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Recurrent uterine artery pseudo-aneurysm requiring repeat embolization during pregnancy – A case report

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ABSTRACT

Background: Uterine artery pseudo-aneurysms (UAP) mainly occur after traumatic delivery or traumatic pregnancy termination. A UAP can be either asymptomatic or present with symptoms such as vaginal bleeding, abdominal pain, hypovolemic shock, or fever when infected. We describe a patient diagnosed with a uterine artery pseudo-aneurysm in pregnancy that required repeat embolization. The patient had no risk factors in her recent medical history. She did, however, undergo in-vitro fertilization with oocyte retrieval performed under transvaginal ultrasonographic guidance. We suggest the hypothesis of formation of the pseudo-aneurysm secondary to vascular injury during oocyte retrieval.
Case Report: A 35-year-old primigravida, who conceived by in-vitro fertilization, presenting with severe abdominal pain at 19 2/7 weeks of gestation. Ultrasound examination with color doppler imaging revealed a hypoechoic lesion with turbulent arterial flow pattern on the lower left side of the uterus. Selective catheterization and subtraction angiography permitted diagnosis of a large pseudo-aneurysm of the left uterine artery. A selective embolization was performed. Recanalization of the embolized artery was confirmed 11 weeks after initial presentation, requiring repeat embolization. A planned caesarean section was performed at 34 weeks of gestation and a healthy boy was born with a birth weight of 2065 g.
Conclusion: Uterine artery pseudo-aneurysm is a rare but potentially life-threatening condition. It can be diagnosed using (doppler) ultrasound, revealing a hypoechoic mass with swirling blood flow. Angiography is the standard reference in diagnosing UAP and may provide definitive treatment. Management with selective unilateral uterine artery embolization appears to be safe in hemodynamically stable patients. It does not compromise uteroplacental circulation and may help to prolong the pregnancy, reducing morbidity associated with preterm birth.

1. Background

A pseudo-aneurysm results from inadequate sealing of a puncture or laceration of the arterial wall, leading to extravasation of blood into periarterial tissues which forms a perfused sac that communicates with the arterial lumen [9,11]. Rupture of a pseudo-aneurysm is unpredictable and represents a major complication. Uterine artery pseudo-aneurysms (UAP) mainly occur after traumatic delivery or traumatic pregnancy termination. Other recognized causes of UAPs include hysterectomy, myomectomy, and cold knife conization [1]. An UAP can be either asymptomatic or present with symptoms such as vaginal bleeding, abdominal pain, hypovolemic shock, or fever when infected.

We report a case of a patient diagnosed with UAP who required repeat embolization in pregnancy.

2. Case Report

A 35-year-old primigravida consulted her obstetrician at 19 2/7 weeks of gestation having experienced mild abdominal pain for a few days. Her recent medical history included a diagnostic laparoscopy because of abdominal pain which revealed a rupture of a hemorrhagic corpus luteal cyst of the left ovary. This pregnancy was conceived by in-vitro fertilization (IVF). Further medical history was unremarkable. Clinical and ultrasound examinations revealed no abnormalities. She was treated with analgesics. The next day, she came to the obstetric department because of sudden onset of severe lower abdominal pain. She presented with nausea, vomiting, diarrhea, and an urge to move. There was no vaginal bleeding. She had a normal blood pressure of 129/82 mmHg and a heart rate of 79 beats per minute. On bimanual palpation, the uterus was hypertonic and painful, and a para-uterine mass

Abbreviations: UAP, Uterine artery pseudo-aneurysm; IVF, in-vitro fertilization; MRI, magnetic resonance imaging.

