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Clinical Paper

Spontaneous rupture of uterine artery in a non-pregnant woman with adenomyosis: a case report and review of current literature

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Abstract

Spontaneous rupture of a uterine artery is a rare occurrence and more often associated with postpartum haemorrhage. It is even more unusual outside pregnancy. In this report, we will describe a case of spontaneous uterine artery rupture in a 40-year-old non-pregnant female with adenomyosis who presented with severe vaginal bleeding. We will also review the aetiology of rupture of uterine artery based on the current literature.

Case report

A 40-year-old woman, para 1, presented overnight to the emergency department of her local hospital with vaginal bleeding. It was her second attendance within 24 hours. She reported onset of menstruation six days previously, and the bleeding was increasingly heavy with clots, associated with intermittent lower abdominal cramps and fainting episodes. Her last pregnancy was more than 15 years ago. She had stopped a combined oral contraceptive pill two months before for a planned discectomy. On examination, she was pale with mildly tender suprapubic region, but her vital signs were stable. Initial blood results were as follows: haemoglobin 110 g/L, white cell count 11.5x10^9/L, platelet count 261x10^9/L. She was initially managed conservatively with tranexamic acid and medroxyprogesterone.

In the next few hours, she had multiple episodes of hypotensive shock and continued to bleed despite medical management. On vaginal examination, cervix felt normal and a significant amount of blood clot was evacuated with active bright red bleeding. Bedside transvaginal ultrasound was performed, and it showed an endometrial thickness of 2 mm, clots in uterus and vagina, and no fibroids or free fluid in the pelvis. Her haemoglobin dropped to 62 g/L. The massive haemorrhage protocol was activated, and she received 8 units of packed red cells, 2 units of fresh frozen plasma and 2 units of cryoprecipitate in total.

She therefore underwent a computed tomography (CT) of abdomen and pelvis with abdominal angiography. CT showed active extravasation of contrast from a branch of the right uterine artery into the endometrial cavity, tracking through the cervix into the vagina vault, but no evidence of a discrete vascular abnormality (Figure 1).

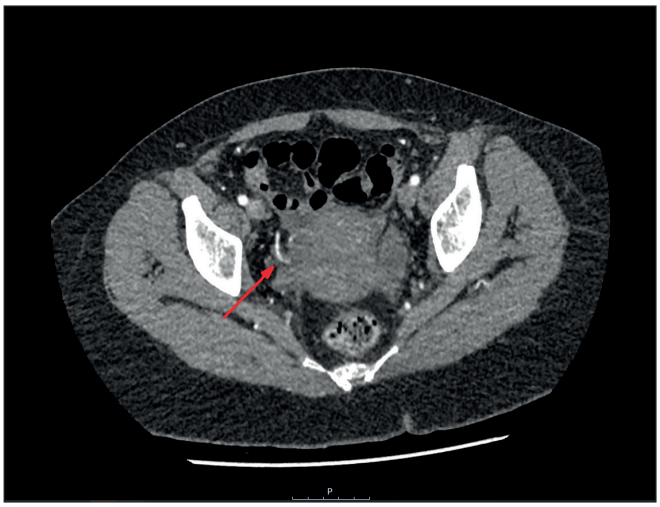

Figure 1.
Arterial phase of CT angiogram showing extravasation of contrast from a branch of the right uterine artery (red arrow) into the endometrial cavity.

Trans-arterial embolisation was considered, but due to the patient's ongoing bleeding and worsening pain, compounded by the delay to transfer to a tertiary centre with interventional radiology, a decision was made to undergo transabdominal hysterectomy. Intra-operative findings included haemoperitoneum, mild features of endometriosis on the uterus and normal adnexae. The procedure was uncomplicated, and her post-operative recovery was largely unremarkable, except for ileus and atelectasis. Histopathological examination of the specimen confirmed adenomyosis and no evidence of malignancy.

Discussion

Aetiology

Spontaneous rupture of uterine artery is a rare occurrence. As such, to date, no systemic review or large studies have been conducted on this. The electronic database PubMed was searched looking for the following terms 'spontaneous uterine artery rupture', 'spontaneous uterine vessel rupture'

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and 'spontaneous uterine artery erosion'. A number of cases have been reported and patients with this condition presented during pregnancy, labour, puerperium[1-8], or up to two years following a gynaecological intervention[9].

