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A Case of Parenchymal Pulmonary Endometriosis

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Abstract

Pulmonary Endometriosis is a rare disease entity and we report a 23-year-old single woman with a history of hemoptysis in association with menstruation. She was previously treated effectively with hormone therapy for 3 months, but decided to undergo surgical resection because of the high cost of hormone therapy. Radiographic finding of the chest showed haziness in the right lower lung field, and chest CT showed a ground-glass appearance in the posterobasal and laterobasal segment. The patient underwent basal segmentectomy of the right lower lobe. There was no incidence of hemoptysis during her menstruation following the operation.

Key Words: Pulmonary endometriosis, hemoptysis

INTRODUCTION

Endometriosis is defined as an extrauterine growth of endometrial tissue and it affects 10-to-15% of women in reproductive age.[1] It is primarily limited in the pelvis but it can also occur in the thoracic cavity as a pleural or parenchymal lesion. The diagnosis of pulmonary endometriosis is usually based on the clinical history of recurrent hemoptysis in association with the menstrual cycle and by pathologic confirmation of endometrial tissue in lung parenchyme. Hormone therapy is frequently used with success, but in the event of relapse, pulmonary resection can be performed following exact localization of the bleeding focus by fiberbronchoscopy and by radiographic method.

CASE REPORT

A 23-year-old single woman was referred for an evaluation of recurrent episodes of hemoptysis which were simultaneous with the beginning of menstruation. Expectoration of 30-to-100 cc of bright red, non-clotting blood was associated with symptoms of cough, nausea and right-side chest tightness. The symptom occurred within the initial 24 hours of each menstrual cycle and lasted for 2 to 3 days and then resolved spontaneously. The patient was asymptomatic in between her menstruation. Apart from two incidences of induced abortion at the ages of 18 and 22 years, she had no specific past medical history. Clinical examination, which included gynecologic examination and pelvic ultrasonography, showed no evidence of extragenital abdominal endometriosis. Laboratory results revealed normal findings and coagulation studies and serum gonadotropin levels were also normal. Computed tomography of the chest, performed during menstruation disclosed a fairly well demarcated area of patchy consolidation and ground-glass opacity suggestive of parenchymal hemorrhage in the posterolateral and laterobasal segments of the right lower lobe (Fig. 1). Follow up chest CT performed 2 weeks later showed a less obvious and almost undetectable lesion in the basal segment (Fig. 2). Fiberoptic bronchoscopy revealed trails of blood clot at the orifice of the laterobasal segmental bronchus of the right lower lobe (Fig. 3). Pulmonary function test showed forced vital capacity of 2.93 liters (73% of predicted value) and forced expiratory volume for one second of 2.88 liters (89% of predicted value). A gynecologist was consulted and pelvic

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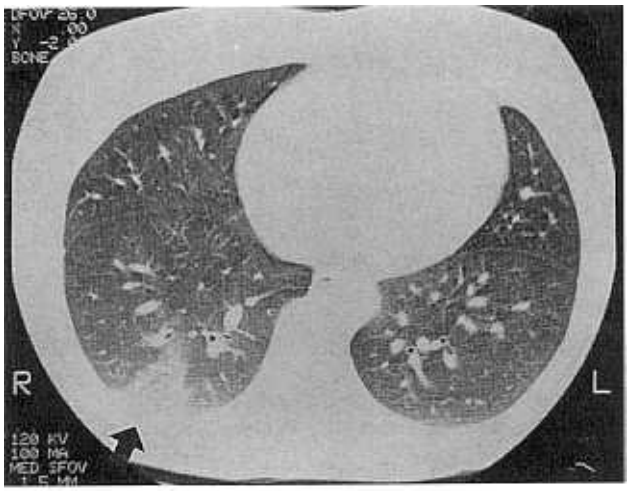

Fig. 1. Chest CT performed during menstruation shows an area of patchy consolidation and surrounding ground-glass opacity suggestive of parenchymal hemorrhage in the laterobasal portion of the right lower lobe.

