

Meigs' Syndrome and Pseudo-Meigs' Syndrome: Report of Four Cases and Literature Reviews

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Abstract

Meigs' syndrome represents a triad of pleural effusion, ascites, and an ovarian tumor, which is usually benign, occurring together. We describe here 2 patients with Meigs' syndrome and 2 patients with pseudo-Meigs' syndrome. Hydrothorax and ascites symptoms in 4 patients are of outstanding performance characteristics of Meigs' syndrome and pseudo-Meigs' syndrome. Ovarian tumors were found by clinical examination and surgically removed. Postoperatively, these signs of hydrothorax and ascites were dissolved completely. Conclusions: These cases highlight the difficulties that may be encountered in the management of patients with Meigs' syndrome, including potential misdiagnosis of the tumor as a malignant ovarian neoplasm that may influence the medical and surgical approach, and the adverse impact that Meigs' syndrome can have on the patient's condition. Clinical doctors should be alert to this kind of disease under pathological diagnosis in the absence of a clear effect of chemotherapy or radiotherapy and to prevent unnecessary harm to the patient.

Keywords

Meigs' Syndrome, Clinicopathology, Diagnosis

1. Introduction

In 1934, Salmon described the association of pleural effusion with benign pelvic tumors. It was not until the re-

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port by Meigs and Cass in 1937 that widespread attention of the medical profession was drawn to the significance of pleural effusion and ascites in benign ovarian fibroma [1]. Rapid resolution of fluid accumulation was observed after tumor resection. The clinical syndrome was termed Meigs syndrome by Rhoads and Terrell [2]. The types of tumors include fibroma, fibroepithelial tumor, thecoma, granulosa-cell tumor, and sclerosing stromal tumor. Later, the same signs in association with benign ovarian teratoma, mucinous cystadenoma, pelvic hemangioma, uterus leiomyoma, and papilloma of fallopian tube are referred to as pseudo-Meigs' syndrome [3]. Meigs' syndrome and pseudo-Meigs' syndrome are a rare disorder. In this study, we described four new cases of Meigs' syndrome and pseudo-Meigs' syndrome to further characterize the clinical and pathological features of this rare entity.

2. Case Presentation

2.1. Case 1

A 29-year-old Chinese woman who presented with distended tender abdomen for a week is being discussed as our first case in this report. Ultrasonography confirmed ascites (**Figure 1**).

Pelvic Magnetic resonance imaging (MRI) examination discovered a 10.0 cm × 9.0 cm × 8.0 cm mass located left under of uterus. Investigations revealed a normal hemogram and serum biochemistry. Gynecological operation discovered a 9.0 cm × 8.0 cm × 5.0 cm solid mass originating from the left ovary and approximately 500 ml ascites volume. The patient underwent left oophorectomy. Histopathological analysis revealed left ovarian thecoma (**Figure 2**).

In the postoperative, there was regression of ascites. The patient's condition has remained satisfactory throughout the 1.5-year follow-up period.

2.2. Case 2

A 46-year-old Chinese woman who presented with chest distress, dyspnea for a month is being discussed as our second case in this report.

There was no history of fever, night sweat or significant weight loss. Ultrasonography examination revealed right pleural effusion. A diagnosis of a tuberculous pleurisy was made in local hospital. Aspiration of right pleural effusion was performed. Cytology of the effusion showed no tumor cells. An abdominal CT scan and ultrasonography revealed ascites and pelvic tumor. Gynecological operation discovered an 8.0 cm × 8.0 cm × 6.0 cm solid mass originating from the left ovary and approximately 3000 ml ascites volume (**Figure 3**).

The patient underwent left oophorectomy. Pathology diagnosis was ovarian fibroma (**Figure 4**).

In the postoperative, there was regression of pleural effusion and ascites. The patient's condition has remained satisfactory throughout the 2-year follow-up period.



Figure 1. Ultrasonography reveals pelvic fluid, and pelvic hypoechoic area in front of the uterus.

