

Case Report

Angioleiomyoma of the scrotal wall

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Received December 14, 2009; accepted May 28, 2010

Abstract

Angioleiomyoma of the scrotum is a rare benign lesion which can mimic a paratesticular tumor. Any solid mass within the scrotum is considered malignant until proven otherwise. Here, we present a case of an angioleiomyoma of the scrotum in a 33-year-old male who presented with painful scrotal mass. Scrotal ultrasonography demonstrated a solid mass in the scrotum, and surgical excision was carried out. Pathologic examination revealed that the tumor was angioleiomyoma.

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Keywords: Angioleiomyoma; Angiomyoma; Leiomyoma; Scrotal; Tumor

1. Introduction

Angioleiomyoma of the scrotum is a rare lesion. There are controversies about the identification of this entity. Smooth muscle hyperplasia, leiomyoma, leiomyomatous hamartoma are synonyms used in the literature. Although this is a rare entity, it should be considered in the differential diagnosis of paratesticular malign tumors.¹ To distinguish it from malignancy, long-term history and normal tumor markers should be kept in mind for initial diagnosis. However, the certain diagnosis is made by pathology. We present a case of angioleiomyoma arising from the scrotal wall.

2. Case report

A 33-year-old male presented with a painful right scrotal mass. The medical history revealed that he had had the scrotal mass for 6 years, whereas the pain had emerged 2 months before presentation. Physical examination revealed a non-tender mass occupying the right hemiscrotum. Scrotal

ultrasonography demonstrated a 7×9 cm heterogeneous solid mass in the scrotum. Serum levels of human beta-chorionic gonadotropin and alpha-fetoprotein were normal (<1.00 mIU/mL and 2.37 IU/mL, respectively). During surgery, right atrophic testis was observed in the inguinal canal and intensive vascular mass was seen in the scrotal wall. The mass was apart from both testicles. The result of intra-operative frozen evaluation was a fibrovascular connective tissue. Scrotal mass was removed followed by right high inguinal orchiectomy because of atrophic testis.

Pathological findings: On longitudinal sectioning, a $2.5 \times 1.5 \times 1$ -cm-long atrophic testicle was detected. Epididymis and vas deferens were normal. The scrotal mass was consisted of hypertrophic smooth muscle fibers intermingled with dilated vascular structures. Immunohistochemical examination revealed strong CD31 and CD 34 reaction for smooth muscle and endothelium (Figs. 1 and 2). The case was reported as angioleiomyoma.

3. Discussion

Intrascrotal lesions with solid component determined on ultrasonography predict malignancy potential. Differential diagnosis is very important in distinguishing between benign and malignant forms.¹ Benign lesions arising from scrotal wall smooth muscle are rare in the literature. Less than fifty cases

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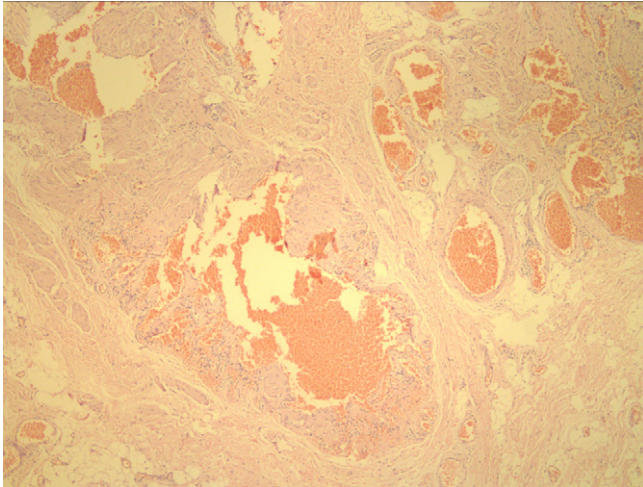


Fig. 1. In histopathological examination, tightly packed asymmetric and thickened vascular network were remarkable (haematoxylin-eosin $\times 40$).

