Uterine arteriovenous fistula and right uterine accessory artery as an extremely rare cause of vaginal bleeding

Abstract

3D Doppler ultrasound scan of the uterus is a usefull tool for the diagnostic of uterine arteriovenous fistula. Uterine artery embolisation is the main conservative approach of the condition wich in the case we present was not successfully because an rare associate condition was overlooked (an accessory uterine artery arising from the descending aorta). The catastrophic menorrhagia that occurred imposed emergency hysterectomy and vascular surgery for the accessory uterine artery ablation.

Keywords: *uterine arteriovenous malformation; uterine artery embolization; massive vaginal bleeding; 3D Color Doppler Ultrasound*

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Case report

A 33 years old woman was admitted in our hospital for heavy vaginal bleeding followed by hemorrhagic shock, which underwent embolization of the uterine artery two months earlier.

The patient had a history of a vaginal delivery followed by two uterine curettages for persistent bleeding: the first one, a couple of hours after delivery, and the second one after the next 5 days. The presence of placental remains at the curettage could not be proved.

Two subsequent pregnancies at 3 and 4 years later ended as early intrauterine embryo demise, and both uterine instrumental evacuation were followed by heavy uterine bleeding.

The patient used contraceptive combined pills for a couple of years but the last three menses were very heavy and lasted more than 7 days.

A transvaginal scan raised the suspicion of an arteriovenous intrauterine fistula. On gray-scale ultrasound we observed a slightly enlarged uterus with a hypo echoic irregular image next to the uterine cavity (Figure 1) which turned to be a vascular structure when Doppler mode was used. Three-dimensional Color Doppler rendering mode revealed a bundle of vessels with arteriovenous communication (Figure 2). Color Doppler mode also showed a very large venous circulation next to the uterus, at the base of both broad ligaments.

The patient had a pelvic MRI that indicated an increased and abnormal vascular pattern in the uterus and in the pelvis (enlarged uterus with many venous dilatations, forming a tumor of 2 cm diameter, with intramiometrial evolution, and also periuterine dilatations, with normal ovaries and cervix. The venous periuterine dilatations were extending to the broad ligaments, around the salpinx, cervix and vagina) (Figure 3).

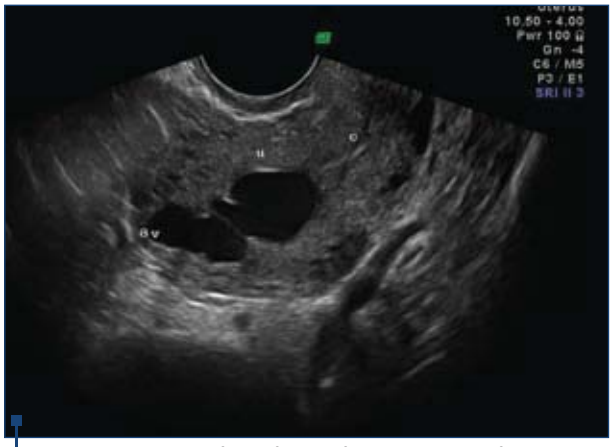

Figure 1. 2D transvaginal scan showing the UAVM as an anechoic structure inside the uterine wall

In order to confirm the diagnosis, the patient was referred to the only service that performed uterine artery embolization and angiography at the same time. According to the angiography protocols, selective catheterization of both uterine arteries and embolization with 2 platinum spirals Matrix 2 Boston scientific of 5X12 respectively 7X20, and additionally gelaspon injection in both arteries were performed (Figure 4).

Transvaginal ultrasound performed 14 days after the embolisation indicated insignificant changes of the uterine malformation image.

In a couple of days after the last ultrasound scan the patient was admitted in the Emergency Unit for massive vaginal bleeding and the aortograhy injection of Pig-tail Fr5, showed arteriovenous malformation of the uterus of 4/3 cm, with main irrigation from an

