CEOG Clinical and Experimental Obstetrics & Gynecology

Case Reports

Uterine arteriovenous fistula after perforation during the placement of an intrauterine device – Minimally invasive treatment using uterine artery embolization

W. Kondo[1], M. Tessmann Zomer[1], F.L. Erzinger[2]

[1] Department of Gynecology, Sugisawa Medical Center, Curitiba, Paraná
[2] Department of Vascular Surgery Instituto da Circulação, Curitiba, Paraná; Marcelino Champagnat Hospital, Curitiba, Paraná (Brazil)

Summary

Uterine arteriovenous fistula (AVF) is a rare, but potentially life-threatening condition. Acquired fistulae may occur as a result of trauma or instrumentation, endometrial carcinoma, gestational trophoblastic disease, and intrauterine devices (IUDs). Herein the authors present the case of a 33-year-old woman with a uterine AVF developing after uterine perforation during the placement of a levonorgestrel IUD. The fistula was diagnosed using color Doppler ultrasonography and angiography and the treatment was conducted by minimally invasive approach using uterine artery embolization.

Key words: Uterine arteriovenous malformation; Arteriovenous fistula; Intrauterine device; Uterine artery embolization.

Introduction

Uterine arteriovenous malformation (AVM) is considered a rare condition; however, it may lead to severe life-threatening situations as a result of profuse or irregular bleeding from the abnormal communication between artery and vein [1]. It may be classified as congenital and acquired [2]: (a) the former develops from failures in the embryological differentiation, leading to multiple abnormal vascular connections [3] and (b) the latter is typically a result of trauma or instrumentation, although it has also been associated with endometrial carcinoma, gestational trophoblastic disease, diethylstilbestrol exposure, and intrauterine devices (IUDs) [4-7].

Historically, the diagnosis of uterine AVM was only possible through exploration per laparotomy or upon pathological examination of the uterine specimen after hysterectomy had been performed. Subsequently, angiography became the gold standard for diagnosis. More recently, color Doppler ultrasonography has been suggested for obtaining a reliable diagnosis [8]. The ultrasonographic diagnosis of AVM is based on the presence of hypoechoic tortuous spaces in the myometrium demonstrating vascular flow as evidenced by color Doppler [1, 9, 10]. MRI and CT scan may be useful to assist the diagnosis in cases where angiography was not able to define adjacent organ involvement [11-13].

Traditionally, hysterectomy was the treatment of choice [1]. Other treatment options include ligation of major vessels, ligation of the iliac artery [14], ligation of AVM vessels [15], and ligation of uterine artery [16]. Recently, uterine artery embolization has been successfully used in more than 50% of the patients [10].

In this paper, the authors report one case of iatrogenic acquired uterine arteriovenous fistula (AVF) after uterine perforation during the placement of an IUD treated by means of uterine artery embolization. The paper was approved by the local Institutional Review Board.

Case Report

A 33-year-old woman, G3 A3, came to the present authors' service with sonographic findings indicating a uterine arteriovenous fistula. Four months prior, she had a uterine perforation during the placement of a levonorgestrel IUD. She presented persistent vaginal bleeding for the following four days and transvaginal ultrasound showed a normal-sized uterus with the IUD penetrating the myometrium at the uterine fundus in the right side. There was a pelvic hematoma measuring 37×24×36 mm at the distal end of the IUD and a moderate amount of free liquid in the pelvic cavity. She underwent a laparoscopic procedure for IUD removal and the uterine defect was closed using zero 25 poliglecaprone suture (Figure 1). She was discharged 12 hours after the procedure.

