Uterine Lipoleiomyoma and Lipoma: A Rare Unique Case Report with Review of Literature

Abstract
Uterine lipoleiomyoma and pure lipoma are uncommon and extremely rare benign lipomatous tumors. Reported incidence is 0.03%–0.25%. Pure cervical lipoma is exceptionally rare. These tumors commonly arise in postmenopausal women. Ultrasonography and computed tomography finding may be nonspecific. Although magnetic resonance imaging can be useful in identifying the fatty nature of the lesion preoperatively, most of the cases postoperatively on histopathological examination. Although various theories of histogenesis of these tumors have been proposed, histogenesis remains to be enigma. These lesions may have coexistent malignancy in uterus, ovaries, and fallopian tubes or may have other metabolic disorders and abnormal estrogen status. Immunohistochemical studies have played an integral role in understanding its complex histogenesis. We did not find any case study in medical literature in which uterine lipoleiomyoma and cervical lipoma have been described in hysterectomy specimen. We report a unique case of a 43-year-old premenopausal woman with complaints of irregular vaginal bleeding and lower abdomen pain. Lipoleiomyoma in uterus corpus and cervical lipoma were diagnosed in hysterectomy specimen.

Keywords: Benign, lipoleiomyoma, lipoma, uterine

Introduction
Benign lipomatous uterine tumors are uncommon and rare. Overall reported incidence of lipoleiomyoma is 0.03%–0.25%. Pure lipomas of uterus are extremely rare, and only few cases have been reported in medical literature, and it is even exceptionally rare in cervix. The exact incidence is not known. Although various theories of histogenesis of these tumors have been proposed, it remains to be enigma. The principle significance of these lesions in uterine wall is that, they may have a coexistent malignancy in uterus, ovaries, and fallopian tubes. These patients may have other metabolic disorders and abnormal estrogen status. We report for the first time a unique a case of a 43-year-old female in whom lipoleiomyoma of the uterine corpus and pure cervical lipoma were diagnosed in hysterectomy specimen.

Case Report
A 43-year-old premenopausal woman with Gravida 3 and Para 3 came to gynecology outpatient unit with complaints of irregular vaginal bleeding and lower abdomen pain since 6 months. There was no significant medical history. On clinical examination, vital signs were normal, and abdomen was soft with no palpable mass. Abdominal and pelvic ultrasonography (USG) was performed and revealed heterogeneous echogenic mass each in uterine corpus (3.5 cm × 2.5 cm × 2.5 cm) and cervix (1.2 cm × 0.8 cm). A diagnosis of leiomyoma in uterine corpus and cervix with degenerative change was made. Surgical specimen of hysterectomy was received for histopathological examination. Specimen showed a well-circumscribed gray-white to yellow tumor mass (3 cm × 2.5 cm) at upper pole of uterine corpus and yellow tumor mass (1 cm × 1 cm) in cervix [Figure 1a].

Histopathology examination of the tumor from uterine corpus showed a tumor composed of lobules of mature adipocytes separated by fibrous septae. At places, thin bundles of smooth muscle cells were seen. Small sized normal blood vessels were seen in Figure 1b. There was no nuclear atypia or increased mitotic figures. A diagnosis of lipoleiomyoma was made. Histopathology of tumor from cervix showed a tumor tissue composed of lobules of mature adipocytes.
thick fibrous septae [Figure 1c], and few entrapped mucosal glands [Figure 1d]. A diagnosis of pure lipoma was made. Immunohistochemical (IHC) stains such as smooth muscle actin (SMA) and vimentin were done in sections of both the tumors to confirm the nature of cells. Smooth muscle cells, endothelial cells from the tumor of uterine corpus showed immunoreactivity with SMA and adipocytes were negative [Figure 2a]. Vimentin staining showed immunoreactivity with smooth muscle, endothelial cells and mature adipocytes, fibrous septae were negative [Figure 2b].

In sections of cervical lipoma, only endothelial cells of blood vessels showed immunoreactivity with SMA, and fibrous septae and mature adipocytes were negative [Figure 2c]. Mature adipocytes showed immunopositivity for vimentin [Figure 2d]. A diagnosis of lipoleiomyoma in the uterine corpus and pure lipoma in the cervix was confirmed.

