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Short communication

Rare association of postmenopausal uterine lipoleiomyoma in a frail woman with deranged lipid profile

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ABSTRACT

Uterine leiomyoma is a common uterine tumor. Because it is a benign uterine tumor, its variants are often underreported by histopathologists. Lipoleiomyoma (LL) is a very rare variant, currently increasingly reported due to increased awareness. Imaging studies can help to locate this tumor but it seldom helps in diagnosing the same. Most of such cases are incidentally/retrospectively reported on gross examination and histopathology after hysterectomy. We report a rare case of lipoleiomyoma occurring in a postmenopausal thin woman with deranged lipid profile.

1. Introduction

Uterine leiomyomas are estrogen-dependent common gynecological benign tumors with prevalence of 25–40% in the reproductive age group. They arise from uterine smooth muscle cells. Based on their location, they are submucosal, intramural, or subserosal types.[1–3] Uterine lipoleiomyomas (LL) are rare benign variants reported in post-/perimenopausal women, and presently their incidence ranges from 0.03% to 0.2%.[1,2] In the current literature, it is probably due to tumor metaplasia or distinct tumor neoplasia within a leiomyoma rather than fatty degeneration, as previously thought.[4] It occurs in patients with deranged lipid profile secondary to estrogen deficiency and is mostly subserosal/intramural in uterine location.[4] Its differential diagnosis includes atypical lipoma, liposarcoma, leiomyosarcoma, and mature ovarian teratoma.[1] So, that is why it is important to diagnose this rare tumor.

2. Case report

A 60-year-old thin, postmenopausal woman presented with increased frequency of per vaginal bleeding since the past six months and distension of abdomen since 28 days. The patient's history revealed that she had attained menarche at the age of 14 years, and had regular menstrual cycles of 4–5 days duration and of moderate intensity at 28 days interval. She attained menopause ten years back and has two healthy children. Her body weight was 46 kg, height was 1.65 m, and BMI was 16.85 kg/m^2.

Gynecological examination revealed no abnormalities of the vulva and cylindrical vaginal portion of the cervix; no evident pathological change was detectable on clinical examination. Findings by ultrasonography (USG) of abdomen and pelvis showed uterus measuring 5.5 cm × 4 cm and homogenous myometrial echo pattern, with endometrial thickness of 4 mm. Three suspicious subserosal fibroids were noted arising from right cornu, left cornu, and anterior wall, measuring approximately 7.1 cm × 2.7 cm, 3.8 cm × 3.1 cm, and 3.4 cm × 3.1 cm, respectively. Bilateral adnexa were unremarkable and there was no free fluid in abdomen. The rest of the abdominal organs and pelvic organs were unremarkable. USG diagnosis was of multiple subserosal fibroids.

All standard serological and hematological parameters were within normal range, except for her lipid profile that showed serum cholesterol at 250 mg/dl, serum HDL at 37 mg/dl, and serum LDL at 135 mg/dl. However, her ECG was normal and she had no chest pain. The patient gave consent for total abdominal hysterectomy with bilateral salpingo-oophorectomy because of multiple leiomyomas.

The histopathologists received total hysterectomy specimen with bilateral adnexa. On gross examination of the specimen, the uterus measured 14 cm × 8 cm × 6 cm and had three subserosal well-circumscribed round masses. The biggest nodule, which was measuring 8 cm in diameter, differed from a typical appearance of uterine leiomyoma by being yellowish-white, lobulated, and having somewhat softer consistency on its cut surface with whorling (Fig. 1). Hard, calcified areas were not noted. Enlarged

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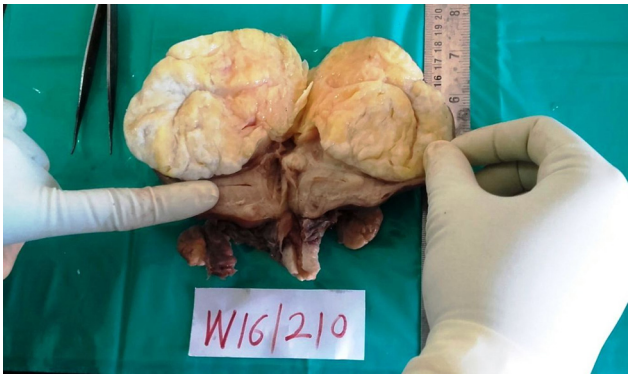

Fig. 1. Cut section of the tumor revealed yellowish-white areas.

fibroid had displaced the endometrial cavity toward cervix. Endometrial cavity measured 2 cm in length. Endometrial thickness measured 4 mm. Endocervical canal measured 2 cm in length. Cervix appeared thickened.

The other two leiomyomas, each of 1.5 cm diameter, showed a coarsely whorled pattern with grayish-white appearance on their cut surface. Right ovary measured 2.5 cm × 2 cm × 1 cm. Cut section of right ovary was solid and unremarkable. Left ovary measured 2.5 cm × 2 cm × 1 cm, the cut section of which was solid with small cystic areas. Right and left fallopian tubes measured 3.5 cm in length each and were unremarkable.