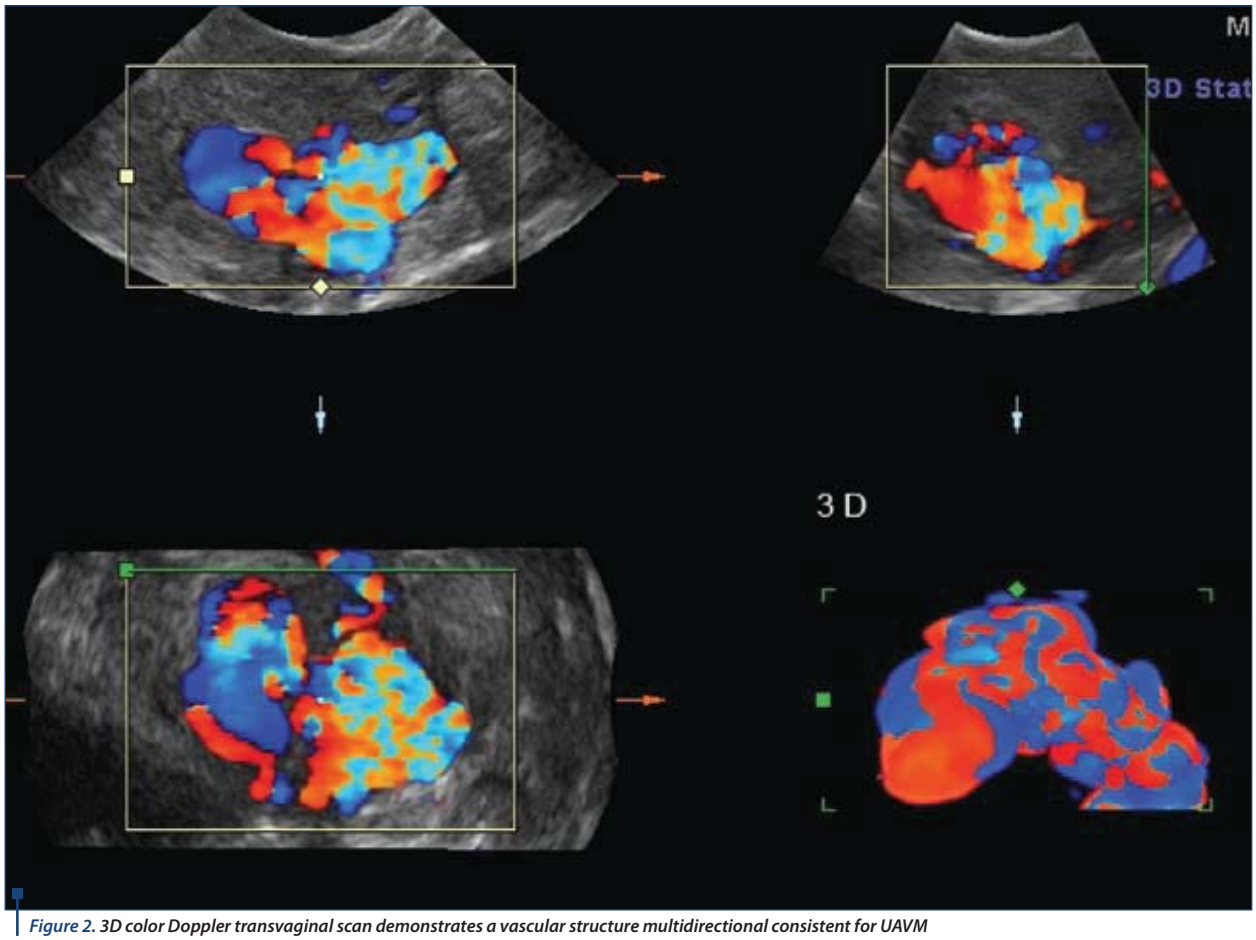

Figure 2. 3D color Doppler transvaginal scan demonstrates a vascular structure multidirectional consistent for UAVM

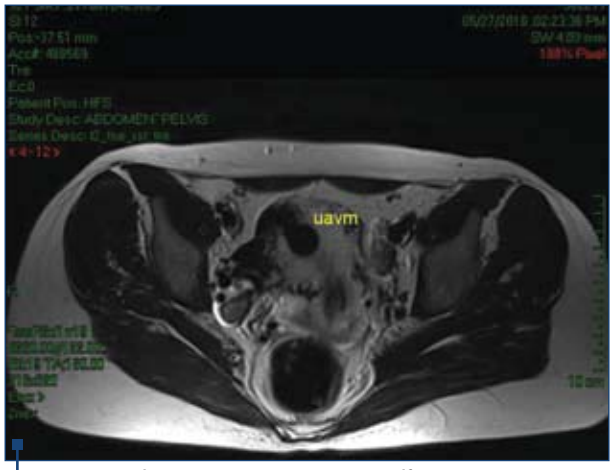

Figure 3. MRI showing uterine arteriovenous malformation

accessory artery arising from the distal aorta under the last lumbar artery on the right and returning circulation via inferior cava vein (Figure 5).

