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CASE REPORT

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Recurrent pleural effusion from ovarian hemangioma: A rare pseudo-Meigs syndrome presentation

Sina Neshat[1] | **Alejandra Yu Lee-Mateus**[1] | **Isabel Fernandez-Bussy**[2] | **Katherine L. Walsh**[1] | **David Abia-Trujillo**[1]

[1]Division of Pulmonary, Allergy, and Sleep Medicine, Mayo Clinic, Jacksonville, Florida, USA

[2]Facultad de Ciencias Médicas, Pontificia Universidad Católica Argentina, Buenos Aires, Argentina

Correspondence
Alejandra Yu Lee-Mateus, Division of Pulmonary, Allergy, and Sleep Medicine, Mayo Clinic, 4500 San Pablo Road, Jacksonville, FL 32224, USA.
Email: yuleemateus.alejandra@mayo.edu

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Abstract

Pleural effusion is a common condition related to various diseases such as heart failure, malignancies, and pneumonia. Ovarian hemangioma is a rare type of female genital tumour and can rarely cause pleural effusion. In this case, we present a 48-year-old female with repeated episodes of recurrent right-sided pleural effusion over 1 year with no clear aetiology. Abdominal computed tomography revealed a large left ovarian mass. After surgical removal of the mass, the repeated pleural effusion episodes ceased, and histopathology analysis reported a rare ovarian hemangioma. Pseudo Meigs' syndrome is a triad of an ovarian tumour, ascites, and hydrothorax that rarely presents with ovarian hemangioma; both effusions are eradicated after removing the tumour.

KEYWORDS

adnexal tumour, pleural effusion, pseudo Meigs' syndrome, thoracocentesis

INTRODUCTION

Pleural effusion is a common condition that refers to the pathological accumulation of fluid in the pleural space. The most common causes of pleural effusion are congestive heart failure, malignancies, pneumonia, and pulmonary embolism.[1]

Ovarian hemangiomas are a rare type of benign vascular tumour of the female genital tract. They usually do not present any symptoms and are diagnosed incidentally during surgery. Ovarian hemangiomas rarely accompany systemic manifestations like pleural effusions.[2] Reported cases of ovarian hemangioma do not exceed 60.[3]

We will introduce a patient with an ovarian hemangioma that presented with recurrent right-sided pleural effusion as the primary symptom and was diagnosed as pseudo-Meigs syndrome.

CASE REPORT

A 48-year-old woman presented to the emergency department in mid-2021 due to shortness of breath and chest pain. She described it as a sharp right-sided constant pain that improved when holding her breath, did not relate to activity, and started 48 h before admission. She had a similar condition in early 2020, which resolved without specific treatment. She also reported abnormal, heavy menstrual bleeding that started 1 year ago, followed by her gynaecologist. Previous endometrial biopsy and pelvic ultrasound were unremarkable, and she was on oral contraceptives. She did not report significant family history of disease or illnesses.

A chest x-ray showed a new moderate-sized right pleural effusion and a stable small left pleural effusion (Figure 1A). A chest computed tomography (CT) scan with contrast confirmed the chest x-ray findings but showed new bilateral pulmonary emboli, more prominent on the left side. One litre of clear yellow fluid was removed by thoracentesis. The patient was discharged home the next day with a diagnosis of pulmonary embolism with no evidence of deep vein thrombosis of the lower extremities. While admitted, she was also consulted by the gynaecology service for her irregular and heavy menses, who recommended an intrauterine device (IUD) to control her abnormal uterine bleeding symptoms. The IUD was inserted 2 months after discharge with no complications.

Six months later, an elevated d-dimer was found on her routine testing despite being on anticoagulation drugs.

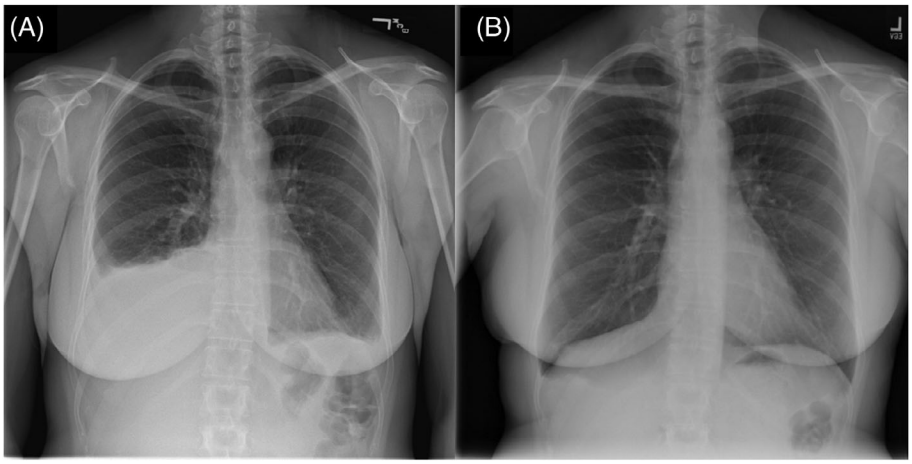

FIGURE 1 (A) Chest X-ray showing a right-sided pleural effusion on initial admission and (B) Chest X-ray showing no evidence of recurrent pleural effusions at 1-month follow-up after removal of ovarian hemangioma

