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Case Report

Uterine lipo-leiomyoma in post-menopausal women: two interesting cases

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ABSTRACT

Lipo-leiomyomas (LL), incidentally found lipomatous variants of uterine leiomyomas do not require surgical intervention unless symptomatic. Large LL of the uterine body is very rare. Majority being benign, these could co-exist with other gynaecological malignancies, metabolic diseases and abnormal oestrogen statuses and do rarely transform to liposarcoma. Here, we presented two interesting cases of postmenopausal women with LL. Ultrasonography and MRI performed in the workup of both, incidentally demonstrated a fatty lesion arising from the uterine corpus, consistent with a lipo-leiomyoma. This case report stresses the need for greater awareness of these variants regarding histogenesis, diagnosis and treatment.

Keywords: Benign, Lipoleiomyoma, Adipocytes, Smooth muscle, Uterine, post-menopausal

INTRODUCTION

Uterine lipomas, first described by Lobstein in 1816, are subdivided into “pure lipomas” and “mixed lipomas” depending upon the absence or presence of muscle tissue.¹ Uterine lipo-leiomyoma (LL) is a rare benign true neoplasm under the umbrella of uterine leiomyoma.

Fatty metamorphosis as known earlier, lipomatous degeneration, hamartoma, and adipose metaplasia are synonyms which mostly occurs in asymptomatic obese perimenopausal or menopausal women.^{2,3}

This case series stresses to create greater awareness of lipo-leiomyoma regarding theories of origin, diagnosis and treatment.

CASE REPORT

Case 1

A 63-years-old, P6, postmenopausal woman presented with mass abdomen and retention urine for 6 months duration. On abdominal examination, a 24 weeks globular, non-tender mass. USG revealed a mass measuring 16 cm×9.1 cm from the fundus. USG and MRI revealed lipo-leiomyoma. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was done on March 8, 2021.

On immune-histochemical analysis, the smooth muscle bundles were immune-reactive for Desmin. The intraoperative and post-operative period was uneventful.

Case 2

A 54-years-old postmenopausal P4 obese with a history of recurrent right lower abdominal pain for 4 months with no history of postmenopausal bleeding, vaginal discharge, dysuria or fever. H/O hypertension and diabetes mellitus and dyslipidemia. Abdominal examination revealed 16 weeks size firm non-tender mass. The gynecological examination did not show any vulval or cervical abnormalities. Pelvic US revealed multiple fibroids, largest, well-demarcated, hyperechoic lesion measuring 7.1×6.6 cm surrounded by a hypoechoic rim which represents a layer of myometrium on the anterior wall of the uterus. MRI confirmed the presence of lipo-leiomyoma (Figure 1). The women underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy on August 16, 2021. Grossly, the well circumscribed mass measured 15.6×9×6 cm. The outer surface was encapsulated, smooth, with few prominent vessels. The cut surface was solid with a yellowish colour (Figure 2). Cervix was unremarkable and the rest of the bilateral adnexa were grossly normal. Microsections of the mass showed interlacing fascicular arrangement of smooth muscle bundle admixed with the clusters of benign mature adipocytes (Figure 3). On IHC, desmin was positive in the smooth muscle component. Based on the morphological and a pathological diagnosis of lipo-leiomyoma of the uterus was rendered (Figure 4). The adipose tissue of the present tumour was free from atypia, and no lipoblasts were seen. The postoperative recovery course was uneventful, discharged on 22 August 2021.

