

# Treatment of uterine artery pseudoaneurysm by embolization or hysteroscopy combined with laparoscopy: two case reports

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## Summary

Uterine artery pseudoaneurysm (UAP) is a rare but potentially life-threatening condition when it ruptures. The primary therapeutic method is embolization, which is approved by most specialists. The authors researched two case reports of UAP. By conducting Doppler ultrasonography and arteriography, two women were diagnosed with UAP after caesarean section, and then arterial embolization was performed for treatment. After embolization, the treatment was successful in one patient but failed in the other one with repeated vaginal bleeding. Therefore, laparoscopy combined with hysteroscopy was performed to the other patient, in which uterine myometrial lesions were located under hysteroscopy and resected by laparoscopy. Finally, the uterine wall defect was reconstructed by suture, and the abnormal vaginal bleeding was successfully stopped after operation. Patients with unexplained abnormal vaginal bleeding, especially after trauma in uterus, should be cared for in case of developing UAP. Embolization is the priority treatment for UAP, while laparoscopy combined with hysteroscopy could be an alternative treatment of unsuccessful embolization.

**Key words:** Uterine artery pseudoaneurysm (UAP); Embolization; Hysteroscopy; Laparoscopy; Uterine myometrial lesion resection; Abnormal uterine artery.

## Introduction

Uterine artery pseudoaneurysm (UAP) rupture has attracted more and more obstetricians' attention. The 24<sup>th</sup> edition of Williams Obstetrics describes a new concept named UAP rupture, which is a possible cause of secondary postpartum hemorrhage (PPH)[1]. PPH remains one of the major causes of maternal death and is classified as primary or secondary. Primary postpartum hemorrhage, defined as more than 500 ml blood loss from the genital tract in first 24 hours after delivery, is most commonly the result of uterine atony [2]. Secondary PPH is defined as a bleeding that occurs more than 24 hours after delivery [3], and the causes of it include infection, involution of placental bed, and retained products of conception [2, 4]. Dohan *et al.* presented an incidence of UAP of 3.06%(18/588) among patients with PPH [5]. A large study showed that the incidence of severe secondary PPH was 0.23% (n = 60/26023), and the UAP occurred in 3.3%[6]. The UAP rupture is an uncommon but important cause of secondary PPH.

After abortion or delivery, the uterine arterial wall is sometimes partly teared or injured. If surrounding tissues cover this blood accumulation and the space still has continuity with the partly injured artery, UAP forms [5, 7]. Hence, when UAP ruptures, massive bleeding may occur. At present, color Doppler ultrasonography is considered to be a convenient and simple method for the diagnosis of UAP, and MRI and CT are both helpful. While the uterine

angiography can provide information regarding the variation of vessels and the relationship between blood vessels and the lesion location, and it is considered to be the gold standard of UAP.

The treatment of the UAP depends mainly on the patients' symptoms, fertility requirement, and the location and size of UAP lesion. At present, the treatments include selective uterine arterial embolization (UAE) and conservative ones. However, there have been no reports of UAP treated by clearance of focal lesion and reconstruction of the uterine cavity under laparoscopy combined with hysteroscopy. The authors report two cases of UAP with massive bleeding, and both were treated with UAE. One of them had repeated vaginal bleeding after embolization, but it was successfully treated by uterine myometrial pseudoaneurysm resection and reconstruct uterine. The authors would like to present their experience in the treatment of the UAP and the treatment of such patients with unsuccessful embolization.

## Case Report

### Case 1

A 30-year-old woman, gravida 1 para 1, was referred to this hospital with torrential vaginal bleeding for two hours. She had delivered a child by caesarean section 31 days prior. Because her temperature reached 38.5°C, intrauterine infection was suspected. After caesarean section and antibiotic treatment, her temperature returned to normal and then she was discharged from hospital.

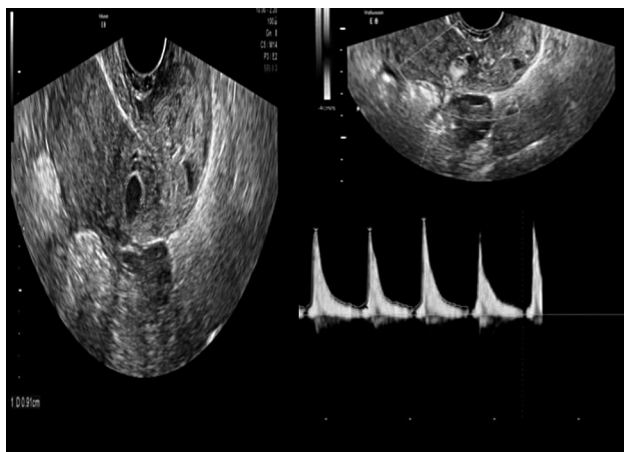


Figure 1. — Left picture is a gray scale ultrasound showing the anechoic area, measuring 8×8 mm. Right picture is a color Doppler ultrasound showing an area with swirling blood flowing with arterial waveforms.