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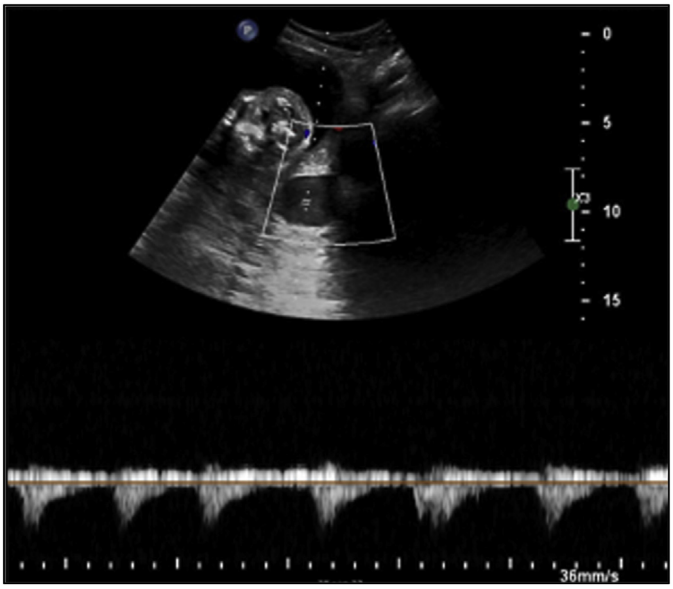

Fig. 1. Ultrasound examination demonstrated flow in the hypo-echoic lesion.

was palpated. The laboratory results showed: hemoglobin 12.3 g/dL, white blood cell count 17.3 × 103/mm, and C-reactive protein 27.4 mg/L. Transabdominal ultrasound showed a normal biophysical profile and no signs of placental abruption. Transvaginal ultrasound with color doppler imaging revealed a hypo-echoic lesion of 3.4 cm on the lower left side of the uterus with a turbulent arterial flow pattern inside the lesion (Fig. 1).

An ovarian (sub) torsion was suspected, and patient underwent diagnostic laparoscopy, during which no abnormalities were observed.

Pain persisted after laparoscopy and the patient underwent emergency magnetic resonance imaging (MRI), which revealed a nodular mass with active bleeding at the level of the cervix (Fig. 2). A pseudo-aneurysm of the uterine artery was suspected.

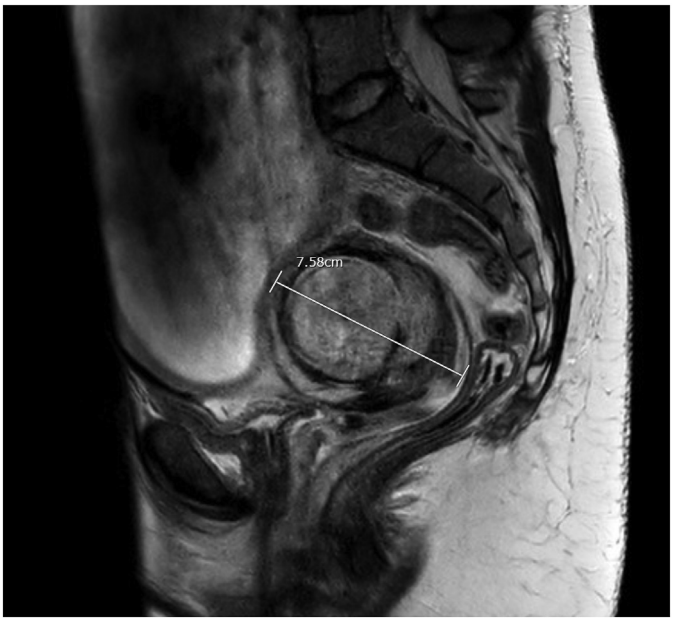

Fig. 2. MRI image (T2) of pseudo-aneurysm before embolization.