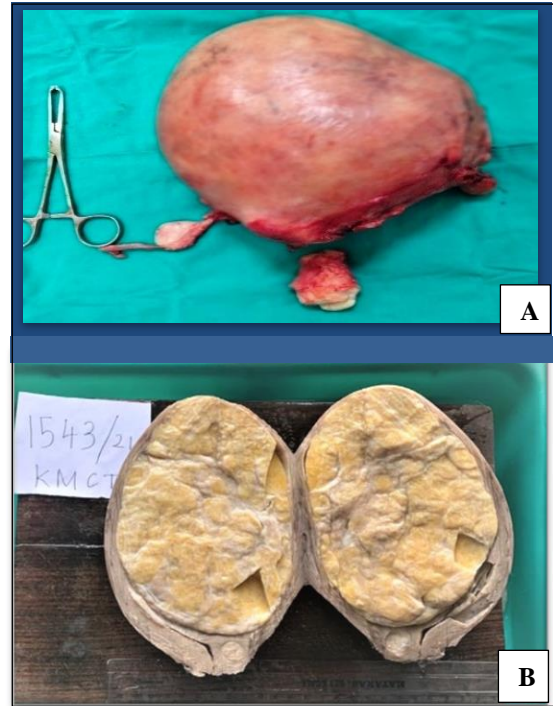


Figure 2: The gross appearance of lipo-leiomyoma; and (B) gross appearance with considerable amounts of fat made the cut surface appear yellow.

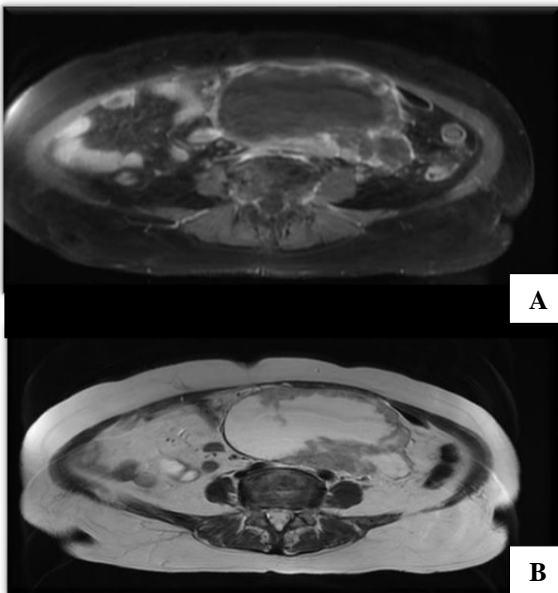


Figure 1: (A) MRI-T, weighted (SE 2000/20) image; and (B) inversion recovery (2000/60; TI=140 msec) image. The change in relative signal intensity of the fatty tumour components follows that of subcutaneous and pelvic fat. The fat composition can be detected effectively by using fat suppression techniques that reduce chemical shift artifacts and associated signal drop.

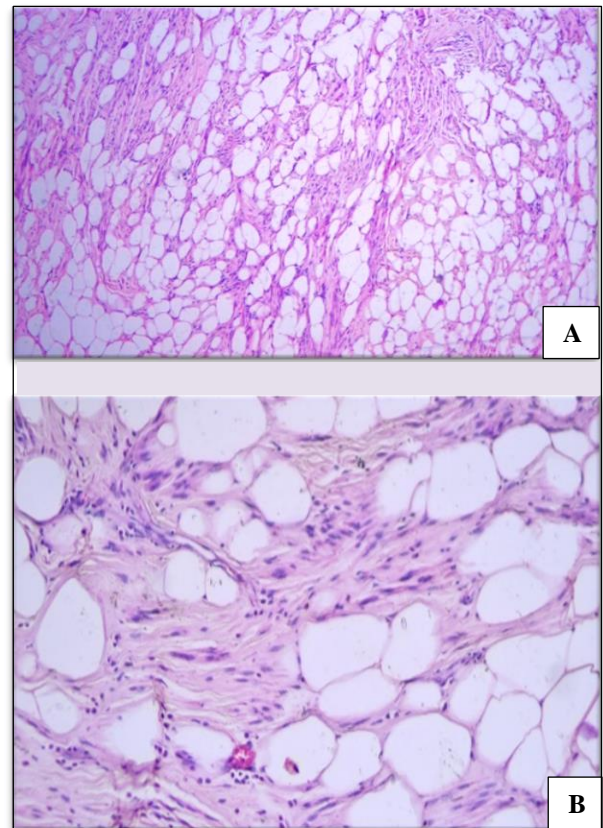


Figure 3: Microsections of the mass showed interlacing fascicular arrangement of smooth muscle bundle admixed with the clusters of mature adipocytes.

