Archives of Gynecology and Obstetrics (2019) 300:841–847
https://doi.org/10.1007/s00404-019-05267-w

REVIEW

A giant superinfected uterine angioleiomyoma with distant septic metastases: an extremely rare presentation of a benign process and a systematic review of the literature

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Received: 26 February 2019 / Accepted: 6 August 2019 / Published online: 21 August 2019

Abstract

Purpose Uterine angioleiomyoma is a rare type of leiomyoma variant and there are few cases reported in the literature. The definitive diagnosis is usually obtained only after the histopathologic examination because there are no specific imaging criteria for this disease. The objective of this article is to review published cases about this clinical condition.

Methods We report a case of giant angioleiomyoma superinfected by *S. agalactiae* with the development of latero-cervical distant metastasis in a premenopausal woman. Firstly, the case herein reported was orientated as an endometrial stroma sarcoma in the peri-operative histologic examination by frozen sections. It was treated with laparotomic total hysterectomy, bilateral salpingo-oophorectomy, inframesocolic omentectomy and pelvic and paraaortic lymph node dissection. Postoperative definitive anatomopathological analyses using a proper immunohistochemical panel revealed a case of uterine angioleiomyoma. We also review other case reports published about this clinical condition.

Results We present the first case reported in the literature, in our knowledge, of a giant angioleiomyoma superinfected by *S. agalactiae* with the development of distant septic metastases. Immunohistochemistry permitted the definitive diagnosis of angioleiomyoma. Treatments previously reported are hysterectomy or tumor resection and any patient recurred.

Conclusions The definitive diagnosis is usually obtained after the definitive histopathologic examination since the use of immunohistochemical study has an important role in this regard. Complete surgical removal of the lesion is the treatment of choice, with no recurrent cases reported to date.

Keywords Uterine angioleiomyoma · Septic distant metastases · Endometrial stroma sarcoma · Immunohistochemical study

Introduction

Angioleiomyoma (AL) is an exceptionally uncommon subtype of leiomyoma formed by smooth muscle cells and thick walled vascular vessels [1]. The etiology of uterine AL remains unclear. World Health Organization (WHO) does not classify uterine AL as a separate entity or leiomyoma [2, 3]. By literature review, we found that all postoperative diagnosis of AL was up to 0.40% cases of uterine leiomyomas [4].

AL has been described more frequently arising in the subcutaneous tissue of lower extremities. It also can be located in the head and neck, in the submandibular gland and retroperitoneum. AL can be rarely found in the female genital tract [5], with less than 20 cases located in the uterine corpus

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described in reviews published in English medical literature until now [3, 6–18]. Cervix [19–21], ovary [22–24] and the broad ligaments [25–29] are the other uncommon sites of involvement.

There are no specific imaging findings and operative studies usually fail to diagnose this entity [5, 18, 30]. In all the reported cases, a definitive diagnosis was made only after thorough histopathologic examination of the mass and immunohistochemical studies. Three histological types have been described in AL, depending on the vascular channels and the thickness of their muscular walls. Capillary or solid type is characterized by capillary of dense structure with narrow vessels interlaced with thick fascicles of smooth muscles. Venous type is branded of thick vessels interspersed with fascicles of smooth muscles, and cavernous type is typed with widened vessels and lesser amount of smooth muscles.

This paper, to the best of our knowledge, is the first case reported in the literature of an adult woman with a giant uterine angioleiomyoma superinfected by *S. agalactiae* with the development of distant septic metastases. In addition, we perform a systematic review of the literature reports on uterine AL.

Case report

A patient aged 47-years was admitted to our hospital after complaining of progressive abdominal distension and the development of a latero-cervical mass suggestive of abscess and fever up to 38°. The patients' medical history was non contributive, except for a positive vaginal *Streptococcus agalactiae* (*S. agalactiae*) culture performed one year ago for vaginal discomfort.

On exam, she was febrile (37.9 °C) and pain free. Her physical examination showed a marked abdominal distension with an umbilical hernia. Vaginal examination proved normal, with no signs of vaginal bleeding or discharge. Abdominal examination suggested the presence of an abdominal mass of roughly 50 cm in diameter which precluded identification of pelvic structures. At cervical level, an erythematous mass could be identified over the right sternoclavicular union. Transvaginal ultrasound (TVUS) showed a normal uterus measuring 100 × 73 × 89 mm, linear endometrium, and a giant cystic mass measuring 540 × 460 mm, with mucinous content and thick walls. Laboratory results were remarkable for; Hemoglobin (Hb) 7.1 g/dl; 12.25 × 10E leucocytes; 776 × 10E9 platelets, prothrombin time 16.9 s (8–13.7) Partial thromboplastin time of 28 s (25–40.6), ferritin levels of 497 ng /ml (normal levels: 25–250); CA − 19–9 Antigen of 7.6 U/mL; CA − 125 antigen 29.0 U/ml; Serum-Human epididymis protein 4 (HE4 239.7 pmol/L (0–100) ROMA index 77.76%. Hepatitis B and C and HIV serological tests were negative.

