

Novel therapeutic strategy for uterine arteriovenous fistulas: case report

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Summary

Background: The major presenting symptom of uterine arteriovenous fistulas is massive, torrential vaginal bleeding, the degree of which often leads to a shock state. **Case:** A 35-year-old woman, gravida 3, para 2 presented with massive vaginal hemorrhage at the first menstruation six months after delivery. Uterine arteriovenous fistulas were diagnosed by color Doppler ultrasonography (US), dynamic computer tomography (CT), and conventional angiography. The patient underwent hysterectomy after bloodstream decrease by bilateral uterine artery embolization. **Conclusion:** The extent of uterine arteriovenous fistulas was diagnosed by color Doppler US, CT, and pelvic angiography, and this precise evaluation led to an adequate therapeutic strategy for uterine arteriovenous fistulas.

Key words: Uterine arteriovenous fistulas; Bilateral uterine artery embolization; Color Doppler ultrasonography; Dynamic computer tomography; Conventional angiography.

Introduction

Uterine arteriovenous fistulas are rare causes of uterine bleeding, but are known as lesions with a risk of unexpected massive vaginal bleeding [1-5]. The degree often leads to a shock state, and there can be the need for blood transfusion [3]. Uterine vascular lesions involve arteriovenous malformations, true aneurysms, pseudoaneurysms, and chorioangioma of the placenta [1]. In a broad sense, arteriovenous malformations have been variably described as cirroid aneurysm, arteriovenous aneurysm, arteriovenous fistula, pulsating angioma and cavernous hemangioma [2, 5]. In a narrow sense, arteriovenous malformations are considered to be mainly congenital. On the other hand, arteriovenous fistulas are considered to usually be acquired [1, 4]. Most reported acquired cases were complicated by a history of cesarean section or uterine endometrial curettage, or the usage of intrauterine contraceptive devices [2-6]. Hormonal dysfunction and an inadequate healing process from previous damage are believed to play some role in the pathogenesis [4]. In addition, uterine arteriovenous fistulas are found in women who have menstruation and can become pregnant [4, 6].

In this case, color Doppler ultrasonography (US), dynamic computer tomography (CT), and conventional angiography were able to define the extent of uterine arteriovenous fistulas, and a novel therapeutic strategy for uterine arteriovenous fistulas was developed: hysterectomy after blood flow decrease by bilateral uterine artery embolization.

Case Report

A 35-year-old woman, gravida 3, para 2 had a history of hydatidiform mole at 22 years old. She presented with massive vaginal hemorrhage at the first menstruation six months after the delivery of her second child. Her blood pressure deteriorated, and the peripheral blood hemoglobin was 4.9 g/dl. Coagulation study results were within the normal range. Color Doppler US showed multiple hypervascular flows in the uterine myometrium (Figure 1). The bleeding was controlled by combination treatment with estrogen and progestin. The peripheral blood hemoglobin recovered up to 9.7 g/dl with oral and parenteral iron without any blood transfusion in about one month. Human chorionic gonadotropin in serum was negative.

To evaluate whether uterine arteriovenous malformations were the cause of the massive uterine bleeding, contrasting CT was taken. The CT revealed a bilateral vascular lump throughout the myometrium to the pelvic and uterine cavities (Figure 2). Most of the vascular lump consisted of expanded veins, but the lump might have involved some arteries according to the early stage of contrasting just after the start of the contrasting



Figure 1. — Multiple anechoic spaces and color signals in a mosaic pattern shown by color Doppler US.

