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Case Report

Serous Papillary Ovarian Type Tumors of the Testis: Two Rare Cases

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Abstract

Ovarian type epithelial high-grade and borderline tumors are rare in the testis and paratesticular tissue in men. Only about 40 cases reported in the literature to date and a few of them were metastatic. Due to rarity, there is no exact consensus in the optimal management of these patients. Herein we report two cases; first with a high-grade metastatic serous papillary carcinoma and another with a borderline serous papillary cystadenoma of testicular tissue.

Keywords: Testis tumors, paratesticular neoplasms, serous papillary ovarian type cancer

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Testicular cancer is the most common solid malignancy in males between the ages of 15 and 35, although it accounts for only 1% of all cancers in men. Germ cell tumors account for 95% of testicular cancers.[1] The unusual histologic type of serous papillary carcinoma of the testis is rare, therefore, standardized staging and treatment options do not exist.[2,3] Hence, sharing experience is critical to determine the best way to diagnose and treat these patients. Herein, we report two patients with ovarian type surface epithelial tumors of testicular tissue.

Case Reports

Case 1 — A 64-year-old male was admitted with a left scrotal swelling present for several weeks. Laboratory tests including tumor markers [beta-hCG<0.100 mIU/mL, CA-19-9<0.6, alpha-fetoprotein (AFP) 4.5 ng/mL] were within normal range, except for elevated carcinoembryonic antigen (CEA, 6.45 ng/mL) and CA-125 (65 U/ml). Scrotal ultrasound demonstrated a 12 cm paratesticular cystic mass with hyperechoic papillary projections protruding into the cyst lumen. A fluorine-18-fluorodeoxyglucose positron emission tomography/computed tomography (FDG PET/CT) revealed multiple pathological intraabdominal, mediastinal, supraclavicular, and cervical lymphadenopathies and a mass in the left testis.

The patient underwent a left radical orchiectomy with the removal of the paratesticular mass (Fig. 1). The supraclavicular lymph node was also excised. Histological examination demonstrated a tumor with predominantly micropapillary, glandular, and cribriform architecture (Fig 2a). Psammomatous calcification was also present. Histopathology of the supraclavicular lymph node revealed a metastasis of the papillary carcinoma (Fig. 2b). Immunohistochemical (IHC) staining demonstrated diffuse positivity for Ber-EP4, estro-

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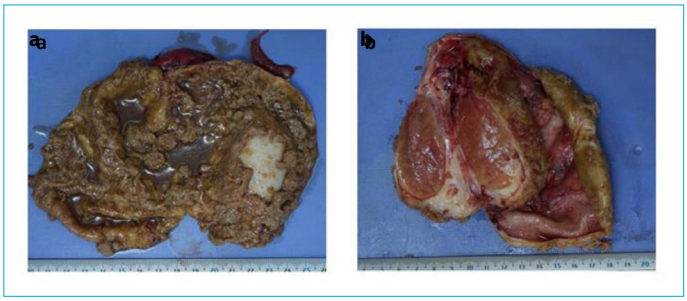


Figure 1. (a) Gross appearance of the tumor. The cystic tumor contained papillary formations. **(b)** Testicular tissue was intact.

gen receptor (ER), CA-125, PAX8, WT1, and CK7 and negative staining for progesterone receptor and calretinin. The histomorphological picture resembled a high-grade ovarian serous carcinoma and a final diagnosis of invasive serous cystadenocarcinoma was made.

Postoperative paclitaxel plus carboplatin was administered for a total of 7 cycles. Afterward, CA-125 regressed to 22 U/ml. A FDG PET/CT showed partial morphological and metabolic regression of the involved lymph nodes. Chemotherapy was discontinued after 7 cycles because of grade 3 neuropathy. Six months later, CA-125 progressed to 60 U/ml and FDG PET/CT showed multiple new and progressing mediastinal and abdominal metastatic lymph nodes. Second-line chemotherapy with gemcitabine plus carboplatin was administered for 8 cycles. After the chemotherapy, CA-125 decreased to normal levels. A FDG PET/CT rescanning for response evaluation demonstrated partial regression of the disease. Following 8 cycles of combination chemotherapy, maintenance therapy with gemcitabine on days 1, 8, and 15 every 28 days was started. After a total of 12 cycles, gemcitabine was discontinued due to stable disease and the patient is being followed up in good health 39 months after the first diagnosis.