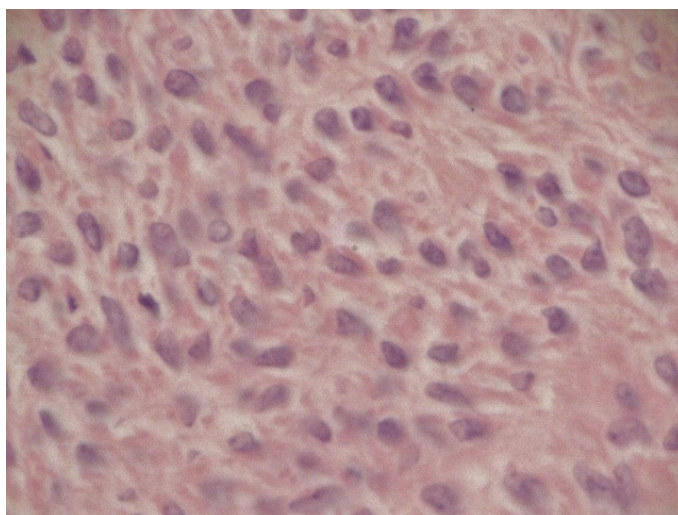


Figure 2. Histology of the ovarian tumor showing benign features such as short spindle cells, round or oval nucleus, cytoplasm rich in collagen fibers are separated (HE, original magnification $\times 400$).

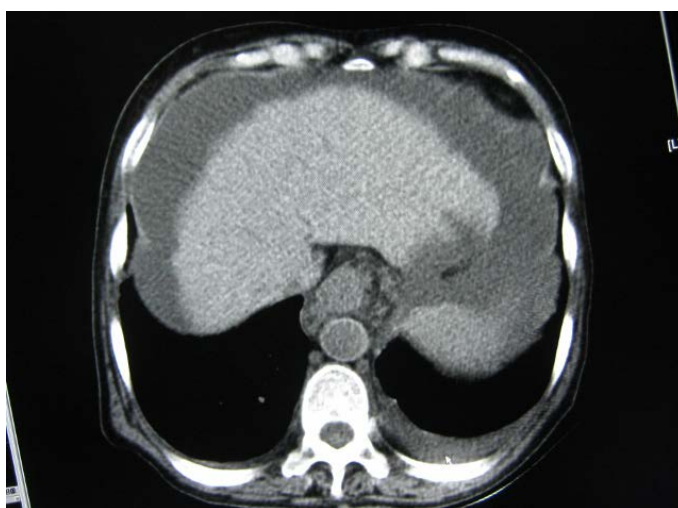


Figure 3. The CT scan shows pleural effusion and ascites.

2.3. Case 3

A 45-year-old woman who presented with chest distress and breathlessness for two months is being discussed as our third case in this report. A diagnosis of a tuberculous pleurisy was made in outside hospital. Repeated Thoracic drainage did not obvious invalid. The patient was turned into our hospital. A systemic examination revealed bilateral pleural effusion and pelvic mass. Serum levels of carcinoembryonic antigen (CEA), carbohydrate antigen 19 to 9 (CA199), and carbohydrate antigen 125 (CA-125) were normal. Gynecological operation discovered a 7.5 cm \times 6.5 cm \times 3.0 cm cystic mass originating from the right ovary and approximately 500 ml ascites volume. The patient underwent right oophorectomy. Pathology diagnosis was ovarian mature cystic teratoma (**Figure 5**).

In the postoperative for tenth day, there was regression of pleural effusion and ascites. The patient's condition has remained satisfactory throughout the 2.5-year follow-up period.

2.4. Case 4

A 72-year-old Chinese postmenopausal woman who presented with progressive abdominal distention for two

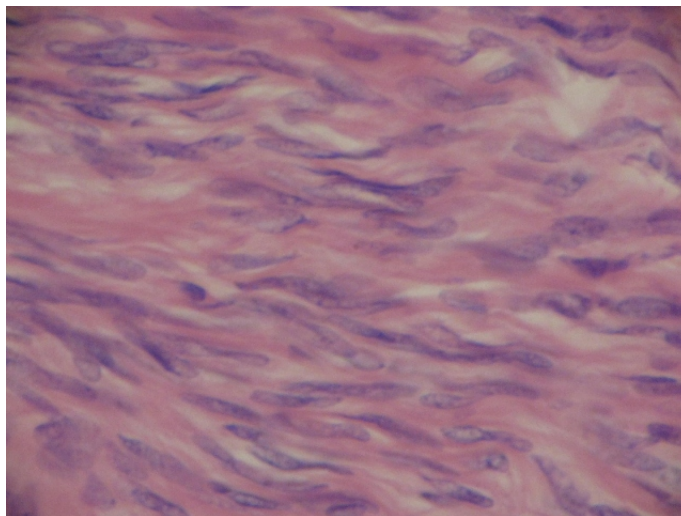


Figure 4. Histology of the ovarian tumor showing that the tumor was large number of spindled cells arranged in fascicles pattern (HE, original magnification $\times 400$).

