Int J Clin Exp Med 2019;12(5):6213-6218
www.ijcem.com /ISSN:1940-5901/IJCEM0084809

Case Report

Prophylactic abdominal aorta balloon placement to control hemorrhage during hysterectomy in a postmenopausal case of uterine arteriovenous malformation: a case report

Yinghan Chen, Tingting Liu, Ling Ouyang

Department of Obstetrics and Gynecology, Shengjing Hospital of China Medical University, Shenyang 110004, China

Received August 30, 2018; Accepted January 10, 2019; Epub May 15, 2019; Published May 30, 2019

Abstract: Uterine arteriovenous malformation (AVM) following gestational trophoblastic neoplasia (GTN) is a rare condition in gynecology, with fewer than 100 cases reported in the literature. It may cause recurrent or life-threatening heavy vaginal bleeding, even after complete resolution of the tumor following chemotherapy. Uterine AVM is usually present in women of reproductive age. This is the first report to describe a postmenopausal woman with uterine AVM associated with GTN. Prior to surgery, the patient had a balloon catheter inserted into the abdominal aorta. A total hysterectomy and bilateral adnexectomy with removal of the uterine AVM was successfully performed without significant blood loss. Preoperative insertion of endovascular balloon occlusion catheters into the abdominal aorta may effectively reduce blood loss and blood transfusion in the uterine AVM surgery.

Keywords: Uterine arteriovenous malformations, gestational trophoblastic neoplasia, balloon occlusion, hysterectomy

Introduction

Arteriovenous malformation (AVM) is an abnormal connection between arteries and veins, not including the capillary system [1]. Common causes of acquired uterine AVM include all kinds of abortion, uterine curettage, caesarean section, myomectomy, cesarean scar pregnancy (CSP), and gestational trophoblastic neoplasia (GTN) [2]. Uterine AVM can cause sudden vaginal bleeding or chronic irregular vaginal bleeding. The bleeding is often recurrent and may occur even after complete tumor remission following chemotherapy [3].

Uterine AVM secondary to GTN is rare and more commonly found in women of childbearing age (the oldest reported age was 49 years) [4, 5]. Since most patients choose to preserve fertility, selective pelvic arteriography and uterine artery embolization are first-line therapies for AVM complicated with vaginal bleeding [6]. Hysterectomy is an effective means for the treatment of heavy bleeding caused by uterine arteriovenous fistula but is mainly suitable for patients without reproductive requirements, poor follow-up conditions, or for whom embolization has failed [7, 8].

Herein, we present a case of a postmenopausal woman with uterine AVM successfully managed by hysterectomy with the prophylactic placement of a balloon catheter into the abdominal aorta. In this case, AVM occurred 35 years after GTN, which is the longest interval between AVM and GTN diagnoses reported to date.

Case report

A female patient, 59 years old, presented with massive vaginal bleeding without obvious causes two months before admission, followed by intermittent vaginal bleeding in greater amounts than menstrual bleeding. Regardless of hemorrhaging, the patient did not present with abdominal pain or any other complaint. Due to her

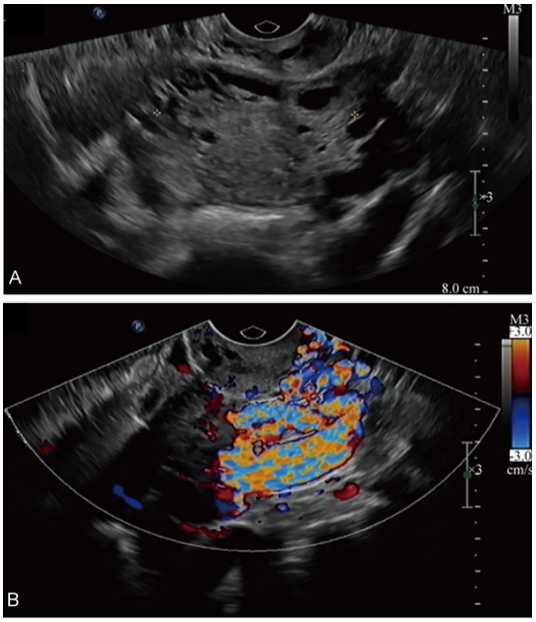

Figure 1. A. Transvaginal US image of the uterus showing multiple cystic structures of several sizes within the uterine myometrium. B. Transvaginal color Doppler sonography demonstrating a tangle of vessels with high-velocity, low resistance turbulent flow producing a color mosaic pattern.

