Rare disease

Uterine artery pseudoaneurysm: a rare cause of secondary postpartum haemorrhage following caesarean section

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Summary

A 30-year-old woman, para 2, who underwent an uneventful repeat elective lower segment caesarean section with sterilisation presented on the 23rd postnatal day to our Obstetric Emergency department with profuse bleeding per vaginum. She had a similar bout of bleeding on the 19th postnatal day and was treated elsewhere with blood transfusion and uterotonics. On ultrasonography, she was diagnosed to have few retained adherent placental bits in the lower part of the uterus with a small left broad ligament haematoma and hence was injected with a single dose of methotrexate. A communication of uterine artery was suspected as the mass was close to the uterus and few pulsations were felt on per vaginal examination while attempting aspiration of haematoma. Uterine artery pseudoaneurysm was diagnosed by Doppler studies and confirmed by contrast enhanced CT. She was treated successfully by bilateral internal iliac artery ligation as facilities for embolisation are not available.

BACKGROUND

Difficulties are encountered in arriving at diagnosis of uterine artery pseudoaneurysm. It is an uncommon cause of secondary postpartum haemorrhage (PPH) and can lead to mortality due to rupture if not recognised and treated.

CASE PRESENTATION

A 30-year-old woman, para 2, with two living children attended our emergency Obstetrics department with complaints of profuse vaginal bleeding since 4 days. There was no history of abdominal pain. Bleeding was bright red to start with and later mixed with few clots. The medical history revealed that she underwent an uneventful elective lower segment caesarean section (LSCS) with sterilisation 23 days ago at some other hospital. On the 19th postnatal day, she had a similar episode of bleeding for which she was transfused with 2 units of packed cells and treated with injections of tranexamic acid and methylergometrine. There was no history suggestive of haemorrhage during or immediately after LSCS and she did not receive any blood

transfusion at the time of surgery. There was no similar history following the first caesarean section. There was no relevant medical, social or family history like bleeding diathesis.

General examination revealed severe pallor, a pulse rate of 90/min and blood pressure (BP) of 150/90 mm Hg. On inspection of abdomen, there was a transverse suprapubic scar with induration. Palpation revealed 14 weeks size uterus which was well contracted. Per speculum examination showed brownish discharge through a healthy cervix and there was no fresh bleeding. A provisional clinical diagnosis of secondary PPH was made and she was investigated for the cause.

Her haemoglobin was 6 g% and bleeding time and clotting time were within normal limits. She was transfused with 1 unit of packed cells and was started on ceftriaxone injection. Ultrasonogram (USG) performed the next day showed an echogenic area in the lower part of the uterus along with an anechoic area of 5 × 6 cm to the left of the uterus (figure 1A,B). Adherent placental bits with

Figure 1 (A) Initial transvaginal scan (TVS) shows an echogenic area in the lower part of the uterus and an anechoic area to its left and below. (B) TVS showing well-circumscribed anechoic area in the myometrium – uterine pseudoaneurysm.

left broad ligament haematoma was diagnosed. She was given a single dose of methotrexate 50 mg intramuscularly for the treatment of adherent placental bits. As the size of the suspected haematoma did not decrease, aspiration was attempted after 5 days under aseptic precautions in the operation theatre. Abdominal and vaginal routes of aspiration were tried without much result except a few cc of dark coloured blood. She was re-evaluated by a careful pelvic examination. The mass in the broad ligament was felt to be continuous with the uterus and few pulsations were felt. Hence, a communication of uterine artery with broad ligament haematoma was suspected and Doppler study was planned. Five days after this, she had another fresh bout of bleeding amounting to 300 ml and her pallor increased and her pulse rose to 110/min but BP was maintained (120/80 mm Hg). One unit of packed cells were transfused.

Doppler study undertaken on the 13th day of admission (36th postnatal day) showed the presence of left uterine artery pseudoaneurysm with flow into the haematoma (figure 2A,B). Contrast enhanced CT (CECT) performed 3 days later confirmed the presence of left uterine artery pseudoaneurysm, which was diagnosed by the presence of strong enhancement of the lesion in the arterial phase. The lesion was round to oval measuring 1.3–1.7 cm (figure 3A,B) and was close to the endometrial cavity surrounded by a rim of hypodensity and clot and was continuous with the reference artery. A final diagnosis of pseudoaneurysm of uterine artery was arrived at.

INVESTIGATIONS

1. Haemogram – 6 g%; peripheral smear – normocytic and normochromic RBC.
2. USG abdomen – echogenic area in the lower segment and anechoic area in the myometrium (figure 1).
3. Doppler studies of uterine arteries – swirling flow into the haematoma (figure 2).
4. CECT pelvis – enhancement of the lesion in the arterial phase (figure 3).
5. Cervical swab culture – no growth.

