Atypical Meigs' syndrome

CE HANDLER, RE FRAY, PD SNASHALL

From the Departments of Neurology, Obstetrics, and Gynaecology and Medicine, Charing Cross Hospital, London

Large blood-stained pleural effusions, especially in young patients, are unusual and may be caused by metastatic disease in the chest, a large pulmonary infarction, and, rarely, pulmonary tuberculosis. Meigs' syndrome is an uncommon cause of pleural effusion associated with ascites and a benign ovarian tumour, most often a fibroma. The fluid is usually clear but occasionally blood-stained. The mechanism of formation of the fluid remains a mystery ever since Meigs drew attention to this syndrome over 40 years ago.

We report a case of atypical Meigs' syndrome, which is defined as the association of ascites and pleural effusion with uterine fibromata. We have found only four such published cases.¹⁻⁷ Patients with atypical Meigs' syndrome may present a diagnostic problem as the clinical picture may masquerade as pulmonary tuberculosis or gynaecological carcinoma with metastatic involvement in the chest. They should, therefore, always be considered for exploratory laparotomy.

Case report

The patient was a 35-year-old West Indian nurse. She was admitted to Charing Cross Hospital in November 1980 with a two-month history of progressively increasing dyspnoea. Despite the use of oral contraceptives, her menstrual loss had been heavy with associated dysmenorrhoea and deep dyspareunia. She denied night sweats, anorexia, or weight loss. She had nursed patients with pulmonary tuberculosis and had not had a BCG vaccination.

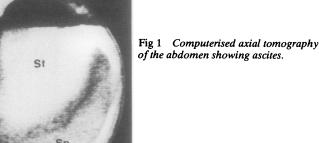
Examination revealed a well but anaemic patient. There was a large right-sided pleural effusion and an irregular pelvic mass. No ascites was clinically detectable.

A chest radiograph confirmed the pleural effusion. Haematological examination showed a haemoglobin of 8.4 g/dl, microcytes on the blood film, and normal platelet count and clotting screen. The erythrocyte sedimentation rate was 45 mm/h and a sickle-cell screening test was negative. Biochemical screening and urinalysis were normal.

Pleural biopsy with chest aspiration was performed with drainage of one litre of dark blood-stained fluid. No organisms were seen on either Gram or Ziehl Niellson preparation. The fluid revealed scanty white cells (80% lymphocytes and 15% atypical mononuclear cells). Lowenstein Jensen culture was negative. The glucose concentration was 3.5 mmol/l (plasma glucose 7.8 mmol/l) and the protein 53 g/l (plasma protein 74 g/l). Histological examination and culture of the pleura were normal. A Mantoux test at 1/100 000 was positive. Repeat cultures of pleural fluid were negative and a further pleural biopsy was normal. A ventilation perfusion lung scan was normal. Repeat chest radiography after a third aspiration showed complete clearing of the right pleural effusion but over the next few days this had partially reaccumulated.

A diagnosis of probable Meigs' syndrome was made and ascites was demonstrated on computerised axial tomography of the abdomen (fig 1).

Laparotomy revealed about 500 ml of blood-stained ascitic fluid and a large fibromyoma 9×9 cm in diameter was detached from the fundus of the uterus. The ovaries and the fallopian tubes were normal as were the liver, gall bladder, kidneys, and peritoneum. Histological examina-



Present address of Dr CE Handler: Cardiac Department, Guy's Hospital, London SEI 9RT.

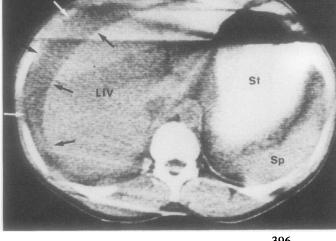




Fig 2 Section of excised uterine fibromyoma.

tion demonstrated that the tumour was a fibromyoma (fig 2). It was haemorrhagic and infarcted and showed hyaline change. The patient made an uneventful post-operative recovery. The pleural effusion had not reaccumulated and when the patient was reviewed one year postoperatively she was symptom free and her chest radiograph was normal.

Discussion

Pleural effusion and ascites in association with uterine fibromyomata is rare. In this case, our initial working diagnoses were metastatic tumour, tuberculosis, or pulmonary infarction. However, we failed to confirm these diagnoses and therefore diagnosed Meigs' syndrome by exclusion. At laparotomy, the ovaries were normal but she had a large uterine fibromyoma, excision of which led to resolution of the patient's ascites and pleural effusion. It is postulated that this case represents atypical Meigs' syndrome.

Meigs reviewed a series of 124 cases, five of which were uterine fibromyomata. In these cases, he suggested the diagnosis of pseudo-Meigs' syndrome. This is an even rarer occurrence. Such cases may masquerade, as in our patient, as a tuberculous pleural effusion,² or resemble inoperable malignant disease of the pelvis with pleural metastases.

The cause of ascites and pleural effusion in both Meigs' syndrome and atypical Meigs' syndrome is obscure.⁸⁻¹⁰ The ascites and pleural effusion are usually clear rather than blood-stained, making our case highly unusual. The pleural effusion is usually on the right side or occasionally left-sided but may be bilateral. The fluids have usually resembled a transudate and a low serum protein has been a contributory factor in the majority of reports but not in our case.

We should like to thank Dr GD Perkin and Mr CD Sims for their permission to report this case. We are grateful to Dr D Katz for his advice with the radiological investigations.

References

- ¹ Meigs JV. Pelvic tumours other than fibromas of the ovary with ascites and hydrothorax. *Obstet Gynaecol* 1954;3:471-86.
- ² Scott JK, Dick JA. Pseudo Meigs' syndrome masquerading as tuberculous effusion. *Tubercle* 1972;53:143-6.
- ³Salmon UJ. Benign pelvic tumours associated with ascites and pleural effusion. *Mt Sinai J Med (NY)* 1934;1:169-72.
- ⁴ Calmenson M, Dockerty MB, Bianco JJ. Certain pelvic tumours associated with ascites and hydrothorax. Surg Gynaecol Obstet 1947;84:181–91.
- ⁵ Martinez-Esteve P, Orrico J. Myoma of the uterus associated with ascites and hydrothorax. *West J Surg* 1950;**58**:28-31.
- Soloman S, Faber SJ. Fibromyomata of the uterus with haemothorax—Meigs' syndrome. Arch Intern Med 1971;127:307-9.
- ⁷ Frank N, Frank KJ. Uterine tumour mimicking Meigs' syndrome (intractable heart failure). J Med Soc N J 1978;70:17–18.
- ⁸ Selye H. A syndrome produced by divine noxious agents. *Nature* (London) 1936;138:32.
- ⁹ Dockerty MB, Masson JC. Ovarian fibromas: a clinical and pathological study of 283 cases. Am J Obstet Gynecol 1944;47:741-52.
- ¹⁰ Rubin IC, Novak J, Squire JJ. Ovarian fibromas and thecal cell tumours, ascites and hydrothorax (Meigs' syndrome). Am J Obstet Gynecol 1944;48:601-16.