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Anticoagulation therapy for a rare case of pseudo-Meigs' syndrome complicated by massive ascites in the postpartum period

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Abstract

Most persistent symptoms of pseudo-Meigs' syndrome (PMS) are alleviated by surgical tumor removal. The present case report suggests that PMS may present with ascites and hypercoagulation and that emergency anticoagulation can improve the patient's condition. We herein describe a postpartum woman with an acute presentation including abdominal pain, ascites, postpartum hemorrhage, and degeneration of a large uterine fibroid. Initial evaluation revealed unexpected massive ascites, pleural effusion, a highly elevated D-dimer level, and a moderately elevated CA125 level. Following anticoagulation therapy, the ascites, abdominal pain, and pleural effusion resolved. There was no recurrence of these symptoms during follow-up, although the large degenerating uterine fibroid and mildly elevated serum CA125 level persisted. Postoperatively, pathological analysis confirmed leiomyoma, the patient's CA125 level returned to normal, and the ascites resolved, meeting the diagnostic criteria for PMS. Further studies are needed to determine whether a hypercoagulable state is common in pregnant patients with PMS and to develop strategies to improve outcomes.

Keywords

Anticoagulation, pseudo-Meigs' syndrome, ascites, postpartum, leiomyoma, high D-dimer level

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Introduction

Meigs' syndrome refers to the presence of pleural effusion and ascites associated with a benign ovarian fibroma. Other benign or malignant ovarian tumors, as well as uterine fibroids, can also be associated with pleural effusion and ascites; this condition is termed pseudo-Meigs' syndrome (PMS).[1] Treatment of PMS is mainly symptomatic and surgical. Symptomatic treatment includes thoracentesis and drainage of ascites to relieve symptoms, along with supportive treatment to improve hypoproteinemia.[2] However, most persistent PMS symptoms require surgical removal of the tumor. Pleural effusion and ascites typically resolve within a few weeks postoperatively, usually without recurrence.[1] We herein present a case of PMS that occurred during the postpartum period. The condition was characterized by abundant ascites and a highly elevated D-dimer level. Our report highlights the clinical characteristics and treatment response in this case. This case offers valuable insights into the individualized treatment of complications related to uterine fibroids during pregnancy.

Case presentation

A 35-year-old woman (gravida 1, para 1, abortion 0) was referred to the obstetric service 15 hours postpartum because of a large amount of ascites, abdominal pain, elevated D-dimer and CA125 levels, and the presence of a uterine myoma. The patient's pregnancy had been unremarkable except for the uterine myoma, which had been noted early in the pregnancy and showed a slight increase in size (up to 6 cm in diameter) later in the pregnancy. This myoma did not affect the course of the pregnancy.

Fifteen hours after delivery, the patient developed sudden abdominal pain following an intramuscular injection of 250 μg Hinmupei for postpartum bleeding. Physical examination revealed fluid waves and shifting dullness, and transabdominal ultrasound confirmed massive ascites and liquefaction of the myoma. A contrast-enhanced computed tomography scan of the chest and abdomen showed no thrombosis in the blood vessels but revealed a large amount of ascites, a small pleural effusion, intestinal wall edema (Figure 1), and a myoma in the posterior uterine wall. The myoma exhibited cystic degeneration at its center (Figure 1), and the ovaries appeared normal.

Because of the large volume of ascites, ultrasound-guided diagnostic drainage was performed and yielded 50 mL of yellowish fluid. The ascitic fluid analysis was consistent with exudate. The Rivalta test was positive, whereas culture and cytology of the ascitic fluid were negative. The albumin level in the ascitic fluid was 4.2 g/L (serum albumin, 20.8 g/L; serum–ascites albumin gradient, 16.6).

