

Spontaneous Rupture of Aneurysms of the Ovarian Artery at Times Remote from Pregnancy

Yuzo Kodaira¹, Taro Iwamura¹, Hiroki Hoshino¹,
Ken Takahashi¹, Yutaka Kawahigashi¹ and Koshi Matsumoto²

¹Department of Surgery, Heisei-Tateishi Hospital

²Department of Diagnostic Pathology, Ebina General Hospital

Abstract

Spontaneous rupture of an aneurysm of the ovarian artery is usually considered a rare complication of pregnancy and the puerperium. However, we observed this emergent condition in a 51-year-old postmenopausal woman. We report here our experiences and consider lessons about diagnosis and management that can be drawn from this case and 5 other published cases in multiparous middle-aged women. These lessons include application of contrast-enhanced computed tomography to focus emergent care, surgical intervention, and association with systemic inflammatory response syndrome. We also consider how the cases might shed new light on the pathogenesis and evolution of this condition.

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Key words: retroperitoneal hemorrhage, ovarian artery, aneurysm

Introduction

Spontaneous retroperitoneal hemorrhage is a surgical emergency usually originating from rupture of an aneurysm of the aorta or the renal artery, malignant and benign retroperitoneal tumors, and disorders of coagulation¹. Rupture of an aneurysm of the ovarian artery rarely causes spontaneous retroperitoneal hemorrhage; only 16 cases having been reported^{2–17}. After the first case was described in 1963, 11 additional cases occurring in late pregnancy or early in the puerperium have been reported^{7–17}. However, 5 cases of retroperitoneal hemorrhage caused by spontaneous rupture of the ovarian artery not associated with these periods of

pregnancy have recently been described^{2–6}.

We describe an additional case of retroperitoneal hemorrhage due to a dissecting aneurysm of the ovarian artery outside the peripartum period. On the basis of this case and previously published cases we describe key clinical features that should alert clinicians to this condition.

Case Report

A 51-year-old woman presented to the emergency room with the right flank pain and lower abdominal pain of sudden onset. On admission, blood pressure was 106/68 mm Hg, heart rate was 96 beats per minute, and body temperature was 37.0°C. The physical examination showed tenderness in the right

Correspondence to Yuzo Kodaira, Department of Surgery, Flowers and Forest Tokyo Hospital, 2–3–6 Nishigahara, Kita-ku, Tokyo 114–0024, Japan
E-mail: kodaira@hanamorithp.jp
Journal Website (<http://www.nms.ac.jp/jnms/>)



Fig. 1 Massive retroperitoneal hemorrhage visualized with contrast-enhanced CT.

lower quadrant of the abdomen but no rebound tenderness or muscular defense of the abdominal wall. The white blood cell count of $22,600/\mu\text{L}$ and C-reactive protein (CRP) concentration of 0.91 mg/dL (normal $<0.45\text{ mg/dL}$) suggested an acute-phase inflammatory change. The hemoglobin concentration was 10.2 g/dL , and the hematocrit was 31.2% . Results of other biochemical studies of the blood and of urinalysis were normal. The x-ray studies of the chest and abdomen showed no abnormalities. The past history included occasional episodes of anxiety treated with sedatives and an untreated myoma of the uterus. The patient was multiparous, with pregnancies having occurred 29, 26, and 18 years earlier with normal gestations and deliveries. The patient had no history of hypertension or cardiovascular disease. She had taken no anticoagulants.

The patient was admitted to the hospital for a suspected acute inflammatory ileocecal lesion. However, shortly after admission, the pain in the right lower quadrant worsened and expanded to the midline. Contrast-enhanced computed tomography (CT) of the abdomen showed an area of heterogeneous density in the retroperitoneal space extending from the inferior pole of the right kidney to the inguinal area. The lesion had a CT value of approximately 70, the same density as blood, and the study was interpreted as showing a massive retroperitoneal hemorrhage (**Fig. 1**). The early phase of enhancement demonstrated sudden discontinuity of the right ovarian artery in the middle of the hematoma.

Based on these images and the presentation, a diagnosis of acute retroperitoneal hemorrhage caused by the bleeding of the right ovarian artery was made, and an emergency laparotomy was performed. The laparotomy revealed extraperitoneal hemorrhage, as expected, and intraperitoneal hemorrhage, apparently caused by extension of bleeding through a tear of the mesenteric membrane. The later finding suggested that the exacerbation and expansion of the abdominal pain at admission were due to intra-abdominal extension of hemorrhage causing, in turn, acute peritonitis.

