Gigantic Lipoleiomyoma of Cervix with Extensive Hyaline Degeneration: A Case Report with Review of Literature

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ABSTRACT

Uterine lipoleiomyomas are rare benign neoplasms of mature smooth muscle and adipose tissue. Although mostly located intramurally within uterine corpus; cervix is rather an unusual site of its origin. Premenopausal or postmenopausal obese ladies commonly suffer with this tumour. Lipoleiomyomas generally grow asymptotically, but larger tumours often elicit symptoms due to pressure effects exerted on adjacent organs. We highlight a giant cervical subserosal lipoleiomyoma in a 48-years-aged menopausal female. She presented with ill-defined lower abdominal pain. Radiologically a circumscribed genital mass was detected, which was removed with total abdominal hysterectomy plus bilateral adnexitomy.

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Introduction
Lipoleiomyoma, an uncommon benign mesenchymal neoplasm, represents 0.03-2.9% of all leiomyomas arising at female genital organs.[1-5] Uterine corpus is its usual site of origin. But, around 6.5-14% of lipoleiomyomas originate in cervix. Broad ligament, peritoneum and ovary are the other secluded locations.[3-5] It is mostly sited intramurally while about 13-20% tumours grow subserosally. Asymptomatic in its evolution, lipoleiomyomas are generally discovered incidentally on surgical specimens.[4,5]

Mean diameter of lipoleiomyomas range from 4.6-5.5 cm.[3,4] Obesity and peri- or postmenopausal age-groups have highest association with this neoplasm. Structurally, it is characterized by an implicit cohabitation of mature smooth muscle fibres and adipose tissue in variable proportions.[6] Compared to pure leiomyomas, degenerative changes rarely appear in lipoleiomyomas.[4]

This report explores a huge subserosal lipoleiomyoma with extensive hyaline degeneration, originating from the cervix of a 48-years-old postmenopausal female.

Case Report
A 48-years-aged, P2+0, three-years-postmenopausal lady attended the gynaecologic out-patients clinic with vague, poorly-localized, lower abdominal pain for past four months. She did not complain about any definite gastrointestinal or genitourinary symptomatic. Even her per-vaginal and per-rectal examinations proved to be disappointing as well.

Next step she underwent abdomino-pelvic ultrasound; which identified a massive heteroechoic mass involving her genital organs, measuring approximately 18×17 cm. Sonographic discrimination between uterine or salpingo-oophoritic echotexture was impossible. Under this obscure clinico-radiologic circumstance, the lady was scheduled for total abdominal hysterectomy with bilateral salpingo-oophorectomy and accordingly the tumour was en masse despatched for histopathological examination.

Grossly a circumscribed, gigantic, multilobulated, subserosal neoplastic mass; spanning 19×18×16 cm, was seen to be circumferentially emanating from cervix; and extended upwards to squeeze the corpus in between tumoural lobules. Its excised surface appeared solid-homogeneous, greyish-white, punctated with pale-yellowish foci. Haemorrhage, necrosis, or calcification was absent. Morphologically, rest of the genitalia maintained its normal architecture (Figure 1). Provisionally, a diagnosis of ‘leiomyoma’ was strongly deliberated.

Microscopically, the tumour was well-delineated and pseudo-encapsulated. It was comprised of uniform spindle-shaped, mature smooth muscle cells arranged in intersecting fascicles; with interspersed benign adipocytes in small groups and singles. The myocytes featured monomorphic elongate cigar-shaped nuclei, fine chromatin, inconspicuous nucleoli and moderate-to-abundant amount of eosinophilic fibrillary cytoplasm (Figure 2). The myogenic component was often replaced by diffuse extracellular collagenous deposits, with minimal alteration in the adipocytic topography (Figure 3). Exuberant cytological atypia or mitosis, necrosis and calcification were imperceptible. Ultimately, the tumour was confidently diagnosed as ‘cervical lipoleiomyoma with extensive hyaline degeneration’. Following an uneventful postoperative outcome, the patient recovered satisfactorily during next six months’ follow-up.

Discussion
Lipoleiomyoma is an extremely uncommon uterine fatty tumour, regarded as a histologic subtype of leiomyoma. Collectively it constitutes less than three per cent of all uterine leiomyomas.[2-5] That is why its true incidence among general population is yet elusive.[5] Among the handful of case series published so far, Akbulut et al.[4] examined maximum number of subjects i.e. 76 cases; followed by Wang et al.[3] (50 cases), Aung et al.[2] (17 cases) and Bolat et al.[5] (10 cases).