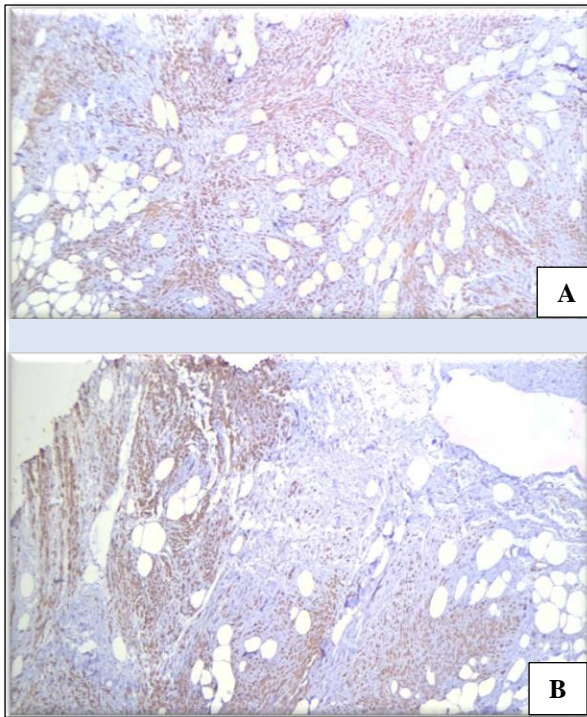


Figure 4: Microscopic examination of a lipo-leiomyoma with a high adipocyte component-desmin.

DISCUSSION

Uterine lipomas, first described by Lobstein in 1816, are subdivided into “pure lipomas” and “mixed lipomas” depending upon the absence or presence of muscle tissue.¹ The spectrum of lipomatous lesions comprises pure lipoma, lipo-leiomyomas, and fibro-lipomyomas. Lipo-leiomyomas are histologically composed of variable amounts of smooth muscle, fat cells, and fibrous tissue.⁴

Theories

Histogenesis of the lipomatous lesions of the uterus includes misplaced embryonic fat cells, adipose metaplasia of stromal or smooth cells, fatty infiltration or degeneration of connective tissue, or pluripotential cells or lipoblasts migrating along uterine arteries and nerves.⁴ Adipocytic metaplasia of uterine smooth muscle cells which can proceed to form localized adipocyte tissue in leiomyoma is the most accepted theory.⁵

Environmental factors in the uterus may play a role in adipocyte differentiation in LL.⁵ Lipomatous metamorphosis could occur in conditions of low oxygen, hypoxia, or serum starvation. Adipocytes have a stimulating effect on leiomyomas via TNF- α , and Wnt-10b. Wnt/ β -catenin signalling in mesenchymal stem cells can regulate adipogenesis (Figure 5 and 6).⁶

Presentation

Most of the patients are usually asymptomatic.⁷ Abnormal uterine bleeding, palpable mass, pelvic discomfort and

heaviness along with urinary frequency, and incontinence could be the presenting features. Cervical LL present with dysuria, urinary urgency, and ureteral obstruction.⁸ Uterine prolapse and inversion caused by a continuous enlargement of the tumour is a severe complication.⁹

Incidence and location

Uterine lipo-leiomyoma is a rare benign true tumour, with an incidence ranging from 0.03% to 0.2%.¹⁰ It was reported that the simultaneous occurrence of a traditional uterine leiomyoma in an analysis of 70 lipo-leiomyoma patients.⁷ Uterine LL are mostly found in the uterine corpus and intramural regions as the cervix or the intravascular region of the broad ligament.^{1,7,11} Extra-pelvic-inguinal, vulva, parasternal have been described. An extrauterine lipo-leiomyoma was also described as a myo-lipoma, a rare benign lesion consisting of smooth muscle and regularly distributed mature adipose.^{12,13}

Associations and size

Metabolic diseases including hyperlipidemia, hypothyroidism, and diabetes mellitus and hyper-estrogenic states are seen with uterine lipo-leiomyoma.¹⁴ Lipo-leiomyomas also co-exist with leiomyomas, adenomyosis endometriosis, endometrial hyperplasia, and polyps in the same patient.^{4,7} These may present as solitary or multiple lesions, with an average size range of 5-10 cm, but it can vary from a few mm to 32 cm in size.¹⁵

