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Case report

Incomplete pseudo-Meigs' syndrome caused by endometrial ovarian cyst: A case report

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ABSTRACT

A 63-year-old female was admitted to our hospital with history of persistent dyspnea. Right pleural effusion and ovarian tumor were discovered, but here were no significant findings on thoracoscopy under local anesthesia. The pleural effusion was suspected to be secondary to Meigs' syndrome, and a diagnosis of endometriotic ovarian cyst was made. Since the pleural effusion resolved after surgery, the patient was diagnosed with incomplete pseudo-Meigs' syndrome. We consider this to be a valuable case, as there are no previously reported cases of pseudo-Meigs' syndrome derived from an endometriotic ovarian cyst, to the best of our knowledge.

1. Introduction

An ovarian fibroma, with pleural and ascitic fluid that disappears after removal of the tumor, is classified as Meigs' syndrome (MS) [1] when the primary tumor is fibroma, thecoma, granulosa cell tumor, or Brenner tumor. With other tumor types, such cases are defined as pseudo-Meigs' syndrome (PMS) [2].

Generally, both pleural effusion and ascites are recognized, but there are reported atypical cases with only pleural effusion, which are called incomplete Meigs' syndrome [3]. Here we report a case of incomplete pseudo-Meigs syndrome which was thought to be caused by an endometriotic cyst. This case was considered valuable, as to the best of our knowledge, there are no previously reported cases secondary to an endometriotic cyst.

2. Case

A 63-year-old woman developed fever and cough, and eight days later, palpitations and dyspnea. She visited our hospital on the same day and was admitted because of right pleural effusion. There was no medical or drug history, and no smoking or drinking history.

Physical findings included height 161.5 cm, weight 70.4 kg, blood pressure 139/87 mmHg, pulse rate 120 beats/minute, SpO2 92% (indoor air), and body temperature 37.0 °C. There were no superficial lymph nodes, and paucity of movements along with absent breath sounds over right hemithorax, the abdomen was flat and soft with no tenderness, and there was no skin rash.

Blood results at the time of admission showed no abnormalities in hepato-renal function, but CRP was as high as 6.16 mg/dl and CA125 increased to 114 U/ml (normal value $\leq$ 35). Various autoantibodies were negative.

Chest radiograph (Fig. 1) showed a large pleural effusion on the right side and mediastinal shift to the left. Pulmonary embolism and malignant disease were suspected and contrast-enhanced chest CT (Fig. 2) was performed for the purpose of systemic screening. A large right pleural effusion was identified, which caused complete right lung collapse and left mediastinal displacement. Contrast-enhanced abdominal CT showed an 18 mm nodule in the right ovary.

Subsequently, pleural effusion evaluation was performed. The chest exudate was yellow and turbid, specific gravity: 1.036, total cell number: 3383/μL (tissue monocyte: 2%, lymphocyte: 95%, neutrophil: 3%), RBC: 22,000/μL, LDH: 214 U/L, TP: 5.0 g/dL, Alb: 2.8 g/dL, AMY: 51 U/L, T-Chol: 142 mg/dL, RF: 2 IU/mL, glucose: 130 mg/dL, hyaluronic acid: 7400 ng/mL, CEA: 0.5 ng/mL, NSE: 2.5 ng/mL, SCC: 32.8 ng/mL, CYFRA: 9.9 ng/mL, Antinuclear antibody <40 times and ADA: 13.8 IU/L.Cytology revealed lymphocytes and mesothelial cells, but no findings suggestive of a malignant tumor.

Based on the above results, Meigs's syndrome was considered to be a differential of right unilateral pleural effusion, so pelvic contrast-enhanced MRI (Fig. 3) was performed for qualitative assessment of the

Abbreviations: Meigs' syndrome, MS; pseudo-Meigs' syndrome, PMS; pseudo-pseudo Meigs' syndrome, PPMS.

