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A Giant Pure Uterine Lipoma: A Case Report

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Authors' contributions

This work was carried out in collaboration between all authors. Author IM managed the acquisition of the data. Author MAB performed the drafting and the analysis of the manuscript. Author FG wrote the first draft. Author SBR managed the conception and design. Author NM performed the histological and immunohistochemistry analysis. Author AB did critical revision of the manuscript. Author BL read and approved the final manuscript.

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Case Study

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ABSTRACT

Aims: The authors present a case of a giant pure lipoma of the uterus revealed by a postmenoposal metrorrhagia and discuss the clinical, radiological, histological characteristics and the different histogenesis hypothesis of this entity.
Case Presentation: We report the case of a 73-year-old woman who presented with a pure uterine lipoma in which a preoperative diagnosis of ovarian teratoma was suggested by computed tomography.
Conclusion: Pure uterine lipoma is extremely rare and only a few cases have been reported in the literature. It usually develops in postmenoposal women. Clinical symptoms and physical signs are similar to those found in leiomyomas. Nevertheless, preoperative diagnosis is difficult and requires pathological confirmation.

Keywords: Pure lipoma; uterine tumor; histogenesis pathology.

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1. INTRODUCTION

The fatty tumors of the uterus are rare, representing approximately 0.03 to 0.12% of hysterectomy. These tumors have been described for the first time by Lobstein in 1816 including a spectrum of histological lesions ranging from lipomatosis in leiomyomas to pure lipomas [1]. Mixed forms (lipoleiomyoma and fibrolipoma) are far the most common. Pure uterine lipoma is extraordinarily rare and few cases have been reported in the literature [2]. The clinical manifestations do not usually differ greatly to those caused by leiomyomas except that they affect women that are somewhat older and normally postmenopausal [1]. Most lipomas are located in the body of the uterine corpus and the size can range from a few millimetres up to several centimeters. Diagnosis is made after a meticulous analysis of the surgical specimen, although some radiological techniques may indicate their existence prior to surgery [2]. Their prognosis is excellent, except in cases associated to malignant intracavitary pathology. Through this observation, we propose to discuss the clinical, radiological and histological features of this exceptional entity.

2. CASE REPORT

A 73 year-old postmenoposal woman, with no particular past medical history, who presented with a sensation of pelvic heaviness recently associated with low abundance bleeding.

Physical examination revealed an abdominal firm, mobile and painless mass. Gynecological examination revealed no abnormalities of the vulva, the cervix and the vagina.

Ultrasound showed a well circumscribed mass measuring 20 x 14 cm, echoic in the center and surrounded by a hypoechoic rim (Fig. 1). Computed tomography revealed a large, well limited, abdomino-pelvic mass, with central and peripheral tissue fatty component, high density areas were observed in this mass; those radiological aspects were evoking an ovarian teratoma (Fig. 2).

Total hysterectomy with bilateral salpingo-oophorectomy was performed. The uterus was increased in size. Both ovaries were in place and without defect. At gross examination, the hysterectomy specimen measured 26 × 24 × 12 cm, with globular enlargement of the corpus. The cut surface showed a well-circumscribed, homogenous yellow mass in the intramural region measuring 20 cm in its greater diameter. No areas of necrosis or hemorrhages were seen.

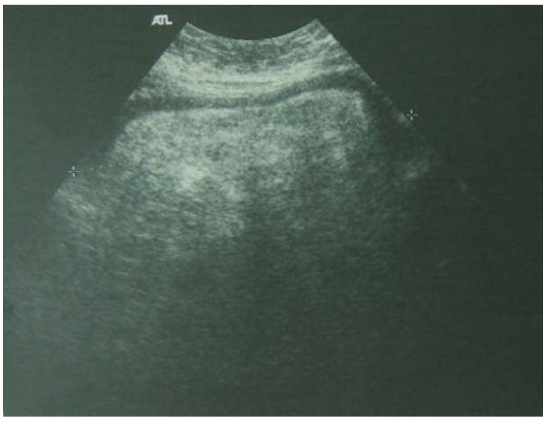

Fig. 1. Ultrasound of the pelvis showed a well-circumscribed, echoic mass with surrounding hypoechoic rim