A thoracic computerized tomography (CT) scan showed an abcesification at the right sternocleidomastoid muscle measuring 16 × 7 × 16mm and another collection in the right paracervical and anterior hemithorax measuring 31 × 76 × 42mm (Fig. 1a). Multiple pathologic lymph nodes at levels IIa–b, III, IV, VII and supraclavicular ipsilateral to the masses were observed. The abdominal and pelvic CT scan showed a giant mass of apparent endometrial origin and mucinous content measuring 400 × 300 × 350 mm (Fig. 1b). No signs of ascites, carcinomatosis or pelvic dissemination were observed.

Transfusion of two units of red blood cell was prescribed and empirical antibiotic treatment with Amoxicillin-clavulanic was started. Afterwards, latero-cervical abscess was debrided. Microbiological analysis showed *S. agalactiae*

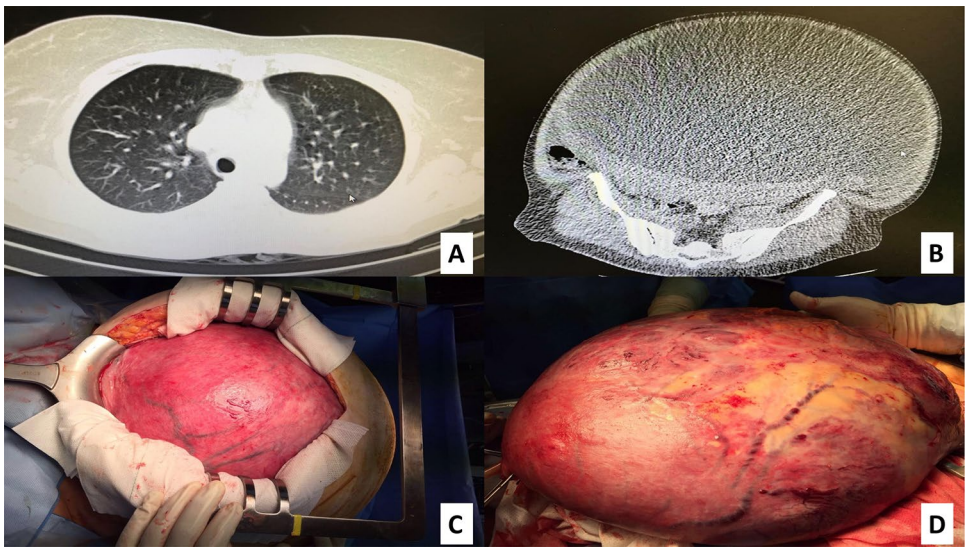

Fig. 1 **a** Abcesification in the right paracervical and anterior hemithorax observed in preoperative CT scan. **b** A giant mass of apparent endometrial origin and mucinous content detected in preoperative CT scan. **c**, **d** Exploratory laparotomy, which confirmed the presence of an uterine giant mass with purulent content

as a causative agent of infection, and the patient continued the treatment with Amoxicillin-clavulanic acid 2 g/8 h for 30 days. Afterwards, an exploratory laparotomy was performed, which confirmed the presence of an uterine giant mass out of which 1600 cc of purulent liquid were extracted (Fig. 1c, d). We also observed enlarged pelvic and paraaortic lymph nodes. No carcinomatosis or distant metastases were observed. Intraoperative histological study by frozen section (FS) revealed a possible Endometrial Stromal Sarcoma. A total hysterectomy, bilateral salpingo-oophorectomy, inframesocolic omentectomy and pelvic and paraaortic lymphadenectomy were performed. Complete cytoreduction was achieved. The patient had an unremarkable postoperative course and was discharged home 5 days after surgery. She prolonged antibiotic treatment with amoxicillin-clavulanic acid for 6 weeks.

