



Spontaneous Rupture of Ovarian Artery Aneurysm in a Postmenopausal Woman: A Case Report and Literature Review

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Spontaneous rupture of an ovarian artery aneurysm is an extremely rare, life-threatening disease and has been reported to be most highly associated with pregnancy. The current study presents a case of intraperitoneal and retroperitoneal hematoma caused by spontaneous rupture of a right ovarian artery aneurysm in a 56-year-old woman. A 56-year-old woman visited the emergency room with right lower quadrant abdominal pain. Contrast-enhanced computed tomography showed a large retroperitoneal and intraperitoneal hematoma and active extravasation of contrast medium in the right retroperitoneum. Consequently, transcatheter arterial embolization was successfully performed. Spontaneous rupture of an ovarian artery aneurysm should be suspected in multiparous women with abdominal or flank pain even if it is unrelated to pregnancy. Suspicion of this entity is needed for earlier diagnosis and management.

Key Words: Aneurysm, Retroperitoneal space, Spontaneous rupture, Therapeutic embolization

INTRODUCTION

Spontaneous rupture of an ovarian artery aneurysm is an extremely rare disease and occurs most often during the intrapartum or puerperium period. This paper presents a case of retroperitoneal and intraperitoneal bleeding caused by spontaneous rupture of a right ovarian artery aneurysm in a 56-year-old postmenopausal woman with well-controlled hypertension. She was diagnosed by contrast-enhanced computed tomography (CT) and treated by transcatheter arterial embolization (TAE).

CASE REPORT

A 56-year-old menopausal woman, gravida 2 para 1, visited the emergency department with right lower quadrant abdominal pain for 3 hours. She had no history of abdominal trauma or physical stress. She was

taking anti-hypertensive medication for well-controlled blood pressure and had not used any anticoagulants. She had a history of 1 vaginal delivery, 1 spontaneous abortion, and menopause at 3 years prior. At presentation, she was hyperventilating due to extreme abdominal pain that radiated through the whole abdominal area including the upper abdomen. She complained of nausea without vomiting.

On arrival, her blood pressure was 96/60 mmHg, pulse rate was 117 beats per minute, and body temperature was 36.7°C. The patient was pale with a distended abdomen. Physical examination was significant for muscle guarding with pain in the right lower quadrant.

Laboratory results showed decreased hemoglobin concentration of 8.6 g/dL and hematocrit of 25.1% and an elevated white blood cell count ($14,500/\text{mm}^3$). The liver enzymes were slightly elevated (aspartate transaminase [AST] 44 U/L and alanine transferase [ALT] 54 U/L). The hemoglobin concentration dropped to 5.9

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Fig. 1. Contrast-enhanced dynamic computed tomography: A large hematoma with active extravasation of intravenous contrast in the right retroperitoneum. A moderate amount of hemoperitoneum also was found in the pelvic cavity and right subphrenic space. (A) Coronal view. (B) Sagittal view.



Fig. 2. Abdominal aortogram: Selective right gonadal arteriogram showing contrast extravasation at the right distal gonadal artery.



Fig. 3. Abdominal aortogram: No abnormalities of the abdominal aorta and other arteries.

g/dL at 2 hours after arrival. Conservative treatment including aggressive fluid therapy and blood component transfusion was administered.

To investigate suspected hemoperitoneum, emergent contrast-enhanced abdominal and pelvic CT was performed. CT showed a large hematoma and active extravasation of intravenous contrast in the right retroperitoneum. A moderate amount of hemoperitoneum was observed in the pelvic cavity and right subphrenic space (Fig. 1). Emergent transfemoral angiography revealed active contrast extravasation suggestive of an aneurysm at the distal portion of the right ovarian artery (Fig. 2). Abdominal aortogram examined the main artery that could cause massive retroperitoneal bleeding, including the abdominal aorta, and there were no

abnormal vascular findings (Fig. 3). Embolization was conducted using diluted glue (1:7) with a concerto coil (2 mm × 4 cm). Abdominal aortic angiogram showed no contrast extravasation after embolization (Fig. 4).

The next day, there was no significant change in amount of right retroperitoneal hematoma or hemoperitoneum and no evidence of active bleeding on CT. The abdominal pain gradually subsided, vital signs stabilized, and hemoglobin concentration increased to 10.1 g/dL. The patient was managed conservatively with antibiotics and did not need further surgical intervention. She was discharged 8 days after embolization and she did not complain of any symptoms during 2 months of follow-up.

DISCUSSION

Retroperitoneal hematoma is a life-threatening emergency event. Abdominal trauma resulting from iatrogenic injury during large vessel surgery is the most common cause of retroperitoneal hemorrhage [1,2]. Furthermore, renal artery rupture, retroperitoneal tumor, and coagulopathic disorder have been suggested as major causes of retroperitoneal hemorrhage [3].