unsuccessful treatment in another hospital, the patient was referred to the Shengjing Hospital, China Medical University. She had undergone uterine aspiration due to "hydatidiform mole" 35 years ago, but the postoperative pathological diagnosis was choriocarcinoma, and she received three courses of intravenous 5-FU chemotherapy (28-30 mg/kg ▪ d for 8-10 days every two weeks). Two years later, the patient had a normal pregnancy and gave a full-term natural birth to a baby. The patient had no additional pregnancies after that and had menopause at the age of 53 years. A gynecological examination revealed visible blood clots in the vagina, bright red blood flowing out from the cervix, a normal-sized cervix, soft and slightly increased uterus, and no abnormality in the bilateral appendage area. This study was conducted in accordance with the declaration of Helsinki. This study was conducted with approval from the Ethics Committee of China Medical University. Written informed consent was obtained from the participant.

Laboratory and imaging findings

The laboratory examination showed a hemoglobin value of 83 g/L, a hematocrit level of 26.6%, a blood human chorionic gonadotrophin (hCG) value of 8.84 mIU/mL, and other serum tumor markers within the normal range. The ultrasound examination revealed the anterior position of the uterus, sized 7×5.7×4.2 cm, and an endometrial thickness of approximately 0.4 cm (**Figure 1**). The left and right ovary sizes were about 2.7×1.7 cm and 2.3×1.3 cm, respectively. Several dilated and circuitous vessels were observed in the bilateral para-uterus and in the intra-myometrium, and the largest diameter was about 1.4 cm (left) and 1.2 cm (right). The blood vessels were filled with multicolored mosaic blood flow signals, showing a turbulent spectrum, which could not be measured. The diagnosis given by ultrasound examination was vasodilatation in the bilateral para-uterus and in the intra-myometrium. Magnetic resonance imaging (MRI) showed disordered, rough, and irregularly distributed blood hypointensity in the uterine myometrium and in the peripheral pelvic plexus on T1 and T2-weighted sequences, especially in the left ovarian plexus. No obvious increase in hypointensity was seen in the myometrium or in the peripheral pelvic cavity. A pelvic digital subtraction angiography showed remarkable thickening and tortuosity of the bilateral uterine arteries, and an extensively deformed vascular mass in the uterine body and on both sides of the uterus, especially on the left side. The blood supply from the bilateral uterine arteries was drained via a large early venous drainage. The draining vein had local aneurysms. The bilateral ovarian arteries were thickened and tortuous, and arteriovenous malformations were observed in the bilateral horns of the uterus (**Figure 2**). These signs were sufficient to make a diagnosis of AVM with hemorrhage.

A single uterine artery embolization was considered to be ineffective for this case, due to the large diameter of the artery and the intense blood flow at the lesion. As the malformation was very extensive, a uterine embolization was not performed. We therefore decided to conduct a total abdominal hysterectomy and a bilateral adnexectomy to remove the uterine AVM. Prior to the surgery, endovascular catheters for balloon occlusion were inserted into the abdominal aorta in an attempt to reduce the risk of intraoperative uncontrolled hemorrhage.

