Acquired Uterine Vascular Malformation: Clinical Outcome of Transarterial Embolization
후천성 자궁 혈관 기형: 경피적 경동맥 색전술의 임상적 결과

Chae Hoon Kang, MD1, Seung Boo Yang, MD2*, Dong Erk Goo, MD2, Yong Jae Kim, MD2, Jae Myung Lee, MD2, Woong Hee Lee, MD2
1Department of Radiology, Gangneung Asan Hospital, Gangneung, Korea
2Department of Radiology, Soonchunhyang University Gumi Hospital, Gumi, Korea

Purpose: To evaluate clinical outcomes of transarterial embolization of bilateral uterine arteries (UAE) in patients with acquired uterine vascular malformation (UVM).

Materials and Methods: This retrospective study was performed on the medical records of all 19 patients who underwent transarterial embolization of bilateral UAE for the treatment of symptomatic UVMs from January 2003 to June 2011. Embolization was performed via the unilateral femoral artery approach with a catheter and angiographic techniques. Clinical success was defined as definitive resolution of abnormal vaginal bleeding. Post-procedural complications included all adverse events related to the embolization procedure.

Results: A total of 20 procedures were performed in 19 patients. One patient required repeat embolization because of incomplete embolization related to prominent high flow malformation. Clinically, in all patients, bleeding was controlled immediately after embolization. No complications occurred in all patients during the follow up period. In all patients who underwent successful UAE, menstrual cycles were normally restored within 1–2 months. Normal pregnancy with term delivery was observed in two of the 19 cases.

Conclusion: Transarterial bilateral UAE is a safe and effective treatment in patients with vaginal bleeding caused by acquired UVM, and it allows the possibility of future pregnancy.

Index terms
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*Corresponding author: Seung Boo Yang, MD
Department of Radiology, Soonchunhyang University Gumi Hospital, 179 1gongdan-ro, Gumi 39371, Korea.
Tel. 82-54-468-9391 Fax. 82-54-464-9300
E-mail: ysbysb@sch.ac.kr

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tion or hysterectomy. Currently, arterial embolization is the preferred treatment because it is minimally invasive and has the potential to preserve fertility.

The purpose of our study is to describe our experience in using transarterial bilateral uterine arteries (UAE) in patients with acquired UVM, and to evaluate the long term clinical outcomes.

**MATERIALS AND METHODS**

From January 2003 to June 2011, a total of 19 patients (mean age: 30.8 years, range: 22–44 years) underwent bilateral UAE for the treatment of a symptomatic UVM. The medical records of all 19 patients were retrospectively reviewed. This study received approval from Soonchunhyang University's institutional Review Board.

All but one patient underwent a single embolization session. All 19 patients initially presented with vaginal bleeding. The recent obstetrical history prior to embolization revealed a history of D & C after missed abortion (n = 12), cesarean section (n = 5), normal vaginal delivery (n = 1), and ectopic pregnancy (n = 1).

All patients underwent abdominal and transvaginal ultrasound examinations or CT or MRI before embolization. Embolization was performed using the unilateral femoral artery approach with a catheter and angiographic techniques. After insertion of a 5-French introducer sheath (Terumo, Tokyo, Japan), bilateral UAE were catheterized using a 5-French Cobra catheter (Cook Medical, Bloomington, IN, USA) under local anesthesia. Depending on the vascular anatomic findings during angiography, decisions were made regarding whether or not to use an additional 2.7-French microcatheter (Progreat; Terumo) to catheterize the feeding uterine artery. Bilateral UAE was performed using gelatin sponge particles made from gelatin sponge sheets (Gelfoam; Upjohn, Kalamazoo, MI, USA) and/or additional microcoils (Tornado; Cook Medical).

Angiographic success was defined as successful embolization of the vascular malformation with stasis of blood flow in the main feeding uterine artery and no residual opacification of the UVM. Ultrasonographic success was defined as eradication of the UVM and restoration of normal uterine vasculature. Clinical success was defined as definitive resolution of abnormal vaginal bleeding. Post-procedural complications included all adverse events related to the embolization procedure.

The medical records were reviewed with respect to post-procedural complications, recurrence of vaginal bleeding, menstrual cycle, pregnancy or delivery.

**RESULTS**

Angiographic success was achieved in all 19 patients. One patient required repeat embolization since the UVM was not completely embolized due to prominent high flow in the first session. Therefore, a total of 20 procedures were performed in 19 patients.

Angiography revealed the presence of a pseudoaneurysm during five procedures (25%) (Fig. 1). During the remaining 15 procedures (75%), hypertrophied main UAE with hypervascular mass-like lesions and early draining into a large pelvic vein were opacified. No direct arteriovenous communication was demonstrated (Fig. 2). During all procedures, no contrast extravasation into the uterine cavity was depicted. The embolic agents used in all embolization sessions were gelatin sponge particles, along with additional microcoils during three procedures due to high flow. On completion angiography, no evidence of non-target embolization or vessel wall damage in the uterine or internal iliac artery was noted.

Duplex ultrasound evaluation performed three days after embolization showed ultrasonographic success in all patients.

