The New Journal of Urology
eISSN 3023-6940
doi: https://doi.org/10.33719/nju1348373

CASE REPORT

Testicular Angiomyolipoma: A Case Report and Review of the Literature

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Submitted: *2023-08-23*
Accepted: *2023-10-19*

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Abstract

Angiomyolipoma (AML) is a benign mesenchymal tumor, that is composed of thick-walled blood vessels, smooth muscle cells and adipose tissue. They are commonly seen in kidneys, testicles are not a typical location for AML's; many solid testicular tumors are germ cell malignancies. Here we report the sixth testicular AML case in the literature, to our knowledge. An 80-year-old male undergone bilateral orchiectomy with the diagnosis of local invasive prostate cancer, without any testicular symptoms. Pathological analysis demonstrated a tumor, involving nearly the whole left testicle, composed of muscle cells, vessels and fat tissue, diagnosed as testicular AML. In this article our case is reported and we also make a literature review about testicular AML's.

Keywords: angiomyolipoma, angiolipoma, testis, orchiectomy

INTRODUCTION

Angiomyolipoma (AML) is a rare tumor of mesenchymal origin, composed of adipose tissue, thick-walled blood vessels, and smooth muscle cells. They are sporadic in most of the cases, however less than a quarter of the cases might be a component of tuberous sclerosis complex (TSC), as a result of mutations in TSC1 or TSC2 tumor suppressor genes. Kidney is the most frequent solid organ that AML's locate, followed by the liver [1]. Testicles are very atypical organs for AML location; to our knowledge only five testicular AML's and an intratesticular angiolipoma without the muscle cell component have been reported [2,3,4,5,8,9]. Here, we report the sixth case which is the second incidental testicular AML.

CASE PRESENTATION

An 80-year-old male patient was admitted to our outpatient clinic with a diagnosis of locally invasive prostate cancer. Preoperative physical examination of the testes was completely normal. Due to his age and stage, bilateral orchiectomy was planned instead of radical surgery for treatment. Testicular imaging was not performed before bilateral orchiectomy because of the absence of pathological findings on physical examination. In the routine pathological examination of the patient after orchiectomy, an incidental angiomyolipoma was detected in the left testicle.

Cite: Altan, SA, Karaman, ZS, Adsan O. Testicular Angiomyolipoma: A Case Report and Review of the Literature. New J Urol. 2024;19(1):34-37. doi: 10.33719/nju1348373

Macroscopic Findings

The dimensions of the right testicle were 8x6x5 cm's, the left one was smaller, 6x4x3 cm's. The right testicle was normal in cross section, however the cross-section of the left testicular specimen was rich in fat tissue and the tumoral lesion was macroscopically involving the whole testicle and the residual testicular tissue was not noticeable.

Microscopic Findings

Microscopic examination showed well circumscribed, yellow colored, tumor which was composed of numerous large vessels, mature fat tissue and bundles of smooth muscle fibers (Figure 1). Smooth muscle fibers were concentrated around the dilated thick-walled hyalinized vessels, and surrounded by adipocyte islands. All three components (angio-myo-lipoma) were mature histologically, and any cytological atypia, mitosis, pleomorphism or necrosis was not detected in the lesional cells. No sarcomatoid changes or hemorrhage were present. There were atrophic remnants of the seminiferous tubules around the tumor. In epididymal sections, lumens of the ducts were empty, any spermatocytes had not been observed.

Immunohistochemistry showed that the tumor cells were actin positive (Figure 2), and HMB-45 negative, only in a very small area showed a pale HMB-45 positivity (Figure 3).

Discussion and Review of the Literature

AML is a benign tumor, more commonly seen in women, and after the fifth decade. The cellular origin of testicular AML remains unknown, perivascular epitheloid cells are thought to be the source [5].

Testis is not a well-known site for AML's, testicular AML is not listed in World Health Organization (WHO) histological types of testicular tumors; they are commonly seen in kidneys (77%), and secondly in liver (14%) [3,6]. Only five cases of testicular AML have been reported to date.

Although AML's are benign tumors, epitheloid AML (EAML) was reported as a metastasizing sub-type, also may present nuclear atypia [6].

Five cases have been reported in the literature, summarized in (Table 1).

The tumor in the present case was nearly involving the whole testicle.

There is not an identical treatment recommendation for testicular AML's, since the majority of testicular tumors are germ cell tumors (GCT), which should be diagnosed and treated promptly; radical orchiectomy is the standard treatment option. Testicular AML might be a challenging pathological diagnosis, as a result of its rarity.