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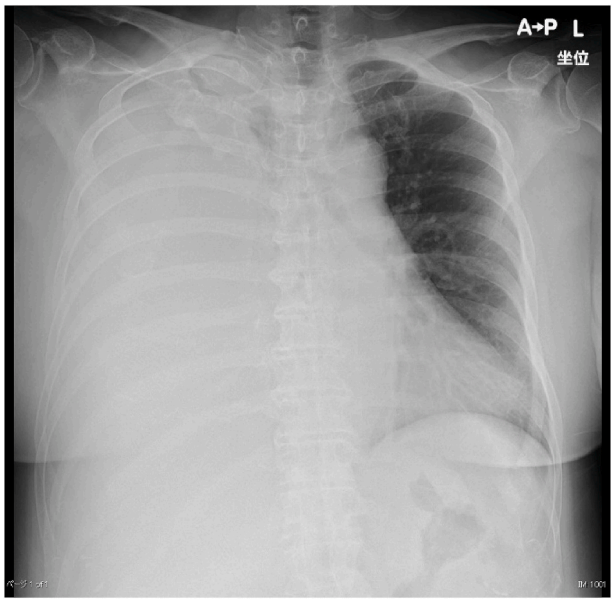
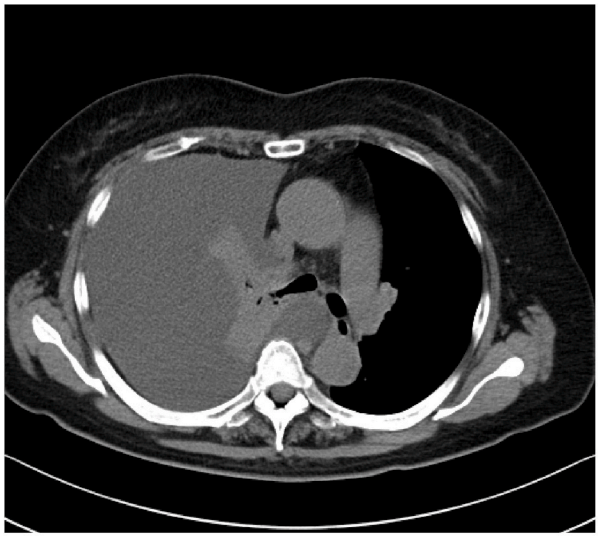


Fig. 1. Chest X-ray at the first visit.



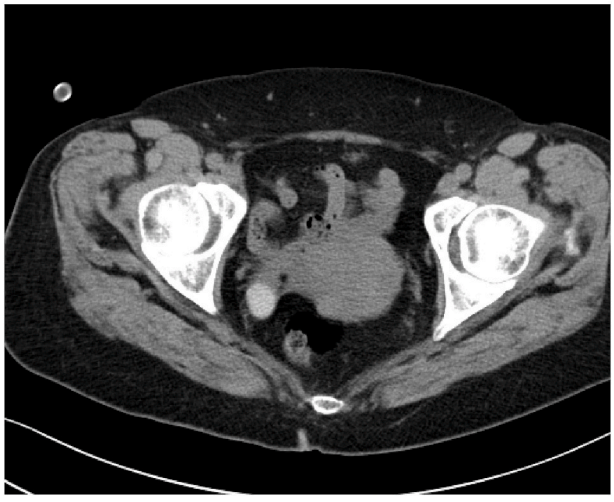
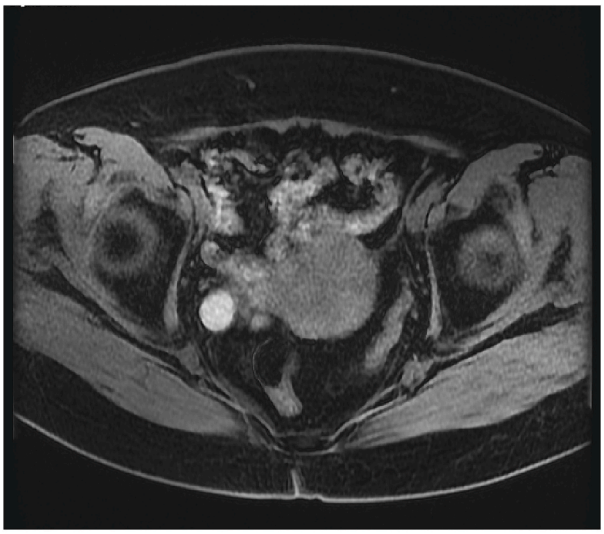


Fig. 2. CT at the first visit.



