

Testicular fibroma: A case report and literature review

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Abstract

Sex cord-stromal tumors are the second most common testicular tumors after germ cell tumors. They account for about 2%–5% of adult testicular tumors. Most of these tumors are benign. The most common sex cord-stromal tumor is Leydig cell tumor. In contrast, testicular fibroma is a very rare type of sex cord-stromal tumors. Histologically, testicular fibromas resemble their ovarian counterparts; however, they are much less common than ovarian fibromas. To the best of our knowledge, <50 cases of testicular fibromas are reported in the English literature. Herein, we report a rare case of testicular fibroma with acellular collagen plaque in a 51-year-old male presenting as a painless testicular mass.

Keywords: Fibroma, sex cord-stromal, testis, thecoma, tunica albuginea

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INTRODUCTION

Testicular sex cord-stromal tumors are divided into pure, mixed, and unclassified tumors according to the 2016 World Health Organization (WHO) classification of tumors of the testis.^[1] The pure tumors include Leydig cell tumor, Sertoli cell tumor, granulosa cell tumor, and tumors in the fibroma-thecoma group. Fibroma and thecoma are closely related tumors, and hence, they are considered under one category in the 2016 WHO classification of tumors of the testis. Testicular fibroma is a rare sex cord-stromal neoplasm derived from either the gonadal stroma or the tunica albuginea.

CASE REPORT

This is a 51-year-old-male smoker with multiple comorbidities including obesity, type 2 diabetes mellitus,

hypertension, ankylosing spondylitis, and chronic interstitial nephritis. He had a past surgical history of bilateral total hip replacement. He was referred to urology with a 2-year history of small painless testicular mass, which recently increased in size. No history of testicular trauma or sign and symptoms of epididymo-orchitis. Ultrasound of scrotum showed a solid heterogeneous mass in the upper pole of the left testis measuring 2.7 cm [Figure 1a and b]. Tumor markers were performed including alpha fetoprotein, beta human chorionic gonadotropin, and lactic dehydrogenase, and all were within the normal limits. Left inguinal radical orchiectomy was performed. Gross examination showed a well-circumscribed intratesticular firm, white, and homogenous mass measuring 3.0 cm × 2.5 cm × 2.5 cm [Figure 2]. There was no attachment to the tunica albuginea. Necrosis and hemorrhage were absent. Microscopic examination showed

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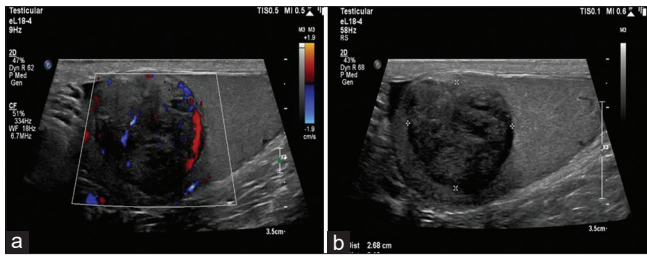


Figure 1: (a and b) Scrotal ultrasonography showing solid heterogeneous mass

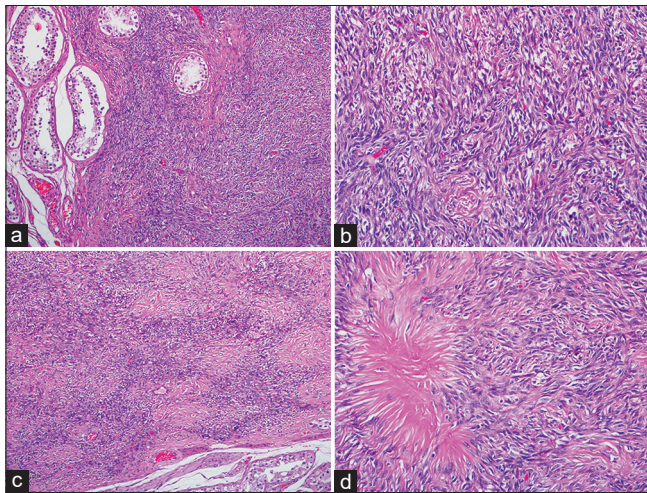


Figure 3: (a) Section from the tumor showing spindle cell proliferation with collagen bundles and entrapped seminiferous tubules at the periphery (Hematoxylin and eosin stain) (b) High power of the tumor with fascicular growth pattern and elongated nuclei. (Hematoxylin and eosin stain) (c and d) Acellular collagen plaques. (Hematoxylin and eosin stain)

an unencapsulated proliferation of spindle cells separated by thick collagen bundles with entrapped seminiferous tubules at the periphery [Figure 3a]. The neoplastic cells were arranged in the fascicular growth pattern with scant cytoplasm and elongated nuclei [Figure 3b]. Acellular collagen plaques were identified [Figure 3c and 3d]. Mitotic figures were up to 5 per 10 high-power fields (HPFs). There was no evidence of increased cellularity and nuclear atypia or necrosis. Entrapped seminiferous tubules are noted at the periphery of the tumor. Immunohistochemical stains were performed on a Ventana Benchmark Ultra autostainer (Ventana Medical System, Tucson, AZ). The tumor cells were positive for calretinin (Ventana; prediluted) and inhibin (Ventana; prediluted) [Figure 4]. The tumor cells were negative for cytokeratin AE3/AE1 (Ventana; prediluted), β -Catenin (Ventana; prediluted), S100 protein (Ventana; prediluted), Melan-A (Ventana; prediluted), CD34 (Ventana; prediluted), STAT-6 (Acris GmbH; 1:50), SMA (Ventana; prediluted), and Desmin (Ventana; prediluted). The diagnosis was testicular fibroma. The patient is under regular follow-up.



