

## Embolization for post-partum rupture of ovarian artery aneurysm: Case report and review

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### Abstract

Spontaneous rupture of an ovarian artery aneurysm most commonly presents with abdominal pain in a multiparous woman in the early post-partum period. Aneurysms of the ovarian artery have been reported in the published work very infrequently. In our case, a 31-year-old multiparous woman experienced sudden left lower quadrant abdominal pain on the second post-partum day. Angiography showed rupture of a left ovarian artery aneurysm, which was successfully embolized using gelatin sponge particles. The patient resumed menstruation 3 months after the embolization and concurrently conceived, ultimately giving birth at term without complications. Interventional radiology appears to be a highly safe and effective technique for diagnosis and management of a ruptured ovarian artery aneurysm with minimal risk of impairing subsequent fertility.

**Key words:** angiography, early post-partum period, fertility, ovarian artery aneurysm, transcatheter arterial embolization.

### Introduction

Spontaneous rupture of an ovarian artery aneurysm is extremely rare; 21 cases have been reported in the English-language published work during the past 5 decades. The episodes tend to occur during pregnancy or in the early post-partum period. Here we report a case of spontaneous rupture of the left ovarian artery in the early post-partum period and also review the published reports about this condition. Written consent was obtained from the patient.

### Case

A 31-year-old woman, gravida 6, para 4, with a history of cesarean delivery 13 years ago, was transferred to our University Hospital complaining of severe lower abdominal pain 2 days after a vaginal delivery at 39

weeks of gestation. Her pregnancy course had been uneventful. In late pregnancy, her blood count showed a white blood cell count of 11 700/mm<sup>3</sup>, hemoglobin 10.7 g/dL, and platelets 270 000/mm<sup>3</sup>. She gave birth to a healthy male infant weighing 2958 g in a local clinic, and the placenta was delivered spontaneously. Examination following delivery revealed a minimal tear of the cervix, which was repaired properly. The estimated blood loss was 378 g.

On the second post-partum day, the patient experienced sudden onset of abdominal pain in the left lower quadrant, and she was immediately transferred to our hospital by ambulance. On arrival, she was alert but appeared uncomfortable due to the pain. At the initial physical examination, her vital signs indicated body temperature, 37.1°C; heart rate, 92 b.p.m.; respiratory rate, 16 breaths/min; and blood pressure, 96/56 mmHg. Abdominal examination showed left

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lower quadrant tenderness without rebound. Initial laboratory test results indicated an elevated white blood cell count of  $16\,700/\text{mm}^3$  and a decreased hemoglobin level of  $7.9\text{ g/dL}$ . Ultrasonographic examination of the abdomen and pelvis demonstrated no intraperitoneal effusion, but an 8-cm-diameter hematoma was visualized inferior to the left kidney. Enhanced computed tomography (CT) of the abdomen and pelvis confirmed a massive hematoma inferior to the left kidney that extended to the left retroperitoneal region (Fig. 1). Tortuous vascular structures were observed beside the hematoma and there was no extravasation.

A reformatted CT scan indicated that the tortuous vessel was the left ovarian artery with an aneurysm formation (Fig. 2). There was no indication of rupture or dehiscence of the uterine scar from the previous cesarean delivery on magnetic resonance imaging (MRI) or ultrasonography, and there were no foci of actual bleeding. Therefore, we selected expectant management.

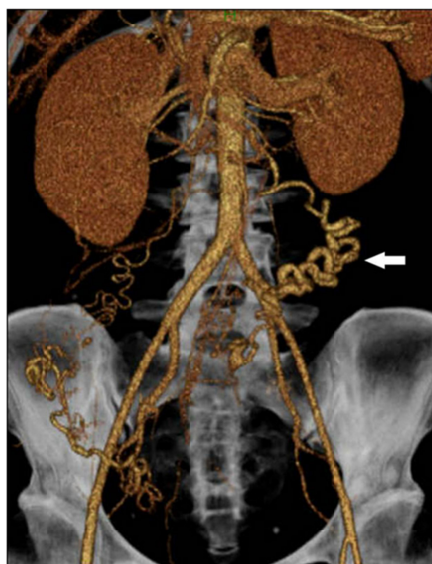
Six days after delivery, the patient's hemoglobin level decreased to  $6.4\text{ g/dL}$ , and she received a blood transfusion. Emergent angiography was undertaken to locate the origin of bleeding. She underwent catheterization through a right femoral approach with 4-Fr catheters (Pig-tail, MIK, Duck head; Medikit) and a 2.5-Fr microcatheter (Renegerd-18; TARGET, Boston Scientific). Angiography revealed the tortuous left

