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Calcifying fibrous tumor: A rare spermatic cord presentation

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ABSTRACT

Calcifying fibrous tumor of the tunica vaginalis is a rare fibrous proliferation, which can sometimes mimic a scrotal malignancy prompting surgical intervention. It has been recognized as a benign lesion, and no recurrence has been described after its resection. Its identification is essential to avoid overtreatment and unnecessary orchiectomy. We herein describe a rare case of calcifying fibrous tumor of the tunica vaginalis in a young patient and review the clinical features of inguinal and scrotal calcifying fibrous tumors to help clinical decisions and encourage a testis sparing surgery.

Introduction

Calcifying fibrous tumor of the tunica vaginal is a rare, benign fibrous proliferation in the tunica vaginalis. Although rare, it represents the second more frequent benign paratesticular lesion, after adenomatoid tumor of the epididymis, comprising 6% of all paratesticular tumor. Surgical excision is the resolutive therapy, and recurrence after its excision has not been reported. However, most clinicians lack experience in the calcifying fibrous tumor due to its low incidence, making preoperative diagnosis difficult and possibly leading to overtreatment and unnecessary orchiectomy.

Case report

A 20-year-old male presented to our outpatient clinic with a complaint of painless left inguinal mass, which slowly increased in size over the previous 5 years. He denied local trauma or other symptoms and had been otherwise healthy. On physical examination, the patient appeared well, both testicles were normal in size and consistency, and no hydrocele was encountered. A firm non-tender left inguinoscrotal nodule was located near the external inguinal ring. There were no inguinal lymphadenopathy or any other findings.

Laboratory investigations revealed normal blood cell count, as well as a serum AFP, LDH, and HCG within the normal ranges. Scrotal and inguinal ultrasound showed a heterogeneous and well-delimited lesion with acoustic shadowing beside the left external inguinal ring measuring 4.2 × 2.7 × 2.6cm, closely related to the spermatic cord. Color Doppler flow imaging showed no blood-flow sign in the nodule (Fig. 1A). The testis and epididymis were normal, and no varicocele or hydrocele was presented.

The patient was subsequently taken to the operating room for inguinal exploration. Upon surgical exploration, a well-circumscribed round-shaped lesion was found into the spermatic cord and resected (Fig. 1B and C). No other lesions or abnormalities were detected, and the testicles were preserved. The pathology report revealed a hypocellular tumor, with a dense well-circumscribed hyalinized collagen, lymphoid aggregates, scattered lymphocyte and plasmocyte infiltration, and dystrophic calcifications, consistent with a calcifying fibrous tumor (Fig. 1D and E).

Discussion

A calcifying fibrous tumor is a rare benign tumor most commonly found in the gastrointestinal tract and pleura. Its occurrence in the tunica vaginalis is even rarer, with just a few cases reported. Due to the variable presentation and histology, these tumors had been received different designations such as fibrous pseudotumor, fibroma, calcifying fibrous pseudotumors, inflammatory pseudotumor, benign fibrous paratesticular tumor, pseudofibromatous periorchitis, nonspecific peritesticular fibrosis, and reactive periorchitis.[1,2] In 2002, the World Health Organization denominated this entity as calcified fibrous tumor.[2] Additionally, Some of these cases previously described in the literature

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may correspond to other entities such as a solitary fibrous tumor or inflammatory myofibroblastic tumor; however, the majority of paratesticular inflammatory pseudotumors and calcified fibrous tumor are currently thought to represent the same lesion in various stages of development, with the predominance of inflammation and myofibroblastic proliferation in early lesions, to heavily collagenized, paucicellular fibrous nodules in more advanced stages.[3]

Clinically, it appears as a slow-growing non-tender mass, with a wide range of size. It can be presented as multiple or solitary, scrotal or inguinal mass, sometimes mimicking a testicular malignancy, which may result in radical surgery. Most reported cases have involved exclusively the tunica vaginalis; occasionally, it can affect the tunica albuginea, epididymis, or spermatic cord as well.[1] Other clinical presentation, much less common, is associated with a thick, firm and fibrotic band involving the tunica vaginalis, which can encase the testis.[1]

Its etiology is poorly understood and its true nature (i.e., reactive vs. neoplastic) is controversial.[4] Since half of the patients have hydroceles and about 30% have a history of trauma or epididymitis, some advocate for a reactive etiology, as opposed to a neoplastic origin.[4-] Based on certain similarities to other fibroinflammatory disorders characterized by infiltrates of IgG4-expressing plasm cells, some studies support the theory of this tumor might be an immunoglobulin G4-related disease.[3]

