CASE REPORT

An exceptional outcome from a rare cause of postpartum haemodynamic collapse

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SUMMARY

We present a case of a 26-year-old woman presenting with haemorrhagic shock without any overt bleeding on postpartum day 2. Work up revealed intra-abdominal haemorrhage from an unclear source in the pelvis. She was intubated, resuscitated with mostly crystalloids, and started on norepinephrine and dopamine, in a rural hospital with limited resources. She was then transferred to the closest tertiary care centre 3 h away by air. On arrival, she was hypothermic (34.9°C) and had developed severe metabolic acidosis (pH 6.89). A massive transfusion protocol was initiated. She underwent an emergency laparotomy and the origin of the active bleeding was believed to be a branch of the right internal iliac artery. Haemostasis was achieved with packing. She was subsequently taken to interventional radiology for angioembolisation of the right uterine artery as an alternative to haemostatic hysterectomy. Her final diagnosis was rupture of an extrauterine uterine artery pseudoaneurysm.

BACKGROUND

Rupture of a uterine artery pseudoaneurysm (UAP) is a rare cause of postpartum haemodynamic collapse. It most commonly occurs as a result of traumatic delivery or traumatic pregnancy termination secondary to shearing forces acting on the walls of the pelvic vessels. This results in a tear that allows blood to escape from the vessel's lumen into the surrounding tissues or adventitia. If the bleeding is contained and the vessel's lumen remains in continuity with the tear, a pseudoaneurysm forms. However, in a recent case series by Baba *et al*,¹ 7 of the 22 women diagnosed with a UAP did not have any history of trauma, leading to the conclusion that other unknown causes may contribute to their formation.

Three types of UAPs exist. The classic form is an intrauterine UAP presenting as postpartum haemorrhage. The second form of UAP is less common. It presents as a spontaneous haemoperitoneum because the UAP is located extrauterine. The diagnosis is often delayed in both types and the outcome fatal or, at a minimum, results in a haemostatic hysterectomy in women of childbearing age. Awareness of this rare disease can lead to early diagnosis and more conservative treatment options. Interventional radiology can be utilised to embolise the culprit vessel and preserve fertility, if the diagnosis is certain and the patient is relatively stable after adequate resuscitation. The third type of UAP described is that found incidentally on ultrasound (US) or colour Doppler at routine postpartum or

postabortal check-ups.¹ The natural history of this third type is unknown and prophylactic angioembolisation is controversial, given the associated risk of deep-vein thrombosis or pulmonary embolism (0.2%) and permanent amenorrhoea (3.9%).¹

CASE PRESENTATION

A 26-year-old woman, gravida 4 para 4, was transferred to a tertiary care hospital on postpartum day 2, for haemorrhagic shock. Following an unremarkable pregnancy, she had undergone a spontaneous vaginal delivery at 36 weeks gestation complicated by retained products of conception requiring manual extraction without instrumentation 4 h after delivery.

Her medical history revealed that her previous deliveries were uncomplicated. This was her third vaginal birth after a caesarean section. She previously abused alcohol and smoked cigarettes. She stopped both during the second month of this gestation. She denied any family history of collagen or vascular diseases.

She reported sudden onset right lower quadrant pain 24 h postdelivery. On physical examination, she appeared distressed with a blood pressure of 75/45 mm Hg and a heart rate of 120 bpm. Her abdomen was soft with right lower quadrant tenderness on palpation and no peritonitis. There was no vaginal bleeding or discharge.

INVESTIGATIONS

The patient's haemoglobin dropped from 83 to 49 g/L. A bedside US showed no retained products. She was transfused 4 units of packed red blood cells (pRBC) and started on clindamycin for suspected postpartum endometritis. After 16 h, she had a blood pressure of 41/31 mm Hg, heart rate of 163 bpm and haemoglobin of 77 g/L. Contrast infused CT scan of the abdomen and pelvis were performed (figures 1 and 2). She continued to deteriorate and developed decreased mental status, hypothermia (34.9°C) and metabolic acidosis (pH 6.89). She was given two doses of tranexamic acid, intubated and started on norepinephrine and dopamine infusions prior to transfer via air ambulance to the closest tertiary care centre 3 h away. She had received a total of 4 units pRBC, 500 cc of fresh frozen plasma and 13 L of crystalloids in the rural hospital.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis for a postpartum patient presenting with haemodynamic collapse and no overt bleeding includes venous thromboembolism,

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Figure 1 Abdominal CT coronal view. An intra-abdominal haematoma (26×17×11 cm) is centred over the inferior vena cava to the right of midline. Large areas of dense contrast enhancement are evident within the haematoma. The uterus is enlarged consistent with the postpartum state.

Figure 2 Abdominal CT sagittal view.

