CASE REPORT

Uterine artery pseudoaneurysm with an anastomotic feeding vessel requiring repeat embolisation

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SUMMARY

Uterine artery pseudoaneurysm (UAP) is a rare cause of delayed postpartum haemorrhage. Early diagnosis and endovascular management are effective in treating this condition. We present the case of a 36-year-old gravida 3, para 2 woman with delayed postpartum haemorrhage and endometritis following a spontaneous vaginal delivery. Ultrasound and catheter angiogram demonstrated a UAP arising from the distal aspect of the left uterine artery. Significant bleed persisted despite selective bilateral uterine artery embolisation. A repeat angiogram confirmed complete occlusion of bilateral uterine arteries, but abdominal aortogram demonstrated that the left ovarian artery was now feeding the pseudoaneurysm. A repeat embolisation procedure was performed to occlude the left ovarian artery. The patient was discharged the following day. Selective arterial embolisation is effective in the management of UAP. Persistent bleeding despite embolisation should raise the suspicion of anastomotic vascular supply and may require repeat embolisation.

BACKGROUND

Postpartum haemorrhage (PPH) is a potentially life-threatening condition that accounts for 25% of maternal deaths worldwide.¹ Delayed PPH is defined as excessive vaginal bleeding occurring between 24 hours after delivery until 6 weeks post partum.^{2–3} Common causes of delayed PPH include retained products of conception, endometritis, subinvolution or involution of the placental bed, arteriovenous malformation and choriocarcinoma.^{4–5}

Uterine artery pseudoaneurysm (UAP) is a rare cause of delayed PPH. Its incidence is estimated to be around 3%.⁶ A UAP is an extraluminal collection of blood that communicates with the main vessel through a defect in the arterial wall.⁷ It can mimic and mask other entities, such as adnexal masses,⁸ septic abortion⁹ or endometritis, as we have reported here. It is thought that UAP develops as a complication of vascular injury resulting from inflammation, trauma or iatrogenic procedures.^{10–11}

CASE PRESENTATION

A 36-year-old gravida 3, para 2 woman was admitted to our institution on postpartum day 9 with a delayed PPH and endometritis. She had a spontaneous vaginal delivery at term with manual removal of the placenta. Estimated blood loss at delivery was average, and she did not experience any significant

bleeding during her postpartum hospital stay. She had previously undergone a manual removal of the placenta following a pregnancy termination at 18-week gestation for trisomy 18. On postpartum day 9 of this current pregnancy, she returned to hospital with increasing vaginal bleeding, subjective fevers and chills, a tender uterus, sonographic evidence of air within the endometrial cavity and a white cell count of $17.1 \times 10^9/L$. The diagnosis of postpartum endometritis was made, and patient was started on intravenous antibiotics.

INVESTIGATIONS

Ultrasound showed a $1.9 \times 1.4 \times 1.7$ cm anechoic area within the endometrium. Colour Doppler demonstrated a swirl of colours (yin-yang sign), and spectral Doppler showed the typical 'to and fro' pattern consistent with UAP. This was surrounded by a heterogeneous area containing multiple air locules, suspicious for retained products/endometritis (figure 1). The patient's haemoglobin dropped acutely from 9.3 to 7.5 g/dL in the first few hours following admission and she was deemed too unstable for the MRI that was planned to fully delineate the above-mentioned intrauterine lesion. The patient was resuscitated with intravenous fluids and emergent angiography was carried out with the intent of performing bilateral uterine artery embolisation. Selective uterine artery angiogram demonstrated a 1×1 cm pseudoaneurysm arising from the distal aspect of the left uterine artery (figure 2).

TREATMENT

Selective bilateral uterine artery embolisation was performed using gelfoam slurry—a non-permanent embolisation material (figure 3). The patient's blood pressure and coagulation factors were normal prior to her embolisation procedure. Although no transfusion was required, the patient continued to bleed on the day following the embolisation procedure. An MRI of pelvis with contrast was performed to assess the uterus and endometrium. This surprisingly revealed a persistently enhancing UAP, thereby suggesting recanalisation of the uterine artery (figure 4). A repeat angiogram, however, showed that both the uterine arteries were still completely occluded (figure 5). Abdominal aortogram was then performed, and it demonstrated that the left ovarian artery was now feeding the UAP and preventing its thrombosis (figure 6). Therefore, the left ovarian artery was selectively embolised; this led to a complete thrombosis of the UAP (figure 7).

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Figure 1 (A) Transabdominal ultrasound with colour Doppler demonstrates an anechoic oval structure projecting into the uterine endometrial cavity. (B) Colour Doppler mode reveal a 'yin-yang sign' in keeping with an arteriovenous malformation.

OUTCOME AND FOLLOW-UP

Following the second embolisation procedure, the patient's bleeding slowed substantially, and she was discharged on postprocedure day 2. She continued to have a healthy postpartum course thereafter and was well at her 6-week follow-up appointment.

DISCUSSION

Selective arterial embolisation is an effective method for the treatment of arterial pseudoaneurysms. However, embolisation failures may still occur in the presence of an alternate feeding vessel. We report a case of UAP after vaginal delivery which required repeat embolisation due to an anastomotic feeding vessel originating from the left ovarian artery.

