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MEIGS SYNDROME MIMICKING MALIGNANCY: A DIAGNOSIS CHALLENGE

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ABSTRACT

Background: Meigs syndrome is a benign ovarian mass associated with pleural effusion and ascites. The ovarian tumor is typically a fibroma, the most common benign pure stromal tumor, with 10-15% of cases presenting with pleural effusion or ascites. **Case Presentation**: Here, we present a Meigs syndrome in a 57-year-old woman. This patient complained of an abdominal mass and massive pleural effusion. Multiple uterine myomas and left ovarian solid tumor were found during laparotomy. Following the removal of ovarian mass, the patient's condition improved, and she was discharged a week after being hospitalized. **Conclusion:** The clinical symptoms of Meigs syndrome are similar to those of malignant disease. However, with optimal treatment, the outcome of Meigs's syndrome is generally satisfactory.

BACKGROUND

Meigs syndrome was first described by Joe Vincent Meigs in 1937 as a triad of a benign ovarian fibroma, ascites and pleural effusion.[1] This syndrome is an uncommon disease characterized by the resolution of both ascites and hydrothorax following tumour removal. Meigs syndrome is occasionally associated with a fibroma-like Brenner or granulosa cell tumor. [1] It is termed as pseudo-Meigs syndrome when it is associated with any other type of ovarian tumor, including mature teratoma, struma ovarii, metastatic ovarian tumor, or leiomyoma. [2]

The diagnosis of Meigs syndrome is challenging due to symptoms that mimic those of disseminated malignant tumour. We are reporting a rare case of Meigs syndrome in a 57-year-old woman who initially presented with massive pleural effusion, initial malignant adenocarcinoma cytology and suspected mesenterial tumour.

CASE PRESENTATION

A 57-year-old female was referred to Universitas Gadjah Mada Academic Hospital with an abdominal mass and right pleural effusion. She reported abdominal enlargement and breathing difficulty over the last four months prior to hospital admission. She denied experiencing rapid weight loss, nausea or vomiting, loss of appetite, or problem in micturition and defecation. There was no history of malignancy, metabolic disease, cardiac disease or hypertension. At a district hospital, physical examination revealed decreased vesicular sound on the right side of the lung and a solid mass palpable on the left upper quadrant of abdomen. Computed tomography (CT) scan showed an intraperitoneal mass in the right abdomen, with differential diagnosis including a mesenteric mass, massive right-sided pleural effusion and ascites. She was then managed by a digestive surgeon and pulmonologist. A pleural puncture was performed and 1000 ml of fluid was evacuated. The level of CEA and AFP were normal, measuring 0.73 ng/mL and 1.13 ng/mL, respectively.

Five days after the first thoracocentesis, she complained of shortness of breath and thorax x-ray showed a massive right-sided pleural effusion (Figure 1A). Approximately 1500 ml of fluid was evacuated from the right pleura. Pleural fluid cytology showed medium to large-sized polymorphic cells cell clusters with papillary, ball formation, acinar patterns. The nuclei were round and oval, with irregular and hyperchromatic nuclear membranes. These cytology results indicate metastatic adenocarcinoma.

The following day, thoracostomy (water sealed drainage (WSD) insertion) was performed, and 1500

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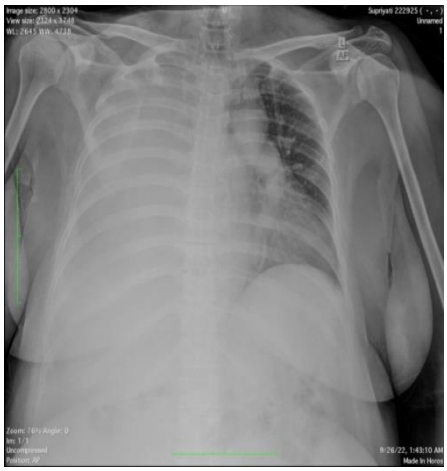
cc of pleural fluid was evacuated. After this procedure, the patient underwent laparotomy to established a definitive diagnosis and to plan further treatment. Approximately 2450 ml of ascites was drained and a mass covered by omentum was found. Sharp adhesiolysis revealed that the mass originated from the ovary. An intraoperative consultation with an obstetrician and gynaecologist was conducted. During pelvic exploration, multiple uterine myomas were found, the right fallopian tube and ovary were normal, and the left ovary was enlarged, measuring 25 x 20 x 15 cm. Myomectomies and left salpingooophorectomy were performed (Figure 1B), and an abdominal drain was also placed.



