

Oncology

Scrotal leiomyoma a rare benign intra-scrotal mass could lead to unnecessary orchiectomy

Ali Ariafar^a, Mohammmdreza Soltani^b, Fatemeh Khajeh^{c,d,**}, Shahryar Zeighami^b, Nima Naghdi sedeh^b, Behnoosh Miladpour^e

^a Urology Oncology Research Center, Shiraz University of Medical Sciences, Iran

^b Shiraz University of Medical Sciences, Urology Department, Iran

^c Shiraz University of Medical Sciences, Pathology Department, Iran

^d Fasa University of Medical Sciences, Pathology Department, Iran

^e Fasa University of Medical Sciences, Urology Department, Iran



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ABSTRACT

Leiomyoma is a benign tumor originating from smooth muscle cell, Mostly from uterus. However, in men is a very rare entity. Scrotal leiomyoma is a very rare tumor. Here we presented a case of scrotal leiomyoma in a 71-year-old man. He presented with a slowly growing, painless mass and heaviness in the left testis for 10 years. Due to huge size, testicular attachment and preoperational diagnosis of atypical leiomyoma/leiomyosarcoma, orchiectomy was performed. Pathology report diagnosed leiomyoma. We suggest frozen section diagnosis as a useful tool, to prevent unnecessary procedure.

Introduction

Leiomyoma is a benign neoplasm of the smooth muscle. Leiomyomas can arise anywhere in the body, but scrotal leiomyoma is a very rare entity which arises from the dartos muscle, a kind of subcutaneous smooth muscle in the scrotum.¹⁻³ In 1937, Stout reported five case of scrotal leiomyoma in the literature. In 1964,² Siegal and Gaffey reported ten cases between 1905 and 1975 from the records of the Mayo Clinic.¹ The smooth muscle neoplasms of the scrotum were first described by Fosters in 1858.⁴ Superficial leiomyoma of the skin and subcutaneous tissue were classified into three groups based on the location of the smooth muscle origin I. Piloileiomyoma (originate from arrector pili muscle), II. Angioleiomyoma (originate from smooth muscle of blood vessel) III. Genital leiomyomas (originating from the smooth muscle of the nipple, vulva and scrotum).⁴ The genital leiomyoma has the lowest incidence rate in all cutaneous ones. Cutaneous leiomyoma consisted of only 5% of all leiomyoma.^{1,5}

Siegel and Gaffey reported the prevalence rate of scrotal leiomyoma as 1/1000 of all scrotal tumors by a review of 11,000 scrotal tumor cases.¹

Case presentation

Our patient is a 71-year-old man who referred to Ali-Asghar clinic affiliated to Shiraz University of Medical Sciences because of the left testicular enlargement and heaviness for 10 years. Physical examination revealed a non-tender, firm semi-mobile mass which was attached to the overlying skin and testis. No lymphadenopathy was identified. Testicular sonography was in favor of a scrotal mass, but because of testicular attachment, orchiectomy was preferred for him with an overlying ellipse of the scrotum. The specimen was sent for pathology examination. Grossly, the tumor was an oval mass measuring 11Cm in greatest diameter, which was attached to the overlying skin. Because of the tumor pressure on the left testis, the left testis became atrophic. Cut sections showed a solid grey-white mass with whorling appearance (Fig. 1). Hematoxylin and eosin stained sections was in favor of benign spindle cell neoplasm. The individual cell showed eosinophilic cytoplasm with elongated blunt ended nucleus in a whorling and interlacing pattern. No mitosis was seen. No nuclear pleomorphism was seen. No infiltrative border was seen (Fig. 2). Immunohistochemistry (IHC) for Smooth Muscle Actin antigen (SMA) and Desmin antigen was positive

* Corresponding author. Fasa University of Medical Sciences, Pathology Department, Fasa Medical School, Fasa University of Medical Science, No: 162, 15th Alley, Zargari Avenue, Shiraz, 7183934735, Iran.

E-mail addresses: Ariafora@sums.ac.ir (A. Ariafar), soltani732003@yahoo.com (M. Soltani), F.khaje@fums.ac.ir (F. Khajeh), Zyghamishahryar@yahoo.com (S. Zeighami), nimanaghdi@yahoo.com (N. Naghdi sedeh), miladpour90@yahoo.com (B. Miladpour).

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Fig. 1. Gross appearance of scrotal leiomyoma, Arrow: shows the tumor attachment to a piece of scrotum.

(Fig. 3). IHC for S-100 protein, CD34 and cytokeratin was negative.

Hence, the diagnosis of smooth muscle origin was approved for this tumor. By considering morphologic criteria and IHC staining, with the diagnosis of benign leiomyoma the patient was discharged, but kept on follow-up.

Discussion

Scrotal leiomyoma is a rare, usually painless, and slow growing mass. These patients usually delay clinic referral with an average of 6–7 years, after they noticed the testicular mass, when the testicular heaviness and large size are the main chief complains.^{1,3,5}

Scrotal smooth muscle tumors are classified into 3 groups, benign, atypical and malignant based on 4 criteria.

