Uterine Arteriovenous Malformation Formed in a Large Uterine Cervical Myoma

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Arteriovenous malformation (AVM) can arise in various organs, particularly the brain, but it is rare in the uterus. Uterine AVM is potentially lethal and is generally associated with uterine trauma, such as dilatation and curettage, therapeutic abortion or uterine surgery. On the other hand, uterine myoma is the most common benign gynecological tumor, but uterine cervical myoma is rare and grows in the extraperitoneal space, with development of complex capillary networks within the tumor. Cervical myoma surgery is therefore a difficult operation with a risk of massive bleeding. We report herein a patient with uterine AVM formed within a large cervical myoma in a postmenopausal woman. The patient was a 55-year-old Japanese woman who complained of lower abdominal distension. Ultrasonography, computed tomography and magnetic resonance imaging showed an 18 × 20-cm uterine cervical tumor with dilatation of numerous vessels. Pelvic angiography was scheduled to provide accurate diagnosis and to minimize intraoperative blood loss. In fact, preoperative pelvic angiography allowed us to identify the true feeding artery and drainage veins. Occlusion of the feeding artery with a balloon device is effective in decreasing intraoperative bleeding. Abdominal total hysterectomy was performed as the surgical management of this uterine AVM. Prophylactic endovascular balloon occlusion of the ipsilateral internal iliac artery reduced the amount of hemorrhage during surgery, although blood transfusion was needed in our patient. In conclusion, preoperative embolosclerotherapy should be considered as a treatment option in patients with AVM present in a large uterine cervical myoma.

Keywords: balloon occlusion; cervical myoma; hysterectomy; pelvic angiography; uterine arteriovenous malformation


Arteriovenous malformation (AVM) can arise in various organs, particularly the brain, but less often in organs, such as the pancreas and liver (Jaeger and Forbes 1946; Talbot and Silverman 1952; Stone et al. 1965; Grannis et al. 1973). Uterine AVM is rare and potentially lethal, and is associated with damage to the uterine tissue, cesarean section, curettage procedures, infection, retained products of conception, trophoblastic disease, choriocarcinoma and other gynecological malignancies, such as cervical carcinoma and endometrial carcinoma (Delaloye et al. 1998). Only one previous report has described AVM arising from a benign uterine tumor, and the patient in that case underwent uterine artery embolization to the symptomatic uterine body myoma, but suffered non-target embolization from an intrafibroid AVM (Anonymous 2009). Uterine myoma is the most common benign gynecological tumor, occurring in about 20% of women of reproductive age, whereas uterine cervical myoma is rare, accounting for about 5% of all myomas (Matsuoka et al. 2010). Uterine cervical myoma displaces the position and course of the bladder, ureter, rectum and uterine vessels, and grows within the retroperitoneal space, with complex capillary networks develop behind the myoma. Removal of cervical myoma is more difficult than removal of fibroids located in the uterine body. AVM arising in a uterine myoma is extremely rare, and no case arising from cervical myoma has been reported. Uterine cervical myoma can lead to highly expanded veins, meaning that surgical procedures run a risk of causing significant bleeding.

We describe herein surgical management of the uterus
AVM formed within a large cervical myoma in a postmenopausal woman using abdominal total hysterectomy combined with preoperative pelvic angiography and prophylactic endovascular balloon occlusion of the ipsilateral internal iliac artery. Preoperative pelvic angiography revealed the true vascular formation of the uterine AVM, facilitating surgical management. AVM in a large uterine cervical leiomyoma is challenging lesions; thus, careful management plan is essential to determine the optimal method of treatment of this rare lesion.

