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Case Report

Rare Case of Metastatic Primary Testicular Angiosarcoma

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Abstract

Testicular angiosarcoma is a very rare malignancy. We present an additional case with multiple distant metastasis. No specific local clinical findings. This proves their aggressive biological behavior. A 50-year-old patient with a single nodule palpable in the left testicle, no redness of the skin, pain or fever. With uneven surface, hard consistency and is slightly painful on palpation. Right testicle physical examination reveals smaller nodule. Ultrasound examination revealed a heterogeneous nodular structure of both testicles. Laboratory investigation - normal blood count, biochemistry and normal values of tumor markers. The patient was informed that both testicles are with pathological findings and bilateral orchiectomy should be performed, but the idea wasn't accepted. A left radical inguinal orchiectomy was performed. Pathohistological and immunohistochemical examination of the specimen revealed primary testicular angiosarcoma. Primary testicular angiosarcoma is an extremely rare histologic subtype of testicular tumors. The presence of multiple general metastasis - cerebral, lungs, suprarenal glands, left kidney and right testis proves their aggressive biological behavior. There were no clinical findings of previous testicular germ cell tumor. The pathohistological identification and immunohistochemical verification are decisive for the diagnosis and the treatment.

Keywords: Angiosarcoma, testicular tumors

INTRODUCTION

Testicular tumors represent 1% to 1,5% of all malignant tumors in males and 5% of the urological tumors (D'aniello et al., 2014).

Primary angiosarcoma, known as hemangiosarcoma is extremely rare subtype testicular tumor, with an aggressive biological behavior, poor therapeutic outcome and prognosis. The survival rate ranging from 6 to 16 months (Young et al., 2010). It develops from the endothelium of blood or lymph vessels. It can be primary testicular angiosarcoma or secondary asa transformation of pre-existing testicular germ cell tumor into angiosarcoma. Only 6 cases are described in the medical literature. The first case was published by Hughes et al. (1991). In 2007 5 more clinical cases have been published (Armah et al., 2007).

Publishing this articleis further enrichment of the scarce medical literature with one more case of rare histological subtype of testicular tumors.

Case presentation

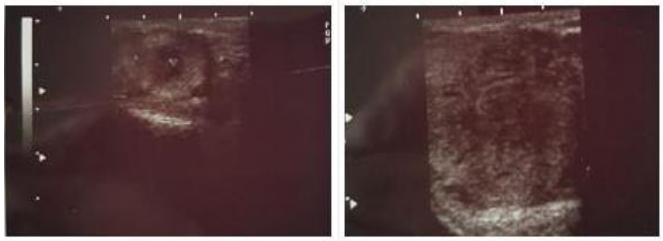
The case focuses on a 50-year-old patient with a palpable tumor in the left testis, no redness of the skin, pain or fever. Treated on an outpatient basis for "epididymitis", with antibiotics and anti-inflammatory

drugs, without improvement in the local finding.

Physical examination reveals a single nodule in the left testicle about 2 cm in diameter. It was with uneven surface, hard consistency and is slightly painful on palpation. The right testicle is with normal size, but a smaller nodule was palpated as well with dimensions of 0.5 cm in diameter.

Laboratory investigation - normal blood count, biochemistry and normal values of tumor markers.

The ultrasound examination revealed a heterogeneous nodular structure of both testicles. Doppler ultrasound wasn't performed by the imaging department because of technical reasons.



A left radical inguinal orchiectomy was performed.

Pathohistological and immunohistochemical examination of the specimen revealed primary testicular angiosarcoma. (Figure 1-4)

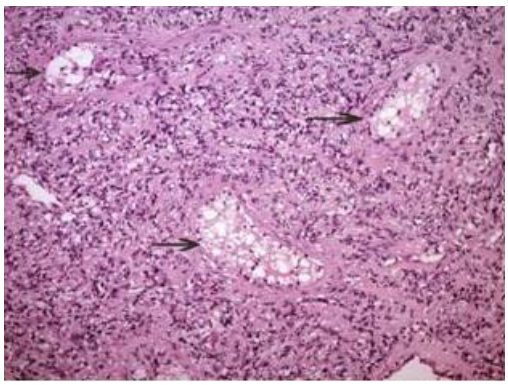


Figure 1. Hematoxylin and eosin staining. Magnification x100. Testicular tissue fully engaged by small and medium-sized vessels. Atrophic glands are marked with arrows.

