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Case Report

Uterine lipoleiomyoma: Case report and review of the literature☆

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ABSTRACT

Uterine lipoleiomyomas are variants of uterine leiomyomas that are often found incidentally, and do not require surgical intervention, unless symptomatic, and thus must be differentiated from lesions that need to be excised. While these tumors are benign, there have been reports of uterine lipoleiomyomas coexisting with other gynecological malignancies, metabolic diseases and abnormal estrogen statuses, as well as going through a malignant transformation into a liposarcoma. Here we present a 58-year-old female that presented with complaints of right upper quadrant abdominal pain. Ultrasonography and computed tomography performed in the workup incidentally demonstrated a fatty lesion arising from the uterine corpus, consistent with a lipoleiomyoma. This report exemplifies the importance of correctly distinguishing between non-malignant and malignant uterine masses in order to provide the correct management, as well as determining the need for further investigation relating to other malignancies, metabolic diseases, abnormal estrogen statuses and being aware of malignant transformation.

Introduction

A uterine lipoleiomyoma is an uncommon variant of the relatively common uterine leiomyoma [1]. Lipoleiomyomas are rare, benign tumors composed of mature adipocytes intermixed with smooth muscle cells. The reported incidence of uterine lipoleiomyomas is 0.03% – 0.2% of all uterine leiomyomas [2–4]. The histogenesis of these benign tumors is still unknown, though, there are several theories supported by immunohistochemical staining [3,5–7]. While these tumors are benign, there have been reports in the literature of uterine lipoleiomyomas coexisting with other gynecological malignancies, as well as with various metabolic diseases and ab-

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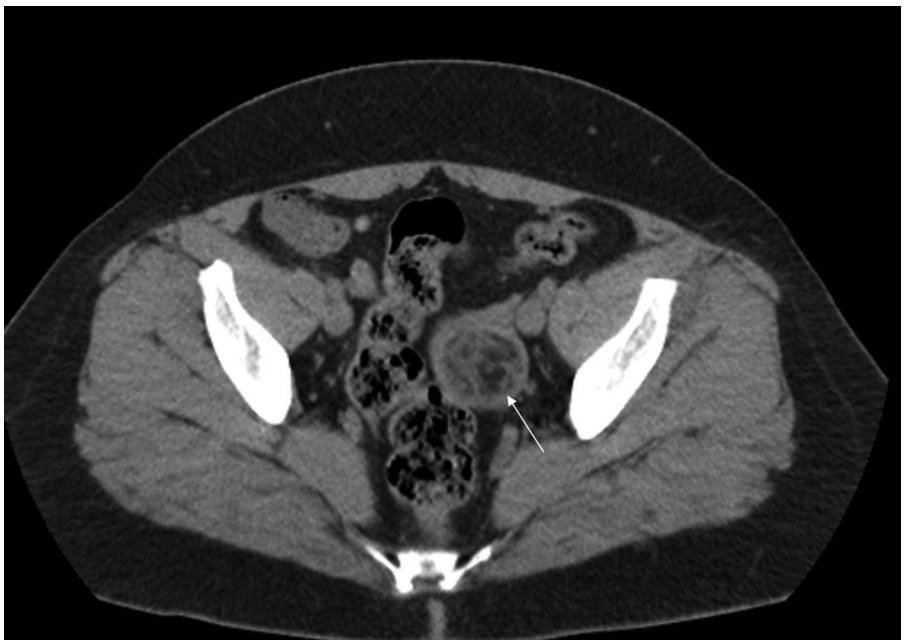

Fig. 1 – Transverse non-contrast CT of the pelvis demonstrating a macroscopic fat containing lesion (White arrow) within the uterine parenchyma, consistent with a lipoleiomyoma.

normal estrogen statuses [8,9].This report aims to describe a case of uterine lipoleiomyoma from initial presentation up to diagnosis and management. Furthermore, this report intends to describe the characteristic appearance of this rare entity on imaging, as well as discuss the theories of histogenesis and the presence of other malignancies and metabolic diseases in the literature over the past ten years.