The mean clinical follow-up period was 20 months, and it ranged from 1 to 42 months. Vaginal bleeding was controlled immediately after embolization in all patients, and none of the patients reported any clinical symptoms associated with UVM during the follow-up period. Minor adverse events consisting of post-embolization pelvic pain were noted in three patients within a week of the procedure. All patients were treated conservatively by intramuscular or intravenous administration of 100 mg tramadol HCL daily (Tradol; Bugwang, Seoul, Korea) for 3–7 days. All patients had complete resolution of their pain within a week.

Recovery of their normal menstrual cycle was seen in all 19 patients. Among these 19 patients, two patients achieved full-term intrauterine pregnancy with uneventful labor. In these cases, the mean time period between embolization and delivery was 20.1 months.
DISCUSSION

Vascular malformation can occur in any part of the body, including the female pelvis. Mulliken and Glowacki (5) proposed the classification of vascular malformation based on their flow characteristics (fast flow or slow flow), and their vascular channel components (capillary, venous, lymphatic, arterial and combined). UVM, a type of fast flow malformation, can affect the pelvic structures such as the pelvic wall, the visceral organs, or both (6, 7). It can form congenitally or can be caused by other secondary causes, and it can be difficult to differentiate between these causes depending on the patient’s history. Congenital UVM arises from abnormal embryologic development of the primitive vascular structure, and it results from multiple abnormal arteriovenous communications. It contains multiple arterial feeders and venous drainage channels, and an intervening nidus (8-11). Acquired UVMs are more common than primary UVMs, and their growth is typically stimulated by trauma such as D & C, or hormonal changes that occur during puberty or pregnancy (12). They can also be associated with malignancies such as endometrial carcinoma or GTD.

Compared with congenital UVMs, acquired UVMs tend to have single or bilateral feeding UAE without an extrauterine artery supply, and do not have a nidus (8). UVMs typically evolve over time, from an asymptomatic lesion to a lesion with active shunting, venous engorgement or hypertension. If they rupture, UVMs can cause life-threatening bleeding; hence, prompt diagnosis and treatment are important.

Currently, the diagnostic modalities available for UVM are ultrasonography (US) and MRI. Color and duplex Doppler US are especially good screening and diagnostic methods (6, 9, 13). The most common Gray-Scale ultrasonographic feature of UVM is the anechoic tubular space in the myometrium without a mass effect, but this feature is nonspecific; hence, the diagnosis of AVM is not based on Gray-Scale ultrasonographic morphology. On color Doppler US, a tangle of tortuous vessels with multidi-

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**Fig. 1.** A 27-year-old woman presented with vaginal bleeding for 2 months after D & E. Gray-Scale ultrasonogram (A) shows a cystic mass in the uterus. Color Doppler ultrasonogram (B) reveals a pseudoaneurysm. Selective left uterine angiogram (C) shows a pseudoaneurysm in the uterus. Completion uterine angiogram (D) after uterine artery embolization with gelatin sponge particles reveals the disappearance of pseudoaneurysm with occlusion of the uterine artery. Color Doppler ultrasonogram (E) obtained two days after embolization reveals complete thrombosis of the pseudoaneurysm.
rectional, high-velocity arterial flow with an asymmetric distribution is observed. Duplex Doppler US examination shows the features of arteriovenous shunting, like fast arterial flow with low resistance (8, 9). MR angiography, performed as a time-resolved contrast-enhanced three-dimensional acquisition, can be useful in evaluating the extent of the lesion and the vascular anatomy around this anomaly. Furthermore, 3D-reconstruction of the vascular structures with elimination of overlapping vessels or other anatomic features allows better depiction of the anatomy of the arterial and venous components (14). However, uterine artery angiography remains the diagnostic tool of choice for UVM, because both diagnosis and immediate embolization can be performed simultaneously by the interventionist (1, 15).

In the past, hysterectomy or surgical ligation of the uterine artery was widely performed for the treatment of UVM (16). However, hysterectomy has a major limitation as it cannot preserve the patient’s fertility after surgery (17-19).

Wu et al. (17) introduced laparoscopic bipolar coagulation of uterine vessels, which can relieve symptoms and has advantages such as shorter surgical times and absence of radiation exposure. However, general anesthesia and laparoscopic surgery are required.

Transarterial embolization of a UVM was first introduced by Forssman et al. (18). The benefits of UAE are that it is minimally invasive, preserves the uterus, requires less hospitalization and results in a shorter recovery period. Currently, hysterectomy is performed to treat uterine AVM only in those women who do not want additional pregnancies or who do not have the capacity to bear children. In addition, many previous studies have revealed that UAE is safer and less invasive than surgical procedures (19-21).

UAE should be performed in bilateral uterine arteries, because the uterine artery has many collateral supply channels and reperfusion from a contralateral uterine artery can occur, which may induce continuous bleeding or rebleeding after embolization (22-24).

