

Large Broad ligament lipoleiomyoma masquerading as an ovarian malignancy: an uncommon tumor in an unusual site: a case report

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Abstract: Lipoleiomyoma is a very uncommon tumor accounting for 0.3 to 2.1% of all uterine leiomyomas and is composed of adipocytes and smooth muscle cells. They are mostly found in the fundus or cervix and extremely rare in the broad ligament. Preoperative diagnosis of lipoleiomyoma of the broad ligament is very difficult due to its similarity to ovarian tumor. I report a case of a 45-year-old premenopausal woman with huge lipoleiomyoma of the broad ligament mimicking an ovarian tumor. I present this case because of its rarity and deceptive clinical presentation along with confusing histopathology.

Keywords: Lipoleiomyoma, broad ligament, ovarian malignancy.

INTRODUCTION

Lipoleiomyoma is a very uncommon tumor accounting for 0.3 to 2.1% of all uterine leiomyomas and is composed of adipocytes and smooth muscle cells [1]. It is most commonly located in uterine corpus although cervical, ovarian, and retroperitoneal locations are also described [1]. Broad ligament lipoleiomyoma is even rarer with few cases (10 cases)(Table1) reported in English literature [1-9]. These tumors may be confused with other conditions such as mature ovarian teratoma, well-differentiated liposarcoma and angiomyolipoma.

I present this case because of its rarity and deceptive clinical presentation along with confusing histopathology.

CASE REPORT

A 45 year old female presented with mass in abdomen and decreased appetite since 2 weeks. USG revealed an abdominopelvic mass with cystic and solid components, multiseptations, internal vascularity and low resistance flow. CT scan revealed a large pelviabdominal cystic mass of size 23x17x17 cm with few solid areas and internal septations within. Post contrast the lesion showed heterogenous enhancement. Tumor biomarkers were within normal limits (CA-125:11.8 U/mL, CEA: 1.96ng/mL, LDH: 600 U/L,AFP:5.42IU/ml,BETA HCG:2IU/L).An ovarian neoplasm was suspected and total abdominal hysterectomy with bilateral salpingoopherectomy was done and sent for frozen section analysis to exclude malignancy.

Table-1: Summary of all the cases of broad ligament lipoleiomyoma reported in English literature till date

	Year of publication	Age in years	Modality of treatment	Tumor size (in cms)	Laterality and location
Case 1	1989	NA	NA	NA	BL
Case 2	2000	43	TAH+BSO with resection of tumor	25	Left, BL
Case 3	2002	68	TAH+BSO	40	BL
Case 4	2002	57	TAH+BSO	8	BL
Case 5	2006	54	NA	4.6	BL
Case 6	2009	63	Laparotomy +en-block excision	20	Right ,BL
Case 7	2010	54	TAH+BSO	8	Left, BL
Case 8	2012	56	TAH+BSO	19	Left, BL
Case 9	2014	55.5	NA	5.5	BL
Case 10	2015	45	TAH+BSO with resection of tumor	17	Left, BL
Present case	2016	45	TAH+BSO	18.5	Left, BL

NA: not available, TAH+BSO: Total abdominal hysterectomy and bilateral salpingoopherectomy, BL:broad ligament



Fig-1:A: Gross examination revealed a large circumscribed mass with a smooth external surface attached to the left parametrium measuring 18.5x 17x 6 cm;
B: Cut section of the mass was solid with cystic areas, yellow white in appearance

