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Giant angioleiomyoma of uterus: A case report with focus on CT imaging

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ABSTRACT

We report a rare case of giant angioleiomyoma located in the uterus and detected in a 37-year-old woman. The uterus is an extremely rare location for angioleiomyoma. The definitive diagnosis is usually obtained only after the histopathologic examination because the imaging criteria are challenging for this disease. We focused our attention on the main computed tomography features able to provide a robust preoperative diagnosis of this rare clinical entity.

Introduction

Angioleiomyoma of uterus is a very rare benign tumor, originating from mesenchymal tissue [1]. It develops in the lower extremities of middle-aged women and rarely affects other body regions [1]. The uterus is an extremely rare location for angioleiomyoma [2,3]. On imaging, the diagnosis is very difficult, and the final diagnosis is usually formulated only when the histopathologic investigation has been performed [1].

We report a rare case of giant angioleiomyoma detected in a 37-year-old woman who came to our attention for anemia, menorrhagia, and a very prominent abdomen.

We focused our attention on the main computed tomography (CT) features useful to provide a robust preoperative diagnosis of this rare clinical entity.

Competing Interests: The authors declare that they have no conflicts of interest.

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Case presentation

A 37-year-old female was admitted to the surgery-gynecology-oncology department with a 1-year history of abdominal distension, menorrhagia, and dyspepsia. A central abdominal mass was evident on clinical examination. The CA-125 and hemoglobin levels were 304 IU/mL (normal < 35 IU/mL) and 9 g/dL, respectively.

Ultrasound scan revealed a large solid abdominal mass, extending from pelvis to epigastric region, suggesting a malignant ovarian tumor or a uterine sarcoma.

An abdomino-pelvic CT with and without contrast medium was performed, showing an oval giant abdominal mass, with dimensions of 32 × 30 × 25 cm, occupying the entire abdomen with suspect origin from uterine fundus.

On contrast-enhanced CT scan, in the arterial and venous phase, the giant mass showed multiple vascular branches. In the late phase, the mass showed an inhomogeneous "sand-like" enhancement (Fig. 1 and Fig. 2).

At CT scan the giant mass was found inseparable from uterine fundus. In addition, a large bilateral pelvic varicocele and an evident uterine arteries hypertrophy were present (Fig. 3). All the abdominal organs were dislocated by the mass without any evidence of infiltration.

All these findings supported the hypothesis of a uterine origin of the mass. Moreover, the abundant presence of vascular structures within the mass suggested the hypotheses of a giant uterine angioleiomyoma.

The laparotomy, performed under general anesthesia, confirmed the uterine origin of the giant mass (as reported in Fig. 4); then, a total hysterectomy was performed. The weight of the giant mass was 12.5 kg.

Macroscopically, the cut surfaces were white with a variegated appearance with pinkish brown and gray areas (Fig. 5A and B); no region of necrosis were found within the lesion. Histologically, the tumor was composed of interlacing fascicles of spindle cells with interspersed abundant thick walled blood vessels (Fig. 5C and D).

Discussion

Angioleiomyoma of the uterus, also known as vascular leiomyoma, is a very rare benign tumor, originating from the mesenchymal tissue and composed of smooth muscle cells and thick-walled vessels [1]. Angioleiomyoma occurs more frequently in the lower extremities and rarely affects other body regions [1]. It represents the 0.34%-0.40% cases of uterine leiomyomas [2].