**Clinical Report**

A 55-year-old Japanese woman (gravida 2, para 2) initially presented to another hospital with distension of the lower abdomen. Ovarian tumor was suspected and she was referred to the Department of Obstetrics and Gynecology at our hospital. Past medical history was unremarkable, with no history of abdominal or pelvic surgery or episodes of genital bleeding. Uterine myoma had been diagnosed at 45 years old, but no treatment was performed, as she remained asymptomatic. She had entered menopause at 50 years old. Internal examination showed an unmovable uterine tumor and marked deviation of the uterine cervix. Transvaginal ultrasonography showed an 18 × 20-cm uterine tumor (Fig. 1A). Color Doppler analysis demonstrated high flow at low velocity in dilated vessels within the uterine tumor. Transabdominal ultrasonography using a 3.5-MHz vector transducer demonstrated a low-echoic lesion and color Doppler analysis demonstrated dilatation of numerous vessels within the tumor (Fig. 1B). Pulsed Doppler evaluation of the identified area revealed a resistance index of 0.52, with a peak systolic velocity of 13.5 cm/s and turbulent flow. Computed tomography (CT) showed multiple expanding vessels within the tumor and the dilation of the right internal iliac arteries and right ovarian vessels (Fig. 2A). Magnetic resonance imaging (MRI) revealed a diffuse uterine cervical tumor with focal hemorrhage (Fig. 2B), while magnetic resonance angiography (MRA) showed dilatation of numerous vessels in the pelvis (Fig. 2C). However, details of communications between vessels were unclear. Laboratory investigations revealed a decreased hemoglobin level of 9.8 g/dl, while fibrin degradation prod...
uct and D-dimer levels were markedly elevated, at 274.0 μg/ml and 95.7 μg/dl, respectively. Lactate dehydrogenase levels were also elevated, at 671 units/ml. The cervical tumor was suspected to contain an AVM or aneurysm and surgical management was considered necessary to clarify the tumor histology. Before hysterectomy, pelvic angiography was scheduled to provide accurate diagnosis and to allow minimization of intraoperative blood loss using balloon occlusion of the internal iliac artery. This was performed using a left common femoral approach. By applying a standard Seldinger technique, the left femoral artery was punctured under local anesthesia, and the right iliac arteries were selected. A catheter was subsequently advanced to the right proximal portion of the internal iliac artery. The arterial phase of right internal iliac angiography showed numerous expanded arteries within the uterine tumor and early drainage of about three veins into the left internal iliac vein, rather than into the right ovarian vessels (Fig. 3A). Flow from the left internal iliac artery was normal and independent of the AVM (Fig. 3B). AVM in the uterine tumor was thus confirmed. A 5.2-Fr catheter with 9-mm occlusive balloons (Selecon MP; Clinical Supply, Gifu, Japan) was positioned with the tip in the proximal portion of the internal iliac artery, just after the bifurcation from the common iliac artery. After inflation of the occlusion balloon, angiography confirmed correct placement of the occlusion balloon into the right internal iliac artery and revealed stagnant flow (Fig. 3C). The balloon was then

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**Fig. 2.** CT, MRI and MRA.  
A) Coronal pelvic CT in the arterial phase shows large enhancing vessels in the uterine cervical tumor and swollen right gonadal veins. B) Sagittal pelvic T2-weighted MRI shows a diffuse uterine cervical tumor. C) MRA shows dilatation of numerous vessels in the pelvis.
deflated and with the arterial sheath and balloon catheter secured in place, and the patient was transferred to the operation room. After abdominal angiography, total hysterectomy was performed. Intraoperative findings showed the uterine corpus on the left posterior side of the cervical tumor (Fig. 4A), with normal bilateral adnexa. As preoperative angiography had shown that the right ovarian veins were not the vessels draining this AVM, the right infundibulopelvic ligament was first doubly ligated and transected. After inflating the internal iliac artery balloon, the peritoneum was incised and both the right uterine and superior vesical arteries were exposed, then clamped, ligated and transected. The right ureter was exposed, and it was laterally displaced by the cervical myoma. Oozing occurred from many right-sided small vessels in the retroperitoneal space dissected from the uterine cervical tumor, bladder and paravaginal vessels and should be controlled. Conversely, the procedure for the left side was performed smoothly. The amount of bleeding was about 2,200 ml by the completion of hysterectomy. After abdominal hysterectomy, the balloon catheters were deflated, and oozing from many dissected vessels around the cervical myoma increased. Total blood loss was approximately 3,800 ml, requiring blood transfusion. On opening of the resected tumor, numerous markedly expanded veins were found (Fig. 4B). Histopathology showed large numbers of morphologically abnor-
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Mal vessels with the presence of both arterial and venous elements (Fig. 5), confirming uterine AVM. The postoperative course was uneventful and the patient was discharged after 9 days.

Discussion

Uterine AVM is considered a rare and life-threatening entity, although its description has been limited to isolated case reports and small case series. Uterine AVM can be classified as congenital or acquired (Vogelzang et al. 1991). Causes of acquired uterine AVM are usually traumatic, resulting from prior dilation and curettage, therapeutic abortion, uterine surgery, and direct uterine trauma, or less commonly retained products of conception, trophoblastic disease, choriocarcinoma, or other gynecologic malignancies (Delaloye et al. 1998). In the present patient, uterine AVM arose in a cervical myoma with many extremely swollen vessels, expanded feeding arteries and drainage veins, and the patient had no history of abdominal or pelvic surgery. AVM has to be differentiated from arteriovenous fistulas and hemangiomas (Game et al. 2002). An arteriovenous fistula is a solitary acquired communication between an artery and a vein secondary to erosion or trauma (Trout et al. 1986). A hemangioma is a congenital neoplasm of vascular tissue involving small vessels (Game et al. 2002). In our patient, the histological features of this tumor were different from hemangioma. Therefore, this tumor was diagnosed as AVM in a cervical leiomyoma. Myoma of the uterine cervix accounts for about 5% of all myomas and enlargement of the cervix causes displacement of the surrounding bladder, ureter and uterine blood vessels. Surgical treatment is thus more difficult than that for uterine corpus myoma. Only one previous report has described AVM arising in a uterine myoma. In that case, the AVM was subtle and non-target embolization resulted in fatal systemic and pulmonary embolism (Stone et al. 1965). Obviously, a detailed preoperative diagnosis using several imaging modalities was not performed. Our clinical findings and the progress of treatment performed in the present patient would thus be valuable for similar patients.