Definitive histologic examination of the endocavitary mass showed a moderately cellular spindle cell tumor with an important vascular pattern. No pleomorphism, mitotic figures, or coagulative necrosis were identified. Immunohistochemical staining showed positivity for smooth muscle markers (smooth muscle actin (SMA), Desmin and h-Caldesmin). Estrogen (high intensity, score 240; 80% positive) and Progesterone (medium intensity, score 160, 80% positive) receptors were present. The Ki-67 labelling index was low (< 5%). The endothelial cells stained with vascular marker, CD34 and CD31. CD10 and HMB 45 markers resulted negative. CD10 negativity and histologic features ruled out an endometrial stromal nodule. Similarly, HMB-45 negativity argued against a Perivascular Epithelioid Cell tumor (PEComa). A diagnosis of AL was offered. Pelvic and paraaortic lymph nodes resulted negative. The omentum presented fibrosis and a xantogranulomatous reaction secondary to ischemic necrosis due to local vascular compression. Final anatomopathological analyses confirmed a giant uterine angioleiomyoma superinfected by *S. agalactiae*. Nowadays, the patient remains disease-free on yearly follow-up for three years.

Discussion

Leiomyomas are the most common uterine neoplasms. There are different variants of leiomyomas and AL differs from them in that they are encapsulated and contain numerous vessels. Angioleiomyoma involving the female genital tract is not frequently encountered and only case reports and occasional studies have been previously published. Although its rareness, we have detected that the total number of AL of the uterus has been underestimated in previous reviews, as we found 34 cases in English literature [3, 4, 6–19, 30–36] (Table 1).

Clinical and radiological differentiation between AL and leiomyoma is difficult [5]. AL commonly present as a well-circumscribed mass arising from the uterine corpus. Few cases with multiple uterine AL have also been reported [11, 15]. AL range from 4 to 32 cm in the maximum dimension in previous reports [3, 7–18]. Herein, we present the biggest AL reported in the literature (40 cm). These tumours can be submucosal, intramural and subserosal [3, 7–18].

Uterine AL has been described more frequently in women ranging from 30–69 years of age, although one reported case described an AL in an adolescent woman [2].

Commonly, patients with AL present with abnormal uterine bleeding (AUB), abdominal pain or abdominal mass. It is speculated that AUB in angioleiomyoma is due to dysregulation of vascular growth factors and their receptors, which regulate the process of angiogenesis. Despite its benignity, literature reports present numerous complications posing a serious threat to the health and life of patients with uterine AL. The high vascularity of these lesions and the accompanying menorrhagia can lead to severe anaemia and consumptive coagulopathy [2, 11]. Rarely the mass may suffer a spontaneous rupture with the development of hemoperitoneum [12] and disordered blood clotting due to consumptive coagulopathy. In some cases degeneration in AL simulates an ovarian neoplasm, especially if the AL is located on the posterior wall of the uterus [10, 15], presenting with pseudo–Meigs syndrome and a raised cancer antigen 125 level [16]. In Table 1 we have summarized the clinical features of the patients with uterine AL in the English literature.

Several imaging techniques (ultrasound, CT scan, Magnetic Resonance Imaging) have been performed in the perioperative study of patients with AL. However, AL has no specific imaging findings and is extremely difficult to distinguish it from other masses originated from smooth muscle cells. Currently, the role of imaging techniques is restricted to the evaluation of the type of lesion and the extent of the tumor [30]. Despite this, imaging techniques may suggest preoperative diagnosis of uterine AL in women with UAB, anaemia and signs of vascular damage in imaging scans [11].

Complete surgical removal of the lesion is the treatment of choice. Total hysterectomy with or without bilateral salpingo-oophorectomy is the treatment of choice and was performed in all the reported cases except few cases that patients underwent myomectomy or tumor resection [4, 14, 14, 19, 31] (Table 1). The decision depends on the patient's symptoms and the patient's desire to preserve fertility. Minimally invasive techniques have also been reported as valid methods for removing the lesions without increasing risk of incomplete excision [31, 37]. All patients reported had an unremarkable postoperative course with resolution of symptoms. No recurrence was reported in any case. Thereby, if completely removed, AL have an excellent prognosis.