When the right colon and the intestinal mesentery were rotated medially, a massive retroperitoneal hematoma weighing approximately $1,000\text{ g}$ was exposed and then removed. These procedures exposed the ovarian artery and vein, which were found to be skewed and ruptured. The vessels were ligated and resected. Examination of the right retroperitoneal space, including the uterus and the ovary, revealed no other abnormality or injury. The postoperative course was unremarkable, and the patient was discharged from the hospital on the 9th postoperative day.

The segment of the ovarian artery removed at laparotomy was approximately 70 mm long, and part of the segment was tangled and had formed nodules. The pathological diagnosis of the specimen was a dissecting aneurysm. Microscopic examination showed dissection of the media in areas where nodules had formed and false lumens filled with clotted blood (**Fig. 2a**). A break of the internal elastic lamina could be seen where the wall had been dissected (**Fig. 2b**).

Discussion

When aneurysms of the ovarian artery form and why rupture is associated with late pregnancy and the early puerperium are poorly understood. Barrett et al. have speculated that pregnancy causes hyperplasia of the intima and changes in the organization and content of the media¹⁸. The hemodynamic and hormonal alterations of pregnancy are the most likely cause of these arterial changes. These pregnancy-related physiologic

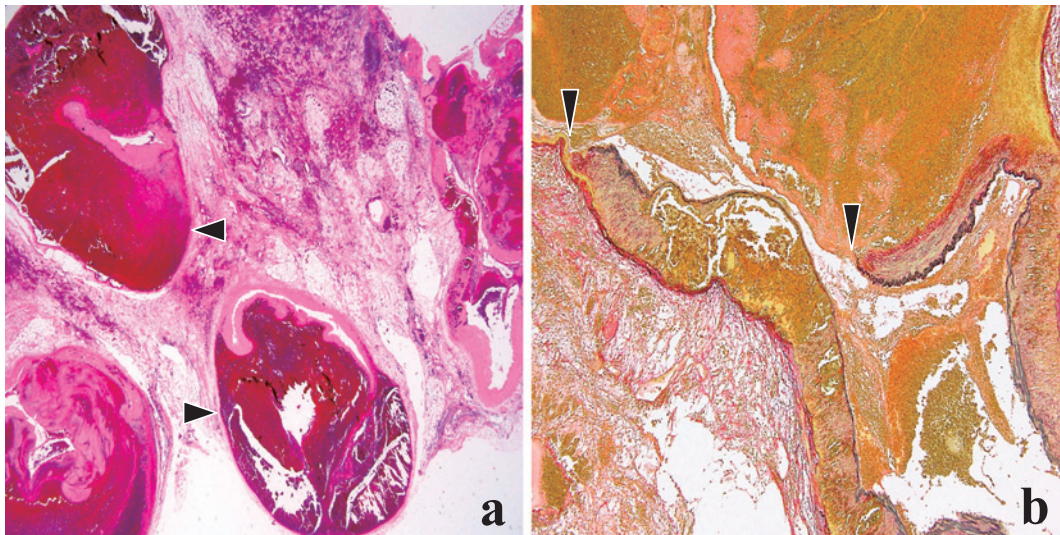


Fig. 2 **a.** Microscopic appearance of a rupture of an aneurysm of the ovarian artery. The image shows dissection of the media and false lumens filled with clotted blood (**arrowheads**). (hematoxylin and eosin, $\times 10$)
b. Disruptions (**arrowheads**) of the internal elastic lamina of the ovarian artery caused by dissection of an aneurysm. (Masson trichrome stain, $\times 25$)

changes might in turn cause formation and rupture of arterial aneurysms. However, the present case and several previously reported cases indicate that an aneurysm of the ovarian artery can rupture at times remote from pregnancy.

