



## Case report

## Embolization of iatrogenic uterine pseudoaneurysm



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## ABSTRACT

Uterine artery pseudoaneurysms (UAPs) are rare vascular lesions that may be life threatening if not diagnosed and properly treated. The clinical presentation of UAPs includes a spectrum of symptoms that are often associated with other and more frequent gynecologic/obstetric pathologies, both with and without vaginal bleeding, and may span from postpartum hemorrhage to the absence of symptoms. We report cases of two patients with UAP, both of whom were diagnosed with ultrasonography and contrast-enhanced computed tomography and successfully treated with transcatheter embolization. The first patient presented delayed hypovolemic shock following surgery for endometriosis, whereas the second patient suffered from postpartum hemorrhage after cesarean section. Diagnosis of UAPs relies on noninvasive imaging; transcatheter arterial embolization is an effective treatment to control bleeding in both hemodynamically stable and unstable patients.

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## Introduction

Uterine artery pseudoaneurysm (UAP) is a rare and severe vascular anomaly resulting from inadequate sealing of a lacerated wall of a uterine artery; it accounts for approximately 3% of cases of postpartum hemorrhage.<sup>1</sup> The aneurysmal sac is typically walled by a single soft tissue layer and sustained by the arterial blood pressure; its rupture is unpredictable and represents a major complication.<sup>1</sup>

UAPs mainly occur after traumatic delivery or traumatic pregnancy termination, comprising 47% of cases of cesarean section, manual removal of placenta, forceps delivery and vacuum extraction, and cold knife conization; other recognized causes of UAPs include hysterectomy and myomectomy.<sup>1,2</sup>

A UAP is rare but not negligible cause of delayed or secondary postpartum hemorrhage<sup>1,2</sup>; a massive uterine hemorrhage from a UAP may require an emergency laparotomy unlike other conditions which may be conservatively treated.<sup>3</sup>

It is important to outline that a UAP may also show atypical clinical features that may be deceiving to obstetrics facing an

emergency.<sup>4,5</sup> As previously stated, a UAP may be either asymptomatic or present with symptoms such as vaginal bleeding, abdominal pain, hypovolemic shock, or fever when infected.<sup>4,6,7</sup> Such a vast array of symptoms can make it difficult to diagnose a UAP, especially in case of a nontraumatic delivery, abortion, or pregnancy termination.<sup>8</sup> The wide spectrum of symptoms associated with a UAP that mimic other conditions makes it a “chameleon” pathology; as a result, UAPs should always be considered in differential diagnoses.<sup>4,5</sup>

In this context, contrast-enhanced computed tomography (CT) should always be considered in diagnostic work up, as it plays a key role in determining digital subtraction angiography (DSA) indication.

Traditional surgical management of UAPs includes revision with packing, bilateral internal iliac, or uterine artery ligation, and, when other treatments fail, hysterectomy. Transcatheter arterial embolization has recently emerged as a safe and highly effective alternative treatment.<sup>1,2</sup>

We report cases of two patients with UAPs, both of whom were successfully treated with transcatheter embolization. In the first case, the patient developed an extrauterine pseudoaneurysm without vaginal bleeding following pelvic surgery for endometriosis, whereas in the second case, the patient experienced a postpartum hemorrhage after Cesarean section.

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## Case Reports

### Case 1

A 34-year-old woman diagnosed with pelvic endometriosis underwent right oophorectomy, left salpingectomy, sigmoid resection, and excision of an endometrioma in the Douglas pouch. Seven days after surgery, the patient suffered from acute abdominal pain and hypovolemic shock without any vaginal bleeding; the patient was transferred to the intensive care unit and stabilized.

A prompt contrast-enhanced CT of the abdomen revealed a left extrauterine UAP measuring 5 cm × 4 cm in diameter (Figure 1A); the diagnosis was thereafter confirmed with an abdominal DSA of the lumbar aorta and a selective catheterization of the left hypogastric artery (Figure 1B).

