

Recurrent ovarian hemorrhage in a patient with aplastic anemia

A case report

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Abstract

Rationale: Recurrent ovarian hemorrhage resulting in ovarian infarction may lead to a life-threatening intraperitoneal hemorrhage in women with bleeding disorders such as aplastic anemia (AA). Moreover, it is seen as ovarian tumors in the diagnosis. The authors report a clinical case with the aim of sharing our experiences and exploring the ways to diagnose, treat, and avoid ovarian hemorrhage.

Patients concerns: A 48-year-old woman with AA had suffered from a serious abdominal distension for the past 24 hours, which had occurred intermittently for the past 15 years.

Diagnoses: Pelvic ultrasonography had revealed a large anechoic area of fluid in the abdomen without any sign of primary hemorrhage each time she had experienced an episode over the past 15 years. The volume of pelvic fluid had decreased after anti-inflammatory and hemostatic treatment. At presentation, the abdominal computed tomography suggested an ovarian tumor with a massive hemoperitoneum (a right ovarian mass, $5.7 \times 5.0 \times 5.0 \text{ cm}^3$ in size, with a large amount of abdominal and pelvic fluid).

Interventions: Surgery was performed with respect to the bilateral uterine adnexa. On laparotomy, there were blood clots of approximately $6.0 \times 6.0 \times 5.0 \text{ cm}^3$ surrounding the right ovary and approximately 400 mL bloody fluid in the abdomen.

Outcomes: The patient recovered without incident and was transferred to a hematology ward 1 week later. Postoperative pathology confirmed hemorrhagic infarction of the right ovary.

Lessons: In conclusion, continuous ovarian bleeding can cause ovarian infarction to women with bleeding disorders and it may be confused with an ovarian tumor. Moreover, an earlier ovariectomy procedure under stable conditions or treatment with gonadotropin-releasing hormone that prevent bleeding via ovulation suppression may be effective for such cases.

Abbreviations: AA = aplastic anemia, CT = computed tomography, GnRH-a = gonadotropin-releasing hormone agonist.

Keywords: aplastic anemia, case report, intraperitoneal hemorrhage, ovarian infarction, pelvic fluid

1. Introduction

Bleeding is a typical symptom of patients with aplastic anemia (AA).^[1] Some reports have described corpus luteum hemorrhage in female patients with AA.^[2] However, few reports have described that AA patients with a large amount of recurrent pelvic fluid caused by ovarian bleeding that resulted in ovarian hemorrhagic infarction. This case describes a woman with AA who had a large amount of recurrent pelvic fluid without any sign of primary hemorrhage. After ovariectomy, pathologist confirmed the hemorrhagic infarction of the right ovary.

2. Case history

A 48-year-old woman with AA was admitted to the gynecological ward, having shown symptoms of abdominal distension for the past 24 hours, which had occurred intermittently for the past 15 years. In those periods, she had suffered from abdominal distention approximately 4 to 5 times per year without inducing; these episodes resulted in more complicated symptoms of nausea, vomiting, and chest distress, which was sometimes severe. Pelvic ultrasonography had showed that there was a large anechoic area of fluid in the abdomen that caused the episode. However, the symptoms lessened after anti-inflammatory and hemostatic treatment. In the past 24 hours, she had again abdominal distention with nauseating, vomiting, and chest distress. Her menstrual cycle had been irregular over the last 4 years. Her last menstrual period had lasted 45 days. Additionally, she denied sexual activity for 19 years after her divorce and had been diagnosed with AA for 27 years. Further, her condition was controlled by taking testosterone to promote hematopoiesis (intramuscular injection of 100 mg, every other day).