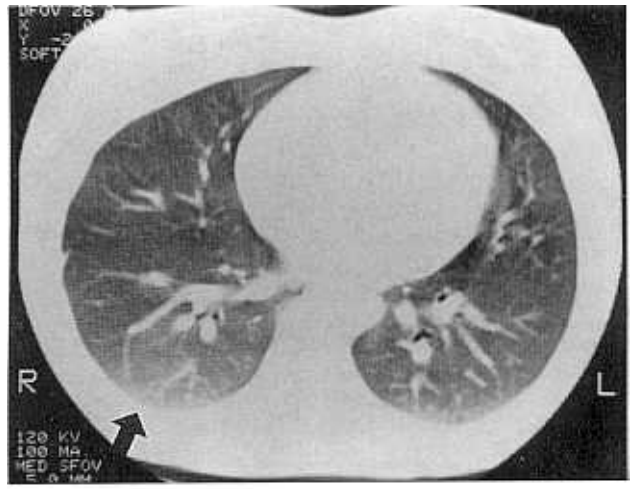

Fig. 2. The lesion in the laterobasal segment of the right lower lobe disappeared in the chest CT taken during the intermenstrual period.

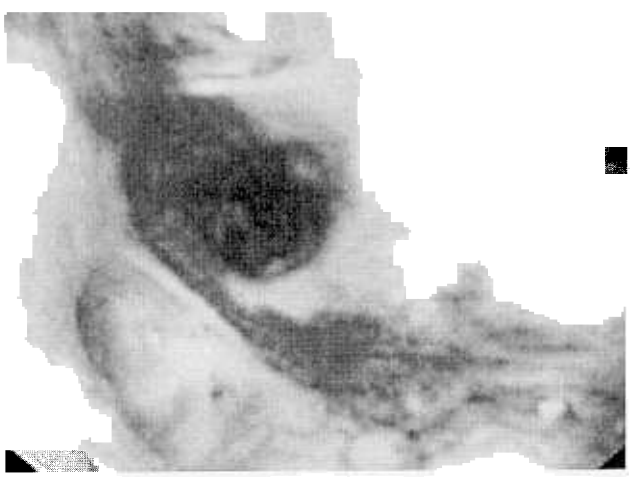

Fig. 3. Fiberoptic bronchoscopy revealed trails of blood at the orifice of the laterobasal and posterobasal segmental bronchi of the right lower lobe.

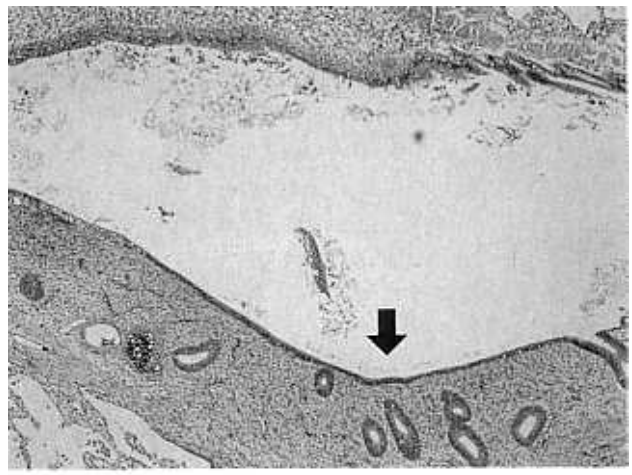

Fig. 4. Respiratory epithelium from the laterobasal segmental bronchus reveals transformation into the endometrial stroma and glands.

examination revealed no evidence of pelvic endometriosis. Assuming that the patient had pulmonary endometriosis, she was treated with Zoladex (1 ample, SQ, monthly) to induce a pseudomenopausal state by decreasing estrogen release by the suppression of pituitary gonadotropin. During three months of hormone therapy, the patient had no episodes of hemoptysis although she encountered side effects from severe hot flashes and palpitation. However, the high cost of treatment ($313.^{00}$/3.6 mg/ample of Zoladex) and the possibility of symptomatic remission after the cessation of hormonal therapy, led her to consider surgical treatment.