Imaging

Provides reproducible and non-invasive maps of tumours and plays a significant role in demonstrating the presence of adipose tissue and confirming the location of the lipo-leiomyoma. The sonographic appearance of leiomyomas is a hyperechoic mass that is partially encased by a hypoechoic rind.¹⁶⁻¹⁸ The rind is thought to represent a layer of the myometrium surrounding the fatty component. No obvious vascularity can be seen in most cases; however, in broad ligament fibroids, vascularity has been documented, along with a bridging vascular signal.¹⁹ Computed tomography (CT) is the main method for evaluation of presenting symptoms.²⁰ CT scans showed a well circumscribed, predominantly fatty mass with areas of non-fat soft tissue density arising from the uterus.^{21,22} However, the disadvantage of CT is its inability to depict detailed uterine anatomy and to identify the origin of the lesion, which can result in confusing an adnexal mass with a uterine lesion.²³ Magnetic resonance imaging is the most beneficial imaging modality in lipo-leiomyomas with hyperintensities on the T1 weighted sequence. Multiplanar capabilities, high sensitivity, and specificity to fat and tissue characterization, is the modality of choice for confirmation of diagnosis. On MRI, the lesion shows high signal intensity on T1- and T2-weighted images, and slightly higher intensity in the skeletal muscles in the same section.²⁴ The fat composition can be detected effectively by using fat suppression techniques that reduce chemical

shift artifacts and associated signal drop.^{25,26} Ando et al concluded that radiologists should undertake two consecutive procedures: (a) evaluation of the presence of increased signal intensity on T1-weighted images; and (b) evaluation of the fat components on fat-suppressed T1-weighted images.²⁵

Demonstration of fatty components of the tumour can also differentiate lipo-leiomyomas from leiomyosarcomas; however, they have low positive predictive value in the differential diagnosis of well-differentiated liposarcomas that mimic lipo-leiomyomas, characterized by nodular septa and prominent foci of high T2 signal as well as prominent areas of enhancement.^{24,25,27} It can be beneficial to identify possible complications caused by mechanical compression of large-size lipo-leiomyomas, such as hydronephrosis or hydroureter. Magnetic resonance spectroscopy, used to evaluate the concentration of metabolites in tissues, has been performed to diagnose pure uterine myomas by the existence of the lipid peaks, and thus, it may be helpful in diagnosing lipo-leiomyomas, which contain relatively more adipocytes.²⁴

Pathologically

Lipomatous tumours of uterus are categorized into three groups- (a) pure lipoma composed only of mature fat cells and is encapsulated; (b) mixed type- lipo-leiomyoma, angiomyolipoma, fibro-myolipoma etc along with various *mesodermal* tissue components adipose tissue, smooth muscles, fibrous component and connective tissue; and (c) malignant neoplasm like liposarcoma consisting of less differentiated fat cells that have undergone sarcomatous change.

Histologically

Lipo-leiomyoma can be differentiated from leiomyoma with fatty degeneration, by the even distribution of adipose tissue admixed with smooth muscle bundle.¹⁵ The adipose component was entirely mature without any lipoblasts, nuclear pleomorphism and prominent mitotic activity. Further, it can be distinguished from leiomyosarcoma by the bland appearance of the nuclei and occasional mitosis in the smooth muscle component.²⁸

Immunohistochemistry-IHC

The smooth muscle cells intervening the adipose tissue will render positive for SMA, desmin, vimentin, while the adipose tissue for vimentin, thus supporting the theory of direct transformation of muscle cells into adipose cells that may originate from the transformation of a totipotent mesenchymal cell IN LL.^{4,29}