Ultrasound usually exhibits a well-delineated lesion with acoustic shadowing due to the dense fibrotic core and its calcification. An associated hydrocele is the most frequently concurrent finding. Magnetic resonance imaging could also be utilized to evaluate these lesions and usually demonstrates low signal intensity on both T1 and T2 weighted images due to fibrosis.[5]

The calcifying fibrous tumors have a clear boundary and lacking capsule. On sectioning, it appears as a solid, firm, and grey-whitish tumor. The size could sharply range. Microscopically, It presents with densely hyalinized collagen along with dystrophic calcifications and lymphoplasmacytic infiltrate.[2] Although immunohistochemical analysis is not mandatory for diagnosis, fibroblasts do express vimentin and factor XIIIa, and CD34 immunoexpression can be seen occasionally.

Surgical excision is the resolutive therapy. Recurrence after its excision has not been reported, strengthening an organ-sparing surgery.[2] If there is any concern for malignancy, a frozen section can be done intraoperatively.

Conclusion

Urologists must be aware of the possibility of calcifying fibrous tumor when assessing a patient with paratesticular lesion. Ultrasound and MRI images and intraoperative pathological frozen section examination can contribute to the surgical management decisions and exclude malignancy. As it is a benign reactive lesion, its diagnostic assumption can avoid unnecessary orchiectomy. Herein, we described an illustrative case to remind of this differential diagnosis.

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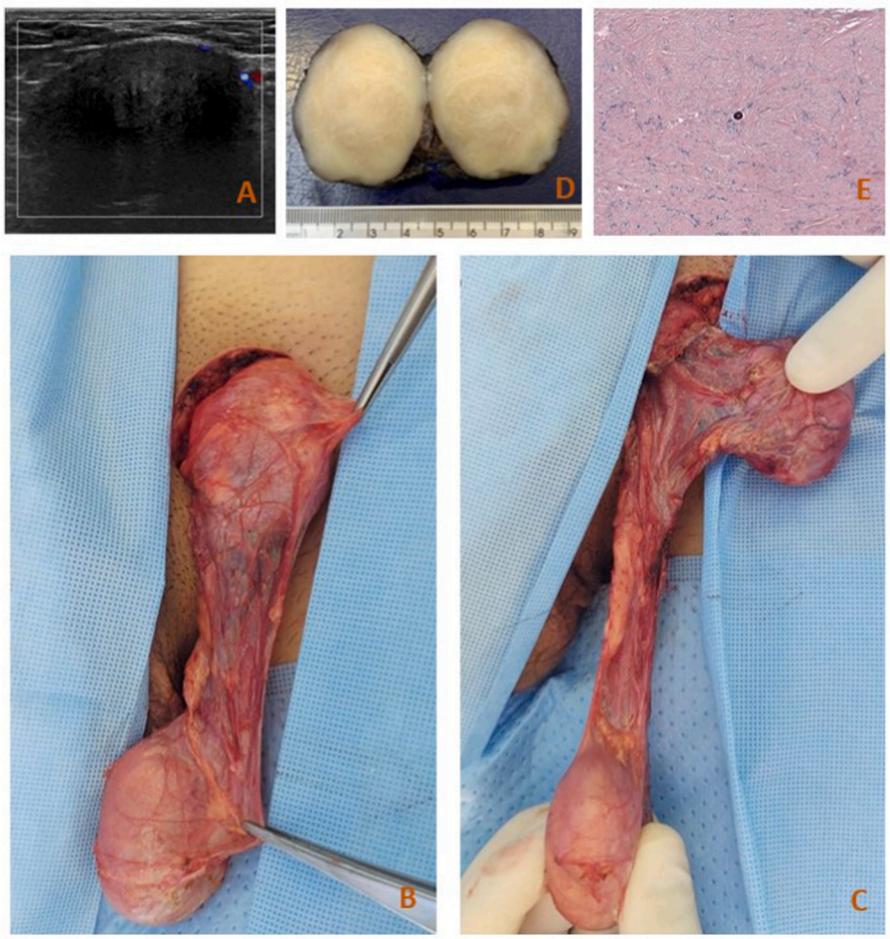

Fig. 1. A: Ultrasound doppler scan revealing a well-delineated, heterogeneous, and hypovascular lesion. **B– C:** Surgical images showing a tumor into the spermatic cord and its vascularization. **D:** Gross specimen of calcifying fibrous tumor of the tunica vaginalis testis. E: Hematoxylin-eosin-stained specimen illustrating hyalinized collagen bands with dystrophic calcification.

Declaration of competing interest

None declared.

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