



# Intra-testicular angioleiomyoma. A case report of a rare male urological entity

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## ABSTRACT

Angioleiomyoma (vascular leiomyoma) is a common benign smooth muscle tumor, often affecting the soft tissue of the extremities, head, and neck regions. However, testicular involvement is an exceedingly rare event. Herein, we present an unusual case of testicular angioleiomyoma. To the best of our knowledge, this is the third reported case in English literature.

## 1. Introduction

Angioleiomyoma is a benign neoplasm, originates from the vascular smooth muscle. Commonly affects people in their third to fifth decades of life.<sup>1</sup> These tumors often occur in the subcutis and deep dermis of the extremities, and to a lesser extent, of the head, neck, and trunk regions.<sup>2</sup> However, benign smooth muscle tumors are exceedingly rare in the testes.

## 2. Case presentation

A 41 year-old-male, medically free, presented to the hospital with left testicular pain. Clinical examination revealed left testicular tenderness and swelling, which did not transilluminate. Ultrasound examination of the left testis showed a well-defined testicular mass, 5.0 cm in the greatest dimension. Serum alpha-fetoprotein and beta-human chorionic gonadotropin were normal. The patient underwent left orchiectomy.

The orchiectomy specimen weighed 30 g, the testis measured (8.0 × 5.5 × 4.0 cm), epididymis measured (1.0 × 1.0 × 0.5), and spermatic cord measured (11 cm in length × 2 cm in diameter).

The cut sections of the testis showed a well-defined, gray-tan, rubbery intra-testicular mass located at the lower pole, measured (5.5 × 5.0 × 4.0 cm). The mass appeared limited to the testis and did not grossly extend into tunica albuginea or the epididymis. The remainder of the testicular parenchyma is grossly unremarkable. The spermatic cord was grossly unremarkable. The mass was well sampled and submitted for pathological examination.

Microscopic examination revealed a well-defined tumor composed of

interlacing fascicles of bland spindle cells with cigar-shaped nuclei, eosinophilic cytoplasm with focal epithelioid morphology (Fig. 1 A). The background showed prominent, variable-sized vascular proliferation and in focal areas, the spindle cells appeared in close vicinity to the vascular wall (Fig. 1 B). Rare mitotic figures were observed. There was no evidence of pleomorphism or necrosis. There was no evidence of Germ cell tumor, or Germ cell neoplasia in situ in the surrounding testicular tissue.

The tumor cells were positive for Caldesmon (Fig. 1 C), smooth muscle actin (SMA), muscle-specific actin (MSA), desmin, and vimentin. CD34 highlights rich vascular background (Fig. 1. D). The tumor cells were negative for HMB45 (Fig. 1E), S100 (Fig. 1F). Melan A, inhibin, myoglobin, Myo D1, myogenin, EMA, and Pankeratin.

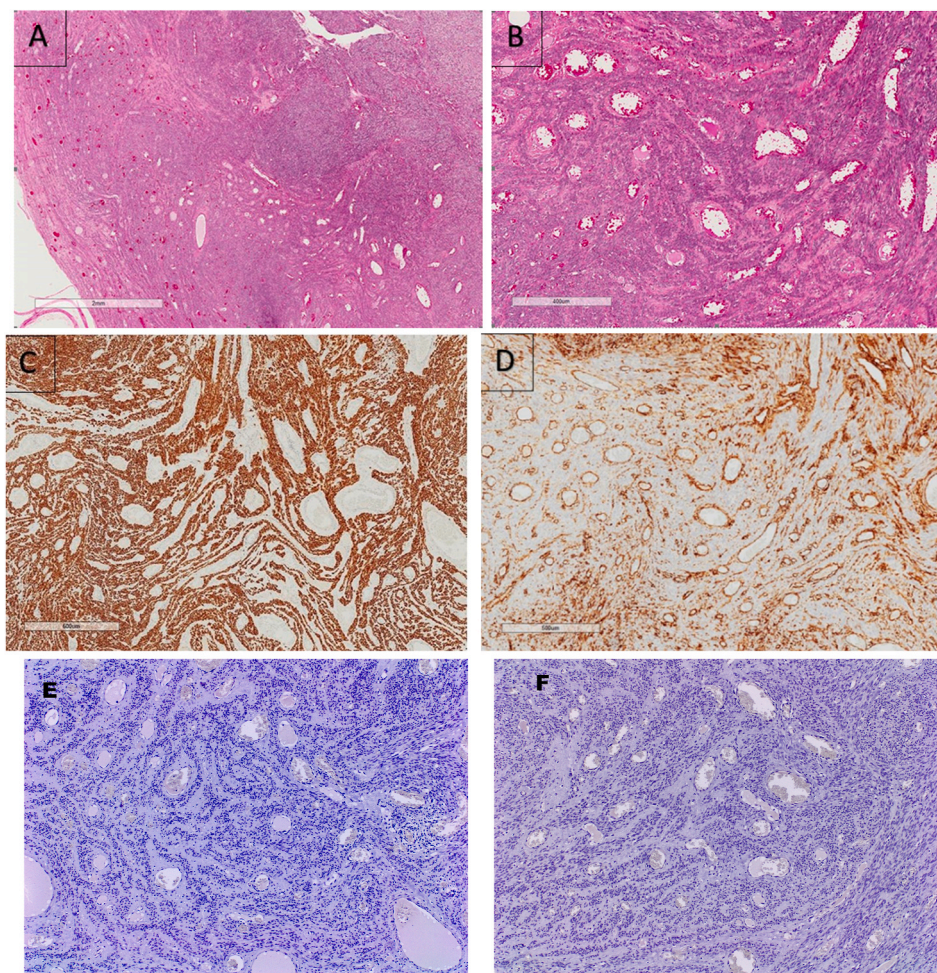
The postoperative period was uneventful, and the patient was discharged home.