She underwent basal segmental resection of the right lower lobe and the gross finding of specimen showed a fan-shaped area of brownish discoloration with distinct margination, probably due to the deposition of hemosiderin in lung parenchyme. Intraoperative fiberbronchoscopic examination was performed and in contrast to the preoperative finding, the focus of bleeding was confirmed at the orifice of the com-

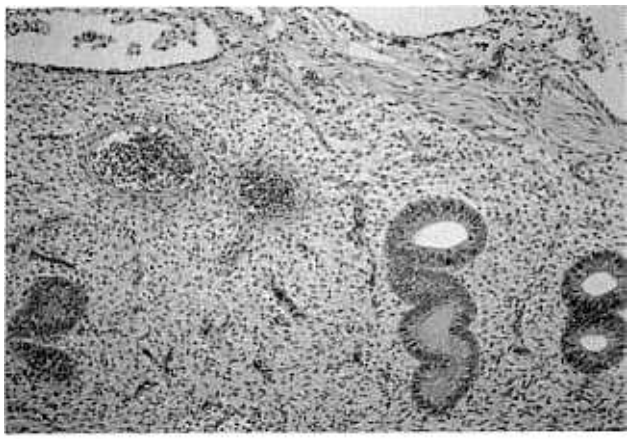

Fig. 5. The endometrial stroma and glands are seen in the proliferative phase (H&E, ×400).

mon trunk of the anterobasal, laterobasal and posterobasal segmental bronchi. A large, non-bleeding superior segment was spared and no other parenchymal or pleural lesions were noted during thoracotomy. Histopathologic examination of the resected specimen revealed a typical finding of pulmonary endometriosis with the respiratory epithelium of the laterobasal segmental bronchus replaced by the endometrial stroma and glands of the early proliferative phase. Infiltration of macrophages and hemosiderin in the alveolar space was also noted (Fig. 4 and 5). The postoperative course was uneventful with no recurrence of hemoptysis during 3 months of followup in the outpatient clinic.

DISCUSSION

A diagnosis of intrathoracic endometriosis has generally been assumed on condition that catamenial hemoptysis is concurrent with a radiologically demonstrated pulmonary lesion. Histopathologic confirmation can be obtained in less than one-third of cases due to poorly localized focus of endometrial tissue. However, diagnosis has been made on the basis of the clinical picture of symptoms associated with the menstruation cycle after excluding other causes of recurrent hemoptysis such as tuberculosis, pneumonia, bronchiectasis, tumors and Goodpasture syndrome.[1]

Two types of thoracic endometriosis, that is pleural and parenchymal disease, have been described.[2] Pleural endometriosis, which is the more common form, usually causes chest pain and dyspnea and may be associated with the pneumothorax or pleural effusion. Parenchymal disease is very rarely seen and less than 20 cases have been reported since it was first reported by O. H. Schwarz in 1938.[1,3] Parenchymal endometriosis has been manifested as either asymptomatic pulmonary nodules or as hemoptysis, pneumothorax or hemothorax during menses.[4] The incidence of parenchymal or pleural lesion seems higher in the right side and parity is not significantly different between the two diseases.[5,6] Most reported cases have occurred in the third and fourth decades, but the patients of pleural disease tend to be younger. Associated pelvic endometriosis has been rare but most patients had a history of prior gynecologic or obstetric procedures.