Discussion

Uterine lipomatous tumors are uncommon neoplasms and are categorized into: (1) Pure lipoma which is composed of only mature fat cells. (2) Lipoleiomyoma, angiomyolipoma, and fibromyolipoma which consists mixture of mature adipose tissue, smooth muscles, fibrous tissue, and other connective tissue elements. (3) Liposarcoma which is malignant neoplasm[11-13]. Lipoleiomyomas can occur anywhere in uterine corpus and cervix. Lipoleiomyomas in extraterine sites such as broad ligament, retroperitoneum, and ovaries have been reported.[12] Uterine pure lipomas are extremely rare.[5-8] These tumors commonly arise in postmenopausal women, and most of them are asymptomatic. Some patients present with symptoms such as pelvic pain, discomfort, and vaginal bleeding.[6,7,11] Preoperative diagnosis of uterine lipomatous tumors is difficult and should be confirmed postoperatively on histopathological examination.[8] Imaging diagnosis can play an important role in determining the fatty nature of the lesion. USG and computed tomography (CT) finding may be nonspecific; magnetic resonance imaging (MRI) can be useful in identifying the fatty nature of the lesion. Although MRI is the modality of choice for the final diagnosis, most of these lesions are diagnosed postoperatively on histopathology.[3,8,11,12] Similar observations were made in our case study. USG diagnosis in our patient was leiomyoma. CT and MRI were not done in patient.

Lipoleiomyomas were previously called as fatty metamorphosis, lipomatous degeneration, hamartoma, and adipose metaplasia. It is now regarded as true neoplasm. Histogenesis of these tumors have been a subject of speculation and is still a mystery.[7] Many theories have been proposed which include lipoblastic differentiation of misplaced embryonic fat cells, metaplastic changes of connective tissue or smooth muscles into fat cells, pluripotent cell migration along the uterine nerve and vessels and fatty infiltration or degeneration of connective tissue.[7,8,12] Fatty degeneration of leiomyoma is very rare and can be differentiated from lipoleiomyoma. In lipoleiomyoma, fat is evenly distributed in the lesion suggesting fat cells as integral part of tumor. Similar observations were made in our case report.

IHC studies have played an integral role in understanding its complex histogenesis. Mignogna et al.[6] reported immunoreactivity of fat cells with vimentin, desmin, and SMA which support the hypothesis of a direct transformation of smooth muscle cell into fat cells. In contrast, Akyildiz et al. reported a case of pure uterine...
lipoma in which fat cells were positive for S-100 and negative for SMA.\[7\] In our case, mature adipocytes from both the tumors showed immunoreactivity with vimentin but not with SMA. Similar observations were made in a study conducted by Sharma and Mandal.\[14\] Cytogenetic studies of uterine lipoleiomyoma suggested that they have pathogenetic origin similar to that of a typical leiomyoma.\[14\]

Some researchers have emphasized that hyperestrogenic state in various metabolic disorders such as hyperlipidemia, hypothyroidism, diabetes mellitus, postmenopausal lipid metabolism changes, and toxemia during pregnancy contribute to its development.\[10,14\]

In our case study, the patient was a 43-year-old female with no significant medical history and laboratory data suggesting metabolic disorders. Coexistent gynecologic malignancy has been reported in few studies.\[10,13\] In our patient, total hysterectomy was done, and there was no evidence of gynecological malignancy. In view of complex histogenesis and association of gynecological malignancies and metabolic disorders in patients of uterine lipoleiomyoma, more large number case studies with IHC findings and follow-up are required to understand histogenesis. Follow-up of our patient revealed no medical complaints since 1 year.

**Conclusion**

Lipoleiomyoma is a rare and commonly occurs in uterine corpus. Pure uterine lipoma is extremely rare. Although MRI can be a useful modality for preoperative diagnosis, most cases are diagnosed postoperatively on histopathology examination. These are benign neoplasms with excellent prognosis. Histogenesis is controversial. Coexistent gynecological malignancies and metabolic disorders in lipomatous uterine tumors warrant detail clinical, pathological evaluation, and follow-up.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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