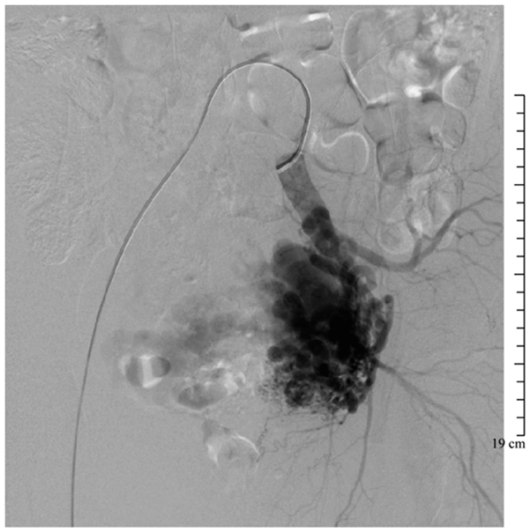

Figure 2. Digital subtraction angiography indicates a uterine vascular abnormality associated with an arteriovenous malformation-like lesion.

Percutaneous abdominal aorta balloon presetting

An 8 F artery sheath (Medtronic Inc., Minneapolis, USA) was inserted into the right femoral artery by a surgeon of the Invasive Technology Department. A 9 mm water film guide wire was introduced into the sheathing canal. With the guidance of the wire, a balloon catheter of 18 mm×40 mm was inserted into the abdominal aorta between the renal artery and the iliac artery, below the renal artery level. After the balloon was inflated to 4 atm, angiography showed no blood flow in the distal end of the abdominal aorta. As blood flow occlusion by the balloon inflation was confirmed, the balloons were deflated in order to avoid the rapid development of collateral circulations.

Intraoperative observation

The whole range of ovarian vessels in the left infundibulopelvic ligament were thickened, extensively dilated, tortuous, and winding into a vascular net. The diameter of the thickened ligament was about 4 cm. The vessels in the left round ligament were also tortuous, thickened, and extensively dilated. The uterine artery, the para-uterine vascular plexus, and the para-cervix vascular plexus in the retroperitoneal space of the left pelvic wall had completely lost their normal forms, and the vessels were tortuous, thickened, extensively dilated, and winding into vascular nets, occupying the left para-uterine and the para-cervix spaces. Similar changes were observed in the vessels of the right infundibulopelvic ligament, the right para-uterus, and the right para-cervix, but the changes on the right side were slighter than those on the left size. Intraoperatively, the dilated vessels in the pelvic cavity were ligated, and the infundibulopelvic ligament, round ligaments in the bilateral pelvic cavity and para-uterine tissues were also dealt with. Following an extensive hysterectomy, the bilateral ureters were isolated, and the uterine vessels, the para-uterine vascular plexus, and the para-cervix vascular plexus of both sides were cut off and ligated. The bilateral main ligaments were cut off and ligated, and the whole uterus and bilateral appendages were removed. During the treatment of the bilateral para-cervix vessels, the abdominal aorta was blocked twice for no more than eight minutes and the interval between the blockages was more than one minute. A total hysterectomy and bilateral adnexectomy with uterine AVM removal were successfully performed in 1 h 10 min, with a blood loss of 20 mL. No blood transfusion was required, and the balloon occlusion catheters were removed after the surgery without any complications.

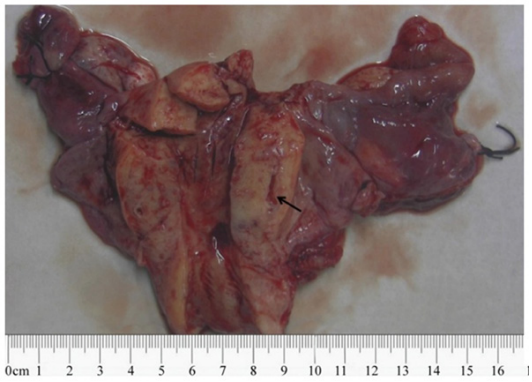

Figure 3. The gross observation of the removed uterus. The arrow shows a cross-section of the obvious thickening intramural blood vessels in the uterus.

Gross observation of the uterus

After the uterine cavity was opened, significantly dilated intramural vessels were visible, and multiple dilated intramural vascular cavities were observed (**Figure 3**). The dilated malformed vessels of different sizes were seen in the bilateral mesosalpinx area and in the para-uterine tissues.