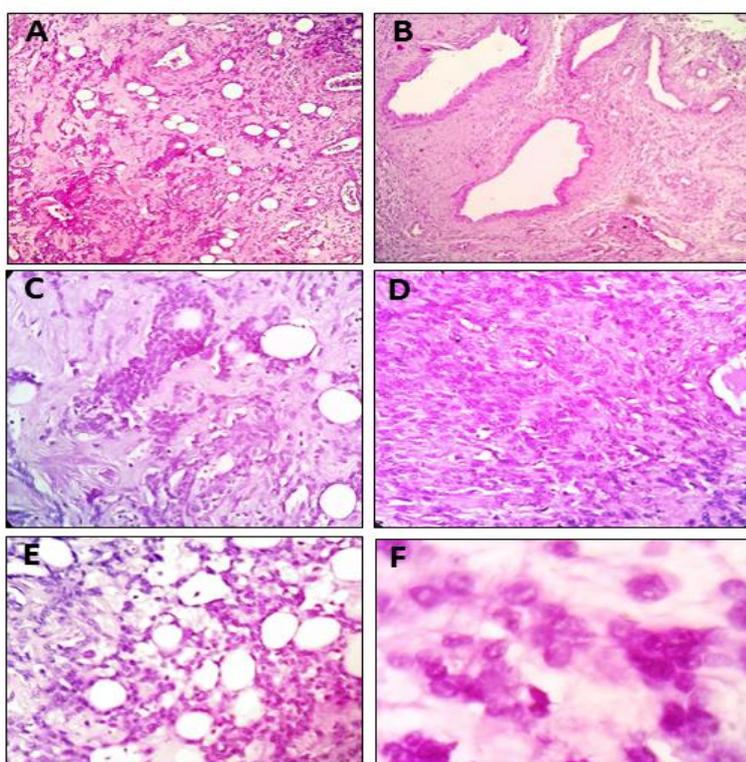


Fig-2: A:ovoid cells arranged in cords and sheets with interspersed fat , H&E, 40X;
B:thick walled vessels a prominent component, H&E, 100X;
C:a higher magnification of the tumor cells along with adipose tissue in a hyalinised stroma, H&E, 100X;
D:Diffuse sheets of spindle cells, H&E, 100X;
E:an area where adipose tissue is prominent , H&E, 100X;
F:cells are uniform with ovoid nuclei with evenly dispersed chromatin and small conspicuous nucleoli,H&E,1000X

Gross examination (Figure 1) revealed a large circumscribed mass with a smooth external surface attached to the left broad ligament measuring 18.5x 17x 6 cm. Both the ovaries were identified separate to the mass with no involvement. Cut section of the mass was solid with microcystic areas filled with myxoid viscous fluid. The uterus, cervix and bilateral ovaries were within normal limits. Microscopic examination

(Figure2:A-F) revealed a well circumscribed tumor with a heterogenous morphology composed of spindle to epithelioid cells arranged in cords, nests and diffuse sheets in a fibromyxoid stroma exhibiting alternate hypercellular and hypocellular areas. At places the stroma was oedematous forming microcystic pattern. The tumor had a rich vasculature composed of vessels of varying calibre scattered throughout with some areas

showing perivascular cuffing. Mature adipocytes were seen throughout the tumor. Bundles of smooth muscle fibres were seen (Figure3). No cellular pleomorphism, nuclear atypia or mitotic activity was seen. Necrosis was absent. Considering the morphology, with the tumor showing muscle, fat as well as rich vasculature, differential diagnosis considered was lipoleiomyoma and angiomyolipoma. IHC markers HMB 45 and Melan A were performed, both of which were negative.

The challenges in this case was the location, the size, the angiomyolipoma like vessels, the microcystic areas, the epithelioid looking cells arranged in cords, making diagnosis difficult. Finally, however the definite presence of thick bundles of smooth muscle lining the mass and the prominent adipocytes combined with negative HMB45 and Melan A helped in rendering a diagnosis of lipoleiomyoma.