1. Size, more than 5 Cm
2. Infiltrated margin
3. Equal or more than 5–10 mitosis per 10 HPF (high power field)
4. Moderate cytologic atypia

The tumors with only any one of the above criteria are benign. The tumors with any two criteria are atypical. The tumors with any three or fulfilling criteria are considered malignant (leiomyosarcoma).⁴

In our case, the tumor size was more than 5cm, but the other criteria, cytologic atypia, mitosis and infiltrated margin are absent; hence, the diagnosis of benign leiomyoma is made. Pathologic diagnosis of smooth muscle tumor and careful examination for presence of the criteria mentioned before is an essential need for diagnosis and subsequent treatment. The pathologist especially must look for mitotic figure because Newman and Fletcher mentioned the presence of any mitotic activity as a criterion of malignant potential.⁵

The painless nature of a scrotal leiomyoma is a reflection of slow growing nature of leiomyoma, which caused pushing the nerve trunk outward, rather than compressing on it.⁵ Because of painless and slow growing nature of the mass, the usual interval between patient recognition and surgical removal is wide from 2 to 20 years with average of 6.7 years.^{1,5} There are a wide range of differential diagnosis such as fibroma and sebaceous cyst. If the mass is painful schwannoma and if accompanied with scrotal ulceration squamous cell carcinoma (SCC) are in the top of the clinical differential diagnosis.^{3,5} If there is any adhesion to the testis, the possibility of atypical leiomyoma or leiomyosarcoma should be considered. The management of benign and atypical leiomyoma is the same, simple excision of the tumor requires long term follow up for atypical leiomyoma. However, leiomyosarcoma needs aggressive management, which requires wide resection with 3–5 cm safe margin. Including subcutaneous tissue, fascia and negative margin confirmation on pathology report.⁵ In the management of leiomyoma,

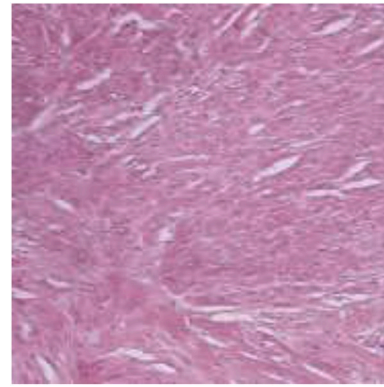


Fig. 2. Microscopic appearance of scrotal leiomyoma, note the bland looking appearance of spindle shape cells arranged in whorling appearance. H& E stain, magnification:400.



Fig. 3. Diffuse positive immunohistochemical (IHC) staining for Smooth Muscle Actin Antigen (SMA) in scrotal leiomyoma, magnification:100.

we should avoid radiation because of inducing effect of malignant transformation in the smooth muscle tumors. In the follow up, we need to rule out the recurrence; if there is recurrence, true investigation should be considered to rule out any malignancy. In our case, we need to consider the other smooth muscle tumors in differential diagnosis of leiomyoma. The essential point is careful pathologic examination to rule out the malignant criteria, especially mitotic activity, to avoid misdiagnosis and erroneous management.

Conclusion

Scrotal leiomyoma is a very rare benign tumor. It is difficult to perform an exact preoperative diagnosis; only histopathological examination can prove the diagnosis of the leiomyoma. In this case, radical orchidectomy was performed because of large intrascrotal mass and possibility of malignant neoplasm, also rarity of reported benign, intrascrotal neoplasm. We suggest preoperative frozen section diagnosis, with re-emphasis to looking for mitosis and cytologic atypia. This needs a close interaction and cooperation between urologist and pathologist. The urologist should also be aware of above mentioned criteria such as size and infiltrated margin to take a good excisional sample for frozen diagnosis. The frozen section diagnosis may be able to prevent unnecessary orchiectomy especially in young men.

Declaration of competing interest

The authors declare that they have no competing interests.

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List of abbreviation:

IHC	Immunohistochemistry
SMA	smooth muscle antigen
HPF	high power field
Cm	Centimeter
SCC	squamous cell carcinoma

Ethics approval and consent to participate

There is no need to ethical approval relevant to our case report. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Consent for publication

A copy of the written consent from the patient for publication of any data or image or video is available for review by the Editor-in-Chief of this journal.

Availability of data and material

All of the data and material is available in our clinic and lab.

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Authorship contributions

All of the authors have contributed in writing the report. Dr SH. Zeighami, Dr A. Ariafa, Dr MR. Soltani and Dr N. Naghdi collected the data and visits the patient. Dr A. Ariafar was the main surgeon responsible for all operations and clinical management of the patients. Dr F. Khajeh was the pathologist and diagnosed the case. Dr B. Miladpour assisted in diagnosis. All authors have read and approved the final manuscript.

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