Table 1. Summary of the 2 publications applying preoperative placements of balloon-occlusion catheters to hysterectomy to treat AVM

Author	Age of patient	Etiological factor	Placement location	Method of application
Soeda et al. 2012 [17]	55	Large cervical myoma	Internal iliac artery	Inflate the balloon during the whole hysterectomy.
Yamamoto et al. 2015 [12]	59	Dilatation and curettage	Uterine artery	Inflate the balloon only when needed.

The patient recovered well and was discharged four days after surgery. No recurrence was reported after up to two years from surgery.

Discussion

For treatment of uterine AVM, selective pelvic arteriography and uterine artery embolization are usually the first choices for young women who want to preserve their reproductive function. Conversely, for women who do not require preservation of the reproductive function, in whom uterine artery embolization failed, or whose follow-up conditions were poor, hysterectomy is an effective means to treat heavy bleeding caused by uterine AVM [7, 9]. Uterine artery embolization is not always successful, and embolization failure may lead to repeated bleeding and the need for another embolization or hysterectomy. In the present case, the patient was not interested in preserving her reproductive function. In addition, due to the large diameter and the high intensity of blood flow at the lesion, a single uterine artery embolization was considered to be unsuitable [10]. Thus, a total hysterectomy was performed.

Hysterectomy of the uterine AVM is not an ordinary hysterectomy, but a high-risk operation [11]. Obviously, vascular malformation and pelvic congestion may lead to severe bleeding during surgery. Unlike the simple benign disease, uterine AVM needs to be treated with radical hysterectomy, as the vessels in the para-uterine and para-cervix tissues are deranged and enlarged. Just like the manifestation in the present case, since the relationship between vascular malformation and ureter is disordered, it is necessary to fully isolate the ureters [12, 13]. In this way, preoperative placement of endovascular balloon occlusion catheters is performed to temporarily block the abdominal aorta during operation, in order to control the bleeding.

Preoperative placement of endovascular balloon occlusion catheters for pelvic surgery has been developed with the aim of decreasing intraoperative blood loss, by means of temporary control of the pelvic circulation [14]. In the field of obstetrics and gynecology, this technique is mainly used in high-risk cesarean sections, such as perilous placenta previa and placenta increta [15, 16]. Indeed, many studies have suggested that the preoperative placement of endovascular balloon occlusion catheters can reduce blood loss and blood transfusion during caesarean sections, but this technique remains little used in gynecological surgical procedures. To the best of our knowledge, only two papers have focused on hysterectomy combined with the preoperative placement of endovascular balloon occlusion catheters (**Table 1**). Soeda et al. first used this technique [17], placing the balloon occlusion catheter into the internal iliac artery and inflating the balloon during hysterectomy. In addition, it has been reported that both the preoperative placement of the balloon into the bilateral uterine arteries and the temporarily inflated occlusion balloons during surgery can help the surgeon navigate and identify the inflow arteries within the vascular tangles. After vascular ligation, the balloon is deflated to ascertain whether the bleeding is stopped [12].

In the present case, we placed the endovascular balloon occlusion catheter within the abdominal aorta. Compared to the uterine or internal iliac artery, placing the balloon occlusion catheter into the abdominal aorta has unique advantages. First, super selective catheterization of the uterine or internal iliac artery requires a bilateral operation, which is a time-consuming procedure, whether it is bilaterally performed before the operation or blocked one by one during the operation. Insertion of the balloon occlusion catheter into the abdominal aorta is a relatively simple procedure, requires less X-ray exposure, and saves more rescue time [18]. Secondly, because of the ectopic blood supply from the external iliac and ovary arteries, simply blocking the bilateral internal iliac arteries or the uterine arteries cannot effectively control the uterine bleeding, but the abdominal aorta balloon occlusion can basi-

cally block most of the pelvic blood supply, preventing a better bleeding control than balloon occlusion of other arteries.