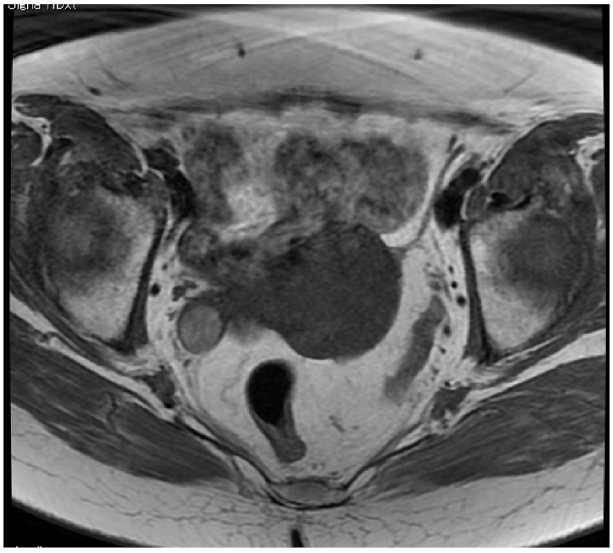


Fig. 3. Pelvic MRI

right ovarian nodule. A 20 mm mono-cystic lesion was found on the right side of the pelvis, with T1-weighted images showing high signal, T2-weighted images showing slightly lower signal, similar to muscle, and diffusion-weighted image showing high signal. A cyst of ovarian thyroidoma and endometriosis was suspected. In addition, multiple low-intensity nodules and tumors with clear boundaries on T2-weighted images of the myometrium were considered uterine fibroids.

A right chest cavity drain (20Fr) was therefore placed on the day of admission, with 1200 ml of pleural effusion drained between the day of admission and the following day, and then spontaneous draining performed with a water seal. From the 3rd to the 10th day of hospitalization, about 180 ml/day of pleural effusion was collected.

Thoracoscopy under local anesthesia (Fig. 4) was performed on the 4th day of hospitalization to search for a potential cause. A slight redness and partial fat-like yellow ridge was found on the pleural surface, but no evidence of ectopic endometriosis was found, nor was there any evidence of malignant tumor metastasis. No apparent small holes were found on the diaphragm surface. Biopsy was performed randomly, and pathological examination showed hyperplasia of pleural tissue and mesothelial cells with mild lymphocytic infiltration, but no atypia. Immunostaining revealed PgR-negative, ER-negative, and CD10-negative nonspecific pleurisy.