In the postoperative follow-up, the patient developed amenorrhea for the following four months. Transvaginal gray-scale ultrasound revealed cystic hypoechoic areas at the uterine wall;

Revised manuscript accepted for publication November 9, 2015

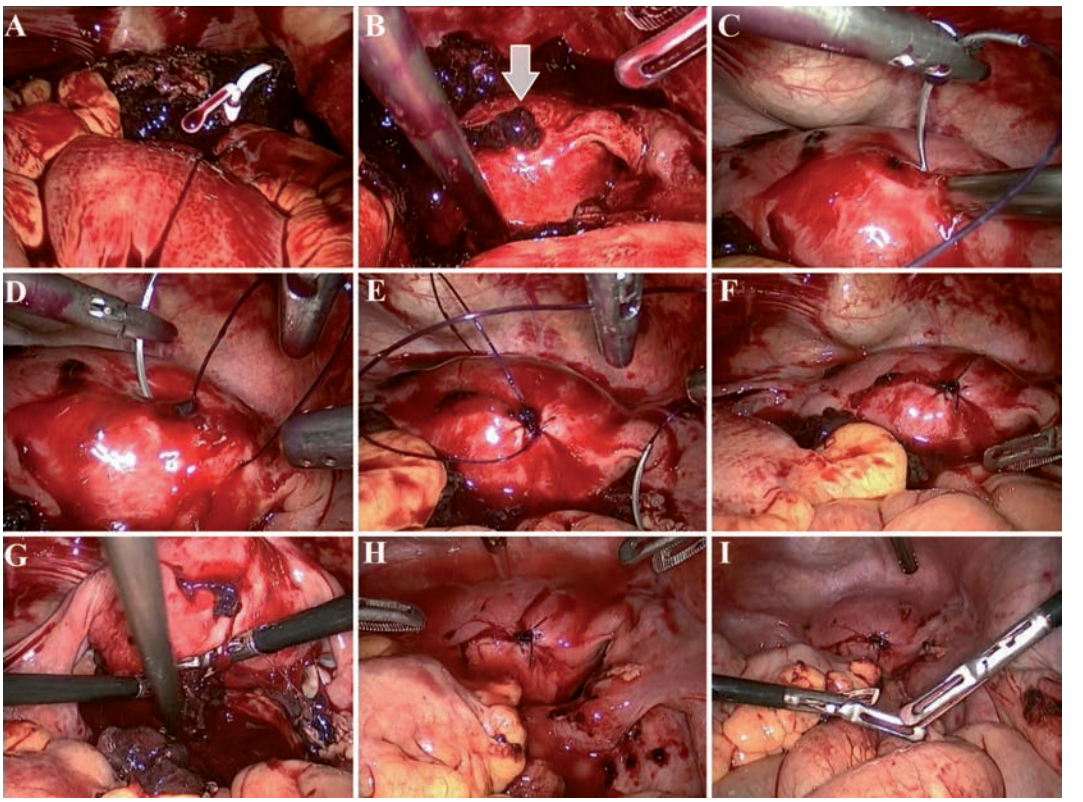

Figure 1. — A: During laparoscopy, the levonorgestrel IUD was found inside the pelvic cavity in the middle of a pelvic hematoma. (B) The hematoma at the uterine fundus. C to F: Suturing the place of perforation at the uterine fundus. G to I: Aspiration of the clots and blood from the pelvic and abdominal cavities and final aspect after the procedure.

