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Epididymal leiomyoadenomatoid tumor: A case report of a rare benign paratesticular mass

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ABSTRACT

Primary tumors of the epididymis are rare and commonly benign in nature. Leiomyoadenomatoid tumors are a rare subvariant of adenomatoid tumors that combines features of leiomyomas. Tumor histology is notable for tubular spaces lined by mesothelial cells with a proliferative spindle cell component. To the best of our knowledge, few cases have been reported in the literature. We report a case of leiomyoadenomatoid tumor in a 58-year-old male.

1. Introduction

Adenomatoid tumors are benign, yet rare tumors of the epididymis and testis derived from mesothelial origin. These neoplasms have also been characterized in the female genital tract, pancreas, liver, mesocolon and adrenal glands. An even rarer subvariant, Leiomyoadenomatoid tumor, has been described as a smooth muscle component that has been incorporated into the neoplasm.[1] Leiomyoadenomatoid tumors were first designated as a "mixed tumor of the epididymis" but were later renamed "leiomyoadenomatoid tumors".[2] In some cases, the adenomatoid component may be overshadowed by the proliferative smooth muscle component making it difficult to diagnose on pathology.[1] Here, we report a case of an epididymal leiomyoadenomatoid tumor in a 58-year-old male.

2. Case presentation

A 58-year-old male with hypertension and recent history of subarachnoid hemorrhage repaired with endovascular coiling presented for evaluation of a right scrotal mass with intermittent right testicular pain for three years. The patient reported a history of scrotal swelling, dysuria, urgency, frequency and intermittent nocturia. Physical examination revealed a firm right multilobular paratesticular mass. Routine laboratory tests including complete blood count, basic metabolic panel and urinalysis were within normal limits.

On ultrasonographic imaging, the swelling consisted of a 2.7 × 3.6 × 3.7 cm inhomogenously echogenic mass with central vascularity (Fig. 1). Differential considerations included adenomatoid tumor, scrotal hemangioma, epididymal leiomyosarcoma, and granulomatous disease. Obtaining the correct diagnosis was essential for determining the approach to treatment. Additional laboratory testing for oncologic biomarkers including prostate specific antigen (PSA), PSA free, CA-125, alpha-fetoprotein, lactate dehydrogenase and human chorionic gonadotropin were all within normal limits.

Right sided transcrotal exploration was subsequently performed revealing a normal appearing testicle with an inferior pole mass originating from the epididymal tail. Intraoperatively the mass was presumed to be a spermatocele and was dissected off the surrounding tissue, excised and sent for pathology. The patient had an uneventful postoperative course.

On macroscopic examination, a solid well-circumscribed mass arising from the epididymal head was observed measuring 3.0 × 2.5 × 2.4 cm. A cut section showed a tan-white whorled appearance without gross visible degenerative changes. The surrounding adnexa were otherwise normal. On immunohistochemical analysis, the smooth muscle component of the specimen stained positively for Desmin and Smooth Muscle Actin. The glandular component showed reactivity with Calretinin, Cytokeratin AE1/AE3, Podoplanin and CD34, but was negative for Wilm's Tumor 1 (Fig. 2). Based upon the two microscopically observed components with immunohistochemical confirmation, a diagnosis of leiomyoadenomatoid tumor was made.

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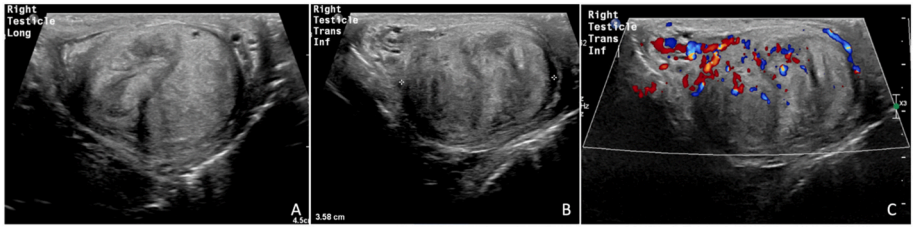

Fig. 1. Ultrasound of the right testicle. (A) Long view of heterogeneous scrotal mass. (B) Transverse view of scrotal mass. (C) Doppler showing increased vascularity.

