CASE REPORT

Acquired vascular malformation in ruptured caesarean section scar: a rare cause of life-threatening vaginal bleeding

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SUMMARY

Uterine arteriovenous malformations (AVM) are rare and potentially life-threatening lesions. Patients present with severe vaginal bleeding which usually does not respond to conservative management and on most of occasions requires radiological or surgical intervention. We report a case of acquired AVM in a ruptured caesarean section scar. The patient presented with life-threatening vaginal bleeding and was treated with gonadotropin releasing hormone agonist and subsequent angioembolisation coiling.

BACKGROUND

Uterine arteriovenous malformations (AVM) are described as cirroid aneurysm, arterio-venous aneurysm, pulsating angioma or cavernous angioma. They cause intractable vaginal bleeding which can be life threatening in some cases and does not respond to medical treatment¹⁻³ They may be congenital or acquired. Acquired AVM may be the consequence of uterine curettage, caesarean section, gestational trophoblastic disease or endometrial carcinoma⁴ We report a case of 30-year-old woman, who came with intermittent episode of heavy vaginal bleeding.

CASE PRESENTATION

A 29-year-old woman para 1+3, presented to the gynaecology outpatient clinic with symptoms of polymenorrhea and menorrhagia for 10 months. She had history of ectopic pregnancy and right-sided salpingectomy 9 years previously. She had an 8 years old live child, born with caesarean section. In the last 2 years, she twice had dilation and curettage (D and C) for two spontaneous first trimester miscarriages. In immediate previous D and C 14 months previously, she had perforation of previous caesarean scar which was later surgically repaired. She started to have intermittent episode of heavy vaginal bleeding after 4 months of scar repair.

The patient's initial ultrasound scanning revealed an irregular anechoic lesion measuring 4.3×3 cm in the lower anterior wall of the uterus (figure 1). On Doppler spectral ultrasound it showed swirling turbulent flow with abnormal predominantly arterial waveform (figure 2). Multiple enlarged venous channels were identified in the left adnexa. A preliminary diagnosis of acquired AVM formation on repaired previously ruptured caesarean scar was made on ultrasound scanning. Findings were

further investigated and confirmed with CT angiography (CTA) of the pelvic region.

On CTA, predominant arterial supply of AVM was noted from left ovarian artery with small branches coming from the left uterine artery and draining venous channels draining into left internal iliac vein (figure 3). During radiological workup, the patient was asymptomatic and had no bleeding episode. She was managed medically with gonadotropin releasing hormone agonist (Zoladex) 3.6 mg/month (3 doses) subcutaneously. But after 3.5 month she presented in emergency department with heavy vaginal bleeding. Her haemoglobin was 7.6 gm/dL and was given 2 units of blood transfusion and was planned for angio-embolisation.

On preliminary angiography, a large AVM was identified within the uterus filling from bilateral uterine and left ovarian arteries and draining into left internal iliac vein. It was successfully embolised with polyvinyl alcohol particles, platinum coils and Histoacryl glue (figures 4–6).

The patient had uneventful recovery after procedure and was symptom free. A month later on follow-up ultrasound scanning, recurrence of AVM was noted at the same location but it was small in size compared with initial AVM and measured 2×1.7 cm. The patient refused further intervention. She was managed medically with oral methergine 0.125 mg twice daily for one month and oral transamine 500 mg thrice daily for 15 days. She had only one episode of vaginal bleeding during this period but remained haemodynamically stable. Six-month follow-up ultrasound scanning revealed spontaneous resolution of the AVM (figure 7). The patient is currently symptom-free.

INVESTIGATIONS

- ▶ Ultrasound Doppler
- ▶ CT angiogram
- ▶ Conventional angiogram

OUTCOME AND FOLLOW-UP

Six-month and 1 year follow-up ultrasounds revealed complete resolution of AVM. The patient is currently symptom-free.

DISCUSSION

The first case of AVM was reported in 1926 by Dubreuil and Loubat.⁵

AVMs are very rare but are potentially life-threatening vascular lesions. They may be

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Figure 1 Ultrasound grey scale image: an anechoic lesion in uterus (white arrow). On colour Doppler ultrasound this is tangle of vessels, suggestive of arteriovenous malformations.

congenital or acquired. Most congenital uterine AVMs are isolated anomalies but can occur in association with AVM at other sites.