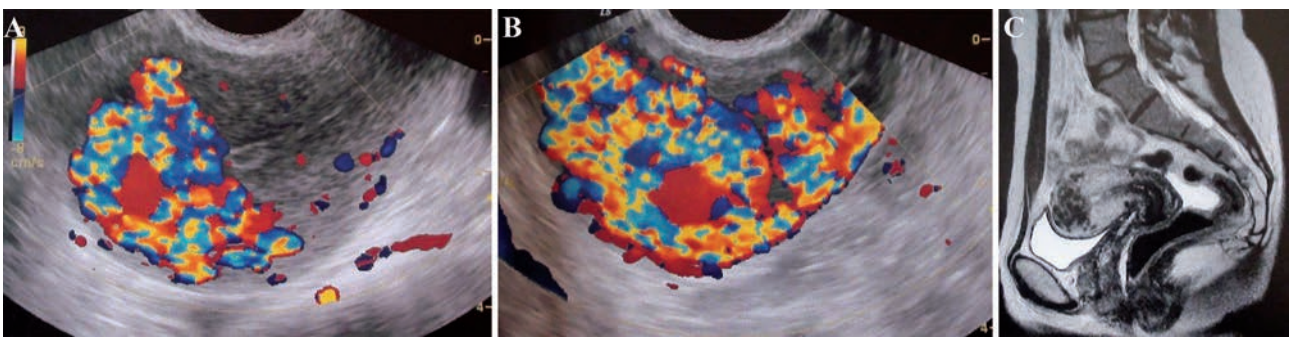

Figure 2. — A and B: Transvaginal ultrasound with color Doppler showing hypervascular areas with turbulent flow at the uterine fundus. C: Pelvic MRI showing the AVF at the uterine fundus.

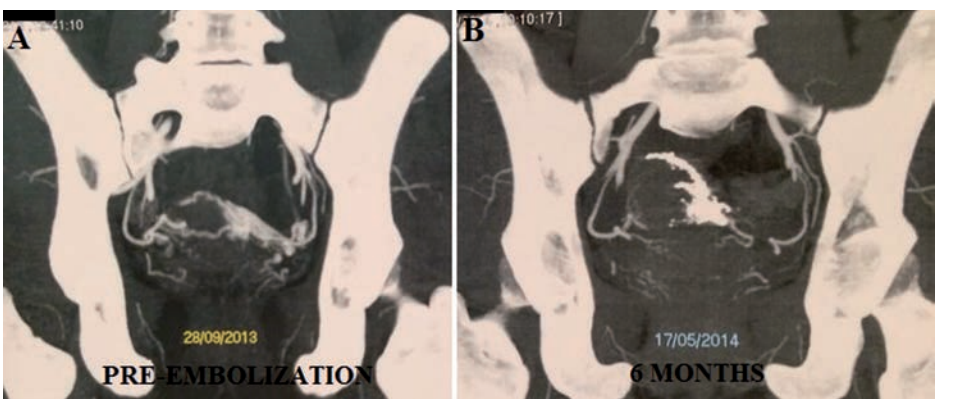

Figure 3. — A: Pre-embolization CT angiography demonstrating the hypervascularization at the left side of the uterus and the uterine fundus, originating from the left uterine artery. (B) Hypervascularization reduction after embolization.

color Doppler showed turbulent flow in the myometrium at the uterine fundus, characteristic features of an AVF (Figures 2A and 2B). Serum level of beta-HCG was below 25 mIU/ml, thus excluding gestational trophoblastic disease.

Pelvic MRI showed periuterine vascular structures affecting the myometrium at the uterine fundus, originating from the superficial layer until the deep layer of the myometrium with loss of the junctional zone (Figure 2C).

CT angiography showed prominence of the uterine arteries, especially at the left side, associated with the presence of vascular structures with significant rise of the arterial phase at the left parametrium and within the myometrium, particularly at the uterine fundus (Figure 3A). These findings were consistent with the possibility of an AVF.

The right femoral artery was punctured under local anesthesia and sedation. After selective catheterization of the left uterine artery, an angiography was performed and confirmed the previous findings of the CT angiography (Figure 4A). Then, a microcatheter was placed inside the artery and the embolization of the left uterine artery was successfully performed using a non-adhesive liquid embolic agent (Figure 4A). The total time of the procedure was 85 minutes.

The patient was discharged on the second day after the procedure using oral analgesia with paracetamol. She was suggested to return to normal activities in 7 days. CT angiography was repeated six months after the embolization confirming the success of the procedure (Figure 3B).

