Case Report

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Rare Case; Primary Epididymal Adenocarcinoma

Anıl Eker[1], Murathan Muratoğlu[1], Ertuğrul Şefik[2], Funda Taşlı[3]

[1]*University of Health Sciences Turkey, İzmir Bozyaka Training and Research Hospital, Clinic of Urology, İzmir, Turkey*
[2]*İzmir Tınaztepe University Faculty of Medicine, Department of Urology, İzmir, Turkey*
[3]*University of Health Sciences Turkey, İzmir Bozyaka Training and Research Hospital, Clinic of Pathology, İzmir, Turkey*

Abstract

Paratesticular masses constitute 2-3% of the scrotal masses. Epididymal masses constitute only 5% of prescrotal masses, and the most common type of tumor is adenotoid tumor, which is benign. Malignancies of the epididymis are rare. Epididymal adenocarcinoma is much less common. It can be primer and metastatic, and there are fewer than 40 cases in the literature in both groups. Primary epididymal adenocarcinoma is extremely rare, with only 23 cases reported. The disease diagnostic process, findings, and treatment are still unknown. In this article, we aimed to present a case of epidimal adenocarcinoma with primary origin from the epididymis.

Keywords: Epididymal adenocarcinoma, paratesticular masses, epididymal cancer

Introduction

As the paratesticular region contains various structures, including the epididymis, spermatic cord, tunica vaginalis, and strong fat-ligament-muscle supporting tissues, it may give rise to a number of tumor types with various behaviors. Paratesticular masses constitute 2-3% of the scrotal masses (1). 30% of paratesticular neoplasms are malignant. Sarcomas are the most common type of tumor and generally originate from the spermatic cord (2). Epididymal masses constitute only 5% of prescrotal masses, and the most common type of tumor is adenotoid tumor, which is benign. Malignancies of the epididymis are rare. Epididymal adenocarcinoma is much less common. It can be primer and metastatic, and there are fewer than 40 cases in the literature in both groups. Primary epididymal adenocarcinoma is extremely rare, with only 23 cases reported (2,3). In this article, we presented a case of epidimal adenocarcinoma with primary origin from the epididymis.

Case Report

Consent has been obtained from the patient that the disease and treatments related to the disease will be shared as scientific publications. A 63 year-old farmer presented with a 1 year history of right inguinal area and scrotal pain. It was found that he had no comorbidities, and he underwent surgery for right inguinal hernia. The physical examination revealed a right scrotal mass that made it impossible to differentiate between the epididymis and testicle. Scrotal Doppler ultrasonography showed a hypervascular right scrotal mass that filled the entire testicle and right hydrocele. Also, a 13 mm inguinal lymph node was observed. The patient's beta-HCG, alpha-fetoprotein (AFP), and lactate dehydrogenase levels were found to be 0.33 mL/U, 5.09 ng/mL, 189 U/L respectively at normal intervals. There was no evidence to suggest infection at laboratory values and physical examination. Thoracic computed tomography (CT) and abdominal CT imaging with testicular tumor preliminary diagnosis did not reveal any lesion that could be considered as metastasis. Right radical orchiectomy was performed in the patient with a preliminary diagnosis of testicular cancer. Pathology specimens evaluated from 2 different centers. The pathology result was spotted epidiymal adenocarcinoma. Immunohistochemically, EMA + CKPAN +, CK7 +, Pax8 +, CD10 +, WT1 +, ER+, calretinin +, p53 +, Ki67+ and CEA - prostate specific antigen (PSA) -, PLAP -, AFP -, bHCG -, inhibin -, CD30 - was spotted (Figure 1). The tumor consisted of solid and papillary areas. It was found to be low grade in the papillary and adenoid areas and high grade in the solid areas (Figure 2). The tumor was limited to the testis and epididymis. The spermatic cord was reported to be not invaded. The lymph node removed from the inguinal region was detected as reactive. The patient's preoperative thorax and abdominal CT images showed no

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Address for Correspondence: Anıl Eker, University of Health Sciences Turkey, Izmir Bozyaka Training and Research Hospital, Clinic of Urology, Izmir, Turkey
Phone: +90 532 051 6110 **E-mail:** anileker993@gmail.com **ORCID-ID:** orcid.org/0000-0003-1603-3336
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evidence of any other adenocancer focus or metastasis that could be the primary source. It was found to be PSA - tapped and excluded the possibility of epidimal metastases of prostate adenocarcinoma. The patient was then referred to clinical oncology. In the 6th postoperative month, positron emission tomography-CT images showed metastases in the retroperitoneal lymph nodes and lung. Six cycles of carboplatin and paclitaxel chemotherapy were administered to the patient. With this treatment, disease remission was achieved.

Discussion

Epididymal adenocarcinoma is a rare condition. It can be primer and metastatic, and there are fewer than 40 cases in the literature in both groups. Primary epididymal adenocarcinoma is extremely rare, with only 23 cases reported (2,3,4). In these cases, patient ages ranged from 27 to 81 years (4). In a review 12 of the 21 reported cases of epididymal adenocarcinoma, the patient's age was greater than 50 (3). In this case, our patient was 63 years old.

