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Leiomyoadenomatoid Tumor Of Uterus: A Case Report Of A Rare Variant Of Adenomatoid Tumor

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Abstract:
Background: *Adenomatoid tumor is a benign tumor occurs in the fallopian tube and uterus in the females and epididymis, spermatic cord in males. Leiomyoadenomatid tumor is a rare variant of adenomatoid tumor and usually an incidental finding in uterus.*
Case report*: A 47-year-old postmenopausal woman presented with per vaginal spotting and underwent hysterectomy with bilateral salphingo-oopherectomy based on ultrasonographic finding of 5cm size solid mass in the left uterine wall. Gross diagnosis of sub serosal fibroid was made which turned out to be a leiomyoadenomatoid tumor of uterus microscopically and was confirmed by immunohistochemistry.*
Conclusion*: This rare variant of leiomyoadenomatoid tumor is reported here for highlighting the dual differentiation nature of the tumor and uncertainity of histiogenesis.*
Keywords*: Adenomatoid tumor, Uterus, Leiomyoma, Mesothelial tumor*

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I. INTRODUCTION

Adenomatoid tumors(AT) are benign neoplasms of mesothelial origin that occur more frequently in the genital tracts of both men and women. In uterus, AT are usually located in the subserosa of the cornual myometrium and presented as intramural mass or pedunculated fibroid depends on the growth pattern. Lesions are typically solitary, and shows interanastomosing pseudo glands or pseudo vascular spaces, and a smooth muscle component is often present. The prominence of smooth muscle component simulates a leiomyoma and the lesion is denoted as a "leiomyoadenomatoid tumor"[1]. These have also been described in extragenital sites such as liver, heart, adrenal gland, mediastinal lymph node, pleura, and pancreas[2]. Adenomatoid component of this tumor may resemble proliferating capillaries and lymphatic ducts, as well as metastatic adenocarcinomas imparting a diagnostic challenge for pathologists also[3]. The true incidence of UATs may be ~1%, which is the incidence reported in the recently available literature. We here in report this rare case of leiomyoadenomatoid tumor of uterus presenting as subserosal fibroid.

II. CASE DETAILS

A 47-year-old postmenopausal female patient presented to the gynaecology outpatient department with the complaint of per vaginal spotting for the past 2 days with no history of pain, passage of clots or white discharge per vaginum. Clinical examination revealed soft, non-tender abdomen, cervix pointing downwards, uterus anteverted, normal size, non-tender, mobile and bilateral fornices were free. Routine blood investigations were normal except for slightly reduced Hemoglobin(Hb). Pelvic ultrasound showed well defined heteroechoic lesion of size 5.2x5.1x3.3cms in left lateral wall of uterus and endometrial thickness was found to be 6.5mm. Based on clinical and radiological findings, the diagnosis was made as leiomyoma in the left lateral wall of the uterus,and hysterectomy with bilateral salphingo-oopherectomy was performed and the specimen was sent for histopathological examination.

On gross examination, uterus with cervix measuring 9.5x8x3cms with a pedunculated subserosal fibroid measuring 5x4.5x4cms in the left lateral wall of the uterus arising from the outer wall of the uterus

extend along left fallopian tube. Cut surface of the mass showed gray-white firm appearance that extends from corneal myometrium, serosa to the sersoal surface of fallopian tube(Figure.1a & 1b).

Gross appearance of endometrium, myometrium, cervix and ovaries with right side fallopian tube were unremarkable. Left sided fallopian tube lumen was patent and subserosal mass of uterus extends along the tubal serosal surface & distorting it.

Microscopic examinations of the mass showed interlacing fascicles of proliferation of benign spindle cells interspersed with many cystic spaces lined by flat to plump cells with eosinophilic cytoplasm. No abnormal mitotic activity or cytological atypia were seen and no necrosis was observed. Stroma shows infiltration of lymphocytes and plasma cells around blood vessels, cystic spaces and also within few cystic spaces. This unusual morphology of leiomyoma created confusion that either tumor had coexisting vascular and lymphatic proliferation or mesothelial origin of tumor with smooth muscle differentiation since tumor projects out and extend along serosal surface of fallopian tube.

So Immunohistochemical investigation was performed using CD34 as endothelial marker and calretinin as mesothelial marker. It revealed intense positivity for calretinin and negative for CD34 that confirmed the mesothelial origin of this tumor.(Figure 3a & 3b). The final histopathological diagnosis of leiomyoadenomatoid tumor arising from left cornual uterine serosa extending along fallopian tube surface was made.