Figure 3 Angiogram of right uterine artery before embolisation. An asymmetrically hypertrophied artery is visible with at least two focal outpouchings of contrast in the distal vessel representing uterine artery pseudoaneurysms (UAPs).

amniotic fluid embolism, sepsis and uterine rupture. The rapid drop in haemoglobin, hypotension and tachycardia seen in our patient was more suggestive of postpartum haemorrhagic shock. The CT revealed contrast extravasation in the lower abdomen with an intact uterus, suggesting intra-abdominal haemorrhage. Causes of postpartum spontaneous haemoperitoneum that have been described in the literature include rupture of uterine vessels, umbilical vein, splenic artery aneurysm, splenic vein, UAP and liver (spontaneous, haematoma, HELLP syndrome, haemangioma, metastases).²

TREATMENT

Given the patient's profound shock and uncertain diagnosis, a midline laparotomy was performed. It revealed a large retroperitoneal nonexpanding haematoma. The uterus, spleen and liver were intact. The haematoma contained 2.7 L of clot. The origin of the active bleeding was suspected to be a branch of the right internal iliac artery but it was not amenable to ligation. The only surgical options were hysterectomy or attempting to pack the abdomen for haemostasis. Haemostasis was successfully achieved with packs and the patient's abdomen was closed primarily. The patient was subsequently transferred to interventional radiology. Bilateral common, external and internal iliac angiograms were performed through the left femoral artery (figure 3). Using an absorbable Gelfoam (Pfizer) slurry, the right uterine artery was embolised to stasis. A postembolisation angiogram showed no active extravasation.

OUTCOME AND FOLLOW-UP

The abdominal packs were removed on postoperative day 2, and the patient was discharged on postoperative day 7.

She was seen in follow-up 8 weeks postoperatively, and her menses had not returned. She was not breast feeding. She was most concerned with her risk of UAP recurrence and future fertility. Neither could be addressed with the current evidence.

DISCUSSION

About 150 cases of spontaneous haemoperitoneum in pregnancy secondary to rupture of the utero-ovarian vessels or uterine varicose veins have been reported.^{3–4} Of these cases, 61% occurred before labour (most in the 3rd trimester), 18% were intrapartum and 21% puerperal.² Hodgkinson⁵ published a case series, in 1952, of 75 women with utero-ovarian vessel rupture, in which three anatomical variations of bleeding were observed: rupture of vessels localised on the uterine surface resulting in intraperitoneal bleeding, rupture of vessels localised between the two leaves of the broad ligament resulting in retroperitoneal bleeding and a combination of both types.^{3–6} The consequences described in Hodgkinson's⁵ series were dramatic, with maternal mortality rates as high as 49% and rising to 76.3% for patients in labour.⁶ More contemporary studies have suggested a reduced maternal mortality rate of 3.6% and fetal mortality rates of 31%; this likely represents advances in resuscitative, anaesthetic and surgical management.

Several hypotheses on the aetiology of utero-ovarian vessel rupture exist. Veins are much more commonly reported to rupture because of their tortuous path and lack of valves in the pelvis. This results in uncontrolled dilation from the increased pressure secondary to the gravid uterus and muscular contractions during labour.² Considering the rarity of the condition, it is hypothesised that additional factors must be present to precipitate haemorrhage, such as arteriovenous malformations, underlying collagen or vascular disorders, adhesions from previous pelvic surgeries or infections, uterine fibroids and extrauterine endometriosis.^{3–7}

The most common location of vein rupture is the broad ligament (78.3%), followed by the back of the uterus (18.3%) and, finally, the front face of the uterus (3.3%).

Similar data do not exist for arterial rupture as it has been reported much less frequently. The most common cause of arterial rupture includes uterine artery aneurysms or pseudoaneurysms. UAPs are estimated to occur in 0.2–0.3% of child-bearing women.¹ Three types of presentations exist, as described previously. Our patient may have acquired a UAP after her first or second pregnancy, and it remained undiagnosed and unruptured.¹ Alternatively, she may have acquired it during her third or current pregnancy and rupture was precipitated by labour requiring manual extraction of retained products. Our hypothesis is that scar tissue formed in the pelvis secondary to her previous caesarean section causing shearing forces during manual extraction of the retained products and rupture of the acquired UAP.

