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Angiomyolipoma of the testis: A case report

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Abstract

Angiomyolipoma AML is a benign mesenchymal tumor composed of abnormal blood vessels, cells with smooth muscle features, and mature adipose tissue in variable proportions. Within this content we present a case of testicular AML in a 75 years old male who presented to his physician complaining of a painless swelling and hardness over his right testis that increased over time, without any history of trauma. Testicular tumor markers were normal, and diagnosis was made based on the histopathological studies post orchidectomy.

Keywords

Angiomyolipoma; testis.

Introduction

Angiomyolipoma, a benign tumor, most commonly occurs in the kidney and the liver [1]. It is the most common benign tumor of the kidney [2]. Most of these tumors occur as a sporadic case, and little of them occur in association with tuberous sclerosis [3]. Extra- renal sites of AML are rare and include ovary, uterus, fallopian tube, skin, spermatic cord, epididymis, hard, soft palate, mediastinum, colon and nasopharynx and other very rare sites [4]. Testicular angiomyolipoma remains very rare and not very well understood and only few cases were reported in the literature.

Case report

A 75-year-old male, with a history of diabetes and hypertension, not previously known for having a urologic disease, presented to his physician with the chief complaint of a painless swelling and hardness over his right testis that appeared few years before the presentation and increased slowly in size over the time to reach its actual size. The patient had no history of blunt trauma. Past medical and surgical history were unremarkable. On physical examination, the patient had a solid mass over his right testis,

with a moderate hydrocele. Left testis was normal. Ultrasound showed a heterogeneous mass within the right testicular capsule. Tumor markers including alpha-fetoprotein (AFP) and beta human chorionic gonadotrophin (β-HCG) were within normal limits. After orchidectomy was done, the gross description revealed an intraparenchymal tumor measuring 3.5 x 3 x 2.6 cm in size. The tumor was well circumscribed, hemorrhagic and focally cystic.

Microscopic examination showed a well circumscribed tumor proliferation made of a mixture of adipose tissue, vessels arranged in lobules and smooth muscle fibers. The tumor was focally hyalinized and cystic.

Immunohistochemistry revealed a positivity of CD34 in the blood vessels component, Desmin and SMA showed a positivity in the smooth muscle component. However, HMB-45 was negative.

No evidence of malignancy was seen.

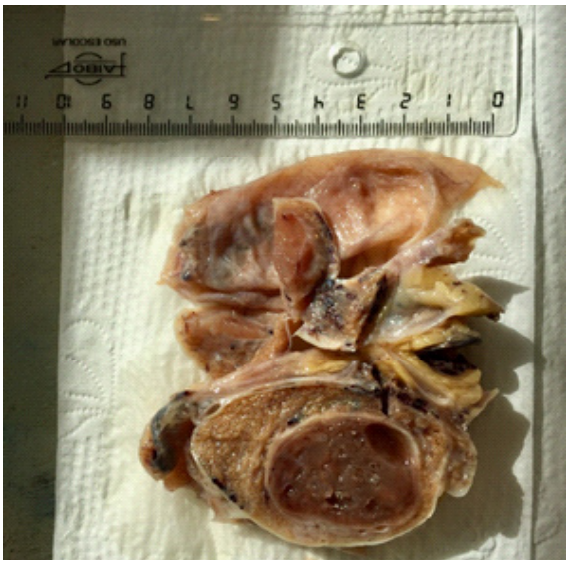

Figure 1: Macroscopic appearance showing a well circumscribed, hemorrhagic and focally cystic tumor