Laboratory findings included an elevated D-dimer level of 48,390 μg/L (reference range, <550 μg/L), normal prothrombin time of 11.0 s (control, 11.7 s), normal activated partial thromboplastin time of 29.5 s (reference range, 20–39.7 s), normal thrombin time of 17.6 s (reference range, 14.7–20.7 s), normal fibrinogen level of 362 mg/dL (reference range, 200–400 mg/dL), elevated CA125 level of 131.99 U/mL (reference range, <35 U/mL), elevated alpha-fetoprotein level of 74.98 U/mL (reference range, <9 U/mL), elevated lactate dehydrogenase level of 316 U/L (reference range, 109–245 U/L), and decreased serum albumin level of 20.9 g/L (reference range, 40.0–55.0 g/L) (Table 1).

A multidisciplinary team was consulted to manage anticoagulation (administered for 1 week) and antibiotic therapy (administered for 2 days). These treatments alleviated the abdominal pain, and the ascites and pleural effusion decreased by day 4.

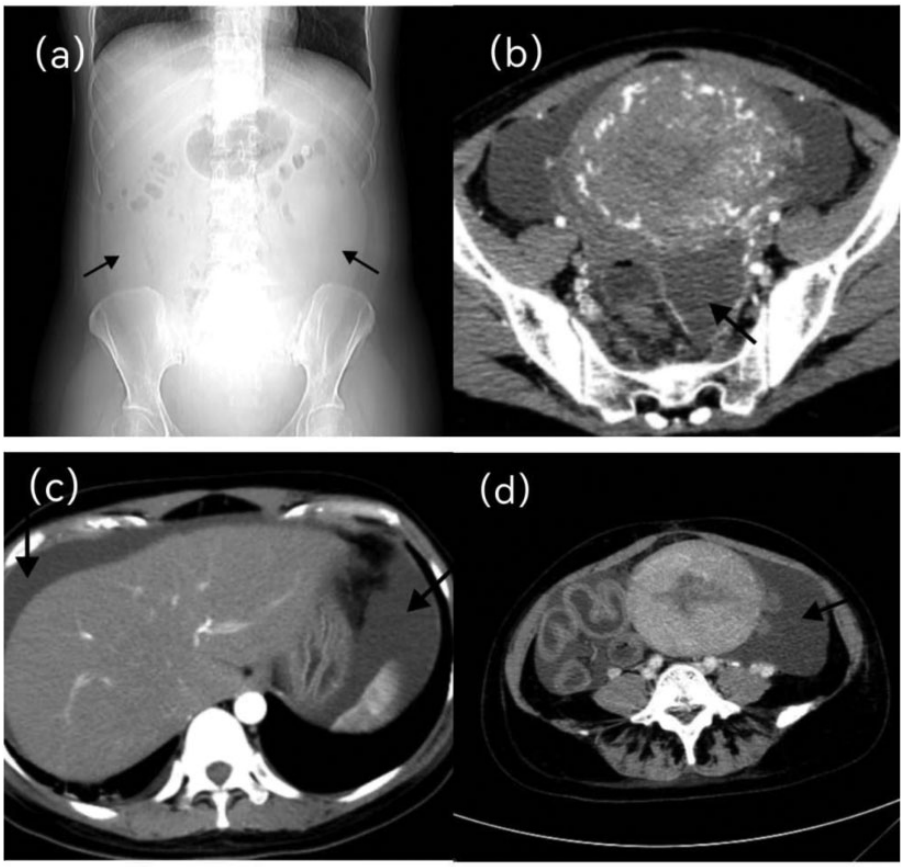

Figure 1. Abdominal contrast-enhanced computed tomography scan demonstrating a large amount of ascites and a solid tumor mass. (a) Large amount of abdominal effusion. (b) Solid tumor mass in the posterior wall of the uterus with cystic degeneration. (c) Abundant ascites in the hepatorenal recess and splenorenal recess and (d) abundant ascites in the pelvic cavity.

The serum albumin level remained unchanged from the pretreatment value, while the D-dimer level decreased. The patient was discharged in stable condition after 7 days.

