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Testicular fibrothecoma: A case report and literature review

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ABSTRACT

Sex cord Stromal are neoplasms containing granulosa, Sertoli, and Leydig cells. The problem usually occurs among females that are hormonally active and is associated to Gorlin-Goltz syndrome. The present study reports a 37-year-old male was referred to Urologist due to a firm right testicular mass for which orchiectomy was performed. Tumor markers were all within normal range. Histopathological examination revealed compact spindle tumor involving the rete-testis in keeping with pure fibrothecoma. The results of the given study are valuable for medical professionals related to the field of urology and pathology.

Introduction

Sex cord and stromal tumors (SCSTs) are rare, representing approximately 4–5% of all gonadal neoplasms. They are neoplasms that contain Leydig cells, Sertoli cells, granulosa cells, or Theca cells. SCSTs are more common in females and are usually associated with Gorlin-Goltz syndrome (nevoid basal cell carcinoma syndrome). They usually present as a hormonally active ovarian mass in post-menopausal females after the age of sixty. Common presentation is like other diseases with an excessive estrogen state (e.g. endometrial hyperplasia, per vaginal bleeding, or adenocarcinoma). Mostly, they present unilaterally, while bilateral and extra gonadal tumors are very rare. Surgical excision is the mainstay of treatment and is mostly curative.[1]

However, few cases of testicular fibrothecoma have been reported in males, and only three case reports of a pure testicular theca cell tumors have been described in the literature.[2] The present study reports a case of pure testicular fibrothecoma.

Case presentation

A 37-year-old male, with no previously diagnosed medical conditions, was referred to Urology clinic as a case of painless firm right testicular mass, first noticed by the patient 9 months prior to presentation. Physical examination showed a hard mass located on the upper pole of the right testis, measuring about 3 cm. The left testis was normal in size, consistency, and with no palpable masses. General physical examination was unremarkable, including secondary sexual characteristics with no evidence of gynecomastia.

Scrotal ultrasound (Fig. 1-A) showed a well-defined hypoechoic vascular solid mass measuring 2.12 × 2 × 1.7cm with no parenchymal echogenicity. The left testis was normal in size with no abnormal echogenicity, mass or vascularity.

Pre-operative testicular tumor markers (Beta-hCG, AFP and LDH) were all within normal physiological limits.

The patient was considered to have a malignant testicular tumor and right inguinal orchiectomy was performed. His post-operative course was smooth, and he was sent home the following day.

Gross Pathological examination (Fig. 1-B) showed a tumor (2.3 × 2 cm) showing well circumscribed tan solid mass involving rete-testis and focally involving the epididymis. Light microscopy showed a compact spindle tumor involving the rete-testis. Higher power magnification showed bland looking spindle cells arranged in interlacing fascicles with no atypia or mitosis seen (Fig. 2). The remaining testicular tissue showed normal spermatogenesis. Immunohistochemical staining (Fig. 3) showed strong positive staining for Calretinin and SMA and weaker staining for inhibin and BCL2. The conclusion was made of a localized testicular fibrothecoma, without tunica albuginea or lymphovascular invasion.

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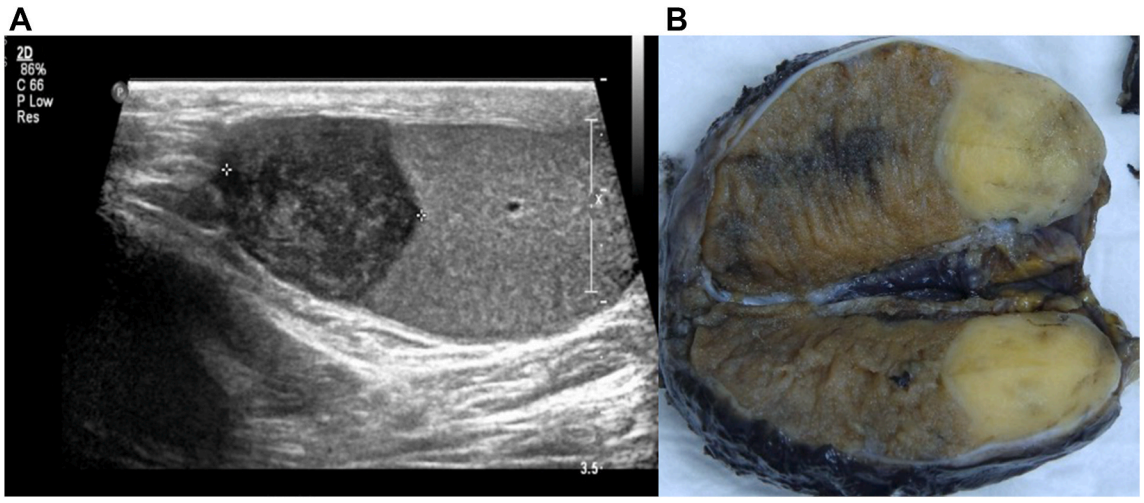