During the examination, she appeared to be pale and had irregular blood circulation (pulse 102/min and blood pressure 90/59 mm Hg). Her abdomen was distended and tense. A gynecological examination revealed a mobile uterus of normal size and a tender right adnexal structure (approximately $5.0 \times 6.0 \times 6.0 \text{ cm}^3$ in size). The pregnancy and tuberculin tests were negative. Trans vaginal pelvic ultrasonography (Fig. 1) revealed a right complex adnexal mass of $5.50 \times 4.33 \times 4.62 \text{ cm}^3$ in size

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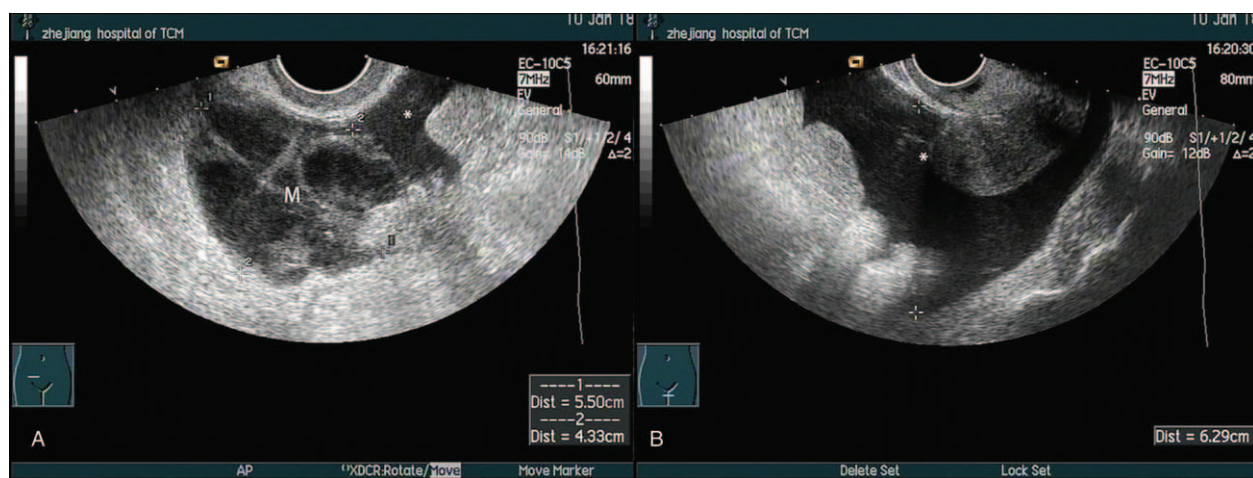


Figure 1. (A) Sagittal ultrasound image showing the complex adnexal mass (M), $5.50 \times 4.33\text{ cm}^2$ in size, and free fluid (*) in the abdomen. (B) Coronal image showing considerable free fluid (*) in the abdomen suggestive of massive hemoperitoneum.

with a large anechoic area of fluid in the abdomen (6.29 cm in depth) showing the right ovarian tumor with hemorrhage. The abdominal computed tomography (CT) (Fig. 2) revealed a right adnexal mass of $5.7 \times 5.0\text{ cm}^2$ in size with a large mass of abdominal and pelvic fluid, as shown on pelvic ultrasonography. The pertinent laboratory findings were as follows: hemoglobin: 96 g/L, platelets: $61 \times 10^9/\text{L}$, total leukocytes: $6.5 \times 10^9/\text{L}$, erythrocyte sedimentation rate: 18 mm/h, prothrombin time: 12.20 seconds, activated partial prothrombin time: 23.50 seconds, cancer antigen 125: 131.2 U/mL, cancer antigen 199: 3.9 U/mL, carcinoembryonic antigen: 1.4 ng/mL, liver function: total protein: 74.5 g/L (normal reference value 65.0–85.0 g/L), and albumin 40.7 g/L (normal reference value 40.0–55.0 g/L). A provisional diagnosis of intraperitoneal hemorrhage was made. After completing a series of 10 units of platelets and intravenous antibiotics (meropenem) for prophylaxis infection, laparotomy was performed after admission when her platelet count was $81 \times 10^9/\text{L}$ following a blood transfusion.