Follow-up included pelvic ultrasound and evaluation of the CA125 level. Although ultrasound showed no increase in ascites, the CA125 level remained elevated (112.4 U/mL). Six months later, exploratory laparoscopy revealed a 7- × 5- × 6-cm myoma with normal-appearing ovaries. Histological examination confirmed a benign leiomyoma with necrosis. The resolution of ascites and pleural effusion, along with these pathological findings, confirmed the diagnosis of PMS.

Discussion

This patient was a postpartum woman with an acute presentation characterized by abdominal pain, ascites, postpartum hemorrhage, and degeneration of a large uterine fibroid. Auxiliary tests revealed an elevated serum CA125 level and a hypercoagulable state. Diagnostic abdominal paracentesis ruled out the presence of cancer cells or

Table 1. Summary of the clinical investigations in the present case.

	Later in pregnancy	Before treatment of ascites
CA125 (U/mL)	28.6	131.99
Albumin (g/L)	31.6	20.8
Urinalysis	No protein	0.24 g/24 h (normal)
N-terminal pro-B-type natriuretic peptide (pg/mL)	Normal	560.8
AST, ALT	Normal	Normal
Echocardiogram	–	Normal cardiac structure and function
D-dimer (μg/L)	1950	48,390
LDH (U/L)	257	529
HBsAg	Negative	Negative
HGB (g/dL)	14.3 (normal)	12.8 (normal)
PT (seconds)	11.2 (normal control: 11.7)	11.0 (normal control: 11.7)
APTT (seconds)	28.0	29.5
TT (seconds)	17.7	17.6
Fbg (mg/dL)	321	362

AST, aspartate aminotransferase; ALT, alanine aminotransferase; LDH, lactate dehydrogenase; HBsAG, hepatitis B surface antigen; HGB, hemoglobin; PT, prothrombin time; APTT, activated partial thromboplastin time; TT, thrombin time; Fbg, fibrinogen.

hemoperitoneum due to uterine rupture, indicating ascites with a high serum–ascites albumin gradient. The ascites could not be attributed to heart failure, nephropathy, or liver insufficiency, nor was it consistent with systemic lupus erythematosus (SLE), which typically presents with polyserositis and multi-system involvement. Non-cirrhotic portal hypertension was considered as a potential cause, requiring the exclusion of intra-abdominal visceral venous thrombosis, a rare condition. Early diagnosis was challenging because common causes vary widely, including coagulation disorders, autoimmune diseases, myeloproliferative disorders, infectious diseases, and medication effects.

Imaging studies showed no thrombosis in the portal vein, hepatic veins, or inferior vena cava, and the liver size did not suggest hepatic venous thrombosis. With input from a multidisciplinary team, the attending physician focused on uterine fibroid degeneration and the hypercoagulable state, initiating anticoagulant and anti-inflammatory treatments. The patient was discharged after the ascites resolved. During a 6-month follow-up, the ascites did not recur, although the serum CA125 level remained mildly elevated and the uterine fibroid continued to degenerate. Malignancy was initially considered, but postoperative pathology confirmed a benign uterine fibroid with ascites as a rare complication. Postoperatively, the patient's CA125 level normalized and the ascites resolved, fulfilling the diagnostic criteria for PMS.

CA125 is the most commonly used marker for epithelial ovarian malignancies. In this case, the association of a uterine fibroid with an elevated serum CA125 level is clinically rare and can easily be misdiagnosed as a malignant tumor. Literature reports indicate that Meigs' syndrome can be accompanied by an elevated CA125 level, with the amount of ascites positively correlating with the CA125 level. However,

the CA125 level alone cannot determine whether a tumor is benign or malignant.[3] The exact mechanism by which PMS leads to elevated CA125 is unclear, but it may be related to increased abdominal pressure caused by tumor growth and CA125 expression in mesothelial cells.[4] Some researchers speculate that this increase is due to inflammation on the surfaces of the pleura and peritoneum caused by the presence of free fluid. Local irritation of the pleura and peritoneum by pleural effusion and ascites can result in a moderately elevated CA125 level.[5,6]