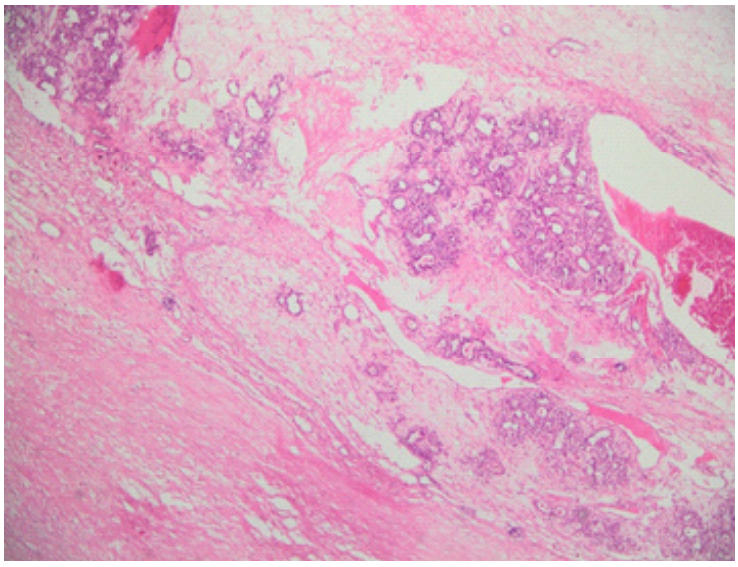

Figure 2: H&E showing the three components of the tumor. (40x magnification)

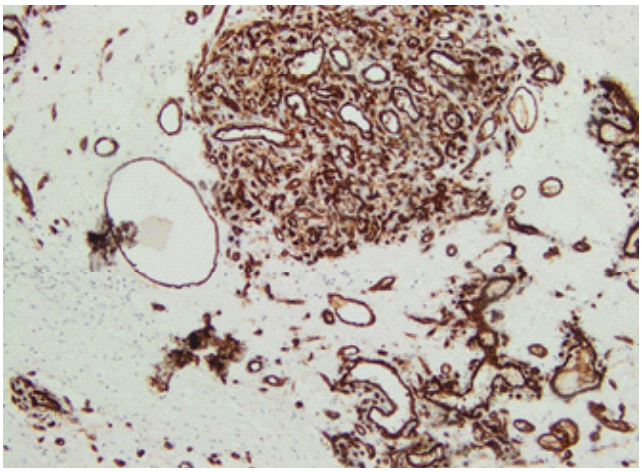

Figure 3: CD34 positive in the blood vessels. (100x magnification)

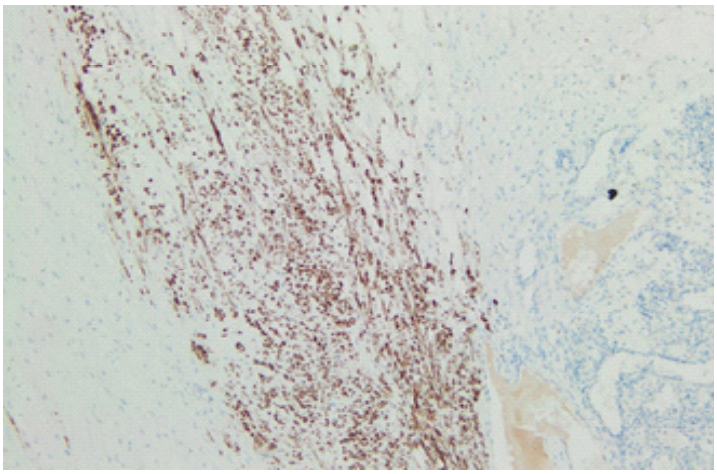

Figure 4: Desmin positive in smooth muscle component. (100x magnification)

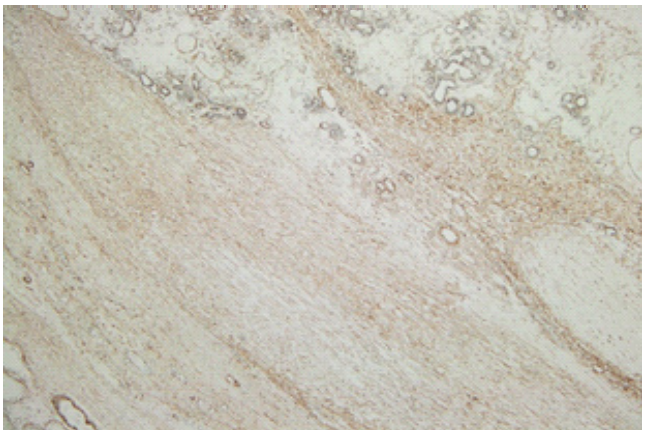

Figure 5: SMA positive in smooth muscle component. (40x magnification)

