A Rare Case of Bilateral Catamenial Pneumothoraces

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Introduction

Catamenial pneumothorax (CP) is an entity of thoracic endometriosis syndrome (TES), a rare form of active extrapelvic endometriosis in or around of the lungs. CP accounts for 73% of TES and responsible for 2-5% of spontaneous pneumothorax of women in 30-40s. In patients presenting with right-sided pneumothorax within 72-96 hours of the onset of menstruation, CP should be strongly considered. Here, we present a rare case of bilateral CP.

Case Presentation

A 41 year-old female with complaints of intermittent periumbilical pain for more than a decade–ascribed to gas pain or reflux–was referred for evaluation of primary peritoneal carcinomatosis. In her 20's, she developed bloody discharge from the umbilicus and underwent a procedure to prevent further episodes. Her pain worsened and became more generally distributed over the abdomen in the few years prior to her current presentation. Worsening symptoms prompted her gynecologist to order an abdomino-pelvic MRI, which showed a moderate amount of ascites. Diagnostic paracentesis revealed serosanguineous fluid with a predominance of RBCs and lymphocytes. Cytology was significant for carcinoma likely of a gynecologic source, and CA-125 was 235U/mL. Family's cancer history was noncontributory.

A staging CT scan showed bilateral pneumothoraces (R40%-L20%) with small bilateral effusions. She was immediately admitted for further management. She reported chronic chest pain and dyspnea, but no new significant changes compared to the preceding week. The first day of her last menstrual period was a few days prior to the CT scan. Her vital signs were stable, and exam was notable for bilaterally decreased breath sounds. A right chest tube was placed, but due to persistent pneumothorax on the right chest, a VATS inspection, pleural biopsies and pleurodesis were performed.

During the VATS inspection, multiple adhesions were noted within the pleural space, as well as dense white fibrotic plaques along the posterior aspect of the thorax. A bluish discoloration was noted on the infra-diaphragmatic portion of the diaphragm. Pathology revealed fibrous plaques with mild chronic inflammation and reactive mesothelial cells. Fluid analysis from the both sides of the chest was also negative for malignancy. She was discharged three days post-VATS and underwent TAH-BSO one month later. Pathology confirmed extensive endometriosis with peritoneal implants–the initial pathology report was subsequently amended.

Discussion

It is often challenging to make a definitive diagnosis of catamenial pneumothorax, because symptoms may not recur with every menstruation and often nebulous. Women in their reproductive years with symptoms of pelvic endometriosis, who report cyclic chest pain, dyspnea, or hemoptysis should direct clinicians to entertain idea of CP. A combinations of VATS pleural inspection, pleurodesis, and postoperative hormonal suppression with estradiol/progestin or GnRH agonists, is a preferred modality of management. Pleurectomy or TAH-BSO especially is considered to be the last resort, as the latter terminate fertility although thought to be highly effective.

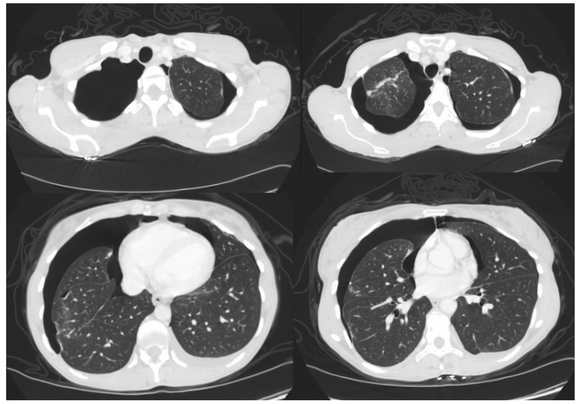

Figure 1. CT Chest with contrast

1. Moderate right and small left pneumothoraces (R40%-L20%).
2. There are a few scattered bilateral pulmonary nodules.

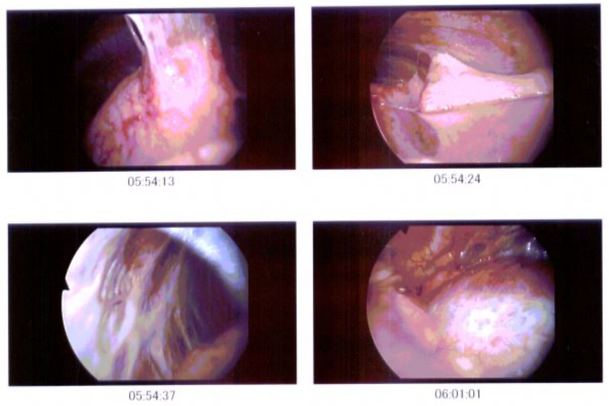

Figure 2. Video-Assisted Thoracoscopy (VATS)

1. Multiple adhesions within the thoracic wall
2. A dense white fibrotic plaque particularly along the posterior aspect of the thorax.
3. White plaquing with a small bluish discoloration over the diaphragm.