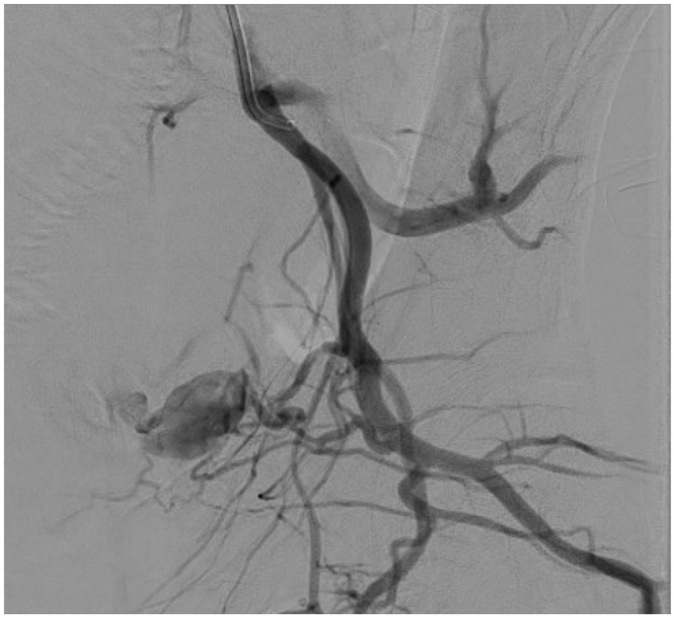

Fig. 3. Subtraction angiography of the left internal iliac artery revealing a large pseudo-aneurysm of the left uterine artery.

Selective catheterization and subtraction angiography of the left internal iliac artery permitted diagnosis of a transection and large pseudo-aneurysm of the left uterine artery (Fig. 3).

A selective embolization of the pseudo-aneurysm was performed. Microsphere particles (Embosphere® microspheres of 700–900 μm diameter) and gelfoam provided insufficient embolization. Complete obliteration of the pseudo-aneurysm was achieved using 3 coils (Complex Helical 4x4mm, Boston Scientific). Angiographic control showed complete occlusion of the left uterine artery (Fig. 4).

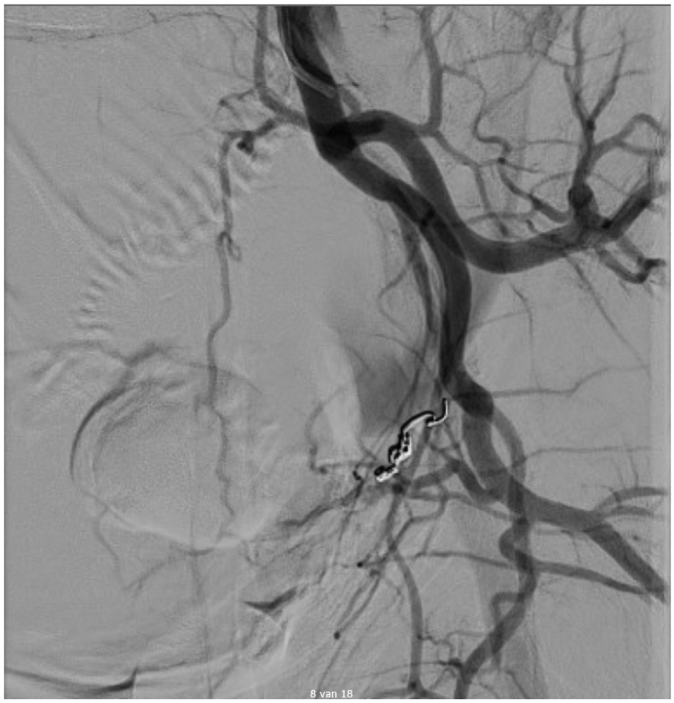

Fig. 4. Pseudo-aneurysm of the uterine artery after coiling.