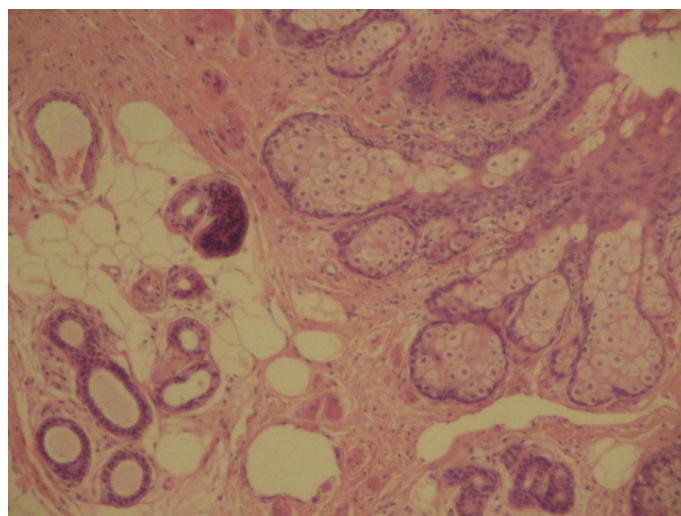


Figure 5. Histology of the ovarian tumor showing benign features such as teratoma ectoderm squamous epithelium, hair follicles, sweat glands, and sebaceous glands and the adipose tissue in the mesoderm(HE, original magnification $\times 100$).

months is being discussed as our fourth case in this report. Before arriving at our hospital, repeated abdominal drainage did not obvious invalid. There was no history of fever, significant weight loss, jaundice. There were no symptoms of hyperthyroidism. Ultrasonography confirmed uncovered substantial ascites and pelvic solid mass. The uterus and ovaries could not be identified separately from the pelvic mass. In addition, the serum CA125 determination revealed 3515 U/MI was found. Repeated ascitic analysis did not reveal any malignant cells. A diagnosis of a malignant ovarian neoplasm with ascites was made. Gynecological operation discovered a mass of multi-nodular originating from the right ovary and approximately 5000 ml ascites volume. The left ovary was atrophic. No peritoneal implant or abdominal lymphadenopathy was seen. The patient underwent bilateral oophorectomy. Pathological gross examination of the right ovarian mass revealed a 8.0 cm \times 7.0 cm \times 7.0 cm mass with a multi-nodular external surface. Cut surface was multicystic with cysts varying from 0.5 - 2.5 cm in diameter filled with a brown jelly-like gelatinous material. On microscopic examination, the right ovarian mass revealed normal ovarian architecture only in one focus with replacement of the rest of the ovarian parenchyma

by benign colloid filled thyroid follicles. No cytological feature of malignancy was seen. No neural tissue, cartilage, or adnexal tissue was seen. Sections from the left ovary showed compressed normal. Pathology diagnosis was right ovarian benign struma (**Figure 6**).

Three weeks after surgery, patients' CA125 had decreased to 592U/ml, and after one month when CA125 had decreased to normal. The patient is on follow-up with no recurrence of her symptoms.

3. Discussion

Meigs' syndrome is a rare disorder. In 1937, Meigs described a syndrome of ovarian fibromas with ascites and hydrothorax that quickly resolved on removal of the fibroma. Meigs separated his definition into true Meigs' syndrome, which he preferred to call Demons-Meigs' syndrome and pseudo-Meigs' syndrome distinguished by different tumor types. Meigs' criteria for diagnosis of Meigs' syndrome include four characteristics: the tumor must be a fibroma or fibroma-like tumor; ascites must be present; a hydrothorax must be present; and the ascites and hydrothorax resolve with removal of the tumor [4]. The diagnosis of pseudo-Meigs' should meet similar criteria, although the tumor type may be any of the types described above. In this paper, the four patients had complete resolution of the ascites and pleural effusion postoperatively. According to definition of Meigs' syndrome, the first patient and second patient are true Meigs' syndrome, the third and the fourth are pseudo-Meigs' syndrome in our cases.

