

A unique case of a serous borderline tumor of the paratestis

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Abstract Serous borderline tumors of the paratestis are histologically identical to their ovarian counterparts and hypothesized to arise from Mullerian metaplasia of the tunica vaginalis. They are exceedingly rare with many cases probably going clinically unnoticed. We present a case of serous borderline tumor of the paratestis, with, to the best of our knowledge, the first published sonographic and magnetic resonance (MR) images of this entity. It was successfully treated with partial orchiectomy with no disease recurrence after one-and-a-half years.

Key Words: Paratestis, scrotal imaging, serous borderline tumor, tunica albuginea

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INTRODUCTION

Serous borderline tumors of the paratestis are histologically identical to their ovarian counterparts and hypothesized to arise from the Mullerian metaplasia of the tunica vaginalis. They are exceedingly rare with many cases probably going clinically unnoticed. Clinical presentation generally consists of a painless single testicular nodule, without any associated symptoms. Imaging features typically consist of cystic components with nodular or papillary projections as well as a variable degree of calcification, vascularity, and enhancement. However, the imaging characteristics typically cannot distinguish them from other benign and malignant lesions and orchiectomy is typically performed. Generally, serous borderline tumors of the paratestis carry a favorable prognosis, with little risk of recurrence or metastasis.

CASE REPORT

A 63-year-old male reported a painless left testicular nodule for one year with recent enlargement. He described no associated

symptoms. Of note, the patient was on topical testosterone replacement for the past nine years. A physical examination revealed two small palpable left testicular nodules, with mild tenderness, upon examination.

The initial sonographic examination demonstrated two contiguous, well-circumscribed, homogeneous, isoechoic nodules, with peripheral calcification and minimal vascularity. The epicenter of these nodules projected within the margin of the testis. However, an echogenic margin supported a tunical origin [Figure 1]. The nodules measured 0.6 × 0.5 × 0.6 cm and 0.7 × 0.6 × 0.6 cm. Given the mild tenderness and negative tumor marker panel (Alpha fetoprotein (AFP), Lactate dehydrogenase (LDH), and the beta subunit of human chorionic gonadotropin (b-hCG)), a trial of oral antibiotics were given and a subsequent follow-up ultrasound (US) was performed three weeks later. The ultrasonographic findings were unchanged and magnetic resonance imaging (MRI) was obtained for tissue characterization and better anatomic localization. The MRI revealed a paratesticular bilobed lesion probably originating from the tunica albuginea, with a predominantly intermediate T1 and T2 signal, with peripheral enhancement [Figure 2]. The imaging characteristics supported a benign extratesticular etiology, such as, an adenomatoid tumor, leiomyoma or a fibrous lesion of the paratesticular tissues.

In view of the low suspicion for malignancy, a scrotal surgical approach was used. The frozen section also favored a benign

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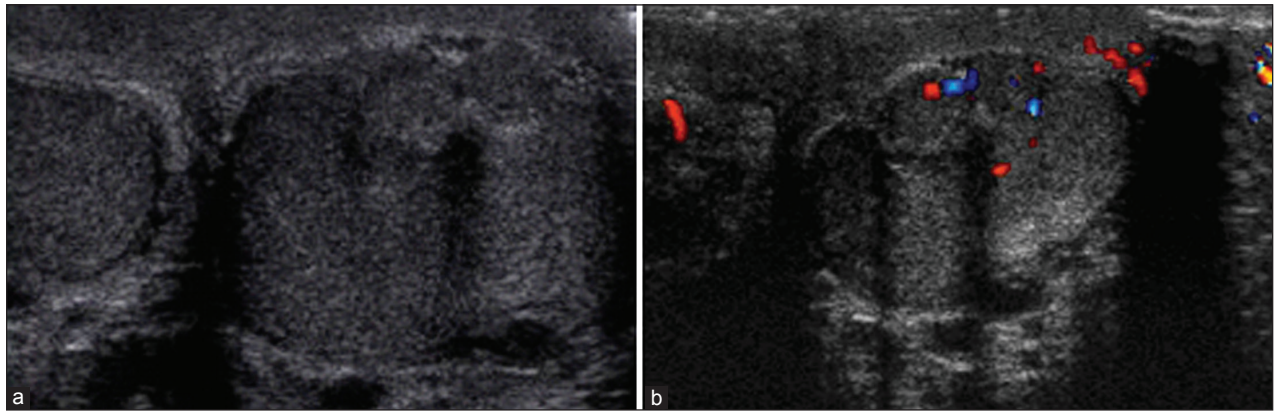


