

Oncology

Leiomyoma of the testis: case report, review of literature, and discussing the role of contrast-enhanced ultrasound

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ARTICLE INFO

Keywords:

Intratesticular leiomyoma
Contrast-enhanced ultrasound

ABSTRACT

Leiomyomas are benign mesenchymal neoplasms that originate from smooth muscle cells. Leiomyoma of the testis is exceedingly rare. We present a case demonstrating a 1.3 cm hypoechoic mass of the left testis but with significant enhancement on contrast-enhanced scrotal ultrasonography concerning for a malignant neoplasm in a 33-year-old Hispanic male. However, his final pathology revealed a benign disease, specifically intratesticular leiomyoma. We discuss the role of contrast-enhanced scrotal ultrasound in this disease process.

Introduction

Leiomyomas are benign mesenchymal neoplasms that originate from smooth muscle cells and most commonly occur in the uterus, small bowel and esophagus, with increased incidence in the fourth and fifth decades of life. Presently, there is a limited number of published reports of leiomyomas affecting the male genitourinary tract. Of these, testicular leiomyomas are exceedingly rare. Here, we present a case of unilateral testicular leiomyoma in an otherwise healthy 33-year-old male presenting with a testicular mass. We discuss the utility of contrast enhanced-scrotal ultrasonography as an adjuvant imaging modality in suspiciously benign testicular lesions.

Case presentation

This is a 33-year-old Hispanic male with no significant past medical history who presented with a 6-month history of a left testicular mass. The patient denied testicular pain, history of testicular trauma or any associated symptoms. On physical exam, both testes were descended with normal spermatic cord structures. A firm, non-tender 1.5 cm mass was palpated on the superior aspect of the left testicle. Otherwise, the remainder of the left testicle was soft. The right testicle was soft, non-tender and without masses. The patient's serum tumor markers (beta-human chorionic gonadotropin, alpha-fetoprotein, lactate dehydrogenase) were all within the normal range.

Scrotal ultrasonography demonstrated a 1.3 × 1.3 cm hypoechoic mass in the left testis with a few foci of calcifications (Fig. 1a). There was

no evidence of increased vascularity or flow on Doppler evaluation (Fig. 1b). This had suggested a possible benign disease process of the testicle. Therefore, we elected to perform scrotal ultrasonography with contrast-enhancement for further evaluation of the mass. Following intravenous injection of contrast, the hypoechoic lesion demonstrated enhancement during arterial and venous phases concerning for a primary malignant neoplasm of the testicle (Fig. 2).

Computed tomography imaging of the chest, abdomen, pelvis was unremarkable for pulmonary disease or retroperitoneal lymphadenopathy. Given the patient's age, physical exam, laboratory evaluation, normal contralateral testicle, and imaging suggestive of testis malignancy, the patient then underwent a left radical orchiectomy. At the time of surgery, a firm lesion was palpable measuring approximately 1.3 × 1.1 × 1.1 cm. Gross pathology of the specimen revealed a tan, well-circumscribed tan lesion 1 cm from the hilum of the testicle with a portion abutting directly underneath the tunica albuginea (Fig. 3). Immunohistochemical stain for SMA was positive on the tumor cells, while S100, Ki67, pancytokeratin and PAP were negative. Histopathological and immunohistochemical examinations of the lesion were characteristic of leiomyoma.

