IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 22, Issue 6 Ser.12 (June. 2023), PP 21-23
www.iosrjournals.org

A Rare Case Of Primary Testicular Leiomyosarcoma

Dr. Meethu John[1], Dr. Veena R[2], Dr. Tejaswini B.N[2], Dr. Sowmya. B. U[2]
[1]Resident Pathology, [2]Consultant Pathologists, HCG Cancer Hospital, Bengaluru.

Abstract
Primary Testicular Leiomyosarcoma Is A Rare Entity, Arising Secondarily Following Exposure Of Radiotherapy, Use Of Anabolic Corticosteroids And Testicular Germ Cell Tumor. Herein We Present A Case In An Elderly Male Who Presented With Left Sided Scrotal Swelling. He Underwent Left High Inguinal Orchidectomy And Histopathologic And Immunostaining Confirmed Primary Leiomyosarcoma.

Keywords : *Testis ,Immunohistochemistry, Leiomyosarcoma, Orchidectomy*

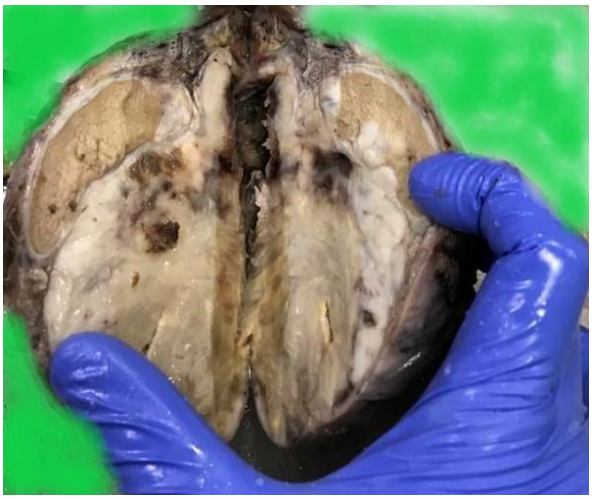
Date of Submission: 15-06-2023 Date of Acceptance: 25-06-2023

I. Introduction

Primary testicular leiomyosarcoma is very rare with only few cases being reported till date. They differ from more common sarcomas of spermatic cord as well as from germ cell tumors. We are presenting an extremely rare occurrence of high grade primary testicular leiomyosarcoma in an elderly male with absence of any predisposing features for this rare malignancy.

II. Case Report

A 60year old man presented with diffuse left sided scrotal swelling since four months. Imaging showed a large lobulated necrotic peripherally enhancing mass lesion suggestive of ? abscess involving testis / ?neoplastic mass with necrosis. Serum tumor markers were normal. High inguinal orchidectomy was performed. Intra-operative findings were of infective necrosis. On gross examination, the testis was enlarged measuring 9.5x6x4.5cm. Cut surface showed a grey white firm lesion with well-defined pushing margins within the testicular parenchyma measuring 9.5x6x4.5cm (Fig 1.). The epididymis and spermatic cord were grossly normal. Microscopy showed a malignant neoplasm formed of fascicles of spindle shaped cells with elongated fusiform nucleus, nuclear hyperchromasia and moderate amount of cytoplasm (fig 2.).



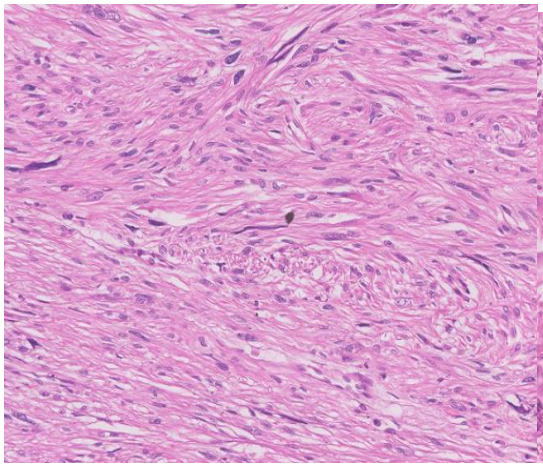


Fig 2.