DIFFERENTIAL DIAGNOSIS

1. Secondary PPH due to retained and adherent placental bits.
2. Postoperative broad ligament haematoma.
3. Puerperal endometritis.

TREATMENT

- Severe anaemia corrected by multiple blood transfusions.
- Laparotomy and bilateral internal iliac artery ligation was undertaken as an elective procedure as facilities for embolisation are not available at our centre and patient did not want to undergo embolisation elsewhere as she could not afford the same.

(At laparotomy, the anterior surface of uterus was densely adherent to the anterior abdominal wall. Right ovary was adherent to the appendix. Peritoneal surface

Figure 2 Doppler studies of uterus. (A) Doppler flow into the enlarged area in the myometrium. (B) Flow during systolic phase as well as diastolic phase swirling flow.

Figure 3 CECT of pelvis enhancement of the lesion during arterial phase and its close proximity to the uterine cavity.

was oedematous and friable. Bilateral internal iliac artery ligation was performed with utmost care, and uterus was not mobilised and no attempt was made to trace the uterine arteries for fear of haemorrhage because of severe pelvic congestion.)

OUTCOME AND FOLLOW-UP

Follow-up USG and Doppler 5 days later and after 2 weeks revealed the absence of anechoic area and flow in the left uterine artery.

DISCUSSION

PPH is the leading cause of maternal mortality in India. Most of the cases are due to primary PPH. Secondary PPH is rare and can be life-threatening occasionally when the cause is not recognised. The common causes of secondary PPH are retained placental bits and postpartum endometritis. Uterine artery pseudoaneurysm is a rare cause of secondary PPH. Diagnostic problems are encountered in arriving at the diagnosis of this condition and if it is not recognised early pseudoaneurysm can rupture and cause massive bleeding leading to morbidity and mortality. So far, 57 cases of pseudoaneurysm of uterine artery were reported in literature.¹ This is the first case in our experience.

A pseudoaneurysm is a haematoma that forms outside the arterial wall as a result of a leaking hole in an artery and it is contained by the surrounding tissues and is in constant communication with the artery. It is also called as false aneurysm and delayed diagnosis results in life-threatening haemorrhage.¹⁻² It can manifest as late as 2 years,³ it can be asymptomatic⁴ or it can manifest with life-threatening vaginal bleeding.⁵ The clinical manifestations reported are delayed postabortal bleeding,³ delayed PPH,⁶ secondary PPH⁷ and postoperative haemorrhage.⁸ Development of pseudoaneurysms is a complication of vascular injury resulting from inflammation, trauma, or iatrogenic causes such as surgical procedures, percutaneous biopsy or drainage.⁹

The most common cause for uterine artery pseudoaneurysm is caesarean section.¹⁻¹⁰ Following caesarean section, it may be expected to occur in the immediate postoperative period in case of angle extension and can be diagnosed in the asymptomatic state itself by USG.⁴ Diagnostic problems occur when the patients are not screened by USG in the postoperative period and when they present late usually due to secondary PPH.⁶⁻⁷ Diagnostic difficulties are because of its rare nature and its presentation of vaginal bleeding. Under the influence of sustained arterial pressure, blood dissects the wall around the damaged artery and forms a perfused sac that communicates with the arterial lumen. While maintaining contact with the parent vessel, extravasating blood dissects through tissues, finally establishing a connection with the uterine cavity, causing a delayed haemorrhage.⁶

The unnecessary procedures that led because of missed diagnosis were repeated curettages⁷ and starting higher antibiotics.¹¹ The vaginal haemorrhage is usually massive and occurs repeatedly as in the present case and most often large number of transfusions are required.⁷⁻¹¹ The gold standard for diagnosis is angiography of pelvic vessels

though it can be diagnosed by the characteristic swirling flow on colour Doppler. The management in modern days is uterine artery embolisation, which was used in almost all the cases reported in recent years, but when facilities are not available as in the present case, bilateral internal iliac artery ligation can be undertaken. It is better to achieve bilateral uterine artery occlusion as there are case reports of reappearance of pseudoaneurysm requiring second time angiography because of collateral circulation.⁶

It is essential to look for pseudoaneurysm in cases of mass in pelvis following LSCS. One should keep in mind this possibility first rather than retained placental bits causing secondary PPH as every Obstetrician ensures that the placenta is completely removed at LSCS before suturing the uterine incision.

Learning points

- Misdiagnosis of uterine artery pseudoaneurysm is very common.
- Pseudoaneurysm of uterine artery has to be suspected whenever difficulties are encountered during caesarean section and a postoperative screening should be undertaken by ultrasound to rule out the same.
- Bilateral internal iliac artery ligation can be undertaken to manage a case of pseudoaneurysm of uterine artery where facilities for embolisation are not available.

Competing interests None.

Patient consent Obtained.

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