CASE REPORT

Decidualised fallopian tube endometriotic implant causing spontaneous haemoperitoneum in a twin pregnancy

Ileera Aggarwal,¹ Pamela Tan,² Manisha Mathur²

¹Department of Gynaecological Oncology, KK Women's and Children Hospital, Singapore, Singapore

²Department of Obstetrics and Gynaecology, KK Women's and Children Hospital, Singapore, Singapore

Correspondence to

Dr Ileera Aggarwal, lmaggar@hotmail.com

Accepted 5 July 2014

SUMMARY

Spontaneous haemoperitoneum in pregnancy (SHiP) is a rare but serious condition associated with an adverse pregnancy outcome. We present a case of previable twin pregnancy presenting with SHiP secondary to endometriosis, necessitating an emergency laparotomy and hysterotomy as a life-saving measure to achieve haemostasis. At laparotomy there was massive haemoperitoneum with active bleeding from the left fallopian tube and a left salpingectomy was performed. Histological examination showed haemorrhagic foci of endometriosis on the fallopian tube exhibiting florid stromal decidual change. We wish to raise awareness of this uncommon but potentially life-threatening condition requiring early recognition and prompt recourse to surgical intervention to minimise the morbidity and mortality.

BACKGROUND

Spontaneous haemoperitoneum in pregnancy (SHiP) is a rare condition associated with a high maternofetal mortality and morbidity.¹ Endometriosis is a major risk factor causing spontaneous haemoperitoneum either via invasion of the adjacent vessels or bleeding from the decidualised endometriosis implants themselves.²

CASE PRESENTATION

A 31-year-old primigravida presented to the labour ward triage at 21 weeks, 6 days gestation with sudden onset of severe upper abdominal pain and one episode of vomiting. The pain started in the upper abdomen 2 h prior to presentation, but later became more generalised in location and constant in nature. There was no history of uterine contractions, leaking liquor or vaginal bleeding. There were no precipitating factors of abdominal trauma, coitus or straining during defecation. The pregnancy had been uneventful till this presentation.

This was a dichorionic diamniotic twin pregnancy, conceived following her first cycle of in vitro fertilisation-intracytoplasmic sperm injection for primary infertility resulting from severe endometriosis. She had a history of therapeutic laparoscopy for severe endometriosis and a 5 cm left-sided endometrioma, 2 years prior to the conception. During the laparoscopy she was noted to have a frozen pelvis with extensive adhesions involving the colon and an obliterated pouch of Douglas. During the procedure, extensive adhesiolysis,

ablation of endometriotic implants and a left ovarian cystectomy was performed.

On admission, she was noted to have a pain score of 9/10. Her vital signs were stable and abdominal palpation revealed generalised tenderness with guarding. Uterus was palpable at fundal height of 24–26 weeks. The cervix was closed and tubular and there was no vaginal bleeding. Cervical motion tenderness was positive.

INVESTIGATIONS

Haemoglobin was 9.9 g/dL, total white cell count $22.64 \times 10^9/L$ and platelets $296 \times 10^9/L$ along with normal liver and renal function, serum amylase and C reactive protein. An abdominopelvic ultrasound showed normal liver, gallbladder, spleen and kidneys. A small amount of free fluid was noted in the perisplenic and perihepatic areas. The appendix could not be fully evaluated. A twin pregnancy with demise of one fetus was noted. The other foetal heart was normal. There was no evidence of a retroplacental clot. Neither of the ovaries was visualised.

Surgical consult was sought. Antibiotics were started in view of high leucocyte count. The pain gradually reduced in intensity with analgesia and became more localised to the left flank area. Over a 24 h period her vital parameters remained stable but the haemoglobin dropped to 7.5 g/dL and she was transfused with packed cells. The urine analysis was normal.