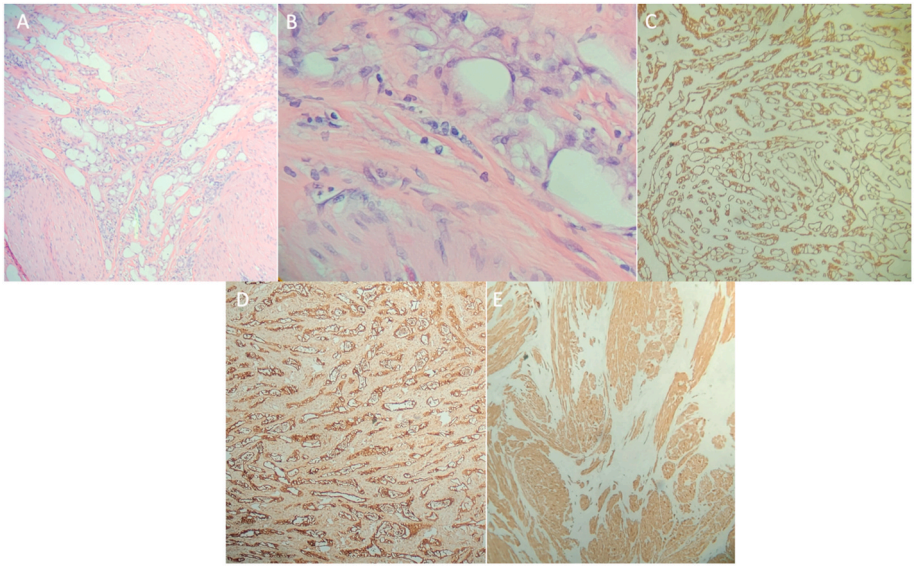

Fig. 2. Microscopic examination of tumor. (A) H&E x100 demonstrating smooth muscle overgrowth. (B) H&E x400 of smooth muscle fibers. (C) Immunohistochemical staining for AE1 and AE2. (D) Immunohistochemical staining for calretinin. (E) Immunohistochemical staining for desmin.

3. Discussion

Adenomatoid tumors originate from the mesothelium and are the most common paratesticular tumor.[3] Leiomyoadenomatoid tumors represent a rare subvariant of adenomatoid tumors which incorporates a smooth muscle component. Although the histopathogenesis of this tumor is not well understood, to date, there have been seven reported cases of leiomyoadenomatoid tumors identified in the literature. A combination of the presence of both adenomatoid tumor and leiomyoma, derivation from progenitor cells capable of differentiation into mesothelium and smooth muscle, and adenomatoid tumor arising in a pre-existing asymptomatic setting of smooth muscle hyperplasia have all been suggested hypotheses of leiomyoadenomatoid tumor origin2. Regardless, exuberant growth of the smooth muscle components may obscure the adenomatoid components of leiomyoadenomatoid tumors leading to their misdiagnosis.[1] These concerns warrant careful pathological review with immunohistochemical confirmation.

Leiomyoadenomatoid tumors within the genitourinary tract of men typically present as painless scrotal masses incidentally found on physical exam or other procedures. On scrotal ultrasonography leiomyoadenomatoid tumors appear heterogenous as well as either hyperechoic or hypoechoic.[2] Scrotal MRI or CT may help distinguish the lesion's characteristics and better assess its relation to the surrounding parenchyma.[4]

Currently, there is no difference in the management of epididymal adenomatoid, leiomyomas and leiomyoadenomatoid tumors.[4] These lesions are benign and are well managed with surgical excision without metastasis or further recurrence.[2] Testicular preservation surgery is the preferred option for management of these lesions where the preoperative differential diagnosis has a benign disease course. Intraoperative frozen sections in select cases have an important role in preventing unnecessary orchiectomies where the differential diagnosis includes malignancy.[3]

To date there are no reported cases of recurrent leiomyoadenomatoid tumors or malignant transformation.[5] At 15 months follow-up, the patient has not exhibited any recurrence. We will continue to follow our patient for any further pathologic developments.

4. Conclusion

Leiomyoadenomatoid tumors are exceedingly rare benign masses that contain adenomatoid and leiomyomatous elements. These lesions have been reported in the epididymis of men as well as the uterus and adnexa in women. We present one such case of leiomyoadenomatoid tumor in the right inferior epididymis of our patient. Rigorous microscopic evaluation and adequate sampling are necessary to avoid misdiagnosis.

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Declaration of competing interest

The authors declare no conflict of interest.

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