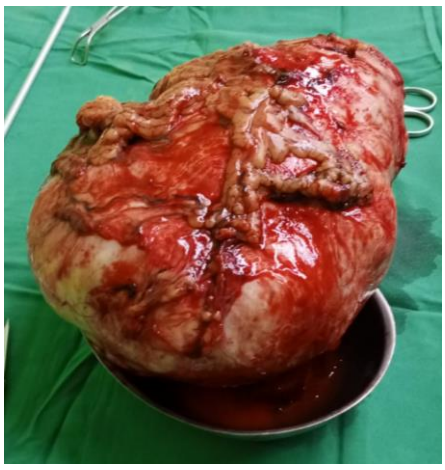


Figure 1. A) Thorax X Ray on admission. B) Left ovary was enlarged and left salphingoophorectomy was performed.

After the surgery, the patient was managed in the intensive care unit (ICU). On the first day, the WSD output was 980 ml of serohemorrhagic fluid, while the abdominal drain output was 580 ml of serohemorrhagic fluid. These fluid outputs decreased on second day to 350 ml and 300 ml, respectively. Cytology of the pleural fluid was repeated and showed several groups of monomorphic mesothelial cells, few lymphocytes and macrophages scattered against a uniform background of erythrocytes, with no malignant cells detected. The patient was discharged from the ICU on the second day. The subsequent histopathological study revealed ovarian tissue with corpus albicans and tumour cells arranged in storiform pattern. The cells were of medium size, with abundant cytoplasm, oval or spindle-shaped nuclei with pointed ends, regular membranes and fine chromatin. Connective tissue stroma underwent hyalinization and hyalin plaque were observed. The conclusion of the histology examination was ovarian fibroma while the uterine tissue was confirmed as uterine myomas. On day five, both the WSD tube and abdominal drain were removed. Following this, a thoracic X-Ray evaluation showed minimal pleural effusion. The patient was discharged after a week of admission with significantly improved clinical condition.

DISCUSSION

Meigs syndrome is a condition characterized by pleural effusion and ascites associated with a benign ovarian tumour. The ascites and the pleural effusion usually resolve after the removal of the ovarian tumour. The majority of ovarian tumours seen in Meigs syndrome is ovarian fibroma. [3] An ovarian fibroma is a sex cord-stromal ovarian tumour that commonly occurs in postmenopausal women, with a mean age of approximately 50 years. The incidence of ovarian fibroma is about 4% of all ovarian tumours. Meigs syndrome is rare, occurring in only 1% to 2% of cases of ovarian fibromas. [4]

The diagnosis of Meigs syndrome is challenging because it mimics abdominal malignancies and other conditions, including ovarian cancer, uterine tumour, gastrointestinal cancer, lung cancer, liver cirrhosis, abdominal tuberculosis, congestive heart failure, and nephrotic syndrome. [5–7] Suspicion of Meigs syndrome can be raised by the presenting clinical symptoms such as dyspnea,

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abdominal enlargement, discomfort and lower-extremity edema.[5] During physical examination, shifting dullness as a sign of ascites and a solid mass may be palpated in the abdomen. Dullness to percussion of the lungs, decreased breath sounds, and a pleural rub during auscultation may also be found. [7] In our case, the patient complained of breathing difficulty and an abdominal mass on palpation. The production of peritoneal fluid in Meigs syndrome is proposed to result from irritation of the peritoneal surfaces by a solid ovarian tumour. Direct pressure on surrounding lymphatics or vessels, hormonal stimulation, or release of inflammatory mediators from the tumour which cause increased capillary permeability are another proposed mechanism. [8] The fluid then enters the unilateral or bilateral pleural cavity through the lymphatic vessels or through a communication between the pleural and the abdominal cavities.