Methods for diagnosing uterine AVM include ultrasonography, CT, MRI, and angiography (Grivell et al. 2005; Cura et al. 2009). Gray-scale ultrasound findings for AVM include multiple anechoic structures with serpentine contours within the myometrium, while the addition of color Doppler provides diagnostic sensitivity and a more accurate, noninvasive method of investigation (Huang et al. 1998). On color Doppler imaging, AVM typically appears as vascular tangles of tortuous vessels, with high-velocity, low-resistance flow (Timmerman et al. 2003). In our case, color Doppler imaging showed vascular tangles of tortuous vessels, but with low-velocity, low-resistance flow. Such findings, however, cannot rule out AVM from other possible
vascular diseases. CT and MRI are very useful in determining the size, extent, and vascularity of AVM and defining the involvement of adjacent organs (Gulati et al. 2000). Digital-subtraction angiography remains the gold standard for diagnosis, with the added advantage of allowing the options of embolization or balloon occlusion as preoperative treatment (Grivell et al. 2005; Cura et al. 2009). However, this is rarely performed for purely diagnostic purposes, due to its invasive nature. In the present case, angiography showed hypertrophy of the right uterine arteries feeding a tortuous, hypertrophic arterial mass with large accessory feeding vessels and early drainage into enlarged hypertrophic left internal iliac veins, revealing that this vascular anomaly in the tumor represented an AVM. Moreover, these findings enabled us to perform initial dissection of the right ovarian artery and vein and ligate the right uterine and superior vesical artery, providing important information for surgical management. Angiography is indeed invasive, but also supplies valuable information for reaching the true diagnosis and planning surgical procedures.

The first treatment option for uterine AVM is hysterec-
omy, while a second option is uterine artery embolization (UAE), which is applied to patients wanting to preserve reproductive capability (Huang et al. 1998). Grivell et al. (2005) stated that with increased experience and the development of embolization techniques and equipment, embolization should become the first choice for treatment in women of all age groups, not only those desiring future fertility. Many cases of acquired uterine AVM are associated with uterine trauma such as obstetric procedures, and such cases typically undergo UAE with gelatin sponge particles. In the present case, the patient was a postmenopausal woman and did not express any desire for preservation of the uterus, moreover vascular anomalies were almost entirely located within the cervical tumor, which showed a smooth surface with no invasion into surrounding organs, so this tumor was considered resectable and hysterectomy was performed to clarify the histology of the tumor.

Surgical management of AVM in various organs requires ligation of the feeding arteries to reduce blood supply to the drainage veins. We planned right uterine artery ligation after transection of the right infundibulopelvic ligament. The large tumor arising from the uterine cervix occupied the entire pelvic cavity and access to the retroperitoneal space was limited. We therefore decided to perform temporary endovascular balloon occlusion of the affected unilateral internal iliac artery for the immediate management of hemorrhage from retroperitoneal arteries during dissection of fibro-fat-ty-connective tissue. The usefulness of temporary endovascular balloon occlusion of bilateral internal iliac arteries has been reported in the management of obstetric hemorrhage due to abnormal placentation (Carnevale et al. 2011), myomectomy (Takeda et al. 2009) and hysterectomy procedures in cases of large cervical myoma (Takeda et al. 2011). In our case, bleeding increased after balloon deflation, indicating that unilateral occlusion of the internal iliac artery was considerably effective in controlling bleeding. However, to achieve much reduction of hemorrhage during hysterectomy, bilateral occlusion of the internal iliac artery or perioperative vascular embolization should be attempted.

Surgical resection of AVMs carries the risk of a massive intraoperative hemorrhage, incomplete removal of the AVM nidus, surrounding organ injury, and high recurrence rates (Do et al. 2012). Therefore, endovascular therapy with various embolic and sclerosing materials, independently or in combination with surgical treatment, has become an accepted therapeutic option (Do et al. 2012). Surgical resection with preoperative intra-arterial embolization with rapid-setting liquids or particles after embolization of the draining vein with larger coils to decrease the blood velocity in AVM (Do et al. 2012) could also have been effectively applied to control bleeding in our case.

Uterine AVM arising in the uterine cervical fibroid is extremely rare and this case represents the first report of a postmenopausal patient who underwent surgical treatment of bulky cervical uterine fibroid with intrafibroid AVM. Preoperative pelvic angiography appears highly useful for diagnosing the true feeding artery and drainage veins and assisting the planning of surgical procedure. Occlusion of the feeding artery with a balloon device is effective in decreasing intraoperative bleeding, but presents a potential risk of bleeding after balloon deflation. AVM in a large uterine cervical myoma is a challenging lesion; namely, careful management plan is essential to achieve a complete cure with acceptable morbidity.

Conflict of Interest

The authors report no conflict of interest.

References


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