



Oncology

Leiomyoadenomatoid tumors: A type of rare benign epididymal tumor

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ABSTRACT

Primary tumors of the epididymis are mostly benign in nature, and the most common type is adenomatoid tumors followed by leiomyomas. Leiomyoadenomatoid tumors are very rare benign epididymal neoplasms composed of two components: gland-like structures lined by cuboidal cells and bundles of smooth muscle components. The goal of treatment is testicular-preserving surgery. A preoperative and intraoperative evaluation plays an important role in proper management. To the best of our knowledge, few cases have been reported in the literature. We report a case of a right epididymal tail leiomyoadenomatoid tumor in a 49-year-old male who underwent *trans*-scrotal exploration and tumor excision.

Introduction

The majority of intrascrotal masses are testicular in origin and usually malignant; paratesticular tumors represent 2–3% of intrascrotal masses and are usually benign. Adenomatoid tumors are a benign mesothelial cell origin that can be seen in male and female genital tracts. Adenomatoid tumors are the most common benign epididymal neoplasms, followed by leiomyomas.^{1,2}

Leiomyoadenomatoid tumors are a rare variant of adenomatoid tumors that are commonly present in middle-aged males and contain both adenomatoid and spindle cell components. This term was defined by Epstein³ in 1991. Clinically, radiological and gross findings cannot distinguish adenomatoid from leiomyoadenomatoid tumors, and careful microscopic examination is essential to identify this variant.⁴ Here, we report a case of an epididymal tail leiomyoadenomatoid tumor in a 49-year-old male.

Case presentation

A 49-year-old male known to have diabetes mellitus presented with a painless right intrascrotal mass that started five years ago and did not increase in size. There was no history of lower urinary tract symptoms and no previous scrotal trauma, genitourinary tract infections or surgeries. Local examination revealed a right lower intrascrotal nontender, round and firm mass approximately 3 × 2 cm separated from the testis.

Nothing significant was found in the rest of the physical exam. Routine laboratory tests, including complete blood count, blood chemistry, urinalysis, serum α -fetoprotein, B-human chorionic gonadotrophin, and alkaline phosphatase, were all within the normal limits.

Scrotal ultrasonography demonstrated a well-defined mildly hyperechoic lesion in the tail of the right epididymis, with mild internal vascularity on color Doppler ultrasound examination (Fig. 1). Scrotal MRI showed a well-defined right hemiscrotum mass measuring 1.9 × 2.3 cm along the posterior inferior aspect of the right testis and separated from it, likely arising from the epididymis tail with partial hypointensity in T2-weighted sequence (Fig. 2). Subsequently, right *trans*-scrotal exploration was performed, during which a benign looking, rounded, encapsulated, and whitish mass was identified arising from the tail of the epididymis, which was dissected from the surrounding tissue and excised. The patient's postoperative course was uneventful.

Microscopically, the tumor was defined by two components: the first consisted of benign cords, nests and tubules, and the second was composed of smooth muscle cells. No nuclear atypia or increasing mitotic activity was observed. Immunohistochemistry studies showed an adenomatoid component stained by calretinin, and the leiomyomatous component revealed a positive reaction for h-caldesmon (Fig. 3).

Finally, based on the two components observed, mesothelial and muscular, the patient was diagnosed with a right epididymal leiomyoadenomatoid tumor. At the 2-month follow-up, the patient was well and without evidence of recurrence.