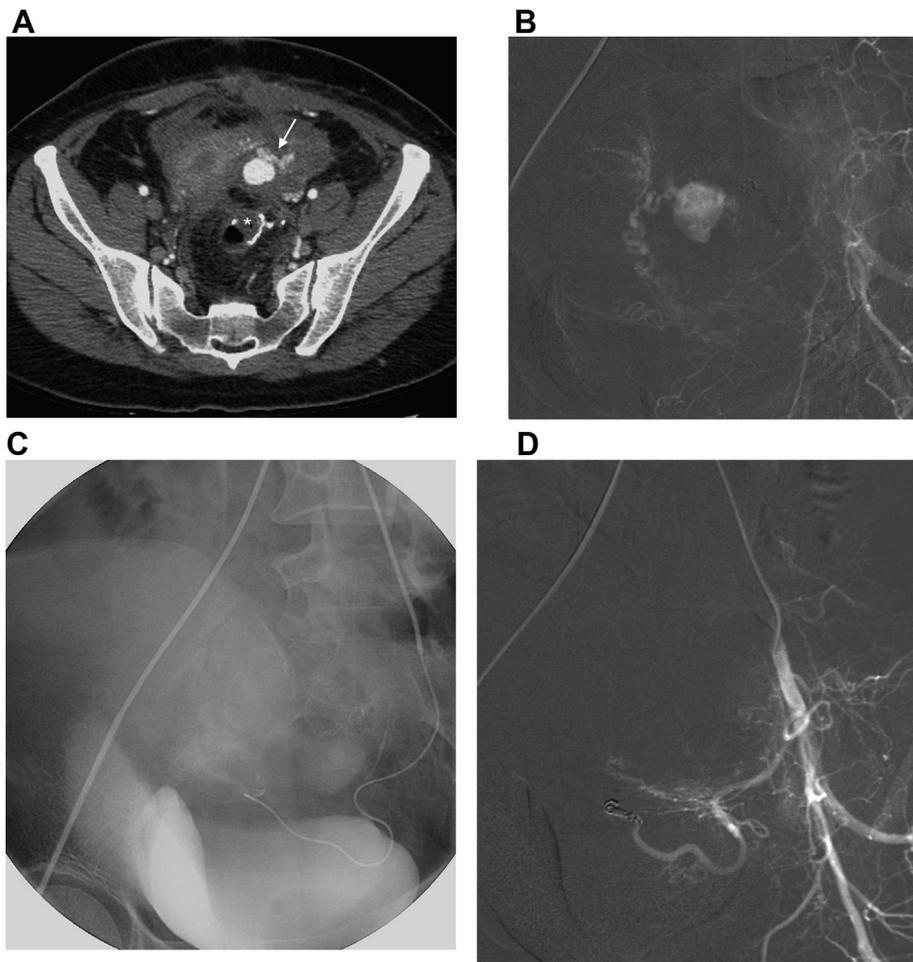
A superselective catheterization of the left uterine artery was performed for embolization with a coaxial microcatheter, the tip of which was advanced as close as possible to the UAP. Unfortunately, because of the rupture of the UAP during vessel negotiation, the patient became hemodynamically unstable, as demonstrated by an immediate arteriography (Figure 1C). Emergency intubation, fluid resuscitation, and a blood transfusion were immediately performed. Vascular occlusion was thereafter accomplished with

polyvinyl alcohol particles (710–1000 μm in size) for distal embolization and with platinum microcoils for proximal embolization.

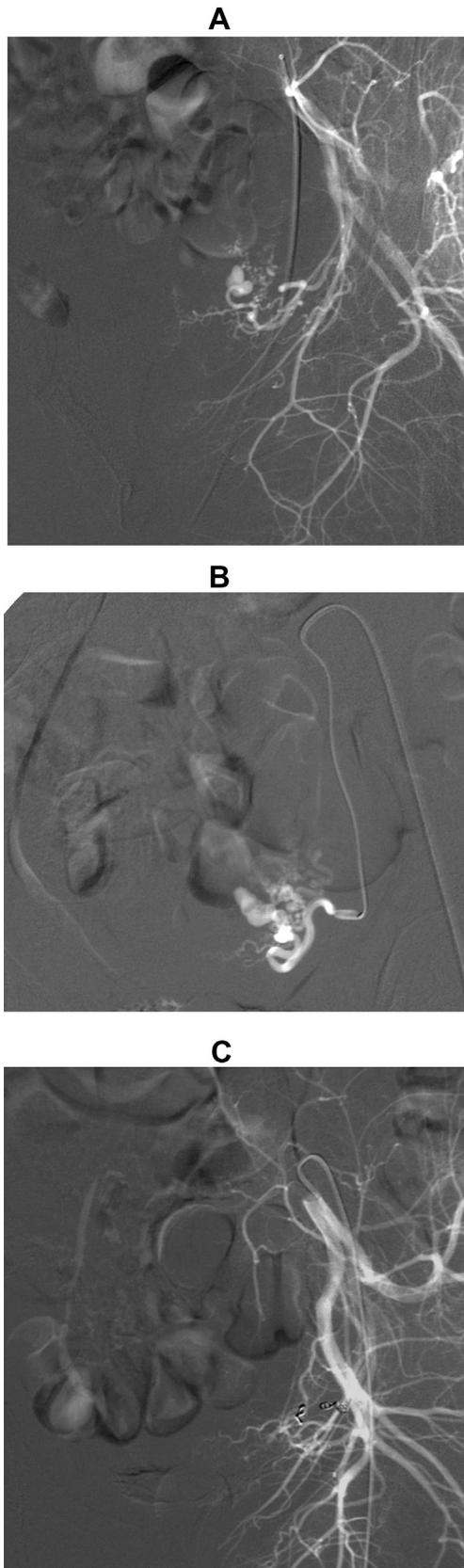
Completion postprocedural DSA of both internal iliac arteries demonstrated the occlusion of the aneurysm and of the supplying artery (Figure 1D). The procedure was uneventful, and the patient rapidly recovered after the embolization and was discharged 1 week later. After an 18-month follow-up period, the patient was in good health. Serial transabdominal and transvaginal pelvic ultrasonographic examinations did not detect any signs of any further issue.