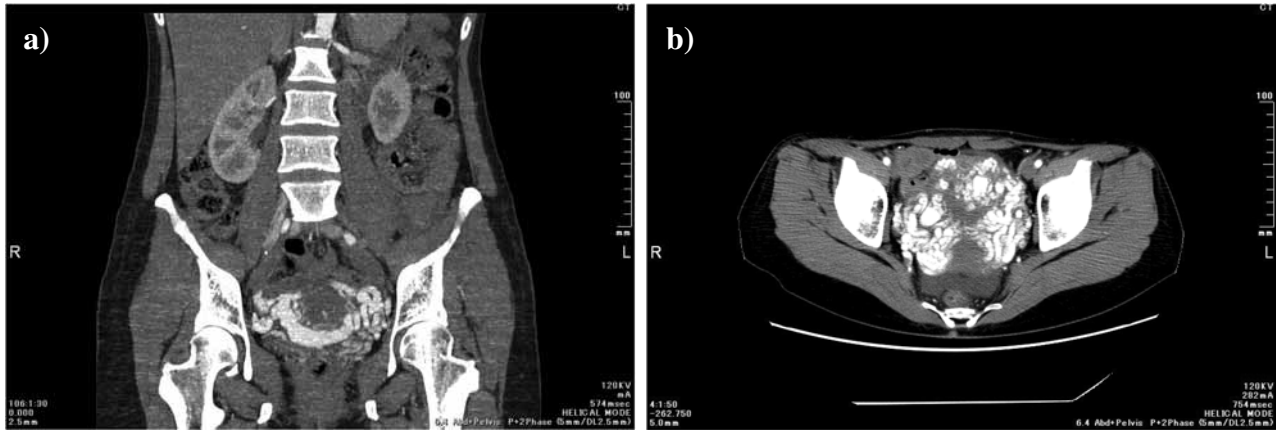


Figure 2. — Bilateral hypervascular mass shown by enhanced CT. a) coronal section; b) horizontal section.

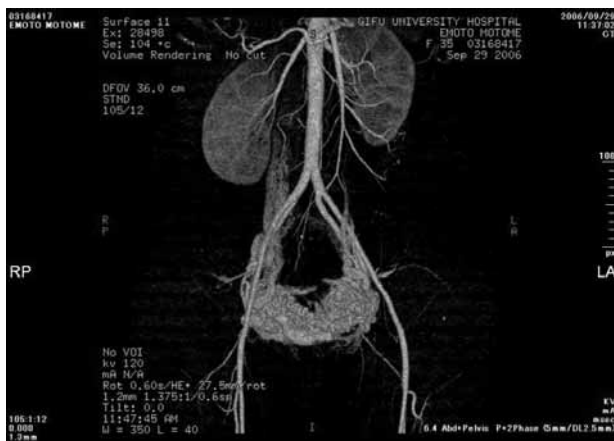


Figure 3. — Vascular lump consisting of expanded veins shown by 3D-CT.

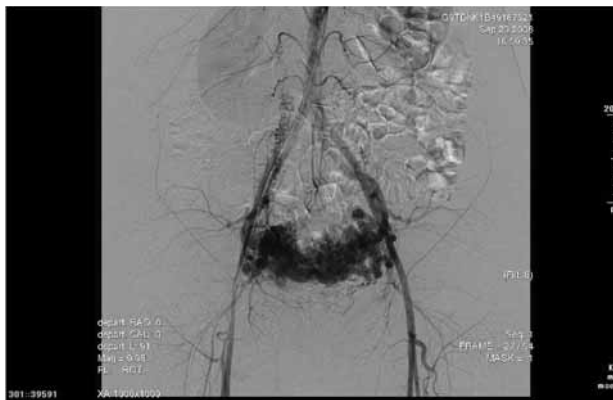


Figure 4. — Uterine arteriovenous fistulas shown by pelvic arteriography.

agent drip infusion (Figure 3). The vascular lump was diagnosed as uterine arteriovenous fistulas. The patient selected hysterectomy because she had already had two children, and wanted to avoid the recurrence of massive uterine bleeding after artery embolization. Pelvic angiography demonstrated hypervascular masses supplied predominantly by bilateral uterine arteries but also by other internal iliac arteries (Figures 4 and 5). To avoid massive bleeding at hysterectomy, preoperative artery embolization was performed for blood flow decrease of

the saturated internal organs detected by contrasting CT (Figure 2).

The post-embolism angiogram showed that the blood flow decreased approximately 90% in the right side and 40% in the left (Figure 6). No immediate complications such as pain and fever occurred after the embolization. The patient underwent hysterectomy with 220 g of bleeding, and afterwards the pelvic vascular lump almost completely disappeared because the uterine arteriovenous fistula, recognized as the nidus, was removed by hysterectomy. Neither symptoms nor abnormal signs, checked by contrasting CT one month after surgery, occurred.

Discussion

The major presenting symptom of uterine arteriovenous fistulas is intermittent and torrential vaginal bleeding, especially torrential bleeding that is believed to occur from the arteries [2, 3]. US is the most common investigation tool, and especially color Doppler US with spectral wave analysis is valuable [1-3, 7]. To evaluate the extent of uterine arteriovenous fistulas, CT is less invasive than pelvic angiography, which can be useful for diagnosis with artery embolization treatment [4]. Reports of good outcomes and successful pregnancies after artery embolization have gradually increased recently, but the long-term success rate of embolization was still reported to be from 79% to 90% [4-6]. Although the degree of uterine bleeding and number of enriched collateral vessels are critical in treating uterine arteriovenous fistulas, the management in patients with symptomatic uterine arteriovenous fistulas depends on the desire for pregnancy [4, 5]. As our patient had no desire to preserve her fertility, but desired a radical cure, she underwent hysterectomy after artery embolization to avoid massive bleeding at hysterectomy.