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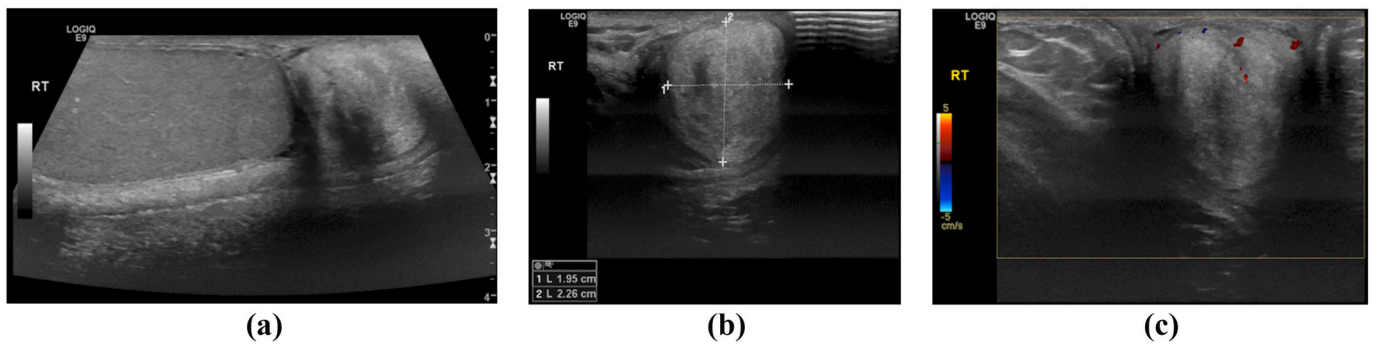
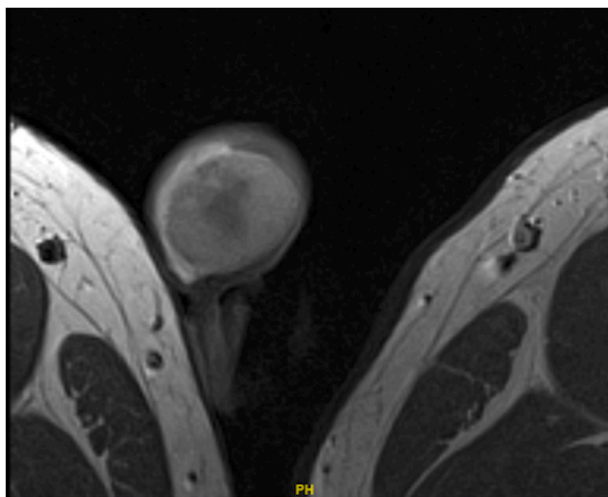
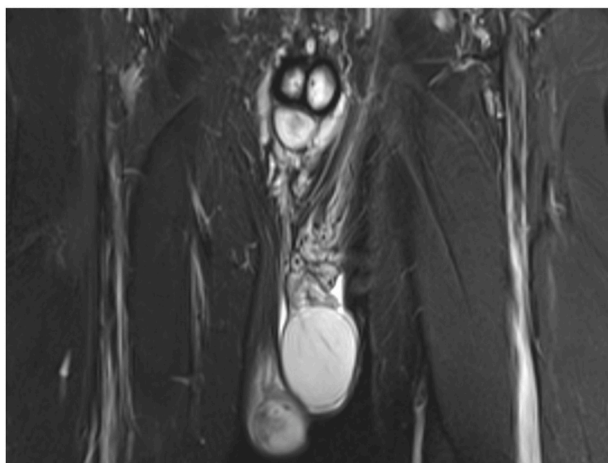


Fig. 1. Testicular U/S: (A) a longitudinal view shows a well-defined hyperechoic lesion inferior to the right testis. (B) The lesion measures 1.9 × 2.2 cm in transverse view. (C) Mild internal lesion vascularity observed.



(a)



(b)

Fig. 2. MRI of the scrotum. (A) Axial view shows a right intrascrotal mass, inferior, posterior and separated from the testis. (B) Partial low signal in T2 demonstrated in coronal view.

Discussion

Epididymal leiomyoadenomatoid tumors commonly develop in middle-aged males. However, patients can present with painless intrascrotal slow-growing masses, which are usually discovered incidentally during physical examination or during other procedures. They behave in

a benign fashion, with no metastasis or recurrence reported.⁴ A round scrotal extratesticular nodule less than five cm is usually encountered during physical examination, and the most common location is the distal part of the epididymis. Scrotal ultrasonography is ideal to differentiate between cystic and solid scrotal masses, but there is no specific characteristic to distinguish benign from malignant lesions.¹ Leiomyoadenomatoid tumors appear as heterogeneous, hypoechoic or hyperechoic lesions on sonography. Scrotal MRI or CT can help in select patients to assess the lesion characteristics and relation to surrounding tissue. FNAC can be a useful technique in epididymal nodules, as it can detect benign and malignant conditions in 90.3% of patients and helps to avoid other aggressive diagnostic procedures.⁵ FNAC can be considered in patients who are highly suspected of infection, with inflammatory nodules and surgical removal not indicated.