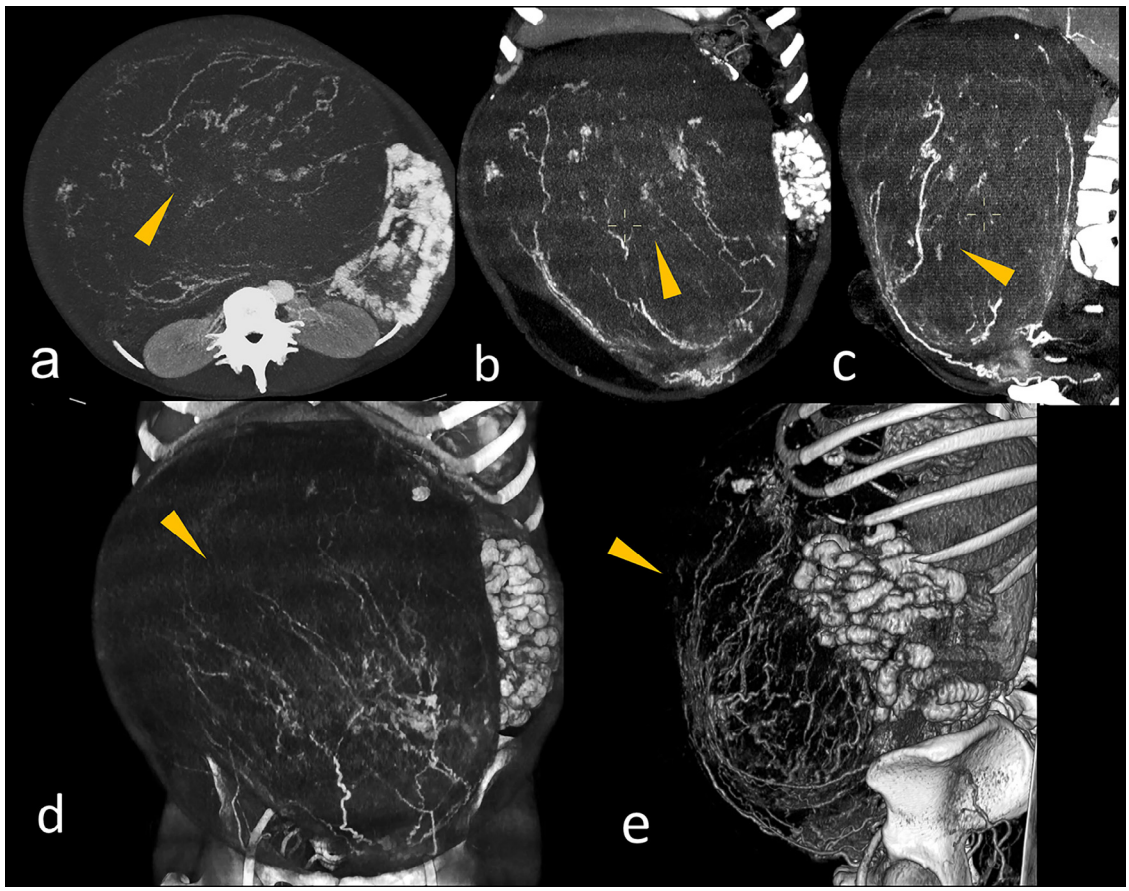

Fig. 1 – Axial (A), coronal (B), and sagittal (C) computed tomography (CT) maximum intensity projection (MIP) images showing multiple vessels crossing the abdominal mass. CT coronal (D) and sagittal (E) volume-rendered image provides good 3D definition of the vascular distribution within the mass.

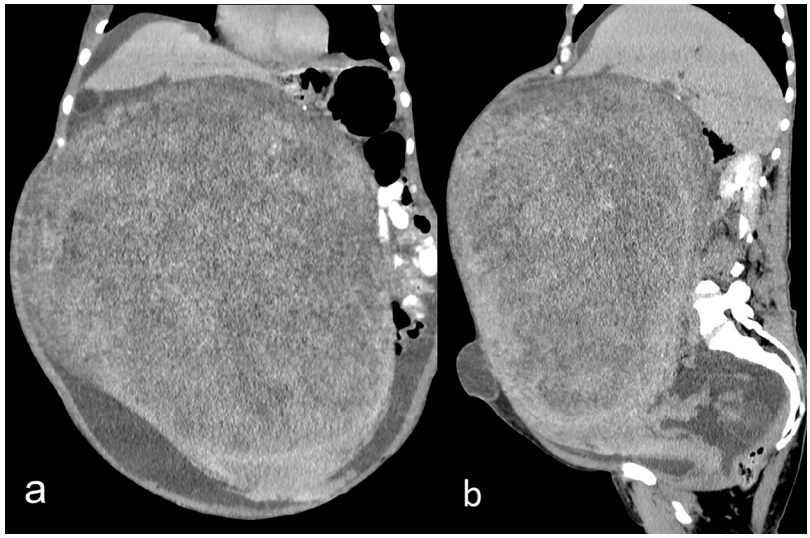

Fig. 2 – Coronal (A) and sagittal (B) ccomputed tomography (CT) images showing a "sand-like" enhancement of the mass in the late phase.

Angioleiomyoma usually develops in middle-aged women [1,3].