Table 1 Cases of uterine angioleiomyoma found on the English literature

N	Ref	Year	Age (years)	Clinical features	Preoperative diagnosis	Concomitant diseases	Surgical treatment	Recur
1	[7]	1990	32	AP	Ovarian cyst	Tuberous sclerosis	Tumor excision	No
2	[9]	1999	41	AM	Leiomyoma	–	TH	No
3	[10]	2001	50	AP and AUB	Ovarian cyst	–	TH	No
4	[11]	2003	50	AM and AUB	Leiomyoma	–	TH + BSO	No
5	[12]	2006	45	AP	Ovarian cancer	Hemoperitoneum (ruptured mass)	TH + BSO	No
6	[14]	2007	33	AUB	leiomyoma	–	TH	No
7	[14]	2007	34	AUB	leiomyoma	–	TH	No
8	[14]	2007	49	AUB	leiomyoma	–	Myomectomy	No
9	[13]	2007	55	AUB	leiomyoma	–	TH + BSO	No
10	[15]	2009	69	AM	Ovarian neoplasm	–	TH + BSO	No
11	[16]	2012	47	Pseudo-Meigs syndrome	Ovarian tumor vs uterine sarcoma	–	TH + BSO	No
12	[17]	2012	38	AM	sarcoma	DIC	TH + BSO	No
13	[37]	2013	41	AUB	Pelvic mass (non filiated origin)	–	Tumor excision	–
14	[34]	2014	55	AUB	Sarcoma	–	TH + BSO + omentectomy + pelvic LND	No
15	[2]	2014	19	AM + AUB	Leiomyoma	–	sTH	No
16	[3]	2014	49	AUB	Ovarian tumor		TH + BSO	No
17	[18]	2015	39	AP and AUB	Leiomyoma	–	TH	No
18	[6]	2015	50	AP	Leiomyoma	–	TH + BSO	No
19	[4]	2016	48	AUB	Leiomyoma submucosa	–	Hysteroscopic myomectomy	No
20	[4]	2016	44	AUB	Leiomyoma	–	Myomectomy	No
21	[4]	2016	60	AUB	Leiomyoma	Endometriosis	TH + BSO	No
22	[4]	2016	43	AUB	Leiomyoma	–	TH and BS	No
23	[4]	2016	48	AUB	Leiomyoma	Endometriosis	TH + BSO	No
24	[4]	2016	47	AUB	Leiomyoma	–	TH	No
25	[4]	2016	46	AUB	Leiomyoma submucosa	Chronic endometritis	Hysteroscopic myomectomy	No
26	[4]	2016	43	AUB	Leiomyoma	–	TH + BSO	No
27	[4]	2016	45	AUB	leiomyoma	Endometriosis	TH + BSO	No
28	[32]	2017	33	AM, AUB and severe anemia	Leiomyoma	–	Myomectomy	No
29	[19]	2018	32	AUB	Leiomyoma	–	Myomectomy	No
30	[19]	2018	46	AUB	Leiomyoma	–	Myometomy	No
31	[19]	2018	37	AUB	Leiomyoma	–	Hysterectomy	No
32	[19]	2018	44	AUB	Leiomyoma	–	Myomectomy	No
33	[30]	2018	37	AUB + AP	Ovarian tumor vs uterine sarcoma	–	TH	–
34	[31]	2018	39	AP	Leiomyoma	–	Myomectomy	No
35	Our case	2019	47	Fever + AM	Endometrial stromal sarcoma	–	TH + BSO + omentectomy + pelvic and paraaortic LND	No

Ref bibliographic reference, *y* years, *recur* recurrence, *AM* abdominal mass, *AP* abdominal pain, *AUB* abnormal uterine bleeding, *vs* versus, *DIC* disseminated intravascular coagulation, *TH* total hysterectomy, *sTH* subtotal hysterectomy, *BSO* bilateral salpingoophorectomy, *BS* bilateral salpingectomy

However, the role of myomectomy or tumor resection alone with the preservation of the uterus has not been studied widely and will need further investigation.

Macroscopically, AL shows tan-white areas interspersed with hemorrhagic areas or may exhibit a marbled appearance with pink-brown and grey areas [3, 7–18]. Some AL are multiloculated and contain blood-filled cysts. Histologically, AL is composed of interlacing fascicles of monotonous spindled smooth muscle cells spinning around the abundant thick-walled blood vessels. In contrast, usual leiomyomas present the density of the vascular network similar to or less than the normal myometrium [38] and are composed of capillaries along with a few arterioles and small arteries. Most of AL rarely show pleomorphism, necrosis or mitotic figures. Only one case with mitosis up to 2 mitoses per 10 high power fields has been reported [14]. If extensive nuclear atypia is observed, extensive sampling including the border of the tumor should be performed to discard increased or atypical mitosis and necrosis to exclude a leiomyosarcoma. Other changes described in this tumor are myxoid change, edema, hyalinization of the stroma and fibrin deposition in the vessel wall [3, 7–18]. Even though, only two cases of uterine AL reported fibrin thrombi within the vessels, which is frequently observed in AL of the skin [17]. In addition, AL may show degenerative changes like large cavernous deformation of the vascular spaces.

