Prompt Diagnosis and Treatment of Uterine Arcuate Artery Pseudoaneurysm: A Case Report and Review of Literature

ABSTRACT
Post-partum haemorrhage is a major determinant of maternal mortality. Traditionally, cases of post-partum haemorrhage caused by arterial injuries were managed by caesarean hysterectomies or bilateral internal iliac artery ligations. The diagnosis of aneurysms or arteriovenous malformations of uterine artery are often missed. Uterine curettage, caesarean section or vaginal delivery can result in uterine vascular anomalies like pseudo aneurysms, arteriovenous malformations, arteriovenous fistula and rupture of uterine vessels. Colour Doppler ultrasound pelvis allows detection of these vascular abnormalities. It helps in differentiating the vascular abnormalities that require embolization from non–vascular abnormalities which can be managed by uterine curettage. Vessel malformations can be treated safely with transcatheter uterine artery embolization, but they can develop disastrous consequences with inadvertent uterine curettage. Transcatheter uterine artery embolization after pelvic angiography is the treatment of choice for uterine artery malformations and it has the advantage of preserving the reproductive capacity. We recommend a routine use of colour Doppler ultrasound pelvis for evaluation of abnormal uterine bleeding.

Key words: Angiography, Doppler, Post partum, Haemorrhage, Uterine, Pseudoanerysm, Embolization, Arteriovenous, Malformation

BACKGROUND
Vascular supply of the uterus is primarily the uterine artery, which approaches the uterus at the level of cervix and lower uterine segment. Branches of uterine artery, the arcuate arteries extend inwards and run circumferentially between the outer and the middle third of myometrium. The radial arteries arise from the arcuate arteries and are directed towards the uterine cavity to become spiral arteries in the endometrium. Uterine venous channels follow a course similar to that of arteries. Colour Doppler ultrasound shows a circular pattern of blood flow signals in the outer myometrium, from the arcuate arteries and venous plexus and a radial pattern of blood flow in the middle and inner myometrium, from the radial and spiral arteries and the accompanying veins.

A uterine artery pseudo aneurysm develops when artery is lacerated or injured. While maintaining contact with the parent vessel, extravasated blood dissects through the tissues. The boundaries of the pseudo aneurysm are constituted by thrombus, as opposed to the three arterial layers of a true aneurysm. An aneurysm expands with each pulsation and it finally ruptures inside the uterine cavity, causes a clinically visible haemorrhage. In the neck of the pseudo aneurysm, the to and fro pattern may be potentially identified on Doppler, because the arterial blood flows like a jet into the aneurysm cavity during the systole, then reverses (backward flow) into the original artery during the diastole. This is in contrast to arteriovenous malformation where blood flows throughout [1]. This pattern is explained by the pressure gradient between a distended high pressure pseudo aneurysm and a low pressure artery during diastole. The feeding arteries and outline can be confirmed by detector Computed Tomography [2].

CASE PRESENTATION
A 25-year-old lady presented to the emergency department with complaints of continuous bleeding per vaginum for 30 days, following oral abortifacients. She was known to be 2 months pregnant but was unsure of the dates of her last menstrual period. She was gravid 2 and parity 1, having had a spontaneous vaginal delivery one year ago. She was breastfeeding her one–year–old child. She was haemodynamically stable. There was a mild pallor. Lower abdomen was soft. On speculum examination, cervical os was found to be closed and bleeding through os was present. On vaginal examination, the uterus was found to be soft, anteverted and slightly bulky. Adnexal palpation revealed normal fornices. Her urinary human chorionic gonadotropin was positive.

Transvaginal ultrasound confirmed a post–aboratal uterus. There was no evidence of residual placental tissue in uterine cavity and endometrial thickness was 9 mm. Grey scale showed a hypoechoic lesion measuring 2 × 1.5 cm in the left lateral wall of uterus. Colour Doppler sonography showed yin and yang blood flow pattern within the body, suggestive of pseudoaneurysm [Table/Fig-1]. CT...
angiography with non ionic contrast showed early contrast filling of the lesion within the uterus [Table/Fig-2]. Volume rendered image and delayed CT image [Table/Fig-3] confirmed the diagnosis of pseudoaneurysm.

To preserve the fertility in this young patient, a transcatheter uterine artery embolization of pseudoaneurysm was planned. She underwent digital subtraction angiography. Arteriography revealed a pseudoaneurysm from the terminal part of left uterine artery. In addition, the left uterine artery was tortuous and hypertrophied. Left uterine artery was selectively embolized with a mixture of gel foam and contrast media. The right uterine artery was also embolized. A post–embolization angiographic study was performed to ensure complete occlusion of vessels. Follow up colour Doppler showed aneurysmal cavity filled with echogenic content, with no evidence of blood flow. The patient is asymptomatic at present.

Post–abortal haemorrhage is a major cause of maternal mortality. Secondary PPH is defined as excessive bleeding which starts after 24 hours of delivery to 6 weeks postpartum [3]. Common causes include retained products of conception, sub–involution of placental bed and endometritis [4]. Rare causes include pseudoaneuerysms of uterine artery, arteriovenous malformations and choriocarcinoma. Colour Doppler should be added to all cases of unexplained bleeding per vagina, as there are cases of retained villi associated with arteriovenous malformations [5].