It is rarely seen and its pathophysiology remains unclear [5]. An ovarian mass and pleural effusion and ascites in a female patients generally suggest a malignancy in clinical. An ovarian mass and an elevated serum CA125 level in a postmenopausal female generally suggest a malignancy. CA125 is the most important clinical marker for the diagnosis, treatment and follow-up of epithelial ovarian cancer. CA125 is now identified as the most widely studied serum biomarker for ovarian tumors [6]. Our fourth patient elevated serum CA125 level is suggestive of an ovarian malignancy. In the literature review, there has only been 10 cases of this tumor, associated with ascites and pleural effusion (Meig's Syndrome) and increased CA125 so far. In such cases, the tumor mimics malignant ovarian tumor [7]. The precise mechanism of serum CA125 level elevation which increased intra-abdominal pressure caused by tumor growth may also elicit mesothelial expression of CA125 [8]. The etiology of the ascitic fluid of Meigs' syndrome is unclear. Some investigators believe that the fluid is transuded from the surface of the tumor. Other proposed mechanisms for the production of ascitic fluid include direct pressure on surrounding lymphatics or vessels, hormonal stimulation, and tumor torsion. The etiology of the hydrothorax is also unclear. Transfer of ascitic fluid via transdiaphragmatic lymphatic channels is the current prevailing theory [2].

In our case 3 and case 4, there were ovarian mature cystic teratoma and ovarian benign struma. Mature cystic teratomas account for approximately 20% of all ovarian tumors. Of these, approximately 15% contain normal

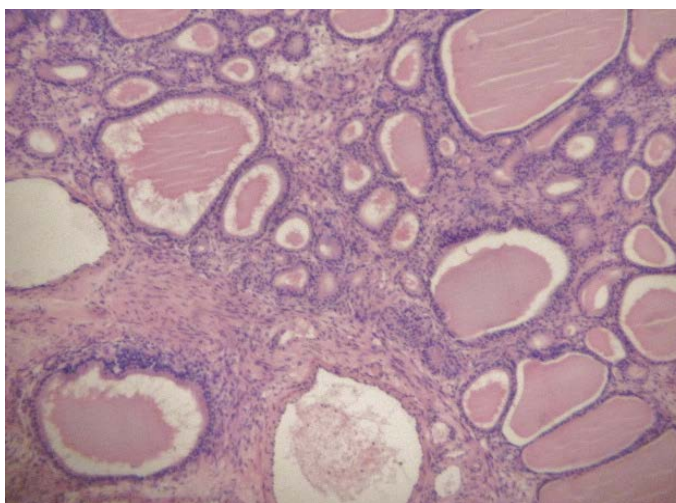


Figure 6. Histology of the ovarian tumor showing such as well-differentiated thyroid follicular, and only a small amount of normal ovarian tissue(HE, original magnification $\times 200$).

thyroid tissue [7]. Struma ovarii is a monodermal variant of ovarian teratoma. Preoperative clinical diagnosis of struma ovarii, however, is very difficult.

Despite containing thyroid tissue, only 5% of struma ovarii have features of hyperthyroidism [7]. Ascites has been reported in one-third of cases [2]. However, uncommon is the association of ascites and hydrothorax with this tumor. Surgical removal ovarian tumor is the treatment of first choice for Meigs' syndrome. In our patient, she presented with substantial ascites and elevated CA125 levels and this condition was rapidly resolved with surgical removal.

In conclusion, Meigs' syndrome is a benign disease with a good prognosis. Although this is a fairly rare syndrome, it should be considered in women presenting with unexplained hydrothorax and ascites. Suspicion of this syndrome allows for prompt diagnosis by ultrasound and subsequent tissue diagnosis. Clinical doctors should be alert to this kind of diseases under pathological diagnosis in the absence of a clear effect of chemotherapy or radiotherapy and to prevent unnecessary harm to the patient.

4. Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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