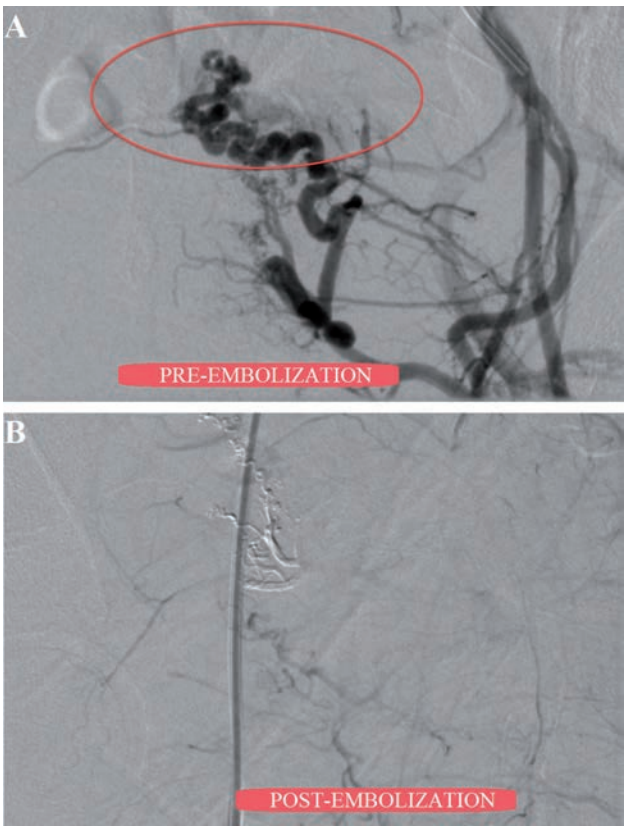

Figure 4. — A: Pre-embolization: aspect of the AVF originating from the left uterine artery. B: Post-embolization radiologic exam with embolic agent showing proximal occlusion of the left branch of the affected segment of the uterine artery with no evidence of the AVF.

Discussion

Uterine perforation during the placement of a levonorgestrel IUD occurs in 0.11% of patients and may be a potentially serious complication of its use [17]. In most cases it may be retrieved from the pelvic/abdominal cavity by laparoscopy but some patients require conversion to laparotomy to repair perforation of adjacent organs [18].

The development a uterine arteriovenous shunt after removal of an embedded IUD has already been described in the medical literature [19]. To the best of the present authors' knowledge, this is the first case reported in the literature of development of uterine AVF after uterine perforation during the placement of levonorgestrel IUD that was removed from the pelvic cavity by laparoscopy.

In the present case, the uterus seemed normal during the laparoscopic retrieval of the levonorgestrel IUD. Although the exact place of the perforation was sutured during the laparoscopic procedure, the AVF subsequently developed at the uterine fundus.

In the majority of the cases, uterine AVFs present as a life-threatening hemorrhage that does not respond to medical treatment [7, 10]. The present patient was asymptomatic, and the uterine AVF was diagnosed during transvaginal ultrasound once she presented amenorrhea, due to the formation of uterine synechiae after the laparoscopic retrieval of the levonorgestrel IUD.

Angiography is considered the gold standard for the definitive diagnosis of AVFs and its goals are to define the vascular anatomy, assess the extent of the vascular fistula, and identify the feeding vessels [1, 20, 21].

Since the first uterine artery embolization procedure reported by Forssman *et al.* [22], there have been numerous reports on the treatment of uterine AVM by this approach [9, 10, 23-25]. Nowadays, uterine artery embolization is the most common treatment option in 59% of the patients with uterine AVM [10]. Several embolic agents may be used: gelfoam, polyvinyl alcohol, steel coils, tris-acryl-gelatin, isobutyl-2-cyanoacrylate, acrylic polyamide, detachable balloons, thrombin, ethanol, and histoacryl [10, 26].