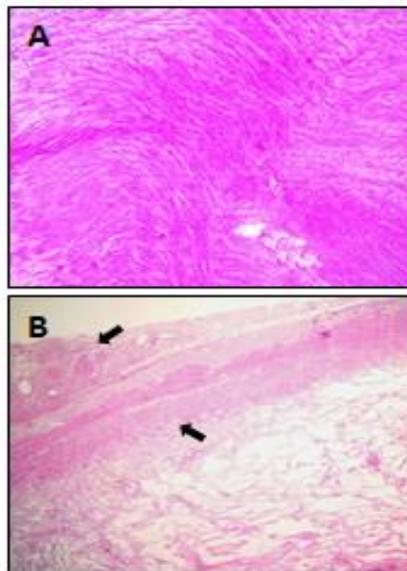


Fig-3A: thick bundles of smooth muscles, H&E, 100X;
B: smooth muscles at the periphery of the tumor (marked by arrows), H&E,100X

DISCUSSION

Lipomatous tumors of the uterus can be divided into three groups – pure lipomas, which are composed of encapsulated mature fat cells, lipomas with several mesodermal components (lipoleiomyomas, angiomyolipomas, and fibromyolipomas), and the exceptional malignant liposarcoma [10]. Lipoleiomyomas were first described by Lobstein in 1816 [11]. It was formerly designated with different names like fatty metamorphosis, lipomatous degeneration, adipose metaplasia, etc but is now considered as a true benign neoplasm [11]. Although the pathogenesis of the lipoleiomyoma is ambiguous, immunohistochemical studies reveal that lipoleiomyoma may arise from metaplasia of pluripotent mesenchymal cells or direct metaplasia of smooth muscle cells [7].

Compared to usual leiomyomas which tend to occur predominantly in women of reproductive age and regress after menopause, the lipoleiomyomas are commonly seen in older women with mean age of the patients in the largest two series so far being 55.4 years [2,12]. In our case, the patient was premenopausal. Most patients are asymptomatic and are detected incidentally, but amid symptomatic ones- pelvic pain, palpable mass or abnormal bleeding are the most frequent

symptoms.[1] Our case presented with a large mass in abdomen and an ovarian malignancy was primarily suspected. The presence of a pelvic mass in a woman is certainly not sufficient to consider ovarian malignancy particularly if it is not associated with ascites and elevated tumor markers. However, the heterogenous solid cystic nature of the lesion on imaging and its adnexal localization as in our case may suggest a malignant ovarian neoplasm that definitely requires surgical exploration with frozen section analysis where available.

Lipoleiomyomas range from 0.5 to 55 cm in size, with a mean of 5.50 cm with only few reported cases of large lipoleiomyomas [8]. Histologically, lipoleiomyoma are composed of an intimate admixture of mature smooth muscle cells and adipocytes, often with the former element predominant.[8] In the present case, there was an amalgamation of many elements- bundles of smooth muscle, adipocytes, epithelioid looking cells as well as abundant vessels. Epithelioid morphology has been reported previously in literature, however unlike the previously reported cases, in our case the cells were devoid of any nuclear atypia [13, 14]. Rare lipoleiomyoma with renal angiomyolipoma-like vascular proliferation have been reported, similar to our case [8, 12].

Although the pathogenesis of uterine lipoleiomyomas is still indefinite, it is known that some metabolic disorders, including hyperlipidemia, hypothyroidism, and diabetes mellitus are associated with lipoleiomyomas [8, 15].

The long-term follow-up of patients with uterine lipoleiomyoma verified that these lesions are benign without any recurrences or disease-related deaths [2].

CONCLUSION

Recognition of this rare tumor especially in an unusual location like broad ligament is of great importance, because it may be confused clinically with an ovarian malignancy and may necessitate surgical intervention not only due to associated symptoms but also to be able to exclude an ovarian malignancy. Histologically it may mimic angiomyolipoma, spindle cell lipoma, angioliipoma, well-differentiated liposarcoma, ovarian teratoma, and benign or malignant degeneration of ordinary leiomyomas so a careful and meticulous examination is essential.

Abbreviations:

USG: ultrasound
CT: computed tomography
CA-125: cancer antigen-125
CEA: carcinoembryonic antigen
LDH: lactate dehydrogenase
AFP: alpha fetoprotein
BETA HCG: beta human chorionic gonadotrophin
IHC: immunohistochemistry
HMB 45: human melanoma black 45
Melan A: melanoma antigen

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