

Arteriovenous Malformation of the Pregnant Uterus

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SUMMARY

Arteriovenous malformation of the pregnant uterus is very rare, and may present with unexplained torrential bleeding. We report a patient with absence of the conventional risk factors, and was saved by quick recourse to hysterectomy to control the bleeding.

KEY WORDS:

Arteriovenous malformation, pregnant uterus

INTRODUCTION

Uterine arteriovenous malformation (AVM) is extremely rare, with only less than 80 cases reported worldwide¹. They characteristically present with unexplained, intermittent and torrential bleeding. We report a rare case of arteriovenous malformation of the pregnant uterus.

CASE REPORT

A 36 year old Para 4 at 16 weeks of gestation was admitted with an innocuous complaint of mild lower abdominal pain. She had three previous uneventful vaginal deliveries and denied any history of uterine surgery, pelvic infection or abdominal trauma. Antenatal follow-up was unremarkable.

She became haemodynamically unstable overnight with massive intra-peritoneal bleeding. At emergency laparotomy, bleeding from a pin-point source at the posterior aspect of

uterus was identified with the surrounding myometrium and other intra-peritoneal organs appearing normal. Fine superficial haemostatic sutures around the bleeding point caused profuse bleeding from needle entry and exit points, with further sutures aggravating the bleeding. Hysterectomy was performed when bilateral uterine artery and internal iliac artery ligation failed to secure haemostasis.

A relaparotomy in the face of continued blood loss, revealed brisk bleeding from both ovarian hila requiring bilateral salphingoopherectomy and intra-abdominal packs to achieve haemostasis. She had further morbidities in the form of adult respiratory distress syndrome, disseminated intravascular coagulopathy and burst abdomen but recovered after appropriate intervention.

Serial sections of the gross specimen showed many small dilated and congested vascular spaces predominantly at the posterior wall of uterus. No evidence of placental hemorrhage, infarction, or retroplacental clots was noted.

Histopathological examination showed many ectatic vessels in the posterior uterine wall and both ovarian hila. It was a combination of thin and thick walled blood vessels (mostly of cavernous type) accompanied by focal areas of hemorrhage. Most of these vessels were malformed and displayed muscular disruption with incomplete muscular wall. In areas where the vessels were much dilated and thin walled, the uterine stroma formed part of the vessel wall. Arterialized veins were also

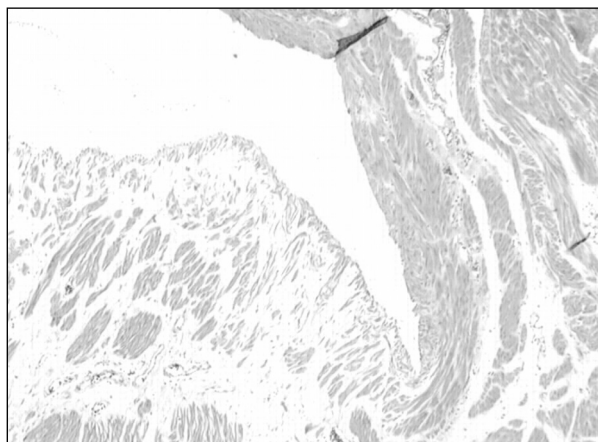


Fig. 1: (H&E X25). An incomplete and very thin venous wall, with part of the wall formed by uterine stroma

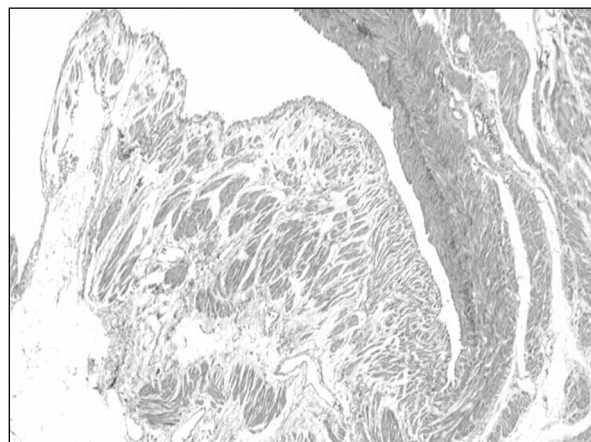


Fig. 2: (EVG stain x 25) Picture showing an arterialized vein. The elastica can be seen beneath the thickened intima

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present. No significant pathology was seen in the placental tissue and the umbilical cord.

DISCUSSION

The histopathological examination was typical for a conclusive diagnosis of AVM of the pregnant uterus. These are abnormal communications between the arteries and veins. The veins receive blood directly from the arteries without the intervening capillary beds and channels. Hence they operate and receive blood at higher pressures. They have intimal fibrous thickening, whereas the arteries are abnormally swollen, thin and lack smooth muscle and elastin. There is abnormal capillary proliferation next to the anomaly.

Events that predispose a uterus to AVM include uterine surgery, endometrial evacuation and infective processes like endometritis, septic abortions and infected retained products of conception. The normal artery-vein channels are disrupted and re-anastomosis occurs in a suboptimal/abnormal tissue milieu. Hormonal changes, mainly pregnancy, menstruation and high dose estrogen-progestin trigger bleeding episodes. However the exact mechanism is still unknown. AVM can also involve the ovaries, as seen in this case.

Other clinical presentations include late post-partum haemorrhage, abnormal bleeding after miscarriage and post-procedural bleed, usually refractory to conventional methods. Most patients have one or more of the following signs and symptoms before they present with bleeding; pelvic pain radiating to the back, vagina or down the posterior aspect of the leg on the same side, pulsatile mass with bruit detectable vaginally or abdominally, leg edema and cardiac decompensation².

Torres *et al* first described the use of grey scale ultrasonography in attempting to diagnose uterine AVM, demonstrating anechoic spaces within myometrium³. The advent of Doppler technology enhanced the diagnostic capabilities. Intensely vascular tangles of tortuous vessels with high velocity and multidirectional flow in a low resistance area within the anechoic spaces are typical.

The gold standard diagnostic tool is angiography, where presence of bilateral hypertrophied uterine vessels, large feeding vessels, and a tortuous hypertrophic arterial mass draining early into massively enlarged hypertrophic veins are very suggestive findings. It also allows for therapeutic intervention in the form of embolization utilizing polyvinyl alcohol particles, in the same setting.

Management principles take into account haemodynamic stability, degree of blood loss and desire for future fertility. After initial stabilization, angiography is advocated if facilities are available (with embolization in the same setting). Only ten successful pregnancies, after embolization have been reported till 2004⁴. Some consider it as an absolute contraindication for pregnancy, citing the inability of the weakened myometrium to contain a pregnancy. Surgical removal of AVM has been attempted with the intent of preserving fertility function. Hysterectomy remains a popular and safe choice.

CONCLUSION

AVM of uterus, although rare, must be considered in unexpected, intermittent torrential bleeding after delivery or a procedure on the uterus. Angiography and embolization should be performed in stable patients when the bleeding is not heavy and when there is a high index of suspicion. Otherwise most cases will only be diagnosed after a hysterectomy.

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