Different hypotheses regarding the pathogenesis of pulmonary endometriosis have been suggested and are still being debated. Pleural endometriosis has been explained by one of the following three mechanisms: Ivanoff's metaplasia theory postulates that the pleura which develop from the coelomic cavity may undergo metaplasia to form pleural endometrial tissue.[6] Sampson's tubal regurgitation theory or Charles transplantation theory propose a retrograde menstruation with transdiaphragmatic passage and subsequent implantation of the stromal and glandular elements of endometrium.[2,7] This is well supported by the surgically recognized findings of associated diaphragmatic endometriosis in the cases of pleural endometriosis.[4] Parenchymal endometriosis, by contrast, is explained by the blood-borne metastases of endometrial tissue and is supported by the histopathologic finding of endovascular endometrial epithelium.[2] Hemoptysis or pneumothorax could result from rupture of the capillaries or alveoli within the lesion due to fluid shifting during menstruation, and it is affected by an increased level of PGF2 causing bronchovascular constriction and subsequent rupture.[8]

Pulmonary endometriosis is usually suspected by typical recurrent symptoms synchronous to their menstruation cycles and confirmed by demonstrating a pathologic finding of endometrial tissue in the parenchymal tissue. The size and severity of the lesion is markedly pronounced at the time of menstruation as compared with that during the intermenstrual period.[9] Fiberoptic bronchoscopy, appropriately timed during the menstrual cycle, is the diagnostic procedure of choice for localizing the focus of bleeding,

although lung biopsy or washings often do not yield a definite tissue diagnosis.[2,10]

Medical therapy has been considered as the first line of treatment, if needed at all. Hormonal therapy with progesterone, Nafarelin, Danazol or Zoladex in order to induce a pseudopregnancy or a pseudomenopausal condition by suppressing pituitary gonadotropin has been performed.[11,12] Most patients show an excellent response while receiving medical treatment, but remission rates after the cessation of therapy vary widely and significant side effects of postmenopausal symptoms are observed in up to 85% of patients.[11] Surgical resection has been considered after the failure of medical therapy and it is a more optimal and definitive treatment measure.[6,13] Wedge resections under thoracoscopic localization of the involved lesion have recently been attempted and successful results without symptomatic remission have been reported.[2,4] Long-term followup and further investigations on the pathogenesis are required.

REFERENCES

1. Grunewald RA, Wiggins J. Pulmonary endometriosis mimicking an acute abdomen. Postgrad Med J 1988;64:865-6.
2. Cassina PC, Hauser M, Kacl G, Imthurn B, Schroder S, Weder W. Catamenial hemoptysis. Diagnosis with MRI. Chest 1997;111:1447-50.
3. Schwarz OH. Endometriosis of the lung. Am J Obstet Gynecol 1938;36:887-9.
4. Foster DC, Stern JL, Buscema J, Rock JA, Woodruff JD. Pleural and parenchymal pulmonary endometriosis. Obstet Gynecol 1981;58:552-6.
5. Elliot DL, Barker AF, Dixon LM. Catamenial hemoptysis: New methods of diagnosis and therapy. Chest 1985;87: 687-8.
6. Karpel JP, Appel D, Merav A. Pulmonary endometriosis. Lung 1985;163:151-9.
7. Sampson JA. Peritoneal endometriosis due to menstrual dissemination of endometrial tissue into the peritoneal cavity. Am J Obstet Gynecol 1927;14:422-69.
8. Balasingham S, Arulkumaran S, Nadarajah K, Jayaratnam FJ. Catamenial pneumothorax. Aust NZ J Obstet Gynecol 1986;26:88-9.
9. Volkart JR. CT findings in pulmonary endometriosis. J Comput Assist Tomogr 1995;19:156-7.
10. Wood DJ, Krishnan K, Stocks P, Morgan E, Ward MJ. Catamenial hemoptysis; a rare cause. Thorax 1993;48: 1048-9.
11. Johnson WM, Tyndal CM. Pulmonary endometriosis. Treatment with danazol. Obstet Gynecol 1987;69:506-7.
12. Lawrence HC. Pulmonary endometriosis in pregnancy. Am J Obstet Gynecol 1988;159:733-4.
13. Horsfield K. Catamenial pleural pain. Eur Respir J 1989; 2:1013-4.