If a patient is haemodynamically stable and a UAP is suspected, imaging can be used to aid in preoperative diagnosis. UAP is classically described on US as a hypoechoic mass, and on colour Doppler as a 'yin-yang' or 'to-and-fro' sign from swirling blood flow within the mass.⁸ The size of the haematoma seen on the CT and found intraoperatively in our patient may have limited the use of US and may explain why no abnormal findings were documented on the bedside pelvic US. A CT scan showing active extravasation of contrast material in a hyperdense mass in the pelvis is consistent with a ruptured UAP although it is not specific. Finally, angiography can be used to both confirm the diagnosis and treat a ruptured UAP. The characteristic findings of a ruptured UAP on angiography include a

sac-like structure and extravasation from this structure. We suspect that angiography failed to demonstrate active extravasation from the sac-like structure in our patient because haemostasis had already been achieved intraoperatively using packing. Angioembolisation can be used to treat ruptured UAP in stable and/or in unstable patients if the diagnosis is highly suspected based on US, CT or angiography.⁹ Díaz-Murillo *et al*⁷ reported the first case using this technique successfully in a patient found to have a bleeding uterine varicocele on emergency caesarean section.

Unfortunately, the diagnosis of UAP is made preoperatively in only 5% of cases presenting with haemodynamic collapse.⁷ This leaves no other options but emergency surgery for repair of the bleeding vessel or haemostatic hysterectomy.⁷ Aggressive resuscitation and exploratory laparotomy for haemostatic control with abdominal packs allowed for angioembolisation as an alternative to hysterectomy. Díaz-Murillo *et al*⁷ recommend bilateral embolisation of the uterine arteries to decrease the risk of rebleeding secondary to formation of collaterals leading to recanalisation of the affected side.⁹ The main advantages of angioembolisation are the decreased risk of serious complications related to major surgery and preservation of fertility.⁷ The main complications associated with uterine artery angioembolisation include pulmonary embolism, deep-vein thrombosis, uterine infarction, transient ovarian failure, bladder wall necrosis and vaginal fistula.⁷ The risk of complications is decreased with unilateral uterine embolisation. We suggest unilateral embolisation if: (1) the cause of bleeding is known; (2) the contralateral uterine artery is normal calibre and does not cross the midline and (3) the patient can be reliably monitored for rebleeding. Unilateral UAP angioembolisation with an absorbable agent decreases the risk of permanent amenorrhoea and uterine necrosis.⁹

Learning points

- ▶ Uterine artery pseudoaneurysm (UAP) is a rare complication of traumatic deliveries, caesarean sections or traumatic abortions.
- ▶ Spontaneous haemoperitoneum occurring secondary to extrauterine UAP can result in life-threatening haemorrhage.
- ▶ Emergency laparotomy with haemostatic hysterectomy has been classically described as the only treatment option for extrauterine UAP.
- ▶ Rapid diagnosis of extrauterine UAP and balanced resuscitation can permit angioembolisation of the culprit vessel through interventional radiology to preserve fertility.
- ▶ The risk of extrauterine UAP recurrence and the natural history of incidentally discovered UAP are unknown.

Contributors MD was responsible for data collection and manuscript preparation. JM was responsible for identification and management of the case as well as for manuscript preparation. JR was responsible for identification and management of the case as well as for manuscript preparation.

Competing interests None declared.

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REFERENCES

- 1 Baba Y, Matsubara S, Kuwata T, *et al*. Uterine artery pseudoaneurysm: not a rare condition occurring after non-traumatic delivery or non-traumatic abortion. *Arch Gynecol Obstet* 2014;20:435–40.
- 2 Giulini S, Zanin R, Volpe A. Hemoperitoneum in pregnancy from a ruptured varix of broad ligament. *Arch Gynecol Obstet* 2010;282:459–61.

- 3 Fan Y, Inocencio G, Azevedo S, *et al.* Spontaneous rupture of utero-ovarian vessels in pregnancy. *Acta Obstet Gynecol Scand* 2013;7:215–18.
- 4 Kapila P. Fatal non-traumatic spontaneous hemoperitoneum in second trimester of pregnancy—autopsy findings. *J Forensic Leg Med* 2011;18:139–40.
- 5 Hodgkinson CP. Hemorrhage from ruptured utero-ovarian veins during pregnancy; unusual syndrome complicating pregnancy. *JAMA* 1952;148:277–8.
- 6 Ginsburg KA, Valdes C, Schinder G. Spontaneous utero-ovarian vessel rupture during pregnancy: three case reports and a review of the literature. *Obstet Gynecol* 1987;69:474–6.
- 7 Díaz-Murillo R, Tobías-González P, López-Magallón S, *et al.* Spontaneous hemoperitoneum due to rupture of uterine varicose veins during labor successfully treated by percutaneous embolization. *Case Rep Obstet Gynecol* 2014;2014:580384.
- 8 Matsubara S. Pseudoaneurysm: a chameleon in obstetrical emergency practice. *Arch Gynecol Obstet* 2011;283:669–70.
- 9 Dohan A, Soyer P, Subhani A, *et al.* Postpartum hemorrhage resulting from pelvic pseudoaneurysm: a retrospective analysis of 588 consecutive cases treated by arterial embolization. *Cardiovasc Intervent Radiol* 2013; 36:1247–55.

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