

CASE REPORT

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Uterine lipoleiomyoma Lipoleiomioma uterino

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ABSTRACT

Lipoleiomyoma is a rare benign uterine neoplasm whose incidence varies between 0.03%-0.2%. This tumor is considered a benign variant of typical uterine leiomyomas. It consists of a variable proportion of mature adipocytes and smooth muscle cells. The etiology may be related to estrogen deficiency occurring after the menopausal transition; it usually appears in obese perimenopausal or menopausal women. The symptomatology is nonspecific, and most are diagnosed incidentally. We present a case of uterine lipoleiomyoma in a 45-year-old patient who consulted for abdominal pain. Ultrasonography showed a tumor in the anterior wall of a homogeneous and well-defined uterus. During laparotomy, a yellowish tumor with a soft texture was found. Total hysterectomy plus oophorosalingectomy was performed. The anatomopathologic diagnosis was uterine lipoleiomyoma.

Key words: Lipoleiomyoma, Uterine tumor, Adipocytes, Smooth muscle tumor

RESUMEN

El lipoleiomioma es una neoplasia uterina benigna poco frecuente cuya incidencia varía entre 0,03% y 0,2%. Este tumor es considerado una variante benigna de los leiomiomas uterinos típicos. Está formado por una proporción variable de adipocitos maduros y células musculares lisas. La etiología puede estar relacionada con la deficiencia de estrógenos que se produce después de la transición menopáusica; generalmente aparece en mujeres obesas perimenopáusicas o menopáusicas. La sintomatología es inespecífica y la mayoría es diagnosticada de forma incidental. Se presenta un caso de lipoleiomioma uterino en paciente de 45 años quien consultó por presentar dolor abdominal. La ecografía mostró tumor en pared anterior de un útero homogéneo y bien definido. Durante la laparotomía se encontró tumor amarillento y de textura blanda. Se realizó histerectomía total más ooforosalingectomía. El diagnóstico anatomopatológico fue de lipoleiomioma uterino.

Palabras clave. Lipoleiomioma, Tumor uterino, Adipocitos, Tumor de músculo liso

INTRODUCTION

Primary uterine lipomatous tumors are infrequent and generally benign. Histologically, these tumors include pure lipomas, lipoleiomyomas, and fibrolipomyomas^(1,2). Uterine lipoleiomyoma, a variant of leiomyoma, has an incidence of approximately 0.03%-0.2% of all uterine tumors⁽³⁾. It is composed of variable amounts of mature adipocytes and smooth muscle cells and its most common location is the uterine corpus, as intramural growths. It has also been described in the cervix, ovaries and retroperitoneum and most cases have been reported in menopausal women⁽⁴⁾. They are difficult to diagnose clinically and can be confused with mature ovarian teratoma, well-differentiated liposarcoma and atypical lipoma^(2,3). Symptomatology is similar to atypical leiomyomas, and they do not require treatment if they are asymptomatic^(1,4). A case of uterine lipoleiomyoma is presented.

CASE REPORT

A 45-year-old woman, nulligesta, consulted for presenting hypogastric pain of 15 days of evolution accompanied by increased abdominal volume and menometrorrhagia for approximately 6 months. The patient reported menarche at 15 years of age with menstrual cycles of 5 to 6 days and an interval of 26 days. She denied medical and surgical history of importance.

Physical examination revealed an enlarged uterus with a hard, mobile, and non-painful tumor that reached two centimeters below the umbilical scar, compatible with an 18-week pregnancy. There were no palpable adenopathies or visceromegaly. The gynecological examination revealed no abnormalities of the vulva, vagina, and cervix. The rest of the physical examination was within normal limits.

Ultrasound showed an enlarged uterus with a 10-millimeter endometrium, which contained a homogeneous, hyperechoic, well-defined tumor measuring 26 x 22 x 15 centimeters on its anterior wall, suggesting the possibility of uterine leiomyoma. Both adnexa had a normal appearance. Doppler evaluation revealed low vascularization of the lesion (Figure 1). Computed tomography images were like those on ultrasound with predominantly fatty component within the lesion. The tumor displaced the urinary bladder, rectum, and other abdominopelvic structures. Routine test results, including tumor markers, were within normal limits. In view of the findings, the patient was scheduled for surgery.