Fig. 2. A 30-year-old woman presented with intermittent vaginal bleeding for 1 month after missed abortion. Transvaginal Gray-Scale (A) and color Doppler (B) ultrasonograms show a tangle of tortuous vessels with increased vascularity in the uterine fundus. Selective left uterine angiogram (C, D) demonstrates an hypertrophied uterine artery and a vascular mass lesion with early venous drainage during the arterial phase. Post-embolization pelvic angiogram (E) shows eradication of the vascular mass and complete occlusion of the uterine artery.
Embolization materials widely used in these procedures are gelatin sponge particles, polyvinyl alcohol (PVA), coils, glues, detachable balloons, or a combination thereof (15, 19, 21-23). Gelatin sponge particles are used as a means of causing mechanical obstruction, slowing blood flow and hastening thrombus formation (25). However, embolization using gelatin sponge particles causes temporary vessel occlusion and allows recanalization in a few weeks, leading to a risk of reperfusion or rebleeding (25). PVA results in a permanent embolic effect by adherence to the vessel wall, causing stagnation of flow, and direct occlusion of the smallest vessels into which it can fit. Coils are also permanent embolic agents available in various sizes and shapes. They induce mechanical thrombosis by themselves and chemical thrombogenesis due to their fibers. However, a potential complication of coil embolization is the occlusion of non-targeted vessels due to migration, which can result in cerebral infarction or pulmonary embolism. This can occur during treatment of high-flow conditions such as AVM or arteriovenous fistula. N-butyl-2-cyanoacrylate (NBCA), commonly used in the treatment of cerebral AVM and spinal tumors, is also used to treat uterine AVM. NBCA is a clear and free-flowing monomer. When it is exposed to water or blood, polymerization is induced, and it results in an intravascular cast. McCormick and Kim (26) reported successful embolization of uterine AVMs with NBCA. The disadvantages of using NBCA are the need for a special syringe and materials and the risk of catheter embedding (25). Ethylene vinyl alcohol copolymer (Onyx is the commercial name) is also widely used for treatment of cerebral AVM. Abbayir et al. (27) reported a case of uterine AVM treated with a combination of Onyx and other materials. However, in our study, the embolic agents used in all embolization sessions were gelatin sponge particles. Only during three procedures, additional microcoils were needed due to high flow malformation. Hence, gelatin sponge particles are adequate embolic agents for management of acquired UVMs.

Kim et al. (28) reported successful restoration of the menstrual cycle within two months in all 19 patients who underwent UAE using gelatin sponge particles or PVA. This result of the previous study is compatible with that of our study.

Peitsidis et al. (20) performed a systematic review of case reports of UVM treatment and identified 17 pregnancies (17%) after UAE for treatment of UVM. We observed normal pregnancy and term delivery in two patients (10.5%), who had undergone bilateral UAE. Maleux et al. (4) estimated that the mean time period from UAE to subsequent pregnancy was 15.6 months in their report. The mean time period between embolization and delivery was 20.1 months in our study. Therefore, our study confirms the previously reported result regarding the potential to become pregnant after particulate UAE (3, 29, 30).

The possible complications associated with UAE are groin hematoma, uterine ischemia, pelvic infections such as tubo-ovarian abscesses, endometritis, and transient or permanent amenorrhea (31). In our study, the number of minor adverse events due to the procedure was low. Pelvic pain was observed during the first week in three patients, but the pain was relieved by conservative treatment. None of the patients presented with amenorrhea, as described in 4% of the cases after bilateral UAE for the treatment of uterine myomas. No major adverse events were observed in our study.

Our study has some limitations. The retrospective design and the small number of patients included in the study do not allow extensive statistical analysis.

In conclusion, transarterial bilateral UAE is a safe and effective treatment in patients with vaginal bleeding caused by acquired UVM. This procedure allows preservation of uterine function with the possibility of future pregnancy. Transarterial bilateral UAE should be considered as a primary treatment option in these patients.

REFERENCES


후천성 자궁 혈관 기형: 경피적 경동맥 색전술의 임상적 결과

강채훈¹ · 양승부²* · 구동억² · 김용재² · 이재명² · 이웅희²

목적: 후천성 자궁 혈관 기형 환자에게 있어서 양측 자궁 동맥에 대한 경동맥 색전술의 임상적 결과를 알아보고자 한다.


결과: 총 19명의 환자에서 20번의 시술이 시행되었다. 첫 번째 시술에서 현저하게 빠른 속도의 기형으로 인해 색전되지 않은 한 명의 환자에서 재시술이 요구되었다. 임상적으로 전 환자에게서 색전술 후 출혈이 즉시 조절되었다. 합병증은 추적 관찰 기간 동안 모든 환자에게서 발생하지 않았다. 성공적인 색전술을 시행받은 환자 모두가 1~2개월 이내에 정상적인 월경 주기에 돌아왔다. 19명 중 2명에게서 정상 임신과 만삭 분만이 이루어졌다.

결론: 후천성 자궁 동맥 기형에 의한 혈출류 환자에게 있어서 양측 자궁 동맥에 대한 경동맥 색전술은 안전하고 효과적인 치료법이며, 향후 임신의 가능성을 열어 준다.

¹강릉아산병원 영상의학과, ²순천향대학교 구미병원 영상의학과