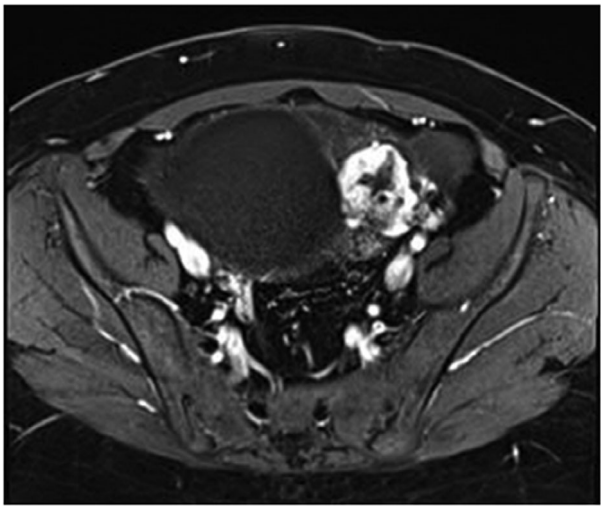

FIGURE 2 Magnetic resonance imaging of a solid, irregular diffusion restricting heterogeneously enhancing mass in the left adnexa, which measured 4.0 × 5.6 × 4.1 cm

Additionally, a new large right pleural effusion and a small amount of upper abdominal ascites, with no evidence of new pulmonary embolus, were seen on chest CT. One and a quarter litre of clear yellow fluid was removed by repeated thoracentesis.

Five months later, she presented recurrent pleural effusion and 850 mls of yellow fluid was removed by thoracentesis from the right pleural space. The fluid sample cytology analysis showed no evidence of malignancy, and cultures were negative for bacterial, fungal, or viral infection. A positron emission tomography scan was performed reporting a large pelvic cystic mass involving the solid components that appeared to originate from the left ovary. Trace perihepatic ascites and moderate right pleural effusion were seen. Subsequently, a magnetic resonance imaging of the pelvis confirmed the presence of the ovarian mass (Figure 2) and laparoscopic surgical removal was performed. Diagnosis was an adnexal hemangioma after gross pathological examinations.

At one-month follow-up, the patient was asymptomatic and well-recovered at her multidisciplinary visits. Her most recent chest x-ray showed no evidence of pleural effusions (Figure 1B).

DISCUSSION

In this case, we faced unexplained recurrent pleural effusions over 1 year, with intervals between the recurrences shortening: an initial relapse time of about 6 months, and later reduced to a 1-month gap to the last recurrence.

Ovarian hemangioma is a rare adnexal tumour. It has a high blood vascular supply and is mainly seen incidentally during surgery or imaging studies. It can sometimes cause systematic manifestations as well.[2] In this case, the left ovarian hemangioma was the principal cause of the recurrent pleural effusions. After the removal of the mass, pleural effusions ceased, and the patient was asymptomatic in her subsequent visits.

Meigs' Syndrome comprises concomitant ovarian fibromas, ascites, and hydrothorax. Conversely, pseudo-Meigs' Syndrome is a similar syndrome, except the tumour involved is different from fibroma.[4] The most common causes of Pseudo-Meigs' syndrome are stromal tumours, teratomas, cystadenomas, uterine leiomyomas, ovarian adenocarcinoma, endometroid carcinoma, and secondary metastatic ovarian tumours. Ovarian hemangiomas are not usually considered a common cause of this syndrome. Most importantly, our patient presented solely with recurrent pleural effusions, in contrast with most of patients with Pseudo-Meigs' syndrome who present with abdominal symptoms.[4] The classic triad of this syndrome also includes elevated CA-125, which our patient also presented and which subsequently declined after removal of the mass.

The cause of ascites and pleural effusion in Meigs' and Pseudo-Meigs' syndromes remains poorly understood. It has been hypothesized that the hydrothorax originates from migration of excessive ascites fluid into the pleural cavity through specific lymphatic channels in the diaphragm. Supporting this theory is the fact that most pleural effusions are almost entirely right-sided, even when the abdominal mass is left-sided, as in our patient. This is similar to hydrothorax from peritoneal dialysis where the movement of the dialysate into the pleural space is right-sided and can be considered as a 'porous diaphragm syndrome'. Yet, pleural effusion can occur in the absence of obvious ascites, as in our case, and vice versa, massive serous ascites does not necessarily translate into pleural effusion.[5]

Pseudo-Meigs' syndrome is an uncommon cause of recurrent pleural effusion, and even rarer from ovarian hemangioma and should be considered in the face unexplained effusion with no other systemic symptoms.

AUTHOR CONTRIBUTIONS

Sina Neshat was responsible for writing the main draft and patient data collection. Alejandra Yu Lee-Mateus revised the manuscript. Isabel Fernandez-Bussy was involved in manuscript writing. Katherine L. Walsh was involved in patient data collection. David Abia-Trujillo was responsible for manuscript review and scientific checking.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

ORCID

Sina Neshat https://orcid.org/0000-0001-6522-1865

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