Based on the variable relationship among smooth muscles and vascular cavities, angioleiomyomas are classified into 3 histologic types: capillary or solid, cavernous, and venous [3]. Angioleiomyomas are well encapsulated and usually their dimensions are smaller than 2 cm, with a range of 0.2-4.3 cm [4].

Patients with uterine angioleiomyoma usually exhibit severe dysmenorrhea, abdominal pain or discomfort, menorrhagia, abdominal mass, and severe anemia [1]. Other possible clinical findings reported in the literature are consumptive coagulopathy, spontaneously ruptured with severe intra-abdominal bleeding, pseudo-Meigs syndrome, and a raised cancer antigen 125 level [5–7].

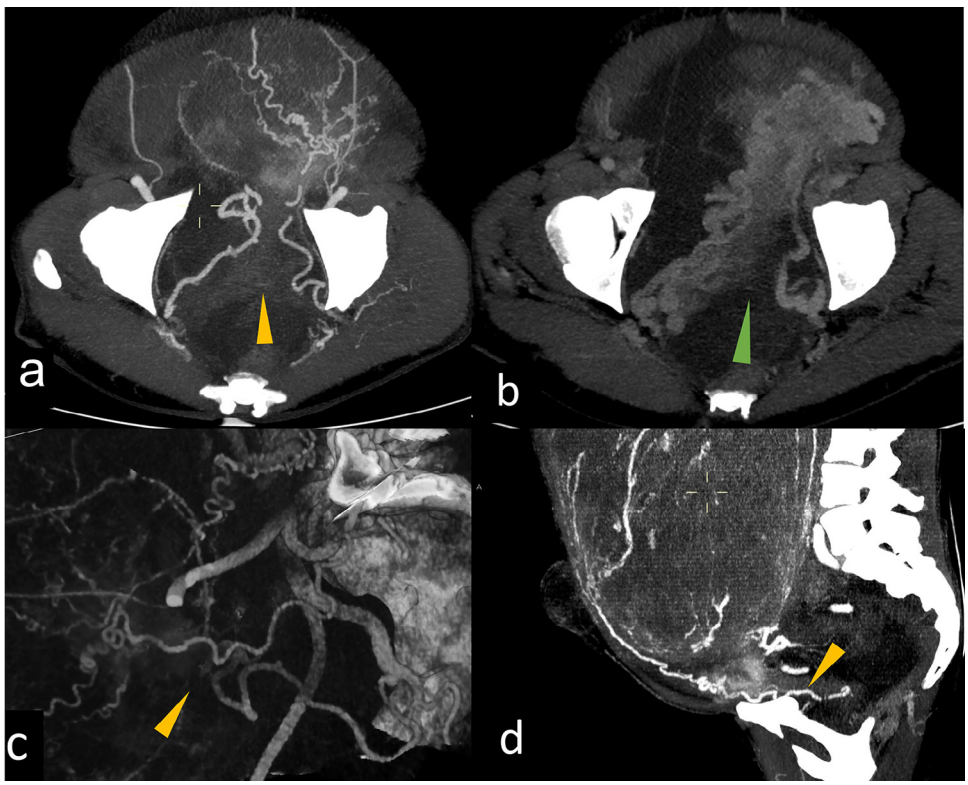

Fig. 3 – Axial computed tomography (CT) maximum intensity projection (MIP) images showing the hypertrophy of uterine arteries (yellow arrowhead in A) and the presence of a large pelvic varicocele (green arrowhead in B). Sagittal CT oblique volume-rendered image (C) provides good 3D definition of the uterine arteries (yellow arrowhead), and sagittal MIP images (E) show uterine artery hypertrophy.