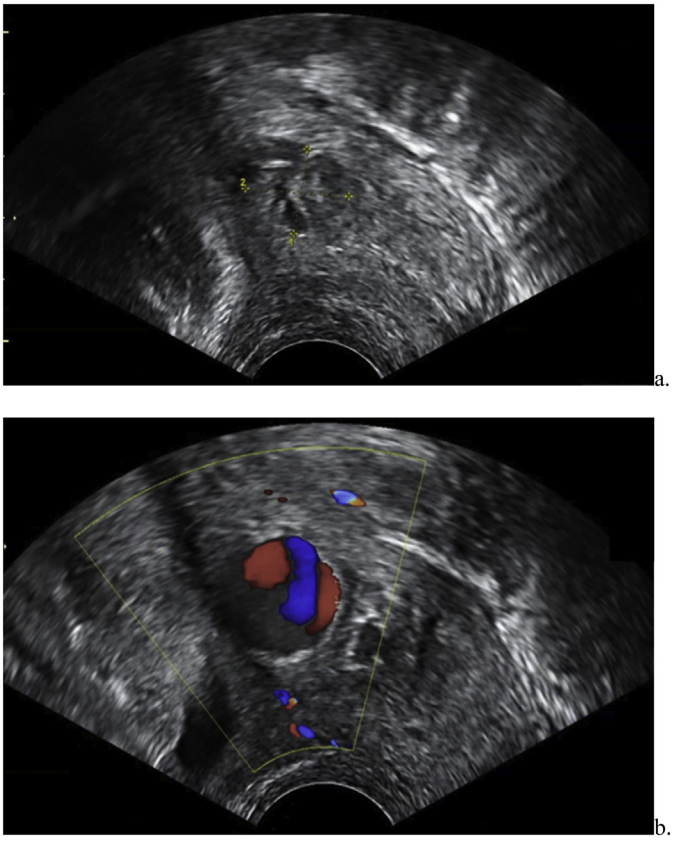

Fig. 5. Coiled uterine artery pseudo-aneurysm (5.a.) inferior to a more superior located para-cervical mass (5.b.) (1.5 × 1.9 cm) with turbulent arterial flow.

Ultrasound examination with color doppler imaging after the procedure showed a hematoma without flow, measuring 5.1 × 5.9 cm. The patient left the hospital 7 days after the procedure. She had regular follow-up in the antenatal clinic.

Five weeks later, at 25 4/7 weeks of gestation, the patient presented with vaginal bleeding. Transvaginal ultrasound revealed a cervical length of 46 mm and a stable non-pulsatile coiled UAP of 3.2 × 2.6 mm. Due to persistent complaints of contractions, the patient received tocolytic drugs for 48 h to allow for administration of antenatal corticosteroids. Blood loss decreased and patient left the hospital 5 days later.

At 30 weeks of gestation, the patient presented with increasing lower abdominal pain and vaginal blood loss. Transvaginal ultrasound with color doppler imaging showed the coiled UAP and a more superior para-cervical lesion (1.5 × 1.9 cm) with turbulent arterial flow (Fig. 5). Imaging, however, did not confirm the diagnosis of recurrent UAP. Because of the discrepancy between MRI and ultrasound, angiography was repeated, during which a recurrent UAP, caused by recanalization, was confirmed (Fig. 6). The recurrent UAP was embolized with glue (Glubran® 0.8 mL). Complete occlusion was confirmed on further angiography.

Because of the complicated course of the pregnancy, persistent pain, and risk of recurrence, a planned caesarean section was performed at 34 weeks of gestation. A caesarean section was opted for to prevent high intra-abdominal pressure during labor and delivery. A healthy boy was born with a birth weight of 2065 g (Apgar 8/10 one minute and 9/10 five minutes after birth). The boy experienced no neonatal morbidities. He was discharged from the neonatal intensive care unit 18 days later. Maternal recovery was uneventful.