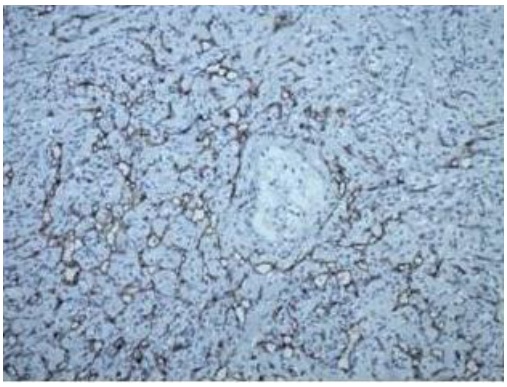


Figure 2. Immunohistochemistry. CD 34 Magnification x100. Positive staining for CD 34 marking endothelial cells. In the middle, an atrophic canal of the testicle.

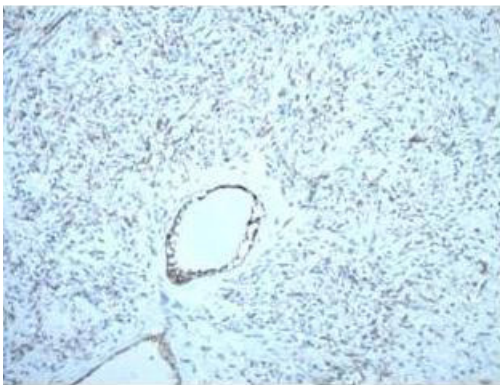


Figure 3. Immunohistochemistry. Vimentin Magnification x100.Positive staining for vimentin, marking the mesenchymal structures - vessels and interstitial tissue.

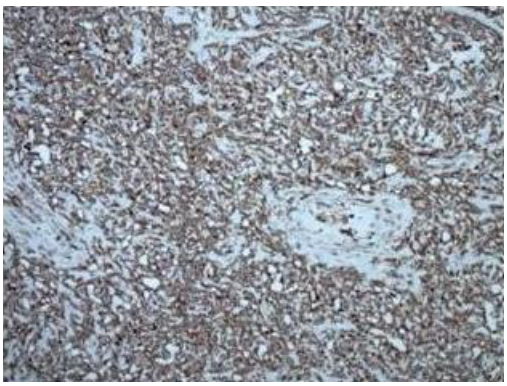


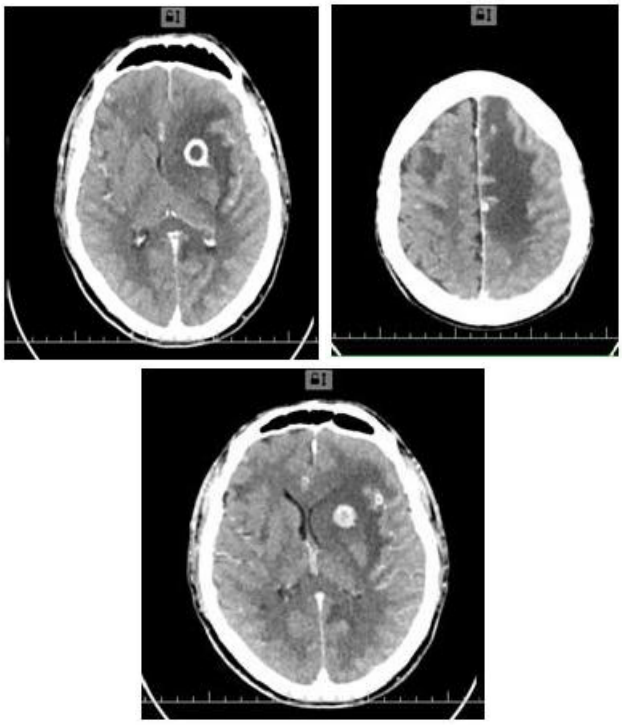
Figure 4. Immunohistochemistry. Cytokeratin AE1 / AE3 Enlarged x100. The epithelial cells of the atrophic testicular glands and some of the tumor cells are positively marked

