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Inflammatory myofibroblastic tumor of spermatic cord

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ABSTRACT

INTRODUCTION: Inflammatory myofibroblastic tumor (IMT) is a neoplasm of intermediate biologic potential. Only a few cases of IMT in the spermatic cord have been reported. It was earlier included in a wide spectrum of reactive and neoplastic lesions called "inflammatory pseudotumors". It commonly presents as a painless scrotal mass, usually in children and young adults.
PRESENTATION OF CASE: We present a case of IMT in the spermatic cord who based on clinical, radiological and cytological findings underwent surgical exploration of left scrotal sac. The mass was separate from the left testis and left epididymis, and was closely adherent to pampiniform plexus of veins. Wide excision of the mass was done. Histology and immunohistochemistry suggested IMT.
DISCUSSION: IMT is a myofibroblastic spindle cell proliferation with chronic inflammatory infiltrate. Surgical exploration is essential as clinically and radiologically benign or malignant nature of mass cannot be distinguished. The diagnosis of IMT is based on the histological features and is substantiated by immunomarkers.
CONCLUSION: In clinically distinct masses, based on frozen section, either tumor excision or radical orchidectomy can be performed. The prognosis is excellent after complete surgical excision of spermatic cord IMT. Careful long-term follow-up is essential, because of the possibility of recurrence, though rare in this site.

1. Introduction

Inflammatory myofibroblastic tumor (IMT) is a neoplasm of intermediate biologic potential.[1] IMT was formerly included in a wide spectrum of reactive and neoplastic lesions known as "inflammatory pseudotumors". They have also been described in the literature under names such as atypical myofibroblastic tumor, pseudosarcomatous myofibroblastic proliferation, pseudosarcoma, plasma cell granuloma, and proliferative funiculitis. Over the last two decades, IMT came to be known as a distinct tumor with its typical clinical, pathological and molecular features. Inflammatory pseudotumor was first described in the lung where it was hypothesized to be a reparative postinflammatory process.[2,3] Its exact etiology still remains unknown. Subsequently similar lesions were reported from extrapulmonary sites also. Accurate data regarding the incidence, prevalence and anatomical distribution of IMT are difficult to obtain due to the use of the terms "inflammatory pseudotumor" and "IMT" interchangeably in the literature. The abdominopelvic region, lung, mediastinum, and retroperitoneum are frequent sites. Only a few cases of IMT in the spermatic cord have been reported.[4–6] We present the case of a young man with an IMT involving the spermatic cord, closely adherent to the pampiniform plexus of veins.

2. Presentation of case

A 22-year-old male presented with a painless left scrotal mass of 7 months duration. There was no history of trauma, fever, recurrent urinary tract or sexually transmitted infections and no past history of exposure to tuberculosis. Physical examination revealing a nontender, firm oval-shaped, 3 cm × 3 cm sized mass with nodular surface was palpable over the superior pole of left testis. It could be separated from left testis. Skin over the mass was normal. The mass was mobile in the inguinal canal but it was restricted in the subcutaneous plane, suggestive of adherence to the cord structures. Hydrocele excluded. There was no inguinal lymphadenopathy. The external genitalia including both testes were normal. Abdominal examination was normal.

Sonography of scrotum showed a solid, heterogeneous mass of size 3 cm × 3 cm with internal vascularity in the left scrotal sac, separate from left testis and left epididymis. The testes and epididymis were normal sized and there was no free fluid in right or left scrotal sac. Abdominal sonography was normal. Fine needle aspiration

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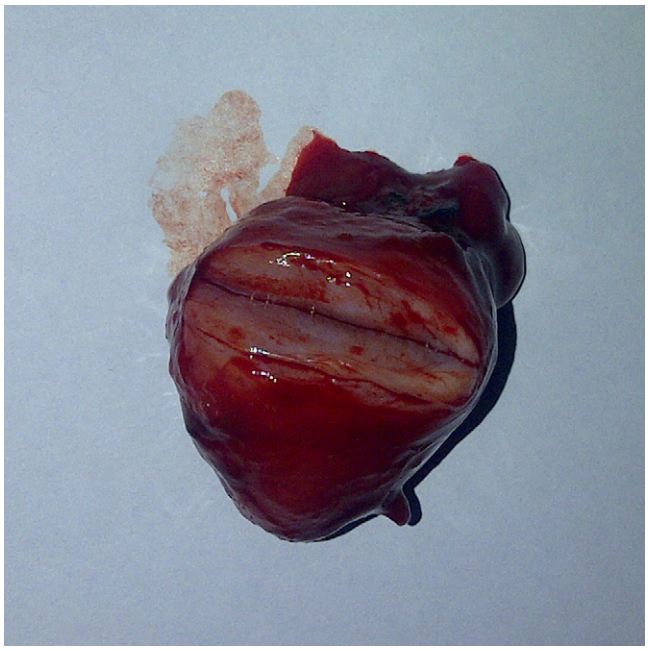

Fig. 1. Gross appearance of globular tumor of 4.5 cm × 3.5 cm × 2 cm size with brown, capsulated external surface and gray–white cut surface. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

cytology of the mass revealed clusters of oval to spindle shaped cells in a proteinaceous background.

Left scrotal exploration demonstrated the tumor, closely adherent to pampiniform plexus of veins. Intra-operative Doppler examination was done to identify the testicular artery. Testicular artery and vas deferens were isolated. Wide excision of the mass with surrounding cremaster muscle and a leash of veins which seemed to enter the mass were excised.

