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Case Report

Paratesticular fibrous pseudotumor unraveling the rare diagnosis on histopathology

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Abstract

Paratesticular fibrous pseudotumor is a rare benign tumor-like lesion that originates mostly from the tunica vaginalis and affects the testicular tunica albuginea, epididymis, and spermatic cord. This tumor-like lesion is thought to be a consequence of a reactive proliferation of inflammatory and fibrous tissue, however exact etiology is still unknown. The diagnosis was mostly made on an excision specimen by histopathological examination.

We describe an extreme rare case report of an Indian male in his 50s complaining of pain, hardening, and swelling of the left testicle for 5 days. Literature attests to its exceptional rarity; only a few examples have been recorded from India, and there are only about 200 cases globally that have been archived. Henceforth, it is worthwhile to share our experience and report our case.

Keywords: Paratesticular tumor, Fibrous Pseudotumor, Tumor like lesions, High resonance sonography

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1. Introduction

Paratesticular fibrous pseudotumor is a rare benign tumor-like lesion that originates mostly from the tunica vaginalis and affects the testicular tunica albuginea, epididymis, and spermatic cord.[1,2] In 1830, Sir Astley Cooper et al. first reported this rare tumor-like condition.[3] Later, in 1904, Balloch gave a detailed case report presentation.[4,5,6,7]

We describe a case of an Indian male in his 50s who had only five days' history of clinical symptoms. Literature attests the exceptional rarity of this tumor like lesion; only a few example have been recorded from India, and there are only about 200 cases globally to date that have been archived.[8] Henceforth, it is worthwhile to share our experience and report our case.

2. Case Report

An Indian male in his 50s came to surgery emergency with complaints of pain, hardening, and swelling of the left testicle for 5 days. The High Resolution Sonography (HRS) report showed larger left testes than right side with increased color flow with few hypoechoic patches noted. The findings suggested orchitis, left side. The right testes appeared normal. The patient was operated on, on the same day of admission; a left orchidectomy was performed under general anaesthesia. The specimen was submitted to the Department of Histopathology for histopathological examination for a definite diagnosis.

The gross specimen consists of an enlarged testis measuring 5.5 x 3.5 x 3 cm and weighing 215 g. The epididymis appears to be necrosed and blackened, and focal ulceration of overlying tunica vaginalis is noted. The rest of the external surface is smooth and shows engorged blood vessels. On serial sectioning, band-like solid paratesticular growth was identified, measuring 5.5 x 3 cm. Growth is gray-white and firm to hard in consistency. Grossly, growth is adherent to tunica vaginalis and abutting the tunica albuginea and appears to infiltrate onto adjacent testicular parenchyma and also encasing the spermatic cord margin. Testis measures

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2.6 x 2.2 cm, and the cut surface is unremarkable grossly. (**Figure 1**)

Five full-thickness slices were sampled along with the spermatic cord margin. No remnants were kept.

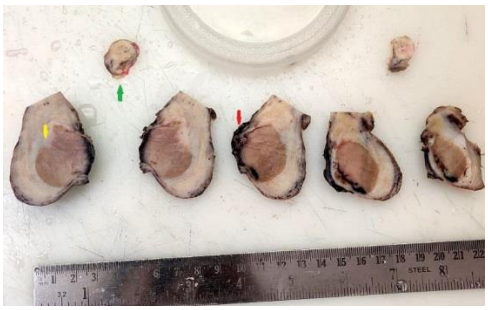

Figure 1: Serial sections of the received gross specimen show a diffuse band-like, tan-colored, firm, and solid tumor-like lesion adhered to the tunica vaginalis and abutting to the testes (yellow arrow). Tunica albuginea is indistinguishable. The tumor-like growth reached up to the surgical margin, encasing the spermatic cord (green arrow). There is an ulceration noted over the epididymis (red arrow).