Histological examination of the biggest nodule showed a mixture of bland, spindle-shaped, smooth muscle cells without nuclear atypia in a whorled pattern with admixed mature adipocytes. The nuclei of the smooth muscles were elongated and had finely dispersed chromatin and small nucleoli. Between these muscle cells, a significant amount of fat cells were visible. The adipose component was entirely mature without any lipoblasts (Fig. 2). Based on the above findings, the tumor was diagnosed as a benign, subserosal lipoleiomyoma. Sections from the other fibroids showed classical histomorphology of conventional subserosal uterine leiomyomata. The endometrium showed atrophic changes. Cervical sections showed features of chronic ectocervicitis and chronic papillary endocervicitis with focal squamous metaplasia.

Sections from both the ovaries showed corpus albicans and left ovary also showed simple serous cyst. Both the fallopian tubes showed congestion of wall.

The patient recovered well surgically. Her lipid profile continued to be deranged postoperatively and so she was started on statin and aspirin drugs. Her lipid profile was normal after 4 weeks of medication with proper diet advised in follow-up by dietician.

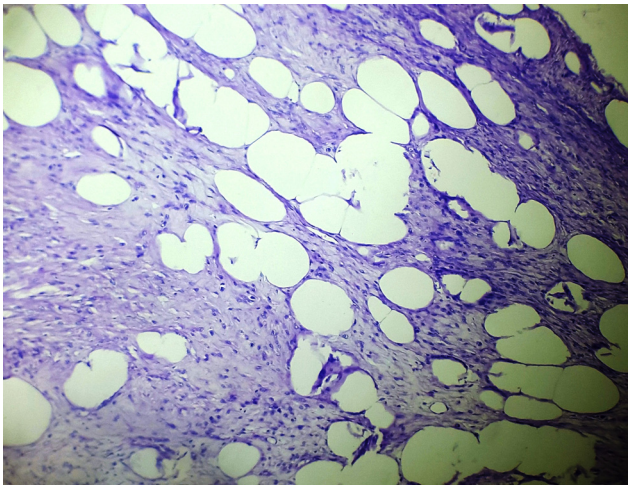

Fig. 2. Smooth muscle bundles with adipocytes (H&E, ×100).

3. Discussion

Myolipoma of soft tissue was first described in 1991 by Meis and Enzinger.[5] These tumors showed characteristic histological findings, being composed of benign smooth muscle and mature adipose tissue, even though the exact adipocyte concentration is not defined. Similar tumors in the uterus are known as lipoleiomyomas.[1,5]

Lipoleiomyomas occur in postmenopausal women with mean size of 5.5 cm diameter. The present case had such a tumor of 8 cm diameter. They may be single or multiple, but more than two lipoleiomyoma nodules in a uterus are not yet reported.[4,6] Also, calcerous degeneration in lipoleiomyoma is not yet reported. Its association with ovarian leiomyoma is reported.[3] The present case had no such ovarian findings.

Apart from common uterine location, LL occurs in different locations like ovaries, cervix, broad ligaments, and retroperitoneum. In uterus, intramural LL location is more common than subserosal or submucosal location.[1,2,4] Uterine LL is known to be associated with adenomyosis, endometriosis, endometrial hyperplasia/polyp, conventional leiomyomas, and various gynecological malignancies.[4] The present case of LL was subserosal in uterine location associated with two subserosal conventional leiomyomas; it is a rare case report where all three nodules were subserosal in uterus (Table 1).

Based on recent literature, it is suggested that lipoleiomyomas result from neometaplasia of uterine smooth muscles into adipocytes or totipotent, multipotential, undifferentiated mesenchymal cells or ectopic embryonic fat cells.[1–4,6,7]

Imaging studies help in exact tumor location with USG as the preferred diagnostic modality and are seldom underreported due to lack of intuition about the same. So, CT and MRI can be used to assist preoperative USG-based diagnosis, as the former helps to know the fat content of the rare tumor.[2,4] Though imaging plays an important role in preoperative diagnosis and localization of the lipoleiomyoma, it is the final pathological examination that confirms the diagnosis by ruling out the other differentials of this tumor.

The differential diagnosis of the lipomatous mass in the pelvis includes benign cystic teratoma, malignant degeneration of cystic teratoma, nonteratomatous lipomatous ovarian tumor, benign pelvic lipomas, liposarcomas, and lipoblastic lymphadenopathy.[1,6,7] So, it is important to diagnose this rare leiomyoma variant.

A number of various lipid metabolic disorders or other associated conditions, which are associated with estrogen deficiency as occurs in peri- or postmenopausal period, possibly promote abnormal intracellular storage of lipids.[1,6,7] That is why, our thin/frail postmenopausal woman had deranged lipid profile with lipoleiomyoma.

Lipoleiomyomas when asymptomatic require no treatment and are clinically similar to leiomyomas. So, it is important to differentiate these tumors from ovarian teratoma, which requires surgical excision. Lipoleiomyomas are benign tumors of the uterus that do not affect mortality.[7]

We conclude that uterine subserosal lipoleiomyoma associated with subserosal location of two conventional leiomyomas is exceedingly rare with clinical features similar to conventional leiomyoma, having intuitive radiological characteristics, retrospective histopathology-based diagnosis, and excellent prognosis. Also, deranged lipid profile is due to estrogen deficiency in postmenopausal frail women, which could be of significance in etiopathogenesis of uterine lipoleiomyomata, needing further