### Case 2

A 21-year-old woman was referred to the Department of Radiology, San Bortolo Hospital for treatment for a UAP that was diagnosed in another hospital. The patient was hospitalized 1 month after a cesarean section for a metrorrhagia not controlled with a conservative treatment. A transvaginal ultrasonographic examination revealed an inhomogeneous fluid collection within the myometrium near the cesarean scar; the following arterial CT detected an intramural pseudoaneurysm measuring 1.5 cm in diameter. The diagnosis was then confirmed with DSA of the left internal iliac artery, performed via the ipsilateral femoral artery (Figure 2A).



**Figure 1.** A 34-year old woman with hemorrhagic shock after pelvic surgery for endometriosis. (A) Contrast-enhanced computed tomography shows a pseudoaneurysm close to the left aspect of the uterus (arrow: enlarged left uterine artery; asterisk: circular stapling of sigmoid resection). (B) Digital subtraction angiography of the left internal iliac artery (left anterior oblique view, late phase) confirms the diagnosis of extrauterine pseudoaneurysm supplied by the left uterine artery. (C) Native X-ray direct image of the pelvis after manual superselective arteriography: intraperitoneal diffusion of the contrast medium without evidence of the pseudoaneurysm. (D) Postembolization digital subtraction angiography shows the persistent occlusion of the pseudoaneurysm.



**Figure 2.** A 21-year old woman with postpartum hemorrhage. (A) Digital subtraction angiography of the left internal iliac artery (left anterior oblique view) shows a uterine pseudoaneurysm. (B) Superselective catheterization of the left uterine artery. (C) Sealing of both the pseudoaneurysm and the feeding artery is demonstrated by arteriographic control after embolization.

Superselective embolization was achieved by employing a microcatheter with polyvinyl alcohol particles (710–1000  $\mu\text{m}$  in size) and two platinum microcoils. The successful occlusion of the UAP was demonstrated by ipsilateral and contralateral hypogastric arteriographies (Figure 2B). The patient is currently in good condition; no vaginal bleeding recurred during the following year. Serial ultrasonographic investigations that followed were normal.

### Discussion

Bleeding from a UAP is unpredictable and may occur at variable time intervals, which could range from weeks to months after its development<sup>1</sup>; furthermore, UAPs may sometimes be clinically asymptomatic and detected incidentally.<sup>4</sup>

UAPs may in some instances rupture within the uterine cavity and thus cause vaginal bleeding.<sup>7–9</sup> In some cases, the rupture involves the peritoneal surface of the uterus only, the hemorrhage may be confined to the abdominal cavity.<sup>7</sup> In the latter case, diagnosis of an intraabdominal hemorrhage should be clinically suspected when sudden abdominal pain and collapse occur. Rupture of a UAP is unpredictable, and conservative management should thus be ruled out in therapeutic protocols. Risk of bleeding is usually directly proportional to both the size of the pseudoaneurysm and to internal sac pressure according to Laplace's law.<sup>10</sup> Moreover, the increased postpartum uterine tone, a physiologic mechanism for controlling the bleeding, may likely contribute to the breaking of a UAP.