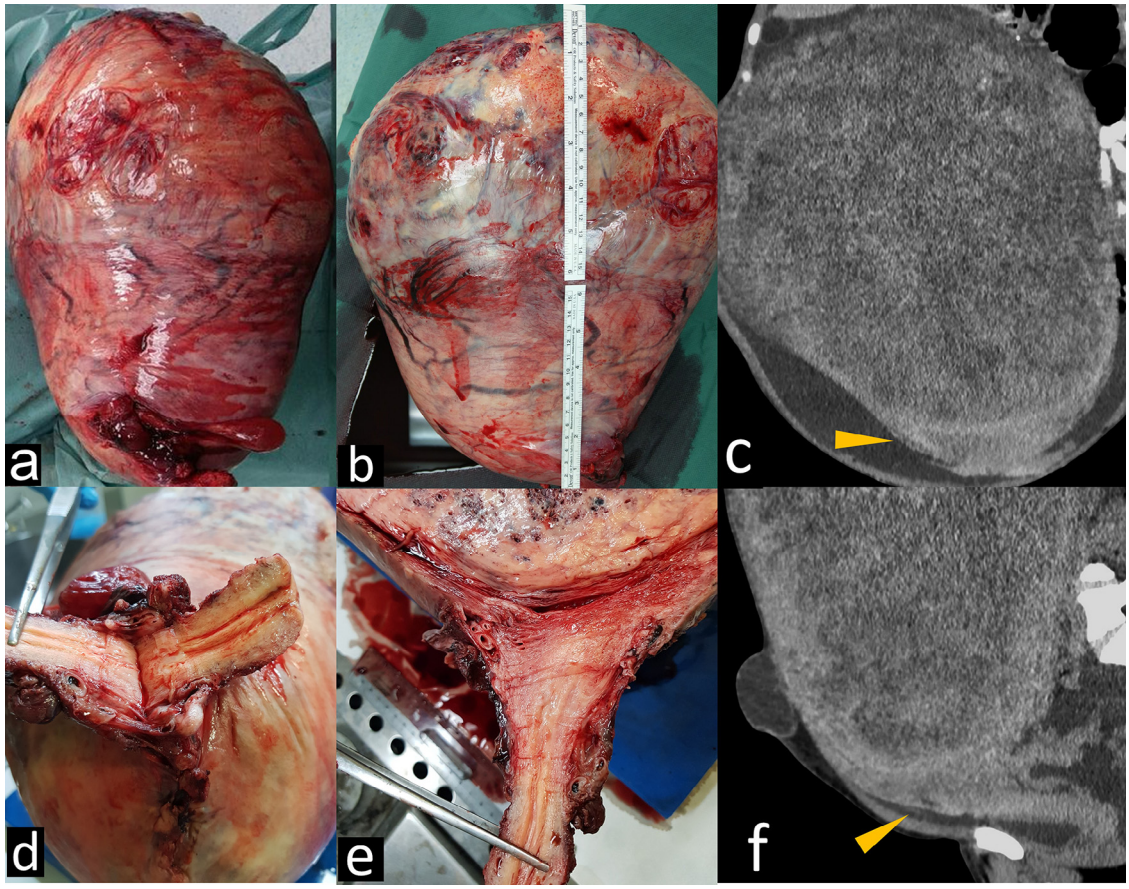

Fig. 4 – Gross photographs (A, B, D, and E) showing a large mass arising from the uterus. The corresponding computed tomography (CT) images, coronal (C) and sagittal (F), show the uterine origin of the mass (yellow arrowhead).

Angioleiomyoma of the uterus ranges from 4 cm to 30 cm in the greatest dimension; but few cases reach size greater than 20 cm [1–4,7–9].

Preoperative CT diagnosis of an angioleiomyoma is extremely difficult; the few cases reported in the literature do not provide common CT characteristics. Hsieh et al. [4] described typical CT findings of angioleiomyoma as a multilobulated mass with solid and laminated configuration, and cystic and multiseptal contents. Other authors do not confirm these findings and reported a single and unilocular uterine mass [3] or a single unilocular necrotic mass [5].

In any case, angioleiomyoma should be included in the differential diagnosis when the CT imaging highlights prominent tortuous vascular-like structures arising from the uterus [4]. However, from the literature, it emerges that a final diagnosis was made only when histopathologic investigation was performed [1,9].

In most cases, the surgical treatment is hysterectomy with or without resection of appendages; myomectomy, when technically possible, is rare, and it is considered only to preserve fertility [9].

To the best of our knowledge, our case reports one of the largest uterine angioleiomyoma described in the literature [1–4,7–9].

In our experience, the CT examination highlighted some findings that allowed us to hypothesize, before the surgery, the uterine origin of the mass and its histotype. First, the use of contrast-enhanced CT revealed the presence of multiple vascular branches within the giant mass (Fig. 1). Second, the CT scan in the late phase showed a inhomogeneous “sand-like” enhancement (Fig. 2). Last, the CT scan revealed the presence of a large bilateral pelvic varicocele and an evident uterine arteries hypertrophy (Fig. 3), associated with a giant mass inseparable from uterus. Although pelvic varicoceles may not necessarily be related to a giant angioleiomyoma, its presence, together with the hypertrophy of uterine arteries, has strengthened the hypothesis of a uterine origin of the tumor.