The massive vaginal bleeding that followed in a couple of minutes after the angiography, imposed emergency surgery (total abdominal hysterectomy and ligation of the accessory uterine artery were carried out). The sequence of surgical procedure was imposed by the necessity of haemostasis and immediate unavailability of the vascular surgeon. The abdomen was opened through a lower midline incision. In the peritoneal cavity we found both fallopian tubes bleeding (about 30 ml), normal size, colorless, uterus (40/30/30 mm) and also normal ovaries; the right infundibulopelvic ligament appeared to be of a much increased volume with many arterial and venous dilations; at this level, an intraperitoneal, right internal parametrial ascending path of a 7 mm diameter artery with origin at 1 cm below aortic bifurcation, on its anterior side with an aneurismal dilation of 5 mm diameter at this level. We performed total hysterectomy with bilateral salpingo-oophorectomy. We proceeded to the dissection, ligation and sectioning of the accessory arterial path followed by its total retroperitoneal excision. We also proceeded to the dissection of the right uterine accessory vein (1 cm diameter) which was flowing into the inferior cava vein, 2 cm next to the right renal vein.

Post-operative evolution was good in terms of surgery, and the patient was discharged after 5 days.

Anatomical examination of the uterine specimen confirmed the arteriovenous malformation in the ute-

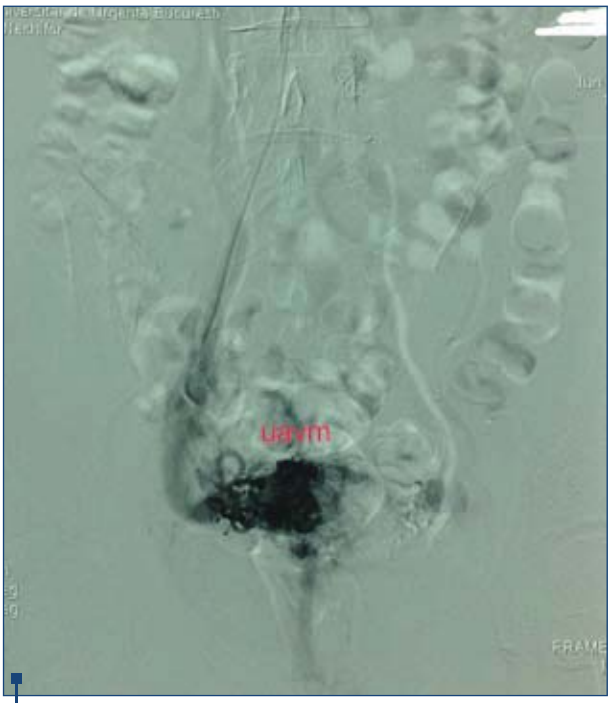

Figure 4. Arteriography of the pelvis after uterine artery embolization

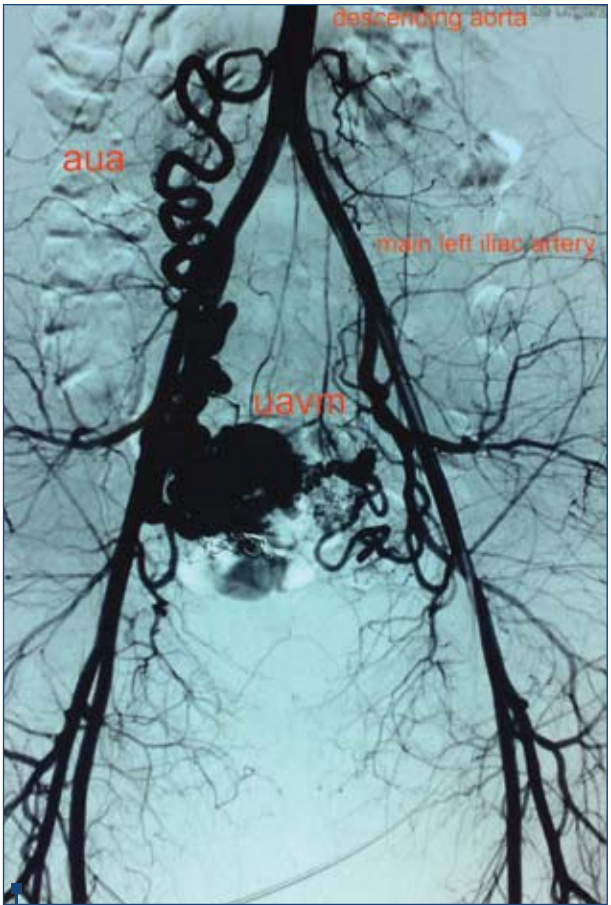

Figure 5. Arteriography of the pelvis after uterine artery embolisation

rine wall communicating with the cavity through a 2 cm square continuity solution.

Discussion

Uterine arteriovenous fistula (UAF), also known as uterine arteriovenous malformation (UAVM) is a rare cause of vaginal bleeding. The literature reports several cases of such rare conditions which can be congenital or acquired (occurring mainly after surgery or obstetrical procedures - trophoblastic disease)[3,5].

UAF can be manifested by abnormal vaginal bleeding with heavy menses or intermenstrual bleeding sometime with late onset after the incriminating event (pregnancy or surgery). Other symptoms include pelvic pain or low urinary tract complaints[2,4]. UAF can be involved in the etiology of pregnancy loss and also in massive bleeding subsequent to pregnancy loss[1]. Literature suggests that menorrhagia or menometrorrhagia is produced by the disruption of the epithelium covering the vessels in certain situations as menstruation or curettage[6].

