Endometriosis-related pneumothorax: Clinicopathologic observations from a newly diagnosed case

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Pneumothorax related to thoracic endometriosis has been generally considered to be a rare entity. Exact causative and pathogenic mechanisms are relatively poorly known, and controversies continue. Very recently, Alifano and coworkers performed a prospective study on spontaneous pneumothorax (SP) in women of reproductive age and found that catamenial pneumothorax (CP), a typical manifestation of thoracic endometriosis, accounted for 25% of all cases of SP referred for surgical intervention. In that study all the patients with CP had diaphragmatic abnormalities (nodules or holes), and endometriosis was proved in all but one case. Treatment involved partial diaphragmatic resection, mechanical pleurodesis, and ovarian suppression therapy.

In the present article the clinicopathologic findings of a newly diagnosed case of endometriosis-related pneumothorax were studied to provide further insight into this condition.

Clinical Summary
A 39-year-old woman with an unremarkable clinical history was hospitalized for dyspnea and right-sided chest pain. Chest radiography revealed a complete right-sided pneumothorax. Tube thoracostomy was performed, obtaining immediate lung re-expansion. Drainage was discontinued on the third day, and the patient was discharged. Because pneumothorax had occurred on the third day of her menstrual cycle, CP was suspected, and therefore the patient was seen at the outpatient clinic on the fourth day of the subsequent cycle. The results of clinical examination and chest radiography were strictly normal. One week thereafter, mild exertional dyspnea appeared. Chest radiography and thoracic computed tomographic scanning revealed a partial pneumothorax. The decision to perform video-assisted thoracoscopy was made. At exploration, both the lung and the parietal pleura were normal, and no pulmonary air leak was identified at lung re-expansion with the cavity filled with saline solution. Exploration of the diaphragm showed multiple infracentimetric holes and brown-violet nodules, involving a 3 × 5–cm area of the tendinous portion of the muscle. On the basis of these findings and in agreement with previous experience, conversion to video-assisted minithoracotomy was made, and a partial diaphragmatic resection, including the pathologic area, was carried out. The defect was repaired with interrupted nonabsorbable sutures. Pleural brushing was performed.

The postoperative course was uneventful, and control inspiratory and expiratory chest radiographs were fully satisfactory. A 6-month ovarian suppression therapy (triptorelin, 3.75 mg [Decapeptyl; Ipsen Biotech, Paris, France], administered intramuscularly once a month) was planned.

Grossly, the resected specimen was an extremely thin portion of diaphragm with several infracentimetric holes and nodules (Figure 1.A). In some areas the whole thickness of the diaphragm was less than 80 μm. At light microscopy, the diaphragm mainly...
consisted of adipose and loose connective tissue with or without a thin strand of dense fibrous tissue. Endometrial implants were found, consisting of foci of endometrial stroma with fresh hemorrhage, often lined with glandular epithelium (Figure 1, B and C).

Discussion

CP (ie, a recurrent pneumothorax occurring within 72 hours from the onset of menses\(^3\)) is generally considered to be a typical presentation of thoracic endometriosis. Several pathogenic mechanisms have been evoked\(^1,3,5\) including sloughing of visceral pleural implants with subsequent air leak, rupture of blebs or peripheral alveoli perhaps subsequent to the bronchiolar spasm caused by paracrine secretion of prostaglandin by lung endometrial implants, and transdiaphragmatic passage of air through diaphragmatic defects. The latter mechanism has been indicated as the most important one in the only prospective study\(^3\) published thus far. Furthermore, a radiologic sign identifying this mechanism has been documented.\(^5\) The diaphragmatic defects are thought to be caused by sloughing of endometrial implants, although congenital defects (congenital porous diaphragm) would be responsible for the disease in some cases.\(^1,6\)

Our case does not fulfill the criteria for definition of CP because only a single episode was verified in the menstrual period. On the other hand, diaphragmatic endometriosis with holes and implants was proved in the absence of any lung abnormality. The first episode was related to the menses: in this period the absence of a cervical mucus plug favors the aspiration of external air and its passage through the genital tract and the peritoneal cavity up to the thorax where a negative pressure exists.\(^1\) In the intermenstrual period air might be forced from outside to the peritoneum through physical activity or sexual intercourse.\(^6,7\) If diaphragmatic defects exist, a pneumothorax might develop, as in our patient. Thus our case might be defined as “thoracic endometriosis-related pneumothorax” rather than CP.

The other interesting feature of our case is the pathologic appearance of the resected diaphragm. Together with the well-known holes and endometrial implants, most of the surface of the resected diaphragm showed signs of involution, being extremely thin and presenting several holes. It has been reported that partial rupture of the diaphragm with intrathoracic liver herniation might be caused by diaphragmatic endometriosis.\(^8\) Our report suggests that this might be caused by progressive coalescence of holes and
thinning of the diaphragm caused by substitution of its normal structure by sloughing endometrial tissue.

This report suggests that thoracic endometriosis should be suspected in any case of recurrent SP occurring in women of reproductive age, especially if right sided, even when episodes occur out of the menstrual period. The report straightens the possible risks related to the lack of recognition, inadequate management, or both of diaphragmatic endometriosis, especially in terms of possible late rupture.

References

Integrated overlapping ventriculoplasty combined with papillary muscle plication for severely dilated heart failure

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We have previously reported on the overlapping cardiac volume reduction (OLCVR) operation and obtained acceptable clinical outcomes. To enhance remodeling effects by changing the shape of the ventricle elliptically, we performed papillary muscle plication (PMP) combined with the OLCVR operation in 8 recent cases. This brief communication reports the favorable early results.

Clinical Summary
Eight patients (5 male and 3 female patients; mean age, 54 ± 6 years) underwent the OLCVR operation combined with PMP from March 2003. Underlying diseases were ischemic dilated cardiomyopathy (ICM) in 4 patients and idiopathic dilated cardiomyopathy (DCM) in 4 patients. All patients had grade 3 (n = 4) to grade 4 (n = 4) mitral regurgitation. The preoperative ejection fraction was 22% ± 5%, and the left ventricular (LV) diastolic dimension was 72 ± 4 mm, as evaluated by means of echocardiography. The LV end-diastolic volume index, assessed by means of left ventriculography, was 198 ± 42 mL/m². Preoperative New York Heart Association functional class was III in 3 patients and IV in 5 patients, including 2 catecholamine and intra-aortic balloon pump (IABP)–dependent patients. Emergency operations were performed for 2 patients.

Informed consent was obtained before the operation and after full explanation. Mitral annuloplasty with an undersized artificial ring was performed in all patients during blood cardioplegic arrest. Next, a 10-cm-long incision was made along the left anterior descending coronary artery in the enlarged LV free wall. Through the incision, PMP was carried out with 3 autologous pericardium-pledgeted mattress sutures. These sutures were placed through the trabeculae around the bases of the anterior and posterior papillary muscles, with the deepest being just below the site of chordal attachment. The left marginal incision was then continuously su-