MS was strongly suspected at the time of admission, and laparoscopic

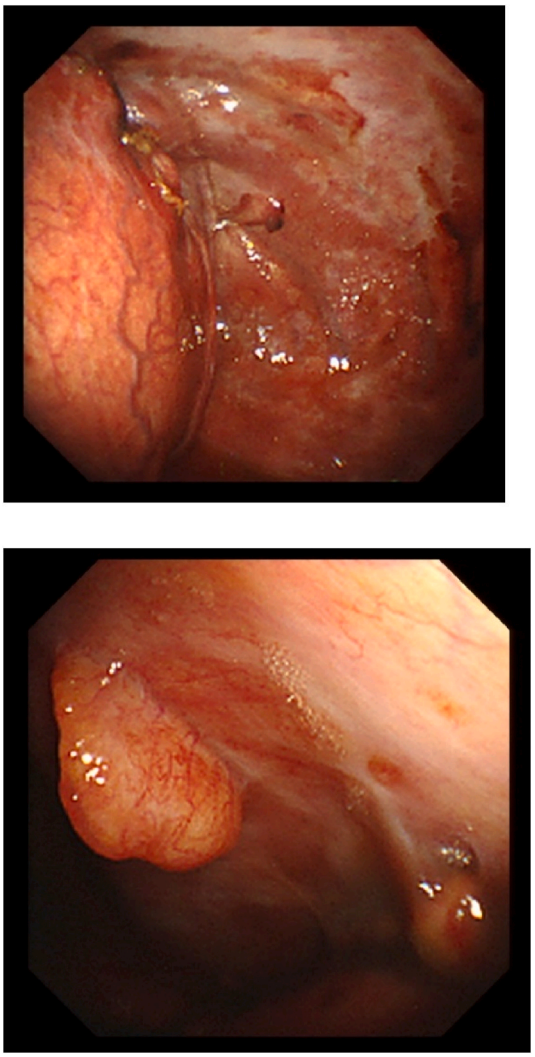


Fig. 4. Thoracoscopy under local anesthesia.

adnexectomy was performed on the right ovarian tumor on the 11th day of admission, with no significant ascites observed during the operation. A cyst was present in the resected right ovary, and histologically, there was a cluster of histiocytes that had phagocytosed hemosiderin on the luminal surface of the cyst wall. The epithelium was completely shed, but an ectopic endometrium was partially observed adjacent to the cyst. Endometrial cyst with endometriosis was diagnosed, consistent with the chocolate cyst due to endometriosis. Although the drain was removed 4 days after surgery, the pleural effusion had resolved on CT examination on the 12th day after surgery, and no re-collection was observed. Based on the above, the patient was diagnosed with incomplete PMS secondary to right endometriotic ovarian cyst.

3. Discussion

Here we report a case of incomplete pseudo-Meigs syndrome, thought to be caused by an endometriotic cyst. To the best of our knowledge, there are no previously reported cases secondary to an endometriotic cyst.

In 1937, Meigs et al. reported 7 cases of pleural effusion and ascites associated with ovarian fibroma that disappeared after tumor removal [1]. These cases were defined as follows: (1) primary tumors are fibroma, thecoma, granulosa cell tumor, Brenner tumor, etc.; (2) with ascites; (3) with pleural effusion; and (4) pleural and ascitic fluid disappears with tumor removal, and does not re-collect. Cases that satisfy all 4 criteria are defined as true MS, but PMS if associated with tumors other than outlined in (1), but satisfying the other criteria [2]. Furthermore, those who present with symptoms of systemic lupus erythematosus and high CA-125 in pleural effusion and ascites are called pseudo-pseudo Meigs' syndrome (PPMS), also known as Tjalma syndrome [4]. Since this case was negative for antinuclear antibody and PPMS was excluded, the patient was diagnosed as incomplete PMS derived from an endometriotic cyst. Although various reports have presented cases with PMS, no reports of cases derived from endometriotic cysts could be found by searching Google Scholar/PubMed.

Endometriosis affects approximately 10% of women of reproductive age. Since it is estrogen-dependent, it resolves spontaneously after menopause, but it may develop or recur. The exact prevalence of endometriosis in menopausal women is unknown, but is estimated to be approximately 2–5%. In an epidemiological survey of about 40,000 endometriosis patients, the proportion of postmenopausal women was 2.6%, including 9 elderly people aged 80 and over [5]. As far as we know from the medical records at the time of the gynecological examination, it seems that there were no particular subjective symptoms.

Riley et al. summarized pleural effusions derived from gynecological diseases [6], and found multiple pleural effusions caused by gynecological diseases other than MS. Different diagnoses included menstrual pneumothorax/thoracic endometriosis syndrome (TES) and ovarian hyperstimulation syndrome (OHSS). Joseph et al. reported 110 cases of TES, with an average age of 35 ± 0.6 years (15–54 years) [7], and the majority younger than this present case. Pleural effusion findings, endoscopic findings on thoracoscopy under local anesthesia, and pleural biopsy specimens showed no endometrial lesions including immunostaining. Pleural effusion due to TES is unlikely because there was no history of pneumothorax or pleural effusion. In addition, OHSS is generally preceded by ascites and may be accompanied by pleural effusion in severe cases, but this case did not present with ascites. In addition, ovulation induction is a major risk factor, and the history differs from this case.