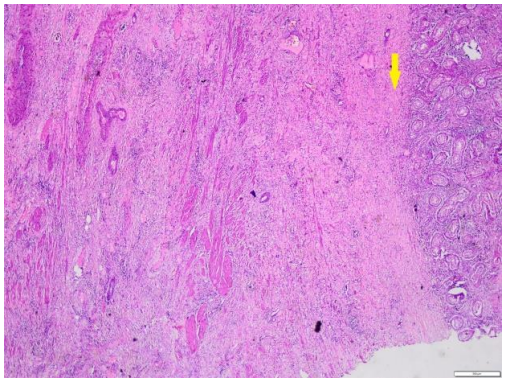

Figure 2: Sections show haphazard proliferation of fibrosis with interspersed fibroblasts, myofibroblasts, and interspersed inflammatory cells with congested blood vessels, arising from the tunica and abutting the testicular parenchyma in a zone-like pattern [H&E, 40X, yellow arrow].

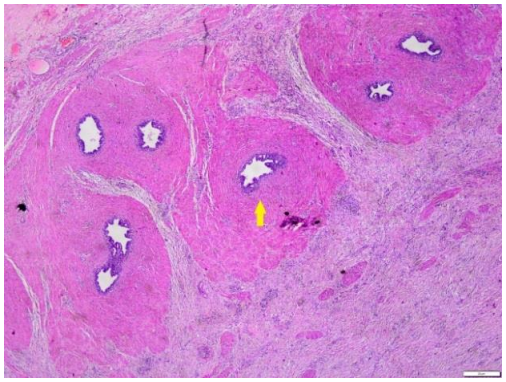

Figure 3: Section show encasement and distortion of Vas deferens mimicking telescoping artifact [H&E, 100X, yellow arrow].

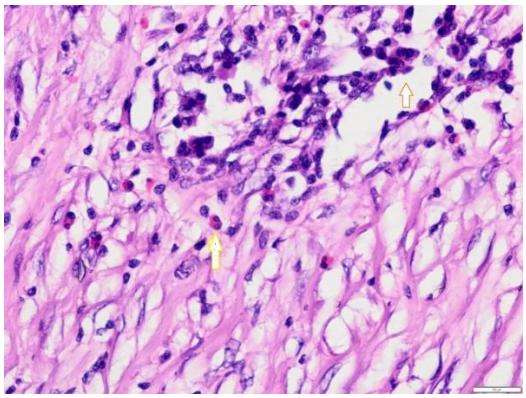

Figure 4: Section shows multifocal areas showing aggregates of plasma cells (red arrow) with their reactive forms and prominence of eosinophils (yellow arrow) [H&E, 400X].

Multiple H&E-stained sections examined under the microscope revealed a tumor-like inflammatory lesion arising from the testicular adnexa, involving the tunica vaginalis, tunica albuginea, and spermatic cord, as well as a part of the epididymis focally. Sections examined from growth show hypercellular to hypocellular areas comprising of proliferating fibroblasts, myofibroblasts, and histiocytes and interspersed dense mixed-cell inflammatory cell infiltrate comprising of neutrophils, lymphocytes, plasma cells, and prominence of eosinophils. Many dilated lymphovascular channels are noted with areas of fibrosis and hemorrhage. Testicular parenchyma, rete testis, and epididymis are markedly infarcted and non-functioning and infiltrated by dense acute inflammatory cell infiltrate at places forming micro-abscesses. Sections examined from the spermatic cord margin show similar histomorphological findings as described above.(**Figure 2**, **Figure 3**, **Figure 4**) No evidence of tubercular granuloma or malignancy was seen in the total 32 sections examined.

Thereafter, a diagnosis of "Paratesticular Fibrous Pseudotumor with Acute Epididymo-Orchitis" was awarded.