MRI of the abdomen and pelvis was undertaken and showed a gravid uterus with two foetuses within, one of which was non-viable. Generalised ascites of heterogenous T1-weighted and T2-weighted signal and a fluid–fluid level in the inferior pelvis adjacent to the rectum was noted (figure 1). Haemoperitoneum could not be excluded, however, a definitive cause was not identified on the study. The visualised uterine outline appeared intact. There was no obvious free intraperitoneal gas. It was difficult to visualise the appendix due to adjacent oedema. The liver and spleen appeared normal and there was no overt adnexal mass.

TREATMENT

As the patient improved clinically, conservative management was continued. However, on day 3 since admission, she reported sudden onset of shortness of breath and increasing abdominal pain. Her oxygen saturation dropped to 90% on room

CrossMark

To cite: Aggarwal I, Tan P, Mathur M. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-205317

Figure 1 Haemoperitoneum on MRI.

air and she was noted to be pale with tachycardia and significant hypotension with a distended and guarded abdomen. A bedside ultrasound revealed free fluid in the paracolic gutters. Decision was made for an urgent exploratory laparotomy on suspicion of ongoing intraperitoneal haemorrhage leading to maternal collapse.

A midline laparotomy was performed. There was a massive haemoperitoneum of 2.2 L with large clots extending to the liver and splenic region. There was a frozen pelvis due to the endometriosis obliterating the pouch of Douglas completely. Active bleeding was noted coming from the left adnexa. However, the exact bleeding point could not be identified initially due to the gravid uterus, adhesions and the distorted anatomy. Decision was therefore made for a hysterotomy to improve access. The left fallopian tube was noted to be distended and stretched due to endometriotic adhesions with active bleeding from it. Left ovary could not be identified clearly. Endometriotic deposits were also noted on fundus and posterior surface of the uterus as well as the terminal ileum with active oozing from the uterine deposits. Salpingectomy was performed to control the bleeding after identification of the left ureter. The uterine incision was closed in two layers and haemostasis was achieved.

OUTCOME AND FOLLOW-UP

The total estimated blood loss during the procedure was 3.5 L and the patient was transfused with 6 pints of packed red blood cells, 1 L of fresh frozen plasma, 1 pint of cryosupernatant plasma and 10 units of cryoprecipitate intraoperatively. The patient recovered well and subsequently discharged on the 12th postoperative day.

Histological examination of the removed fallopian tube showed haemorrhagic foci of endometriosis exhibiting florid stromal decidual change and dispersed variably atrophic dilated glands lined by flat epithelium (figures 2 and 3).

DISCUSSION

Advances in assisted reproductive techniques have allowed sub-fertile women with endometriosis to get pregnant. Data emerging from studies of pregnant women with endometriosis has increased our awareness of complications occurring in this cohort.³ Adverse pregnancy outcomes including rare SHiP, obstetrical bleeding and preterm birth have been linked to endometriosis.⁴ The exact pathophysiological mechanism is not very clear. Various theories have been proposed ranging from

Figure 2 Histology: tubal serosal endometriosis haemorrhage.

progesterone resistance in eutopic endometrium to decidualisation of ectopic endometrium.⁵

Ectopic decidualisation (formation of ectopic decidua) during pregnancy is a well-documented phenomenon due to the effect of progesterone on either the normal subcoelomic mesothelium of peritoneal cavity or on endometriotic deposits in ovary, fallopian tube, posterior cul de sac, uterosacrals and even appendix, ileum and ureter.⁶ Increased angiogenesis, inflammation and invasiveness of these endometriotic implants have been suggested as a cause of SHiP. Intrusion of decidualised stroma into the vessel wall can cause narrowing of the vessel leading to increased pressure, weakness and eventually rupture. In our case above, we hypothesise that decidualisation of the vascular endometriotic implant on the tube could have weakened the area. As the uterus enlarged, there was excessive traction on the severely adherent decidualised tube causing it to rupture leading to massive haemoperitoneum. Bleeding endometriotic implants noted at multiple sites in the pelvis also contributed.

