Case Report

Pregnancy-unrelated spontaneous rupture of a right ovarian artery aneurysm✩,✩✩,★★

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ABSTRACT

Spontaneous rupture of an ovarian artery aneurysm is extremely rare. It can lead to retroperitoneal hemorrhage that is often life-threatening. We report a case of pregnancy-unrelated spontaneous rupture of a right ovarian artery aneurysm in a multiparous woman. A 29-year-old woman, gravida 3, para 3, whose latest pregnancy involved uneventful gestation and delivery 2 years previously, was admitted for right flank pain. The urine test result for pregnancy was negative. Computed tomography revealed a large retroperitoneal hematoma and right ovarian artery aneurysm with contrast extravasation. After selective angiography, embolization of the right ovarian artery was successfully achieved using microcoils. Diagnostic angiography with subsequent transcatheter arterial embolization is an effective and less invasive technique for the management of ovarian artery aneurysm.

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Introduction

Although spontaneous rupture of an ovarian artery aneurysm (OAA) is extremely rare, it can lead to retroperitoneal hemorrhage that is often life-threatening [1]. Most cases of spontaneous rupture of an OAA are related to pregnancy and occur during the peripartum or postpartum period. However, spontaneous rupture of an OAA unrelated to pregnancy has been described in a few reports [1].

Recently, transcatheter arterial embolization (TAE) has been increasingly performed to manage ruptured OAA; however, various treatment options, including observation and surgical repair, are available [1,2].

In this paper, we report a case of pregnancy-unrelated spontaneous rupture of an OAA in a young multiparous...

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A 29-year-old woman, gravida 3, para 3, was transferred to the emergency room for progressively worsening right flank pain that suddenly occurred 2 days previously. The patient had no history of hypertension, gynecologic disease, abdominal surgery, or abdominal trauma. Her last pregnancy was 2 years ago, with an uneventful gestation and delivery. The most recent menstruation period was over 3 weeks prior, with no change in flow and duration. Her family history was negative for genetic or connective tissue disorders. On admission, the patient was alert but anemic. Her blood pressure was 70 of 50 mm Hg, pulse rate 110 beats/min, and body temperature 36.9°C. The abdomen was slightly distended, and palpation revealed tenderness with mild muscle guarding in the right lateral abdomen. Hematological examination revealed a red blood cell count of $305 \times 10^9$/mm$^3$, a hemoglobin concentration (Hb) of 9.6 g/dL, and a white blood cell count of 15,060/mm$^3$. Laboratory data, including coagulation studies, were within the normal ranges. The urine test result for pregnancy was negative. Abdominal ultrasonography revealed a large retroperitoneal mass adjacent to the right kidney. Emergent computed tomography (CT) of the abdomen and pelvis revealed a large retroperitoneal hematoma ($10 \times 13$ cm) with extravasation of contrast extending from the inferior aspect of the right kidney to the right pelvic regions and a right ovarian artery with an aneurysm at its middle portion in the center of the hematoma (Fig. 1). These findings suggest that the retroperitoneal hematoma resulted from a rupture of the right OAA. No other vascular abnormality, such as a left OAA or renal or splenic artery aneurysm, was detected on CT. To identify the origin of the bleeding and manage the bleeding less invasively with TAE, selective angiography of the right ovarian artery was performed immediately after CT under aggressive resuscitation with intravenous infusion of packed red blood cells and crystalloid. Transfemoral selective angiography showed a tortuous and segmentally dilated right ovarian artery with a saccular aneurysm ($2.6 \times 2.3$ cm) (Fig. 2). Subsequently, embolization of the right ovarian artery was performed using microcoils. An angiographic catheter (5Fr, Type Micaelson, Medikit Tokyo) was introduced into the right ovarian artery using an AQUA V-III guiding wire (Cardinal Health Japan, Tokyo). A Carnelian PIXIE ER MSV110S Microcatheter (Tokai Medical Products, Aichi, Japan) was introduced, and then 4 microcoils (C-stopper coil 0.014" filling coil (Piolax Medical Device, Kanagawa, Japan), Hilal embolization microcoil, Tornado® embolization coil, and Nester® embolization coil (Cook Medical, Bloomington, IN, USA) were placed in the artery proximal and distal to the aneurysm. Postembolization angiography demonstrated complete occlusion of the artery and disappearance of extravasation of contrast (Fig. 2). The patient required a transfusion of 2 units of packed red blood cells to decrease Hb concentration to 8.7 g/dL on postembolization day 1. However, the patient’s recovery was uneventful. CT performed on post-embolization days 4, 11, and 38 showed progressive shrinking of the retroperitoneal hematoma without extravasation of contrast (Fig. 3). The patient is well with regular menstruation one year after TAE.
Fig. 2 – Transfemoral selective angiography shows a tortuous and segmentally dilated right ovarian artery with the saccular aneurysm at its middle portion (A). Postembolization angiography shows complete occlusion of the artery and the disappearance of extravasation of the contrast medium (B).

Fig. 3 – Postoperative computed tomography (11 days (A) and 38 days (B) after TAE) shows a shrinkage of the retroperitoneal hematoma. (White arrow).

**Discussion**

Spontaneous retroperitoneal hemorrhage or hematoma is a life-threatening complication of various clinical pathologies, including vascular diseases, rheumatologic disorders, renal tumors, adrenal tumors, trauma, and coagulopathy. Among vascular diseases, rupture of an abdominal aortic aneurysm and aneurysm of a visceral artery, such as the renal artery or splenic artery, is recognized as the common cause of retroperitoneal hemorrhage. In contrast, spontaneous rupture of an OAA is a rare cause of retroperitoneal hemorrhage [1].