3. Discussion

The etiology of paratesticular fibrous pseudotumor is not fully understood, but trauma-induced inflammation, infective pathology, or inflammatory hydrocele might play a role in its development.[9] In recent years, few studies stated its association with serum IgG4-related disease (IgG4-RD), induces sclerosis and plasma cell-rich infiltrate, which show IgG4 positivity.[10,11,12]

The epididymis was found to be involved in our case report. However, epididymis involvement is rare in this pseudotumor, seen in less than 10% of all reported cases.[13]

This tumor-like lesion is thought to be a consequence of a reactive proliferation of inflammatory and fibrous tissue.[13]

The diagnosis was mostly made on an excision specimen by histopathologic examination.[13]

It is most commonly seen in young and middle-aged males, with an average age of 30 years, but reports show a wide range of age presentation from 7 to 95 years.[1,13] It presents as a painless single or multiple discrete nodular lesions varying from 0.5 to 8 cm in size or diffuse multinodular hemiscrotal mass.[8] In our case, it presents as a diffuse sheath like thickened paratesticular tumor like lesion abutting and pushing testis. Detachment of nodules can lead to free floating 'pearls' seen in grossing and also can be detected on USG.[8] The consistency is usually hard, and patients encounter difficulty in sectioning while grossing, which makes it difficult to differentiate from sarcomas.[8] The lesion was found to be associated with hydrocele in up to half of the cases.[8,9]

Ultrasonography (USG) is the commonest initial imaging modality used mostly. The USG findings usually showed well-defined, homogenously hypoechoic or hyperechoic extratesticular lesions.[4,8] Magnetic resonance imaging (MRI),with its classical findings being a very low signal density on T2-weighted images, an intermediate signal density on T1-weighted images, and no or minimal non-homogenous contrast enhancement, is thought to be a better investigation offered to the patient in a diagnostic dilemma.[14]

However, high-resolution sonography is a cost-effective, less time-consuming, and easily available modality to rule out malignancy. Likewise, in our case, HRS showed an inflammatory cause, i.e., orchitis, and exclude out the malignancy which later show concordance with final histopathological diagnosis.

Excision of affected testis and spermatic cord (if involved) is sufficient for the treatment of paratesticular fibrous pseudotumor.[1,8] An intraoperative frozen section can be performed for the spermatic cord margin to avoid chances of malignancy, as most of the time this entity is a mimicker on radiology, and to avoid unnecessary radical surgeries.[1,8]

This tumor-like lesion can show different gross presentations as multinodular, well-delineated, oval, and freely mobile nodules, often with diffuse fibrosis of the tunics, and may involve epididymis and spermatic cord.[15] The microscopic examination demonstrated hyalinized tissue and fibroblasts in a richly vascularized stroma, with collagen bundles and calcification foci (noted in few studies) and inflammatory cells comprising lymphocytes, plasma cells, and a few eosinophils and neutrophils.[9] The inflammation may vary from sparse to intense. Immunohistochemistry (IHC) is non-specific for paratesticular fibrous pseudotumor; however, it can be applied if available and to reduce the subjectivity of different diagnostic opinions. IHC can identify fibroblasts and is positive for vimentin, smooth muscle-specific actin, and common muscle actin and negative for S-100, keratin, and desmin.[8,16]

The prognosis of this pseudotumor is excellent to good, if not associated with IgG4-related disease, and there is no reported evidence of recurrence in many studies.[1,16]

4. Conclusion

Paratesticular fibrous pseudotumor is a tumor-like lesion, an extremely rare entity involving the testicular adnexa and rarely involving the testis; however, epidymymal and spermatic cord involvement are noted. The commonest etio-pathogenesis postulated is an inflammatory-reactive process due to trauma, and close association with hydrocele is well documented. HRS is recommended for diagnostic modality to reduce over diagnosis of this benign entity as malignancy on simple USG. MRI is also recommended in difficult cases but mostly gives similar results as HRS; however, it is costly, time-consuming, and less available in developing countries for low-income patients. Excision is the mainstay treatment; an intra-operative frozen section of the spermatic cord margin can be performed in doubtful cases. Histopathological examination is the only key to unlocking the correct diagnosis. However, IHC is not recommended but can be performed if available.

5. Source of Funding

None.

6. Conflict of Interest

None.

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