Journal of Korean Medical Science
Vol. 6, No. 2, 187-190 June 1991

Arteriovenous Malformation of the Uterus
—A Cause of Massive Operative Bleeding—

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Arteriovenous malformations of the uterus are extremely rare and they occur either in congenital or acquired forms. The most common clinical presentation is abnormal uterine bleeding, which may be aggravated by therapeutic curettage. Because of their rare incidence and clinical importance in management of patients, we report a case of arteriovenous malformation causing serious bleeding during a hysterectomy for uterine leiomyoma. The patient was a 47-year-old multiparous woman who had a history of chronic vaginal bleeding for one year. Numerous anomalous blood vessels draining into the right and left uterine arteries were found on the anterior wall of the uterus and parametrium.

Key Words : *Arteriovenous malformation, Uterus, Operative bleeding.*

INTRODUCTION

Arteriovenous malformations of the uterus are rare lesions which have not yet been well described in common obstetric and gynecologic textbooks(Blaunstein, 1982 ; Fox, 1982). The recognition of these malformations is particularly important to clinicians because of the potential hazards of curettage in control of uterine bleeding. Pathologically, these lesions should be differentiated from the highly vascularized leiomyoma. We present here a case of arteriovenous malformation involving the anterior wall of the uterus and parametrium, which is responsible for serious bleeding during hysterectomy for leiomyoma.

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CASE HISTORY

The patient was a 47-year-old multiparous woman(3-0-4-3) who had a history of chronic vaginal bleeding since November 1989. One year later, endometrial curettage was done at a general hospital because of repeated vaginal bleeding without stopping. Under the impression of uterine myoma, the patient was transferred to Korea University Hospital. She had a history of anticoagulant therapy for ischemic heart disease for an unknown period. At the time of her admission to this hospital, her hemoglobin and hematocrit were 9.3 g/dl and 26.9%, respectively. On operation, the uterus was slightly enlarged, and there was adhesion between the anterior uterine fundus and right bladder dome. Both adnexae were free from any lesions. On dissection of the adhesion and clamping both uterine arteries, massive bleeding occurred. The subtotally resected uterus was submitted for pathologic examination to find any lesions responsible for the massive bleeding. The bleeding was controlled by

gauze packing for one and a half hours. The postoperative course was uneventful and the patient was discharged.

PATHOLOGIC FINDINGS

The subtotally resected uterus and the attached right adnexa were removed. The uterus measured 6×4×3.5㎝ and weighed 100 g. The previousely bisected uterus showed a 3㎝ infarcted intramural myoma in the anterior fundus. On the anterior fundic portion, markedly dilated right and left uterine arteries were found. On tracing the arteries, both had several tributaries and drained to the myometrium. The anterior half of myometrium contained a conglomerated mass of blood vessels varying in size and thickness with intervening hypertrophied muscle fibers.

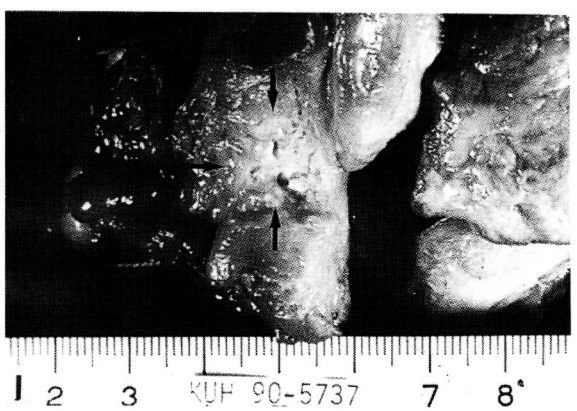

Fig. 1. An ill-defined conglomerated mass of various sized blood vessels(arrows) in the myometrium is shown.

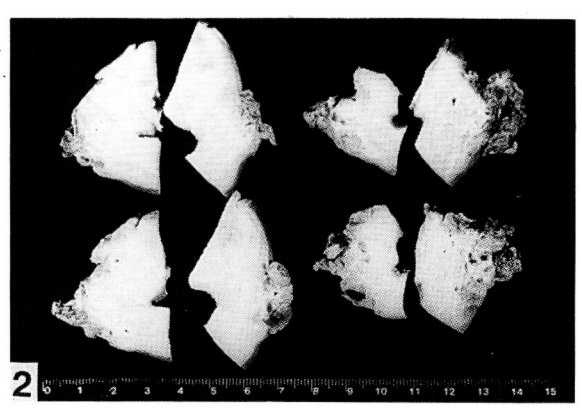

Fig. 2. Small and large blood vessels found in the myometrium and parametrium.

Fig. 3. Irregular-shaped blood vessels are gr ouped within the myometrium(Masson-Trich rome, 1x original magnification).

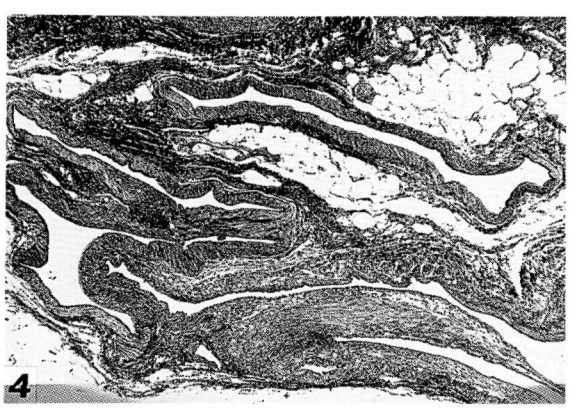

Fig. 4. Medium-to-large arteries and veins, with varying thickness of the wall are seen in the parametrium(H-E, x 40).