Differential diagnosis

It include benign cystic teratoma, malignant degeneration of cystic teratoma, non-teratoma, lipomatous ovarian

tumour, pelvic lipomas and liposarcomas, and pelvic fibromatosis.⁷

Rate of growth

This is higher for lipo-leiomyomas than for conventional uterine fibroids, the latter of which tend to regress after menopause, it is necessary to document tumor size and assess growth stability at regular follow-up intervals, which may require shortening the interval or changing the treatment. Uterine lipo-leiomyomas are variants of uterine leiomyomas and are characterized by progressive enlargement that can occur even after menopause.³⁰ The phenomenon of tumour-to-tumour metastasis (TTM) has been documented in uterine lipoleiomyomas.³¹

Surgical options

Depend on tumour size, location, number of lesions, patient preference, and available surgical facilities. Hysterectomy may be offered to patients who do not wish to preserve their uterus, according to standard uterine myomas guidelines.³² Myomectomy of very large fibroids carries an increased risk of serious perioperative complications such as excessive bleeding.³⁴

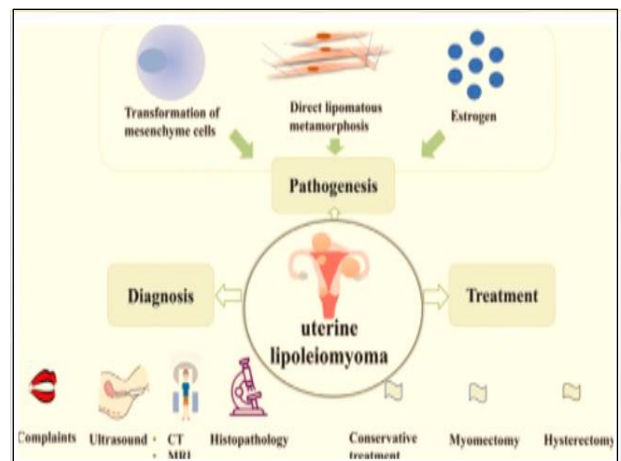


Figure 5: Histogenesis of lipomatous lesions.^{4,5}

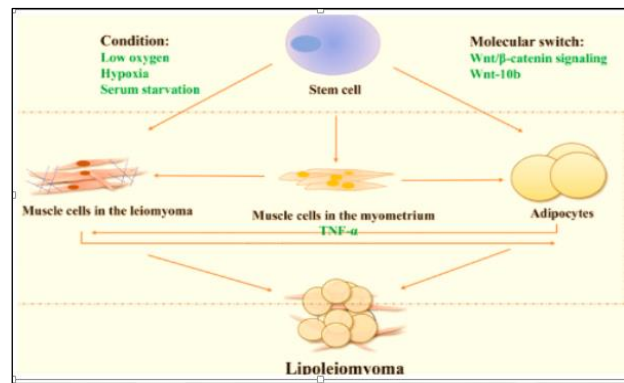


Figure 6: Environmental factors affecting differentiation leading to lipo-leiomyoma uterus.^{5,6}

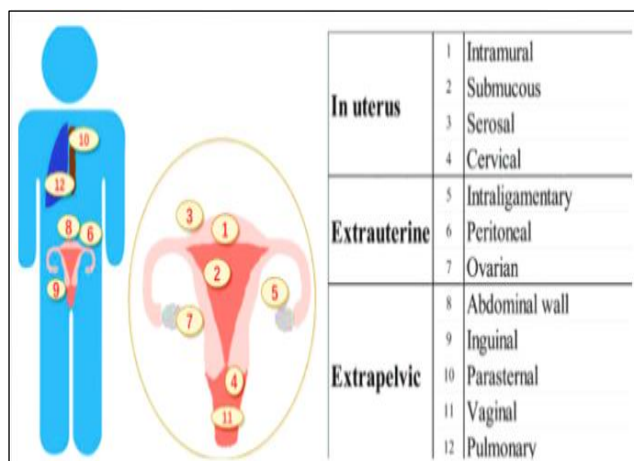


Figure 7: Location of lipo-leiomyomas.^{12,13}

CONCLUSION

Awareness of the lipomatous lesions of the uterine cavity is important as clinical symptoms of lipo-leiomyoma are very similar to leiomyoma but have a distinctive radiological, histological, and immunohistochemical characteristics. The purpose of this case report is to promote greater awareness of lipo-leiomyoma characteristics with a focus on histogenesis, diagnosis, and treatment.

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