

# Catamenial right haemothorax due to endometriosis: two case reports

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## Introduction

The presence of ectopic functional endometrial tissue may occur in 5–15% of females, especially those within the reproductive age group. Pelvic and rarely extrapelvic structures, including in the thoracic region, are the site of involvement in most reported cases.<sup>1–3</sup> Thoracic endometriosis may involve either or both the pleura and the lung parenchyma. Pleural endometriosis manifests as catamenial pneumothorax or haemothorax ('catamenial' means simultaneous with menstruation). Thoracic endometriosis, especially cases involving the pleura, typically occurs in women in their mid-30s,<sup>3,4</sup> patients usually present with chest pain and dyspnoea over the first 1–2 days of menstruation due to right-sided haemothorax or pneumothorax.<sup>2–4</sup>

Endometriosis involving the pelvic and thoracic region poses a diagnostic challenge especially in settings with few facilities and expertise for imaging and histological evaluation. Treatment options include medical therapies, ultrasound-guided chemical pleurodesis, surgery, or a combination of approaches.<sup>5–8</sup> There is a dearth of reports in the literature on extrapelvic endometriosis with thoracic involvement, especially from sub-Saharan Africa. Here, therefore, we report two cases of catamenial right-sided hydrothorax due to endometriosis in female patients of reproductive age who presented for care in a tertiary health institution in north-eastern Nigeria.

**Case 1.** A 31-year-old housewife reported a long history of cough, shortness of breath, shoulder pain, abdominal and right-sided pleuritic chest pain that was often severe during her monthly menstruation and occasionally necessitated hospitalisation. She had no other constitutional symptoms. On examination she was dyspnoeic with a respiratory rate of 18 breaths per minute, stony dull sounds on percussion, and decreased breath sounds over the entire right hemithorax consistent with pleural effusion. The remainder of the physical examination was normal. Haematological and biochemical investigations were within normal limits. Chest radiograph (Figure 1 a)

revealed complete opacification of the right hemithorax with shift of the mediastinum to the contralateral side. The left lung and the thoracic cage appeared normal. Ultrasound-guided thoracentesis drained haemorrhagic fluid and cytology revealed red blood cells and inflammatory cells. Microbiologic examination, including stains for acid-fast bacilli and routine bacterial cultures, was negative. The patient was diagnosed with a massive right haemothorax and a therapeutic thoracentesis was performed, which drained 2.3 litres of blood. Repeat chest radiograph (Figure 1 b) after the procedure revealed normal lung fields. There was repetitive catamenial accumulation of pleural fluid during four menstrual cycles, confirmed by serial chest radiographs, which were also drained accordingly. Computed tomography (CT) scans of the chest on the third day of menstruation during the second cycle after the onset of the investigation showed extensive right pleural effusion without mass lesion in the thorax (Figure 2). Abdominal ultrasound scan and CT scan of the pelvis showed a bulky irregular uterus (13 × 7 × 6 cm), with an irregular, mixed-echogenicity mass measuring about 4 × 4 × 3 cm in the posterior wall, without calcifications. The endometrial plate measured about 1 cm in thickness and bowed anteriorly due to the mass. The adnexae and the pouch of Douglas were normal. Initial assessment of uterine fibroid was made, but laparotomy revealed a frozen pelvis with extensive endometriosis and adenomyosis in the posterior wall of the uterus. Histology confirmed pelvic endometriosis. After detailed counselling, the patient was placed on danazol 400 mg twice daily, which resulted in remarkable improvement. She was discharged a month later,

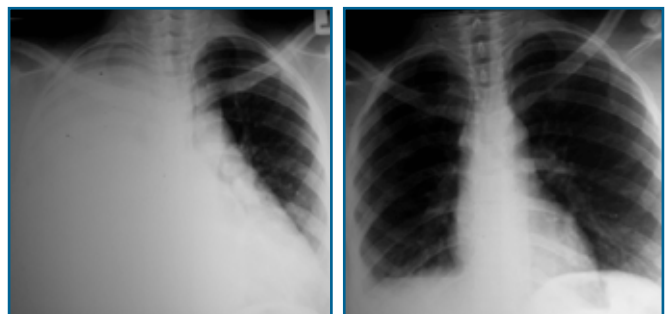


Figure 1 (a)

Figure 1 (b)

Figure 1. (a) Chest radiograph of the patient showing complete opacification of the right hemithorax with shift of the mediastinum to the contralateral side; the left lungs and thoracic cage are normal. (b) Repeat chest radiograph after thoracentesis and drainage showing normal lung fields, heart, and rib cage.