Conclusion

The extent of the uterine arteriovenous fistulas was diagnosed by color Doppler US, CT, and pelvic angiography, and this precise evaluation led to an adequate therapeutic strategy.

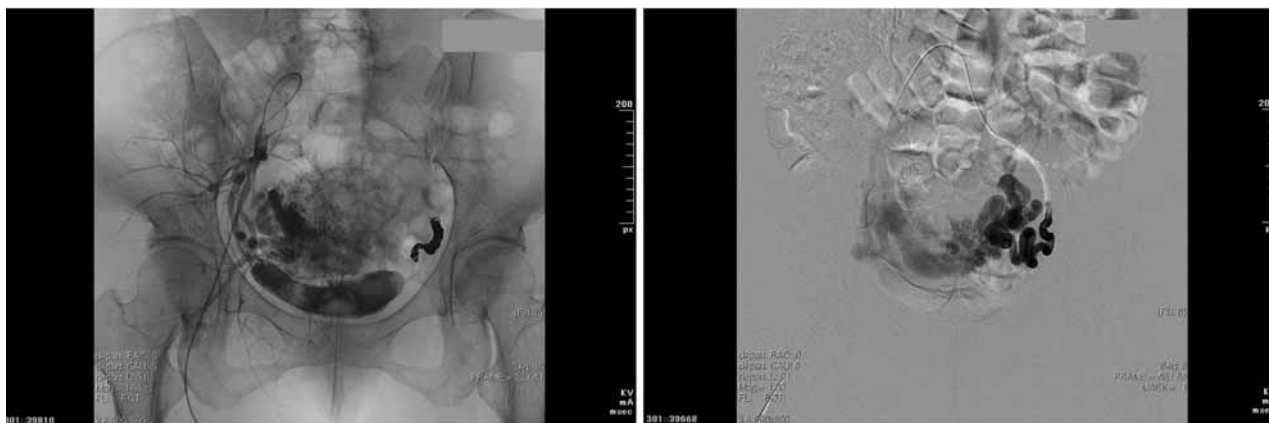


Figure 5. — Uterine arteriovenous fistulas before embolization shown by arteriography.

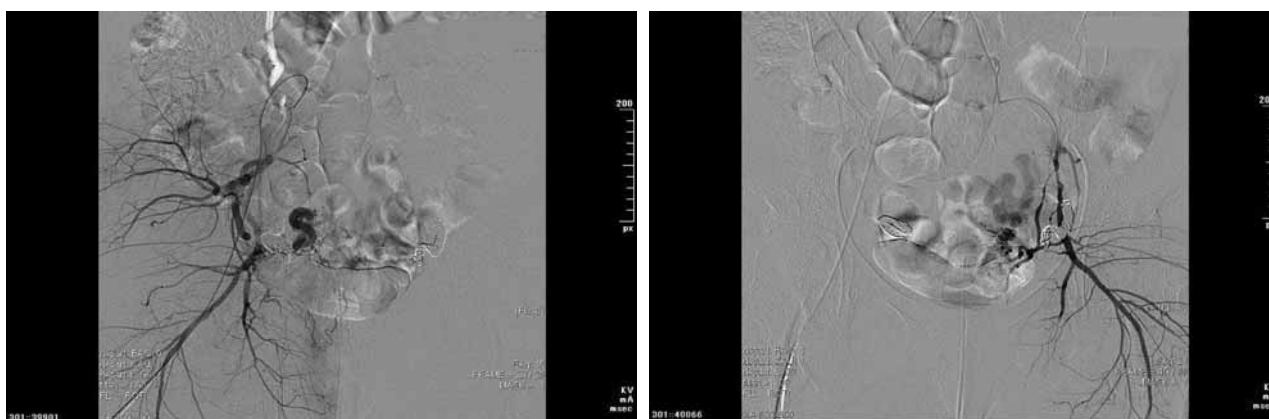


Figure 6. — Remaining collateral arteries after embolization shown by arteriography.

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