Uterine angioleiomyoma causing severe abnormal uterine bleeding

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Summary

Background: Angioleiomyoma or angiomyoma or vascular leiomyoma is an unusual benign mesenchymal neoplasm. The authors present a rare case of large uterine angioleiomyoma causing severe abnormal uterine bleeding. *Case:* The patient, a 53-year-old, gravida 2, para 2, premenopausal Greek woman presented with a complaint of severe abnormal uterine bleeding. On gynecologic examination there was a palpable pelvic mass. Preoperative computer tomography (CT) of the abdomen and pelvis revealed an intra-abdominal mass 25 x 15 cm with abnormally increased vascularization. She underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy, total omentectomy and elective pelvic lymph node dissection. Histopathology revealed uterine angioleiomyoma. Follow up 84 months after initial surgery showed no evidence of recurrence. *Conclusion:* Despite the type of surgery, patients with uterine angioleiomyoma have very low risk of recurrence and excellent prognosis.

Key words: Angioleiomyoma; Angiomyoma; Vascular leiomyoma; Uterus.

Introduction

Angioleiomyoma or angiomyoma or vascular leiomyoma is an unusual benign mesenchymal neoplasm [1. It is composed of smooth muscle cells and contains thick-walled vessels [1]. It most commonly occurs in the skin of lower extremities, head and trunk [2]. Uterine angioleiomyoma is extremely rare and only few cases have been reported in the English literature [1, 3-11].

The authors' aim was to present a rare case of large uterine angioleiomyoma causing severe abnormal uterine bleeding and review current literature.

Case Report

The patient, a 53-year-old, gravida 2, para 2 premenopausal Greek woman presented to the 2nd Department of Gynaecology of St. Savvas Anticancer-Oncologic Hospital, with a complaint of severe abnormal uterine bleeding during the last six months. Her past surgical history was unremarkable. She was taking medication for arterial hypertension. Her family history revealed no evidence of cancer among the first-degree relatives.

On gynecologic examination, there was a palpable pelvic mass. There were no palpable inguinal lymph nodes and the rest of pelvic examination was normal.

Preoperative computer tomography (CT) of the abdomen and pelvis, abdominal and transvaginal ultrasound (U/S) revealed an intra-abdominal mass 25 x 15 cm with abnormally increased vascularization. Preoperative CT of the chest, chest X-ray, intravenous pyelography (IVP), colonoscopy, and urethrocystoscopy were normal. Preoperative CA-125 was 8.7 U/ml. Preoperative hemoglobin was 8 g/dl.

Exploratory laparotomy recognized a solid mass 25 x 15 x 12 cm with many dilated vessels on the right sidewall of the uterus. There were firm adhesions between tumor and bladder that rendered excision difficult. Frozen section showed mesenchymal tumor with suspicion of malignancy. The patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy, total omentectomy, and elective pelvic lymph node dissection.

Multiple histopathological sections taken from the whole specimen showed: interlacing smooth muscle bundles without atypia or necrosis, intermingled with abundant thick-walled vessels (Figure 1). Also, the vascular and spindle cell components were immunoreactive for vimentin and smooth muscle actin. The omentum and all removed pelvic lymph nodes were negative for malignant disease. The peritoneal washing smear was negative for malignant cells. The final diagnosis was uterine angioleiomyoma.

The postoperative period was uneventful. Follow up 84 months after initial surgery, with CT of the abdomen and pelvis, abdominal U/S, and chest X-ray showed no evidence of recurrence.

Discussion

Angioleiomyoma is an unusual benign mesenchymal neoplasm. It most commonly occurs in the subcutis of the lower extremities [1,2]. It can also be located in the head and neck region, even in the submandibular gland [1,2,12,13].

Only few cases of angioleiomyoma of the female genital tract have been reported in the English literature [1,3-11,14-16]. Almost all of them located in uterus and only two cases located in ovary [1,3-11,14,15]. Perhaps they are underreported, because in many cases they are also recognized as benign leiomyomas with an unusual histological appearance [6].