Figure 1: Transaxial sonographic image at the level of the lower testis: (a) Two subcentimeter contiguous isoechoic nodules at the periphery of the left testis demonstrate fine peripheral calcifications with acoustic shadowing. Although the epicenter of the lesions projects within the margin of the testis, the echogenic margin supports a tunical origin. (b) Doppler imaging shows minimal peripheral vascularity

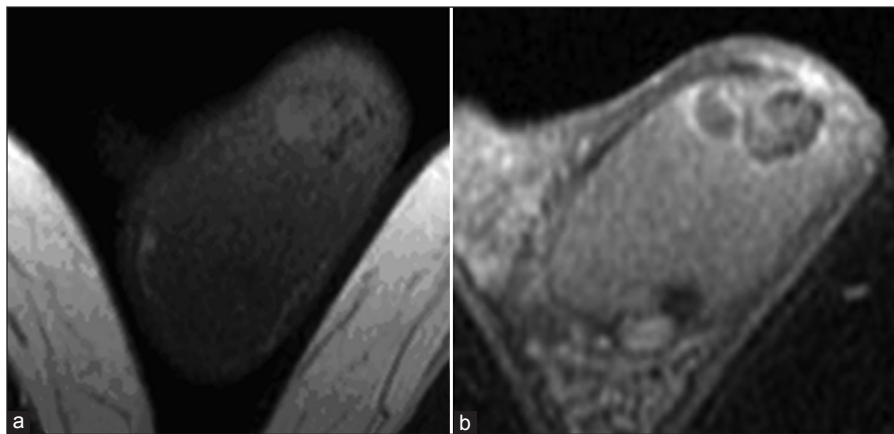


Figure 2: Axial T1 pre- and post-contrast images: (a) T1-weighted image demonstrates two small juxtaped intermediate signal lesions at the periphery of the left testis. (b) T1 post-contrast image with fat suppression demonstrates peripheral enhancement of the contiguous lesions

versus low malignant potential lesion, and therefore, a partial orchiectomy was performed. The gross specimen comprised of a yellow-tan, slightly papillary lesion, measuring $1.8 \times 1.0 \times 0.6$ cm, with negative tumor margins. Microscopically, the lesion contained cystic spaces with small papillary projections lined by cuboidal cells. Focal, small, psammoma calcifications were present. There was no evidence of mitoses or necrosis [Figure 3a]. The neoplastic cells were strongly positive for MOC-31, estrogen receptor [Figure 3b], cytokeratin 7, and WT1. Additionally, there was focal positive staining for calretinin and cytokeratin 5/6, and negative for vimentin. Combined with the tumor location, this pattern of immunostaining was consistent with a serous borderline tumor of the paratestis.

Follow-up sonographic examination at one-and-a-half years demonstrated a postoperative change without evidence for recurrence.