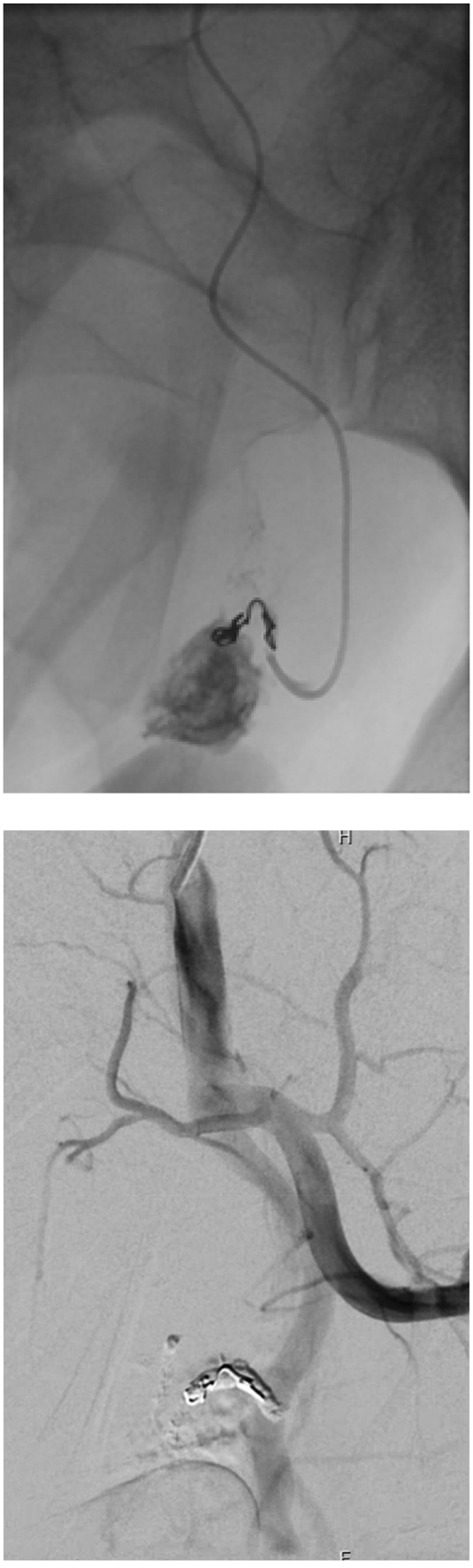

Fig. 6. Recurrent uterine artery pseudo-aneurysm before (up) and after (down) embolization.

3. Discussion and Conclusion

Uterine artery pseudo-aneurysms have been described as a consequence of caesarean section, dilatation and curettage, hysterectomy, myomectomy, and vaginal delivery. [2]

The patient in this case had none of these risk factors in her medical history. She did, however, undergo an IVF procedure with oocyte retrieval performed under transvaginal ultrasonographic guidance. This procedure is known to have a very low complication rate. Vascular, gastrointestinal and genitourinary injuries are extremely rare. To our knowledge, only two cases of pseudo-aneurysm of pelvic vessels after oocyte retrieval have been reported [3,4]. Both reported cases of pseudo-aneurysms after an IVF cycle had been diagnosed during pregnancy. Hormonal, hemodynamic and mechanical changes during pregnancy might contribute to the diagnosis during pregnancy [3,4,10]. The patients with UAP diagnosed during pregnancy described in the case reports by Laubach et al. [5] and Amana et al. [6] also became pregnant through IVF; however, the authors found this origin to be less probable because of the past medical history of their patients.

Failure of uterine artery embolization can be multifactorial. Potential causes include recanalization of embolized arteries, incomplete embolization, collateral uterine arterial supply, disseminated intravascular coagulation, arterial vasospasm and vasodilatation. [7] In the present case, a recanalization of the embolized artery was confirmed.

To our knowledge this is the first reported case of recanalization of a UAP requiring repeat embolization during pregnancy. Radiation dose risk for mother and fetus and the need for diagnostic or therapeutic imaging should be weighed in an individualized risk assessment. Due to the paucity of data there is no evidence supporting the ideal timing and mode of delivery in the current pregnancy, or the management of subsequent conception. If another IVF cycle is planned, caution should evidently be exercised to avoid vascular injury during oocyte retrieval [3].

Uterine artery pseudo-aneurysm is a rare but potentially life-threatening condition. It can be diagnosed using (doppler) ultrasound, revealing a hypoechoic mass with swirling blood flow. Angiography is the standard reference in diagnosing UAP and may provide definitive treatment. Management with selective unilateral uterine artery embolization appears to be safe in hemodynamically stable patients. It does not compromise uteroplacental circulation and may help to prolong the pregnancy, reducing morbidity associated with preterm birth [2,8].

Contributors

Astrid Mulkers was involved in the study design and manuscript preparation and revision.

Kathleen Podevyn was involved in manuscript revision.

Isabelle Dehaene was involved in the study design and manuscript preparation and revision.

All authors read and approved the final manuscript.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

Obtained.

Provenance and Peer Review

This case report was peer reviewed.

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