During laparotomy, a 20 x 18 x 8 centimeter yellowish, soft-textured tumor was observed protruding into the abdominal cavity. Total hysterectomy with bilateral oophorosalingectomy was performed. There was no evidence of lymphadenopathy, and the pelvic organs were

macroscopically normal. Transoperative blood loss was minimal, and the surgery was completed without complications.

On macroscopic examination, the uterus showed a smooth, subserosal, homogeneous, grayish-white tumor with internal yellow areas on cross-section (Figure 2). Both ovaries and fallopian tubes were normal. Histological study of the specimen showed a large cellular proliferation of mature adipose predominance without cellular atypia, together with smooth muscle cells and large intercellular edema (Figure 3). The adipo-

FIGURE 2. MACROSCOPIC IMAGE OF UTERINE LIPOLEIOMYOMA.



FIGURE 1. ULTRASOUND IMAGES OF UTERINE LIPOLEIOMYOMA. A) HOMOGENEOUS HYPERECHOGENIC AND WELL-DEFINED TUMOR. B) DOPPLER ULTRASOUND SHOWING LOW VASCULAR FLOW WITHIN THE TUMOR.

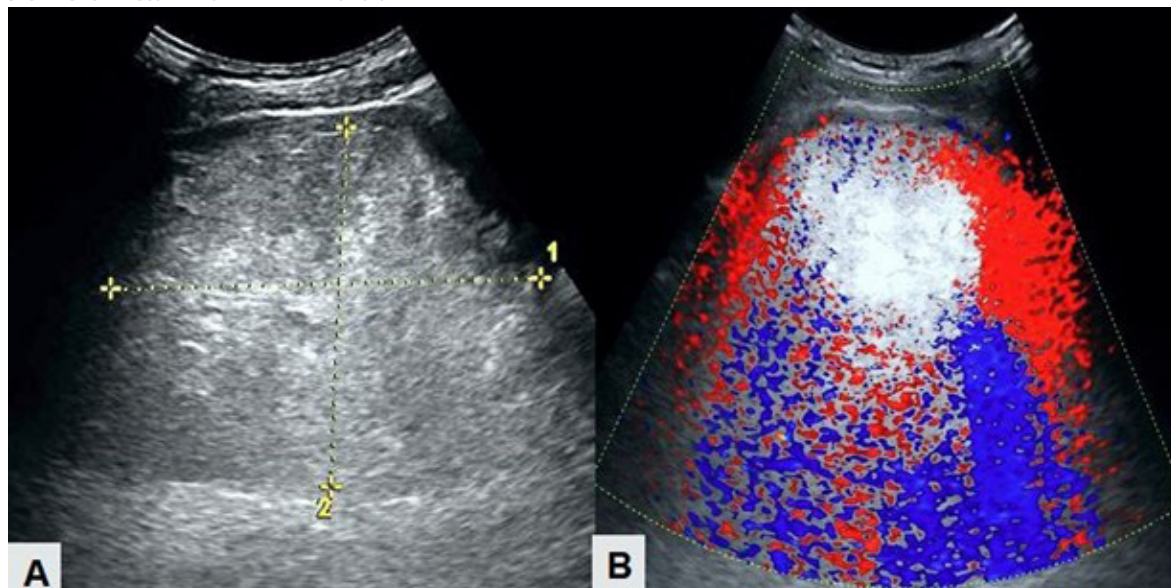
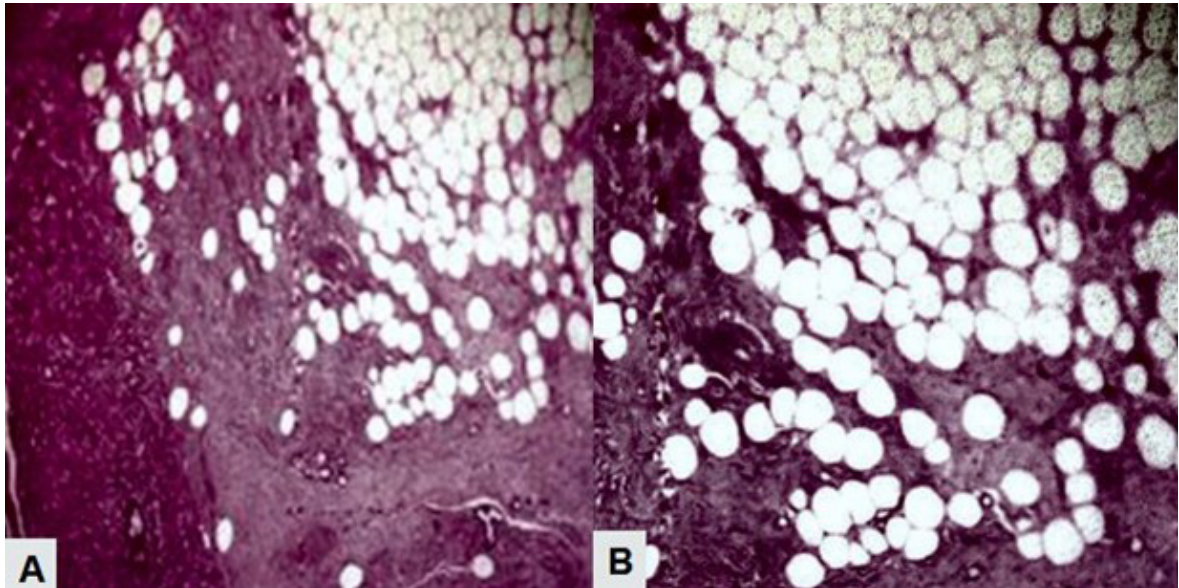