Fig. 1. A: Scrotal Ultrasound showing well-defined hypoechoic vascular solid mass measuring 2.12 x 2 x 1.7cm with no parenchymal echogenicity. B: Gross image of tumor (2.3 x 2 cm) showing well circumscribed tan solid mass involving rete-testis and focally epididymis.

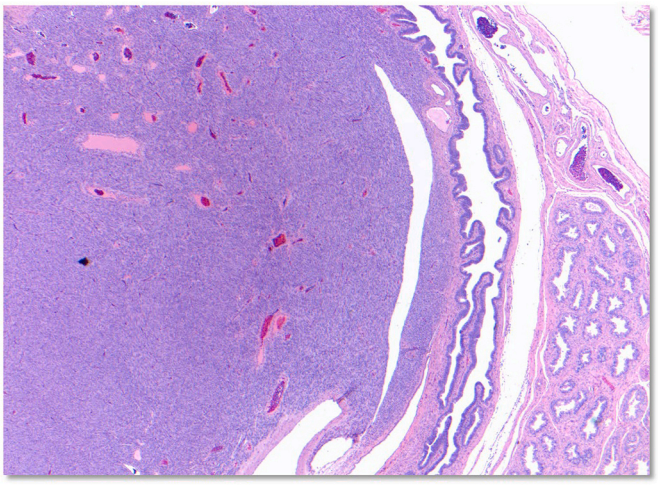

Fig. 2. Low power image showing compact spindle tumor involving the rete-testis, picture in right square is a higher power of tumor showing bland looking spindle cells arranged in interlacing fascicles with no atypia or mitosis seen.

Discussion

Testicular theca cell tumor/fibrothecoma is a rare spindle cell neoplasm with paucity in reported cases. Collins and Symington reported 4 cases of fibrothecoma in 1964.[3] In 1997, Jones and colleagues have utilized the definition "fibroma of gonadal stromal origin" to report 3 cases with identical morphological appearance to those reported in our case.[4] Almost all SCSTs have a benign course. In 1984, Eble et al. reported the only malignant SCST reported in the literature, which metastasized to the retroperitoneal space and resulted in death 13 months after orchiectomy.[5]

In several patients, testicular fibrothecoma was present at the age above 40 years, commonly with a painless testicular mass and sometime with scrotal pain or heaviness that is difficult to preoperatively distinguish from malignancy.[2] The significance of this case report is to spread the knowledge of this rare benign disease by Urologists and Pathologists, as almost all reported cases in the literature (with only one exception) [8] ended up with radical inguinal orchiectomy.

Diagnostic workup includes physical examination, laboratory testing and imaging. Physical examination usually reveals a firm or hard testicular mass, indistinguishable from malignant testicular tumors. Tumor markers (Beta-hCG, LDH, AFP) are usually within normal physiological ranges. Serum testosterone and estrogen may be elevated in some hormone secreting tumors. However, hormone levels are not part of the pre-operative laboratory assessment in common urological practice due to the rare entity of the disease and the low index of suspicion. Imaging in any patient with the symptoms or signs should almost always include a testicular ultrasound, and although it is usually diagnostic of a testicular mass, it is unable to differentiate between different kinds of testicular tumors. In most cases of SCSTs, surgical excision is usually curative, and the disease course is almost always benign. With early diagnosis and treatment, the disease-related complications are minimal, and the prognosis is excellent.

Conclusion

Based on the above discussion, the study concludes that Fibrothecomas is a rare and distinct entity of sex cord/stromal tumors of the testes and have an excellent prognosis. Awareness of such entity is of clinical importance to Urologists and Pathologists alike.