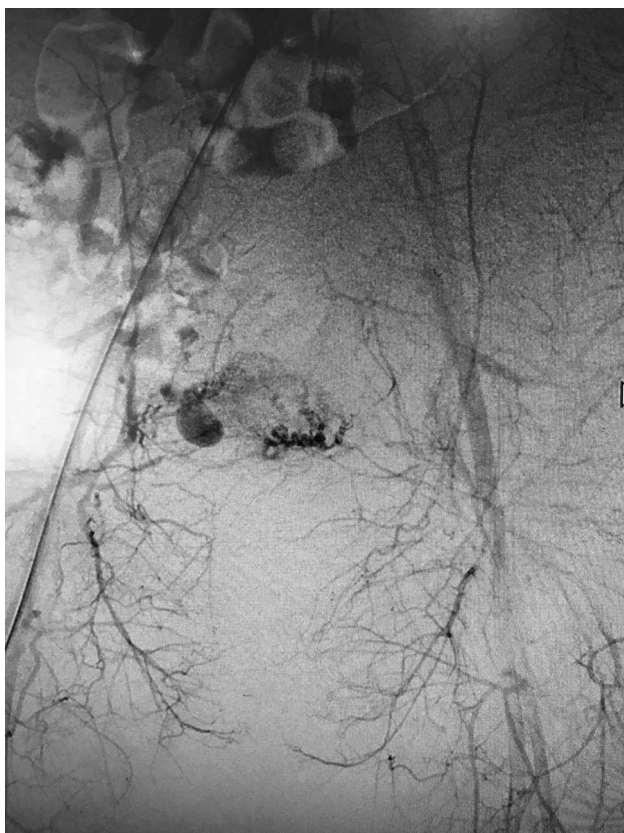


Figure 2. — DSA shows that a cystic cavity in utero is filled with extravasation of contrast material.

Postpartum she performed common breast-feeding and her menstruation did not return. An emergency ultrasound showed that the cesarean section incision had an anechoic area, which measured 8×8 mm. Color Doppler showed an area with swirling blood flow



Figure 3. — Hysteroscopy showing the bare artery with regular pulsation.

and arterial waveforms (Figure 1). She had accelerated breathing and heart rate with hemoglobin 66 g/l, then she received a transfusion of three units of concentrated red blood cells combined with injected oxytocin and hemostatics. Her vital signs of patient were stable, but the patient appeared with a continuous small amount vaginal bleeding. An emergency angiography and UAE were performed. Pelvic arteriography demonstrated the dilated right uterine arteries feeding a sac, confirming the diagnosis of UAP. Gelatin sponge was used for embolism of bilateral uterine arteries, and the patient had no significant vaginal bleeding one week after operation. The patient was followed until 12 months after discharge, and her menstruation returned to normal. Massive vaginal bleeding did not recur.

#### Case 2

A 32-year-old patient, gravid 2 para 1, visited this hospital with a history of torrential vaginal bleeding for two hours and one episode of syncope. Six months prior, she underwent cesarean section because of macrosomia. After operation, the patient had a fever for two days, with the highest temperature reaching up to 38°C, and was discharged from the hospital when her temperature returned to normal after antibiotic treatment. Postpartum she performed breast-feeding and her menstruation did not return. Urgent blood routine showed hemoglobin concentration of 94 g/l. An emergency pelvic ultrasonogram revealed that the uterine vascular area changed in the myometrium and the area measured an inner diameter of 9.1 mm. Color Doppler showed that vascular area appeared high speed with a swirl flowing signal, and Doppler spectrum prompted arterial blood flow waveform. The diagnosis of UAP was considered. After five days of conservative treatment with oxytocin and hemostatic, the patient still had a large amount of vaginal bleeding. Hemoglobin level was at 75 g/l. She received a total of two units of red blood cells. After consultation with interventional radiologist, bilateral UAE were done by gelatin sponge. Pelvic digital subtraction angiography (DSA) was performed and showed a cystic cavity filled with extravasation of contrast material indicating a rupture of blood vessel (Figure 2). Then the diagnosis of UAP was confirmed. After a bilateral UAE,

the vaginal bleeding stopped. However on the third day after UAE, the patient experienced a small amount of vaginal bleeding. Since the gelatin sponge used in arterial embolization is a temporary vascular embolization agent, UAE could fail and the patient had the possibility of further bleeding. Moreover, this patient was expected to preserve fertility. Hence, the surgical procedures including hysteroscopy was used to locate, then resect the lesions in the myometrium under laparoscopy. Hysteroscopy showed the bare artery with regular pulsation near to the right side of intracervical mouth, and the lesions measured about 1×1cm (Figure 3). Then switch to laparoscopy showed that the shape of uterus was normal. Laparoscopy exposed the UAP lesions located by hysteroscopy. After the lesions were removed and parametrial vessels were occluded, uterus was reconstructed with sutures. After applying hysteroscopy again, the bare vessels near the intracervical mouth disappeared, and there were no intraoperative complications. After the operation, two-dimensional ultrasonography and color Doppler ultrasound did not show any cavity change or vessels blood sign in myometrium. The patient recovered well after operation and normal menstruation returned one month later. Within 12 months' follow-up after operation, the women did not have any massive bleeding, and the menstrual cycle became regular and normal.

