Spermatic cord dedifferentiated liposarcoma: Review of literature and case report
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Abstract
Spermatic cord malignancies are a scarce modality and liposarcoma of spermatic cord is even a rarer condition encountered. Liposarcoma is usually a slowly progressive, non-tender, well circumscribed mass of variable shapes owing to conformity to fascial compartments. We are reporting a case of 65-year-old male, with a two-month history of initially tender and later non-tender mass in the scrotum, above the testis. Ultrasonography showed a right mid inguinal mass measuring 6x3x3 cm at the superior pole of the right testis and small fluid around the right testis. Fine needle aspiration cytology (FNAC) of the right inguinal mass revealed a spindle cell neoplasm. The patient underwent right inguinal radical orchiectomy with local wide excision of the sarcoma of the spermatic cord origin. Final histopathology confirmed dedifferentiated liposarcoma. No adjuvant treatment was offered and the patient was put on surveillance. Follow-up of more than 10 months has not revealed any local recurrence, regional or non-regional lymph nodes, or systemic metastasis.

Keywords: Liposarcoma testes, Spermatic cord tumours, Testis neoplasms, Testicular sarcoma.

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Introduction
Liposarcoma of spermatic cord is a fairly uncommon malignancy. Only 7% to 10% of intrascrotal cancers are primary para-testicular tumours. The paratesticular liposarcoma (LPS), leiomyosarcoma (LMS), and rhabdomyosarcoma (RMS) are histological subgroups of spermatic cord malignant tumours.¹ In adulthood, the spermatic cord is the site of more than 75% of these lesions, 20% of which are liposarcomas. The median age of diagnoses is 68 years (between 58–78).² Paratesticular liposarcomas have four major histological subtypes: dedifferentiated, pleomorphic, myxoid or round cell, and well-differentiated. The three most common locations of involvement are the lower limbs (41%), the retroperitoneal areas/para-aortic areas (19%), and the inguino-femoral regions (12%).

Liposarcoma usually appears as slow-growing, non-tender, well-circumscribed, palpable mass as large as 10 cm in diameter. Some patients who have retroperitoneal disease may end-up with diffuse abdominal involvement. Liposarcomas may sometimes have very awkward fusiform or disc like shapes due to conformity to the fascial compartments. Independent predictors of prognosis include lymph node involvement, histologic type, tumour grade, and stage. The type of surgical resection affects the local outcome, irrespective of the subtype of liposarcoma and negative surgical margin is directly associated with increased disease specific survival.³ The best treatment is a radical orchiectomy. So far, no accepted adjuvant therapy has been established.²

We present the diagnostic workup, treatment, and follow-up of a case of Dedifferentiated Liposarcoma originating from the spermatic cord in a 65-year-old male patient to help identify this highly unusual disease and its follow-up.

Case Report
A 65-year-old male, married and having four children, known diabetic and hypertensive, presented to the oncology out-patient department (OPD) of Combined Military Hospital (CMH) Rawalpindi on February 02, 2023, with the complaint of a hard mass in the right hemiscrotum since two months whose size was gradually increasing. Initially, the patient had a mild pain on touch which later settled and became painless. There was no history of trauma to scrotal area, repeated urinary tract infections, any sexually transmitted diseases, undescended testis, or any scrotal/groin surgical intervention. The patient had no family history of cancers. On clinical examination, the abdomen was non-tender and soft, and both the testes were normal. There was an approximately 3x4 cm firm, non-tender, well-circumscribed, mobile right scrotal mass which seemed to be abutting or originating from spermatic cord. Ultrasound doppler of the right groin area disclosed an ovoid, supra-testicular echogenic focal lesion with coarse internal echoes in the right mid-inguinal region.
along the right spermatic cord with volume 20-25 ml (Figure). A cyst measuring 10x9 mm could be seen at superior pole of the right testis and small amount of fluid around the right testis—epididymal cyst.