III. DISCUSSION:

In 1916, Sakaguchi first proposed the term 'Adenomyomata' before Golden and Ash renamed it as 'Adenomatoid tumour' [4]. Adenomatoid tumor is a rare benign neoplasm arising from mesothelial cells and was described later by Masson and Evans in 1942[5]. According to study conducted by Nakayama H et al, the incidence of AT is 1.2% and were incidental findings in hysterectomy specimens of adult women[3]. AT can occur in organs close to mesothelium lined surfaces most commonly in genital tract of both the gender. Extra genital sites have also been reported particularly in the vicinity of serosal membranes, adrenal gland, lymphnode[6]. The patients age is ranged from 24 to 76years [7]. In uterus, AT is typically solitary, subserosal or myometrial masses often located near the cornu of uterus but sometimes diffuse, multifocal masses can also be seen[5]. The tumors are usually small in size <4cm, but sometimes can be large (>9cm) [8]. The tumor in our case is located in left lateral wall of uterine cornu and extend along the serosa of fallopian tube and size is >5cm.

Microscopically, the adenomatoid tumor can have adenoid, angiomatoid, solid and cystic growth patterns or a combination of more than one type [5]. Due to these varying architectural patterns, these tumors can be mistaken for hemangioma, lymphangioma and epitheloidhemangioendothelioma. In uterus hyperplastic smooth muscle component is there in the background of adenomatoid tumor.

'Leiomyoadenomatoid tumor'(LMAT), which is a morphological variant of adenomatoid tumor with prominent smooth muscle component, was first introduced by Epstein in 1992[9]. Sometimes extensive smooth muscle overgrowth may obscure the adenomatoid tumor resulting in misdiagnosis of leiomyoma with malignant infiltrates. In our case, we have predominant cystic pattern resembling lymphangioma among the proliferating smooth muscle elements. The presence of bland, flattened lining cells of cystic pattern argued against it being metastatic adenocarcinoma. In addition, the lining cells of cystic pattern showed plump nature at places again arouse the suspicion of mesothelial origin of this component rather than endothelial cells and so the possibility of leiomyoadenomatoid tumor to be ruled out. Immunohistochemistry have confirmed the mesothelial origin of this tumor in our case by positive staining with calretinin which was consistent with an adenomatoid tumor and negative staining with endothelial marker CD34.

Extensive literature search showed only 19 cases of leiomyoadenomatoid tumor of uterus were reported worldwide. In India only six cases have been reported by Rajan et al[5](3 cases), Mathew et al[10](1 case), Sarma et al[11] (1 case), Bahuguna et al [2](1 case) and ours is the seventh case of this rare tumor entity.

We concluded that the leiomyoadenomatoid tumor is an unusual morphological presentation of AT with benign tumor with excellent prognosis. Hence local resection is preferred for management. In no instances was there a recurrence or metastases reported [12].

The histiogenesis of this biphasic uterine tumor is still debatable whether originated from mesothelial cells with smooth muscle differentiation or from uterine smooth muscle with mesothelial differentiation[13].

IV. CONCLUSION

In summary, Leiomyoadenomatoidtumour is a rare variant of adenomatoid tumor arising from mesothelium with dual component of smooth muscle and cystic adenomatoid mesothelial elements. It grossly mimics as leiomyoma and microscopically as infiltrating malignancy into smooth muscle. This case report highlights the peculiar morphology and the possibility of this rare benign tumor of uterus which may be misinterpreted as malignancy. However, this dual differentiation in this benign tumor created confusion regarding the histiogenesis of this tumor. So detailed morphological findings, along with aid of immunohistochemistry, are useful to diagnose this rare separate entity from classical adenomatoid tumor.