In the present patient, uterine artery embolization was conducted using a non-adhesive liquid embolic agent comprised of ethylene vinyl alcohol copolymer dissolved in dimethyl sulfoxide, and suspended micronized tantalum powder to provide contrast for visualization under fluoroscopy. Onyx® 18 It is safer than other liquid agents because it allows for a slow and controlled injection due to its high viscosity and longer time for polymerization.

Minor complications have been reported after uterine artery embolization, and these relate to minor pelvic pain and postoperative fever [9, 20]. Recurrence after embolization occurred

in 17% of the cases in the review from Peitsidis *et al.* [10].

A tendency of uterine AVMs to proliferate during pregnancy has been reported. Some authors consider the presence of a uterine AVM an absolute contraindication for pregnancy [26]. In the systematic review of Peitsidis *et al.* [10], 17 pregnancies were reported after uterine artery embolizations (17%). All pregnancies resulted in viable healthy neonates, except in one case in which total abdominal hysterectomy was performed in a uterus at 7.4 weeks of gestation due to uncontrolled heavy bleeding [27]. The mean time of delay from treatment to subsequent pregnancy was 15.7 ± 11.7 months with range (2 to 36) months.

In conclusion, uterine AVF may be a potentially serious complication of uterine perforation during IUD placement. The diagnosis of such entity may be done by means of color Doppler ultrasonography and/or angiography and the minimally invasive approach using uterine artery embolization should be considered the first choice of treatment.

References

[1] Hoffman M.K., Meilstrup J.W., Shackelford D.P., Kaminski P.F.: "Arteriovenous malformations of the uterus: an uncommon cause of vaginal bleeding". *Obstet. Gynecol. Surv.*, 1997, *52*, 736.

[2] Beller U., Rosen R.J., Beckman E.M., Markoff G., Berenstein A.: "Congenital arteriovenous malformation of the female pelvis: a gynecologic perspective". *Am. J. Obstet. Gynecol*., 1988, *159*, 1153.

[3] Kasznica J., Nisar N.: "Congenital vascular malformation of the uterus in a stillborn: a case report". *Hum Pathol*., 1995, *26*, 240.

[4] Frencken V.A., Landman G.H.: "Cirsoid aneurysm of the uterus: specific arteriographic diagnosis; report of a case". *Am. J. Roentgenol. Radium. Ther. Nucl. Med*., 1965, *95*, 775.

[5] Cockshott W.P., Hendrickse J.P.: "Persistent arteriovenous fistulae following chemotherapy of malignant trophoblastic disease". *Radiology*, 1967, *88*, 329.

[6] McLachlan M.S., Bird C.C., Naiem E.A., Scott J.S.: "Uterine cirsoid aneurysm". *Br. J. Obstet. Gynaecol*., 1978, *85*, 390.

[7] Cura M., Martinez N., Cura A., Dalsaso T.J., Elmerhi F.: "Arteriovenous malformations of the uterus". *Acta Radiol*., 2009, *50*, 823.

[8] Manolitsas T., Hurley V., Gilford E.: "Uterine arteriovenous malformation - a rare cause of uterine haemorrhage". *Aust. N. Z. J. Obstet. Gynaecol*., 1994, *34*, 197.

[9] Maleux G., Timmerman D., Heye S., Wilms G.: "Acquired uterine vascular malformations: radiological and clinical outcome after transcatheter embolotherapy". *Eur. Radiol*., 2006, *16*, 299.

[10] Peitsidis P., Manolakos E., Tsekoura V., Kreienberg R., Schwentner L.: "Uterine arteriovenous malformations induced after diagnostic curettage: a systematic review". *Arch. Gynecol. Obstet*., 2011, *284*, 1137.

[11] Elia G., Counsell C., Singer S.J.: "Uterine artery malformation as a hidden cause of severe uterine bleeding. A case report". *J. Reprod. Med*., 2001, *46*, 398.

