

## **EMBOLIZATION OF AN INTRAUTERINE ARTERIOVENOUS MALFORMATION - A CASE REPORT -**

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### **ABSTRACT:**

*PURPOSE. TO REPORT A CASE OF A YOUNG WOMAN WITH RECURENT EPISODES OF HEAVY VAGINAL BLEEDING THAT ALTERNATED WITH PERIODS OF MODERATE BLEEDING. THE PATIENT HAS BEEN DIAGNOSED WITH UTERINE ARTERIOVENOUS MALFORMATION IN THE RADIOLOGY DEPARTMENT OF THE UNIVERSITY EMERGENCY HOSPITAL BUCHAREST. GONADOTROPIN-RELEASING HORMONE AGONISTS HAVE BEEN USED AS AN ADJUNCT TO EMBOLIZATION. SUBSEQUENT UTERINE VEINS EMBOLIZATION RESULTED IN COMPLETE DISAPPEARANCE OF THE ARTERIO-VEINUS MALFORMATION, AND NORMAL CYCLES WERE RESUMED TWO MONTHS LATER.*

*DESIGN. CASE REPORT*

*PATIENT. 28 YEARS OLD WOMAN WITH RECURRENT METRORRHAGIAS*

*EXPECTED OUTCOME. DISAPPEARANCE OF AN INTRAUTERINE ARTERIOVENOUS MALFORMATION TREATMENT. GONADOTROPIN-RELEASING HORMONE AGONISTS AND UTERINE VEINS EMBOLIZATION*

*DIAGNOSIS. UTERINE ARTERIOVENOUS MALFORMATION*

*CONCLUSION. SUBSEQUENT UTERINE VEINS EMBOLIZATION RESULTED IN COMPLETE DISAPPEARANCE OF THE ARTERIO-VEINUS MALFORMATION, AND NORMAL CYCLES WERE RESUMED TWO MONTHS LATER.*

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**KEY WORDS:** ARTERIO-VEINUS MALFORMATION, UTERINE VEINS EMBOLIZATION.

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**DISCLOSURE:** All authors contributed equally in developing this study.

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## INTRODUCTION

Arterio-venous malformations (AVMs) can occur in any organ in the body, including the pelvic vasculature and rarely in the uterus. The first case of AVM was reported in 1926<sup>8</sup>. Uterine arteriovenous malformation is a rare but potential life-threatening source of bleeding. These consist a mixture of arterial, venous and small capillary-like channels with fistulous connections. Uterine AVMs can be acquired or congenital<sup>9</sup>. AVMs have been reported in patients from 18 to 72 years old but only rarely in nulliparous women<sup>10</sup>.

Congenital uterine AVMs originate from an abnormality in the embryological development of primitive vascular structures, resulting in multiple abnormal communications between veins and arteries<sup>11</sup>.

Acquired uterine AVMs result especially from dilatation and curettage, direct uterine trauma or uterine surgery. Very rare acquired AVMs result from endometrial or carcinoma and gestational trophoblastic disease<sup>6</sup>. Acquired AVMs are small arteriovenous fistulas between myometrial venous plexus and intramural arterial branches. They appear as a vascular tangle<sup>12</sup>.

The classical symptomatology of uterine AVMs is severe uterine bleeding with no obvious cause, and the bleeding results from a spontaneous vessel rupture or triggered by a dilatation and curettage. The precise diagnosis is very important. Before modern diagnostic methods, diagnosis was made after hysterectomy and histopathologic examination. Today, angiography is the gold standard for diagnosis<sup>13</sup>.

Doppler ultrasonography is also one of the best noninvasive technique, and uterine AVMs show increased vascularity on Doppler and appear as nonspecific heterogeneous or anechoic spaces in the myometrium on gray scale ultrasound<sup>14</sup>.

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In most cases, AVMs are first visualized with sonography because of its widespread use. On angiography, AVMs appear as a complex tangle of vessels supplied by enlarged feeding arteries and show early venous drainage during the arterial phase<sup>16</sup>. Computed tomography scanning with contrast and magnetic resonance imaging are the modalities of

<sup>8</sup> Dubreuil, G., Loubat, E. Aneurysme crivoide de l'uterus. *Ann Anat Pathol.*3:697-718. (1926)

<sup>9</sup> Majmudar, Bhagirath, Nadereh Ghane, Ira R. Horowitz, and David Graham. "Uterine arteriovenous malformation necessitating hysterectomy with bilateral salpingo-oophorectomy in a young pregnant patient." *Archives of pathology & laboratory medicine* 122, no. 9 (1998): 842; Huang, Madeline W., Derek Muradali, W. A. Thurston, Peter N. Burns, and Stephanie R. Wilson. "Uterine arteriovenous malformations: gray-scale and Doppler US features with MR imaging correlation." *Radiology* 206, no. 1 (1998): 115-123.

<sup>10</sup> Diwan, Rajni V., James N. Brennan, Mostafa A. Selim, Thomas L. McGrew, Fouad A. Rashad, Marilou U. Rustia, and Errol M. Bellon. "Sonographic diagnosis of arteriovenous malformation of the uterus and pelvis." *Journal of Clinical Ultrasound* 11, no. 5 (1983): 295-298.