Case report

A 58-year-old postmenopausal female presented with complaints of right upper quadrant abdominal pain. The patient's BMI at the visit was 31.73 kg/m^2. The patient's past medical history was mostly unremarkable other than Sjögren's Syndrome. An abdominal and pelvic CT demonstrated a heterogenous lesion measuring 4.3 × 4.0 × 3.8 cm (Fig. 1). The CT scan showed fatty areas that originated from the uterus evidenced by the "claw sign" of the uterine parenchyma (Fig. 2), and this was thought to be a lipoleiomyoma or an ovarian dermoid cyst. The remainder of the CT was unremarkable other than ectopic position and malrotation of her right kidney, as well as a thickened endometrial stripe measuring 1.6 cm. A transvaginal ultrasound was then performed and showed an avascular, solid hyperechoic mass in the lower posterior uterine wall measuring 4.0 × 3.8 × 3.7 cm (Fig. 3), which was consistent with a diagnosis of lipoleiomyoma. After imaging, the patient was referred to an outside clinic for OB/GYN consultation.

Upon consultation, a hysterectomy with bilateral salpingo-oophorectomy was performed. Found in the removed tissue was a minute focus of atypical endometrial hyperplasia, an adenofibromatous polyp measuring 4 cm in greatest diameter, and multiple leiomyomas, including 1 lipoleiomyoma, measuring 4.4 × 3.4 × 3.5 cm, thus confirming the diagnosis. The patient's cervix and bilateral fallopian tubes and ovaries were found to have no significant histologic abnormalities.

We obtained written informed consent from the patient for publication of this case prior to beginning this report.

Discussion

Lipoleiomyomas are lipomatous variants of uterine leiomyomas. They are typically incidental findings that most commonly occur in postmenopausal women. Akbulut et al. reported a mean age of 55.49 years old based on 70 patients with lipoleiomyomas, and these patients are typically obese and asymptomatic [6]. In our case study, the patient had a BMI of 31.73 kg/m^2 and was age 58 at the time of diagnosis, which is consistent with the recent literature of a postmenopausal, obese woman developing a lipoleiomyoma. If these patients are symptomatic, they most commonly present similar to uterine leiomyomas, with symptoms such as menstrual disturbances, pelvic pain or pressure, constipation or incontinence, as well as a palpable mass if the lesion is large enough [1,9].

Lipoleiomyomas are most commonly seen on imaging in the uterine corpus, but there have been reports in the literature of lipoleiomyomas occurring in the broad ligament, cervix and retroperitoneum [3,4,6,9]. The differential diagnosis for a fat containing lesion of the uterus includes a lipoleiomyoma, mature ovarian teratoma, benign lipoma and benign or malignant degeneration of ordinary leiomyomas [7,9]. Distinguishing a lipoleiomyoma from a lesion that needs to be excised, such as a mature ovarian teratoma, is essential

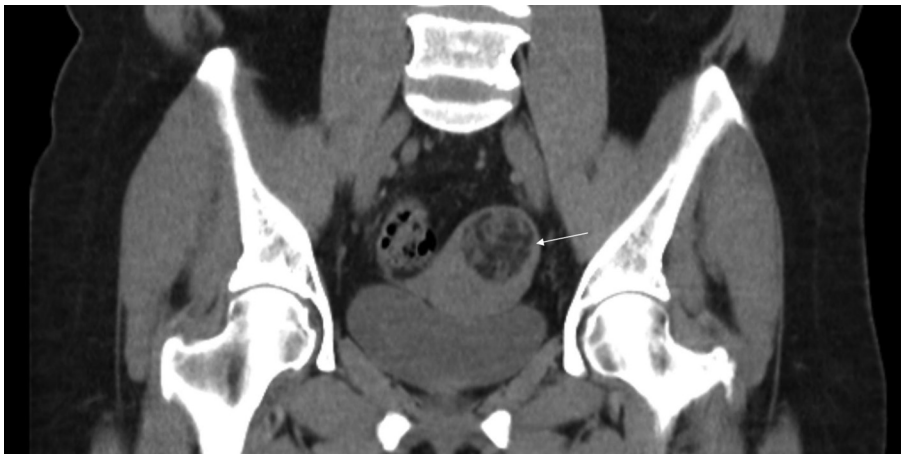

Fig. 2 – Coronal non-contrast CT revealing uterine parenchyma surrounding the low-density fat containing lesion compatible with a "claw sign." (White arrow).