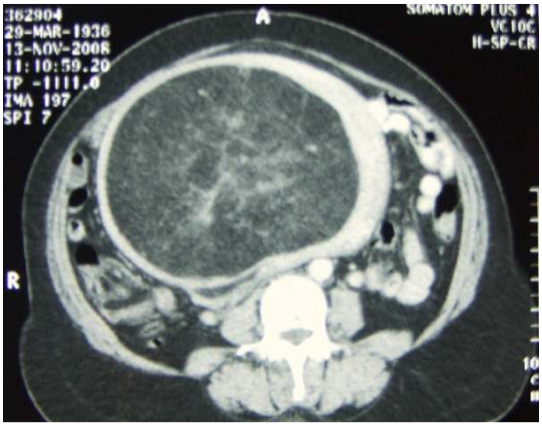

Fig. 2. Computed tomography revealed a regular mass with a heterogenous density of fat and a periphery tissular density

The histopathological examination showed a thin atrophic endometrium. The intramural tumor was composed of only mature adipocytes, separated by thin connective vascular septa (Fig. 3). There were no cytonuclear atypia or mitosis, No smooth muscle cells or fibrous elements or lipoblasts were seen within the tumor (Fig. 4). Adipocytes were positive to smooth muscle actin (Fig. 5).

The diagnosis of primary pure uterine lipoma was made and the postoperative course was favorable. Up to now, the patient is doing well at the 6 years follow-up.

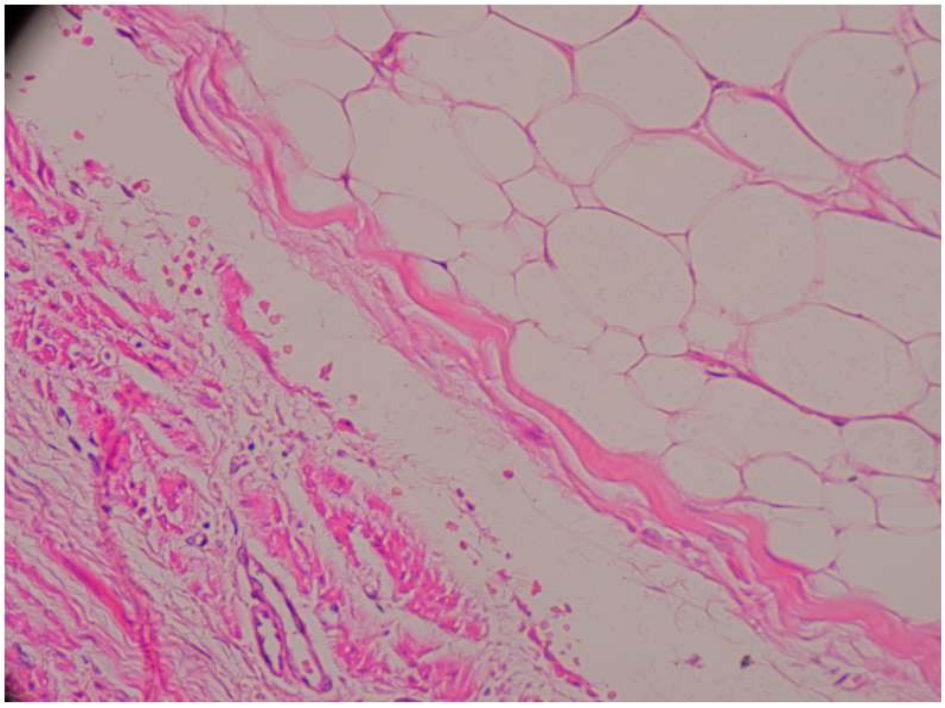

Fig. 3. A benign tumour composed of mature adipocytes separated by thin fibrous septa and peripherally delimited by smooth muscle cells. (Hematoxylin and Eosin, original magnification 400X)