After the radical inguinal orchiectomya psycho-emotional disorders were noticed with manifested changes in the patient's behavior such as depression, lack of communication and motivation, anxiety, refusal to eat and

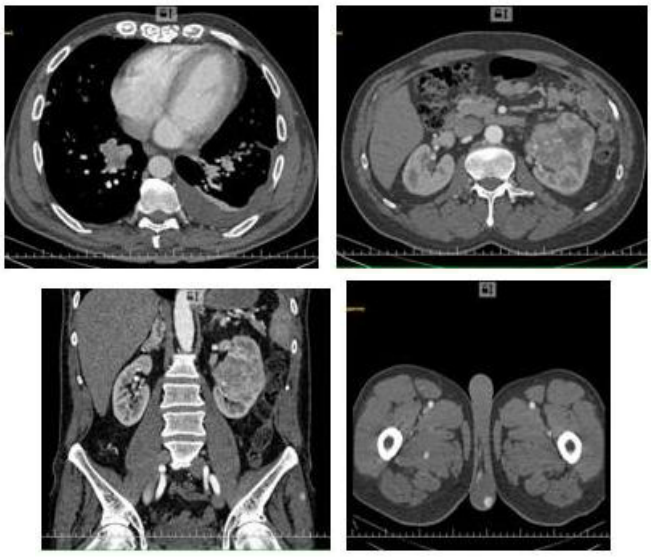
adynamia, a CT scan investigation was performed. It revealed:

CT scan of the brain

Presence of annular hyperdense metastatic lesions in the area of the basal ganglia in the left cerebral hemisphere with a large area of perifocal edema and dislocation of the midline.



CT scan of the abdomen and the pelvis proved the existence of metastatic lesions in the lungs, suprarenal glands, left kidney and the right testis.



DISCUSSION

Patients under 30 years of age with testicular angiosarcoma were found to have evidence of germ cell carcinomas. Patients over the age of 60haveno data of preexisting GCT, but develop primary testicular angiosarcoma.

The most important point here is the size of the testicular tumors at the time of diagnosis. Testicular tumors are known to be painless masses, which is the main factor responsible for patients' late presentation to a physician. Initiatives are required to increase public awareness of the importance of early diagnosis in testicular cancer, especially men between the ages of 15 and 45 years (Sarier et al., 2020).

Primary testicular angiosarcoma is often presented with a local finding and a variety of clinical manifestations and symptoms depending on the localization of the metastasis and the stage of the disease. Patho-histological examination and immunohistochemical verification is crucial for the diagnostic differentiation of primary testicular angiosarcoma from hemangiomas, anaplastic melanoma and epithelial carcinoma (Walker et al., 2011).

Surgical treatment: Inguinal radical orchiectomy is the only therapeutic option.

Targeted radiotherapy is applicable for single meta-static lesions, and in case of disseminated metastases - palliative chemotherapy. Testicular angiosarcomas have a grim prognosis and poor therapeutic outcome.

CONCLUSION

Primary testicular angiosarcoma is an extremely rare histologic subtype of testicular tumors. The presence of multiple generalized metastasis - cerebral, lungs, supra-renal glands, left kidney and right testis proves their aggressive biological behavior. There were no clinical findings of previous testicular germ cell tumor. The pathohistological identification and immunohistochemical verification are decisive for the diagnosis and the treatment. Testicular angiosarcomas have a poor prognosis and therapeutic outcome.

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