FNAC of the right inguinal mass revealed it to be a Spindle cell neoplasm. Metastatic workup done with CT scan of the chest, abdomen and pelvis with contrast enhancement was negative. The patient underwent right radical orchiectomy with excision of the sarcoma of the cord. Histopathology report showed a specimen measuring 15x5x3.5 cm, while the testis was normal. On the proximal end of the specimen there was a mass of 9x3.5x3 cm which was 2.5 cm from the resection margin of the cord. Conventional Leiomyosarcoma grade II, with a mitotic rate of 15 per 10 hpf (high power field), lymphovascular space invasion was not seen, while there was necrosis in individual cells. Spermatic cord margin was negative for sarcoma. Distance of spermatic cord margin from tumour was 2.5 cm.

Immunohistochemistry showed CDK4 and P16 positive. MDM-2 gene amplification by fluorescence in situ hybridisation (FISH) was requested and it was amplified. So it was labelled as De-differentiated Liposarcoma. The patient was not offered any adjuvant chemotherapy or radiotherapy and was put on follow-up. Follow-up surveillance after three months of surgery with contrast enhanced CT scan of the chest, abdomen, and pelvis did not show any local recurrence, loco-regional lymph nodes, or visceral metastasis. Follow-up ultrasonography of the scrotum, abdomen and pelvis after four months of surgery was unremarkable. The patient is well after more than 10 months of follow-up.

**Discussion**

Liposarcoma of spermatic cord is an uncommon aggressive soft tissue cancer with a dismal prognosis. It originates from mesenchymal tissue and can be seen in all of the body’s fat cells. Sarcomas make up the majority of malignant paratesticular tumours, whereas 5–7% are liposarcomas. Patients of all ages can develop primary spermatic cord tumours, which are uncommon but clinically severe urologic diseases. They are the most prevalent paratesticular tumours and often manifest as hard, palpable, slow-growing paratesticular masses. Even though lipomas make up the majority of spermatic cord tumours and are therefore benign, 25% are potentially fatal malignant tumours. Liposarcomas, leiomyosarcomas, rhabdomyosarcomas, malignant fibrous histiocytomas, and fibrosarcomas are the most often reported malignant histological forms. There may be some uncommon histopathological features such as a combination of major pleomorphic, minor sclerosing, and well-differentiated components (a mixed type liposarcoma).

Liposarcomas typically have a capability of local spread. It sometimes involves the para-aortic nodes and can metastasise. When pre-operatively suspected or diagnosed, radical surgery with wide local excision (orchiectomy) and proximal high spermatic cord ligation is recommended as was done in this case. The benefit of adjuvant chemotherapy is not proven and there is no consensus.

Several writers have suggested combining surgery and radiotherapy, particularly for round cell or pleomorphic liposarcoma subtypes and in circumstances where the margins are ambiguous as a result of partial tumour removal. This patient had radical orchiectomy and high proximal spermatic cord ligation. Although the tumour was de-differentiated, the margins were clear on histology, without any loco-regional nodal involvement or metastasis,
so no adjuvant radiotherapy was offered. Adjuvant chemotherapy was not offered in this case although some studies suggest Doxorubicin based chemotherapy to improve recurrence-free survival in all patients and improved overall survival only in adult males. Due to high propensity of loco-regional recurrence of liposarcoma (55 to 70%), a long-term follow-up is obligatory.

Conclusion

Liposarcoma of spermatic cord is a very rare condition but the suspicion of liposarcoma should be kept in mind as a differential in patients having recurrent inguinal hernias. Awareness among surgeons is necessary to keep this differential in mind. The rarity of disease results in difficulty in diagnosis and poses a challenge to treat. Radical Orchiectomy with a higher proximal ligation of spermatic cord is mandated. Efficacy of the adjuvant local and systemic therapies has not been established. It is necessary to have routine post-operative follow-ups to check for recurrences. Optimal management can be sorted out by the help of further studies.

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References