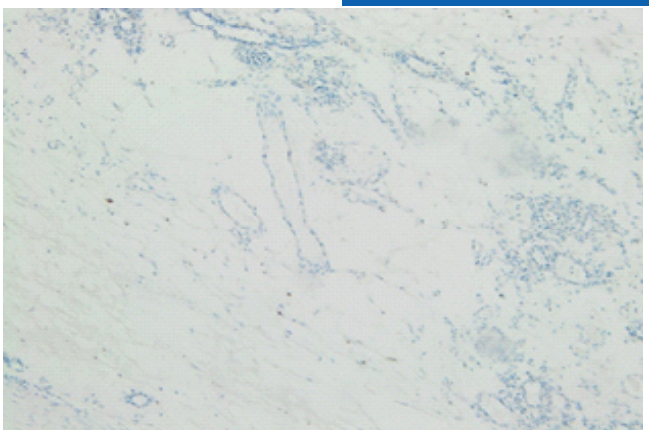

Figure 6: HMB-45 was negative. (100x magnification)

Discussion

Angiomyolipoma is a benign tumor that primarily involves the kidneys and the liver being the second most common site. AML is the most common benign tumor of the kidney. This tumor is described to occur in many other extra-renal locations including the skin, ovary, soft palate, colon, nasopharynx and many other sites. AML is also described as a sporadic case in most of the cases and associated with tuberous sclerosis in fewer cases. This tumor consists of blood vessels, adipose tissue and smooth muscle [1-4]. Angiomyolipoma was previously considered as a form of hamartoma, but is now known to be a tumor that arises from the perivascular epithelioid cells (PECs) and is classified as part of the tumors termed "PEComas" which can affect many organs including the kidney, lung and testis. PECs express myogenic and melanocytic markers [5]. Our case presented with a testicular painless mass that increased in size over the time and tumor markers were within normal limits, which pushed to histopathological investigations after surgery was done and revealed the true nature of the tumor.

Computed tomography (CT) and ultrasound imaging are useful in making the diagnosis of AML in nearly all cases before doing any surgical procedure. Since most AMLs highly contain fat, it makes a particular pattern on CT scan [6]. However, in our case only an ultrasound was performed, and it showed a heterogeneous mass in the right testicle. The histopathologic studies performed provided the final diagnosis.

Considering immunohistochemical staining, smooth muscle actin (SMA) and endothelial marker (CD34) are usually expressed in angiomyolipoma [7]. In our case, SMA and CD34 were used and showed positivity for smooth muscle component and blood vessels in the tumor respectively. Studies showed that HMB-45, which is a melanoma-associated antigen, is only positive in renal and liver AML. Oppositely, HMB-45 showed negativity in skin and testicular AML. Consequently, testicular AML and typical AML in the kidney might be different biologically [8]. In our case, HMB-45 was performed and was in fact, negative.

Most of AMLs are benign tumors, but some cases of malignant AMLs have been reported for renal and extrarenal AML. Tumors with dominant pleomorphic epithelioid components, which demonstrate intense HMB-45 expression, as well as the existence of local invasion and high mitotic activity may predict a poor outcome [8].

Treatment is usually by surgical excision. Some studies found that when conservative surgery is not possible due to a large size tumor, radical orchiectomy should be done. However, when the tumor is small and benign, organ sparing surgery could be attempted [9].

To our knowledge, published studies concerning primary testicular angiomyolipoma are few in numbers, so little is known about this tumor and further studies should be done to determine the nature of this tumor and to upgrade the management and treatment modalities. What differentiate our study from the few previously published studies on testicular AML, is that the patient is 75 years old and is considered old-aged adult while in the previous studies all patients were middle-aged adult or young-adults. This finding could be very important to show what are the age groups affected by testicular AMLs, with patients from every age group already reported in the literature, meaning that testicular AML could affect the young and the elderly.

Conclusion

In conclusion, angiomyolipoma of the testis is a very rare benign tumor that is diagnosed using immunohistochemical studies and treated accordingly to each case.

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