The role of tumor biomarkers in Meigs syndrome is debatable. Elevated levels of CA125 have been reported in several cases of Meigs syndrome. [9,10] Immunohistochemical studies suggest that the increased CA125 in Meigs syndrome is secondary to peritoneal inflammation and mesothelial expression of the antigen. [6] In our case, CA125 was not evaluated because the tumour appeared solid instead of cystic and was suspected to be of gastrointestinal origin. Therefore, CEA and AFP were measured and were within normal limits. [11]

Imaging assessment is recommended for a more precise characterization and to clarify the etiology of the triad in Meigs syndrome. A thoracic X-ray and CT may be used to confirm the presence of a pleural effusion and to exclude any pulmonary malignancy. [12] The possibility of a malignant pleural effusion should be considered in all patients with undiagnosed exudative pleural effusions. The simplest way to establish the diagnosis of a malignant pleural effusion is through pleural fluid cytology. Detailed analyses of pleural effusion can provide valuable information about its properties and etiologies. In our case, the first pleural cytology suggested a malignant origin, whereas subsequent analysis showed no malignant cells. Thus, repeated cytological examination is recommended in the diagnosis of Meigs Syndrome. [13]

Although ultrasound is a readily accessible and cost-effective modality for diagnosing abdominal and pelvic masses, it [may] still be inadequate for a proper diagnosis. In particular, the ultrasound appearance of fibromas is nonspecific. Ovarian fibromas typically appear as solid, hypoechoic masses with attenuation of the ultrasound beam, without increased vascularity. [12,14] To distinguish between benign and malignant tumours of the ovary using ultrasound, the IOTA (International Ovarian Tumour Analysis) simple rules can be applied. [15] Magnetic resonance imaging (MRI) and CT scan of the abdomen and thorax are recommended for a suspicious pelvic mass to exclude metastases and lymphadenopathy. [7] Ovarian fibroma usually display low intensity on T1-weighted images, and marked hypo-intensity on T2-weighted images; they also show heterogeneous and mild-to-moderate enhancement with contrast medium. [16] Despite these imaging techniques, only histological examination of the excised tumour can confirm the diagnosis of Meigs syndrome. The CT scan of our patient showed an intraabdominal mass with a differential diagnosis of a mesenteric mass. During the surgery, the mass was covered by omentum, which matched the suspicion of a mesenteric mass on the CT scan. However, the mass originated from the left ovary and the pathological examination confirmed it as an ovarian fibroma.

Because of the rare occurrence of Meigs syndrome, it is [often] subject to misdiagnosis or delayed treatment. Patients frequently seek treatment in the emergency department due to symptoms caused by hydrothorax or ascites. The patient in this case was initially managed by a pulmonologist and a digestive surgeon. Although the clinical manifestations of Meigs syndrome are similar to those of malignant tumors, it is a benign condition, and surgical resection is the main treatment. The prognosis of this disease is usually favourable. Following the resection of the tumour, symptoms typically resolve gradually, and the rate of relapse is very low.

CONCLUSION

The clinical presentation of postmenopausal women with a solid abdominal mass, ascites, and pleural effusion is highly suspicious for a malignant tumour. However, Meigs syndrome should be considered, as it can present with the similar

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symptoms, and the prognosis of this disease is usually satisfactory after removal of the ovarian tumor.

ETHICAL APPROVAL

There is no ethical approval.

CONFLICTS OF INTEREST

The authors declare no conflict of interest.

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AUTHOR CONTRIBUTIONS

Conceptualization, analysis, data curation, writing—original draft preparation, RW; resources, investigation, writing—review and editing, supervision, YT.

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