

Thoracic Endometriosis: A Report of Two Cases

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Summary

We describe two patients with recurrent hemopneumothorax associated with pelvic endometriosis. The first patient is a 37-year-old nulliparous lady with recurrent bilateral hemopneumothorax. She had a past history endometriosis 10 years earlier. Laparoscopy and biopsy confirmed widespread endometriosis including in the omentum. Recurrence of the hemopneumothorax stopped after danazol therapy suggesting thoracic endometriosis as the cause of hemopneumothorax. The second lady is 47-years old with 2 children. She first presented with hemopneumothorax associated with menstrual period but ultrasound of pelvis did not reveal evidence of endometriosis. However, when she presented with a second episode of hemopneumothorax one year later, she was confirmed to have endometriosis and no further recurrence after treatment with Gonadotropin-releasing hormone analogue.

Key Words: Thoracic endometriosis, Catamenial, Hemopneumothorax

Introduction

Thoracic abnormalities associated with endometriosis were first described by Maurer et al in 1958¹. The monthly periodicity of the symptoms led to the coining of "catamenial" (in Greek-monthly). Thoracic endometriosis can involve the lung parenchyma causing catamenial haemoptysis or more often it involve the pleura causing pneumothorax. Review of 110 cases found that thoracic endometriosis most commonly presents with pneumothorax (73%) followed by hemothorax (14%), haemoptysis (7%) and lung nodules (6%) of cases². The right side is more often involved (90% of cases). Bilateral hemopneumothorax is rarely described. A review of the literature found only one case of bilateral hemopneumothorax had been reported³. We present two cases of catamenial hemopneumothorax (one bilateral) associated with pelvic endometriosis that responded to danazol therapy suggestive of pleural endometriosis.

Case 1

UK, a 37-year-old Malay lady presented in January 1998 with cough and breathlessness of 5 months duration. There was no fever, loss of weight or anorexia. She has primary infertility of 14 years. In 1988 she was found to have endometriosis and treated with Danazol but defaulted after 9 months.

Chest x-ray showed bilateral hydropneumothorax. Pleural tap showed stale bloody aspirate. The problem recurred requiring chest aspirations in February and May 1998. Pleural fluid for cytology, Mycobacterium cultures and pleural biopsy was negative. Laboratory investigations revealed normal full blood count, erythrocyte sedimentation rate, renal and liver profile, and coagulation studies. Bronchoscopy showed no growth or active bleeding.

Pelvic ultrasound revealed a bulky uterus with a left adnexal mass. Diagnostic laparoscopy followed by laparotomy was performed. Dense adhesions

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involving both adnexa, bladder and pouch of Douglas together with left hydrosalpinx, ovarian cyst, multiple fibroids and omental endometrioma were noted. Adhesiolysis, myomectomy, left cystectomy were performed. Histopathological examination confirmed leiomyoma, adenomyosis and endometriosis.

She was initially started on Danazol 200 mg bd. Since then (till last review in March 2002) there was no recurrence of the hemopneumothorax although there was a recurrence of pelvic endometriosis in February 2000 (two years later) which resolved after gonadotropin-releasing hormone (GRH) analogue injections.

Case 2

RS, a 47-year lady (Para 2, last childbirth 15 years ago), was first admitted in April 2001 with history of recurrent chest discomfort, which worsened during menses for about a year. A chest x-ray showed a right pneumothorax with pleural effusion. A chest tube drained stale blood which was negative for cytology. She improved after chest drainage. Ultrasound of the abdomen and pelvis was normal.

She was readmitted one year later (March 2002) with recurrent right hemopneumothorax. She was otherwise well. Repeat ultrasound of the abdomen revealed a small hypoechoic mass in the right adnexae suggestive of endometrioma. On diagnostic laparoscopy endometriotic spots were seen on the posterior wall of the uterus with dense adhesions obliterating the pouch of Douglas. There was bowel adhesions noted on the anterior abdominal wall. The right ovary was cystic but there was no evidence of endometriosis. The hydropneumothorax was drained again and she was treated with GRH-analogue injections. To date (August 2002), there is no recurrent of the problem.

Discussion

It is often difficult to diagnose thoracic or pulmonary endometriosis this results in a delay in diagnosis in the majority of the cases⁴. In their review, Hibbard et al recorded that only 17 of the 46 cases of thoracic endometriosis were histologically proven⁴. Although our two cases presented were not histologically confirmed, the association of recurrent catamenial (periodic association with menses) hemopneumothorax with pelvic endometriosis, which responded dramatically to danazol and GRH-analogue therapy, clinches the diagnosis. It is interesting to note that the second patient had no evidence of endometriosis during the first admission for hemopneumothorax but had significant pelvic endometriosis one year later. On the other hand, the first case presented with chest problems ten years after diagnosed pelvic endometriosis.

There are several hypotheses for the cause of pleural endometriosis. It may result either from local metaplasia of the celomic epithelium (the metaplasia theory) or from retrograde menses with a transdiaphragmatic passage and the subsequent implantation of the endometrium inside the thoracic cavity (the transplantation theory).

The management of thoracic endometriosis is directed to decrease estrogen secretion by hysterectomy with oophorectomy or hormonal therapy. At present, danazol is considered the first-line treatment for all forms of pulmonary endometriosis.

Practitioners should have a high index of suspicion for endometriosis in those with recurrent pneumothorax or hemothorax.

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