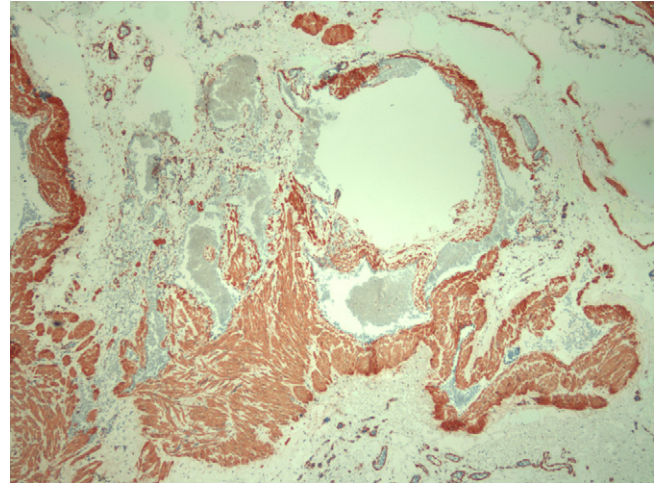


Fig. 2. Vessels and smooth muscle fibers have positive reaction with Actin (immunohistochemistry, smooth muscle actin, $\times 40$).

have been published to date.^{2–4} Most benign lesions derived from the spermatic cord and epididymis. Eleven percent of benign epididymal tumors are leiomyomas.⁵ Because of their embryological background, tumors arising from the male genitalia structures are mesodermal in origin.⁶

These benign lesions tend to be asymptomatic and painless.⁷ Although the patient in this report had painful scrotal mass for 2 months, he said that he had had painless scrotal mass for 6 years. In the literature, patients with 10-, 20-, or even 30-year histories before surgical excision have been described.⁵ Tumor markers of testicular cancer were normal in our case and other cases in the literature.^{5–8}

Ultrasound scan can provide useful information in the diagnosis of scrotal mass. However, it is difficult to reliably identify malignant scrotal mass on the basis of sonographic features alone.^{9,10} Radical orchiectomy may be required, because preoperative and intra-operative findings could not be helpful in excluding malignancy.⁸ However, frozen section is helpful to discriminate malignancy from benign lesion to prevent unnecessary orchiectomy.

In conclusion, to our knowledge, only a few cases of scrotal leiomyoma have been reported in the literature so far. Although this is a benign lesion, it mimics malignancy because of solid components on the basis of ultrasonography. Long-term history and normal tumor markers are essential characteristics to distinguish benign tumors from malignancy;

however, definitive diagnosis is established by pathologic evaluation.

References

1. Barton JH, Davis Jr CJ, Sesterhenn IA, Mostofi FK. Smooth muscle hyperplasia of the testicular adnexa clinically mimicking neoplasia: clinicopathologic study of sixteen cases. *Am J Surg Pathol* 1999;**23**: 903–9.
2. Kato Y, Hori J, Taniguchi N, Hashimoto H, Kaneko S, Yachiku S. Solitary genital leiomyoma of the tunica dartos: a case report and review of the literature in Japan. *Hinyokika Kiyo* 2005;**51**:699–701.
3. Cabello Benavente R, López Martínez-Bernal B, Verdú Tartajo F, Monzó JI, Castaño González I, Moralejo Gárate M, et al. Giant bizarre scrotal leiomyoma. *Arch Esp Urol* 2004;**57**:847–51.
4. Kim NR, Sung CO, Han J. Bizarre leiomyoma of the scrotum. *J Korean Med Sci* 2003;**18**:452–4.
5. Yeh HC, Wu WJ, Lee YC, Chang TH, Huang CH, Li CC. Leiomyoma of the epididymis: a case report. *Kaohsiung J Med Sci* 2006;**22**:519–23.
6. Ghei M, Arun B, Maraj BH, Miller RA, Nathan S. Case report: angio-leiomyoma of the spermatic cord: a rare scrotal mass. *Int Urol Nephrol* 2005;**37**:731–2.
7. Masood J, Voulgaris S, Atkinson P, Carr TW. A rare symplastic or bizarre leiomyoma of the scrotum: a case report and review of the literature. *Cases J* 2008;**9**:381.
8. Minami M, Inoue W, Uchida M. Leiomyoma of the scrotum: a case report. *Hinyokika Kiyo* 1999;**45**:207–9.
9. Hertzberg BS, Kliever MA, Hertzberg MA, Distell BM. Epididymal leiomyoma: sonographic features. *J Ultrasound Med* 1996;**15**:797–9.
10. Leonhardt WC, Gooding GA. Sonography of epididymal leiomyoma. *Urology* 1993;**41**:262–4.