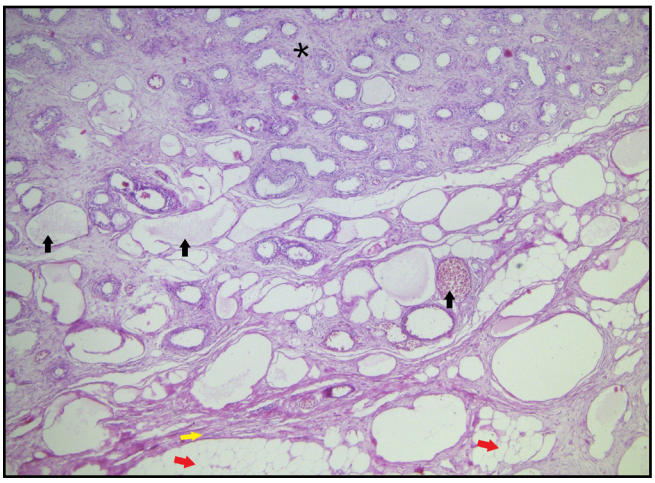

Figure 1. Large vessels, mature fat tissue and smooth muscle fibers. Black arrows refer to vessels, yellow arrow refer to smooth muscle cells, asterisk refers to normal testicular tissue, red arrows refer to fat tissue.

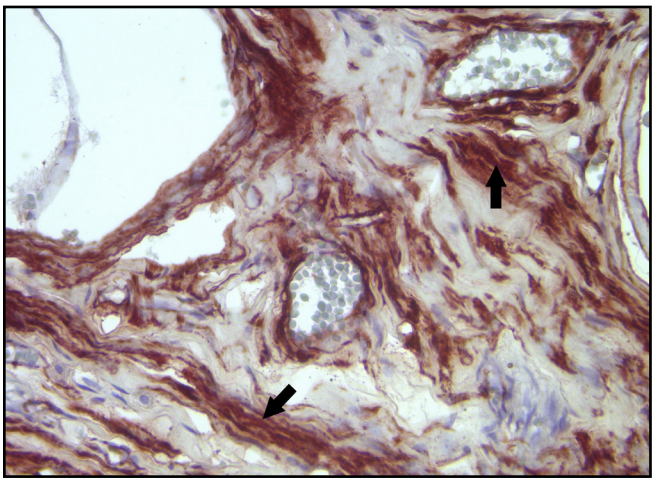

Figure 2. Actin, chromogen DAB. Black arrows refer to smooth muscle cells.

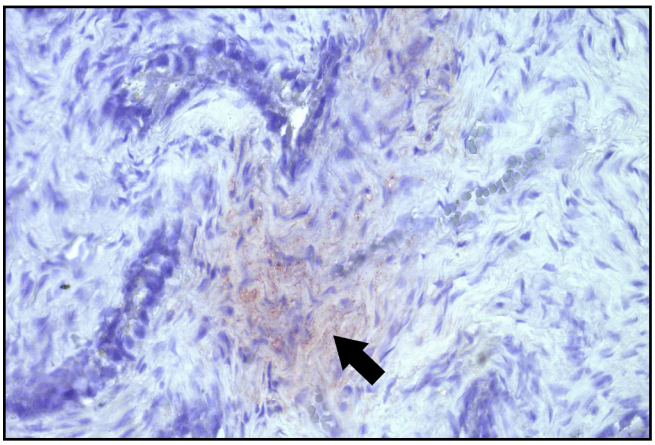

Figure 3. HMB-45, chromogen DAB. Black arrow refers to the pale HMB-45 positive area.

Ultrasound is generally the choice of imaging of the testicles, besides the physical examination. Other imaging modalities are seldom used. In our case orchiectomy is performed because of the diagnosis of local invasive prostate cancer, so neither detailed examination of testicles nor the ultrasound had not been performed. Although perioperatively any tumoral lesions were not visible or palpable in the testicles.

The role of frozen section analysis is limited in testicular lesions. All large testicular tumors are being treated by radical orchiectomy as a standard treatment modality. However, if the lesion is non-palpable, incidentally diagnosed in a routine imaging, the patient does not belong to any risk groups for testicular cancer, and the tumor is small enough (<1.5 cm's in diameter), orchiectomy may be an overtreatment, thus intraoperative frozen section may be an option in these cases [5].