It is a well-circumscribed or encapsulated tumor and contains minimal collagen [7]. It is composed of smooth muscle cells and contains thick-walled vessels [1]. The nature of vessels remains controversial [1]. An appreciable number

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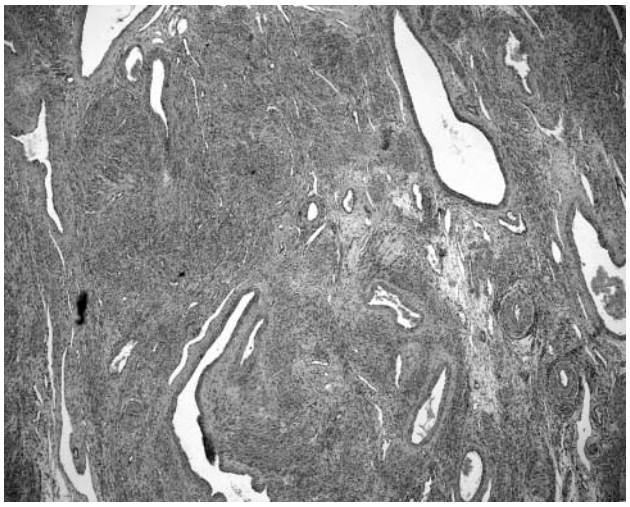

Figure 1. — Uterine angioleiomyoma: interlacing smooth muscle bundles intermingled with abundant thick-walled vessels (Haematoxylin - Eosin x 40).

of angioleiomyomas are not true tumors but rather instances of vascular malformation [2, 17]. Angioleiomyoma most likely derives from tunica media of small blood vessels or from arteriovenous anastomoses [1,2,7,17,18]. Also some cases of angioleiomyomas are hamartomas, because they have variable proportions of mature adipose tissue, smooth muscle cells and anomalous thick-walled vessels [2].

They are classified into three histological subtypes: capillary or solid (66%), cavernous (11%) and venous (23%) [2,14,18]. Classification is based on the variable relationship among smooth muscles and vascular cavities of different shapes [4]. However, histological subtypes have no clinical significance [2].

It usually develops in women between the fourth and sixth decade of life [1,2,19]. Degenerative changes in angioleiomyomas are due to ischemia and they depend on the degree and rapidity of the onset of vascular insufficiency [2]. Myxoid and hyaline changes are the most common forms of degeneration [2,18].

The clinical presentation of uterine angioleiomyoma is usually nonspecific. The most common presenting symptoms and signs are: abdominal/pelvic pain, abdominal/pelvic mass and abnormal uterine bleeding [4,7,20]. The present patient presented with a large uterine angioleiomyoma causing severe abnormal uterine bleeding.

The exact mechanism of abdominal/pelvic pain in patients with uterine angioleiomyomas, remains inconclusive [1]. Perhaps it is pain-related with local ischemia from vessel contraction [1,2,18].

Also uterine angioleiomyomas that contain venous plexuses and dysregulation of growth factors and/or their receptors that regulate angiogenesis or have other effects on vascular structures, are responsible for abnormal uterine bleeding [1,4,21,22].

Usually uterine angioleiomyomas are small in size [7]. However in some cases they have an unusual large size, causing pain, and/or severe abnormal uterine bleeding [4,7,8]. The present patient presented with a large uterine angioleiomyoma causing severe abnormal uterine bleeding. Despite the large size of tumor, the authors did not find degenerative changes on histopathological examination.

The nonspecific nature of symptoms, signs, and imaging findings of uterine angioleiomyomas renders preoperative diagnosis exceptional [23]. All reported cases are diagnosed after surgery [7,23].

Treatment of choice in patients with uterine angioleiomyoma is complete surgical excision [1,4,7]. Either angioleiomyomectomy or hysterectomy, are effective treatment approaches with excellent results [1,4,7]. The present patient underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy, total omentectomy, and elective pelvic lymph node dissection because frozen section revealed mesenchymal tumor with suspicion of malignancy.

The present patient is well with no evidence of recurrence, 84 months after initial surgery. Her prolonged survival is in accordance with current literature [2,6,7]. Despite the type of surgery, patients with uterine angioleiomyoma have very low risk of recurrence and excellent prognosis [2,6,7].

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