Brown *et al* first reported its use in 1979 for the treatment of an extrauterine pelvic haematoma after three failed surgical attempts to control the bleeding.¹² The procedure has since been widely used for management of arterial pseudoaneurysms.¹³ There have been several case reports of UAP requiring embolisation during pregnancy,¹⁴ following abortions,¹⁵ traumatic deliveries or caesarean sections,^{10 13} cervical conisation,¹⁶ myomectomies¹⁷ and hysterectomies.¹⁸ It is thought that traumatic procedures may cause injury to the uterine

artery wall, leading to extravasation of blood into periarterial tissues, thereby creating a perfused sac. This sac then communicates directly with the arterial lumen and forms a pseudoaneurysm.¹⁵ Baba *et al* retrospectively analysed data involving 50 women with angiographically proven UAP and suggested that all patients with an actively bleeding UAP warranted immediate embolisation.¹⁹ Arterial embolisation can be performed using gelatin sponge alone, or in addition to metallic coils or *n*-butyl-2-cyanoacrylate.⁶ There is no evidence on the best type of embolisation substance to use. The choice is therefore at the discretion of the interventional radiologist. Despite potential complications of arterial embolisation including fever, ischaemia, transient to permanent ovarian failure, bladder wall necrosis, vaginal fistula, muscle pain, neurological damage and perforation of surrounding vessels, uterine artery embolisation is effective in the management of UAP and has the added advantage of preserving future fertility when compared with hysterectomy.²⁰ Alternatives to selective arterial embolisation include expectant management,¹⁹ laparoscopic uterine artery ligation with UAP excision²¹ and hysterectomy for patients who have completed their family. Expectant management should be reserved for patients with no current bleeding and no sonographic evidence of any intrauterine hypoechoic mass.¹⁹ One recent case report demonstrated the feasibility of using an intrauterine balloon tamponade test for small UAPs

Figure 2 Pelvic angiogram demonstrates a pseudoaneurysm (see arrow) arising from the distal branches of the left uterine artery.

Figure 3 Post bilateral uterine artery embolisation with gelfoam of (A) right and (B) left uterine arteries.

Figure 4 Circumscribed homogeneously enhancing intraluminal mass connected with a thin vascular pedicle to the spiral arteries consistent with a pseudoaneurysm that recanalised despite previous gelfoam embolisation. The arrow points to the pseudoaneurysm.

(<13 mm) to promote thrombosis prior to dilatation and evacuation.²² But this procedure carries potential risk of UAP rupture during balloon insertion into the uterus, particularly if the UAP is projecting into the endometrial cavity, as it was in our case.

Failure of uterine artery embolisation can be multifactorial in aetiology. Potential causes include incomplete embolisation, haemodynamic instability causing arterial vasospasm, recanalisation of embolised arteries, prominent utero-ovarian anastomosis, disseminated intravascular coagulation and arterial vasodilation caused by postembolisation intravenous fluids and blood transfusion therapy.^{23–25} The pseudoaneurysm in our report had a dual blood supply from both the

Figure 5 Persistent occlusion of previously (24 hours prior) gelfoam embolised right (A) and left (B) uterine arteries. The previously demonstrated pseudoaneurysm is no longer identified on the left (B) on selective bilateral internal iliac angiograms.

Figure 6 Pseudoaneurysm fed by the left ovarian artery arising from the left renal artery. Pseudoaneurysm successfully embolised using glue/lipiodol (1:3). (A) Origin of the ovarian artery arising from the left renal artery. (B) Left ovarian artery feeding the pseudoaneurysm via vascular anastomosis. (C) Complete occlusion of left ovarian artery and the pseudoaneurysm. The arrows in (A) and (B) demonstrate the vessels feeding the pseudoaneurysm, whereas the arrow in (C) shows the occluded pseudoaneurysm.

uterine artery and a branch of the ovarian artery. This was the reason for the failure of the primary embolisation procedure. Previous studies have reported that the primary site of arterial pseudoaneurysms is usually within the uterine arteries (75%), with other feeding vessels such as the ovarian, internal pudendal or obturator arteries being the exception rather than the norm.^{6,9,26} Generally, embolisation failures can be managed with repeat embolisation. To prevent secondary embolisations, Pelage *et al* recommended flush aortography with possible superselective ovarian arteriograms to assess for ovarian artery anastomoses.²⁷ Direct speculum examination during the initial embolisation procedure is also important to allow for early detection of any active bleeders.²⁸ A recent case report also described the use of intra-arterial nitroglycerin during embolisation for severe PPH with uterine artery vasospasm.²⁸ Barring one case report of a UAP which sourced from an anastomotic ovarian artery, the need for ovarian arteriogram during routine embolisation procedures has rarely been discussed in the literature.¹⁹

In the present case, we report a patient who required repeated embolisation due to an anastomotic feeding vessel originating from the ovarian artery. In order to prevent similar embolisation failures, we would recommend that a direct speculum examination and an angiogram of the ovarian arteries be performed at the end of routine uterine artery embolisation procedures to allow for early detection of ongoing bleeders.

Figure 7 Left renal angiogram and abdominal aortogram demonstrate complete exclusion of the left uterine artery pseudoaneurysm.

Learning points

- Selective arterial embolisation is an effective treatment for uterine artery pseudoaneurysm (UAP). However, an anastomotic feeding vessel may cause embolisation failure.
- If bleeding persists following embolisation, it is important to determine if the embolised artery has recanalised, or if another anastomotic vessel is responsible.
- A hidden anastomotic feeding vessel from the ovarian artery may cause unsuccessful embolisation of a UAP. An angiogram of the ovarian arteries should be performed at the end of uterine artery embolisation procedures to prevent embolisation failure.

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