CASE REPORT

An unusual cause of fetomaternal distress at term: uterine vessel rupture in pregnancy

Guy Anthony Calcott,¹ Mohammed Gaber,² Jonathan Freedman,³ Shalini Patni⁴

¹Department of Obstetrics and Gynaecology, Shrewsbury and Telford Hospitals NHS Trust, Telford, UK

²Department of Obstetrics and Gynaecology, Heart of England NHS Trust, Birmingham, UK

³Department of Interventional Radiology, Heart of England NHS Trust, Birmingham, UK

⁴Department of Feto-maternal Medicine, Heart of England NHS Trust, Birmingham, UK

Correspondence to
Dr Guy Anthony Calcott,
gacalcott@doctors.org.uk

Accepted 14 August 2015

SUMMARY

This report describes a case of spontaneous bleeding from uterine vessels presenting as hyperstimulation and fetomaternal distress at term. A 40-week primigravida woman underwent an emergency caesarean section for fetal distress, which unexpectedly revealed a spontaneous haemoperitoneum. Clinical assessment and investigations postoperatively gave a diagnosis of a right-sided uterine artery aneurysm that, it was believed, had ruptured, causing the haemoperitoneum. This was successfully treated postnatally using interventional radiological techniques leading to expeditious maternal recovery and discharge home.

BACKGROUND

This case highlights the value of interventional radiology in obstetrics in cases of postpartum haemorrhage (PPH), and the importance of considering it as a treatment modality. In appropriately selected cases, it allows rapid treatment of bleeding with quick recovery, while avoiding a potential return to theatre and further surgery with its attendant risks and morbidity.

CASE PRESENTATION

A 33-year-old primigravida was booked at 12 weeks gestation as a low-risk pregnancy. The medical history was unremarkable apart from depression treated with fluoxetine. There was no other gynaecological or medical history of note, and no history of pelvic surgery. Her antenatal period included an admission at 32 weeks gestation with contractions and positive Actim Partus, for which she was given dexamethasone. She was discharged home after two nights and the antenatal course was otherwise uneventful.

She presented to the hospital at 40 weeks gestation with a history of spontaneous onset of contractions. She was assessed by the midwife and her cervix was closed. She was encouraged to go home with advice to return if pains become frequent and stronger, as per standard practice.

The patient then re-presented 2 days later in the early morning hours, with a history of frequent, strong and painful contractions. She was assessed by the midwife and found to have a heart rate of 107 bpm, blood pressure 121/98 mm Hg, temperature 35.6°C and respiratory rate 16 breaths/min. She was started promptly on continuous fetal monitoring and an obstetric review was requested on concerns about the maternal condition.

On assessment by the obstetrician, the patient looked pale, distressed and unwell. She reported

constant abdominal pain and contractions for 48 h. She did not report vaginal bleeding and described normal fetal movements. There was no history of ruptured membranes. She was clammy and her heart rate was now regular at 120 bpm. Abdominally, the uterus felt soft between regular contractions. The contractions were occurring every 1–2 min (7 contractions in 10 min). The abdomen was tender on examination. On vaginal assessment, the cervix was closed and there was no evidence of vaginal bleeding.

The fetal monitoring trace was pathological, with reduced variability and atypical variable decelerations. Immediate intravenous access and fluid resuscitation were started with an initial diagnosis of hyperstimulation. Haemoglobin on venous blood gas was 120 g/L and maternal lactate 4.2 mmol/L. As there was no vaginal bleeding, terbutaline 250 µg was given subcutaneously. The hyperstimulation continued with a pathological cardiotocography and the patient was transferred to theatre for category 1 caesarean section under general anaesthetic with a working diagnosis of concealed abruption.

Intraoperatively, there was haemoperitoneum immediately evident on entering the abdomen, estimated to be 400 mL at the time. A lower segment caesarean section was performed. The baby was born with Apgar scores of 5, 6 and 9 at 1, 5 and 10 min, respectively, and went on to recover well. Cord gases were reassuring with arterial pH: 7.31, base excess –6.4 and venous pH: 7.32, base excess –6.7.