Leiomyoadenomatoid tumors are rare benign neoplasms with the presence of two components: an adenomatoid component in the form of gland-like structures lined by single flat or cuboidal cells intermixed with a smooth muscle component in the form of spindle cells. The histogenesis of this entity is debatable. It may represent a collision neoplasm, a variant of adenomatoid tumor associated with smooth muscle proliferation or adenomatoid tumor arising in the background of smooth muscle hyperplasia. Importantly, abundant growth of smooth muscle components can sometimes obscure the adenomatoid component, which might lead to misdiagnosis of leiomyoma or smooth muscle malignancy.⁴

There is no difference in treatment when dealing with epididymal adenomatoid tumors, leiomyomas or leiomyoadenomatoid tumors. As the most common paratesticular tumor is benign in nature, tumor excision with testicular-preserving surgery should be a preferred option taking into consideration a preoperative diagnosis, and intraoperative FSA in selected cases has an important role in preventing unnecessary orchidectomies in cases where differential diagnosis is not achievable.¹ Although an inguinal approach is the standard in any intrascrotal solid tumor, Beccia DJ² found that multiple factors can be helpful to organize a clinical approach of nontesticular tumor management, including patient age, location of the tumor and mode of presentation. A *trans*-scrotal approach is sufficient in patients more than 50 years old with painless epididymal mass because it is likely to have either a benign tumor or metastasis from another primary tumor in which a cure is not achievable. In middle-aged men between 20 and 50 years of age who present with a painless epididymal mass, a scrotal approach can be performed if a benign disease is suggested by lesion finding, such as if it is located in the lower, left side, multiples or bilateral lesions. Otherwise, any child or unresolved painful epididymal mass should undergo an inguinal approach to allow for radical orchidectomy if malignancy is found. In our case, a *trans*-scrotal approach with tumor excision was performed because a benign tumor was highly suspected based on the presentation and clinical and radiological findings.