FIGURE 3. MICROSCOPIC IMAGE OF UTERINE LIPOLEIOMYOMA. A) PROLIFERATION OF SPINDLE-SHAPED SMOOTH MUSCLE CELLS MIXED WITH MATURE ADIPOCYTES (HEMATOXYLIN-EOSIN STAIN, 10X). B) MATURE ADIPOCYTES WITHOUT CELLULAR ATYPIA MIXED WITH SMOOTH MUSCLE CELLS (HEMATOXYLIN-EOSIN STAIN, 40X).



cytes were mature and without evidence of lipoblasts, mitosis, necrosis or angiomatosis. On immunohistochemistry, the cells were positive for smooth muscle actin but negative for HMB-45. The endometrium showed simple hyperplastic changes without atypia. The anatomopathologic diagnosis was uterine lipoleiomyoma.

The patient had an uncomplicated postoperative period and was discharged on the third day of surgery. After 12 months of follow-up there was no evidence of tumor recurrence.

DISCUSSION

Uterine lipomatous tumors are divided into 3 groups: pure lipomas (composed of mature adipose tissue), lipoleiomyomas (composed of a mixture of smooth muscle cells and mature adipose tissue) and malignant liposarcomas⁽⁵⁾. Lipoleiomyoma of the uterus was first described as soft tissue myolipoma in 1991⁽¹⁾. Like the typical leiomyoma, it is a benign and well-defined lesion, but with a large amount of adipose tissue⁽⁴⁻⁶⁾.

The pathogenesis of lipoleiomyoma is unknown. Lipomatous metaplasia or pluripotent mesenchymal cells have been proposed as hypotheses to explain its origin. Some authors have also proposed vascular proliferation like that observed in renal angiomyolipomas as a possible cause^(7,8). However, fatty metaplasia is the most accepted

theory, since the tumors lack lipoblasts, adipocyte atypia or smooth muscle cells and necrosis.