## Discussion

Uterine vascular abnormalities, including UAP, uterine artery malformation (UAM), or uterine artery fistula (UAF) occur after surgical trauma, including caesarean section, dilatation and curettage, and other pelvic operations. Since pseudoaneurysm is not surrounded by arterial wall, it differs from the true aneurysm [8, 9]. However, UAP is considered to occur not only after trauma like cesarean section but also after non-traumatic vaginal delivery [10]. The mechanism of UAP occurrence is not clear. Some authors consider that is not definite to distinguish between delivery associated or surgery associated UAP, whereas hemodynamic change during delivery and pregnancy may cause UAP [11]. Under sustained arterial pressure, blood can strike the wall around the damaged artery, resulting in generating a sac which communicates with the arterial lumen. Extravasating blood can establish a sac in myometrium, leading to the formation of UAP and resulting in second PPH. Matsubara *et al.* assessed the infection which caused pseudoaneurysm rupture and resulted in massive bleeding [1]. In the present case, both patients underwent cesarean section and had a fever before or after it. It maybe testify that the delivery itself, trauma, and infection are the significant causes of UAP.

At present, some non-invasive diagnostic methods have been proposed to diagnose uterine vascular abnormalities, such as color Doppler ultrasound, CT, and MRI. However, pelvic angiography is considered the gold-standard diagnostic method [5, 8]. With the development of color Doppler ultrasound, it is more and more used for diagnosing UAP. Some patients without abnormal haemorrhage were diagnosed UAP by just by color Doppler ultrasound. Typical findings are that the cystic area is filled with a tur-

bulent and multidirectional blood flow pattern with artery waveform, and the peak systolic velocity in the feeding artery is high [12]. MRI and CT can provide the location and accurate data about the size of the lesion. Pelvic angiography not only indicates the location, but also confirms the original artery of lesion, and especially shows extravasation of contrast material [6]. In the present cases, the diagnosis of UAP was firstly suspected by color Doppler ultrasound and then confirmed by angiography.

Treatment of UAP is based on clinical symptoms, the patient's age, and fertility desire. Not all patients who are diagnosed with UAP require surgical treatment. Hironori *et al.* reported that three cases with small UAP with lesions varying from 10-15 mm and with no clinical symptoms, and that spontaneously resolved without any treatments. One of the three patients became pregnant again, and the UAP recurred and spontaneously resolved once again [10, 13]. Baba *et al.* proposed that UAP sac disappearance without surgery treatment maybe because of thrombosis in the UAP sac [14]. Considering the risk of instable haemodynamic, indications of conservative treatments for UAP should be held seriously.

The optimal treatment is the selective UAE, because it is highly effective for controlling haemorrhage and avoiding potential complications of open surgery [7, 15]. Several agents used for embolization are gelatin sponge, polyvinyl alcohol particles, metallic coils or N-butyl cyanoacrylate (NBCA), which are divided into temporary or permanent embolization substances. A few cases reported that the second embolization and the failure of embolization usually occurred at the time of absorption of the embolization substance, abundant collateral circulation and revascularization [6, 16, 17]. Some studies reported complications of UAE, such as pelvic pain, nausea, generalized malaise, low-grade fever, mild leukocytosis, and a moderate rise of C-reactive protein. After embolization, the reduction of uterine blood perfusion could be one of the reasons for transient or permanent amenorrhea, abortion and infertility [18]. In the present two cases, both of them underwent bilateral UAE, one patient was successful for hemostasis but the other had failed.

Traditional surgical methods for UAP include internal iliac artery ligation and hysterectomy. After internal iliac artery ligation, the success rate to hemostasis is uncertain because of the collateral circulation, however, there is a successful report about it [19].

Compared with traditional surgical methods, laparoscopy and hysteroscopy are the minimally invasive methods. Pamplona *et al.* reported the possibility of UAP rupture without vaginal bleeding because the injured artery was positioned away from the endometrium [20]. The present authors thought that the UAP rupture with vaginal bleeding had the lesions near the endometrium. There is a report regarding the use of hysteroscopy to identify the uterine vascular abnormalities and to locate the lesion of uterine artery



malformation [21]. In addition, some authors treat the uterine vascular abnormalities by laparoscopy [18]. In the present second case, the patient underwent UAE but failed, and she and her family refused the second UAE. With the permission of the patient and her family, the authors treated the patient by hysteroscopy combined with laparoscopy. Firstly under the hysteroscopy, blood vessels and arterial pulsation can be clearly seen, including the position of the lesion. Then the lesion was removed and the uterine cavity was reconstructed by laparoscopy. Finally the authors used hysteroscopy to further checking and did not observe any lesion.

In conclusion, the treatment of patients with UAP should vary according to various circumstances. UAE is a good therapeutic option for controlling haemorrhage, however, hysteroscopy combined with laparoscopy can be suitable for re-extravasation bleeding after embolization. This method is minimally invasive, effective, and can retain uterine integrity, which is in line with precision medicine.

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