Case Report

Transcatheter Embolization of Uterine Arteriovenous Malformation: Report of 2 Cases and Review of Literature

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ABSTRACT  Arteriovenous malformation (AVM) of the uterus is a rare cause of menorrhagia and may at times lead to life-threatening hemorrhage. The clinical findings may not always be reliable in the diagnosis of uterine AVM, and a high index of suspicion is important because, unlike many other causes of menorrhagia, curettage may paradoxically aggravate the bleeding. Herein are described the cases of 2 patients with uterine AVM with abnormal vaginal bleeding. Both had a history of abortion followed by dilation and curettage. In both patients, the diagnosis of uterine AVM was established at Doppler flow ultrasonography. Treatment using transcatheter embolization was successful, and both patients had normal menstrual cycles at follow-up. One patient delivered a healthy baby 2½ years after transcatheter embolization. Journal of Minimally Invasive Gynecology (2011) 18, 812–819 © 2011 AAGL. All rights reserved.

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Uterine arteriovenous malformation (AVM) is a rare cause of menorrhagia that may manifest with life-threatening uterine hemorrhage [1,2]. Unlike other conditions, curettage is not therapeutic and may aggravate the bleeding [3,4]. The clinical diagnosis of uterine AVM is often unreliable. However, correct diagnosis can be reliably made using Doppler flow ultrasonography [5]. Other imaging methods including computed tomography and magnetic resonance imaging (MRI) may be used to complement the diagnosis in difficult cases and to characterize the architectural details of the AVM [6–8]. Herein are presented the cases of 2 patients with uterine AVM successfully treated using transcatheter embolization (TCE), a newer treatment method that has been used in several reported cases [3,9–11]. The literature about uterine AVM is also briefly reviewed.

Case Reports

Case 1

A 30-year-old woman, para 1 with 2 missed abortions, came to our hospital with a history of continuous vaginal bleeding for 3 months that did not respond to hormone therapy. Menarche occurred at age 13 years, and cycles were every 30 days and lasted for 4 days. The patient had 1 living child, 2½ years old, who had been delivered via lower segment cesarean section. In addition, she had 2 missed abortions, at 4 years and 1½ years previously; dilation and evacuation (D&E) was performed both times. Because of continuous bleeding via the vagina, a diagnostic D&E had been performed 1 month previously, and the histopathologic report stated proliferative endometrium. Bleeding had worsened after D&E. Subsequent ultrasonography of the pelvis revealed a hypoechoic area in the posterior myometrium, with internal vascularity. Color flow imaging demonstrated...
a florid color pattern with mosaic aliasing continuing into the right adnexal vessels and right uterine artery.

At examination, the patient was severely anemic, with normal general and systemic findings. The abdomen was soft, with no organomegaly. Vaginal examination demonstrated active bleeding from a healthy cervix. Uterus was of normal size, anteverted, and freely mobile, and the fornices were clear. Hemoglobin concentration was 7g/dL, and platelet count and coagulation parameters were normal. The provisional diagnosis of a large uterine AVM was made, which was confirmed at pelvic angiography performed before TCE of the AVM. The pelvic angiogram showed a large AVM in the right posterior part of the body and fundus fed by both uterine arteries and the right ovarian artery. The right uterine artery was embolized with glue (n-butyl cyanoacrylate, Samarth Life Sciences Pvt. Ltd., Mumbai, India; and iodized oil [Lipiodol], Guerbet Laboratories, Aulnay-sous-Bois, France). The small residual shunt was treated via embolization of the right ovarian and left uterine arteries using an absorbable gelatin sponge (AbGel; Sri Gopal Krishna Labs Pvt. Ltd., Mumbai, India) and transfusion of packed cells. After the procedure, bleeding via the vagina did not stop immediately; therefore, the patient was given progesterone (Syserone 5 mg once daily) for 21 days. After this cycle, her menstrual periods were normal for 2½ years, when she conceived again. She delivered normally a healthy baby a week after she returned to her homeland.
Case 2

