

Bilateral Thoracic Endometriosis Affecting the Lung and Diaphragm

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ABSTRACT

Introduction: Endometriosis of the lung and the diaphragm is rare. Patients may present with symptoms such as shortness of breath, chest pain, and shoulder pain or they may be asymptomatic. Of note, there have been few reports of bilateral catamenial disease, and no reports, to our knowledge, of bilateral pathology proven pulmonary parenchymal endometriosis.

Case: A 43-year-old with stage IV endometriosis and large leiomyoma underwent a laparoscopic hysterectomy and treatment of endometrial lesions in 2005. In March and April of 2011, she presented with bilateral pneumothoraces. She subsequently underwent video-assisted thoracoscopy as well as resection and fulguration of bilateral lung and diaphragmatic endometriosis. Pathology confirmed endometrial implants in the lung parenchyma bilaterally.

Conclusion: Catamenial pneumothorax is the most common presentation of thoracic endometriosis. However, bilateral catamenial pneumothoraces are rare. To the best of our knowledge, this case reflects the first report of pathology proven bilateral lung and diaphragm endometriosis.

Key Words: Endometriosis, Catamenial pneumothorax, Leiomyoma.

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INTRODUCTION

Endometriosis of the lung parenchyma was first described by Schwarz in 1938.¹ Spontaneous pneumothorax associated with menstrual cycles (catamenial pneumothorax) was described as early as 1958.^{2,3} To our knowledge, the current case represents the first report of pathology proven bilateral pulmonary parenchymal endometriosis.

CASE REPORT

A 43-year-old Asian American female, with a history of stage IV endometriosis and large leiomyoma, was treated by total laparoscopic hysterectomy and treatment of endometriotic lesions in 2005.⁴ Subsequently, in March and April of 2011, she developed bilateral spontaneous pneumothoraces. Both instances were preceded by typical symptoms she associated with her pre-hysterectomy menstrual cycles.

The patient underwent a combined bilateral video-assisted thoracoscopic surgery (VATS) and video-assisted laparoscopic surgery (VALS) for treatment of endometriosis. We have previously described our standard approach to thoracic endometriosis.⁵ Thoracoscopy revealed multiple hemorrhagic areas on the right lung with adhesions to the chest wall, which were resected. Other superficial areas of endometriosis affecting the right chest wall and right hemi-diaphragm were treated using plasma energy.⁶ Mechanical pleurodesis was performed on the right pleura. Examination of the left lung revealed an apical bleb consistent with possible endometriosis that was resected. Endometriosis of the left hemi-diaphragm appeared extensive and was excised with the assist of an Endo-GIA stapler. Chest tubes were inserted bilaterally. Upon completion of the thoracoscopy, a video-assisted laparoscopic bilateral salpingo-oophorectomy with treatment of endometriosis within the pelvis was performed. Further examination of the abdominal portion of the diaphragm revealed no evidence of endometriosis.

On postoperative day 2, acute onset chest pain prompted a computed topography scan, which revealed a defect in the left hemi-diaphragm. The patient was taken urgently to the operating room for thoracotomy and repair of a diaphragmatic defect. The patient

did well thereafter and was discharged home on hospital day 6 without any further complications.

Histologic examination of lesions from both lungs demonstrated well-circumscribed intra-parenchymal foci of proliferative-phase endometrial glands set in a fibrotic stroma. An admixture of blood and hemosiderin-laden macrophages was present within the lesional glands and the surrounding alveolar spaces. In total, 3 of the 6 biopsied foci of grossly similar lesions demonstrated definite endometriosis, while the remaining samples showed only hemorrhage and fibrosis despite exhaustive sectioning, compatible with previous reports (only ~1/3 of foci are histologically proven).⁷

The patient gave consent to publish these features of her case.

DISCUSSION

Thoracic endometriosis is a rare manifestation of extra-genital endometriosis.¹ Up to 60% of cases will be associated with pelvic endometriosis.^{1,8,9} Lesions on the diaphragm and visceral pleura are the most commonly described sites (38.8% and 29.6%, respectively).⁸ Lesions of the parenchyma are uncommonly encountered.^{1,9} Bilateral thoracic endometriosis is rare.

The exact pathophysiology of catamenial pneumothorax remains unclear. Three main theories have been proposed.¹⁰ The theory of retrograde menstruation suggests endometrial cells enter the peritoneal cavity and then enter the pleural space through lymphatic channels, diaphragmatic fenestrations or hematogenously as with metastatic disease. This theory was first described by Schron and Ruysh over 237 years ago.¹¹ The hormonal model implicates high prostaglandin F₂ at ovulation, which may result in vasospasm and associated ischemia in the lungs. This, in turn and in combination with prostaglandin-induced bronchospasm, may result in alveolar rupture and subsequent pneumothorax.¹² Finally, the anatomic model suggests that the loss of the cervical mucus plug during menses results in communication between the environment, peritoneal cavity, and subsequently the pleural space.¹³

Diagnosis of thoracic endometriosis weighs heavily on clinical suspicion.⁹ Most patients will present with symptoms consistent with catamenial pneumothorax: shortness of breath, cough, and pleurisy. Chest radiograph, CT, MRI, thoracentesis, and bronchoscopy have been deemed useful in evaluating thoracic endometriosis. However, video-assisted thoracoscopic surgery

(VATS) remains the gold standard for both definitive diagnosis and surgical treatment.¹³

Medical treatment is the first step in the management of symptoms; however, it can be expensive and recurrence is high with discontinuation. By contrast, chemical pleurodesis, pleurectomy, and segmental resection have all proven successful in the resolution of symptoms.¹⁴ The introduction of video-assisted endoscopic surgery has revolutionized modern day surgery.¹⁵ In the same vein, the use of VATS allows for direct visualization of implants and nodules throughout the thoracic cavity, and the ability to resect apical blebs, parenchymal, and diaphragmatic implants. Superficial implants can be treated using bipolar, CO₂ laser or plasma energy. Exploratory thoracotomy, previously used regularly for diagnosis and treatment, now is reserved for cases in which minimally invasive techniques fail.

In this case, we used a multidisciplinary approach of VATS and VALS, thus optimally addressing pelvic, thoracic cavity, and subdiaphragmatic regions in a single operation.⁵ The minimally invasive, combined approach to thoracic endometriosis is gaining momentum as the mainstream surgical option for patients.

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