The source of bleeding in cases of SHiP may not be easily recognisable. In a review of 20 publications, the bleeding was described as venous in 80%, arterial in 16% or unknown in 1% cases.² The most common site of bleeding was noted to be posterior surface of uterus or parametria. Rupture of utero-ovarian veins, uterine vein, erosion of uterine artery by endometriosis, rupture of ovarian endometrioma, invasion of ileum by invasive

Figure 3 Histology: mesosalpinx endometriosis.

endometriotic implant have all been implicated as cause of spontaneous haemoperitoneum in pregnancy.^{7–11} There is only one other reported case of massive haemoperitoneum from endometriosis of the fallopian tube, which occurred in a non-pregnant woman and was mistaken as a ruptured ectopic pregnancy.¹² Another case has been reported by Reif *et al* where a left adnexal tumour was the source of spontaneous haemoperitoneum in a twin pregnancy. At laparotomy, the conglomerate mass was identified to be left tube, adhesion and ruptured left ovarian cyst.¹³ Spontaneous haemoperitoneum due to rupture of utero-ovarian vessels is reported to occur most commonly in the third trimester and labour, possibly due to increased intra-abdominal pressure during labour or manouevres such as vomiting, straining during defecation, etc. In the case above, however it occurred earlier at a pre-viable stage of pregnancy. There were no precipitating factors which could explain the sudden haemoperitoneum.

Preoperative diagnosis of SHiP can be challenging. Non-specific symptoms of acute abdominal pain, vomiting and maternal anaemia could signal haemoperitoneum, especially in a case with prior history of endometriosis.¹⁴ Brosens *et al*² reviewed 25 cases of spontaneous haemoperitoneum in pregnancy and reported that 72% were nulliparous, with a mean age of 30. In all the cases, ultrasound examination failed to diagnose intraperitoneal bleed. Differential diagnoses cited were uterine rupture, HELLP syndrome, placental abruption, bleeding ectopic decidual, splenic and hepatic artery aneurysm rupture and perforated appendicitis. MRI can help characterise the abdominopelvic anatomy better, helping to rule out a surgical cause of the acute abdomen, for example, perforated appendix. In the presented case, use of analgesia led to a decrease in pain and continued conservative management. We wish to highlight that in cases of pain with unknown aetiology, analgesia should be used cautiously as it might delay the diagnosis as well as prompt management.

Laparotomy becomes necessary in most of these cases either after maternal collapse or due to fetal distress. A midline incision for laparotomy can allow adequate access to explore the whole abdomen and pelvis. Hysterotomy had to be performed in our case to ascertain the source of bleeding. Although

Learning points

- ▶ Spontaneous haemoperitoneum in pregnancy is a rare condition associated with a high maternofetal mortality and morbidity.
- ▶ Pelvic endometriosis is a major risk factor and more cases can be expected with increasing use of assisted reproductive techniques.
- ▶ High index of suspicion in pregnant women with history of endometriosis presenting with acute abdominal pain and hypovolemia can lead to prompt diagnosis and treatment.
- ▶ Early recourse to surgical treatment improves maternal survival though perinatal morbidity remains high.

preservation of a pre-viable pregnancy is desirable, often delivery of the foetus is required for identification and control of bleeding site.^{15 16} Fortunately, we were able to preserve the uterus in our case.

CONCLUSION

Spontaneous haemoperitoneum in pregnancy is a rare but catastrophic event. Although improvement in resuscitative and anaesthetic techniques has significantly reduced the maternal mortality (49.3–4% between 1950 and 1985), perinatal mortality still remains high (31%).¹⁷ Increased awareness, especially in a pregnant patient with history of endometriosis, is paramount for prompt diagnosis and management.

Acknowledgements The authors wish to thank Dr Jennifer Teo for providing the histopathology slides.