Acquired AVM may be the consequence of uterine curettage, spontaneous first trimester abortion, caesarian section, gestational trophoblastic disease or endometrial carcinoma.⁶⁻⁷ These are reported in women aged between 18 and 72 years. The true incidence of the uterine malformations is not known.

Doppler ultrasound is the first non-invasive diagnostic modality for its detection and characterisation. Further evaluation and characterisation can be performed on CT with CTA and MRI. Conventional angiography is the gold standard for diagnosis and catheter embolisation is becoming the effective first-treating option.⁸⁻¹⁰

An extensive systematic review and meta analysis performed by Peitsidis *et al*¹¹ on iatrogenic acquired AVM included 91

studies, 85 case reports and 6 observational studies. In 17 patients (17%), recurrence occurred after treatment with uterine artery embolisation, similar to our case in which postembolisation AVM had reoccurred.

They further reported that ultrasound imaging was applied in 86 patients (86%), and ultrasound combined with angiography was performed in 51 patients (51%). Peitsidis *et al*⁶ reported that uterine artery embolisation (UAE) was the most frequent option provided in 59 patients (59%). Total abdominal hysterectomy was performed in 29 patients (29%). Spontaneous resolution of AVM occurred in 6 patients (6%). Twenty-four articles reported pregnancies in 27 patients (27%).

Akbayir *et al*¹¹ reported a case of caesarean scar pregnancy with AVM. This 38 year-old woman with a history of two caesarean sections and one D and C, presented with heavy painless vaginal bleeding. Diagnosis of ectopic pregnancy was

Figure 2 On Doppler spectral ultrasound scanning, swirling turbulent flow with abnormal waveform predominantly arterial noted within this lesion (white arrow).

Figure 3 CT Angiography and three-dimensional reconstruction, uterine vascular malformation (long arrow). Predominant arterial supply of this arteriovenous malformations was noted from left ovarian artery (curved arrow) and with small branches coming from the left uterine artery (arrow head). Early filling of draining left iliac vein (short arrow).

Figure 5 Conventional angiogram, predominant arterial supply of this arteriovenous malformations was noted from left ovarian artery (white long arrow).

confirmed with ultrasound scanning and elevated β -human chorionic goadotropin levels. Ectopic pregnancy was managed with methotrexate. After 43 days, an AVM developed at the site of scar pregnancy, which was diagnosed with colour Doppler imaging. The diagnosis was confirmed with angiography and successful bilateral UAE was performed.

Molvi SN *et al*⁹ described two cases of uterine AVM presenting with abnormal vaginal bleeding. Both patients had D and C after abortion. Uterine AVM were diagnosed on Doppler ultrasound scanning. They were successfully managed with trans-catheter embolisation.

In our case, patient developed uterine caesarean scar perforation because of two D and C's later after repair of scar perforation she developed AVM at the site of surgically repaired

scar. This patient was successfully treated with angio embolisation and adjuvant gonadotropin agonist and had uneventful recovery.

Figure 4 Super selective angiogram of left uterine artery: hypervascular tangle of vessels (white arrow).

Figure 6 Postangioembolisation showed completely embolised arteriovenous malformations (white arrow).

Figure 7 Complete disappearance of arteriovenous malformations on ultrasound (white arrow).

Learning points

- ▶ Clinical suspicion of arteriovenous malformations should be high when patients present with intractable vaginal bleeding and have previous history of uterine surgical intervention or trauma.
- ▶ Ultrasound Doppler should be considered as primary modality in investigation.
- ▶ Conventional angiography can be considered as effective treatment option and gold standard diagnostic tool.
- ▶ Timely and accurate recognition are crucial to avoid morbidity and mortality related to this rare but treacherous disease.

Contributors KH and IM were involved in initial diagnosis and further radiology workup. RS was involved in angiography work up. AA was primary physician.

Competing interests None.

Patient consent Obtained.

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