Case Report

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Rare Cause of Testicular Mass: Adenomatoid Tumor of the Testis

Şeref Coşer[1], Halil İbrahim İvelik[1], Gizem Akkaş Akgün[2], Mustafa Mehdi Er[1], İbrahim Güven Kartal[1]

[1]*Kütahya University of Health Sciences, Faculty of Medicine, Department of Urology, Kütahya, Turkey*
[2]*Kütahya University of Health Sciences, Faculty of Medicine, Department of Pathology, Kütahya, Turkey*

Abstract

Adenomatoid tumors are rare benign neoplasms. In this case report, a 38-year-old patient was diagnosed with an intratesticular adenomatoid tumor following orchiectomy because of a suspicious mass in the testis. Adenomatoid tumors, which are most commonly observed in paratesticular tissues, can also be seen as testicular masses that cannot be distinguished from malignant solid testicular masses with clinical findings and imaging methods, causing many unnecessary orchiectomies. When evaluated together with previous cases, adenomatoid tumors do not show clinically aggressive behavior.
Keywords: Adenomatoid tumor, testicular mass, benign tumor

Introduction

Adenomatoid tumors are rare benign tumors of the male and female genital system that are commonly located in paratesticular tissues in men. Adenomatoid tumors, which constitute approximately 32% of paratesticular masses, rarely present with intratesticular localization (1). To date, 15 cases of intratesticular adenomatoid tumor have been described. Typically, they appear between the third and fifth decades of life. These benign tumors are most commonly observed in Caucasians, followed by African Americans (2,3). In this case report, the management of a patient diagnosed with testicular adenomatoid tumor, which could not be distinguished from a malignant tumor preoperatively, is presented.

Case Report

A 38-year-old male patient presented with a complaint of right scrotal pain that had been ongoing for 2 weeks. No history of trauma. Physical examination revealed palpable firmness in the lower pole of the right testicle. Scrotal Doppler ultrasonography revealed a well-defined isoechoic solid lesion with a hypoechoic halo measuring 9x8 mm in size, located in the lower pole of the right testicle. Testicular tumor markers (alphafetoprotein, beta human corionicgonadotropin, lactate dehydrogenase) were within normal limits. Magnetic resonance imaging (MRI) of the scrotum performed at an external center showed a well-defined lesion measuring 14x13 mm with a central cystic - necrotic appearance and a periphery showing intense contrast enhancement, which extended caudally to the testis in the lower pole of the right testicle (Figure 1). Thoracoabdominopelvic computed tomography performed for staging did not reveal any evidence of metastasis. After the patient was informed about the testicular tumors, he underwent right radical inguinal orchiectomy. Histopathological examination revealed a relatively well-defined 1.5 cm lesion with a central hemorrhagic area and a cream-white periphery located 0.5 cm away from the capsule in the lower pole of the testicle. In the serial section examination of the lesion, irregularly defined cell infiltration was observed between the seminiferous tubules and rete testis (Figure 2A). Large cytoplasmic, spindle-polymorphic nuclei with distinct nucleoli were observed in the stroma, and the cells had slightly atypical features. Some cells had a wide eosinophilic cytoplasm, whereas others had a wide vacuolar cytoplasm (Figure 2B). There was no mitosis, lymphovascular invasion, or perineural invasion. No tumor was observed at the surgical margins. Immunohistochemical examination showed positive staining for calretinin (Figure 2C), vimentin, PanCK (Figure 2D), BRAP1, and S100, whereas it was negative for inhibin, CD34, and HBME-1.

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Address for Correspondence: Halil İbrahim İvelik, Kütahya University of Health Sciences, Faculty of Medicine, Department of Urology, Kütahya, Turkey
Phone: +90 542 361 87 13 **E-mail:** halib_ive@hotmail.com **ORCID-ID:** orcid.org/0000-0001-5298-0045
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After the pathological diagnosis, the patient was recommended for follow-up. No problems were detected in the patient's outpatient clinic controls at 3 and 6 months. Patient consent was obtained for the case reports to be published for academic purposes.