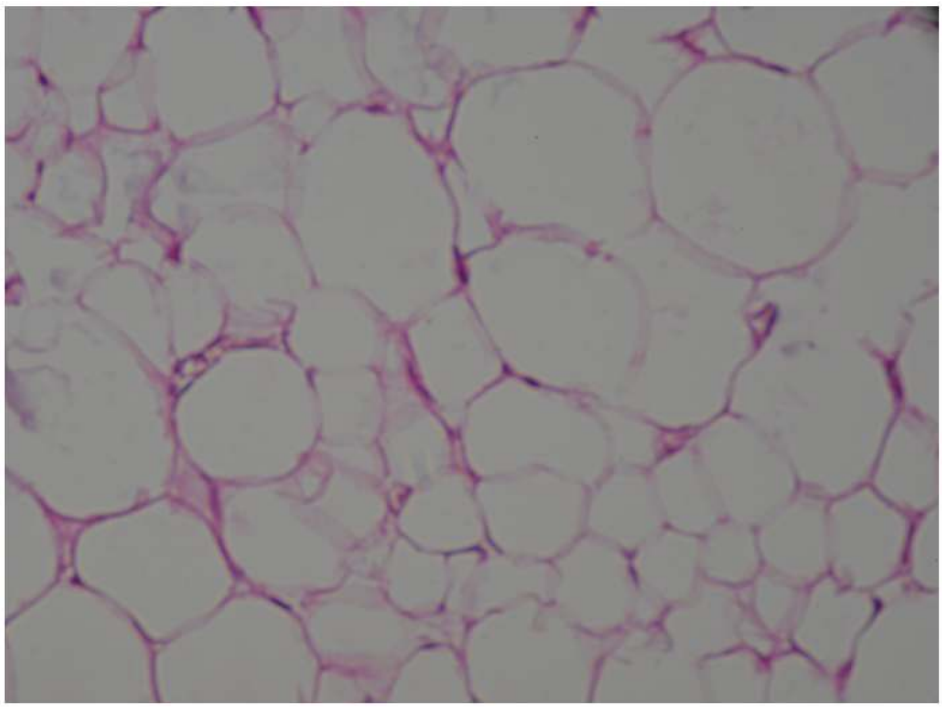

Fig. 4. The adipose mass was composed of large mature adipocytes (Hematoxylin and Eosin, original magnification 400X)

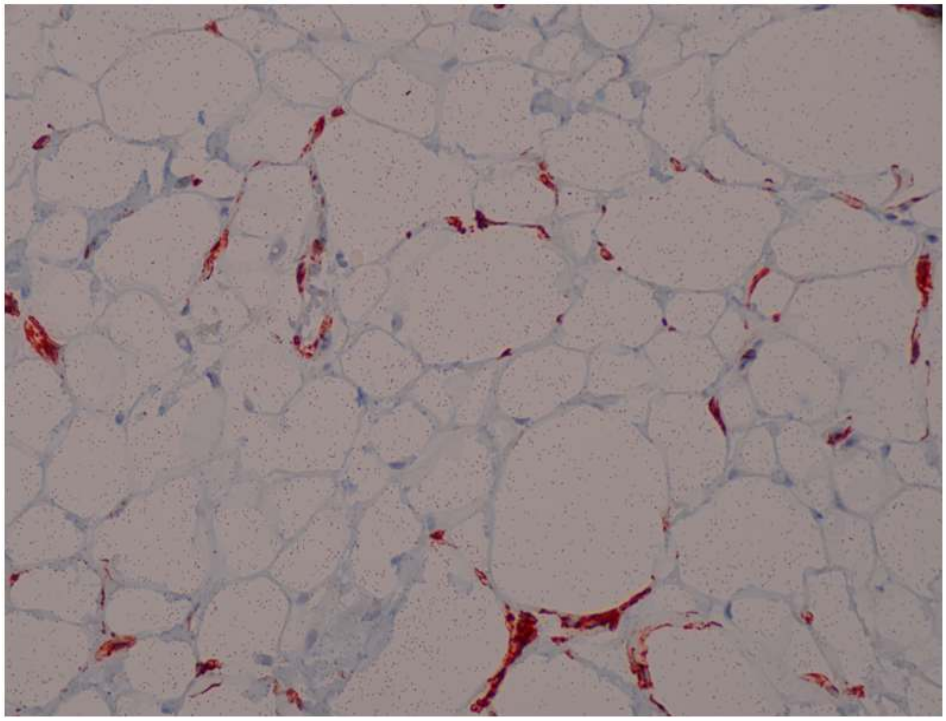

Fig. 5. AML staining at 400 magnification showed strong cytoplasmic staining

3. DISCUSSION

Lipomatous tumors of the uterus are rare benign tumors. Depending on the presence of smooth muscle cells and fibrous tissue, tumors were classified into pure forms and mixed forms. Mixed structures combine in variable proportions smooth muscle cells, fibroblasts and mature adipocytes hence the name lipoleiomyoma and fibrolipoma. They are far the most common [2]. Pure uterine lipoma is exceptional with rare cases reported in the literature.

They usually occur in asymptomatic, obese perimenopausal or postmenopausal women from 50 to 70 years old and may be associated to leiomyomas whose clinical history could not be distinguished [2]. In our case the patient was symptomatic, she presented with a sensation of pelvic heaviness associated with low abundance bleeding.

A preoperative diagnosis may be possible. The ultrasound shows a well limited, hyperechoic mass, surrounded by a hypoechoic rim. The peripheral hypoechoic correspond to the layer of smooth muscle fibers organizing around the fat. However, the sonographic appearance is nonspecific and can be seen in a fatty degenerative changes of a leiomyoma or in dermoid cyst of the ovary [3,4].