Endothelial marker (CD34) and smooth muscle actin are the immunohistochemical (IHC) markers that are typically expressed in all AML's, we used the actin, and the result was positive. Another IHC marker that we use, HMB-45 is a monoclonal antibody that reacts against an antigen present in melanocytic tumors such as melanomas, and HMB stands for Human Melanoma Black. It is interestingly positive in renal and liver AML's, and negative in cutaneous and testicular AML's [5]. Renal positive HMB-45 used in differential diagnosis of resected benign renal specimens [7]. Our case showed general HMB-45 negativity, except a focal pale positive area, that shored up our diagnosis of testicular AML. It is thought that intense HMB-45 expression in kidney or liver AML cases is related to poor outcomes such as malignancies [3]. In our case HMB-45 was negative, which may indicate testicular AML's do not carry the risk of malignancy. That can lead us to think there might be significant biological differences between testicular AML and typical AML in the kidney [3]. In the cases mentioned above HMB-45 negativity was observed. Further research is required in order to make concrete deductions.

Because of the lacking cumulative data about testicular AML's, long term follow-up might be a safe option, however our patient died in several months after bilateral orchiectomy because of prostate cancer.

Despite of the fact that ages of the patients differ significantly according to the literature, because of the limited number of observed cases, making a decision about age group for testicular AMLs can not be done.

Based on observation, some cases presented with symptoms such as pain and swelling while others presented with no symptoms. Also some symptoms such as pain and swelling are common for testicular diseases. Thus testicular AML should be considered during diagnosis.

Table 1. Features of the reported testicular AML cases

Publication	Age	Symptoms	Size	Additional pathology
Lane TM et al. (2004)	56	no (incidental)	1.6 cm at the upper pole of right testicle	left hydrocele
Saito M et al. (2008)	22	pain + swelling	3.0 cm irregular margin tumour at left testicle	para-aortic LAP (1 cm)
Giulianelli R et al. (2012)	53	pain + swelling	not identified, left testicle	not present
Ceifo W et al. (2015)	25	pain + swelling	not identified, right testicle	right atrophic testicle
Waked H et al. (2020)	75	Painless swelling + hardness	3,5x3x2,6 cm, right testicle	Moderate hydrocele
Presented case	80	no (incidental)	not identified, left testicle	prostate cancer

CONCLUSION

Testicular AML is a very rare tumor; however, it may be in the differential diagnosis list of an urologist or pathologist in case of an atypical testicular mass. Each case should be diagnosed, treated and followed up individually according to clinical and pathological findings.

ACKNOWLEDGMENTS

Conflict of Interest: Authors declare no conflict of interest

Funding: None.

REFERENCES

[1] Lin C, Jin L, Yang Y, Ding Y, Wu X, Ni L, et al. Tuberous sclerosis-associated renal angiomyolipoma: A report of two cases and review of the literature. Molecular and Clinical Oncology 2017;7:706–8. https://doi.org/10.3892/mco.2017.1377

[2] Lane T, Masood J, Shah NA, Koye B, Hill JO. Angiomyolipoma of the testis. The Journal of Urology 2004. https://doi.org/10.1097/01.ju.0000107361.95413.a4

[3] Saito M, Yuasa T, Nanjo H, Tsuchiya N, Satoh S, Habuchi T. A case of testicular angiomyolipoma. International Journal of Urology 2008;15:185–7. https://doi.org/10.1111/j.1442-2042.2007.01955.x

[4] Giulianelli R, Albanesi L, Attisani F, Brunori S, Gentile B, Mavilla L et al. A case of angiomyolipoma of the spermatic cord and testicle. Arch Ital Urol Androl. 2012;84(3):165-166. Retrieved from: https://pubmed.ncbi.nlm.nih.gov/23210412/

[5] W C, Qadri IA, Malik A. Angiomyolipoma of the testis: a case report. Medical & Surgical Urology 2015. https://doi.org/10.4172/2168-9857.1000155

[6] Yang L, Feng X, Shen SS, Shan L, Zhang H, Liu X, et al. Clinicopathological analysis of 156 patients with angiomyolipoma originating from different organs. Oncology Letters 2012;3:586–90. https://doi.org/10.3892/ol.2012.554

[7] Yaldiz M, Kilinc N, Ozdemir E. Strong association of HMB-45 expression with renal angiomyolipoma. Saudi Med J. 2004;25(8):1020-1023. Retrieved from: https://pubmed.ncbi.nlm.nih.gov/15322591/

[8] Kalyvas V, Gkekas C, Papadopoulos DP, Malioris A, Milias S, Papathanasiou M, et al. Intratesticular angiolipoma: a rare case of adipose tissue presence in the testis. Case Reports in Urology 2019. https://doi.org/10.1155/2019/7606530

[9] Waked H, Zaarour M, Aftimos G. Angiomyolipoma of the testis: A case report. Open J Clin Med Case Rep. 2020; 1660. Retrieved from: https://www.researchgate.net/publication/344043132_Angiomyolipoma_of_the_testis_A_case_report