Most ruptures of an ovarian artery aneurysm are related to pregnancy and occur during the peripartum or postpartum period. The hemodynamic and hormonal changes during pregnancy appear to cause arterial



Fig. 4. Abdominal aortogram: After selective embolization of right gonadal artery.

alterations that can lead to new aneurysm formation and/or weakening of a preexisting aneurysm [4]. Postpartum uterine involution induces a dextrorotated uterus to return to the normal prepregnant position, and the associated physiological changes can be involved in the formation of ovarian artery aneurysms. The clinical manifestation is often nonspecific and can be erroneously diagnosed as placental abruption or renal calculus.

Based on a MEDLINE search of the English language literature from 1995 to 2020, only 9 cases of spontaneous rupture of an ovarian artery aneurysm not related directly to pregnancy have been published (Table 1) [5-13]. The ages of the patients ranged from 35 to 69 years (mean, 51.2 years), and 5 of 9 patients were postmenopausal. All reported cases occurred in multiparous women, as with our case.

Most previously reported ruptures of an ovarian artery aneurysm occurred in multiparous or grand multiparous woman, suggesting that repeat pregnancy is a risk factor for rupture of an ovarian artery aneurysm [14]. The repeated hemodynamic and endocrine changes during multiple pregnancies could be the cause of arterial change that can exacerbate an aneurysm or rupture a pre-existing aneurysm [10]. Of the 9 cases, 4 had hypertension, including ours. High blood pressure might be a risk factor for spontaneous rupture of ovarian artery aneurysm. Although we could not investigate the recent fluctuation of blood pressure in our case, such fluctuation might cause aneurysm rupture [10].

Because spontaneous rupture of an ovarian artery aneurysm is rare, it might be underdiagnosed. The most common presenting symptom of rupture of ovarian

Table 1. Present case and 9 reported cases of spontaneous rupture of ovarian artery

Age (y)	Obstetrical status	Menopausal status	Underlying disease	Location	Treatment	Reference
53	G1P1	Postmenopause	Not available	Left	Laparotomy	Manabe et al. (2002) [13]
55	G2P2	Postmenopause	None	Right	TAE	Nakajo et al. (2005) [11]
69	G3P3	Postmenopause	Rheumatoid arthritis, osteoporosis, hypertension, bipolar disorder	Left	TAE	Kirk et al. (2009) [9]
46	G3P2	Premenopause	Hypertension	Left	Laparotomy	Chao and Chen (2009) [10]
48	G2P2	Premenopause	None	Left	Laparotomy	Tsai and Lien (2009) [8]
51	G3P3	Postmenopause	None	Right	Laparotomy	Kodaira et al. (2014) [12]
45	G3P3	Premenopause	None	Right	TAE	Toyoshima et al. (2015) [7]
35	Multiparous	Premenopause	HIV, hypertension, type 2 diabetes	Right	TAE	Zorrilla et al. (2019) [6]
59	Not available	Postmenopause	Chronic lymphocytic leukemia, hypertension	Right	TAE	Herskowitz et al. (2020) [5]

G: gravida, P: para, HIV: human immunodeficiency virus, TAE: transcatheter arterial embolization.

artery aneurysm is acute abdominal and flank pain with hemodynamic instability. In earlier reports, aneurysm rupture was diagnosed mostly by exploratory laparotomy. However, CT and angiography recently have improved the diagnostic process and location of hemorrhage. Contrast-enhanced abdominal CT can be helpful for rapid diagnosis and identification of bleeding focus. In our case, hematoma was found not only in the retroperitoneum, but also in the intraperitoneum and could be diagnosed on CT.

According to previously published literature [1,6,7], most of the spontaneous rupture of ovarian artery aneurysm requires immediate management because they were accompanied by massive bleeding and life-threatening. Of the 9 published cases, 5 underwent successful TAE. Since TAE was used to successfully manage a ruptured ovarian artery aneurysm in 1990 [15], it has been used as a treatment option along with ligation of the ovarian artery through laparotomy. Nevertheless, exploratory laparotomy should be performed with failed TAE or if the patient is unstable hemodynamically.

In conclusion, we reported a rare case of spontaneous rupture of an ovarian artery aneurysm presenting as a retroperitoneal hematoma that was not associated with pregnancy. Diagnostic angiography followed by TAE can be an effective therapeutic procedure for such an event. In women presenting with severe acute pain of the abdomen and flank, rupture of an ovarian artery should be suspected, especially in multiparous women with retroperitoneal hematoma.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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