Despite the various CT patterns of uterine angioleiomyoma described in literature, the role of CT imaging remains essential in the differential diagnosis of this condition. Because of the frequent degenerative changes in the course of uterine angioleiomyoma, a sonographic examination (usually the first imaging approach) may show a large inhomogeneous abdominal mass that could lead to a misdiagnosis, erroneously suggesting a possible ovarian origin of the mass. For this reason, it is mandatory to perform a CT examination to obtain a more accurate characterization of the mass, in terms of site of origin, vascularization, and relationships with surrounding abdominal organs.

In conclusion, in our experience, the diagnosis of a giant uterine angioleiomyoma should be considered when, in a well-demarcated uterine soft tissue mass, the CT imaging

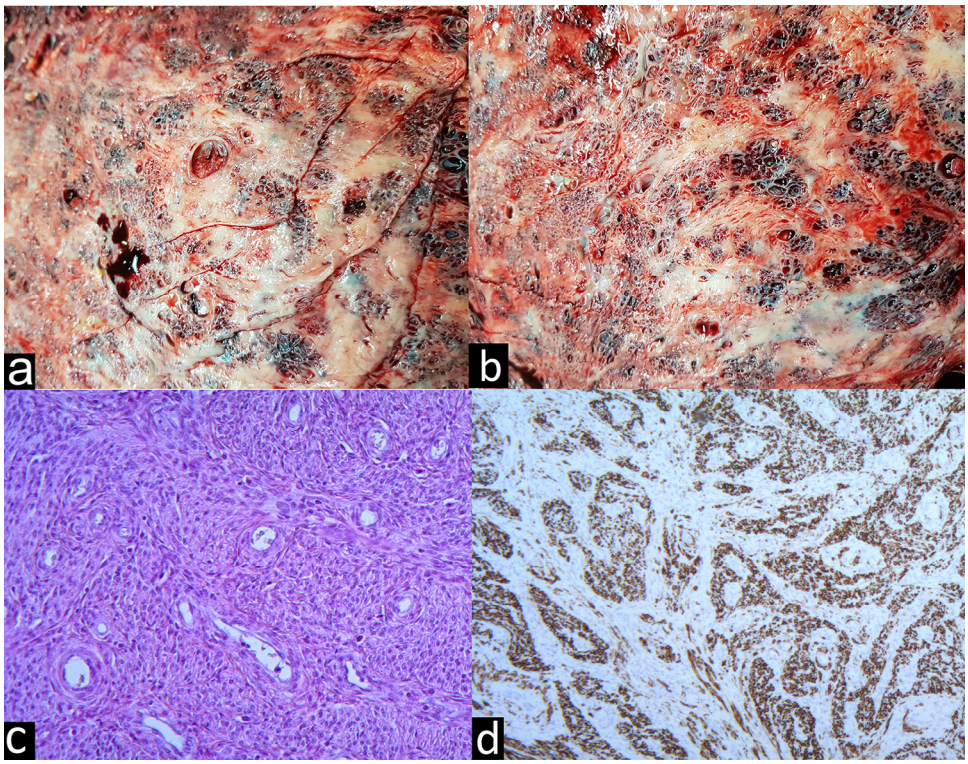

Fig. 5 – Macroscopic images (A) and (B) of uterine angioleiomyoma showing the white cut surfaces with a variegated appearance with pinkish brown and gray areas; multiple vessels are recognizable. Microscopic images of uterine angioleiomyoma showing (C) interlacing fascicles of spindle cells with numerous interspersed vascular channels (hematoxylin-eosin, original magnification ×10) and (D) desmin positive (immunoperoxidase stain for desmin, original magnification ×10).

shows a prominent, tortuous, vascular-like enhancing structures; a sand-like enhancement; and a large, bilateral pelvic varicocele with an evident hypertrophy of the uterine arteries.

Compliance with Ethical Standards: This study received no funding.

Ethical Approval: All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Consent

Written informed consent was obtained from the patient for the publication of this report and any accompanying images.

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