On exploratory laparotomy, there were blood clots of approximately $6.0 \times 6.0 \times 5.0\text{ cm}^3$ surrounding the right ovary and approximately 400 mL bloody fluid in the abdomen. There was no obvious rupture on ovarian surface. The uterus, bilateral

fallopian tubes, and left ovary were otherwise normal. The bilateral adnexa were successfully resected. Intraoperative rapid frozen pathology suggested a hemorrhagic infarction of the right ovary. After releasing the details of her case to her parents, we ended the surgery and the intraoperative blood loss was only 100 mL. The patient received 12 units of platelets and 190 mL fresh frozen plasma transfusion during the operation. She was transferred to the intensive care unit and received intravenous meropenem for gram-positive bacteria infection prophylaxis. The next day, she was transferred back to a general ward. She was moved and referred to the hematology ward 1 week after surgery. Histopathological evaluation (Fig. 3) revealed a hemorrhagic infarction of the right ovary. We reported the case at 3 months following surgery, and the patient had recovered very well without abdominal distention. Pelvic ultrasonography showed nothing abnormal.

3. Discussion

Anemia, hemorrhage, and infection are the typical symptoms of patients with AA, but generally such patients do not have long-standing illness.^[1] Almost all patients have a hemorrhagic



Figure 2. The abdominal computed tomography showing a right adnexal mass (M), $5.86 \times 5.23\text{ cm}^2$ in size, with a large abdominal and pelvic fluid (*).

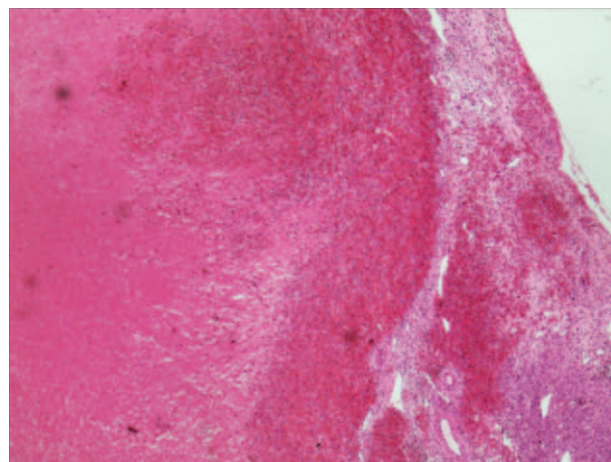


Figure 3. Pathological section showing a hemorrhagic infarction of the right ovary.

tendency. More than 60% of patients have visceral hemorrhage, such as gastrointestinal, fundus, and cranial internal hemorrhage. There are several case reports of corpus luteum hemorrhage in patients with AA.^[3–5] The difference between those 3 reports and this case is that our patient had suffered from a large amount of recurrent pelvic fluid for 15 years without signs of corpus luteum rupture or other obvious primary hemorrhage in pelvic ultrasonography. In addition, the case described here had a recurrent onset whereas the other cases were accidental.

In this case, the transvaginal pelvic ultrasonography and abdominal CT revealed a right adnexal mass with a large amount of abdominal and pelvic fluid. It may be confused upon diagnosis as an ovarian tumor. The ovariectomy and pathology confirmed hemorrhagic infarction of the right ovary.

We concluded that the ovary might bleed during ovulation, which can cause a recurrent large amount of pelvic fluid in women with AA, resulting in ovarian infarction. This may lead to a life-threatening hemoperitoneum in women with bleeding disorders.^[6] Although the advice for surgery was clear and precise, it was suspected that the patient was actively experiencing hemorrhage in the abdomen. The low platelet count secondary to AA made emergency surgery risky and potentially fatal. The current case suggests that a life-threatening intraperitoneal hemorrhage following ovulation may be another cause of death for female patients with AA. It is thus necessary to perform early detection and treatment. Nikolov et al^[6] achieved satisfactory efficacy by using gonadotropin-releasing hormone agonist (GnRH-a) to treat severe uterine hemorrhages, caused by

congenital aplastic and hypoplastic anemias in adolescents. We infer that the following 2 methods may be effective for avoiding recurrent catastrophic bleeding complications in women with AA: first, earlier ovariectomy should be conducting under stable conditions, and second, GnRH-a treatment can help prevent bleeding via ovulation suppression. However, intensive research is needed to verify these methods.

Author contributions

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