<sup>11</sup> Huang, Madeline W., Derek Muradali, W. A. Thurston, Peter N. Burns, and Stephanie R. Wilson. "Uterine arteriovenous malformations: gray-scale and Doppler US features with MR imaging correlation." *Radiology* 206, no. 1 (1998): 115-123.

<sup>12</sup> Chen, Yan, Guoyun Wang, Fubo Xie, Bo Wang, Guowei Tao, and Beihua Kong. "Embolization of uterine arteriovenous malformation." *Iranian journal of reproductive medicine* 11, no. 2 (2013): 159

<sup>13</sup> Grivell, Rosalie M., Kym M. Reid, and Amy Mellor. "Uterine arteriovenous malformations: a review of the current literature." *Obstetrical & gynecological survey* 60, no. 11 (2005): 761-767.

<sup>14</sup> Müngen, E. "Vascular abnormalities of the uterus: have we recently over-diagnosed them?." *Ultrasound in Obstetrics & Gynecology* 21, no. 6 (2003): 529-531.

<sup>15</sup> Müngen, E. "Vascular abnormalities of the uterus: have we recently over-diagnosed them?." *Ultrasound in Obstetrics & Gynecology* 21, no. 6 (2003): 529-531

<sup>16</sup> Torres, William E., Peter J. Sones, and F. Maynard Thames. "Ultrasound appearance of a pelvic arteriovenous malformation." *Journal of Clinical Ultrasound* 7, no. 5 (1979): 383-385.



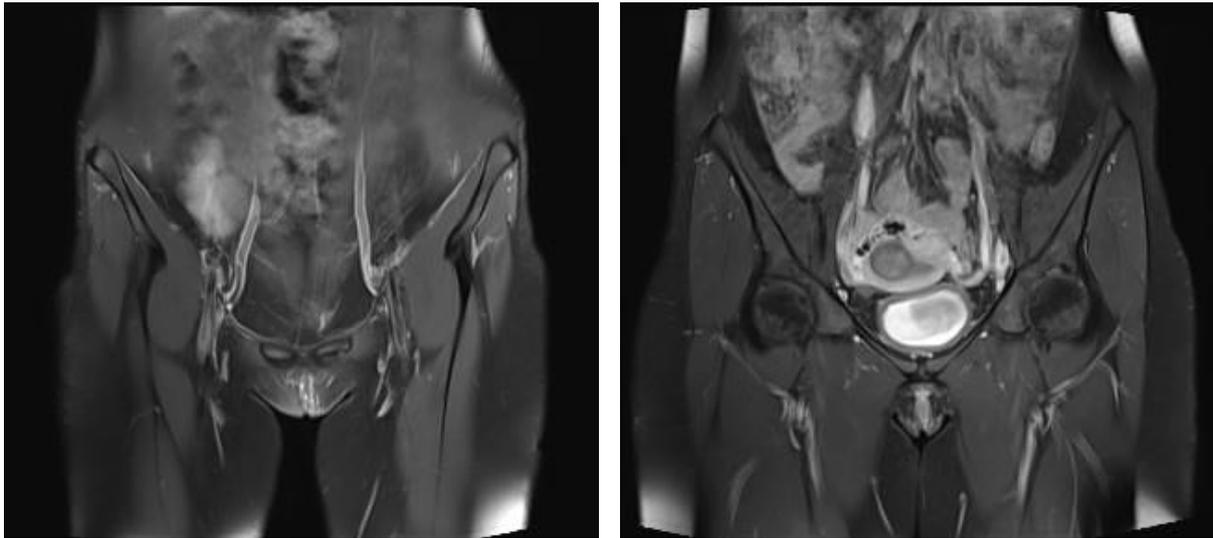


Figure 2 – MRI imaging features of the intrauterine arteriovenous malformation

For confirmation of the diagnosis an angiogram was performed. During hospitalization, the patient was hemodynamically stable with a hemoglobin level of 9.4 g/dL. The uterine artery angiogram confirmed the presence of an arteriovenous malformation in the fundal region of the uterus.



Figure 3 – Angiogram – note the reflux of the contrast substance in the right uterine veins when catheterizing the left uterine artery and the arterio-venous intrauteine malformation

We diagnosed this vascular malformation and recommended gonadotropin-releasing hormone agonists therapy and reevaluation after 3 months.

After informed consent was obtained, a regional anesthetic technique was performed. The right common femoral artery was accessed and a 5F glide catheter was placed through a 5F sheath. Contrast injection demonstrated a serpiginous and dilated arterio-venous structure at the level of the fundus of the uterus. After uterine veins embolization, the patient experienced severe lower abdominal discomfort that required high doses of painkiller and anti-inflammatory medication.