Computed tomography scans, on the other hand is more specific. It shows a very limited fat density mass without being able to determine its exact location especially for large tumors. These are mistaken for adnexal or intra-abdominal tumors as in our case the tumor was diagnosed as a teratoma [3-5]. High density areas might be seen in the tumor, as in our case, as they correspond histologically to the fibro-vascular septa between the adipocytic lobules.

Magnetic resonance imaging is the best imaging modality; it confirms the lipomatous nature of the mass and its exact location and distinguishes between mixed and pure types. However, Lipoma signal is characteristic with high signal intensity on T1-weighted and T2 and a complete loss of signal on saturated fat sequences. It also shows the presence of a peripheral hypointense signal corresponding to the fibromuscular pseudocapsule [6].

Although the preoperative diagnosis is possible, histopathological examination is mandatory to rule out malignancy [6].

The pathological diagnosis is usually easy, suggested at gross examination by the existence of a well circumscribed homogeneous tumor, round or oval in shape with a yellowish color.

It is usually intramural, more rarely serous or intracervical [1-7]. Lipoma often measures between 5 and 10 cm. However, the size may vary from few millimeters to 32 cm [6]. The tumor described in this observation is giant measuring 20 cm in diameter.

Histological examination shows exclusively mature adipocytes, without atypia or mitosis, separated by thin fibro-vascular septa and peripherally delimited by smooth muscle cells and clearly demarcated from its surrounding tissue. On immunohistochemistry, they are positive for Vimentin and S-100 protein. Positivity for smooth muscle markers is also found like in our case [8]. Although it occurs in postmenopausal women, a nuclear positivity for estrogen and progesterone receptors were noted [2,9].

The differential diagnosis of a pure lipoma arises mainly with mixed forms of lipomatous tumors (lipoleiomyoma and fibrolipoma).

Currently, there are no established strict criteria to differentiate pure lipoma from mixed lipomatous tumors [2]. However, many authors consider that the presence of mature adipocytes lines with a peripheral layer of smooth muscle cells is the key to diagnosis [6-9]. However, uterine lipomalike liposarcoma is another differential diagnosis that have been reported in 9 which is characterized by the presence of lipoblasts, cells with bizarre nuclei, mitosis and areas of necrosis [2]. In our case we carefully analysed the tumour histopathologically, observing the absence of lipoblasts. Moreover, nuclear atypia, cellular pleomorphism and mitotic figures were not detected. Therefore, we were able to exclude the diagnosis of a lipoma-like liposarcoma [10].

As fat cells are natively absent from the myometrium, various theories of histogenesis have been proposed and it currently remains unclear and debated even with the support of immunohistochemistry and molecular analysis. Lipoblastic differentiation of misplaced embryonal mesodermal rests, as well as lipoblast or pluripotential cell migration along the uterine nerve and vessels, are supposed mechanisms [11]. Recently, some authors suggested that fatty metaplasia of the connective tissue or the smooth muscle cells seems to be the most plausible histogenetic cause involved in the development of uterine lipomas since adipocytes may express AML in immunohistochemistry as in our case [6]. Smooth muscle cells in the surrounding tissue were reactive to actin, desmin, and vimentin. Estrogen receptor, and progesterone receptor were present; focal actin and desmin were found in granular or filament form in the cytoplasm of the fat cells [12].

In recent years, characteristic chromosomal abnormalities have been found in adipose tumors. Lipomas are frequently characterized by aberrations of the 12q13 approximately q15 chromosomal region and often by rearrangements of the HMGA2 gene. These rearrangements include the formation of chimeric genes that fuse the 50 region of HMGA2 with a variety of partners, such as LPP (3q28) or NFIB (9p22) [6].

Uterine lipomas have an excellent prognosis and can be considered for the differential diagnosis of uterine mass in postmenopausal women, surgical excision is the best treatment [4].

4. CONCLUSION

Pure uterine lipoma is extremely rare and only a few cases have been reported in the literature. It usually develops in postmenoposal women. Clinical symptoms and physical signs are similar to those found in leiomyomas. Nevertheless, preoperative diagnosis is difficult and requires pathological confirmation. This report provides an update of the most current findings and adds to the available data concerning this tumor.

CONSENT

All authors declare that written informed consent was obtained from the patient (or other approved parties) for publication of this paper and accompanying images.

ETHICAL APPROVAL

All authors hereby declare that all experiments have been examined and approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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