A 45-year-old woman, gravida 4, para 0, had a history of irregular periods with heavy bleeding for 5 to 6 years. She had 4 spontaneous early pregnancy losses, followed by D&E each time; the last curettage was performed 6 years previously. Her cycles were every 30 to 60 days, for 4 to 5 days; the last menstrual period was 2 months previously. The patient also had a history of hypertension and hypothyroidism. General and systemic examinations yielded unremarkable findings. Vaginal examination revealed that the cervix was hypertrophied and congested; the uterus was irregular, anteverted, and 14 to 16 weeks of gestation in size; and pulsations were felt in both fornices. The hemoglobin concentration was 9.2 g/dL with normal platelet count and coagulation parameters. Pelvic ultrasound demonstrated a bulky uterus and markedly tortuous and dilated vessels in the myometrium and bilateral adnexae. At pelvic MRI, multiple tortuous flow voids were observed involving the uterus, bilateral adnexae, perivesical region, and pelvic cavity, along with a large venous pouch in the left adnexal region, and dilated and tortuous left ovarian and bilateral uterine arteries (Fig. 2). A pelvic angiogram showed fistulous communication on the left fed by branches of the left ovarian artery and draining into the pelvic veins, with fistulous channels observed near the inferior aspect of the left sacroiliac joint, from both uterine arteries to the pelvic veins (Fig. 3). Uterine artery embolization was performed in 2 stages using bilateral femoral arterial access. The left ovarian arterial feeders were embolized using coils (Cook, Inc., Bloomington, IN), and the left and right uterine arterial feeders were embolized using glue; the large venous pouch on the left side of uterus was embolized using glue after direct puncture under fluoroscopic guidance (Fig. 4). After the procedure, the patient experienced moderately severe pelvic pain (pain score of 5 on a scale of 0 to 10), for which acetaminophen 325 mg with tramadol hydrochloride 37.5 mg (Ultracet) was prescribed twice daily for 5 days, and half that dosage for another 5 days. The pain subsided completely within 10 days. The patient received low-molecular-weight heparin for 3 weeks to minimize the risk of extending thrombosis in the deep pelvic veins. She was well at 1-year follow-up, with regular menstrual periods.

Discussion

Uterine AVM is a rare lesion with considerable risk potential. There are no published reports of the incidence of uterine AVM in the literature. In the series by O’Brien et al [12], uterine AVM was observed in 4.5% of 464 women aged 18 to 41 years in whom a pelvic sonographic examination was performed because of pelvic bleeding. The lesion can be congenital or acquired. Congenital uterine AVMs have multiple feeding arteries, a central tangle of vessels, and numerous large draining veins; these result from abnormal embryologic development of primitive vascular structures and tend to invade the surrounding structures [11,13–15]. Most congenital uterine AVMs are isolated anomalies, but can occur in association with AVMs at other sites [16,17]. Acquired uterine AVM is the predominant type of uterine AVM. It consists of multiple small arteriovenous fistulas between intramural arterial branches and the myometrial venous plexus, tends to have single or bilateral uterine artery feeders without an extraterine arterial supply, and does not have a characteristic nidus [1,2,18,19]. Uterine AVM has been reported after D&C [20], therapeutic abortion [21], uterine surgery [22,23], cesarean section [24], direct uterine trauma [18], gestational trophoblastic disease [25], and exposure to diethylstilbestrol [26]; uterine AVM has also been reported after procedures such as sterilization [27] and hysterectomy [28]. Both of our patients had a history of D&EE.

Patients with uterine AVM may be completely asymptomatic but typically present with intermittent, torrential vaginal bleeding, often in the setting of recurrent spontaneous abortion with normal β-human chorionic gonadotropin (β-hCG) concentration. Menorrhagia can at times be massive and life-threatening, and is thought to occur when the abnormal vessels are exposed from sloughing of the endometrium during menstruation or D&E [20,29]. Other symptoms may include throbbing discomfort in the
lower abdomen, urinary frequency or incontinence, and dyspareunia [1,30]. Some patients may have strong pelvic pulsations after exercise [5]. Systemic hypotension caused by blood pooling within the AVM and even cardiac failure have been described [11].

Clinical diagnosis of uterine AVM is often difficult, and requires a high index of suspicion. Previously, diagnosis of AVM was usually made at angiography, laparotomy, or pathologic analysis for investigation of abnormal vaginal bleeding. Currently, transvaginal ultrasonography is the imaging method of choice [31]. Typical features at gray-scale ultrasound include tortuous anechoic spaces in the myometrium without mass effect. Other features may include myometrial inhomogeneity, an intramural mass mimicking a myoma, and a large bulky cervix that mimics a cervical myoma or carcinoma [13,32]. However, such findings can also be observed in patients with positive β-hCG including those with retained products of conception, hydatidiform mole, or even intrauterine or ectopic pregnancy, with or without concomitant AVM [6,13,31–34]. Thus, the diagnosis of uterine AVM cannot be made on the basis of gray-scale findings alone, and color and spectral Doppler ultrasound should always be performed. The latter help in differentiating AVM by showing high-flow velocity (typically 4- to 6-fold higher than observed in normal myometrial vessels) with low resistance and mixing of arterial and venous waveforms [6,12,20,21]. The arteriovenous shunting in an AVM must be differentiated from malignant arteriovenous shunting