ovarian artery with segmental dilatations, suggesting that the ovarian artery aneurysms remained. Although there was no extravasation of the contrast medium, selective embolization of the left ovarian artery using gelatin sponge particles was performed. After embolization, an angiogram showed complete exclusion of the aneurysm, and the patient's anemia stopped progressing. Follow-up study by ultrasound examination revealed a shrinking left retroperitoneal hematoma.

The patient had an uneventful postoperative course and was discharged 14 days after the embolization. Enhanced CT 2 months after embolization demonstrated a small hematoma in the retroperitoneal space, and did not show any other aneurysm or vascular malformation. She resumed menstruation 3 months after the embolization, and she concurrently became pregnant. Twelve months after the embolization, an elective cesarean delivery was performed at 39 weeks of gestation in the local clinic. The post-partum period was uneventful.



**Figure 1** Abdominal computed tomography scan after injection of iodine contrast media on admission at 2 days after delivery. Transverse plane showed a large left retroperitoneal hematoma (arrows). Tortuous vascular structures on the left side of uterus (arrow head) suggested vascular aneurysm. No extravasation was found in this image.



**Figure 2** Reformatted computed tomography scan on admission at 2 days after delivery revealed the left tortuous ovarian artery with aneurysm formation (arrow).

## Discussion

The spontaneous rupture of an ovarian artery aneurysm is extremely rare. Our review of the published work revealed 21 cases of ovarian artery aneurysm dating back to 1963, as shown in Table 1.<sup>1-21</sup> The age of the subjects ranged from 23 to 69 years, and 14 of the 21 cases were associated with pregnancy. In 10 of those 14, the rupture of the ovarian artery aneurysm occurred within the first 5 days after delivery. In our case, the patient had sudden onset of abdominal pain 2 days after delivery, which is consistent with previous reports.

The physiology and anatomy of the cardiovascular system dramatically change during pregnancy. The enlarging uterus induces local fluctuations in blood pressure in the aorta and ovarian arteries, and dilation of the pelvic arteries occurs in conjunction with the increased uterine blood flow. It is widely accepted that involution of the uterus and the return of other genital organs to their prepregnant state in the post-partum period takes 3-4 weeks. Two mechanisms may be involved with the formation of an ovarian artery aneurysm during this time. First, failure of puerperal involution of a segment of ovarian artery after the pregnancy may induce subsequent aneurysm formation.<sup>4</sup> Second, there may be pregnancy-related changes in the vascular walls.<sup>1,3,4</sup> In a combination of animal and human studies, arterial changes were correlated with high levels of circulating steroid hormones during pregnancy.<sup>22</sup> Moreover, all of the 14 previously reported cases that were associated with pregnancy occurred in multiparous or grand multiparous women, just as in our case, suggesting that repeated pregnancy is a risk factor for the development of an ovarian artery aneurysm.

In the present case, the ovarian artery aneurysm during the post-partum period occurred on the left side. However, it is notable that in previous reports, 11 of the 14 cases of aneurysm associated with pregnancy were located in the right ovarian artery. Although the cause of ovarian artery aneurysms during the post-partum period remains poorly understood, there may be a connection to the physiological rotation of the gravid uterus. Post-partum uterine involution induces a dextrorotated uterus to return to the normal prepregnant position, and the associated physiological changes may be involved in the formation of ovarian artery aneurysms on the right side.

The rupture of an ovarian artery aneurysm should be considered in cases of flank pain or abdominal pain in the early post-partum period, and the diagnosis can be confirmed by a reformatted CT scan. Selective angiog-

raphy is a useful tool both to identify the bleeding from the aneurysm and to manage the bleeding via transcatheter arterial embolization (TAE). In eight of the 21 cases, TAE of ovarian arteries was performed, as shown in Table 1. In two of those eight, the emergent laparotomy was performed following TAE and the ovarian artery was ligated, because embolization was unsuccessful. In our case, the patient was discharged 14 days after TAE with an uneventful recovery. Over the past 3 decades, the role of TAE has evolved from a novel treatment to a major option in the management of obstetric hemorrhage.<sup>23</sup> Interventional radiology has modified this attitude further by offering a minimally invasive alternative of controlling active hemorrhage.

