

Pleural fluid accumulation due to intra-abdominal endometriosis: a case report and review of the literature

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Abstract

A case is presented of massive ascites and right sided pleural effusion caused by endometriosis. The final diagnosis was not made for a considerable time. Massive ascites and a right sided pleural effusion caused by endometriosis is rare, with fewer than 10 reports in the literature worldwide. Physicians should be aware of this potentially treatable cause, having excluded other possibilities such as malignancy and tuberculosis.

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Keywords: endometriosis, pleural effusion, ascites.

A 30 year old AfroCaribbean woman presented with a six month history of general malaise and abdominal distension. On examination she had ascites and a large right sided pleural effusion. Computed tomographic scanning, ultrasound imaging, and chest radiography (fig 1) confirmed a large right pleural effusion with no mass lesion, a 2 cm pericardial effusion, and gross abdominal ascites with the suggestion of

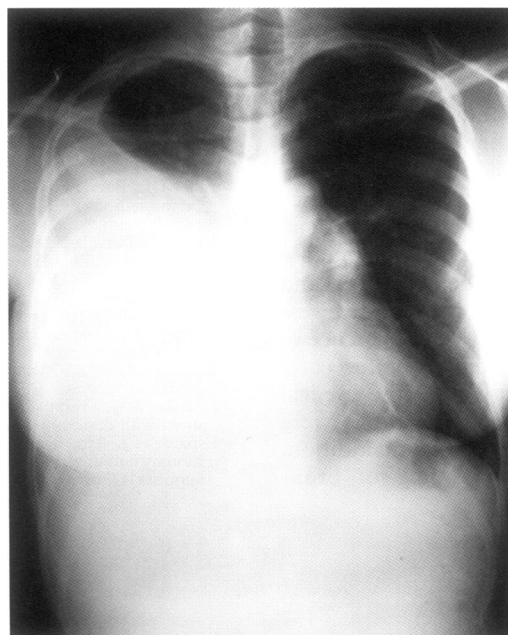


Figure 1 Chest radiograph at presentation.

right ovarian pathology compatible with Meigs' syndrome.

Two years previously she had presented with a burst chocolate cyst. Laparoscopy was performed, the cyst was removed, and a diagnosis of endometriosis was made from laparoscopic appearances, although no endometrial tissue was found in the biopsy samples. She was treated for endometriosis with danazol for nine months and remained well until she presented with the ascites and pleural effusion.

Diagnostic aspirates revealed chocolate coloured ascitic fluid and bloodstained pleural fluid, both exudates. Bacterial and tuberculosis cultures were negative, and the pleural fluid alpha-fetoprotein (AFP) and human chorionic gonadotrophin (HCG) levels were normal. Cytological examination of the pleural fluid showed poorly differentiated adenocarcinoma cells; ascitic fluid cytology was negative. Three litres of bloodstained pleural fluid were drained from the chest. Bronchoscopic examination was normal and medial thoracoscopy showed a normal pleural cavity with no evidence of pleural endometriosis and pleural biopsy samples were normal. Sputum culture and cytology was negative. At presentation full blood count, ESR, clotting screen, routine biochemistry, thyroid function, serum complement levels and immunoglobulins were all normal. HIV and hepatitis B serology was negative, and Tine test grade 2. Carcinoembryonic antigen (CEA) levels were normal, but the CA-125 antigen level was slightly raised at 49 kU/l (upper limit of normal 35).

Laparoscopy showed minimal endometriosis, a left ovarian mass, and perihepatic adhesions. Laparotomy was performed in view of the apparent ovarian mass and the left ovary was removed and a right ovarian wedge biopsy sample was taken. Histological examination showed normal ovaries and peritoneal endometriosis. A six month course of the gonadotrophin releasing hormone (GnRH) agonist leuprorelin acetate was given for endometriosis.

Pleural and ascitic aspirates performed twice over the next year showed no growth on bacterial and tuberculosis cultures and the results of cytological examination were negative. Several pleural biopsy specimens revealed no evidence of malignancy. Repeat surgical thoracoscopy performed at another hospital was normal, and surgical pleurodesis was performed. The pleural fluid reaccumulated over the next eight months.

Two years after presentation she still had a right pleural effusion, a pericardial effusion, and marked ascites. Pleural and ascitic fluid cultures for bacteria and tuberculosis were negative, and cytological examination revealed no neoplastic cells. Four litres of bloodstained pleural fluid were aspirated. Pleural biopsy samples showed fibrous thickened pleura and one possible granuloma, ZN stain for tuberculosis was negative, and bronchoscopic examination was normal.