Angiography was considered to be the gold standard diagnostic method for UAF but because of its invasive characteristic it was gradually replaced by Color Doppler ultrasound scan[7]. Gray-scale transvaginal or transbdominal ultrasound scan displays anechoic intrauterine masses of irregular shape that can be isolated from the endometrium, localized in the uterine wall thickness[4]. In UAF the diagnose can be made by transvaginal Color Doppler ultrasound that indicates a bundle of blue and red threads with circulation in both senses, high velocity and low resistance[5,6].

Considering the obstetrical history of the patient, we can presume that the fistula had a traumatic ethiology, being produced after the vaginal birth when the two curettage were performed. The fistula could also explain the spontaneous abortion and the heavy bleeding that followed. ■

References

1. Arredondo-Soberon F, Loret deMola JR, Shlansky-Goldberg R, Tureck RW. Uterine arteriovenous malformation in a patient with recurrent pregnancy loss and a bicornuate uterus: a case report. J Reprod Med 1997; 42: 239-243.
2. Huang MW,Muradali D, Thurston WA, Burns PN,Wilson SR.Uterine arteriovenous malformations: gray-scale and Doppler US features withMR imaging correlation. Radiology 1998; 206: 115-123.
3. S.M. Kelly, A.M. Belli, S. Campbell Arteriovenous malformation of the uterus associated with secondary postpartum hemorrhage Ultrasound Obstet Gynecol 2003; 21: 602-605.
4. Mungen E, Yergok Y, Ertekin A, Erg ur, Ucmakli E, Ayta¸ clar S. Color Doppler sonographic features of uterine arteriovenous malformations: a report of two cases. Ultrasound Obstet Gynecol 1997; 10: 215-219.
5. Sugiyama T, Honda S, Kataoka A, Komai K, Izumi S, Yakushiji M. Diagnosis of uterine arteriovenous malformation by color pulsed Doppler ultrasonography. Ultrasound Obstet Gynecol 1996; 8: 359-360.
6. Timmerman D, Van den Bosch T, Peeraer K, Debrouwere E, Van Schoubroeck D, Stockx L, Spitz B. Vascular malformations in the uterus: ultrasonographic diagnosis and conservative management. Eur J Obstet Gynecol Reprod Biol 2000; 92: 171-178.
7. Timmerman D,Wauters J, Van Calenbergh S, Van Schoubroeck D,Maleux G, Van den Bosch T, Spitz B. Color Doppler imaging is a valuable tool for the diagnosis and management of uterine vascular malformations. Ultrasound Obstet Gynecol 2003; 21:570-577.