DISCUSSION

Given the rarity of serous borderline tumors of the paratestis,

few studies have been published. The largest case series by McClure consisted of seven cases. The mean patient age was 56 years and average tumor size at presentation was 3.5 cm. Patients typically reported a painless single testicular nodule without any associated symptoms. However, occasional dull pain and/or swelling accompanied the lesion. Laboratory serum tumor markers are typically negative, other than an occasional elevated CA-125.^[1]

Hormonal influences regarding the risk of developing serous borderline tumors of the paratestis are unknown, and therefore, the use of long-term topical testosterone in our patient is of unclear significance. However, their ovarian counterparts have been linked to oral contraceptive use in a Swedish case — control study, demonstrating an odds ratio of 1.69 (95% CI 1.06-2.79) in postmenopausal women.^[2]

To the best of our knowledge, there are no reported cases describing the imaging characteristics of serous borderline tumors of the paratestis. The lesion in our case was relatively

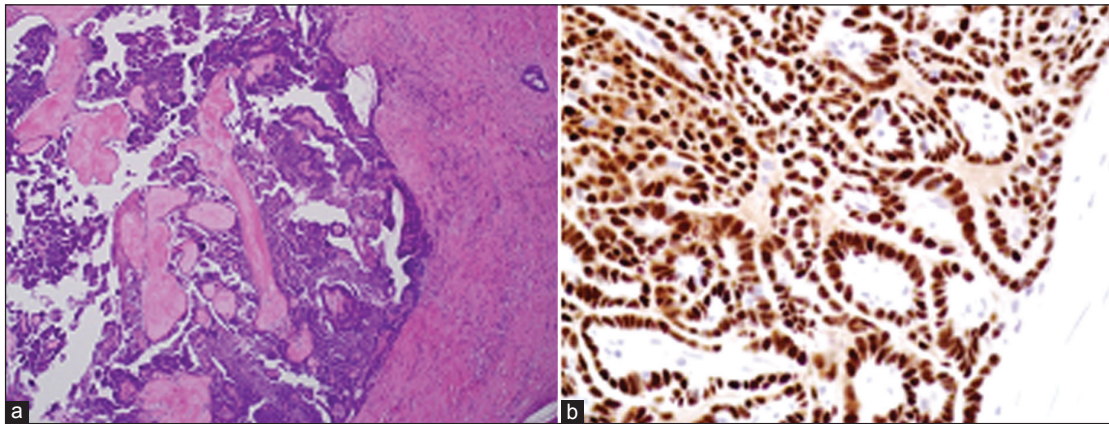


Figure 3: (a) Hematoxylin and eosin stained section at 60x demonstrates papillary projections lined by cuboidal cells projecting into cystic spaces. (b) Immunoperoxidase estrogen-receptor nuclear stained section at 500X demonstrates strongly positive neoplastic cells

homogeneous, with peripheral vascularity on both US and MR imaging. This was surprising, as the majority of serous borderline tumors were cystic in nature.^[3] Additionally, scant calcification was present in our lesion, as has been reported in other more common extratesticular lesions, such as, leiomyoma and fibrous lesions.^[4] However, there were no specific imaging features that allowed differentiation. Adenomatoid tumor and malignant processes, such as, primary or metastatic adenocarcinoma, serous papillary cancer or malignant mesothelioma can demonstrate a similar imaging appearance.

Given the lack of propensity of serous borderline tumors of the paratestis to recur or metastasize, neither pre- nor postoperative surveillance imaging is generally performed. McClure's case series yielded no cases of recurrent or metastatic disease at follow up from four months to eighteen years. Our patient had no clinical evidence of recurrence or metastatic disease one-and-a-half years post-treatment. However, a recent meta-analysis approximated a stage I ovarian serous borderline tumor recurrence rate of 0.27%.^[5] Given the small sample size of known serous borderline tumors of the paratestis, the remote possibility of recurrence cannot be entirely ruled out. Most serum tumor markers for testicular neoplasms including AFP, LDH, and b-hCG, have not been linked with serous borderline tumors of the paratestis. Given the occasional elevation of CA-125 at presentation, post-treatment monitoring may be appropriate in select cases. However, testicular examinations

performed by either the patient or physician play a central role in follow-up.

The sonographic and MR appearance of the serous borderline tumor in our case was similar to more common extratesticular lesions, such as, an adenomatoid tumor, leiomyoma or fibrous lesion. Although rare, this entity must be considered in the differential diagnosis of extratesticular scrotal lesions. However, given the nonspecific imaging characteristics, surgical management is warranted.

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