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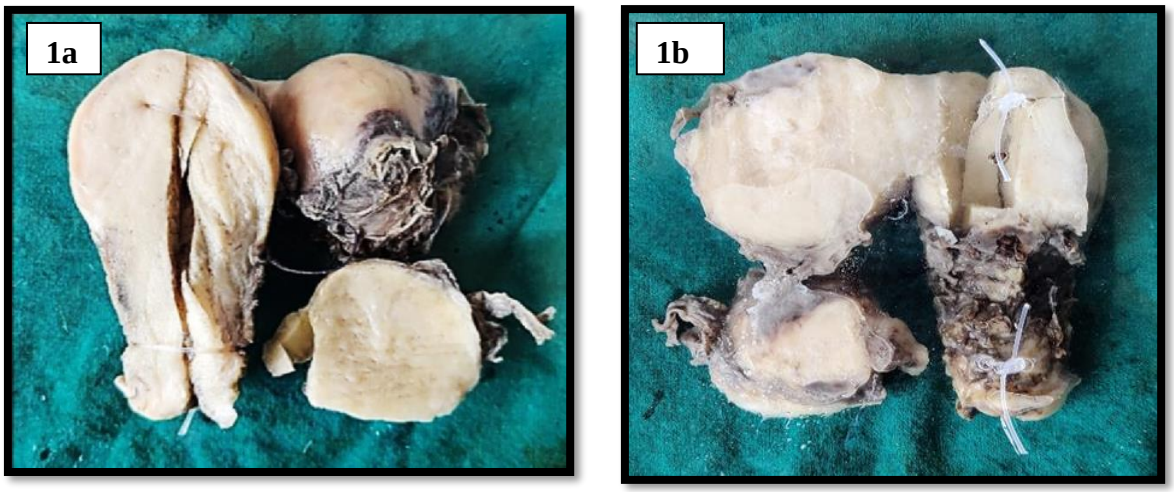
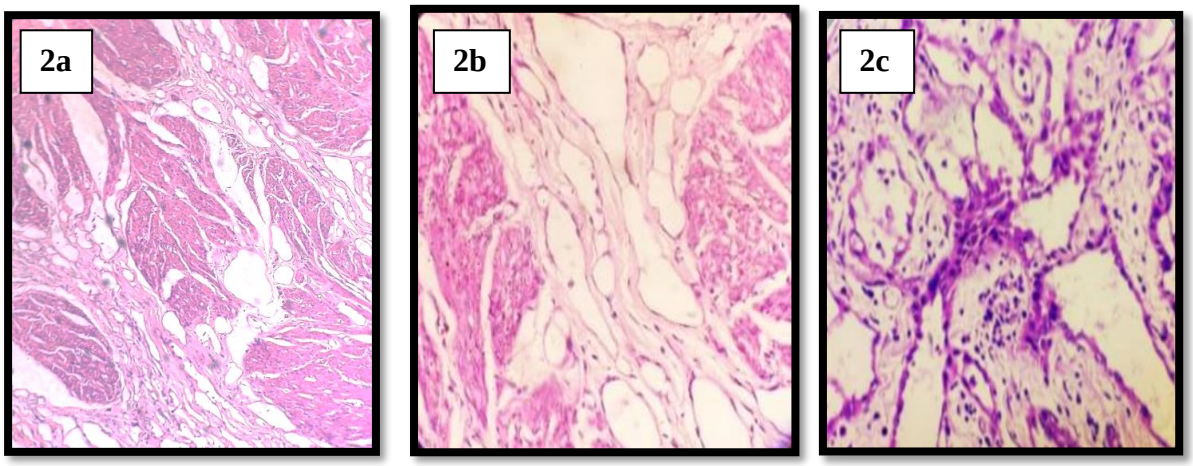
FIGURES:
Gross images:



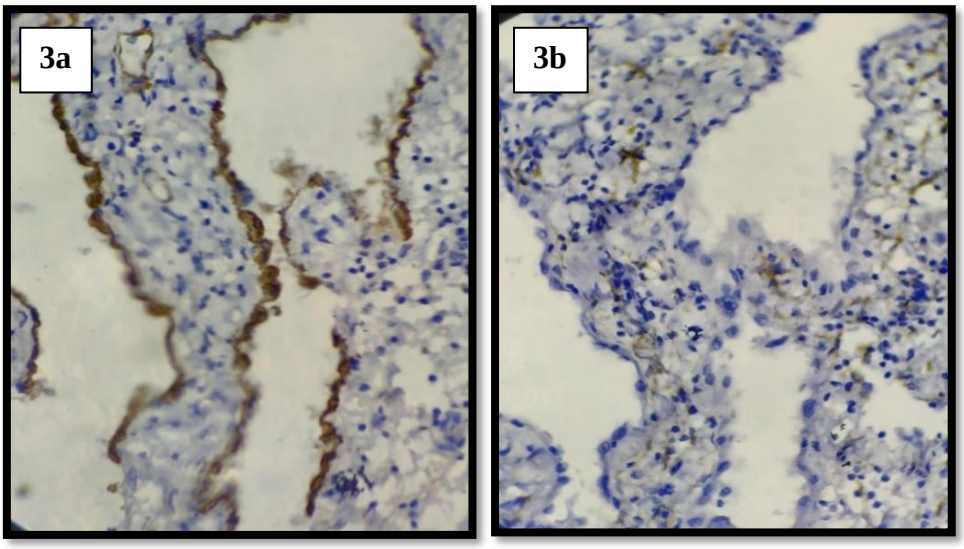
1. **Fig.1(a). Gross image of uterus with subserosal mass extending from corneal end of uterus to fallopian tube. Cut surface is solid as leiomyoma**
2. **Fig.1(b). Posterior aspect of uterus showing pedunculated subserosal fibroid in the left lateral wall of the uterus arising from the outer wall of the uterus extending along left fallopian tube**

Microscopic findings:



1. **Fig.2(a&b): Microphotograph showing dual differentiation of smooth muscle cells and cystic mesothelial elements (Haematoxylin & Eosin staining x 4(a) & x 100(b))**
2. **Fig.2(c): Microphotograph showing flat to plump mesothelial lining of cystic components. (Haematoxylin & Eosin staining x 400)**

Immunohistochemistry study:



1. **Fig.3(a): Microphotograph showing intense positive staining for Calretinin by cells lining cystic component of tumor(Immunohistochemistry x 400)**
2. **Fig.3(b): Microphotograph showing weak/absence staining for CD34 by cells lining the cystic component of tumor(Immunohistochemistry x 400)**