In 1979, Brown et al., reported the first case of selective arterial embolization which was done to successfully treat an extra–uterine pelvic haematoma after three failed surgical attempts which were made to control bleeding [6]. Arterial embolization has been successfully used to control post–partum bleeding which results from uterine atony, placenta accreta and vaginal haematomata. The efficiency and safety of selective uterine artery embolization was evaluated by Pelage et al., in women with delayed post–partum haemorrhage [7]. In their series of 14 women, 2 were found to have pseudoaneuerysms. Immediate resolution of external bleeding was observed after embolization. No complication related to invasive treatment was reported, while other authors have described complications, including muscle pain and bladder necrosis [8].

A true aneurysm has all three layers of arterial wall, whereas a pseudoaneurysm has a wall which is made of thrombus. The differential diagnosis of pseudoaneurysm includes acquired arteriovenous malformation, arteriovenous fistula and direct vessel rupture. Arteriovenous malformations are characterized by multiple communications of varying sizes between arteries and veins, which can be congenital or acquired. Congenital uterine malformations are caused by abnormalities in the embryonic development of primitive vascular structures, whereas acquired arteriovenous malformations consist of multiple small arteriovenous fistulas between intramural arterial branches and venous plexus. Acquired arteriovenous malformations commonly occur following dilatation and curettage, uterine surgery or trauma to the uterus.

Colour Doppler demonstrates to and fro sign in the neck of the pseudoaneurysms and yin yang sign in the body of the pseudoaneurysm. Arteriovenous malformations are characterized by marked aliasing on colour flow doppler and early venous drainage on spectral doppler evaluation.

In a small series of women who underwent embolotherapy for obstetrical haemorrhage, all three women who attempted conception after embolization were found to be successful. Of the 3 women, 2 had bilateral uterine artery embolization [9]. Jung Hyek Kwon et al., in their series of 24 iatrogenic uterine arterial injuries, found 7 cases with pseudoaneurysms, nine with acquired arteriovenous malformations, 6 cases of combined arteriovenous malformations and pseudoaneurysms and two cases of uterine branch rupture. Uterine artery embolization was successful in 22 cases. Two cases were managed by hysterectomy after embolization failure. Causes were failure were summed due to extra feeding arteries such as internal pudendal artery, ovarian artery and inferior epigastric artery [5].

Burshell demonstrated that bilateral internal iliac ligation was more effective in reducing pulse pressure than unilateral ligation [10]. Redistribution and hypoxia induced neovascularization allows bleeding to occur, as there is a rich symmetrical supply to all pelvic organs bilaterally. Hence, bilateral uterine artery embolization is safe and more advantageous than unilateral embolization. Cross filling from contralateral side may not be evident immediately at initially therapy. Patient should be followed with colour Doppler immediately after embolotherapy and thereafter, at 3 months intervals for 1 year and then yearly for three years.
In their study on 24 cases, JH Kwon et al., found no recurrences. Sometimes, ultrasound cannot demonstrate retained villi or extra uterine feeding arteries. Therefore, serum beta test and a meticulous search for possible feeding arteries during angiography are recommended.

Initial pelvic angiography is followed by selective internal iliac angiography. Embolic materials are carefully introduced into uterine artery or other feeding artery until stasis of blood. Ipsilateral internal iliac angiography is repeated to exclude additional feeding arteries which become apparent only after the major feeding vessel is occluded. Embolization of contralateral uterine artery is performed to prevent cross filling. If bleeding does not stop, other feeding arteries such as ovarian artery, inferior epigastric artery or middle sacral artery should be examined.

For occlusion of proximal vessels in cases of pseudoaneurysms arising from larger branches or large arterovenous fistulas, metallic coils are preferred because of risk of shunting of particulate embolic materials via fistulas into the systemic circulation [11]. Temporary occlusion by using absorbable gelatin sponge and maintaining an extensive collateral circulation, helps in preserving menstrual cycle and fertility.

Pseudoaneurysm is an important vascular abnormality, especially following lower section Caesarean section. Uterine artery embolization is a safe technique and it prevents hysterectomy [12]. Our case demonstrated the importance of routine use of Colour Doppler ultrasound study of pelvis in diagnosing the cause of abnormal uterine bleeding. Uterine arcuate artery aneurysm can often be missed, leading to unfavourable patient outcome.

CONCLUSION

With improved Colour Doppler facilities are combined with clinical expertise, maternal mortality caused by post–abortal haemorrhage can be minimized. Transcatheter uterine artery embolization after pelvic angiography is the treatment of choice for uterine artery malformations and it has the advantage of preserving the reproductive capacity. We recommend a routine use of colour Doppler ultrasound pelvis for evaluation of abnormal uterine bleeding.

LEARNING POINTS

- In women with unexplained vaginal bleeding following normal delivery or abortion, a diagnosis of uterine artery arteriovenous malformation or aneurysm should be considered.
- Uterine artery abnormalities can be detected by colour Doppler and confirmed with angiography.
- Transcatheter arterial embolization is the therapy of choice for uterine artery malformations.
- Embolization of contralateral uterine artery is performed because of possibility of cross-filling, which may not be evident at the time of initial therapy.

CONSENT

We have obtained the patient’s consent for the case report.

COMPETING INTERESTS

We do not have any commercial association that might pose a conflict of interest in connection with the manuscript. We certify that neither this manuscript nor one with substantially similar content under our authorship has been published or is being considered for publication elsewhere.

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