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CASE REPORT

Spontaneously Ruptured Uterine Angioleiomyoma

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Angioleiomyoma is an uncommon type of leiomyoma of the uterus that originates from smooth muscle cells and contains thick-walled vessels. A 45-year-old woman with the complaint of lower abdominal pain was admitted to the hospital. In the operation a ruptured, bleeding uterine tumor was seen. She underwent total hysterectomy and bilateral salpingo-oophorectomy. On pathologic examination of the specimen, the tumor was diagnosed as angioleiomyoma. Here, an unusual clinical presentation of uterine angioleiomyoma was reported. (Pathology Oncology Research Vol 12, No 1, 50–51)

Key words: angioleiomyoma, uterus, genital tract

Introduction

Angioleiomyoma is a benign mesenchymal neoplasm that is composed of smooth muscle cells and thick-walled vessels. Angioleiomyoma is usually found in the skin of the lower extremities and only a few cases of uterine angioleiomyoma have been reported[1]. We present here a spontaneously ruptured uterine angioleiomyoma in a woman presenting with severe intraabdominal bleeding.

Case report

A 45-year-old multipara premenopausal woman complaining of lower abdominal pain, nausea and vomiting was referred from local state hospital to our emergency department. She was on medication for hypertension. Clinical examination revealed a painful suspicious mass in the right adnexal region and culdocentesis revealed intraabdominal bleeding. Color Doppler ultrasonography showed a mass with abnormally increased vascularization in the right adnexal region and intraperitoneal free fluid within the abdomen.

Hemoperitoneum was encountered at the surgery and approximately 1500 cc of blood was aspirated. Approximately 6.5 cm at its greater diameter, a leiomyoma-like ruptured mass with vascular structures, originating from uterine fundus, was still bleeding, which was not associated with a specific vessel. The mass was not communicating with the uterine cavity. Two units of whole blood were transfused intraoperatively. A total abdominal hysterectomy and bilateral salpingo-oophorectomy was carried out. The postoperative period was uneventful.

Grossly, a 184 g uterus, measuring 8x7x3 cm, and a well-circumscribed tumoral mass with blood-filled cystic spaces, measuring 6.5 cm in diameter, with rupture, was found. The cut surface was white-yellow in color and exhibited hemorrhage.

On microscopic examination, the nodule was composed of interlacing smooth muscle bundles, intermingled with abundant thick-walled vessels (*Figure 1*). There were no mitotic figures, pleomorphism or necrosis. The vascular and spindle cell components were immunoreactive for vimentin and smooth muscle actin with no reactivity to desmin and S100. The final histopathologic diagnosis was angioleiomyoma. There was no evidence of malignancy. Ten months after surgery the patient is alive with no evidence of disease.

Discussion

Leiomyomas are the most common uterine neoplasms.[2] However, there are several uncommon specific types of leiomyomas. Although the karyotype evaluation did not fit classical subgroups of ordinary uterine leiomyoma,[3]

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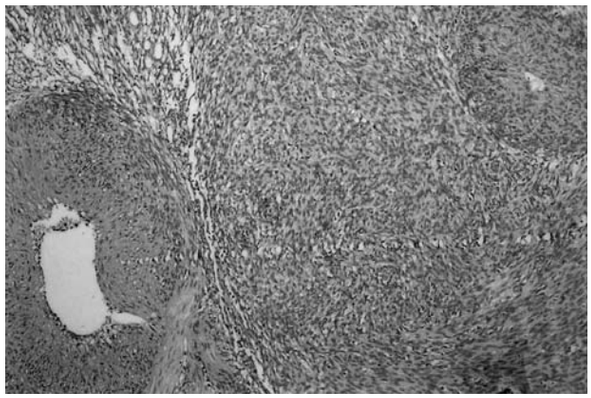

Figure 1. *Microscopic examination reveals numerous thick-walled vessels interspersed between smooth muscle cells (HE, x100)*

angioleiomyoma, a solitary form of leiomyoma, usually occurs in the subcutis, most often in the lower extremities.[1] It can also be located in the head and neck region, even in the submandibular gland.[4,5] Few cases of angioleiomyoma have been described at sites other than the extremities and head. According to Hsieh et al., the number of uterine angioleiomyomas reported was around six.[6]

Angioleiomyoma is a benign neoplasm that is composed of smooth muscle cells and thick-walled vessels. It develops usually between the fourth and sixth decades. It may be small and painless when it is located in the head and neck region.[4] However, pain is the dominant clinical feature of the uterine angioleiomyoma as it was in our case. The mechanism of pain is inconclusive, it may be attributable to local ischemia from vessel contraction.[4] Uterine angioleiomyoma can cause severe menorrhagia and can be huge and multiple.[6] It was stated that local dysregulation of the vascular structures in the uterus is responsible for abnormal bleeding.[7] Leiomyomas contain venous plexuses, and certain growth factors have been suggested as candidates that cause abnormal bleeding. Our patient was on medication for hypertension, therefore, heavy bleeding may have been due to hypertension.

Grossly, the tumor presents as circumscribed, gray-white nodules. Sometimes the tumoral mass can have dilated vessels that can be mistaken for multiloculated and multiseptated ovarian tumor or adenomyosis.[8] In our case nodular, cystic and ruptured mass was found. Microscopically, the well-demarcated nodule was composed of whorled, anastomosing fascicles of uniform, fusiform smooth muscle cells with thick-walled vessels. Areas of myxoid changes, hyalinization, calcification, and fat may be seen. Mitotic figures and necrosis are infrequent. Angioleiomyomas are classified into three histological types: capillary or solid, cavernous, and venous.[6] The nature of vessels is controversial. Some authors suggested that these tumors are probably derived from arteriovenous anastomoses.

As in our case, clinical diagnosis may be difficult, especially when degenerative changes occur. Preoperatively, our presumable diagnosis was a malignant gynecological tumor or ectopic pregnancy because of heavy bleeding. There was no evidence of malignancy in our case. It can be difficult to distinguish an angioleiomyoma from a hemangioma or an arteriovenous malformation if the vascular component predominates. Angioleiomyomas are well circumscribed neoplasms that contain at least foci of typical spindled smooth muscle cells.[2] Hemangiomas are rare in the uterus, and tend to be poorly defined grossly and microscopically. Special stains for smooth muscle cells, such as actin, and vessel markers as CD34 and CD31 are necessary to differentiate angioleiomyoma from other neoplasms such as angiofibroma, fibroma, angiomyolipoma, angiomyofibroblastoma. Angiomyofibroblastoma is positive for vimentin, desmin, but negative for smooth muscle actin.

Surgical excision is the treatment of choice. Either angiomyomectomy or simple hysterectomy for women who do not wish to have more children proved to be an effective treatment. In our patient hysterectomy and bilateral salpingo-oophorectomy was performed.

Briefly, angioleiomyoma should be included in the differential diagnosis of a multicystic mass located in the pelvis. It should be considered when prominent tortuous vascular-like structures are noted on CT examination arising from the uterus.[6] As in our case, it should be kept in mind when there is intraabdominal bleeding, because uterine angioleiomyoma may manifest unusual presentations.

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