Meth et al. described a young pregnant patient with a large pulmonary embolism and wheezing who developed pulmonary edema the day after presentation. She also had leukocytosis and subsequently had a positive methacholine challenge. The relationship between the embolism, roentgenographic findings, bronchospasm, and pregnancy are not entirely clear.

Cases presented by Hyers et al., Dombret et al., and Alpert et al. all involved massive pulmonary emboli, with edema occurring only in the lung zones with preserved perfusion, and normal or elevated pulmonary artery pressures. The postulated mechanism in these cases included hyperperfusion with microvascular damage. Other cases have been presented in which pulmonary edema developed in areas of lung reperfused by surgery or thrombolytic therapy. Presumably the increased vascular permeability was due to ischemic, surgical, or reperfusion injury to the endothelium in the reperfused area.

The effects of pulmonary embolization have been studied extensively in sheep by Ohkuda et al. and Flick et al. They have found that edema occurs in areas of preserved perfusion and may be related to released humoral factors, as leukocyte-depleted sheep have significantly less edema with equivalent pulmonary embolization and similar increments in pulmonary artery pressure than nondepleted sheep.

The development of pulmonary edema after microembolization has also been studied in dogs, and it has been found to be dependent on fibrinogen, as defibrinogenation prevented the development of pulmonary edema in a study by Johnson and Malik. They postulated that the release of humoral factors after fibrin entrapment in the lung leads to increased pulmonary vascular permeability and subsequent pulmonary edema.

Our patient, in contrast to other reported patients, had neither cardiac dysfunction nor massive embolization. We were unable to find any similar cases reported in the literature. We believe this case supports the hypothesis that even following a small pulmonary embolism, local release of humoral factors may result in extravasation of fluid across pulmonary capillary membranes, either because of increased pulmonary vascular permeability or dilatation of the precapillary vessels with a transient increase in hydrostatic pressure in the microcirculatory bed. Although the precise mechanism of pulmonary edema in pulmonary embolism remains to be discovered, this case illustrates that pulmonary embolism should be included in the differential diagnosis of pulmonary edema.

REFERENCES

Concurrent Catamenial Hemothorax and Hemopneumothorax*

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We describe a case of catamenial hemothorax and hemopneumothorax occurring on both sides simultaneously; the patient responded remarkably with danazol therapy. To our knowledge, this is previously unreported in the literature.

(Chest 1993; 103:646-48)

Thoracic abnormalities associated with endometriosis were first described by Maurer et al. in 1958. The monthly periodicity of the symptoms led to the coinage of "catamenial" (in Greek—monthly). Endometriosis can involve the pleura, the lung parenchyma, and the airway. The term catamenial hemoptysis was first proposed by Rodman and Jones. In 1972, Lillington et al. described catamenial pneumothorax. Catamenial hemothorax is also well documented. The concurrent presence of catamenial pleural effusion on one side and catamenial hydropneumothorax on the other side was not encountered in our literature search. Hence, possibly, this is the first report.

CASE REPORT

A 28-year-old unmarried woman, admitted to the hospital for repair of a left femoral hernia, was referred for preoperative evaluation of respiratory symptoms. She gave a history of recurrent episodes of cough and dyspnea of 2½ years' duration. On examination, the patient was pale, had hepatomegaly, bilateral pleural effusion (left more than right), and a pelvic mass palpable per abdomen. Vaginal examination revealed a 10×15-cm mass on the left side contiguous with the uterus.

Results of a urinalysis and complete blood cell count were normal. Sputum was negative for acid-fast bacilli by direct smear. Sputum cytologic study for malignant cells was negative. The LE cell, ANA, and rheumatoid factor were negative. Blood chemistry results were normal except for elevated SGOT (90 IU/L) and SGPT (94 IU/L). A tuberculin test with 1 TU PPD was negative (2 mm). Echocardiography was within normal limits. Chest roentgenograms were consistent with the diagnosis of bilateral pleural effusion. Thyroid hormone

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assay results were normal. Pulmonary function test revealed gross restriction. Ultrasound scan of the abdomen showed a 6-cm irregular mass in the pelvis in relation to the uterus. Bronchoscopy result was normal. A biopsy specimen was taken from a soft left supraclavicular lymph node and it showed only reactive changes. 