The microscopic differential diagnoses include endometrial stromal nodule, fibroma, angiofibroma, angiolipoma, angiomyofibroblastoma and PEComa. FS performed in intraoperative anatomopathological study has limitations that should be noted as limited quantity of tissue analysed, the presence of bloated cell morphology and the presence of freezing artifacts that may difficult the correct diagnosis of AL. Although each one of these entities has its typical histologic characteristics, overlapping histopathological features may exist and the immunohistochemical staining may be required to establish the correct diagnosis (Table 2). AL show diffuse and strong positivity for SMA, desmin, h-caldesmon and progesterone receptors and a low Ki-67 labeling index. Weak positivity for CD10 and oestrogen receptor is observed, while endothelial cells express positivity for CD31 and CD34. It is a differentiating element from other neoplasms like fibroma, angiofibroma, angiolipoma and angiomyofibroblastoma. Angiomyofibroblastomas are vimentin- and desmin-positive but smooth muscle actin-(SMA) negative [12]. PEComas are positive for HMB-45 and MART-1 [4, 19].

Genetic bases of the AL cell evolution are not well known yet. Cytogenetic analysis performed in a 41-year-old woman with uterine AL revealed the following karyotypic abnormality: 46,X,t(X;11)(p11.4;p15)/46, idem,inv(2)(p15q13)/46, idem,inv(2)(p15q13),t(5;20)(q13;q13.2) [9]. There is no more information in the current literature.

The case here reported presents singular characteristics. To the best of our knowledge this is the biggest AL described in the literature and is the first case reported of an adult woman with an AL superinfected by *S. agalactiae* with the development of distant septic metastases. The invasion of the myometrium by blood vessels could explain the passage of *S. agalactiae* into the bloodstream deriving from vaginal colonization, and the ensuing infection of the latero-cervical tissues. Moreover, an endometrial stromal sarcoma was diagnosed in the intraoperative anatomopathological study which implied an overtreatment in our patient. Therefore, it is important for clinicians and pathologists to recognize this rare benign entity and differentiate it from malignant neoplasms when the tumor shows significant cytologic atypia or raised cancer antigen 125 levels by thorough sampling.

In conclusion, AL is an extremely rare benign tumor that arises from smooth muscle cells and presents abundant thick-walled vessels in contrast with usual leiomyomas. Preoperative diagnosis of AL is extremely hard, as there are no specific clinical or imaging findings of this entity. Hence, it is important for the clinician and pathologist to recognize this uncommon benign tumor and differentiate it from its mimickers by histopathologic analysis and, when required, using a proper immunohistochemistry panel. Due to its rareness and the limitations

Table 2 Immunohistochemical differential diagnostics among Angioleiomyoma and its morphologic Mimikers (Endometrial Stromal Nodule, Angiomyofibroblastoma, and Perivascular Epithelioid Cell Tumor)

	SMA	Desmin	h-Caldesm	Calponin	CD31 CD34	CD10	WT1	HMB45	MART-1	Vimentin	MITF
AL	+	+	+	+	+	– / f. w +	–	–	–	+	–
EST	– / f. w-m +	– / f. w +	–	–	–	+	+	–	–	+	–
AMFB	–	+	–	–	–	–	–	–	–	+	–
PEComa	+	+	+	–	–	–	–	+	+	+	+

AL angioleiomyoma, *EST* endometrial stromal tumor, *AMFB* angiomiofibroblastoma, *PEComa* Perivascular Epithelioid Cell neoplasm, *SMA* Smooth muscle actine, *h-Caldesm* h-Caldesmon, *MITF* Miroftalmia transcriptor factor, *f.* focal, *w.* weak, *m.* moderate, + positive, – negative

of FS, gynecologic oncologist should submit the patient to cytoreductive surgery if malignancy is firmly suspected, because a delay in treatment may entail a negative impact on our patients' survival.

Author contributions JLS-I: Project development, Data Collection, Manuscript writing. SC: Project development, Data Collection, Manuscript writing, Manuscript editing. MC-A: Manuscript editing. MC-S: Data Collection. S Cabrera: Data Collection. LI-H: Data Collection, Manuscript writing. MAP-B: Manuscript editing. SM-C: Data collection. AG-M: Final Manuscript editing.

Compliance with ethical standards

Conflict of interest There are no conflicts of interests for this manuscript.

Informed consent The authors thank the patient for giving permission for publishing this case.

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