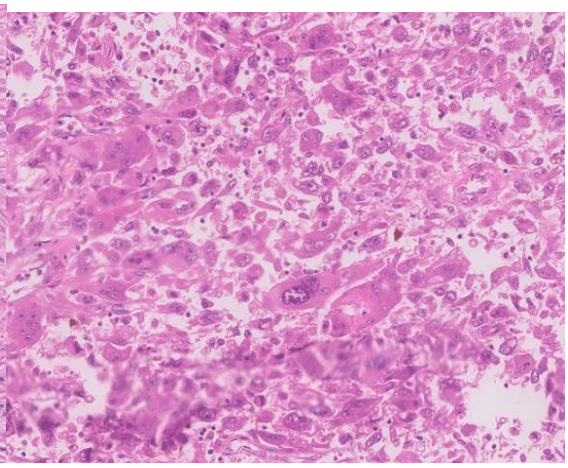


Fig 3.

Many scattered bizarre mononuclear and multinucleated giant cells were seen with a few osteoclastic giant cells (Fig. 3). The stroma was collagenous. There were 12-13 mitosis / 10HPF . Necrosis was seen. The lesion had well circumscribed margins and was seen infiltrating testicular parenchyma. Many plump cells of epithelioid morphology were also seen. No lymphovascular invasion / perineural invasion was noted. Adjacent epididymis and spermatic cord was free of tumor. The resection margin of spermatic cord was unremarkable. Morphological features suggested a diagnosis of High Grade Malignant tumor with Biphasic morphology. Two possibilities were considered: Leiomyosarcoma and Mesothelioma. Immunostaining showed diffuse positivity for SMA, Calponin and H-Caldesmon with focal positivity for panCK. The tumor cells were negative for Calretinin, Inhibin, S100 and CD34. A diagnosis of Primary High grade Leiomyosarcoma of Testis was made.

III. Discussion

Leiomyosarcoma is a rare condition not often encountered in the Genito-urinary system. When presented, most of them arise from the soft tissue of the spermatic cord and the rest originate from the epididymis or dartos muscle of the scrotum (3). Although approximately 100 Para testicular leiomyosarcoma have been reported in the literature (4), fewer than 10 cases of primary leiomyosarcoma of the testis have been mentioned (2, 5-10).

Primary leiomyosarcoma of the testis mostly occurs in young men in whom there is an associated history of high-doses of anabolic steroids (5) or chronic inflammation (6).

The clinical presentation of this tumor is similar to other testicular malignancies. Testicular leiomyosarcoma is diagnosed as a local disease probably due to the fact that it is easily recognized and tends to be slow-growing (10)

Leiomyosarcoma is a soft-tissue tumor arising from smooth muscle cells of mesenchymal origin. Its origin is from contractile cells in the tunica propria of the seminiferous tubules, the muscular layer of blood vessels, and smooth muscle elements in the tunica albuginea (2). The hormonal stimulation of the proliferation of smooth muscle cells is suggested to have a role in the carcinogenesis of leiomyosarcoma (5).

The clinical and biological behavior of these tumors is unpredictable ; however, the high mitotic activity is considered an important criteria for malignancy. Histologically, this type of tumor appears as malignant smooth muscle spindle cells with atypical nuclei that selectively stain with antibodies to smooth muscle actin, Calponin, H-Caldesmon, Desmin and vimentin, but not to S100, CD34, CD-68 and HMB-45.

As the number of reported cases is not significant, standard therapy for this tumor is difficult to recommend. Radical orchidectomy followed by close follow up is the treatment of choice for primary leiomyosarcoma of the testis. Retroperitoneal lymphadenectomy is not recommended since even in the presence of widespread metastatic disease (1). In addition, leiomyosarcomas seem to be radio-resistant as well as chemo-resistant(3)

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