Discussion

Intratesticular leiomyoma of the testicle, including bilateral leiomyomas exceedingly rare but have been reported in the literature. Ultrasonography has long been considered the gold standard in the evaluation of testicular masses given its widespread availability and

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<https://doi.org/10.1016/j.eucr.2020.101168>

Received 12 February 2020; Received in revised form 28 February 2020; Accepted 2 March 2020

Available online 1 April 2020

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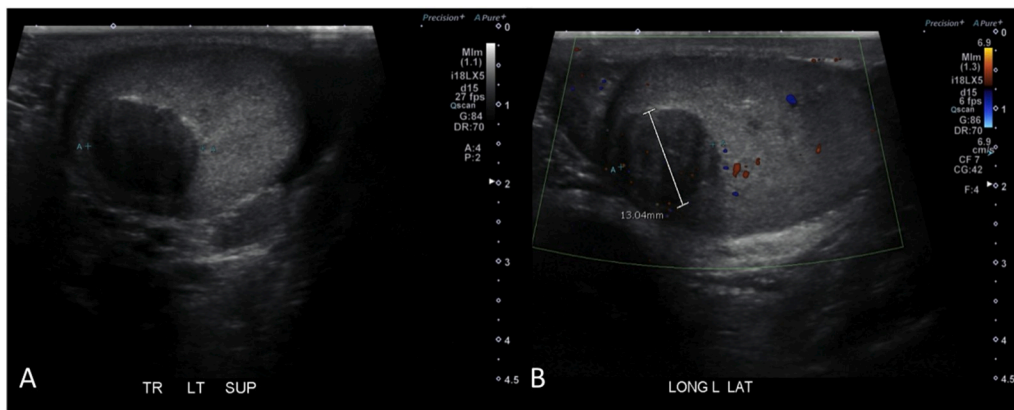


Fig. 1. A) Scrotal ultrasonography demonstrating a 1.3 cm × 1.3 cm hypoechoic lesion in the left testis. B) Color Doppler sonogram of the lesion demonstrates normal flow. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

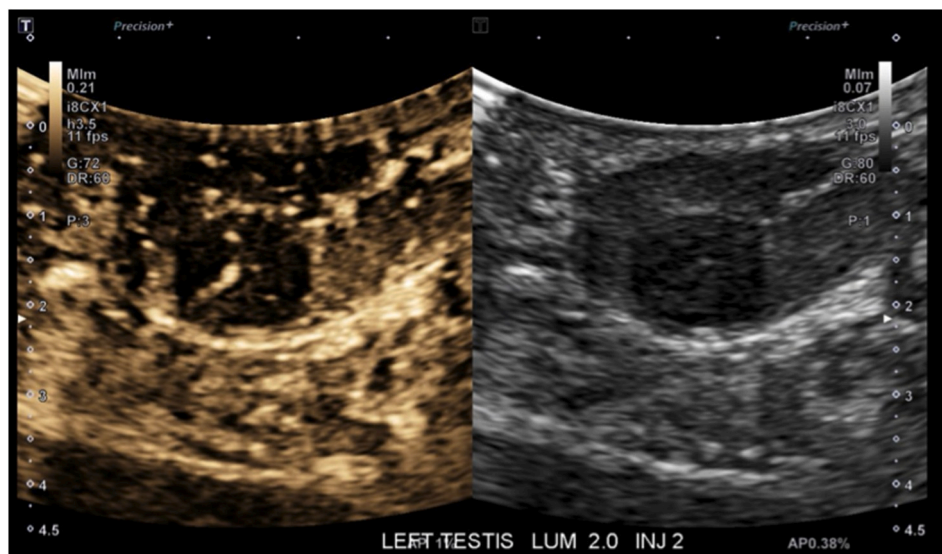


Fig. 2. Contrast-enhanced sonogram demonstrates contrast-enhancement during arterial and venous phases within the hypoechoic mass.