Contributors All the authors were involved in the clinical management as well as researching and preparing the manuscript.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Ginsburg KA, Valdes C, Schnider G. Spontaneous utero-ovarian vessel rupture during pregnancy: three case reports and a review of the literature. *Obstet Gynecol* 1987;69:474–6.
- 2 Brosens IA, Fusi L, Brosens JJ. Endometriosis is a risk factor for spontaneous hemoperitoneum during pregnancy. *Fertil Steril* 2009;92:1243–5.
- 3 Stephansson O, Kieler H, Granath F, *et al*. Endometriosis, assisted reproduction technology, and risk of adverse pregnancy outcome. *Hum Reprod* 2009;24:2341–7.
- 4 Falconer H. Pregnancy outcomes in women with endometriosis. *Semin Reprod Med* 2013;31:178–82.
- 5 Brosens I, Brosens JJ, Fusi L, *et al*. Risk of adverse pregnancy outcome in endometriosis. *Fertil Steril* 2012;98:30–5.
- 6 O'Leary SM. Ectopic decidualization causing massive postpartum intraperitoneal hemorrhage. *Obstet Gynecol* 2006;108:776–9.
- 7 Inoue T, Moriwaki T, Niki I. Endometriosis and spontaneous rupture of utero-ovarian vessels during pregnancy. *Lancet* 1992;340:240–1.
- 8 Renuka T, Dhaliwal LK, Gupta I. Hemorrhage from ruptured utero-ovarian veins during pregnancy. *Int J Gynaecol Obstet* 1998;60:167–8.
- 9 Nishikawa A, Kondoh E, Hamanishi J, *et al*. Ileal perforation and massive intestinal haemorrhage from endometriosis in pregnancy: case report and literature review. *Eur J Obstet Gynecol Reprod Biol* 2013;170:20–4.
- 10 Mizumoto Y, Furuya K, Kikuchi Y, *et al*. Spontaneous rupture of the uterine vessels in a pregnancy complicated by endometriosis. *Acta Obstet Gynecol Scand* 1996;75:860–2.
- 11 Janicki TI, David LJ, Skaf R. Massive and acute hemoperitoneum due to rupture of the uterine artery by erosion from an endometriotic lesion. *Fertil Steril* 2002;78:879–81.

Patient's perspective

I write this in remembrance of my twin angels, whom I dearly miss. The sudden onset of severe abdominal pain was our first clue to an underlying serious problem, in a pregnancy that had been completely uneventful so far. We sought immediate medical attention and went through numerous ultrasound examinations and even MRI. But none of them could give a clue as to what the exact cause might be. Unfortunately, only a laparotomy, along with a hysterotomy, was to help both identify the problem and be the solution. It is also hard to imagine what might have transpired had I been further away from medical care. I firmly believe that it was the immediate medical attention and surgery that I received within minutes, which helped save my life. As for the future, I sincerely hope advances in medical science can help control endometriosis and prevent rare but devastating complications such as this from occurring during pregnancy, which is often precious and achieved in the first place after much difficulty in many of us with endometriosis.

- 12 Harmanli OH, Chatwani A, Caya JG. Massive hemoperitoneum from endometriosis of the fallopian tube. A case report. *J Reprod Med* 1998;43:716–18.
- 13 Reif P, Schöll W, Klaritsch P, *et al.* Rupture of endometriotic ovarian cyst causes acute hemoperitoneum in twin pregnancy. *Fertil Steril* 2011; 95:2125.e1–3.
- 14 Williamson H, Indusekhar R, Clark A, *et al.* Spontaneous severe haemoperitoneum in the third trimester leading to intrauterine death: case report. *Case Rep Obstet Gynecol* 2011;2011:173097.
- 15 Aziz U, Kulkarni A, Lazic D, *et al.* Spontaneous rupture of the uterine vessels in pregnancy. *Obstet Gynecol* 2004;103:1089–91.
- 16 Leung WC, Leung TW, Lam YH. Haemoperitoneum due to cornual endometriosis during pregnancy resulting in intrauterine death. *Aust N Z J Obstet Gynaecol* 1998;38:156–7.
- 17 Hodgkinson CP, Christensen RC. Hemorrhage from ruptured uteroovarian veins during pregnancy; report of 3 cases and review of the literature. *Am J Obstet Gynecol* 1950;59:1112–17.

Copyright 2014 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