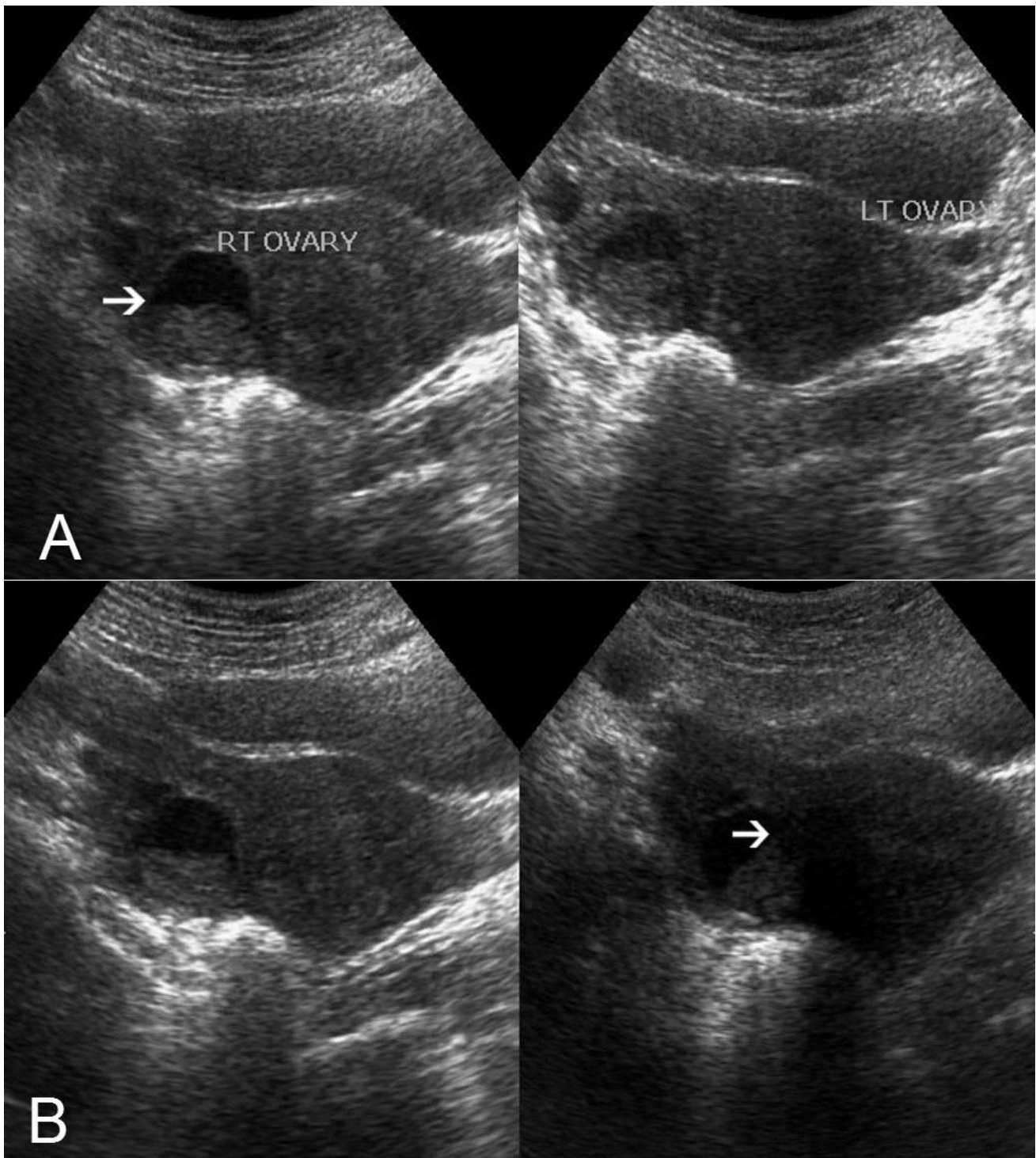


Figure 1: 34 yr old female with hemorrhagic cyst in right ovary: grayscale USG [curvilinear probe, 2-5 Mhz] axial images [a] showing cystic lesion with internal echoes & fluid level in the right ovary [arrow]. The left ovary is normal. Figure 1b: grayscale USG [curvilinear probe, 2-5 Mhz] axial images on left lateral position showing movement of the fluid level.

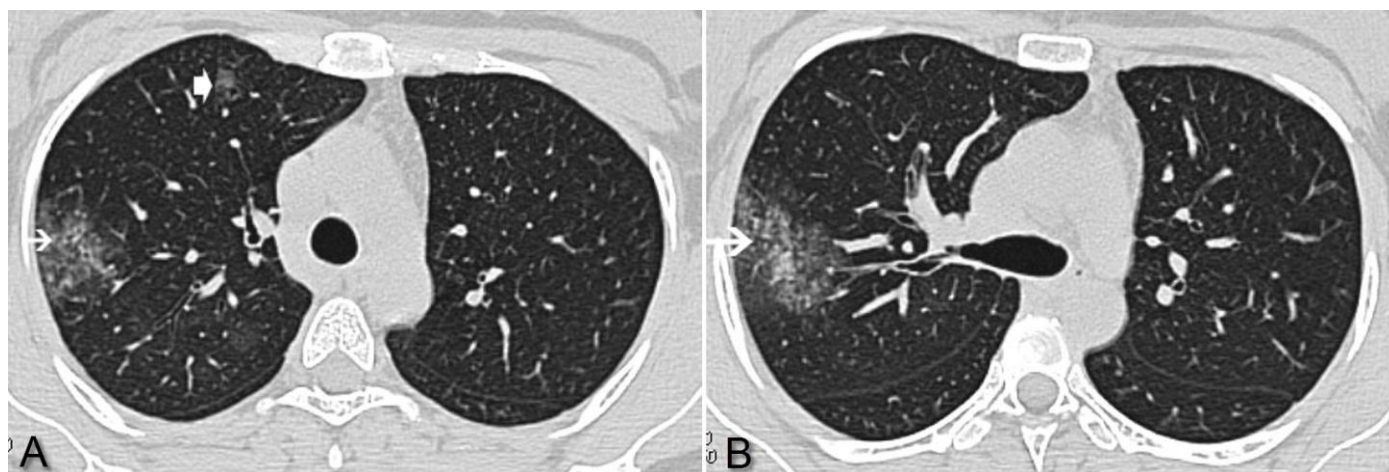


Figure 2: 34 yr old female with pulmonary endometriosis: HRCT axial images GE CT Bright Speed (120KV, 200mAs, 1.625mm x10mm interval, high resolution, non contrast) [a,b] showing ground glass attenuation in posterior segment right upper lobe [arrow], another focal ground glass attenuation is located in anterior segment of right upper lobe [thick arrow].

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Etiology	Thoracic endometriosis is a disorder of unknown etiology where there is metaplastic implantation of endometrial tissue in the thorax.
Incidence	Endometriosis is found predominantly in women of childbearing age and is estimated to affect approximately 5–10% of the female population. Thoracic endometriosis is a rare manifestation of endometriosis whose incidence is unknown. 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%].
Age predilection	Women of reproductive age group
Treatment	Medical therapy is the first line management. Standard mode of treatment is use of oral contraceptives for initial therapy and Danazol in case of recurrence. Surgical lines of management include localization of the ectopic endometrial implant in the lung or pleura and their removal.
Imaging findings	Focal ground glass attenuation with or without nodules on HRCT. Focal T2 hyperintense lesions on MRI that increases in size at the time of menstruation and shows pronounced uptake of contrast agent as compared with that in the intermenstrual period.

Table 1: Summary table of Thoracic Endometriosis

Disease	Clinical	Plain x ray	USG	CT	MRI
Thoracic endometriosis Pleural type:	Catamenial pneumothorax, hemoptysis, Chest pain,	Pneumothorax	Chest: free air detected in between parietal and visceral pleura. Pelvis: ovarian endometriosis	HRCT: pneumothorax, hemothorax	Linear T2 hypointensity [free air] seen along the chest wall.
Pulmonary type:	hemoptysis	Faint reticular/ground glass opacity		focal ground glass attenuation, ±nodules	T2 hyperintense lesion showing increase in size and enhancement during menstruation
Eosinophilic Pneumonia	Peripheral blood eosinophilia	Peripheral consolidation	Localised Consolidation seen on imaging along the chest wall	HRCT: nodular ground glass opacity in the periphery of lung	Peripheral focal areas of T1 hyperintensity corresponding to the site of consolidation, which shows enhancement on post contrast T1 images.
BOOP/COP	Shortness of breath, non-productive cough, Reduced FEV1 due to variable cause	bilateral ground-glass opacity or consolidation	Peripherally based consolidation can be seen as hypoechoic lesion with air bronchograms	HRCT: reverse halo sign (central nodular ground-glass opacity surrounded by a ring-shaped lesion with higher attenuation),	Multiple areas of enhancement seen on T1 post contrast images corresponding to areas of consolidation.

Table 2: Clinical and imaging features of thoracic endometriosis and its differentials

ABBREVIATIONS

BOOP = Bronchiolitis obliterans organizing pneumonia
 COP = Cryptogenic organizing pneumonia
 CT = Computed Tomography
 HRCT = High Resolution Computed Tomography
 TES = Thoracic Endometriosis Syndrome
 USG = Ultrasonography

KEYWORDS

TES; Thoracic Endometriosis Syndrome; Catamenial hemoptysis; Catamenial pneumothorax

ACKNOWLEDGEMENTS

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