Hemorrhagic fluid was aspirated from both sides, but the fluid kept on reaccumulating, requiring repeated aspiration. Repeated examination for malignant cells and culture for microbes were negative. Five pleural biopsies were done that failed to reveal any malignant infiltration of the pleura and were inconclusive. A presumptive diagnosis of Pseudomeig’s syndrome was made. The patient underwent a laparotomy. About 100 ml of hemorrhagic ascitic fluid was obtained which on cytologic examination revealed only RBCs. There were multiple uterine fibroids and another on the round ligament, the largest being 10 × 10 cm. The left ovary had a small cyst that contained clear fluid. Evidence of adenomyosis was present with dark hemorrhagic spots on the myometrium. Myomectomy was done. Histologic examination of the surgical specimen showed leiomyoma with adenomyosis.

The patient had remarkable improvement and had no dyspnea for two months; chest roentgenogram showed clearing of the effusion. Three months later, the patient again presented with exertional dyspnea. Chest roentgenogram showed evidence of hydropneumothorax and small rounded pleural opacities on the right side and pleural effusion on the left (Fig 1). Three days later, 250 ml of hemorrhagic fluid and 300 ml of air were aspirated from the right side. The patient was kept under observation, and serial roentgenograms and physical examination revealed collection of pleural fluid coinciding with menstruation and spontaneous resolution within 10 to 12 days after menstruation. During the next menstrual period, 300 ml of hemorrhagic fluid was aspirated from the left pleural space on the second day. Cytologic examination of the fluid showed the effusion to be consistent with menstrual fluid. The patient was started on a regimen of danazol 200 mg three times daily. She improved with clearing of the effusion and on follow-up for the last six months, no recurrence was noted.

**DISCUSSION**

The pleura is the most commonly involved structure in thoracic endometriosis. Catamenial hemothorax is a rare entity suggesting intrapulmonary or bronchial involvement. The occurrence of hemothorax has been reviewed by Wilkins et al. In the 15 cases, the effusions were closer in appearance to gross blood as in our patient. Pleural effusion was present in every case, only on the right side, in contrast to the bilateral hemothorax in our patient. The majority are nulliparous and they do not notice an association between dyspnea and menstrual bleeding.

The mechanisms postulated for the occurrence of hemothorax and pneumothorax with pleural endometriosis differ. The hemothorax occurs mainly from the pleural lesions that arise from transscocelic spread of stromal and glandular tissue elements, from pelvic uterine or extraterine endometrial tissue. Various mechanisms have been postulated for the occurrence of catamenial pneumothorax. Some of them are leakage of air from peritoneal cavity, focal thoracic implants that either break down during menstruation or swell in response to hormonal changes causing check valve obstruction of terminal bronchioles, factor-mediated mechanisms (PG-F2), and exacerbation of asthma. It is possible that several of the above-mentioned mechanisms play a role in any individual patient. The various methods in arriving at a diagnosis include a detailed history, evidence of pelvic endometriosis, CT of the thorax to locate endometrial implants and CT-guided biopsy, pleuroscopic examination for implants and cytologic examination of the pleural fluid, and recognition of endometrial epithelial cells and hemosiderin-laden histiocytic cells.

The management of individual episodes of hemothorax is identical to that of effusion of any etiology. Definite cure is not possible by surgical ablation due to the disseminated nature of the lesion. Hysterectomy with oophorectomy or hormonal therapy are directed to decrease estrogen secretion. The most common treatment is administration of oral contraceptives to suppress ovulation. The alkyl derivatives of testosterone—ethisterone and danazol—are very effective in all forms of endometriosis, especially with extraterine manifestations. The endocrine pharmacology of danazol is postulated as a selective antagonadotropic with mild androgenic effect. The dosage regimen has varied from 200 to 800 mg/day for an average of 12 to 14 months. At present, danazol is considered the first-line treatment for all forms of pulmonary endometriosis. Pleurodesis or pleurectomy are suitable alternatives for pleural involvement. Other forms of hormonal manipulations are less effective and should be used as supplementary measures when the above management fails to achieve a satisfactory result.