There was no evidence of placental abruption following delivery of the placenta. On abdominal survey, there was no uterine rupture or bleeding from the upper abdominal viscera. The operation was attended by the local gynaecological oncology lead and, on further inspection, bleeding from the posterior aspect of the right broad ligament was noted. No vascular anomaly was positively identified at this point. As the bleeding was from within the pelvic structures, the operating gynaecologist felt positioned to proceed without the assistance of a general surgeon or vascular specialist. Sutures were placed across the area of bleeding with haemostasis achieved. An intraperitoneal drain was inserted and the abdomen was closed as per standard practice.

Postoperatively, the patient received a total of 2 units of packed red cells and 2 units of fresh frozen plasma (FFP). She developed low-grade fever and was started on oral antibiotics. Day 2 postoperatively, she reported worsening abdominal pain. Her respiratory rate was 27 breaths/min with heart rate of 98 bpm. Her abdomen was soft but

CrossMark

To cite: Calcott GA, Gaber M, Freedman J, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-209004

distended, with evidence of rebound tenderness without guarding. The total drain output since delivery was approximately 150 mL of serosanguinous effluent.

INVESTIGATIONS

The patient underwent a contrast CT scan, which reported a right broad ligament haematoma 9 cm×3.4 cm extending to the pouch of Douglas, with bowel distension. General surgical review offered a diagnosis of paralytic ileus, and it was felt that this could be secondary to the operation itself along with the broad ligament haematoma. A conservative approach was adopted in the first instance, with consideration to drainage should the clinical picture fail to improve. The patient subsequently opened her bowels and her abdominal distension improved. She continued to look very pale and on-going bleeding was then suspected. The initial CT images were reviewed again by the obstetric consultant with the reporting radiology consultant to search for any suggestion of continued bleeding. A blush of contrast was then seen in the right adnexum (figure 1). This was thought to represent an actively bleeding vessel from within the pelvic plexus. After discussion within the team, it was felt that return to theatre for a laparotomy under such circumstances could make it difficult to identify this vessel and a better alternative would be to obtain input from the interventional radiologist for the unit. An urgent referral was made and pelvic angiography was conducted.

TREATMENT

The left common femoral artery was accessed under ultrasound guidance. Selective catheterisation of the right internal iliac artery was performed with a 5 Fr RIM catheter (COOK Group, Bloomington, Indiana, USA). Diagnostic digital subtraction angiography performed from this location demonstrated a contained rupture of an aneurysm of the horizontal segment of the right uterine artery (figure 2A). The right uterine artery was superselectively catheterised with a 2.7 Fr microcatheter. This resulted in active extravasation of contrast (figure 2B). The aneurysm and proximal horizontal segment of the uterine artery were immediately embolised using 4×8 mm×14 cm micro-Nester coils (COOK Group, Bloomington, Indiana, USA). Subsequent angiography revealed no further extravasation of contrast and successful occlusion of the distal right uterine artery (figure 3A, B). The patient remained haemodynamically stable throughout the procedure.

Figure 1 Contrast-enhanced CT of the abdomen and pelvis showing a blush of contrast in the right adnexum.

Figure 2 (A) Diagnostic angiogram showing contained contrast extravasation in an aneurysm of the right uterine artery. (B) Fluoroscopic frame grab showing superselective catheterisation of the right uterine artery with active extravasation of contrast from the aneurysm.

OUTCOME AND FOLLOW-UP

Following embolisation, the patient recovered well, and her clinical condition improved rapidly. She was discharged on oral ferrous sulfate 6 days after the procedure. She remained well on follow-up at 2 weeks postdischarge and also on telephone follow-up at 3 months.

