Extraperitoneal Endometriosis, Catamenial Pneumothoracies, And Review of the Literature

June 28, 2011 | Laparoscopy [1], Contraception [2], Pelvic Pain [3], Endometriosis [4]

Objective: To present a case of catamenial pneumothorax and diaphragmatic endometriosis that was managed thoracoscopically. A review of the literature is also presented. A 28-year-old woman initially presented with bloody stools and chronic constipation. During a review of systems, the patient described monthly chest pain associated with her menses. The initial workup included a pre-operative chest x-ray that revealed a right pneumothorax and colonoscopy that revealed biopsy-proven endometriosis of the sigmoid colon.

Abstract

Objective: To present a case of catamenial pneumothorax and diaphragmatic endometriosis that was managed thoracoscopically. A review of the literature is also presented. A 28-year-old woman initially presented with bloody stools and chronic constipation. During a review of systems, the patient described monthly chest pain associated with her menses. The initial workup included a pre-operative chest x-ray that revealed a right pneumothorax and colonoscopy that revealed biopsy-proven endometriosis of the sigmoid colon. An abdominal-pelvic CT scan revealed a 30% right pneumothorax and bilateral complex cystic and solid adnexal lesions. A chest tube was placed followed by resolution of the pneumothorax. High-resolution chest CT was normal. The patient underwent elective right thoracoscopy. A 1.5 cm x 1.0 cm lesion on the right hemidiaphragm was excised with endoscopic shears and histologically confirmed to be endometriosis. A 2.0 cm x 1.5 cm x 1.0 cm wedge section of right apical lung tissue containing a bleb was resected using the endoscopic stapler. This specimen also contained endometriosis. Pulmonary endometriosis may present with chest pain, shortness of breath, or hemoptysis associated with menstrual cycles. This case emphasizes the importance of a careful review of systems in patients with known endometriosis. Several cases of catamenial pneumothorax have been described in the literature. There are a few theories explaining the etiology of extra peritoneal endometriosis and the mechanisms for symptomatology. Management which has traditionally included medical and open surgical options now includes and endoscopic alternative and all of its known benefits. The first report of catamenial pneumothorax appeared in the literature in 1958 (Maurer, et al.). There have been over one hundred cases reported. Diagnosis is often made based on the patient's symptoms. Extra peritoneal endometriosis has also been identified in the extremities and in the male urinary system (Nunn,1949; Patel et al,1982; Gitelis et al,1985; Das Gupta et al,1985; Oliker and Harris,1971; Pinkert et al,1979; Schrodt et al,1980; Martin and Hauck,1985). There are several theories that attempt to explain these extra pelvic sites of endometriosis. One theory may adequately support one site but doesn't explain another. Treatment has included both medical and surgical options. With the development of advanced endoscopic techniques, pulmonary endometriosis can now be treated thoracoscopically with excellent results.

Case Reports

A 28-year-old woman initially presented with bloody stools and chronic constipation. During a review of systems, the patient described monthly right-sided chest pain associated with her menses. She also described pelvic pain with her menses that had worsened over the previous two months. Physical examination revealed a healthy-appearing young female. On pelvic exam, the rectum felt adherent to the posterior cervix and uterus between the uterosacral ligaments. Rectal exam revealed a nodular lesion on the anterior rectum at approximately 10cm. She was evaluated by a gastroenterologist and underwent colonoscopy. A submucosal appearing lesion at 20cm from the anal verge was biopsied and histologically reported as submucosal endometriosis overlying colonic mucosa with polyoid hyperplastic change and lymphoid aggregates. The initial workup included a pre-operative chest x-ray that revealed a right pneumothorax. An abdominal-pelvic CT scan revealed a 30% right pneumothorax and bilateral complex cystic and solid adnexal lesions. A chest tube was placed resolving the pneumothorax. High-resolution chest CT was normal. A diagnosis of catamenial pneumothorax was made based on the patient's history of chest pain associated with the onset of her menstrual cycles and objective findings on radiologic studies. The patient underwent elective...
right thoracoscopy. A 1.5 cm x 1.0 cm lesion on the right hemidiaphragm was observed. Using endoscopic shears, the lesion was excised in entirety. The diaphragm was repaired with interrupted 0 Ethibond sutures placed thoracoscopically and tied in succession. Next, a bleb of the right upper lobe was identified.

A 2.0 cm x 1.5 cm x 1.0 cm wedge section of right apical lung tissue containing the bleb was resected using the endoscopic stapler. Both specimens were evaluated histologically and noted to have endometrial glands and stroma within the tissue confirming extra peritoneal endometriosis.
Discussion
Seventy-five percent of pulmonary endometriosis will present as catamenial pneumothorax, 10.7% as cyclical hemothorax, and 8.3% as hemoptysis. The rest will remain asymptomatic nodules and may be discovered incidentally (Karpel et al, 1985). In general, endometriosis of the pleura or diaphragm will present as cyclic pain or dyspnea associated with a pneumothorax that is usually right-sided. Parenchymal endometriosis is usually asymptomatic and may be associated with cyclic hemoptysis (Foster et al, 1981). Diagnosis of catamenial pneumothorax is most often made clinically, after the exclusion of other causes. Although not in the case presented here, computed tomography (CT) and magnetic resonance imaging (MRI) have been useful in locating the pulmonary endometriosis (Hertzanu et al, 1987; Volkart, 1995; Cassina et al, 1997). MRI appears to be superior when differentiating between pleural and parenchymal disease (Cassina et al, 1997). The mechanism for air entry into the thorax remains controversial. Diaphragmatic fenestrations are seen in only 19-33% of patients with catamenial pneumothorax (Schoenfeld et al, 1986). There is no obvious defect in the majority of patients but microscopic passages between the peritoneal cavity and pleural cavity have been documented using 131I-labeled albumin injected into the peritoneal cavity (Rosenshein et al, 1979). It has been suggested that the genital tract may be a source of air entry (Crutcher et al, 1967). The ball-valve theory proposed by Lillington suggests that air enters the pleural space as endometriosis invades the visceral pleura and impinges upon a small bronchiole (Lillington et al, 1972). Air can enter the space, but because the implant functions as a one-way valve, the air cannot escape. Rossi and Goplerud (1974) hypothesized the entry of air into the pleural space results from rupture of alveoli in response to vasospasm and bronchospasm due to elevated prostaglandins during menses.
In 1927 John Sampson published his classic paper coining the term "endometriosis" and establishing retrograde flow of endometrial tissue into the peritoneal cavity as the cause of this disease. There are two main problems with Sampson's theory: 1) many women have retrograde menstrual flow and do not develop endometriosis and 2) his theory does not explain how endometriosis occurs in sites remote from the pelvis. Other theories were developed.

The coelomic metaplasia theory best explains the presence of endometriosis in the extremities. During embryologic development, the coelomic epithelium is adjacent to the mesenchymal limb buds. If the differentiating coelomic cells are incorrectly integrated into the limb buds, under certain circumstances, they may be stimulated to develop into endometrial tissue responsive to steroid hormones (Suginami, 1991). This theory is also most plausible for explaining the presence of endometriosis found in the male urinary system, usually associated with high dose estrogen treatment (Oliker and Harris, 1971; Pinkert et al, 1979; Schrodt et al, 1980; Martin and Hauck, 1985). These theories can also be applied to pulmonary endometriosis. The pleura arises from coelomic epithelium and may too be stimulated under appropriate circumstances to develop into endometrial tissue (Suginami, 1991). Others have suggested that endometrial cell seeding from retrograde flow...
occurs after passage through diaphragmatic fenestrations, but diaphragmatic defects have been found in only 15-33% of patients with pleural endometriosis (Slasky et al,1982;Foster et al,1981). Vascular/lymphatic transport is another classic theory. Hobbs and Bortnick (1940) investigated this theory by injecting into the ear vein of rabbits, endometrial tissue from the same animal. They found pulmonary endometriosis developed in 79% (19/24) of the animals. Extra pelvic endometriosis and catamenial pneumothoraces have been treated with a variety of medical therapies. Successful results have been reported with danazol and gonadotropin-releasing hormone agonists (GnRH agonists) (Koizumi et al,1999; Morita et al,1997; Shek et al,1995). They are similar in their abilities to suppress disease, but both offer undesirable side effects (Mann et al,1986). The most common side effects of danazol are weight gain, fluid retention, fatigue, acne, oily skin, hot flushes, growth of facial hair, emotional lability, and possible irreversible deepening of the voice. The hypoestrogenic state induced by GnRH agonists may cause hot flushes, vaginal dryness, and bone loss. The bone loss is reversible and can be decreased with progestational or postmenopausal estrogen-progestin add-back therapy (Cedars et al,1990; Surrey and Judd,1992; Friedman and Hornstein,1993). Other commonly used regimens for the treatment of pelvic endometriosis are continuous oral contraceptive pills (no "pill-free" week), medroxyprogesterone acetate 30mg/d, and megestrol acetate 40mg/d. All of these drug therapies suppress endometriosis, but do not cure the disease. Thus the reason for failures or "recurrences." The only definitive cure for endometriosis is surgical resection or complete destruction of the endometriotic lesion. Surgical management does not include bilateral salpingo-oophorectomy. Removal of the ovaries will not ensure relief of symptoms. Estrogen is also produced by peripheral conversion of androgens (primarily androstenedione of adrenal and ovarian origin). This conversion is catalyzed by aromatase. Aberrant aromatase expression has been identified in endometriotic implants causing local production of estradiol within the implant (Noble et al,1996). Therefore, medical therapy remains a first-line treatment option, but it is not unreasonable to proceed directly to surgical management for more definitive treatment. Now with the ability to perform the procedure endoscopically, surgical management becomes an even more attractive option.

**References:**
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**References**
continuous infusion of a gonadotropin-releasing hormone agonist compared to treatment with a progesterational steroid. J Clin Endocrinol Metab 1986;63:1277-1283. ([Medline])

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