The balloon occlusion catheter should be inserted into the abdominal aorta below the renal artery to avoid renal ischemia. In addition, the blocking time should not be too long; otherwise, it will affect the blood supply of the lower extremities and other organs and eventually result in thrombosis and ischemia-reperfusion injury [19]. To date, there is no uniform standard regarding when and how long the balloon must be filled. It is generally advocated that the time of single abdominal aorta occlusion must be less than 10 minutes, with intervals of more than one minute [20]. In our case, the abdominal aorta was blocked twice for no more than eight minutes during treatment of the bilateral para-cervix vessels. The surgical procedure was simple, without active bleeding. For uterine AVM hysterectomy, the preoperative insertion of endovascular balloon occlusion catheters is safe and effective.

The preoperative insertion of endovascular balloon occlusion catheters can make the operating site clean and facilitate the procedure. It is only a disconnection method, not a hemostasis method. Due to low perfusion or blocked perfusion, the bleeding site may not be obvious, and hemostasis may not be sufficient during the operation, which may lead to bleeding after removal of the occlusion balloon catheter. The successful cases reported previously are more successful in “successful disconnection” plus “satisfactory hemostasis”. Thus, intraoperative bleeding should be assessed comprehensively for thorough hemostasis.

To the best of our knowledge, the present study reports the longest interval between the AVM and GTN diagnoses. Previous reports showed that the longest interval between uterine AVM and last pregnancy was 27 years [12, 21]. A 59-year-old woman had a secondary uterine AVM after normal delivery. The intervals between the GTN and uterine AVM diagnoses are varied, with the longest period of 156 months [4]. In our case, the AVM diagnosis was made 35 years after the choriocarcinoma diagnosis and 33 years after the last normal pregnancy. Therefore, uterine AVM should be considered for older or menopausal patients with a history of unexplained abnormal vaginal bleeding after gestational trophoblastic disease.

Conclusion

In summary, we present a histologically confirmed uterine AVM after GTN in a postmenopausal woman. Due to the risk of life-threatening heavy bleeding, the uterine AVM diagnosis should be considered in patients with history of recurrent unexplained abnormal vaginal bleeding after GTN, even if the patient has been in menopause for many years or is older than 60 years. Meanwhile, we have proposed that the preoperative placement of a temporary balloon-occlusion catheter into the abdominal aorta should be considered when hysterectomy is planned for cases with uterine AVM.

Disclosure of conflict of interest

None.

Address correspondence to: Ling Ouyang, Department of Obstetrics and Gynecology, Shengjing Hospital of China Medical University, Shenyang 110004, China. Tel: 86 24 96615 851964; Fax: 86 24 96615 43813; E-mail: cnlingouyang@126.com

References

[1] Das CJ, Rathinam D, Manchanda S and Srivastava DN. Endovascular uterine artery interventions. Indian J Radiol Imaging 2017; 27: 488-495.

[2] Sexton JA and Ricotta JJ. Endovascular approaches to arteriovenous fistula. Adv Surg 2011; 45: 83-100.

[3] Szpera-Goździewicz A, Gruca-Stryjak K, Bręborowicz GH and Ropacka-Lesiak M. Uterine arteriovenous malformation-diagnosis and management. Ginekol Pol 2018; 89: 276-279.

[4] Touhami O, Gregoire J, Noel P, Trinh XB and Plante M. Uterine arteriovenous malformations following gestational trophoblastic neoplasia: a systematic review. Eur J Obstet Gynecol Reprod Biol 2014; 181: 54-59.

[5] Bucha A, Chawla SK and Sethi N. Uterine arteriovenous malformation: a rare cause of abnormal uterine bleeding in a post-menopausal female. Med J Armed Forces India 2016; 72: S210-S212.

[6] Lalitha N, Seetha P, Shanmugasundaram R and Rajendiran G. Uterine arteriovenous malformation: case series and literature review. J Obstet Gynaecol India 2016; 66: 282-286.