Due to the wide spectrum of clinical presentation,<sup>4–8</sup> the differential diagnosis of postpartum hemorrhage should always consider a UAP.<sup>2</sup>

Because of their high sensitivity in demonstrating blood flow characteristically swirling within an anechoic sac-like structure, ultrasonography, and color Doppler sonography are well established and effective tools for diagnosing UAPs,<sup>1,2</sup> although it must be noted that UAPs may show complex sonographic pattern that may lead to a misdiagnosis.<sup>7</sup> Color Doppler ultrasonography also suffers from limitations in detecting the vessel supplying the pseudoaneurysm, whose identification affects the treatment planning.<sup>9</sup> Contrast-enhanced CT is able to rule out more frequent causes of delayed postpartum hemorrhage and identify a UAP<sup>1,2</sup>; moreover, because of the possibility of image reformations, CT angiography may depict the feeding artery.<sup>1</sup>

Angiography remains the standard reference in diagnosing a UAP and may provide a definitive treatment. Since the first case reported by Brown et al<sup>11</sup> in 1979, arterial uterine embolization has been successfully performed to control postpartum bleeding caused by atony of the uterus, placenta accreta, extrauterine pregnancy, vulvar and vaginal hematomas, and UAP. The safety and effectiveness of selective uterine artery embolization were demonstrated by many reports<sup>1–11</sup>; procedural complications are rare and include pelvic organ ischemia, sciatic and perineal nerve neuropathy, muscle pain, and postembolization syndrome.<sup>12</sup>

Despite the fact that bilateral embolization of uterine arteries has been advocated as the treatment of choice to prevent rebleeding,<sup>8</sup> unilateral embolization may be effective for a complete and persistent hemostasis.<sup>6,7</sup> Nevertheless, a contralateral internal iliac arteriography should always be performed after a unilateral embolization to avoid the possibility of reversed perfusion of the pseudoaneurysm.

A unilateral procedure has been suggested in order to preserve reproductive function<sup>13,14</sup>; however, infertility in up to 43% of cases treated with embolization have been reported by Sathe et al<sup>15</sup> in a recent systematic review on management of postpartum hemorrhage. These authors pointed out a low strength of evidence of a negative effect on fertility in women with a previous embolization

for a postpartum hemorrhage but also a greater infertility rate in this group compared with the general population. As a result, despite the high success rate of embolization in stopping bleeding, the uncertain evidence of embolization with respect to future fertility suggests that treatment decisions should be considered on a case-by-case basis and available management options.<sup>15</sup>

Embolization of the anterior division of internal iliac artery has been claimed for a fast procedure in case of emergencies.<sup>16</sup> In our experience, superselective catheterization of the uterine artery with the coaxial technique, when not prevented by an adverse vascular anatomy, is a time-effective procedure; furthermore, when particulate materials are used, this procedure may also allow achievement of the persistent occlusion of arterial cross-over anastomoses.

Coils may be used for embolization of the aneurysmal sac only, for parent artery occlusion with inflow/outflow vessel occlusion or both sac embolization and for parent artery occlusion with inflow/outflow vessel occlusion.<sup>16</sup> However, because of tortuosity of the uterine artery, very distal coil deployment is not always feasible and particles should be chosen first. Because of the risk of uterus necrosis,<sup>1</sup> larger particles should be preferred for the procedure, even in case of a ruptured UAP; additional coil embolization can be suggested to prevent the recanalization of the feeding artery.

Kovo et al<sup>17</sup> reported a case of a UAP successfully treated with ultrasound-guided percutaneous thrombin injection; a transvesical approach may alternatively be used to improve the detectability of the pseudoaneurysm and guarantee a safe puncture.<sup>18</sup> However, this technique should be restricted only to patients with a good acoustic window; moreover, contrary to superficial pseudoaneurysms with a single small neck, such as those involving the femoral artery, percutaneous treatment of a high-flow UAP may be questionable as it may neither guarantee the complete occlusion of the aneurysmal lumen nor prevent an inadvertent embolization of the injected material.

In order to prevent the UAP rupture that may be caused by direct injury by the guide-wire or by excessive intra-arterial pressure during contrast medium injection, the interventional radiologist must be very careful during the angiographic maneuver. In case of a UAP rupture during an arteriography, immediate embolization may save the patient's life.

In conclusion, transcatheter embolization is an effective and safe treatment for UAPs for both hemodynamically stable and unstable patients, and should be considered as the treatment of choice both because of its high success rate and because of its low complication rate. Due to the unanswered questions related to fertility impairments following pelvic embolization, elective embolization of a

UAP should be considered on a case-by-case basis only in the young female population.

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