Krenke used a database to search for MS-related items from 1940 to 2013, extracted only those that could be examined, and reported pleural effusion status [8]. There were various MS subtypes depending on the tumor and storage pattern associated with MS, and those with only ascites or pleural effusion were defined as incomplete PMS. Pleural effusion of PMS is often right unilateral, followed by bilateral. Pleural effusion was exudative in 70–80%, and this case also had pleural effusion. In this case, the number of pleural effusion RBCs was high, but this was not described in the previous report. In general, there were no reports of bloody pleural effusion due to Meigs's syndrome-related disease, and this case also had yellow opaque pleural effusion macroscopically. Thoracoscopic endoscopic findings revealed pleurisy, suggesting that bleeding was enhanced during thoracentesis. Incomplete PMS with only pleural effusion was rare, with 23 cases (18 benign tumors, 5 malignant tumors) out of a total of 447 cases including MS/PMS/PPMS, or 5.1% of the total. Of these, only 4.0% occurred when limited to benign tumors, as in this case.

Several hypotheses have been suggested for the production of ascites in MS, such as mechanical stimulation of the peritoneum around pelvic tumors, congestion of veins and lymph vessels associated with the tumor itself, and production of various interleukins [1,9]. Pleural effusion is thought to occur secondarily via congenital defects or diaphragmatic lymphatic vessels that tend to be common on the right side allowing ascitic fluid to pass through the diaphragm into the pleural cavity [10].

Miyoshi et al. attempted to confirm diaphragmatic trafficking using contrast-enhanced ultrasound with the second-generation contrast agent, perflubutane (Sonazoid®). They reported the results of 17 cases of hepatic pleural effusion [11], with 11 cases (64.7%) where a jet of inflow of contrast medium was observed, and 5 cases with punctate migration. It is speculated that ascites also migrated into the thoracic cavity due to the same mechanism in this case. In addition, Agaranoff et al. reported two cases of incomplete PMS with only pleural effusion.

The possible mechanism was the presence of a small amount of ascites that was difficult to identify on imaging. It is speculated that the ascites disappeared completely due to the continuity [12].

Thoracoscopic observations of MS/PMS cases such as this case revealed pleurisy, and Poumon et al. reported findings of pleural edema and capillary hyperplasia [13]. Cho et al. also reported a case with nodules on the diaphragm, but concluded that pathologically it was a reactive proliferation of mesothelial cells and pleural tissue associated with lymphatic hyperplasia on the diaphragm surface [14].

Previously reported nonspecific pleurisy may explain the fever seen in this case. In addition, some of the mechanisms that cause ascites have been reported to be derived from cytokines produced by tumors that can also cause fever.

Although thoracoscopy only revealed nonspecific pleurisy, if MS/PMS is suspected by diagnostic imaging, thoracoscopy under local anesthesia can provide a lot of information, including pathological findings, which may be useful in excluding malignant disease. Even if PET-CT is performed, it may be difficult to judge whether it is benign or malignant due to the above-mentioned inflammation. Although useful because of strong suspicion of malignancy in the presence of overt distant metastases, standard treatment for ovarian cancer is surgical staging and volume reduction. If a Meigs-related disease is suspected, it is of little significance to perform PET-CT preoperatively, and if it is ovarian cancer, postoperative imaging may be acceptable.

In conclusion, we present a case of incomplete pseudo-Meigs' syndrome that was thought to have been caused by an endometrial ovarian cyst. If pleural effusion with a pelvic tumor is found by diagnostic imaging and MS or PMS is strongly suspected, surgery may be prioritized if there are no findings suggestive of malignant disease after thoracoscopy.

Ethics approval and consent to participate

Not applicable.

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Authors' contributions

TK contributed to decision of treatment, collecting clinical data, data analysis, and writing the manuscript. TU, YF, TS, WY, and JN contributed to discussions about the patient. All authors read and approved the final manuscript.

Consent for publication

Written informed consent for publication of clinical details and clinical images was obtained from the patient.

Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analyzed.

Declaration of competing interest

The authors declare no conflicts of interest associated with this manuscript.

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