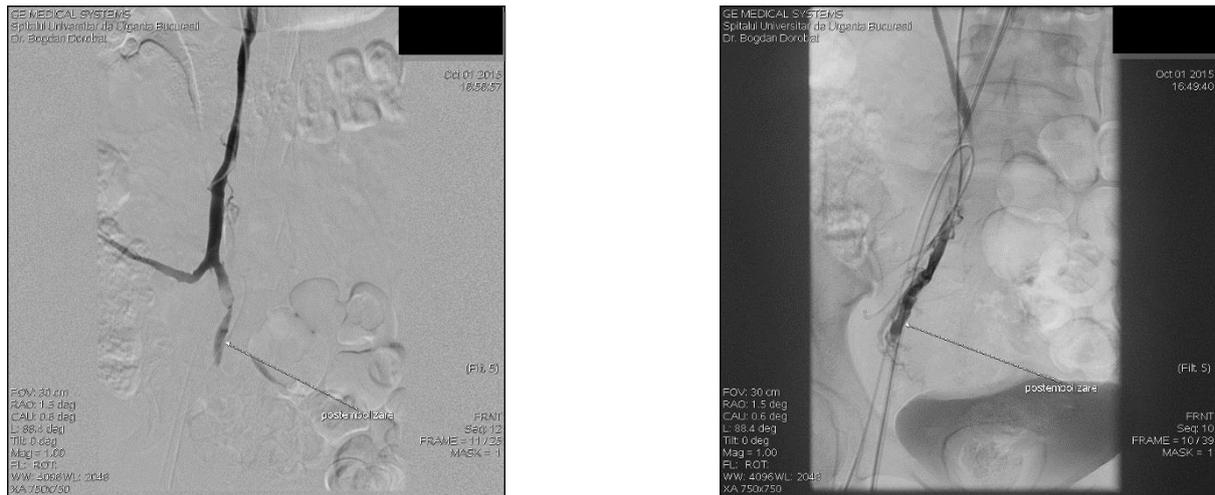


Figure 4 – Angiogram after uterine veins embolization

At a follow-up visit at 1 month after embolization using color Doppler imaging we detected reduced blood flow and a reduced tumor at the level of the uterus. The menstrual cycle of the patient returned to normal two months after the procedure. These results confirmed that the uterine veins embolization was efficient.

## DISCUSSIONS

Dubreuil and Loubat reported the first case of intra-uterine AVM in 1926<sup>18</sup>. Uterine AVMs are uncommon and the true incidence is unknown. The precise diagnosis of AVM is of vital importance because patients present with vaginal bleeding which can cause hemodynamic instability<sup>19</sup>.

We report the case of a patient with a congenital form of uterine AVM given the absence of uterine trauma. However, it is unclear why the patient had no symptoms until now. We can only suspect that the AVM was not superficial or in contact with the basal layer of the endometrium. Traditionally AVMs have been diagnosed by laparotomy or after hysterectomy. Nowadays digital subtraction angiography remains the gold standard of diagnosis<sup>20</sup>.

<sup>18</sup> Fleming, H., A. G. Östör, H. Pickel, and D. W. Fortune. "Arteriovenous malformations of the uterus." *Obstetrics & Gynecology* 73, no. 2 (1989): 209-214.

<sup>19</sup> Singh, N., R. Tripathi, Y. M. Mala, S. Tyagi, S. Tyagi, and C. Singh. "Varied presentation of uterine arteriovenous malformations and their management by uterine artery embolisation." *Journal of Obstetrics & Gynaecology* 34, no. 1 (2014): 104-106.

<sup>20</sup> Milingos, D., D. Doumplis, K. Sieunarine, P. Savage, A. D. Lawson, and J. R. Smith. "Uterine arteriovenous malformation: fertility-sparing surgery using unilateral ligation of uterine artery and ovarian ligament." *International Journal of Gynecological Cancer* 17, no. 3 (2007): 735-737

Gonadotropin-releasing hormone agonists have been used as an adjunct to embolization and in our case 3 months of therapy reduced the size of a uterine AVM from 2.1/2.4 cm to 1/0.5 cm. Subsequent uterine veins embolization resulted in complete disappearance of the AVM, and normal cycles were resumed 3 months later. Especially in young women who desire to preserve fertility, angiographic uterine veins embolization is the preferred therapy for uterine AVMs, because it does not appear to interfere with the menstrual cycle or pregnancy. Other surgical managements are coagulation of the AVM under hysteroscopic guidance, laparoscopic bipolar coagulation of uterine vessels, surgical removal of an AVM and ligation of the uterine artery<sup>21</sup>.

### **CONCLUSION**

AVMs can occur in any organ in the body, including the pelvic vasculature and uterus. We report the case of a young patient with a congenital intrauterine arterio-venous malformation with no symptoms until the age of 28 who was treated using a combined method: gonadotropin-releasing hormone agonists therapy followed by uterine veins embolization.

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<sup>21</sup> Milingos, D., D. Doumplis, K. Sieunarine, P. Savage, A. D. Lawson, and J. R. Smith. "Uterine arteriovenous malformation: fertility-sparing surgery using unilateral ligation of uterine artery and ovarian ligament." *International Journal of Gynecological Cancer* 17, no. 3 (2007): 735-737; Dubreuil, G., Loubat, E. Aneurysme crisoide de l'uterus. *Ann Anat Pathol.*3:697-718. (1926)

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