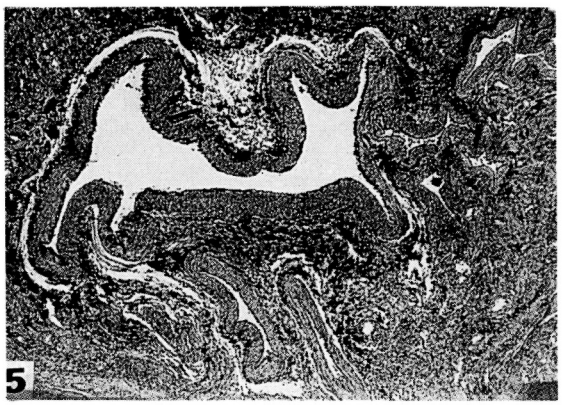

Fig. 5. Malformed blood vessels are grouped in the myometrium(H-E, x 40).

The anterior parametrium close to the uterine arteries contained various sized blood vessels as well(Fig.1 and 2).

Histologically, the myometrium and parametrium contained a mixture of small and large blood vessels(Fig.3). Most of the vessels were muscular arteries and veins with varying degrees of intimal thickening, irregular thinning and thickening of the media(Fig.4 and 5.) Elastic stain showed fragmentation, duplication and loss of internal elastic membrane. The intervening smooth muscle fibers were hypertrophied such as muscular hypertrophy in the adenomyosis.

DISCUSSION

Although uterine leiomyomas showing prominent vascular proliferation are not uncommon, genuine vascular lesions are extremely rare. Among these vascular lesions, 37 cases of arteriovenous malformations have been described in English medical literature as cirsoid aneurysm(Hibbard et al., 1972 ; McLachlan, 1978), arteriovenous fistula(Fulmer et al., 1970 ; Gaylis et al., 1973 ; Antebi et al., 1975 ; Forssman et al, 1982), hemangioma(Dawood et al., 1972), vascular malformation(Follen et al., 1985), arteriovenous sinus(Howard, 1968) and later as arteriovenous malformation (Szalay et al.,1979 ; Ghosh, 1986 ; Poppe et al., 1987 ; Fleming et al., 1989). A single case of arteriovenous malformation causing massive postpartum bleeding has been reported in Korea(Kim et al., 1990).

The ages of the patients ranged from 18 to 72 years. The most common clinical presentation was menometrorrhagia or postmenopausal bleeding which was difficult to control and which was even aggravated by curettage. Sometimes, uterine enlargement, palpable pulsation and audible murmur with or without thril on the abdomen were presenting symptoms. The case reported by Bottomley and Whitehouse(1975) was regarded as congenital in origin. However, most of the reported cases were acquired in association with previous surgery(Fulmer et al., 1970 ; Saunders and Ellis., 1972; Gaylis et al., 1973: Antebi et al., 1974), trophoblastic disease (Hibbard et al., 1972: McLachlan, 1978), or diethylstilbestrol exposure(Follen et al., 1985). Although our patient had a history of abortions, immediate responsible factors for this malformation were not identified. Her intermittent vaginal bleeding was not controlled by repeated curettage.

Pathologically, vascular malformations are usually described as the cirsoid type composed of multiple arteriovenous fistula within a conglomeration of dilated arterial and venous channels. Several vessels, such as the uterine, ovarian, and hypogastric arteries, may supply the vascular malformation ; they are dilated and elongated. The corresponding vessels may be also dilated. Like the cases in the other locations, a distinction between arteries and veins has proven to be difficult because of the intimal thickening of the veins as a result of direct transmission of high arterial blood pressure (Fleming et al., 1989). Uterine arteriovenous malformations should be differentiated from the vascular leiomyomas which have a prominent vascular proliferation. In the latter, the blood vessels are capillary in size rather than large vessels. In our case, the intervening smooth muscle fibers between the vessels were hypertrophied similar to the muscular hypertrophy in adenomyosis, without circumscription as in leiomyomas.

The diagnosis of this type of vascular malformation is usually made retrospectively after hysterectomy, but it may be also made by uterine angiography(Ghosh, 1986 ;

Fleming et al., 1989) and ultrasonography (Poppe et al., 1987 ; Fleming et al., 1989).

Most of the reported cases were treated by total hysterectomy. However, some cases have been treated by arterial embolization (Forssman et al., 1982 ; Ghosh, 1986 ; Poppe et al., 1987) and ligation with or without excision of arteriovenous malformation(Bottomly and Whitehouse, 1975). Preoperative diagnosis by angiography and successful treatment by embolization may be considered one of the therapeutuc modalities in women who desire to retain childbearing potential. Endometrial curettage must be carefully done because it may aggravate the bleeding. Arteriovenous malformation in our case was not recognized before hysterectomy for leiomyoma.

The recognition of uterine arteriovenous malformation in cases of abnormal uterine bleeding not controlled by curettage can make the appropriate treatment of the patient possible.

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