A one month trial of prednisolone 30 mg per day had no effect on the fluid accumulation. The investigations were repeated and bacterial, tuberculous, and fungal cultures and cytology

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were negative, as were pleural fluid levels of rheumatoid factor, antinuclear factor, AFP, and CEA. The dsDNA, antinuclear antibodies, antineutrophil cytoplasmic antibodies, serum angiotensin converting enzyme, rheumatoid factor, AFP, CEA, and HCG levels were normal. The CA-125 level was 36 kU/l, ESR and CRP levels were normal. Sputum, stool and urine cultures were negative for bacteria and tuberculosis, as were stool and sputum samples for ova, cysts and parasites and strongyloides culture. A 24 hour terminal urine examination for schistosomal ova was negative, and serological tests for filarial, schistosomal, strongyloides, amoebic, and hydatid antibodies were negative. An excision biopsy sample of a persistent small mass in the right groin revealed endometriosis. Two litres of ascitic fluid and two litres of pleural fluid were aspirated to relieve discomfort.

After two and a half years of observation and extensive investigation it was felt that malignancy and tuberculosis had been effectively excluded despite the original positive cytological examination of the pleural fluid. It was considered that, in view of the persistent blood-stained fluid, the diagnosis was endometriosis. She was commenced on a six month course of monthly intramuscular injections of leuporelin acetate 3.75 mg. Two months later she developed a moderate pneumothorax in addition to the pleural fluid. Both were drained and a pleurectomy was performed, the histology of which showed no features diagnostic of endometriosis.

It is now nine months since the pleurectomy and the course of leuporelin was completed four months ago. A recent chest radiograph shows reaccumulation of the pleural fluid and her abdomen remains distended with ascites. She remains otherwise well and is extremely reluctant to have any more surgery at present.

Discussion

Massive ascites and right sided pleural effusions caused by intra-abdominal endometriosis is a rare but recognised phenomenon, with fewer than 10 cases in the literature since the first report by Brews in 1954.¹ Despite the fact that patients may present to a respiratory physician, these reports are mainly in the gynaecological literature. This case is typical in that the average age of presentation with ascites is 32 years,² 70% of subjects are black, and it is more common in nulliparous women.²⁻⁴

The most common presentation is increasing abdominal girth with chocolate coloured⁵ or bloodstained⁶ ascitic fluid. The average amount of ascites is 3.3 litres,⁴ the maximum reported being 10 litres.⁷ Ascites may result from rupture of endometriosis or chocolate cysts leading to peritoneal irritation,⁵ or it may occur in a similar way to that in Meigs' syndrome.³ Alternatively,

blocked lymphatic vessels secondary to adhesions may prevent escape of peritoneal fluid.

The pleural effusion generally occurs on the right,^{3,8} although Yu and Grimes reported bilateral pleural effusions.⁹ Pleural endometriosis is well recognised⁸ but, since our patient had a normal thoracoscopic examination on two occasions with normal pleural histology from the pleurectomy sample, pleural endometriosis is unlikely to have been responsible for the pleural effusion unless she had undetected deposits of endometrial tissue. It has been suggested that the right pleural effusion is caused by a communication between the pleural and peritoneal cavities⁸ and in one case three diaphragmatic perforations were seen at thoracoscopy.⁸ This would explain the likely mechanism of fluid reaccumulation in our patient and why surgical pleurodesis failed. Transdiaphragmatic lymphatic vessels are thought to be the source of pleural fluid in Meigs' syndrome and an alternative suggestion for the origin of the pleural fluid in cases of gross ascites associated with a pleural effusion is overloading of the lymphatic drainage of the thoracic cavity.^{9,10}

The definitive treatment for ascites is total abdominal hysterectomy and salpingo-oophorectomy.^{4,5} When accompanied by a pleural effusion thoracotomy and pleurectomy may be curative.⁸ Hormonal treatments – including progestogens,^{6,7} danazol,¹⁰ and the GnRH agonist leuporelin⁹ – have been successful. Radiation castration was the treatment of choice prior to this,¹ and others have used hormonal therapy in combination with surgery.^{2,3} Our patient was treated with danazol and leuporelin acetate with no clear response, and both pleurodesis and pleurectomy have also failed.

In a menstruating woman with persistent bloodstained pleural fluid endometriosis must be considered in the differential diagnosis once more common causes are excluded. Few such cases have been reported, and its rarity makes it difficult to define the best therapeutic approach.

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