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Case Report

An Atypical Presentation of A Case of Endometriosis

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Abstract

Bloody pleural effusion may be a clinical complication of many diseases like malignancy, trauma, pulmonary infarction, ovarian pathology, certain infections etc. Rarely pulmonary, endometriosis can present with haemothorax & pose a diagnostic problem to clinicians. Histopathological confirmation is difficult, since bleeding site is not easy to locate. However, a presumptive diagnosis of pulmonary endometriosis can be made with a typical clinical history. We report a case of pleural endometriosis in a 28 years infertile lady presented with catamenial haemothorax occurring in the first 3 days of menstruation over a 3 months period associated with right shoulder pain & progressive shortness of breath for last 6 years. She was treated with continuous low dose oral contraceptive pill & there was no evidence of recurrent haemothorax during 9 months of follow up period.

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Introduction

'Catamenial' means simultaneous with menses[1]. Catamenial haemothorax or haemopneumothorax is a very rare cause of spontaneous recurring haemothorax in a woman of reproductive age. Most endometriosis induced haemothorax involves right sided pleura with periodic symptoms like right sided shoulder pain & shortness of breath. In addition, concurrent ascites is often present[2]. We herein report a case of pleural endometriosis presenting with catamenial haemothorax with Right sided shoulder pain & progressive shortness of breath.

Case report

A 30 years old infertile lady admitted in a private hospital with severe right sided chest pain & breathlessness occurring within 3 days of menstrual cycle. Her chest pain was not associated with fever cough, haemoptysis or significant weight loss. She had no history suggestive of heart disease or trauma. She also noticed cyclical upper abdominal pain for 11 years & cyclical pain in right shoulder & progressive tightness of chest for last 6 years. Her menstrual cycle was regular with progressive heavy flow. She had no dyspareunia, her bowel & bladder habit was normal. She gave history of hospitalization for the same complains 2 months back & radio-logically diagnosed as a case of Rt-sided pleural effusion. One liter of haemorrhagic fluid was aspirated & patient was discharged with antitubercular drug empirically. As her condition was not improved, she discontinued the drug.

On examination, patient was mildly anaemic, normotensive, non-icteric and other general

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parameters were normal. Examination of respiratory system reveled – respiratory rate 20/min., chest movement & vocal fremitus diminished at the right chest. On percussion, it was dull from 3rd intercostal space to downwards. Her breath sound was diminished in the above mentioned area. There was no added sound. Examination of CVS disclosed no abnormality. Lower abdomen was soft & slightly tender. Fluid thrill was absent but shifting dullness was present, no organs were enlarged.

Pelvic examination had no remarkable findings, only fornices were slightly tender. Examination of other systems revealed no abnormality.

Laboratory studies showed, Hb-9.8 gm/dl, ESR-40 mm in 1st hour. Complete blood count, serum electrolytes, serum glucose liver function tests all were within normal limit. Chest radiograph (CXR) disclosed right sided pleural effusion. ECG was normal. Echocardiogram showed normal echo with good LV function. USG showed normal pelvic organs with mild to moderate amount of free fluid in peritoneal cavity, cul-de-sac was empty. Both pleural & peritoneal fluids were aspirated & sent for biochemical tests, cytology & AFB staining. On naked eye fluids were haemorrhagic. Microscopical examination of pleural fluid showed plenty of RBC, few neutrophils & lymphocytes, reactive mesothelial cells, no malignant cells, AFB staining was negative. Biochemical study showed sugar 90 mg/dl. & protein 0.4 gm/dl.

A 'Presumptive' diagnosis of pleural endometriosis was made & patient was discharged with continued low dose oral contraceptive pill. One month later, patient was clinically improved. No menstruation, no abdominal pain, no chest pain. CXR showed pleural effusion decreased in amount. After 3 months, USG of whole abdomen showed normal study, CXR showed normal findings. After 9 months – patient stopped OCP. Her menstrual cycle established. Till now patient is doing fine. She feels slight lower abdominal pain during menstruation but no chest pain or breathlessness.

To the best of our knowledge, bloody pleural effusion associated with endometriosis was first described by Brews in 1954[3]. Bloody pleural effusion is commonly found in situations of malignancy & tuberculosis[4]. In this case we excluded malignancy & tuberculosis by means of – negative findings in CXR & repeated cytologic studies.