It is important to note that an elevated serum CA125 level can be associated not only with tumors but also with non-tumorous conditions such as tuberculosis, nephrotic syndrome, connective tissue diseases, and others. These conditions are also common causes of serous cavity effusion, and ascites itself may lead to mild elevations in serum CA125. In patients with SLE, pleural effusion, ascites, and elevated CA125 can occur independently of ovarian tumors, a condition referred to as pseudo-PMS or Tjalma syndrome.[7] Some literature has proposed stricter diagnostic criteria for this syndrome, which include the gradual onset of painless polyserositis in patients with SLE and an elevated CA125 level.[8] The inflammatory response in SLE can lead to plasma cell aggregation, and the deposition of immune complexes on the peritoneum can cause local inflammation or peritoneal vasculitis, leading to ascites.[9]

In cases of benign PMS, the pleural effusion and ascites typically resolve spontaneously after the lesion is removed, and the CA125 level decreases, indicating a good prognosis. Treatment for pseudo-PMS focuses on addressing the underlying SLE.[10] In our case, the CA125 level returned to normal, and the ascites disappeared after surgery, meeting the diagnostic criteria for Meigs' syndrome. However, for patients presenting with pleural effusion, ascites, and elevated serum CA125, it is crucial to differentiate between autoimmune diseases, infections, and other causes. When clinical manifestations are atypical or the response to treatment is poor, heightened vigilance is required. In the future, testing for immune-related markers in serous cavity effusions and conducting tissue biopsies may provide more assistance in diagnosis and prognosis.

Potential mechanisms of PMS reportedly include obstruction of lymphatic reflux and paraneoplastic syndrome caused by the release of vasculitic factors.[2,4] In this case, the patient was a postpartum woman with concurrent degeneration of a large uterine fibroid. Uterine fibroid degeneration refers to loss of the fibroid's original typical structure. During pregnancy, red degeneration is the most common type of fibroid degeneration, whereas cystic changes result from the further progression of hyaline degeneration in uterine fibroids.[11] Pain symptoms typically arise from necrotic obstruction of the fibroid, insufficient blood supply to the growing uterus, and prostaglandin release due to cell damage.[12] Especially in the mid to late stages of pregnancy, small blood vessels within the fibroid undergo abnormal degenerative changes. Combined with the hypercoagulable state seen in pregnancy, this leads to circulatory disorders within the fibroid, resulting in thrombus formation, hemolysis, and hemoglobin infiltration into the muscle layer.[13]

In the present case, blood loss from postpartum hemorrhage, leading to blood concentration, and the use of uterotonic drugs may have exacerbated the condition. Heparin, as an anticoagulant, is commonly used to prevent thrombus formation. It reduces blood viscosity, promotes blood flow, inhibits inflammatory reactions, and improves blood perfusion in fibroids, thereby achieving therapeutic effects. This may explain the resolution of acute ascites and abdominal pain following anticoagulant

therapy, as well as the decrease in the D-dimer level.

In conclusion, new-onset ascites during the postpartum period is a rare occurrence. While some forms of ascites have benign causes and carry a good prognosis, others may signal aggressive diseases or be associated with serious complications that can be life-threatening if not recognized and treated promptly. In this case, the patient developed acute ascites in the postpartum period, with symptoms improving following anticoagulation treatment. Although early diagnosis and treatment of atypical symptoms can be challenging and may not be explained by a single cause, this case offers valuable insights into the individualized treatment of complications related to uterine fibroids during pregnancy.

The reporting of this study conforms to the CARE guidelines.[14]

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Author contributions

XL was the primary contributor to the manuscript writing, while BZ was responsible for editing and reviewing the manuscript. Both authors read and approved the final version of the manuscript.

Ethics statement

We obtained written informed consent for publication from the patient described in this report. Institutional review board approval was not required because of the retrospective nature of the study.

Data availability statement

The data that support the findings of this study are available from the corresponding author upon reasonable request.

Declaration of conflicting interests

The authors declare that there is no conflict of interest.

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