Author contributions

Mohammad Alkhamees: conceptualization, methodology, writing-reviewing and editing. Saad M Abumelha: conceptualization, supervision. Tarek Mansi: writing- original draft preparation and data curation. Noura Al-Oudah: investigation and validation.

Ethical approval and consent to participate

An informed consent was obtained regarding the patient's participation in this study.

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Declaration of competing interest

The authors declare that there is no conflict of interest regarding the publication of this article.

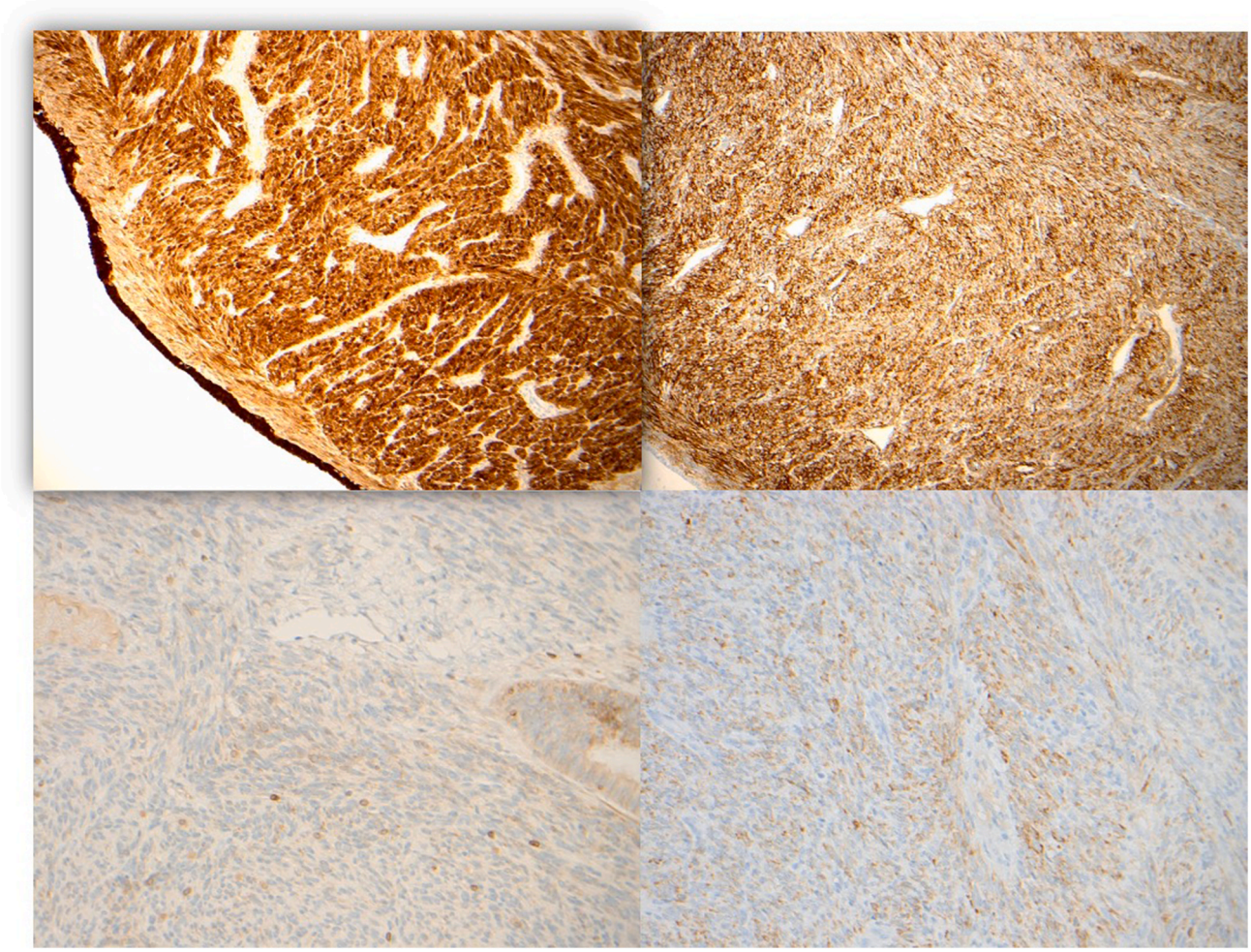

Fig. 3. Immunohistochemical staining of tumor cells show strong positive staining for Calretinin and SMA and weaker staining for inhibin and BCL2.

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