Most cases of spontaneous rupture of an OAA have occurred in late pregnancy or the early postpartum period;
Table 1 – Reported cases of spontaneous rupture of ovarian artery aneurysms.

| Case | Age (years) | Gravida/Para | Ova. art | Onset | Treatment |
|------|-------------|--------------|----------|-------|-----------|
| Pregnancy-related rupture of OAA | 1 | 29 | G4P4 | L | 2d postpartum | laparotomy |
| | 2 | 35 | G6P3 | L | 4d postpartum | laparotomy |
| | 3 | 38 | G6P6 | R | During delivery | laparotomy |
| | 4 | 32 | G3P3 | L | 4d postpartum | laparotomy |
| | 5 | 35 | G3P3 | R | 4d postpartum | laparotomy |
| | 6 | 26 | G5P4 | R | 1d postpartum | laparotomy |
| | 7 | 23 | N/A | R | 1m postpartum | laparotomy |
| | 8 | 31 | G4P3 | R | 39w of gestation | laparotomy |
| | 9 | 36 | G5P5 | R | 4d postpartum | TAE |
| | 10 | 38 | G3P2 | R | during delivery | laparotomy |
| | 11 | 38 | G3P2 | R | 4d postpartum | TAE |
| | 12 | 38 | G12P11 | R | 3d postpartum | laparotomy |
| | 13 | 37 | P4 | L | 39w of gestation | laparotomy |
| | 14 | 30 | G5P5 | L | 5h postpartum | laparotomy |
| | 15 | 39 | G5P4 | R | 5d postpartum | TAE |
| | 16 | 32 | P4 | L | 2d postpartum | laparotomy→TAE |
| | 17 | 37 | G4P4 | L | 4d postpartum | TAE |
| | 18 | 31 | G6P4 | L | 2d postpartum | TAE |
| | 19 | 38 | P5 | L | 4d postpartum | TAE→laparotomy |
| | 20 | 31 | G6P5 | R | 5d postpartum | TAE |
| Pregnancy-unrelated rupture of OAA |  |  |  |  |  |
| 21 | 45 | G6P5 | L | follicular phase | laparotomy |
| 22 | 53 | G1P1 | L | postmenopause | laparotomy |
| 23 | 55 | G2P2 | R | postmenopause | TAE |
| 24 | 46 | G3P2 | L | 2d menstruation | TAE→laparotomy |
| 25 | 69 | G3P3 | L | postmenopause | TAE |
| 26 | 48 | G2P2 | L | 2d menstruation | TAE→laparotomy |
| 27 | 51 | G3P3 | R | postmenopause | TAE |
| 28 | 45 | G3P3 | R | 3d menstruation | TAE |
| 29 | 52 | G2P2 | R | postmenopause | TAE |
| 30 | 35 | Multiparous | R | ? | TAE |
| 31 | 80 | G9P6 | L | postmenopause | TAE |
| Case | 29 | G3P3 | R | luteal phase | TAE |

Ova: ovarian, OAA: ovarian artery aneurysm, TAE: transcatheter arterial embolization, d: days, w: weeks, R: right, L: left.

* Laparotomy for hematoma evacuation.
** Failed TAE.
hemorrhage in our patient. Subsequent selective angiography was also helpful in identifying the bleeding location and managing bleeding with TAE. Considering the frequency of rupture at the time of presentation and the uncertain natural history of an OAA, surgical intervention should be performed to prevent further enlargement and rupture of the aneurysm in women of childbearing age and women with a multiparous history when an OAA is identified.

Before 1990, surgical repair (adnexectomy and ligation of the ovarian artery) was mainly performed in all patients; however, TAE has been increasingly employed as the treatment of choice over the past 3 decades [1,3]. TAE was used in 16 (72.7%) of 22 patients with rupture of the OAA after 1990, although 2 patients required surgical repair for failed TAE [1,3]. We also performed TAE of the right ovarian artery using microcoils immediately after identifying the origin of the bleeding using selective angiography. The commencement of treatment with TAE when making the diagnosis of ruptured OAA by selective angiography and less invasiveness of TAE, compared with surgical repair, is a major advantage because most patients with this pathology might be in shock. Furthermore, TAE is applicable when the origin of bleeding is uncertain on CT. However, the optimal treatment strategy for a ruptured OAA is controversial because the patient’s condition before treatment varies among patients but is often unstable. Given the promptness and less invasiveness of endovascular treatment, TAE should be considered for patients with a ruptured OAA, particularly those with a stable hemodynamic condition.

Three types of materials, gelatin sponge particles (GSPs), microcoils, and N-butyl-2-cyanoacrylate (NBCA), were used for embolization. A study evaluating TAE outcomes using these materials concluded that although TAE with microcoils required the greatest amount of time, TAE with microcoils or NBCA was more effective and feasible than that with GSPs in terms of primary hemostasis and prevention of recurrent bleeding. Additionally, the study suggested that microcoils were particularly suitable for arresting arterial bleeding arising directly from the main branch of the aorta [9].

Although a case of successful pregnancy following unilateral ovarian artery embolization of a ruptured OAA was reported [10], further studies are required to understand fecundity following unilateral or bilateral ovarian artery embolization.

In conclusion, we report a case of pregnancy-unrelated spontaneous rupture of a right OAA that occurred in a young multiparous patient. Rupture of an OAA should be considered a differential diagnosis in multiparous women complaining of abdominal pain, even in non-pregnant women. Diagnostic angiography combined with subsequent TAE is considered an effective and less invasive technique for managing ruptured OAA.

The current study was approved by our institutional research ethics board of St. Mary’s Hospital (21-0401). Written informed consent was obtained from the patient for the publication of this case report.

Patient consent
The patient provided informed consent for the publication of her data.

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Ethics declarations

Ethics approval and consent to participate