![Case 2. Pelvic angiographic findings. (A) Digital subtraction angiogram in anteroposterior view demonstrating enlarged right uterine artery feeding a large uterine arteriovenous malformation (AVM; arrow). Large fistulous communications are observed draining into the pelvic veins. (B) Digital subtraction angiogram in anteroposterior view showing enlarged left uterine artery feeding the uterine AVM (white arrow). A large venous pouch (black arrow) on the left side is also observed. (C) Conventional angiogram in anteroposterior projection showing an enlarged left ovarian artery feeding the uterine AVM. (D) Magnetic resonance image of the pelvis showing multiple tortuous flow voids involving the uterus, both adnexae, the perivesical region, and the pelvic cavity. A large venous pouch in the adnexal region (arrow) that was observed on the angiogram of the left uterine artery is also clearly visible in this magnetic resonance image. OA = ovarian artery; UA = uterine artery.](image-url)
observed in neoplasia; neoplasms characteristically have low-volume high-velocity flow with drainage into a confined venous space, whereas AVM drains into a large low-pressure venous pool [35].

While rapid recognition of uterine AVM as the cause of bleeding is critical because uterine instrumentation may aggravate the condition [23], it is important not to overdiagnose the entity in the postpartum and post-abortion periods because many so-called uterine AVMs have spontaneously resolved at follow-up imaging [36]; further, withholding curettage for fear of heavy bleeding related to a possible AVM may result in unnecessary blood loss due to the presence of retained products of conception. In selected cases (as in our second patient), other imaging methods such as computed tomography and MRI may be used to establish the diagnosis. These imaging methods will also help to determine the size, extent, and vascularity of the lesion as well as involvement of adjacent organs [6–8]. Spin-echo MRI sequences typically demonstrate multiple flow-related signal voids within the lesion, corresponding to the tangle of vessels seen at Doppler interrogation [7,37]. Although contrast medium–enhanced angiography has been the conventional criterion standard diagnostic test, it is rarely used at the present time except when embolization therapy is being performed.

Contemporary management of uterine AVMs is dictated by the clinical status of the patient, age of the patient, site and size of the lesion, and future pregnancy aspirations. Isolated episodes of bleeding in a hemodynamically stable patient may be safely treated conservatively because many of these remain asymptomatic and may regress spontaneously [38]. In the series reported by Timmerman et al [37],

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**Fig. 4**

(A and B) Post-embolization digital subtraction angiograms in anteroposterior view showing obliteration of the arteriovenous malformation (AVM) after selective embolization of the right and left uterine arteries using glue. (C) Post-embolization radiograph of the pelvis in anteroposterior view showing the final glue cast of the AVM after successful embolotherapy. See text for details. UA = uterine artery.
of 9 patients with uterine AVMs diagnosed at ultrasonogra-
phy, 6 experienced spontaneous resolution, 2 patients with hydatidiform mole required chemotherapy, after which the AVMs resolved, and only 1 required embolization. Congenital AVMs do not regress spontaneously [16]. The medical management of AVMs may include therapy using estrogens, methylerygonovine, danazol, and progastrolidins [1,29,39,40]. The potential mechanisms by which pharmacotherapy may be helpful have not been elucidated, and have been postulated to involve covering the bleeding vessels with proliferative endometrium facilitated by estrogen therapy and reduction in the blood flow to and collapse of the AVM using methylerygonovine maleate [41]. Pregnancy after conservative medical management of AVM has been reported [38,41].

Large AVMs, especially those involving the subendome-
trial tissue, usually require surgical intervention. The com-
monly used surgical treatments include ligation of feeding
vessels, resection of the uterine lesion, and oversewing of
the lesion at laparotomy. Coagulation of the AVM under hys-
teroscopic guidance or of uterine vessels during laparoscopy
has also been used to treat uterine AVMs [42,43]. Currently,
hysterectomy to treat AVM is performed only in women who
do not need or wish fertility preservation. Transcatheter
arterial embolization is a minimally invasive treatment
option with potential to preserve fertility because it does
not seem to interfere with the menstrual cycle or pregnancy [3,9,10]. The need for angiography and TCE is
not accurately predicted by the size of the AVM at imaging, and are generally performed if the patient has
anaemia, is hemodynamically unstable, or has recurrent
bleeding. The procedure involves selective cannulation of
the uterine arteries, usually from femoral arterial access at
the groin with the patient under local anesthesia. The
standard agents used for percutaneous treatment of AVMs
is n-butyl cyanoacrylate, an agent that enables controlled
and permanent obliteration of the AVM. We used glue to
embolize the right uterine artery in our first patient, and the
bilateral uterine arteries in our second patient because
these were the main arterial feeders of the AVMs.