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Figure 2. Non-contrast enhanced chest CT scan at second menstrual period showing right haemothorax with marked compression of the lung.

at which time the chest radiograph revealed no pleural effusion. Subsequently, she remained asymptomatic. Hence, a retrospective clinical diagnosis of thoracic endometriosis was made.

**Case 2.** A 27-year-old woman presented with an 8-year history of recurrent lower abdominal pain and pleuritic chest pain without other constitutional symptoms. The symptoms were typically severe at the onset of menstruation and subsided afterwards. She was evaluated at a secondary health facility, where she was given anti-tuberculous medications for 9 months for possible pulmonary tuberculosis complicated by pleural effusion, without significant improvement in her symptoms. She had been married for 10 years and did not have any children.

On examination, she had dullness to percussion and reduced breath sounds over the right middle and lower lung zones. The left lung field was normal. The rest of the physical examination was unremarkable. Chest radiograph (Figure 3) showed a homogeneous opacity involving the right middle and lower zones with obliteration of the adjacent cardiac and diaphragmatic borders, tracking along the lateral chest wall, with a shift of the heart and the mediastinum to the contralateral side. Further investigation using high-resolution computed tomography (HRCT) of the lungs showed an isodense pleural fluid collection with a wedge-shaped hyperdense mass in contact with the visceral pleural surface on the right. Pelvic ultrasound showed a slightly bulky uterus with a well-defined mass of mixed echogenicity measuring 4.4 × 2.5 cm in its posterior wall with a fluid collection in the pelvic cavity. A diagnosis of catamenial right haemothorax in a woman with pelvic endometriosis was made. Under ultrasound guidance, approximately 10 ml

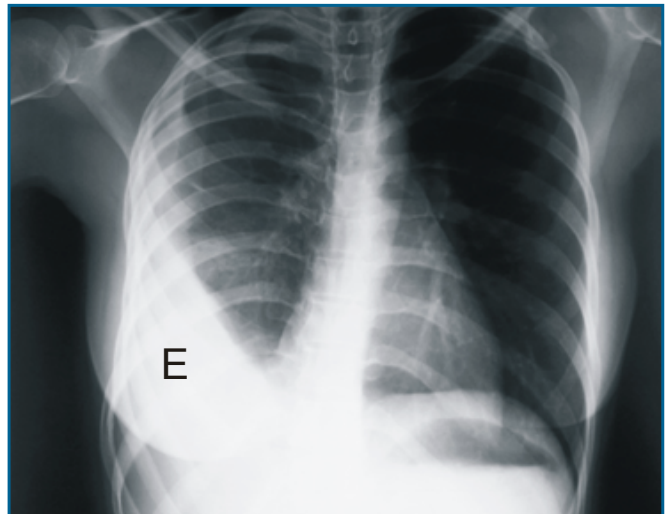


Figure 3. Posterioranterior (PA) chest radiograph showing a homogenous opacity (E) in the right lower and mid zone with obliteration of the adjacent diaphragmatic lower cardiac outline and tracking along the lateral chest wall.

of haemorrhagic, chocolate-coloured fluid was aspirated from both the pleural and pelvic cavities. Acid-fast bacilli smear and routine bacterial cultures were both negative. Cytology revealed endometrial cells without evidence of malignancy. Biopsy for histology confirmed endometriosis in both the pelvic and the pleural cavities. The patient was counselled and placed on twice-daily danazol 400 mg for 2 weeks. She had an ultrasound-guided pleural fluid drainage in which 650 ml of fluid were drained from the pleural cavity and the cavity was ablated with 99% ethanol. This was followed by another session of ultrasound-guided pelvic fluid drainage. About 1.5 litres of a similar chocolate-coloured fluid were drained from the pelvic cavity and ablation was performed with 99% ethanol as a sclerosant. The patient showed a remarkable improvement in her condition as evidenced by the disappearance of the pelvic mass and pleural fluid, and improvement in her clinical condition.

## Discussion

Endometriosis is most commonly confined to the pelvis (pelvic endometriosis), involving structures of the peritoneal cavity, ovaries, and uterosacral ligaments. However, it may rarely occur in extrapelvic locations such as the umbilicus, abdominal scars, breasts, extremities, pleural cavity, and the lungs.<sup>1-3,6</sup> Extrapelvic endometriosis poses a diagnostic and therapeutic challenge, especially in settings where facilities and expertise for imaging and histological evaluation are limited. Thoracic endometriosis may involve either the pleura or lung parenchyma or both. Pleural endometriosis manifests with chest pain and dyspnoea as catamenial pneumothorax or haemothorax. Parenchymal endometriosis, on the other hand, usually presents with chest pain, dyspnoea and haemoptysis. Women of reproductive age, especially those in their mid-30s, are most commonly affected.<sup>8-10</sup> Exacerbation of symptoms coincides with onset of menstruation,

