Adenomatoid tumor of the testis: A rare case report

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ABSTRACT

Adenomatoid tumors (AT) are benign tumors commonly found in paratesticular tissues. However, intratesticular AT are rare. Clinically and radiologically, the AT of the testis imitates the malignant neoplasia of the testis. Here, we present a case of the intratesticular AT.

1. Introduction

Adenomatoid tumors (AT) are commonly found in paratesticular tissues accounting for approximately 32% of the paratesticular masses. Other locations of AT in men are spermatic cord, tunica albuginea, ejaculatory ducts, and rarely intratesticular. The typical locations in women are fallopian tubes and uterus. In addition, the extragenital AT has been reported in the omentum, umbilical skin, mesentery of the small intestine, pancreas, mediastinal lymph nodes, pleura, and heart. These tumors are mesothelial in origin.

The AT typically presents between the third to fifth decade of life. The ethnic races most predisposed to this tumor are Caucasian followed by, Black (14%) and Oriental (0.5%).

The AT involving testicular parenchyma is rare, and approximately 14 cases of intratesticular AT are reported to date.

2. Case presentation

A male in his late 40s presented with left testicular painless swelling, which was present for more than a year. There is no history of any trauma or sudden enlargement of the swelling. There is no personal or family history of testicular malignancy. He has a palpably normal right testis, and the left testis has a firm, non-tender palpable mass near the upper pole. To evaluate the testicular mass suspicious for testicular malignancy, he underwent an ultrasound of the scrotum and testicular tumor markers: Alpha-fetoprotein, beta-human chorionic gonadotropin, and lactate dehydrogenase. The testicular tumor markers were within the normal range. Ultrasound of the scrotum was suggestive of normal findings on the right testicle, and the left testicle showed a solid lesion peripherally and posteriorly (Fig. 1). The Staging work-up with a CT scan of the abdomen and pelvis did not show any metastasis. Given the suspicion for testicular malignancy, the patient underwent preoperative counseling followed by left high inguinal radical orchiectomy.

Histologic examination reveals a relatively well-circumscribed, non-encapsulated 2.0 cm lesion located at the periphery of the testicular parenchyma (Fig. 2-A). It consists of cords and nests of bland-looking epithelioid cells with round to oval nuclei surrounded by fibrous stroma. The cells have abundant eosinophilic cytoplasm with frequent vacuoles. No atypia or mitotic figures are seen (Fig. 2-B). The surgical margins of resection are free of tumor. There is complete spermatogenesis in the seminiferous tubules of the background testicular parenchyma with no evidence of germ cell neoplasia in situ.

Immunostains were performed to show that the tumor cells are diffusely positive for calretinin (Fig. 2-C), D2-40 (Fig. 2-D), WT-1, CKAEL, and Cam5.2. They are negative for inhibin, SALL4, OCT3/4, MOC31, BerEP4, CD34, and CD68. The background testicular parenchyma is also negative for OCT3/4 confirming the absence of germ cell neoplasia in situ. Additional immunohistochemical stains performed showed the tumor is negative for CD31 and CK5/6 with a low Ki67 proliferation index (<5%). These findings confirm the diagnosis of AT.

3. Discussion

Golden and Ash coined the term Adenomatoid tumor to describe...
benign tumors with glandular pattern localized in the genitourinary system.

Clinically, AT of testis present as testicular lump found incidentally. Occasionally there is an associated hydrocele or periorchitis. The AT of the testis does not produce any characteristic radiological appearance to distinguish it from malignant testicular neoplasia. It can present a hypoechoic, isoechoic, or hyperechoic appearance on the testicular ultrasonography. The testicular tumor markers are within a normal range. So, to summarize, testicular AT has clinical and radiological characteristics similar to malignant testicular neoplasia.

Pathologically, on gross examination, AT are usually small, solid, firm, grayish-white to tan, and poorly to well-circumscribed masses. On
microscopy, AT typically presents cords, tubules, and small clusters of cuboidal, dense eosinophilic cells with frequent cytoplasmic vacuolization. The architectural arrangement and vacuolated cytoplasm are clues to the diagnosis. In most cases, the mitoses are not present. Stroma is generally fibrous and occasionally hyalinized or contains smooth muscle. The fibrous stroma often contains scattered lymphocytes or lymphoid aggregates, typically towards the periphery of the tumor. Generally, the AT is unencapsulated and occasionally invades surrounding parenchyma, which is unusual considering the benign nature of the tumor.\textsuperscript{3,4}

The histologic differential diagnosis of AT of the testis is broad, but high in the list are sex cord stromal tumors (particularly Sertoli cell tumors), malignant mesothelioma (MM), metastatic tumors, vascular lesions, and germ cell tumors (particularly yolk sac tumors), among others. All these neoplasms can have similar architectural patterns that include cords, tubules, gland-like architecture, and epithelioid cells. The use of customized immunohistochemical panels complements thorough clinical, gross, and morphologic assessment. In contrast to AT, sex cord stromal tumors are positive for inhibin and Melan A; yolk sac tumors express SALL4, Glypican-3, alpha-fetoprotein, and vascular neoplasm are positive for CD31, CD34, FLI-1. Metastatic lesions can be more challenging, and their immunohistochemical profile varies depending on the primary origin. MM is an important diagnosis to exclude. The distinction relies heavily on gross and morphologic evaluation, as their immunoprofile overlaps with AT. MM more often are larger lesions with a destructive growth pattern and greater cytologic atypia, including the presence of mitoses.

The AT is an important differential diagnosis to consider in patients with testicular lesions. In carefully selected patients, intraoperative frozen section guided partial orchiectomy can be considered.

However, with clinico-radiological parameters similar to the malignant testicular lesions, a majority of the patients with AT of testis have undergone radical high inguinal orchiectomy. The AT is not known to recur, and high inguinal radical orchiectomy is diagnostic as well as therapeutic. Also, the literature does not recommend follow-up serial imaging or monitoring the testicular tumor markers after radical orchiectomy.\textsuperscript{5}

4. Conclusion

AT involving testicular parenchyma is a rare benign tumor of the testis. It is challenging to clinically distinguish the AT of testis from malignant neoplasia of testis due to identical clinical and radiological parameters. Hence, most patients have been diagnosed post radical orchiectomy on pathological evaluation.

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Declaration of competing interest

None.

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