Figure 2: Gross photograph of testis showing an intratesticular well-circumscribed gray white tumor

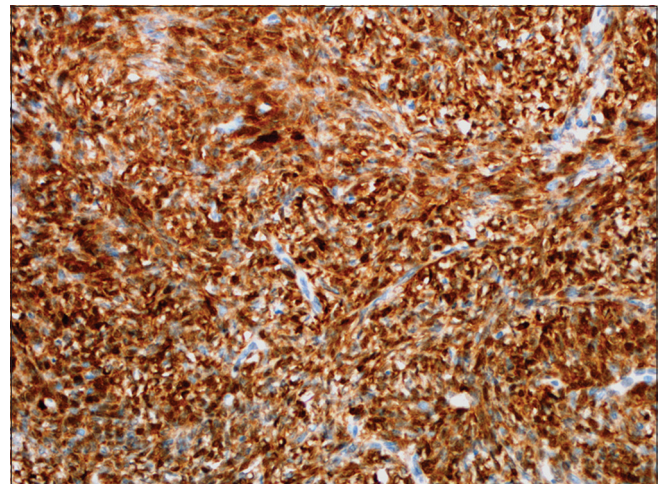


Figure 4: Calretinin immunohistochemical stain with strong positivity

A computed tomography scan of the abdomen and pelvis was performed after 6 months of the surgery, and there was no evidence of recurrence or metastasis.

DISCUSSION

Testicular fibroma is a rare sex cord-stromal neoplasm. In 1997, Jones *et al.* proposed the term fibroma of gonadal stromal origin for neoplasms that arise within the testicular parenchyma and simulate ovarian fibromas.^[2] Fibroma and thecoma are closely related tumors, so the term fibrothecoma is commonly used to cover both entities.^[1,3] The terms fibroma of gonadal stromal origin, fibroma, and fibrothecoma are generally used to describe same entity.^[1] Only few cases of testicular fibroma have been published in the literature.^[2-6] The largest case series was reported in 2013 by Zhang *et al.* where they studied 16 cases of testicular fibrothecoma.^[3] Testicular fibroma is usually present as a slowly growing painless unilateral

testicular mass in most of patients as in our case. They are present at a wide range of age from 16 to 69 years with a mean of 44 years.^[3] Macroscopically, testicular fibromas are well circumscribed tumors with a firm white-tan cut surface and have variable size from 0.5 to 7.6 cm in maximum dimension.^[3] They often abut the tunica albuginea; however, some are centrally located within the testicular parenchyma.^[1-3] Microscopically, fibromas are unencapsulated cellular spindle cell tumors that composed of fibroblasts with fascicular or storiform growth patterns associated with intervening collagen. Acellular collagen plaques are rarely seen as in the current case.^[2] The cells have elongated cytologically bland nuclei; however, focal epithelioid component may occur.^[3] Most cases have up to 5 mitoses/10 HPFs, and only rare cases may have up to 10 mitoses/10 HPFs.^[2,3] Although some tumors may show worrisome features such as hypercellularity and increased mitotic activity, they still have benign behavior. Zhang *et al.* reported two cases with 9–10 mitoses/10 HPFs, one died of other causes 5 years and 8 months and the other had no evidence of disease at 4 years and 10 months.^[3] Immunohistochemically, fibromas are often positive for inhibin and calretinin with variable expression of Melan-A, SMA, S100, CD34, BCL2, cytokeratin, S100 protein, and EMA.^[1-3]

The differential diagnosis of testicular fibroma includes Sertoli cell tumor, adult granulosa cell tumor, myoid gonadal stromal tumor, unclassified sex cord-stromal tumor, and solitary fibrous tumor. Sertoli cell tumor may have ovoid cells that resemble fibroma; however, tubular differentiation is usually present at least focally. Immunohistochemistry is also helpful for the diagnosis of Sertoli cell tumor where majority of these tumors show nuclear expression of β -catenin.^[7] Adult granulosa cell tumor of the testis is extremely rare. It has similar features of ovarian counterpart with different patterns of growth including diffuse, insular, spindled, trabecular, and pseudopapillary. Call-Exner bodies are helpful clue to the diagnosis as well as nuclear grooves of tumor cells.^[8] Myoid gonadal stromal tumor has the features of smooth muscle and gonadal stroma which can resemble testicular fibroma. It is composed of spindle cells with elongated nuclei and occasional nuclear grooves arranged in fascicles with admixed variably ectatic blood vessels.^[1,9] However, the tumor cells are diffusely positive for S100 protein, smooth muscle actin, FOXL2, and steroidogenic factor 1 and negative for calretinin and h-Caldesmon. Inhibin can be focally positivity.^[9] Fibrous stroma of unclassified sex cord stromal tumor may have similarity with testicular fibroma; however, the presence of other component of epithelial

cells of sex cord type is a helpful feature of this tumor which can be highlighted by reticulum stain.^[1,3]

Solitary fibrous tumor is another rare differential diagnosis that may have a similar morphology to testicular fibroma; however, it is usually present as paratesticular mass. A characteristic patternless pattern of growth with “stage horn” appearance of blood vessels as well as positivity for CD34 and STAT6 and lack of expression of inhibin and calretinin are typical features of solitary fibrous tumor.^[10] Finally, surgical excision seems to be a curative treatment of testicular fibroma.

CONCLUSION

Testicular fibroma is a rare benign sex cord-stromal tumor with characteristic morphological features and immunohistochemical staining profile. Despite some worrisome features that present in few testicular fibromas, they behave in a benign fashion.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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