Macroscopically, the mass was globular, gray–brown, measuring 4.5 cm × 3.5 cm × 2 cm with capsulated external surface (Fig. 1). Its cut surface was gray–white, firm to hard, partially embedded. Microscopic examination revealed inflammatory cells, predominantly plasma cells and lymphocytes admixed with myofibroblasts and fibroblasts in a stroma of abundant hyalinised collagen (Fig. 2).

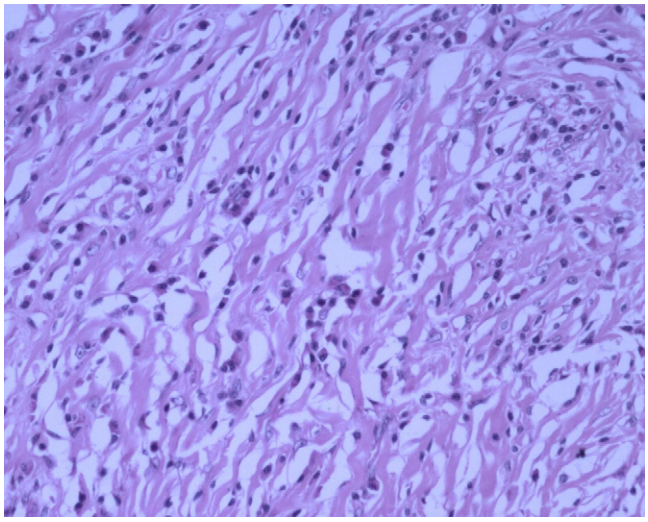

Fig. 2. Histologically tumor is composed of proliferation of spindle cells in abundant hyalinised collagen, admixed with inflammatory cells, predominantly plasma cells and lymphocytes. Hematoxylin–eosin stain, original magnification 200×.

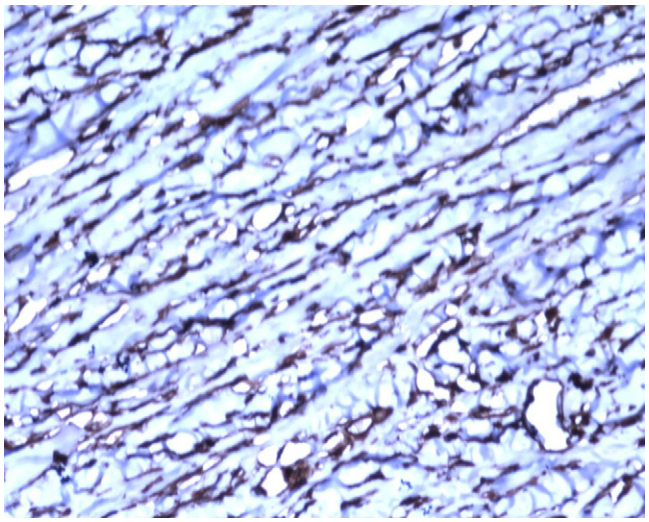

Fig. 3. (Immunostain) Immunostaining showing spindle myoepithelial cells positive for vimentin.

Focal dystrophic calcification was seen. There was no evidence of necrosis or atypia. Immunohistochemically, the tumor was strongly positive for vimentin (Fig. 3), and focally positive for CD34. Epithelial membrane antigen was positive in the plasma cells. Staining for anaplastic lymphoma kinase (ALK) (Fig. 4), smooth muscle actin (Fig. 5), desmin, S100 and cytokeratin were negative. The findings were consistent with an IMT arising from the spermatic cord. Serum IgG4 level was 1.49 g/L which is normal (reference: 0.03–2 g/L), thus excluding IgG4-related sclerosing disease.

3. Discussion

IMT is characteristically composed of spindle myoepithelial cell proliferation accompanied by inflammatory infiltrate of plasma cells, lymphocytes and eosinophils. It usually occurs in the soft tissues and viscera of children and young adults. However, proliferative funiculitis (this term was earlier used to denote an IMT involving the spermatic cord) preferentially affects men of middle to advanced age.[4–6] IMT may span the entire age range. The clinical presentation depends on the site of origin. In spermatic cord IMT, the presentation is usually a painless scrotal mass of variable duration.

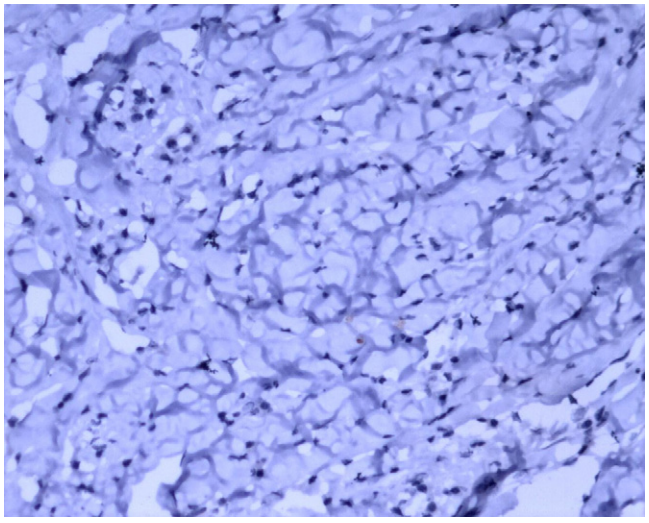

Fig. 4. (Immunostain) Immunostaining negative for ALK.