In relation to pregnancy, the exact aetiology is not well understood[3], but factors including pressure dynamics, anatomic disruptions and hormonal changes were proposed[10]. Other causes that have been identified[3] include aneurysm and pseudoaneurysm, congenital abnormality, vascular defect caused by inflammation or oestrogen[7], erosion from endometriosis[11-14], iatrogenic damage[15-18], and termination of pregnancy[9,19]. As described in Williams Obstetrics, uterine artery pseudoaneurysm is a known rare cause of postpartum haemorrhage[20]. A case series in China concluded that pseudoaneurysm can develop after traumatic pelvic operations and non-traumatic delivery/abortion[21]. Another observational study in Japan estimated the incidence of uterine artery pseudoaneurysm to be 0.3-0.6% of deliveries and suggested that some may be undetected due to the absence of massive bleeding and/or spontaneous resolution[22]. A case series in Pakistan[23] suggested that bleeding as a result of acquired and iatrogenic uterine vascular abnormalities can be attributed to previous caesarean section, gestational trophoblastic disease, pelvic tumours[24-28] and malignancies[29,30], pelvic infection, and dilatation and curettage.

Of all reported cases, uterine artery rupture that are truly spontaneous (i.e. unprovoked by pregnancy or trauma) are exceptionally rare. There was one case where no cause was identified[31], and few cases where the haemoperitoneum occurred as a complication of rupture of a massive fibroid[24,32,33]. In some case reports, local erosion of the uterine artery by an endometriotic lesion was described in non-gravid women[11,12,34]. Given the similar histopathology, we speculate that the adenomyosis in our case behaves similarly to this endometriosis case, causing local ulceration to the adjacent uterine artery, leading to eventual rupture and subsequent haemorrhagic event. In one case in Japan with similar presentation, a patient had a cardiac arrest secondary to severe haemorrhage from severe adenomyosis with fibroid[33].

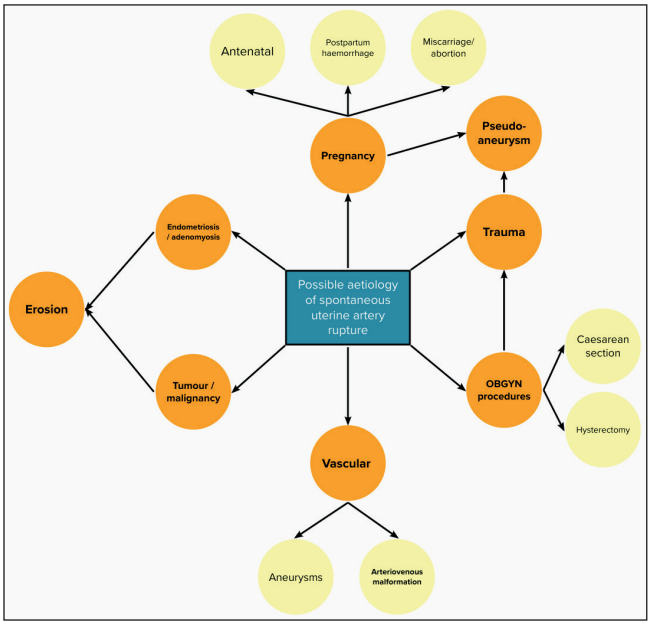

Figure 2.
Possible aetiology of spontaneous rupture of uterine artery. Eventual rupture of the artery can be multifactorial and causes can be interlinked (as demonstrated by the arrows). We deduce, in our case, the patient had a rupture of uterine artery secondary to erosion by adenomyosis.

Based on the literature and cases discussed, we have summarised the possible aetiology of spontaneous uterine artery rupture in Figure 2.

Treatment

In patients with severe haemorrhage such as ours, resuscitation and maintaining haemodynamic stability is the initial management. In terms of definitive treatment, trans-arterial embolisation is the treatment of choice for uterine artery rupture secondary to pseudoaneurysm[35-38], and this would also have been an appropriate treatment for the patient described in this case report. However, due to the lack of interventional radiology facilities in our local hospital, deterioration of the patient and completion of child-bearing, the more invasive management of hysterectomy was performed.

Conclusion

Spontaneous rupture of uterine artery is a rare event but an emergency with high mortality rate. Clinical suspicion in patients with unexplained ongoing severe vaginal bleeding should prompt further investigations and urgent input from obstetrics and gynaecology and interventional radiology. We suggest that, in patients with history of adenomyosis and endometriosis, the threshold for further investigations should be lower to allow rapid recognition and treatment.

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