While rupture of aneurysms of the ovarian artery is closely associated with pregnancy, what can be said about formation of the aneurysm? The lack of reports of aneurysms of the ovarian artery in nulliparous women and of spontaneous aneurysms of the testicular artery suggests that pregnancy is involved in aneurysm formation¹⁹. In fact, the present report and several earlier reports of ruptures occurring at times remote from pregnancy are consistent with an alternative theory that pregnancy causes these aneurysms to form but not necessarily to rupture. The recent observation of Kirk et al. seems to address this point³. These authors have reported on a 69-year-old multiparous woman with a ruptured left ovarian artery aneurysm in whom an intact aneurysm of the contralateral ovarian artery was incidentally found. Because of a paucity of cases, the nature of ovarian artery aneurysms such as incidence, risk factors of the formation, trigger for the rupture remains unclear. Further accumulation of case reports or angiographic analysis of ovarian arteries in large

numbers of parous and nulliparous women may help elucidate the nature of aneurysms.

The effects of contrast-enhanced CT on the diagnosis and management of retroperitoneal hemorrhage has, in our experience, been critical. In the present case, contrast-enhanced CT revealed both the retroperitoneal hemorrhage and the discontinuity of the right ovarian artery. As in the present case and in several earlier cases²⁻⁶, contrast-enhanced CT provides rapid and accurate diagnostic information in the emergency setting. Before CT was available, ovarian aneurysms were diagnosed during exploratory surgery². These cases thus demonstrate how enhanced imaging can focus emergency procedures on the pathologic lesion and might allow some surgical procedures to be avoided.

Our case and 5 other cases summarized in **Table I** offer clues to how the clinical presentation evolves. Rupture of an aneurysm of the ovarian artery was associated with an increase in the white blood cell count in each of 3 cases for which information is available and with hemodynamic changes in 3 of 5 cases. These findings, in the absence of infection, are consistent with the systemic inflammatory response syndrome (SIRS). Lappen et al. have recently reported a high incidence of SIRS in patients with chorioamnionitis, many without detectable evidence

Table 1 Rupture of aneurysms of the ovarian artery at times remote from pregnancy

Case	1	2	3	4	5	6
Age (years)	46	69	48	55	53	51
Obstetrical status*	G3P2	G3P3	G2P2	G2P2	G1P1	G3P3
Side	left	left	left	right	left	right
Treatment	laparotomy endovascular: failed	endovascular	laparotomy endovascular: failed	endovascular	laparotomy	laparotomy
Hemodynamic manifestation**	yes	no	no	no	yes	yes
White blood cells (number/ μ L)	not described	not described	11,640	not described	13,700	22,600
Reference	2	3	4	5	6	present case

*Obstetric status: G=gravida; P=para

**Hemodynamic manifestation is defined as systolic blood pressure<90 mm Hg or heart rate>100 beats per minute or both

of sepsis²⁰. What causes SIRS in individuals with local infection, or in the absence of infection, has been a subject of keen inquiry. We have previously reported that heparan sulfate, a glycosaminoglycan released from damaged tissues, such as vascular endothelial cells, can stimulate inflammatory cells and incite the clinical picture of SIRS²¹⁻²³. If, the rupture of an ovarian artery aneurysm is indeed associated with SIRS, as our experience suggests, a question would be whether rupture causes SIRS or whether SIRS facilitates rupture and the subsequent changes. We should note the mechanism that SIRS can be induced by tissue damage itself through the activation of inflammatory cells without infections. Otherwise, delayed or wrong diagnostic work-up can be made, with the consequence that initial treatments are mismanaged like as administration of antibiotics alone.

In the present case, emergency laparotomy was necessitated by rapid progression of the abdominal findings caused by extension of bleeding from the retroperitoneal space to the intraperitoneal space and by hemodynamic changes. Laparotomy is a reliable emergency treatment for spontaneous or traumatic retroperitoneal hemorrhage²⁴. Clearly, severe hemodynamic disorders and evidence of panperitonitis will usually necessitate laparotomy. Key questions are whether and when laparotomy can be avoided. Transarterial embolization has recently been reported to be an effective treatment for active arterial bleeding^{2-5,7,9,11}. Embolization of the

ovarian artery was successful in 5 of 7 reported cases. Furthermore, Guillem et al. have reported a case in which transarterial embolization was combined with retroperitoneal endoscopy to drain a hematoma⁹. Chao and Chen suggest that transarterial embolization is useful in cases without hemodynamic instability². Still, minimally invasive approaches are reported to have failed^{6,7} when microcatheters were difficult to insert into small-caliber arteries because of spasm and tortuousness. Therefore, preparations for emergent laparotomy should be made when invasive intervention is planned.

Conflict of Interest: The authors indicated no conflicts of interest.

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