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**FIGURE 1.** Chest roentgenogram showing hydropneumothorax and pleural deposits on the right and pleural effusion on the left.
Visceral leishmaniasis is increasingly reported in immunocompromised patients, including patients with AIDS. We report a case of visceral leishmaniasis in an AIDS patient who presented with pulmonary symptoms and bilateral pleural effusions. Histologic evaluation of pleural fluid and bone marrow revealed histiocytes with intracellular Leishmania amastigotes. Visceral leishmaniasis should be considered in AIDS patients with a significant travel history who present with unexplained pulmonary symptoms.

(Chest 1993; 103:648-49)

Infections with protozoal pathogens are common in AIDS. Recently, there have been increasing reports of reactivation of visceral leishmaniasis (kala-azar) in AIDS patients.1-3 We report an unusual case of visceral leishmaniasis in an AIDS patient manifesting as bilateral pleural effusions with no apparent splenomegaly.

**Case Report**

A 46-year-old male homosexual with diffuse cutaneous Kaposi's sarcoma had a two-month history of progressive weight loss, dyspnea, and cough productive of brownish sputum. Previous infections included hepatitis B, gonorrhea, amebiasis and giardiasis. He had traveled to Manila, Borneo, Saudi Arabia, India, Indonesia, Morocco and Cairo, and had lived in Athens, Greece, for five years prior to his return to Rochester, NY, for hospitalization.

On admission, oral temperature was 35.2°C and diffuse bluish-colored nodules and plaques were noted on the skin. Diffuse adenopathy was present, as well as a 3 x 4-cm pedunculated nodular mass in his posterior pharynx. There were clear signs of bilateral pleural effusion. Cardiac examination was normal. His liver span was 14 cm, but no splenomegaly or abdominal masses were detected. His stools were guaiac-positive. Pitting edema (2+) was present below the knees. Neurologic examination was, with the exception of diffuse weakness, within normal limits. The white blood cell count was 2.1 thousand/cu mm with 80 percent polymorphonuclear cells, 16 percent lymphocytes and 4 percent monocytes. The hematocrit was 23 percent. Clinical chemistry values were normal. Sputum cultures grew 1+ yeast and herpes simplex virus. While breathing room air, he had a PaO2 of 77 mm Hg, and a chest x-ray film showed large bilateral pleural effusions. Thoracentesis revealed a serosanguineous fluid with 111,000 red and 467 white blood cells per cubic millimeter with a predominance of histiocytes. Blood chemistry analysis of pleural fluid included a glucose value of 113 mg/dl and a protein value of 4.8 g/dl and an LDH level of 158 IU/L (serum LDH, 170 IU/L). Histiocytes in the pleural fluid contained abundant Leishmania amastigotes (Fig 1), from which Leishmania promastigotes were cultured. Growth was insufficient to allow speciation. Bone marrow examination revealed macrophages containing similar intracellular amastigotes. The patient refused further evaluation and was treated with sodium stibogluconate (Pentostam) and periodic therapeutic thoracentesis.

Initially, pleural fluid decreased and after 10 days intracellular amastigotes no longer could be detected in the pleural fluid. However, the patient then developed elevated pancreatic enzyme levels and pleural fluid began to reaccumulate. Subsequently, his hematocrit value decreased and he developed hypotension, bilateral alveolar infiltrates and intractable seizures. The patient and his family refused all investigational efforts, and he expired less than one month after admission. Terminal blood cultures grew both Klebsiella pneumoniae and Mycobacterium avium intracellularare. An autopsy was refused.

**Discussion**

Visceral leishmaniasis, usually caused by L. donovani or

**Acquired Immunodeficiency Syndrome-Related Visceral Leishmaniasis Presenting in a Pleural Effusion**

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