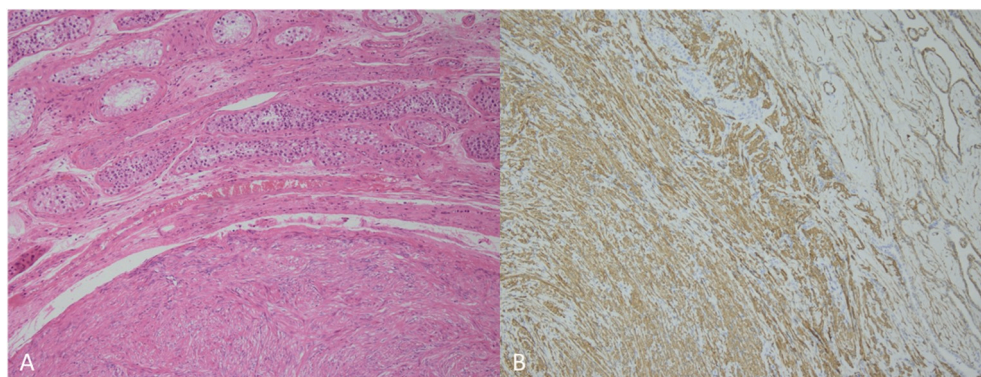


Fig. 3. A) The low power images demonstrates a well circumscribed proliferation of interlacing fascicles of eosinophilic spindle cells abutting the normal testis parenchyma and extending into the rete testis. B) The high power image demonstrates bland spindle cells that have fine nuclear chromatin without nuclear pleomorphism. Mitotic figures are rare to absent. There is no coagulative tumor necrosis present.

safety profile. Ultrasonographic features that may suggest the diagnosis of leiomyoma are the presence of a well-defined round or ovoid mass within an otherwise normal testis and no evidence of increased vascularity or flow on Doppler evaluation.¹ This patient’s ultrasound finding of a well-defined hypoechoic mass is consistent with several case reports of intratesticular leiomyoma.^{1–3} This patient’s lesion also demonstrated

evidence of testicular microlithiasis. While testicular microlithiasis often raises suspicion for malignancy, this case is the first to describe ultrasound findings testicular calcification in the setting of benign leiomyoma.

In this case, contrast-enhanced scrotal ultrasonography was utilized as an adjuvant imaging modality given suspicion for a benign disease

process, which to our knowledge, has yet to be described in previous case reports.^{1–3} Contrast-enhanced ultrasonography has been considered most valuable in the evaluation of testicular malignancy by detecting the absence of intralesional flow, which is most suggestive of a benign process.⁴ However in contrary to this principle, our patient's lesion demonstrated contrast enhancement during arterial and venous phases concerning for malignancy, however final pathologic diagnosis revealed a benign, rare testicular lesion.

In this setting, we performed a radical orchiectomy rather than a partial orchiectomy as the patient had an uninvolved contralateral testicle. This management option is consistent with all other published case reports in which radical orchiectomy was performed as the treatment of choice.^{1–3} Current recommendations for partial orchiectomy include tumor in a solitary testicle, synchronous bilateral testis tumors and tumors with high probability of being benign.⁵ The differential for a benign testicular lesion include teratoma, benign sex cord-stromal tumor (Leydig and Sertoli cell tumors), epidermoid cyst, and lipoma. However, due to the rarity of benign testicular tumors, many surgeons will opt to perform a radical orchiectomy in lieu of a partial orchiectomy. No studies have yet to identify predictors of benign testicular lesions.

O'Brien et al. and Thomas et al. both describe an intratesticular leiomyoma with "moderate" flow on color Doppler ultrasound, whereas our patient's lesion did not demonstrate a significant increase in flow on color Doppler ultrasound.^{1,2} The discrepancy in findings on conventional and Doppler ultrasound between reported cases suggests that testicular leiomyomas may have unique findings on contrast-enhanced ultrasound when compared to other sonographic techniques. Further research is necessary to better characterize the role of contrast enhanced ultrasonography in the evaluation of benign testicular lesions. We

describe our experience with contrast enhancement in a well-defined round or ovoid mass within an otherwise normal testis and negative tumor markers.

Conclusion

In conclusion, radical orchiectomy currently remains the surgical option of choice for intratesticular leiomyoma given the limited ability of imaging to distinguish a benign from a malignant tumor. Contrast-enhanced ultrasound may aid in identifying a unique sonographic profile for this rare lesion.

Declaration of competing interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

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