CASE REPORT

Minimally Invasive Thoracoscopic Mesh Repair of Diaphragmatic Fenestrations for Catamenial Pneumothorax Due to Likely Thoracic Endometriosis: A Case Report

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INTRODUCTION

Catamenial pneumothorax (CP) is a spontaneous pneumothorax (SP) that occurs in women within 72 hours of the onset of menstruation [1, 2]. Historically it is a rare event with an incidence of 2.8-5.6%, although recent studies suggest a higher incidence (18-33%) [1-3]. Patients present with relatively mild symptoms hence the diagnosis is often missed.

CASE REPORT

A 40-year-old female non-smoker presented with an 8 month history of recurrent peri menstrual right sided pleuritic chest pain and associated dyspnea which would resolve spontaneously several days after her monthly menses. During the most recent episode, a chest radiograph (CXR) revealed a SP suggesting a CP and an intercostal chest drain was inserted. Pleuroscopy performed by a chest physician under sedation and local anaesthesia revealed no pleural or lung abnormality. However video assisted thoracoscopic (VATS) surgery a week later during her menses, revealed multiple diaphragmatic fenestrations (Figure 1). In addition to a standard apical bullectomy and parietal pleurectomy, pleural abrasion was performed over the diaphragm and a non-absorbable PTFE mesh was carefully positioned and secured in place to cover the multiple diaphragmatic pores (Figure 2). The patient's recovery was uneventful and she was discharged home four days later with dydrogesterone 10 mg BD for suspected thoracic endometriosis. Pleural and lung histology revealed subpleural fibrosis with acute and chronic inflammation but no evidence of ectopic endometrial deposits. The patient remains symptom free at six months.

DISCUSSION

Pathophysiology

The exact pathogenesis of a CP remains elusive. Thoracic endometriosis with ectopic tissue in the parietal or visceral pleura, diaphragm, and/or lung parenchyma occurs in approximately 50% of cases [2]. Three mechanisms postulated for the passage of endometrial cells to the thorax include; coelomic metaplasia, tissue embolization and retrograde menstruation. Coelomic metaplasia, the pathological differentiation of pleural precursor cells into endometrial cells can occur due to a common embryonic origin but does not account for the right-sided predominance or parenchymal endometriosis [2]. A more plausible aetiology is embolization of endometrial tissue following uterine trauma or manipulation. Endometrial cells travel to the lung parenchyma via lymphovascular routes and to the diaphragm and pleura by transperitoneal-transdiaphragmatic migration via the right paracolic gutter [2, 4]. Finally, retrograde menstruation due to fallopian tube reflux can result in peritoneal endometrial implants that migrate to the chest via the transperitoneal-transdiaphragmatic route.

The most favoured theory as to how endometriosis causes a SP is the transdiaphragmatic passage of air. During menses, the absent cervical mucous plug allows air to enter the peritoneum via the uterus and fallopian tubes. Uterine contraction, physical effort or sexual intercourse forces air into the peritoneum which then enters the thorax via diaphragmatic pores or fenestrations. Fenestrations which are mostly right sided can be congenital or acquired. The latter is due to endometriosis of ectopic tissue. Diaphragmatic defects have been found in up to 72.5% of patients [1-5] and is a likely aetiology.

Visceral pleural endometrial implants may slough away during menses, breach the visceral pleura allowing air to move from the lung into the pleural space. This occurs in 15% of cases due to visceral pleura endometriosis [2-4]. CP may also arise from lung parenchymal endometrial implants that cause swelling of the terminal bronchioles with distal hyperinflation and rupture producing a SP. Lung parenchymal endometriosis however is rare and often bilateral.

Our case was most likely due to transdiaphragmatic passage of peritoneal air that accumulated peri menstrually, in the absence of a cervical mucous plug, via the multiple diaphragmatic fenestrations. The fenestrations were most likely due to ectopic endometriosis as the pores were very inflamed and actively bleeding despite the negative biopsy.

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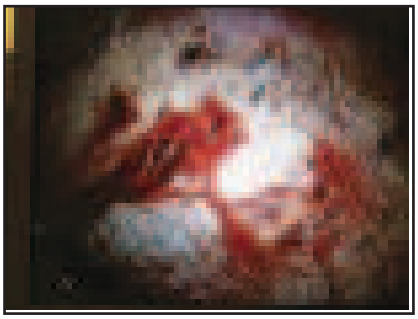

Fig. 1 : Multiple diaphragmatic fenestrations with active bleeding.

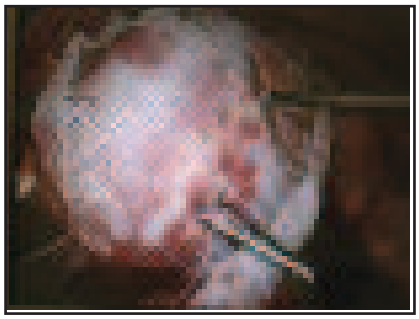

Fig. 2 : Thoracoscopic VATS placement of a non-absorbable PTFE mesh over the right hemidiaphragm.

Congenital fenestrations would not appear inflamed and usually present at an earlier age and are unrelated to the menses.

Diagnosis & Investigation

A CP should be considered for any right-sided recurrent peri-menstrual SP confirmed by CXR. VATS is the gold-standard technique to directly visualise intra-thoracic pathology and should ideally be carried out at the time of menses [1, 2]. In particular, the ipsilateral hemidiaphragm must be carefully inspected for any perforations or ectopic implants and biopsies taken. A negative biopsy however does not rule out endometriosis. In this patient, despite the clinical suspicion of possible CP, the initial medical pleuroscopy performed a week earlier revealed no obvious abnormalities. Subsequent VATS was diagnostic perhaps aided by the fact it was performed peri-menstrually.

Treatment

Definitive treatment requires a dual surgical and medical (hormonal) approach. Ectopic endometrial implants within the visceral pleura may be excised by wedge resection [1, 2]. Optimal treatment of diaphragmatic fenestrations remains contentious. Two strategies are advocated; resection of fenestrations and suturing of holes, or the placement of an artificial mesh. Resection removes sites of endometriosis, and prevents recurrence. However, it is difficult to remove all sources of endometriosis which are often widespread. Placement of an artificial mesh over the entire hemidiaphragm reinforces the diaphragmatic surface, closes perforations and induces adhesions with the lung base. This technique is associated with excellent freedom from recurrence and some suggest all patients be treated with a mesh as endometriosis and diaphragmatic defects may not be present or apparent at surgery [4,5]. The diffuse and friable widespread fenestrations we encountered precluded excision so we elected to use a mesh. Pleural abrasion of the remaining unaffected portion of the hemidiaphragm and a parietal pleurectomy was also routinely performed. A final option for patients with recalcitrant symptoms due to suspected transdiaphragmatic movement of air bubbles is fallopian tube ligation but this is best reserved for patients with no desire for a future pregnancy.

Medical treatment with GnRH agonists suppresses ovarian hormonal (FSH and LH) support to the endometrial tissue [1-3]. Medical treatment alone does not appear to be effective, with recurrent SP rates as high as 50%. Concomitant surgery and hormone therapy (usually for six months) significantly reduces this risk [2, 5].

CONCLUSION

CP must be considered in any pre-menopausal woman who reports even mild dyspnea or right sided pleuritic chest pain around the time of her menses. Diagnostic pleuroscopy or VATS evaluation is best performed at this time. The diaphragm must be carefully inspected for defects, fenestrations or ectopic implants. For widespread defects, a definitive mesh repair can be performed through a minimally invasive VATS approach and the best results are achieved with synchronous hormone suppression therapy.

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