often regresses afterwards, and typically recurs with each menstrual cycle.<sup>3,10,11</sup> Most documented cases of thoracic endometriosis present with either catamenial pneumothorax or hydropneumothorax; however, our first case presented with pelvic endometriosis and isolated haemothorax without a thoracic lesion which is rather unusual.<sup>8-10</sup> Our second patient had both pelvic and pleural endometriosis lesions. However, consistent with the majority of cases reported in the literature, right-sided lesions were observed in both patients.

Although the pathogenesis of pulmonary endometriosis is not well understood, three main theories have been hypothesised.

1. Sampson theorised that menstrual blood with endometrial fragments could regurgitate from the Fallopian tube into the peritoneal cavity. This blood could find its way into the subphrenic space and pass through the diaphragmatic fenestrations into the pleural cavity.<sup>3,8-11</sup>
2. Ivanoff theorised that irritant blood with endometrial fragments could pass through pleural fenestrations and produce metaplasia of the pleural surface, which is histologically similar to that of the peritoneum.<sup>3,8-11</sup>
3. Others theorised that obstetrical and gynaecological procedures that disrupt endometrial blood vessels and lymphatics allow lymphovascular entry of endometrial tissue causing parenchymal disease. This theory is supported by studies that find an association between pulmonary endometriosis and certain forms of endometrial trauma.<sup>3,8-11</sup>

Initial imaging evaluation involves ultrasonography scan (USS) and chest radiography. Further investigations include HRCT scan and magnetic resonance imaging (MRI).<sup>3</sup> Reported HRCT and chest radiographic findings of pulmonary endometriosis include ill-defined opacities of varying sizes, nodules, and areas of consolidation surrounding the nodules, thin-walled cavities, and bullae. These lesions may vary in size during the menstrual cycle and may disappear after the cessation of menstruation. Our patients had massive right-sided effusions, without evidence of consolidation or cavitation. In pleural endometriosis, both HRCT and chest radiographs are usually normal except during menstruation, at which time pleural effusion, pneumothorax, or hydropneumothorax may be present. In rare instances it may present with diaphragmatic surface irregularity or opacities.<sup>2-4,9,11</sup> Both pleural and parenchymal endometriosis are usually unilateral and right-sided lesions are present in 90% of reported cases, which is in keeping with findings in our patients.<sup>1-3,6</sup> Studies indicate that parenchymal endometriosis is more commonly associated with prior uterine surgery than pleural disease.<sup>1,4,6</sup>

Histologic evidence of functioning endometrial tissue confirms the diagnosis of endometriosis; however, this is not always possible due to difficulty in isolating the endometrial tissue. Instead, the diagnosis is usually made on clinical grounds with exclusion of other pulmonary diseases. Thus, a constellation of recurrent symptoms concurrent with the menses, pathologic visualisation of

pulmonary haemorrhage, compatible radiologic features, and exclusion of other diseases makes the diagnosis of endometriosis likely, as in our case series.<sup>3-6,9</sup>

Pelvic ultrasound scan is essential in all patients with thoracic endometriosis as it may coexist with pelvic endometriosis.<sup>3</sup> In our cases, uterine masses were noted that were validated histologically as endometriosis. Treatment options include medical therapy, ultrasound-guided chemical pleurodesis, surgery, or combination therapies.<sup>4-7</sup> However, the treatment strategy remains controversial due to a paucity of therapeutic trials. Medical approaches focus on the suppression of endometrial tissue by blocking the action of oestrogens, but recurrence rates are very high.<sup>10</sup> Also, virilisation, weight gain, and climacteric symptoms make hormonal therapy a less attractive option. Other treatment options include hysterectomy with salpingo-oophorectomy, chemical pleurodesis, lung-sparing segmentectomy and video-assisted thoracoscopic surgery.<sup>3,11,12</sup> In our patients, catamenial haemothorax coexisting with pelvic endometriosis responded favourably to ultrasound-guided sclerosant therapy and anti-oestrogen (danazol). However, our observation is anecdotal, being restricted to two cases. Thus larger prospective studies are needed to validate or refute our treatment modality.

## Conclusions

Endometriosis should be considered as a differential diagnosis of pleural effusion in females of reproductive age, especially when the symptoms are exacerbated by menses. Catamenial haemothorax coexisting with pelvic endometriosis responded favourably to ultrasound-guided sclerosant therapy and danazol in our patients.

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