Discussion

Adenomatoid tumors were first described as a group of benign tumors with a glandular pattern localized in the urogenital system in 1945 (4). Adenomatoid tumors are quite rare in the testis and are most commonly presented in the epididymis (77%). In men, other urogenital localizations where they are observed include the spermatic cord, tunica albuginea, and ejaculatory ducts, whereas in women, typical sites of occurrence are the uterus and fallopian tubes. In addition to these sites, they can also appear in extragenital regions such as the pleura, heart, omentum, mesentery, and mediastinal lymph nodes (3,4). They usually appear as painless hard nodules measuring less than 2 cm, detected incidentally on physical examination. However, sometimes mild pain or accompanying conditions such as hydrocele or periorchitis may also be present. Tumor markers for testicular tumors were negative in all cases. The radiographic appearance of adenomatoid tumors is non-specific, and they appear as hypoechoic, isoechoic, and hyperechoic lesions on testicular ultrasonography. In scrotal MRI, no distinguishing appearance from malignant neoplasms of the testis, as seen in ultrasonography, is observed. In conclusion, adenomatoid tumors of the testis have clinical and radiological characteristics similar to those of malignant testicular neoplasms. Although the origin of adenomatoid tumors is controversial, studies using electron microscopy and immunohistochemical staining suggest that they are of mesothelial origin. Macroscopic evaluation of the specimen shows adenomatoid tumors as small, solid, hard, gray-white, well-defined nodules. On microscopic evaluation, the tumor consists of cuboidal, vacuolated, eosinophilic cells that form dilated tubules, cords, and cell clusters within the fibrous stroma. The vacuolization observed in the cytoplasm is specific to adenomatoid tumors. Mitoses are not observed. In the differential diagnosis of adenomatoid tumors of the testis, metastatic tumors, sex cord-stromal tumors, malignant mesothelioma, and vascular lesions should be considered (5). Although clinical and morphological features are decisive in making a differential diagnosis, specialized immunohistochemical markers are also used. In adenomatoid tumors, positivity for calretinin, vimentin, cytokeratin, WT1, and EMA is observed (5). Unlike sex cord stromal tumors, inhibin negativity is observed in adenomatoid tumors. Negativity for CD31, CD34, and FLI-1 can also be used to distinguish them from vascular neoplasms. Unlike malignant mesotheliomas, adenomatoid tumors do not exhibit mitoses or necrosis, and they have a more destructive growth pattern. Because of the similarity of their appearance to malignant testicular tumors based on clinical and radiological imaging, radical inguinal orchiectomy is widely performed for treating adenomatoid tumors. However, testis-sparing surgery may also be considered among the treatment options in selected cases suspected of having a benign tumor, with intraoperative frozen biopsy taking priority (6,7). There is currently limited data on the recurrence and malignant degeneration of adenomatoid tumors in the literature (8,9). These views are not recommended to be followed by serial imaging methods and keys of tumor markers (10). In this case, similar to our cells, the main problem is that the clinical and radiological features of adenomatoid tumors can hardly be distinguished from malignant neoplasms. For this reason, the possibility of a good tumor should always be kept in mind in testicular masses, thus preventing the appearance of unnecessary orchiectomies.

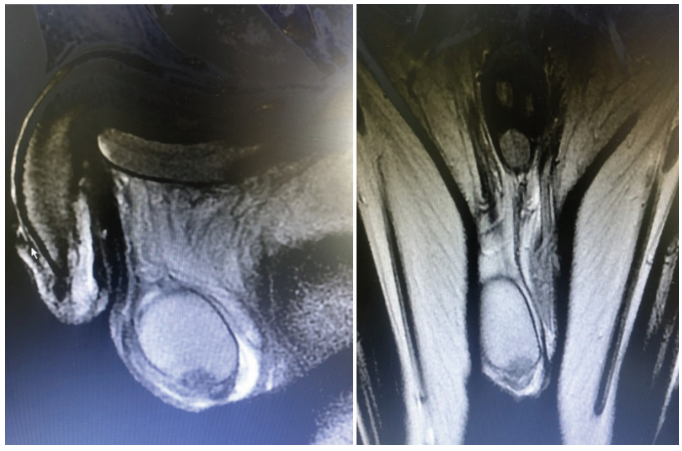

Figure 1. Scrotal magnetic resonance imaging of the tumor: Well-defined lesion located in the lower pole of the right testis

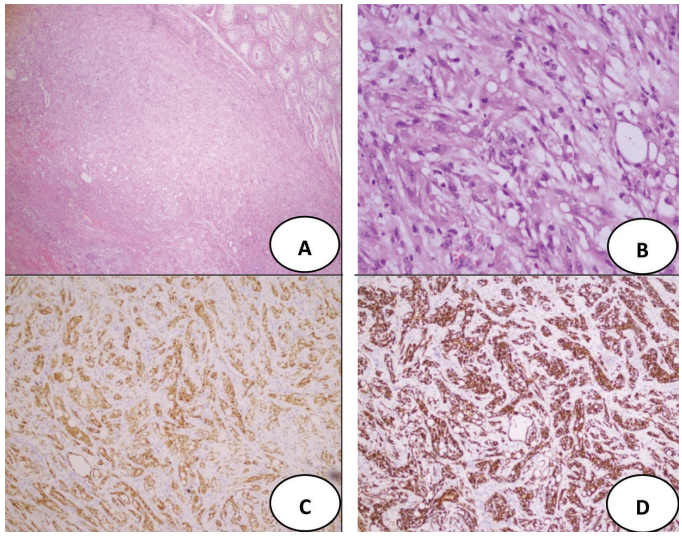

Figure 2. A) Nodular tumoral lesion with a smooth boundary separated from the surrounding testicular tissue (HEx40). **B)** Cells with eosinophilic, spiky cytoplasm, vesicular structure, thin chromatin condensation, punctate nucleoli, and minimal atypia (HEx100). **C)** Calretinin (x100). **D)** Pankeratin (x100)

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Ethics

Informed Consent: Patient consent was obtained for the case reports to be published for academic purposes.

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Authorship Contributions

Surgical and Medical Practices: Ş.C., İ.G.K., Concept: Ş.C., M.M.E., Design: H.İ.İ., İ.G.K., Data Collection or Processing: H.İ.İ., M.M.E., Analysis or Interpretation: İ.G.K., G.A.A.,, Literature Search: Ş.C., İ.G.K., Writing: Ş.C., H.İ.İ.

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