Two types of thoracic endometriosis have been described. Pleural & parenchymal. Pleural endometriosis is the common of two forms. It usually causes chest pain & dyspnoea within 1-2 days of the onset of menstruation & may be associated with simultaneous pneumothorax. Over 90% cases are right sided. The majority of the patients have pelvic endometriosis. Parenchymal endometriosis usually results in catamenial haemoptysis & usually have a history of pelvic surgery or vaginal delivery. Sometimes it may be discovered as asymptomatic pulmonary nodule on chest radiograph[5-10].

There are 3 main theories of pathogenesis for thoracic endometriosis[11-14].

1. Sampson theorized that menstrual blood with endometrial fragments could regurgitate from fallopian tube into peritoneal cavity. This blood could find its way to the subphrenic space & pass through the diaphragmatic fenestrations to the pleural cavity.
2. Ivanoff theorized that irritant blood with endometrial fragments could pass through such fenestrations & produce metaplasia of pleural surface which is histologically similar to that of peritoneum.

However these 2 theories do not explain parenchymal disease.

3. Some theorize that obstetrical & gynaecological procedures that disrupt endometrial blood vessels & lymphatics may allow lymphovascular entry of endometrial tissue causing parenchymal disease. This observation is linked with common association of pulmonary endometriosis & certain forms of endometrial trauma.

The pleural space is a dynamic space with continuous movement of fluids[15]. Fluid accumulation in the pleural space is prevented by the presence of lymphatic vessels in the parietal pleura which communicates with the pleural space[16]. Fluid accumulates in the pleural space in certain disease states only when the lymphatics are affected, the hydrostatic forces change or the oncotic forces decrease[17].

Tuberculosis should always be suspected in patients who present with a chronic effusion. Patient with tubercular effusion typically present with an acute febrile illness with non productive cough (94% cases) & pleuritic chest pain (78% cases)[18].

Tubercular pleural fluid is usually straw colored, exudative (high protein > 5.0g/dl in 50-70% cases) & lymphocyte predominant with cell count of 1000-6000/mm^3. Having more than 5% mesothelial cells is rare & more than 10% eosinophils usually excludes the diagnosis of tuberculosis[19]. Radiologically, tubercular effusion are unilateral in 95% of the cases & parenchymal disease as well as pleural effusion in 50% of the proven cases[20]. Tubercular effusion should resolve within 2 months of treatment. In our case, the patient had bloody pleural effusion without fever & cough and had no improvement after antitubercular therapy.

Malignancy is a common cause of bloody effusions. Lung cancer in the most common cause (36%) & breast cancer is the 2nd most common cause (25%). Ovarian cancer accounts for 5% of malignant effusion[21]. Malignancy was excluded in our patient.

Pleural effusion can also occur in presence of other ovarian pathology. Meigs' syndrome refers to ovarian fibromas, teratomas or grauulosa cell tumors that are associated with pleural effusions. Pseudo Meigs is associated with benign ovarian cysts, uterine leiomyomas or teratomas[22], but in our patient pelvic organs were normal.

The most likely explanation for this findings in our patient is endometriosis, which appear on the pleural surface probably secondary to fluid movement through the fenestration in diaphragm or by metaplasia of pleural surface.

Treatment of pulmonary endometriosis aims at abolishing or suppressing the endometrial tissue and preventing further pelvic seeding[23]. Medical treatment, when effective, supports the diagnosis if pathologic confirmation of endometriosis cannot be obtained. Medical therapy centres on suppressing the function of the ectopic endometrium by ovarian estrogen secretion. Agents such as oral contraceptives, progestin, danazol or gonadotrophin-releasing hormone agonists can be used therapeutically. However, recurrence rates greater than 50% have been reported with hormonal therapy, due to incomplete suppression of ectopic foci and the possibility of perpetual pelvic seeding[24]. Chemical pleurodesis is effective in preventing catamenial pneumothorax and haemothorax. Pleurodesis can be achieved via tube thoracostomy and the application of talc, or with surgical procedures utilizing pleural abrasion and partial pleurectomy with or without talc. Monthly chest pain may persist even after pleurodesis[25]. Pelvic symptoms which are unresponsive to hormonal manipulation can be treated with gynecologic surgery including hysterectomy with bilateral salpingo-oophorectomy. However, there may be recurrences if estrogen replacement therapy is used postoperatively, since it may reactivate quiescent thoracic endometrial tissue implants[26].

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