[12] Kwon J.H., Kim G.S.: "Obstetric iatrogenic arterial injuries of the uterus: diagnosis with US and treatment with transcatheter arterial embolization". *Radiographics*., 2002, *22*, 35.

[13] Al-Shekaili K.R., Bhatnagar G., Ramadhan F.A., Al-Zadjali N.: "Arteriovenous malformation of uterus". *Indian J. Pathol. Microbiol*., 2011, *54*, 187.

[14] Antebi E., Adar R., Deutsch V., Mozes M.: "Arteriovenous fistula complicating gynecologic operations". *Obstet. Gynecol*., 1974, *44*, 858.

[15] Chen A.C., Sheu M.H., Hung J.H., Yu K.J.: "Three-dimensional magnetic resonance angiography guidance for conservative surgical management of pelvic arteriorvenous malformation". *Acta Obstet. Gynecol. Scand*., 2003, *82*, 302.

[16] Halperin R., Schneider D., Maymon R., Peer A., Pansky M., Herman A.: "Arteriovenous malformation after uterine curettage: a report of 3 cases". *J. Reprod. Med*., 2007, *52*, 445.

[17] Heinemann K., Westhoff C.L., Grimes D.A., Moehner S.: "Intrauterine Devices and the Risk of Uterine Perforations: Final Results From the EURAS-IUD Study". *Obstet. Gynecol*., 2014, *123*, 3S.

[18] Kho K.A., Chamsy D.J.: "Perforated intraperitoneal intrauterine contraceptive devices: diagnosis, management, and clinical outcomes". *J. Minim. Invasive Gynecol*., 2014, *21*, 596.

[19] Mustonen M., Kauppila A., Lahti R.: "Uterine arteriovenous shunt after removal of an embedded IUD: a case report". *Int. J. Gynaecol. Obstet*., 1978, *16*, 83.

[20] Ghai S., Rajan D.K., Asch M.R., Muradali D., Simons M.E., Ter-Brugge K.G.: "Efficacy of embolization in traumatic uterine vascular malformations". *J. Vasc. Interv. Radiol*., 2003, *14*, 1401.

[21] Maldonado J., Perez C., Rodriguez W.: "Profuse vaginal bleeding seven weeks following induced abortion". *AJR Am. J. Roentgenol*., 2008, *191*, S79.

[22] Forssman L., Lundberg J., Scherstén T.: "Conservative treatment of uterine arteriovenous fistula". *Acta Obstet. Gynecol. Scand*., 1982, *61*, 85.

[23] Batch T., De Graef M., Kassem Z., Otchinnikoff S., Dechaud H., Maubon A., Rouanet J.P.: "Acquired uterine arteriovenous fistula: report of one case". *J. Radiol*., 2004, *85*, 649.

[24] Yang J.J., Xiang Y., Wan X.R., Yang X.Y.: "Diagnosis and management of uterine arteriovenous fistulas with massive vaginal bleeding". *Int. J. Gynaecol. Obstet*., 2005, *89*, 114.

[25] Hasegawa A., Sasaki H., Wada-Hiraike O., Osuga Y., Yano T., Usman S.M., et al.: "Uterine arteriovenous fistula treated with repetitive transcatheterembolization: case report". *J. Minim. Invasive Gynecol*., 2012, *19*, 780.

[26] Grivell R.M., Reid K.M., Mellor A.: "Uterine arteriovenous malformations: a review of the current literature". *Obstet. Gynecol. Surv*., 2005, *60*, 761.

[27] Phoon C.F., Lee S.L., Kanagaligam D., Tan H.C., Tan H.K.: "Subsequent pregnancy outcome after embolisation of uterine arteriovenous malformation". *Ultrasound. Obstet. Gynecol*., 2006, *28*, 601.

Address reprint requests to:
W. KONDO, M.D.
Av. Iguaçu 1236, sala 114
Curitiba, Paraná (Brazil)
e-mail: williamkondo@yahoo.com