The clinical manifestations of lipoleiomyoma are uncertain due to its low incidence. Unlike typical leiomyomas that tend to occur predominantly in women of reproductive age and diminish at menopause, these tumors are very slow growing, largely asymptomatic, and continue to grow after menopause⁽⁹⁾. Therefore, lipoleiomyoma is most common in perimenopausal and menopausal women between 50 and 70 years of age^(5,6). In general, most patients are asymptomatic, and the diagnosis is made incidentally. However, some present with symptoms similar to typical leiomyomas of comparable size, such as abdomino-pelvic discomfort, heaviness, and genital bleeding⁽²⁻⁴⁾. In cases of acute and persistent abdominal pain, the possibility of torsion should be considered⁽¹⁰⁾.

Patients with uterine lipoleiomyomas usually have associated metabolic disorders, such as dyslipidemia, diabetes mellitus, or hypothyroidism. These metabolic disorders, together with estrogen deficiency occurring in the peri- or menopausal period, seem to promote abnormal intracellular storage of lipids in adipocytes within the tumor. However, the precise reason for this association is still unknown⁽¹¹⁾. This hypothesis is consistent with the advanced age of most patients at the time of diagnosis⁽³⁾. However, the present case did not present any of the metabolic disorders mentioned.



Imaging diagnosis of lipoleiomyoma is challenging, as it can sometimes resemble both a typical leiomyoma and a mature ovarian teratoma, depending on its location^(8,12). The characteristic ultrasound image is of a well-defined heterogeneous lesion containing hyperechoic areas due to adipose tissue. Therefore, accurate diagnosis depends on the correct identification of the tumor components⁽⁷⁾. Magnetic resonance imaging and computed tomography allow the determination of the location of the tumor and the nature of its contents⁽⁴⁾. Although some features in the different imaging modalities may suggest the possible diagnosis of these tumors, the precise diagnosis is based on histopathological findings⁽¹³⁾.

Lipoleiomyomas present characteristic histological findings, as they are composed of adipocytes, hyperplastic smooth muscle cells and fibrous tissue. The combination of mature adipocytes and smooth muscle cells is necessary to correctly assign this condition⁽³⁾. Among lipoleiomyomas, the distribution of the adipose component is highly variable and ranges from uniform distribution within the tumor to focal concentrations in small areas. Therefore, there is no specific percentage of mature adipocytes to confirm the diagnosis. On the other hand, tumors with few microscopic foci of adipose tissue are similar to typical leiomyomas, while those with large amounts of adipose components have yellowish coloration and lobulated cut surface⁽⁵⁾.

The differential diagnosis of uterine lipoleiomyomas includes benign cystic teratoma, malignant degeneration of cystic teratoma, non-teratomatous ovarian lipomatous tumor, benign pelvic lipomas, liposarcomas and lipoblastic lymphadenopathies⁽¹⁴⁾. An association has been described between uterine lipomatous tumors with both endometrial carcinomas and lipoleiomyosarcomas arising from uterine lipoleiomyomas⁽¹¹⁾.

Small, asymptomatic lipoleiomyomas are managed similarly to typical leiomyomas. Surgery is only indicated in larger lesions that produce compression to neighboring organs and/or menstrual alterations^(3,4). They have an excellent prognosis, since, to date, there are no reports of tumor recurrence after hysterectomy. Long-term follow-up of patients has shown that these tumors are benign, with no evidence of recurrence and are not associated with mortality, if diagnosed as a single pelvic pathology⁽⁴⁾. However,

there are reports of lipoleiomyosarcomas arising from uterine lipoleiomyomas and cases of intravenous lipoleiomyomatosis, so patients should undergo frequent clinical and pathologic evaluations to detect any coexisting gynecologic neoplasms⁽¹⁵⁾.

In conclusion, uterine lipoleiomyomas are extraordinarily rare, with clinical manifestations similar to typical leiomyomas. They are usually asymptomatic, so diagnosis can be difficult. In case of diagnostic doubt in the ultrasound evaluation, computed tomography and/or magnetic resonance imaging allow differentiation of the adipose component within the tumor. The definitive diagnosis is based on anatomopathologic findings that show the presence of mature adipose tissue and smooth muscle cells in different proportions without cellular atypia. Surgery is only indicated to alleviate the associated symptoms.

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