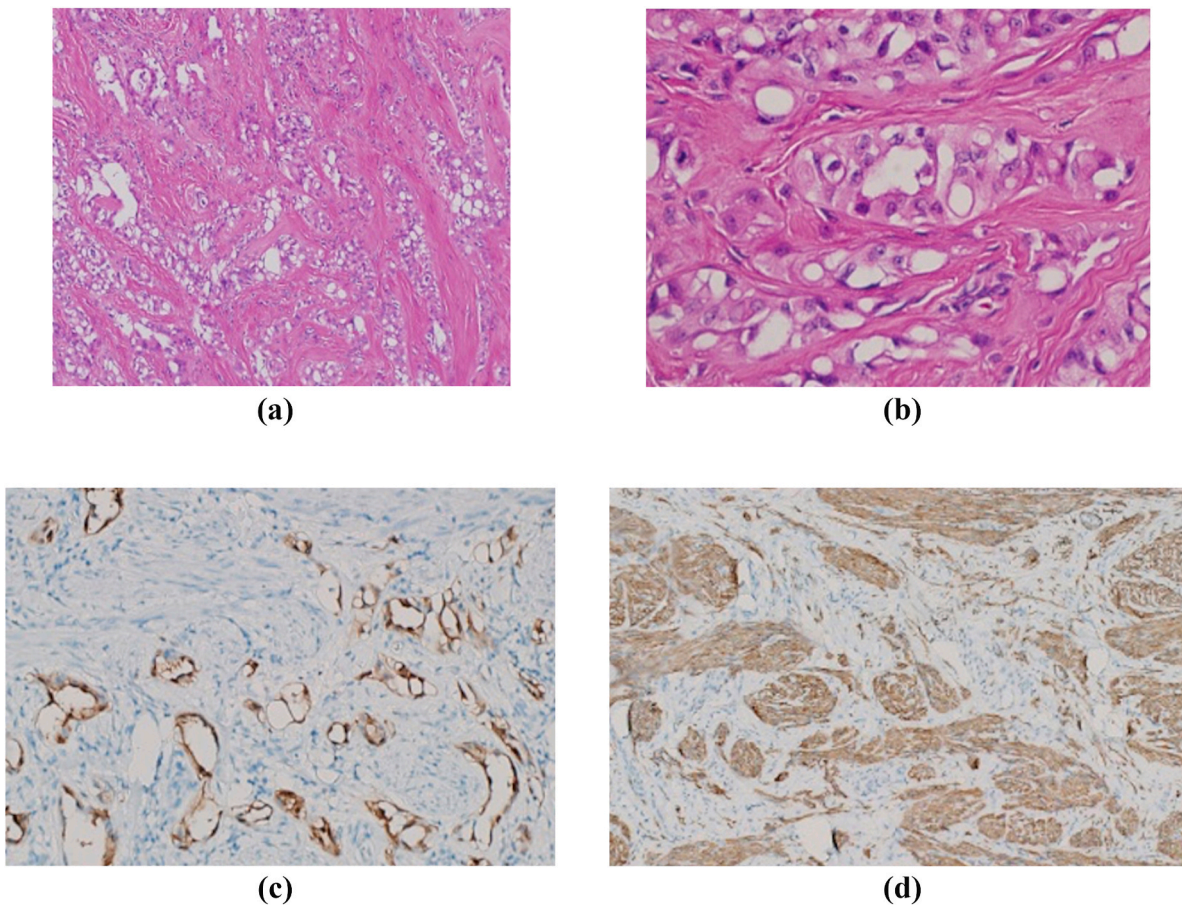


Fig. 3. Leiomyoadenomatoid tumor: (A&B) X100 and 400X H&E staining shows the neoplasm arranged in tubules or gland-like structures along with cords as seen in the high power view. These tubules were lined with a single layer of cuboidal cells, moderate to abundant eosinophilic cytoplasm with single to multiple vacuoles, and a few cells in the background have Signet ring cell morphology. The background stroma show intersecting bundles of smooth muscle cells (form of bland spindle cell). (C) Calretinine immunostaining highlighting the adenomatoid component of the tumor. (D) h-caldesmon highlights the leiomyomatous component.

Conclusion

Epididymal leiomyoadenomatoid tumors are very rare benign neoplasms that contain adenomatoid and smooth muscle components. The mode of presentation, preoperative work-up and careful intraoperative evaluation were essential for proper management. A meticulous microscopic examination with an experienced pathologist is important to diagnose this rare type of neoplasm.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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Declaration of competing interest

The authors declare that there are no conflicts of interest regarding the publication of this paper.

References

1. Urkmez A, Akan S, Ozsoy E, Sahin A, Koca O, Ozturk MI. Diagnosis and treatment of paratesticular adenomatoid tumors. *J Coll Physicians Surg Pak.* 2018;28(9):S217–S219.
2. Beccia DJ, Krane RJ, Olsson CA. Clinical management of non-testicular intrascrotal tumors. *J Urol.* 1976;116(4):476–479.
3. Epstein JI. *Differential Diagnosis in Pathology: Urologic Disorders.* Igaku-Shoin medical Pub; 1992:173–174.
4. Shehabeldin A, Al Sanna G, El-Zaateri Z, et al. Leiomyoadenomatoid tumor of epididymis: a variant of adenomatoid tumor. *Ann Clin Lab Sci.* 2020;50(6):813–817.
5. Gupta N, Rajwanshi A, Srinivasan R, Nijhawan R. Fine needle aspiration of epididymal nodules in Chandigarh, north India: an audit of 228 cases. *Cytopathology.* 2006;17(4):195–198.