[7] Yoon DJ, Jones M, Taani JA, Buhimschi C and Dowell JD. A systematic review of acquired uterine arteriovenous malformations: pathophysiology, diagnosis, and transcatheter treatment. AJP Rep 2016; 6: e6-e14.

[8] Barral PA, Saeed-Kilani M, Tradi F, Dabadie A, Izaaryene J, Soussan J, Bartoli JM and Vidal V. Transcatheter arterial embolization with ethylene vinyl alcohol copolymer (Onyx) for the treatment of hemorrhage due to uterine arteriovenous malformations. Diagn Interv Imaging 2017; 98: 415-421.
[9] Yang JJ, Xiang Y, Wan XR and Yang XY. Diagnosis and management of uterine arteriovenous fistulas with massive vaginal bleeding. Int J Gynaecol Obstet 2005; 89: 114-119.
[10] Mekaru K, Oishi S, Akamine K, Heshiki C and Aoki Y. Spontaneous regression of uterine arteriovenous malformations with conservative management. Case Rep Obstet Gynecol 2017; 2017: 6437670.
[11] Qiu Z, Hu J, Wu J and Chen L. Prophylactic temporary abdominal aorta balloon occlusion in women with placenta previa accretism during late gestation. Medicine (Baltimore) 2017; 96: e8681.
[12] Yamamoto N, Koga K, Akahane M, Wada-Hiraike O, Fujii T and Osuga Y. Temporary balloon occlusion of the uterine arteries to control hemorrhage during hysterectomy in a case of uterine arteriovenous fistula. J Obstet Gynaecol Res 2015; 41: 314-318.
[13] Saeed Kilani M, Lepennec V, Petit P, Magalon G, Casanova D, Bartoli JM and Vidal V. Embolization of peripheral high-flow arteriovenous malformations with Onyx. Diagn Interv Imaging 2017; 98: 217-226.
[14] Wei X, Zhang J, Chu Q, Du Y, Xing N, Xu X, Zhou Y and Zhang W. Prophylactic abdominal aorta balloon occlusion during caesarean section: a retrospective case series. Int J Obstet Anesth 2016; 27: 3-8.
[15] Duan XH, Wang YL, Han XW, Chen ZM, Chu QJ, Wang L and Hai DD. Caesarean section combined with temporary aortic balloon occlusion followed by uterine artery embolisation for the management of placenta accreta. Clin Radiol 2015; 70: 932-937.
[16] Sun W, Duan S, Xin G, Xiao J, Hong F, Hong H, Wu Y and Xu Y. Safety and efficacy of preoperative abdominal Aortic balloon occlusion in placenta increta and/or percreta. J Surg Res 2018; 222: 75-84.
[17] Soeda S, Ushijima J, Furukawa S, Miyajima M, Sakuma K, Watanabe T, Miyazaki M, Hashimoto Y, Nishiyama H and Fujimori K. Uterine arteriovenous malformation formed in a large uterine cervical myoma. Tohoku J Exp Med 2012; 228: 181-187.
[18] Søvik E, Stokkeland P, Storm BS, Asheim P and Bolås O. The use of aortic occlusion balloon catheter without fluoroscopy for life-threatening post-partum haemorrhage. Acta Anaesthesiol Scand 2012; 56: 388-393.
[19] Masamoto H, Uehara H, Gibo M, Okubo E, Sakumoto K and Aoki Y. Elective use of aortic balloon occlusion in cesarean hysterectomy for placenta previa percreta. Gynecol Obstet Invest 2009; 67: 92-95.
[20] Xie L, Wang Y, Luo FY, Man YC and Zhao XL. Prophylactic use of an infrarenal abdominal aorta balloon catheter in pregnancies complicated by placenta accreta. J Obstet Gynaecol 2017; 37: 557-561.
[21] Sato E, Nakayama K, Nakamura K, Ishikawa M, Katagiri H and Kyo S. A case with life-threatening uterine bleeding due to postmenopausal uterine arteriovenous malformation. BMC Womens Health 2015; 15: 10.