Likewise, the venous pouch on the left side in our second
patient was also embozled using glue because of its fairly
large size. Other agents such as absorbable gelatin
sponges, microfibrillar collagen, and polyvinyl alcohol
have also been successfully used for embolization,
especially when the target of embolization is more distal
and further catheter advancement is difficult or impossible.
Stainless steel coils are also commonly used for arterial
embolization, but have the disadvantage that often several
such coils are needed for obliteration of the AVM, and the
necessity for repeated introduction of coils increases the
procedure time and the risk of air introduction.

The technical success rate for TCE has been reported to
be 79% to greater than 90% [1,10,44]. In some patients,
multiple sessions of embolization may be needed (as in
our second patient). Embolization failure may be
attributed in part to the presence of adenomyosis. but is
more often due to development of collateral vessels. Failure of embolization therapy can be managed with
hysterectomy [18,42] or with uterine artery and ovarian
ligament ligation when uterine preservation is desired [43].
Przybojewski and Sadler [45] reported a novel image-
guided management of a uterine AVM after failed TCE in
which they directly injected embolization material into the
nidus of the AVM under ultrasound guidance and fluoro-
scopy after exposing the uterus surgically; the patient had
a successful term pregnancy afterward. When embolization
must be postponed, therapy with gonadotropin-releasing
hormone agonists may be used to reduce the size of the
AVM; such adjunctive therapy may increase the chances
of successful TCE [46].

After successful embolization of a uterine AVM, hypo-
vascularity of involved areas could, in theory, affect placen-
tation and fetal growth; yet, several successful intrauterine
pregnancies after TCE of uterine AVMs have been reported
[19,47,48] including a successful twin pregnancy [49],
which suggests that adequate collateral blood supply can
develop to support a full-term pregnancy. Normal placental
blood flow has been documented after previous TCE to treat
uterine AVM [19]. In the study by O’Brien et al [12], normal
menstrual cycles returned 2 months after TCE, and 5 patients
became pregnant. Both of our patients had menses within 2
months of TCE therapy, and the first patient also delivered
a healthy baby.

With experienced operators, TCE is generally safe. Minor
complications including hematomata, urinary tract infection,
retention of urine, and vessel or nerve injury at the vascular
puncture site are common and require only mild supportive
care or careful observation [50]. Varying degrees of pelvic
pain are also common in the immediate postembolization
period. Pain after TCE is probably due to ischemia produced
by the embolization procedure, usually peaks on the first day
after the procedure, responds well to analgesic therapy, and
resolves in about a week [51]. Severe persistent pain neces-
sitating administration of narcotic therapy may be due to is-
chemia or infarction of viable uterine tissue [52,53]. The
moderate pain after TCE of the uterine arteries in our
second patient could have been due to the large area of
ischemia produced in a 16-week gestation uterus size. As
in other arterial embolization cases, a “postembolization
syndrome” consisting of pelvic pain, fever, leukocytosis
(without left shift), nausea, vomiting, and malaise may
sometimes develop after uterine artery embolization, and of-
ten requires hospitalization. Patients who develop or con-
tinue to have symptoms consistent with postembolization
syndrome with foul-smelling vaginal discharge beyond
1 week after TCE should be evaluated for intrauterine infec-
tion. More serious complications after TCE of the uterine
arteries are rare. Pelvic infection occurs sporadically after
TCE, has been more often reported in cases of TCE
performed to treat uterine myomas, is associated with signif-
ificant morbidity, and may require emergency hysterectomy
fatal sepsis after TCE has also been reported. Urinary necrosis is another rare life-threatening complication that mandates prompt treatment with antibiotics and hysterectomy. Loss of ovarian function can rarely develop after uterine artery embolization, in particular in women older than 45 years because of their more abundant uterine-ovarian arterial anastomoses compared with younger women. Other serious potential complications of TCE may include perianal skin sloughing, uterovaginal and rectovaginal fistulas, neurologic deficits in the lower extremities, deep venous thrombosis, and pulmonary embolism. There were no serious complications in our patients.

## Conclusion

Uterine AVM is a rare but potentially serious cause of abnormal vaginal bleeding. Clinical findings in such cases are unreliable, and uterine curettage is not diagnostic and may even aggravate it at times; therefore, a high index of clinical suspicion is required. Diagnosis should be considered in all patients of reproductive age who have abnormal vaginal bleeding and negative h-hCG test results. Endovaginal ultrasoundography is the imaging method of choice, and color and spectral Doppler imaging should be used routinely to enable the correct diagnosis. Transcatheter embolization is an excellent treatment option with a high success rate, low complication rate, and ability to preserve fertility.

## References