DISCUSSION

This is a rare and interesting case of what appears to be a spontaneous rupture of the uterine artery in pregnancy from what the authors believe to be a uterine artery aneurysm. While a pseudoaneurysm may remain a possibility on radiographic findings alone, it is felt that this is less likely in the clinical context, as it would not account for the initial presentation and, also, it would be unusual for it to form in such a short time frame post-operatively. While pseudoaneurysm is a recognised complication following a caesarean section, it typically presents as late PPH,^{1 2} as opposed to acute antepartum or intrapartum bleeding. Published cases on antepartum rupture of the uterine vessels are rarer,^{3 4} with previous cases being associated with ectopic decidual tissue⁵ and endometriosis.⁶ Aneurysms of the uterine artery are indeed unusual, with few reports of identification in the antenatal period.⁷ They have not previously been reported to present in the manner described.

Figure 3 Diagnostic digital subtraction angiography ((A) performed via microcatheter in the right uterine artery, and (B) performed from RIM catheter in the right internal iliac artery) showing coil embolisation of the aneurysm and uterine artery, and no further contrast extravasation.

Traditional management of such cases involving rupture of the uterine vessels is surgical. This would be through a laparotomy, which would often entail either a hysterectomy or ligation of the uterine or iliac arteries. This case, however, adds to the body of evidence supporting the use of real-time radiological techniques in the diagnosis and management of PPH, in particular, acute single vessel bleeding. There is also a report where identification of a uterine artery aneurysm earlier in the antenatal course has resulted in the aneurysm being embolised in a timely manner, with successful continuation of the pregnancy.⁷

Learning points

- ▶ Interventional radiology is worth considering in such cases of suspected postoperative bleeding, as it has the advantage of avoiding surgery, preventing visceral injuries and circumventing the associated morbidity of a laparotomy.
- ▶ Interventional radiology is a useful tool in cases where access to the bleeding vessel/s is difficult through an open procedure (laparotomy).
- ▶ Interventional radiology is minimally invasive, with low infection risk, short hospital stay and quick recovery.
- ▶ Where spontaneous bleeding is identified intraoperatively but a specific vessel cannot be identified, intraoperative opinion of a vascular specialist should be considered. Where this is not available, intraoperative or early postoperative angiography with or without embolisation can be used to investigate for underlying vascular anomaly and to confidently achieve haemostasis.

The use of angiography and embolisation in this case allowed successful treatment and stabilisation of the underlying condition with no significant added morbidity, and, undoubtedly, reduced the length of stay compared to what otherwise would have been a repeat laparotomy. The authors would encourage readers to consider the use of angiographic techniques (either CT or real-time) in the diagnosis and treatment of secondary PPH, particularly after an operative delivery.

Contributors All the authors were involved in the clinical care of this patient. The first and final draft were prepared by GAC with input from MG and JF relevant to their involvement in the clinical care. SP supervised and reviewed the report throughout.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Dasari P, Maurya DK, Mascarenhas M. Uterine artery pseudoaneurysm: a rare cause of secondary postpartum haemorrhage following caesarean section. *BMJ Case Rep* 2011;2011:pii: bcr0120113709.
- 2 Yi SW, Lee JH. Uterine pseudoaneurysm leakage may cause delayed postpartum haemorrhage: multidetector CT with angiography and transcatheter uterine arterial embolization. *J Obstet Gynaecol* 2012;32:552–5.
- 3 Duhan N, Sangwan N, Rajotia N, et al. Spontaneous uterine artery rupture at delivery. *J Obstet Gynaecol India* 2013;63:72–3.
- 4 O'Connell MP, Prendiville W. Spontaneous uterine artery rupture: a rare cause of abdominal pain in pregnancy. *J Obstet Gynaecol* 2005;25:511–12.
- 5 Azzam HM, Daayana L, Bancroft K, et al. Spontaneous rupture of the uterine artery in pregnancy. *Sci J Med Clin Trials* 2012;2012: 2pp.
- 6 Aziz U, Kulkarni A, Lazic D, et al. Spontaneous rupture of the uterine vessels in pregnancy. *Obstet Gynecol* 2004;103(5 Pt 2):1089–91.
- 7 Cornette J, van der Wilk E, Janssen NM, et al. Uterine artery pseudoaneurysm requiring embolization during pregnancy. *Obstet Gynecol* 2014;123 (2 Pt 2 Suppl 2):453–6.

Copyright 2015 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow