A Case of Giant Mesenchymal Uterine Tumor: Lipoleiomyoma

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Patient: Female, 66-year-old
Final Diagnosis: Lipoleiomyoma
Symptoms: Abdominal pain • pollakiuria and abnormal vaginal bleeding
Medication: —
Clinical Procedure: —
Specialty: Obstetrics and Gynecology

Objective: Rare disease
Background: Uterine lipoleiomyoma is a rare benign tumor composed of varying proportions of smooth muscle fibers and mature adipocytes, without identification of hemorrhage, necrosis, or cyto-architectural atypia. It is part of the leiomyomas category, with an incidence of 0.03-0.2%. The pathogenesis of this lesional category is still unclear, but there are several theories that could explain the occurrence. Magnetic resonance imaging is the most useful diagnostic imaging method. There are a number of pathologies whose exclusion is necessary, with the differential diagnosis being made mainly based on microscopic examination and completed with immunohistochemical tests. Their treatment, when necessary, is surgical, with an excellent post-therapeutic evolution and prognosis.

Case Report: We present the case of an elderly, postmenopausal patient who presented with abdominal pain, abnormal vaginal bleeding, and pollakiuria. The associated pathologies of the patient correspond to those mentioned in the literature, the particularities of the case being given by the large size of the tumor and the association with 2 other typical leiomyomas. Immunohistochemical markers used to exclude other diagnoses (desmin, h-caldesmon, S100, calretinin, MDM2, CD34) confirmed the diagnosis of uterine lipoleiomyoma. Because the patient was symptomatic and a large nodular mass was identified by ultrasound, surgical treatment was performed.

Conclusions: Although it is a benign lesion with an excellent prognosis, the pathogenetic mechanisms are not fully known. Theories of pathogenesis range from misplacing embryonic adipocytes to connective tissue fatty degeneration, and further studies are needed to establish the origin of this lesion.

Keywords: Adipocytes • Myocytes, Smooth Muscle • Postmenopause • Uterus

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Background

Primary uterine lipoid tumors are rare entities, the spectrum of which includes lipoma, lipoleiomyoma, and liposarcoma [1]. Lipoleiomyoma is a rare, benign tumor, made up of a mixture of mature adipocytes and smooth muscle fibers [2]. Although it usually occurs in the retroperitoneum and in the abdominal cavity, in the uterus it occurs most frequently intramurally, on the posterior face [3,4]. Imaging examinations have an important role in identifying the fat component and establishing the topography of the lesion, magnetic resonance imaging being the best diagnostic method [1,2]. From a therapeutic point of view, their treatment depends on the symptoms and the size of the tumor, with the asymptomatic ones not needing surgical treatment [5,6].

Case Report

A woman with previously diagnosed high blood pressure, grade 2 obesity, hypothyroidism, and hyperlipidemia presented to the hospital for abdominal pain, pollakiuria, and abnormal vaginal bleeding. The ultrasound examination showed an enlarged uterus, with the presence of a hyperechoic nodular lesion, with a maximum diameter of 20 cm, for which a total hysterectomy with bilateral anexectomy was performed.

The macroscopic examination of the uterus highlighted the presence of 3 intramural nodular lesions with a diameter of 4-18 cm, the largest being located at the level of the posterior face, on section having a whitish-yellow, lobed aspect, with areas of bleeding or necrosis.

Microscopic examination of the nodular lesion with maximum diameter revealed a proliferation of mature fat cells with lobular disposition, separated by thin bands of connective tissue and focally interspersed with smooth muscle fibers, the appearance being compatible with the diagnosis of lipoleiomyoma (Figure 1). The other 2 nodular lesions were typical leiomyomas with hyaline degeneration.

For lipoleiomyoma, we assessed the following immunohistochemical markers: desmin, h-caldesmon, S100, calretinin, MDM2, and CD34. We found a diffuse, intensely positive, cytoplasmic reaction of desmin and h-caldesmon in smooth muscle fibers, a diffuse, moderately positive reaction for S100 in the membrane of fat cells a diffuse, an intense positive reaction of calretinin in the nuclei of adipose cells, and a negative reaction of CD34 and MDM2 (Figure 2). The immunohistochemical panel confirms the initial microscopic diagnosis.

Discussions

Primary uterine lipomatous tumors can be classified into 3 groups: the first is pure lipomas; the second has an added mesodermal component with lipoleiomyoma, angioleiomyoma, and fibromyolipoma; and the last category is liposarcoma [5,6].

Lopstein first described this type of tumor in 1916 [7]. Uterine lipoleiomyoma is a rare tumor, the incidence of which varies from 0.03% to 0.2% [5]. It most commonly occurs in peri-or postmenopausal obese women, most being asymptomatic [5,7,8]. It can be located both in the uterine body and in the cervix, but cases with retroperitoneal, ovarian, and broad ligamentary location have been reported [4,8]. The symptoms are nonspecific, similar to that of typical leiomyomas, including abdominal pain, discomfort, vaginal bleeding, pollakiuria, incontinence, and a feeling of pressure [2,5,7]. It usually presents as a single tumor mass, with a diameter ranging from 0.5 cm to 55 cm, with an average of 5.5 cm [7]. In the present case, the nodular lesion was located at the level of the uterine posterior wall, having a maximum diameter of 18 cm.

Regarding pathogenetic mechanisms, there are several theories that could explain the presence of adipocytes in association with smooth muscle fibers [7]. These theories include misplacement of embryonic adipocytes, muscle cell metaplasia, differentiation of connective tissue cells into adipocytes, lipocytic differentiation of specific primitive cells in connective tissue, intruterine perivascular adipose cells, fatty infiltration, and connective tissue degeneration [7]. In addition, some metabolic disorders associated with lipoleiomyoma are known, including diabetes, hyperlipidemia, and hypothyroidism [9]. There are also studies that support the role of hyperestrogenism in their development [10,11]. In our case, the patient...
Figure 2. Immunohistochemical features of lipoleiomyoma. (A) Diffuse, moderately positive, cytoplasmic reaction of h-caldesmon in smooth muscle fibers (IHC stain, Ob ×200). (B) Diffuse, moderately positive, cytoplasmic reaction of desmin in smooth muscle fibers (IHC stain, Ob ×200). (C) Diffuse, moderately positive reaction for S100 in the membrane of fat cells (IHC stain, Ob ×200). (D) Diffuse, moderately positive reaction for calretinin in the nuclei of fat cells (IHC stain, Ob ×200). (E) Negative reaction for CD34 in the tumor (IHC stain, Ob ×200). (F) Negative reaction for MDM2 in the lipoleiomyoma (IHC stain, Ob ×200).
had grade 2 obesity, hypothyroidism, and hyperlipidemia, consistent with the clinical picture mentioned in the literature.

Ultrasound imaging, computed tomography, and magnetic resonance imaging can be used to determine the topography of the lesion [1]. Imaging methods suggest the diagnosis by highlighting the adipose component [4]. For differential diagnosis, a histopathological examination is necessary, as imaging alone cannot establish a definite diagnosis [7]. Magnetic resonance imaging is the most useful method of diagnosis, highlighting signals of increased intensity in both T1- and T2-weighted images characteristic of adipose tissue, which can also be confirmed by fat-suppression sequences [1]. In addition, it allows the visualization of the septa inside the tumor mass and of a peripheral frame with a low signal intensity that corresponds to the pseudocapsule, thus being a superior method to computed tomography and ultrasound [12].

The main pathological entities with which the differential diagnosis is made are from the gynecological sphere: mature cystic ovarian teratoma, well-differentiated liposarcoma, atypical lipoma, pelvic fibromatosis, carcinosarcoma with heterologous liposarcomatous differentiation, and leiomyoma with adipose degeneration, in which, unlike lipoleiomyoma, fat is evenly distributed as an integral part of the tumor [1,2,5,8,13].

The mature cystic ovarian teratoma is the most common germ cell tumor, being characterized by the presence of at least 2 types of mature, well-differentiated germ cells [14]. The most frequent types of germ cells are the ectodermal and mesodermal ones, the microscopic expression being represented by the skin, hair, adipose tissue, and muscle fibers [14].

Regarding atypical lipomatous tumor/well-differentiated liposarcoma, immunohistochemical expression is characterized by positive immunoreexpression of MDM2 and S-100 [15].

Fibromatosis is a benign proliferation of fibroblasts, but with an aggressive, infiltrative growth pattern [16].

Carcinosarcoma is a malignant mixed Müllerian tumor, defined by the presence of high-grade malignant epithelial and mesenchymal components [17]. The mesenchymal component may be homologous or heterologous, the latter also including liposarcoma [17].

In our case, the tumor was located in the uterine body, was well defined, and consisted of mature adipocytes and smooth muscle fibers. Microscopic features associated with the immunohistochemical profile confirmed the diagnosis of uterine lipoleiomyoma, excluding other possible neoplasms.

Coexistence with other gynecological diseases has been reported in the literature, such as adenomyosis, endometriosis, endometrial polyp, and endometrial hyperplasia [1,18]. In the study conducted by Akbulut et al, which included 70 patients, 17.1% had associated malignancies in the endometrium, ovary, and mammary gland [7]. Also, Aung et al reported the coexistence of gynecological cancers in 11% of cases, and in the study of Wang et al, 10 cases were identified that combined gynecological malignant neoplasms with lipoleiomyoma [5,18]. In the present case, the association with 2 other leiomyomas with hyaline degeneration was observed. The particularity of the case consists in the important dimensions of the described neoplasm, which was significantly larger than the average diameter reported in the literature.

Therapeutically, asymptomatic lipoleiomyoma does not require surgical treatment, and in symptomatic cases, the options for surgical treatment are myomectomy, hysterectomy, myolysis, tumor embolization, and radiofrequency ablation [5,6].

Conclusions

We can conclude that uterine lipoleiomyoma is a rare, benign tumor, which occurs most frequently in obese, peri- or postmenopausal patients, and may have symptoms similar to typical leiomyoma. Theories of the pathogenesis range from misplacing embryonic adipocytes to connective tissue fatty degeneration, and further studies are needed to establish the origin of this lesion. Magnetic resonance imaging is the most useful diagnostic imaging method. As a benign tumor, the prognosis is excellent, with patient follow-up being necessary in case of association with malignant tumors.

Declaration of Figures’ Authenticity

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