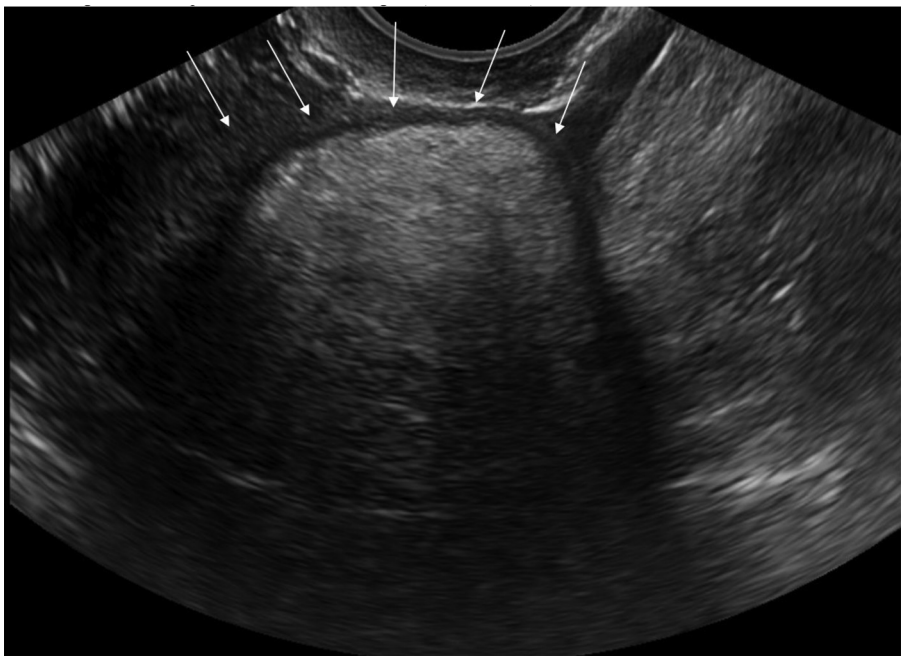

Fig. 3 – Ultrasonography showing the isoechoic/hypoechoic uterine parenchyma surrounding the echogenic fat containing lipoleiomyoma with "claw sign". (White arrows).

in sparing patients from unnecessary invasive procedures. On sonography, lipoleiomyomas typically appear as hyperechoic avascular masses with a hypoechoic rim from the uterine myometrium, as seen in our patient. However, ultrasound is not always able to definitively show the location of the lesion. In contrast to sonography, CT can more definitively show the uterine origin of the mass demonstrated by the claw sign representing the rim of myometrium surrounding the fatty tumor. On CT, the masses have intralesional macroscopic fatty contents. An MRI can also be done to show the fatty content of the tumor, and fat suppression techniques can be used to confirm the fat within the tumor [4,7,9,10].

There have been multiple reports in the literature of lipoleiomyomas coexisting with gynecological malignancies. Oh et al. included six cases of lipoleiomyomas with 1 patient having coexistent stage 1A1 cervical cancer [4]. Akbulut et al. reported twelve gynecologic malignancies among the seventy patients with lipoleiomyomas and reporting gynecologic malignancy in 11% –20% of lipoleiomyomas [6]. Another study reported three patients where liposarcoma arose in uterine lipoleiomyomas [11]. There was no gynecological malignancies found on imaging within our case study. These three studies in the last ten years do not connect lipoleiomyomas with malignancy, however, there must be concern for it when a

lipoleiomyoma is found on imaging, and a thorough search for a second lesion should be conducted.