The histogenesis and nomenclature of ‘lipoleiomyoma’ has long been debated. Previous erroneous theorems...
Fig. 2: Cervical lipoleiomyoma: Microscopically, intersecting fascicles of smooth muscle cells with intervening adipocytes in singles as well as in groups [H&E, 100x]; [inset] The myocytes have uniform cigar-shaped nuclei, fine chromatin, inconspicuous nucleoli and moderate-to-abundant amount of eosinophilic fibrillar cytoplasm [H&E, 400x].

Fig. 3: Cervical lipoleiomyoma with hyaline degeneration: Replacement of the myogenic component by dense collagenous deposits [H&E, 100x].

like embryonic adipocyte misplacement, choristoma, hamartoma, or lipomatous degeneration, is currently obsolete. Nowadays, most researchers opine in favour of its metaplastic derivation from leiomyomas. Back in 1916, detailed histopathology of lipoleiomyoma was first highlighted by Lopstein. But, the term 'lipoleiomyoma' was popularized decades later, by Willen et al. and Pounder, when they classified the uterine fatty tumours into ‘lipoma’ and ‘mixed lipoma/leiomyoma’ i.e. lipoleiomyoma. Moreover, Havel et al. demonstrated definite monoclonality in these tumours. Altogether these evidences substantiate its true neoplastic nature.

The mean age of patients from the series of Aung et al., Wang et al. and Akbulut et al. were 59.9, 53.9 and 55.49 years respectively. But, rarely it has also been reported in premenopausal youngers. Lipoleiomyomas commonly arise intramurally within uterine corpus, while the submucosal origination is most unusual. Cervix has been the site of origin in 7/50, 1/10 and 5/76 cases from the respective observations by Wang et al., Bolat et al. and Akbulut et al. Other uncommon locations include broad ligament and retroperitoneum. The discussed 48-years-old lady presented with a cervical subserosal lipoleiomyoma, which radiologically simulated a genital mass without any indication towards exact site of origin.

Uterine lipoleiomyomas are generally asymptomatic, discovered incidentally on surgical specimens. Sometimes it evolve with pain, bleeding, mass, urinary frequency or a leiomyoma-like pressure-sensation. Accordingly, the discussed patient complained about abdomino-pelvic pain at presentation. The diameter of lipoleiomyomas hardly exceeds 6 cm. In this respect Kim et al. and Kalyankar et al. notified two exceptionally large tumours, measuring beyond 15 cm. Such an extraordinarily huge mass was again encountered in present case. Lipoleiomyomas usually present as solitary lesion, while multifocality being a rarity. Moreover cervical fibroids may often enclose the cervix, simultaneously compress and displace the corpus superiorly. Such cases symptomatize from mechanical pressure exerted on adjacent organs. Similarly, the lipoleiomyoma in discussed patient developed circumferentially from cervix, extended upwards to squeeze uterus and subsequently caused pain symptoms.

Microscopically, lipoleiomyomas harbour a variable admixture of muscular and fatty components. Akbulut et al. subjectively classified it into grades I-III; as higher grades correspond to adipocytic abundance. Likewise in present case, the lipomatous component was outnumbered by myogenic tissue, corresponding to grade I lipoleiomyoma. Normally necro-degenerative changes sparsely involve lipoleiomyomas. But similar to discussed case, Bolat et al. also detected prominent hyaline degeneration within three such lesions.

Immunohistochemical (IHC) expression of vimentin, desmin, S-100, ER, PR and Ki-67 in lipoleiomyomas, has been implicated by several researchers to support its metaplastic histogenesis. But diagnostic implementation of IHC, has no extra advantage over routine haematoxylin-eosin stained method. That’s for IHC was not considered in this reported case.

Histological differential diagnoses of lipoleiomyomas include: spindle cell lipoma, angiolipoma, angiomyolipoma,
and well-differentiated liposarcoma.\textsuperscript{4,5} However, the absence of lipoblasts, atypia, necrosis or mitotic activity; paucity of angiomatous elements; and its characteristic clinicopathological presentation virtually clinched the diagnosis of lipoleiomyoma in present case.

Conclusion
Cervical lipoleiomyoma with extensive hyaline degeneration is a rare benign entity. Perimenopausal and postmenopausal women are most commonly affected. Although usually asymptomatic, larger tumours frequently elicit pain symptoms. Moreover, huge size often creates confusion on ultrasonography regarding its source organ. However, this dilemma is easily settled intraoperatively and final diagnosis can be confidently rendered on the basis of histopathological examination alone.

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References