## 3. Discussion

Benign smooth muscle tumors are classified based on histological features into leiomyoma and angioleiomyoma. Angioleiomyoma is a common benign smooth muscle tumor usually arising from the vascular smooth muscle, commonly in the third to fifth decades of life. These tumors are typically painful and often occur in the subcutis and deep dermis of the extremities, and to a lesser extent, of the head, neck, and trunk regions. They also have been reported in unusual sites, including the oral cavity and auricle.<sup>3</sup>

Benign smooth muscle tumors can theoretically arise anywhere in the body. The origin of intratesticular angioleiomyoma is controversial. Possible sites of origin include vascular smooth muscle cells and

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**Fig. 1.** Hematoxylin and eosin stained sections (H&E) and immunohistochemistry studies. (A): well defined tumor composed of bland spindle cells admixed with variable sized blood vessels (H&E; 10×); (B): Tumor cells arrange in fascicular pattern and appeared in close vicinity to the vascular wall (H&E; 20×); (C): Tumor cells shows diffuse cytoplasmic expression for h-Cal-desmon (20×); (D): CD34 (20×) highlights vascular-rich background. (E&F): HMB45 and S100 (20×) respectively with negative expression.

contractile cells of the seminiferous tubules.

Intratesticular benign smooth muscle tumors are extremely rare, with only a few cases of testicular leiomyoma reported in the English literature. An extensive search in the literature performed; we could identify only two reported cases of intratesticular angioleiomyoma by Robert Lavis and Goran Stimac. Both cases were solitary. No known association with similar pathology at other body sites or contralateral testis.

The differential diagnosis of testicular tumors with spindle cell morphology includes a wide variety of benign and malignant neoplasms such as leiomyoma, angioleiomyoma, sex cord stromal tumor, peripheral nerve sheath tumors, pecoma, melanoma, sarcoma and somatic-type malignancy in germ cell tumor.

Radiologically, the angioleiomyoma at other body sites usually manifests as a well-defined multi-lobulated mass with solid and cystic areas. Nevertheless, in the testis, there is no definite diagnostic radiological feature to arrive at the correct diagnosis angioleiomyoma or to differentiate it from germ cell tumors.

In the light of the documented low sensitivity and specificity of the contemporary diagnostic radiological modalities to accurately delineate intratesticular masses into benign and malignant categories<sup>4</sup> and the substantial rate of malignancy documented for the intratesticular masses, up to 90% in some studies,<sup>5</sup> the surgical excision of intra-testicular masses remains the mainstay of treatment.

Given the limited number of reported cases of intratesticular angioleiomyoma, it is difficult to predict the clinical behavior, the malignant potential, and the prognosis for these tumors. Therefore, radical orchiectomy remains the treatment of choice for such lesions.

Nevertheless, more studies are warranted to further investigate the rule of partial orchiectomy as a better treatment modality.

#### 4. Conclusion

Although benign testicular smooth muscle tumors are exceedingly rare, it should be considered in the differential diagnosis of the intratesticular masses. Further analysis of large number of cases is required to further delineate the clinical behavior of testicular angioleiomyomas.

#### Patient consent statement

This is a single case report with no identifiable patient information/ characteristics are included in the case report. Therefore, there is no need to obtain the patient consent.

#### Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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