The treatment for lipoleiomyomas depends on the presenting symptoms. If asymptomatic, no treatment is needed, as lipoleiomyomas can be managed conservatively. Although, these lesions can be managed conservatively in asymptomatic patients, there is one report in the literature where 3 patients with lipoleiomyomas developed liposarcomas within their lipoleiomyoma, which means observation is necessary for changes [11]. In symptomatic patients, the most common treatment performed in the literature is a hysterectomy [3,4,10,12,13].

Lipoleiomyomas have also been hypothesized to be more common in patients with metabolic disorders with a hyperestrogenic state, such as hyperlipidemia, hypothyroidism, diabetes mellitus and postmenopausal lipid metabolism changes [3,5,6,8,9]. Out of seventy patients in the study done by Akbulut et al., eight patients had diabetes mellitus, six had hypothyroidism, as well as fifty-three of the seventy had other lesions that are associated with hyperestrogenic states, including adenomyosis, endometriosis, endometrial hyperplasia, polyps and gynecologic carcinomas [6]. Karaman et al. reported a lipoleiomyoma in a single patient with a ten year history of diabetes mellitus and hypercholesterolemia, as well as Sharma et al. reported ten lipoleiomyomas with 2 having diabetes mellitus, two with hypothyroidism and one with high triglyceride levels [1,9]. Although these reports show a possible link, there are multiple case reports in the literature where the patient does not have a metabolic disorder [3–5,7]. The increased risk of lipoleiomyomas in postmenopausal women is due to lipid metabolic changes during this period, as well as increased plasma levels of lipids if the patient is obese, which could lead to increased likelihood of development [1,5]. Our patient did have endometrial stripe thickening, however, there was no diagnosis of any metabolic disorder. While the link between lipoleiomyomas and gynecological malignancy, estrogenic status and metabolic disorders have not been statistically proven on a large-scale study, the finding of adipocytes in a leiomyoma should warrant a further evaluation for malignancy, estrogen status and metabolic disorders.

The histogenesis of a lipoleiomyoma is still a mystery, but it is regarded as a distinctive, true neoplasm [5]. Possible theories include lipoblastic differentiation of misplaced embryonic fat cells, metaplastic changes of connective tissue or smooth muscle fibers into adipocytes, and finally, pluripotent cell migration along the uterine nerve and vessels and fatty infiltration [2,9]. These theories have been developed through immunohistochemical staining done on lipoleiomyomas. Terada et al. demonstrated the adipocytes of the tumors stained positively for vimentin, S100 protein, as well as estrogen and progesterone receptors, but produced a negative stain for alpha-smooth muscle actin and desmin, whereas the smooth muscle portion of the tumor stained positive for vimentin, desmin, alpha-smooth muscle actin, estrogen and progesterone receptors [5]. The positivity for estrogen and progesterone receptors supports the fatty tissue being related to the female genital organs. Another study comprised of 76 lipoleiomyomas showed adipocytes staining positive for desmin and vimentin, as well as estrogen and progesterone receptors, which supports smooth muscle cells transforming into adipose cells [6]. Sharma et al. and Chandawale et al. also indicated adipose cells positively staining for vimentin in each of their patients [3,9]. Based on the commonality of positive staining of adipocytes for vimentin in the recent literature, their shared conclusion is that lipoleiomyomas probably result from smooth muscle cell metaplasia into fat cells that possibly originates from a totipotent mesenchymal cell [3,5,6,9].

Conclusion

Uterine lipoleiomyomas are uncommon variants of uterine leiomyomas. Lipoleiomyomas are composed of mature adipocytes with intermixed smooth muscle cells. These tumors tend to occur more often in postmenopausal women. Although the histogenesis has not been fully elucidated, multiple reports in the last ten years have concluded that positive adipocyte staining for vimentin point to lipoleiomyomas most likely resulting from smooth muscle cell metaplasia into fat cells that possibly originates from a totipotent mesenchymal cell. After reviewing the literature, there are reports of lipoleiomyomas coexisting with gynecologic malignancies, as well as with estrogen changes and metabolic disorders, meaning that a further evaluation may be warranted when a fatty uterine tumor is discovered.

Patient consent

We obtained written informed consent from the patient for publication of this case prior to beginning this report.

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