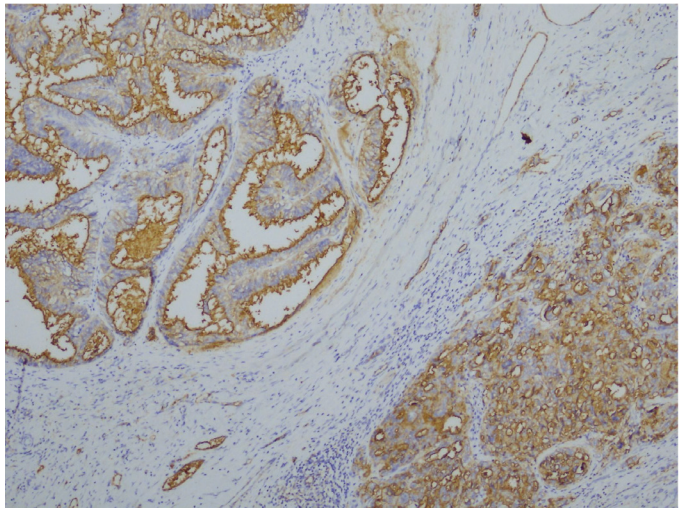

Figure 1. Immunohistochemically EMA + stained epidymal adenocarcinoma cells

EMA: Epithelial membrane antigen

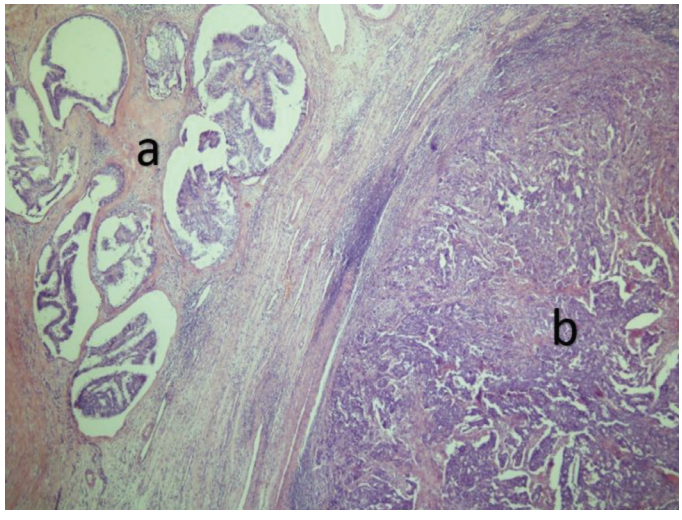

Figure 2. Epididymal adenocarcinoma cells consisting of papillary (a) and solid (b) areas

According to a review, scrotal swelling and palpable mass are the most common findings. Almost one-third of patients complain of scrotal pain (4). In some cases, the disease is accompanied by hydrocele (5). In the latest review 38.5% of patients have hydrocele (4).

Histologically, the tumor can contain papillary, tubular and solid areas. Immunohistochemically, epithelial tumor markers, such as cytokeratin and epithelial membrane antigen (EMA), are positive (3,4,5). Epididymal adenocarcinoma can be metastatic from other organ adenocarcinomas such as prostate, gastrointestinal system, and renal cell carcinoma. To differentiate the lesions from other organ adenocarcinomas, immunohistochemical markers must be used. To exclude prostate adenocarcinoma PSA staining and exclude renal cell cystadenocarcinoma metastasis CD10, CK7 staining must be performed. If epididymal adenocarcinoma PSA staining is found to be negative and CK7 and CD10 must be found positive. Also, PLAP, AFP, and bHCG must be found negative (4,6).

In this case, histologically, the tumor consisted of solid and papillary areas. It was found to be low grade in the papillary and adenoid areas and high grade in the solid areas. Immunohistochemically, EMA (+), CD10 (+), CK7 (+), PSA (-), PLAP (-), AFP (-) and bHCG were also found to be negative. In the literature, most of the cases were calretinin negative (4). However, in this case, calretinin was found positive.

Epididymal lymph node drainage occurs in the pelvic and retroperitoneal lymph nodes. Therefore, the inguinal lymph node dissection is unnecessary. In this case, inguinal lymph node dissection pathology was found benign (4).

Adjuvant treatment is uncertain because of the lack of literature. The prognostic factors of the disease are also uncertain. However, metastasis is the most common cause of death after surgery. It has been reported in bone, liver, spleen, lung, pelvic, and retroperitoneal lymph node metastases. The effects of radiotherapy and chemotherapy are unclear. Platinum-based chemotherapy was the first choice treatment of reported cases with advanced disease, and a positive effect on disease progression was observed (4). In this case, after lung and bone metastasis, 6 cycles of carboplatin and paclitaxel chemotherapy were administered to the patient. With this treatment, disease remission was achieved. The patient is followed up every 3 months. Oncological outcomes are not yet predictable.

Conclusion

Epididymal adenocarcinoma is a very rare malignancy. There are not enough data of diagnosis, differential diagnosis, pathological findings, treatment, and prognosis of the disease. the disease progresses aggressively and becomes metastatic in the early period, but there are not enough data in the literature related to its treatment. The effects of chemotherapy and radiotherapy are uncertain. In some cases, chemotherapy and radiotherapy are not responding, but in this case, the patient was fully cured with chemotherapy. As the number of reported cases increases, our knowledge of the disease will increase.

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Ethics

Informed Consent: Consent has been obtained from the patient that the disease and treatments related to the disease will be shared as scientific publications.

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Authorship Contributions

Surgical and Medical Practices: A.E., E.Ş., Concept: M.M., Design: E.Ş., F.T., Data Collection or Processing: A.E., Literature Search: A.E., F.T., Writing: M.M., E.Ş.

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