The impact of TAE on fecundity has not been studied in great detail. Several studies have reported cases with uneventful pregnancy outcomes,<sup>24-29</sup> and the fertility of patients who underwent embolization of uterine arteries does not appear to be statistically different from that of the general population.<sup>30</sup> In our case, the patient conceived 3 months after the embolization, and uneventfully gave birth at term. There are no studies evaluating the effect of ovarian artery embolization on ovarian function. However, a follow-up study of nontarget ovarian artery embolization following uterine artery embolization suggests that most patients do not develop ovarian failure.<sup>31</sup> To the best of our knowledge, the present case is the first report of a successful pregnancy following unilateral ovarian artery embolization of a ruptured ovarian artery aneurysm. Larger prospective studies are required to understand fecundity following unilateral or bilateral ovarian artery embolization.

In conclusion, the rupture of an ovarian artery aneurysm should be suspected in a multiparous patient presenting with flank pain or abdominal pain in the early post-partum period. Interventional radiology is an effective technique for both diagnosis and management of a ruptured ovarian artery aneurysm, and it may be the best option for patients who wish to preserve their fecundity.

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## Disclosure

The authors have no conflicts of interest related to this article.

**Table 1** Reported cases of ruptured ovarian artery aneurysm

Reference	Year	Age (years)	Obstetric status	Onset day of pregnancy-related case	Side	Treatment	Treatment outcome
1	1963	29	4G4P	Post-partum day 2	Left	Laparotomy	Success
2	1967	35	6G3P	Post-partum day 4	Left	Laparotomy	Success
3	1975	38	6G6P	During delivery	Right	Laparotomy	Success
4	1976	32	2G2P	Post-partum day 4	Right	Laparotomy	Success
5	1977	26	5G4P	Post-partum day 1	Right	Laparotomy	Success
6	Mojab and Rodriguez <sup>6</sup>	1977	—	Post-partum month 1	Right	Laparotomy	Success
7	Siu <i>et al.</i> <sup>7</sup>	1986	6G5P	—	Left	Laparotomy	Success
8	Hogdall <i>et al.</i> <sup>8</sup>	1989	4C3P	39w of gestation	Right	Laparotomy	Success
9	King <sup>9</sup>	1990	5G5P	Post-partum day 4	Right	Embolization	Success
10	Belfort <i>et al.</i> <sup>10</sup>	1993	3G2P	Post-partum day 0	Right	Laparotomy	Success
11	Guillem <i>et al.</i> <sup>11</sup>	1999	3G2P	Post-partum day 4	Right	Embolization	Success
12	Blachar <i>et al.</i> <sup>12</sup>	2000	12G11P	Post-partum day 3	Right	Laparotomy	Success
13	Panoskalis <i>et al.</i> <sup>13</sup>	2000	4P	39w of gestation	Left	Laparotomy	Success
14	Manabe <i>et al.</i> <sup>14</sup>	2002	—	—	Left	Laparotomy	Success
15	Nakajo <i>et al.</i> <sup>15</sup>	2005	2G2P	—	Right	Embolization	Success
16	Kale <i>et al.</i> <sup>16</sup>	2005	5G5P	—	Left	Laparotomy	Success
17	Rathod <i>et al.</i> <sup>17</sup>	2005	—	—	Right	Embolization	Success
18	Poiblanco <i>et al.</i> <sup>18</sup>	2008	5G4P	Post-partum day 0	Right	Embolization	Success
19	Kirk <i>et al.</i> <sup>19</sup>	2009	3G3P	Post-partum day 5	Left	Embolization	Success
20	Tsai and Lien <sup>20</sup>	2009	2G2P	—	Left	Embolization	Success
21	Chao and Chen <sup>21</sup>	2009	3G2P	—	Left	Embolization and laparotomy	Embolization was unsuccessful
22	Our case	2014	6G4P	Post-partum day 2	Left	Embolization and laparotomy	Embolization was unsuccessful

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