Case 2 — A 35-year-old man presented with a complaint of new-onset left testicular swelling. Laboratory investigations including AFP, beta-hCG, CEA, and CA-125 were within normal levels. Ultrasonography showed a hyperdense mass lesion in the paratesticular area. With a preliminary diagnosis of paratesticular tumor, the patient underwent inguinal orchiectomy. Macroscopically, the papillary mass lesion was located between the tunica albuginea and tunica vaginalis. It seemed to be related to the epididymis (Fig. 3a). Microscopically, the tumor exhibited papillae with fibrovascular cores lined by a single layer of cuboidal epithelium (Fig. 3b). The ciliated and hobnail cells were clues to serous differentiation. Minimal atypia was seen in tumor cells with no invasion. However, atypia and increased cellularity were evident in one focus (Fig. 3c). The Ki-67 proliferation index of this focus was high. Tumor cells had diffuse

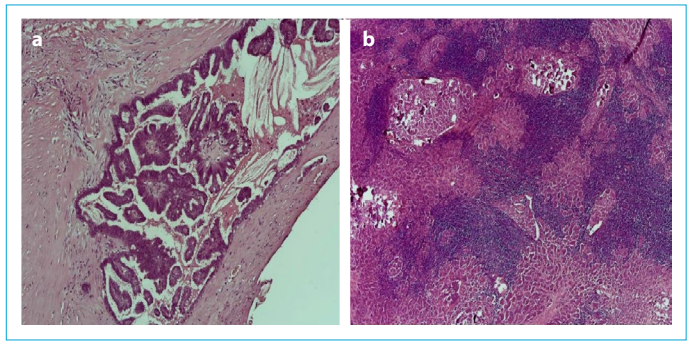


Figure 2. (a) Solid area shows tubulopapillary pattern in the cystic tumor. **(b)** Lymph node excisional biopsy showed malignant cells arranged in micropapillary and tubular pattern with psammomatous calcification.

staining for WT1, PAX8, CK7, ER, CA-125 and were negative for SALL4, OCT4, calretinin, and cytokeratin 5/6. A final diagnosis of borderline serous papillary cystadenoma was made. Staging CT of the chest and abdomen showed no evidence of metastatic disease. Therefore, active surveillance was initiated. At 15 months of follow-up, the patient remained free of recurrence or metastasis.

Discussion

Serous papillary carcinoma which derived from Mullerian elements is a rare malignancy in males. Its origin is not fully understood.[4] In this paper, we presented two patients whose tumors originated from different regions of the Mullerian tract; one from paratesticular tissue, the other from tunica albuginea. These tumors can present as benign, borderline, or malignant character and have similar morphologic and IHC features as their ovarian counterparts.[5]

The signs of presentation in the previously reported cases include hydrocele,[6] testicular fullness or mass,[5] and lymph node swelling.[7] Our two cases presented with testicular swelling despite different clinical stages. With regard to

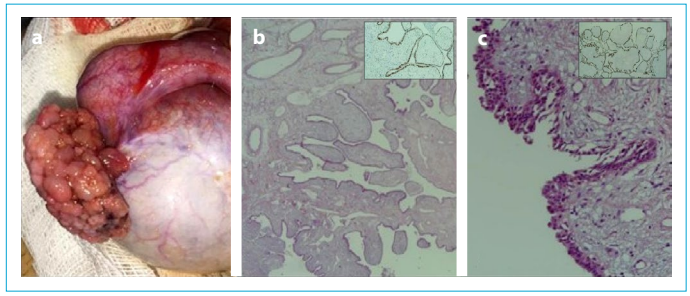


Figure 3. Gross and microscopic appearance of the borderline serous papillary tumor. Macroscopically, the papillary mass was located between the tunica albuginea and tunica vaginalis **(a)**. Microscopically, tumor exhibit papillae with cores lined by a single layer of cuboidal epithelium **(b)**. The atypia and cellularity increased in a focus **(c)**. Insets show immunohistochemical staining for ER **(b)** and CA-125 **(c)**.

staging, the FDG PET/CT imaging was used in our first case and demonstrated metabolically active metastases. This suggests the usefulness of this method in detecting and assessing nodal or distant metastatic disease, in both the initial staging and post-operative surveillance period. The use of FDG PET/CT has previously been described in only one case.[8]

Primary treatment of these tumors has been previously suggested as radical inguinal orchiectomy with hemi-scrotectomy. Blumberg HM et al. who has described the first case of metastatic serous papillary adenocarcinoma of the testis reported that chemotherapy is not efficient due to high resistance.[9] Mermershtain W et al. also indicated that there is not effect of chemotherapy in the adjuvant or palliative settings.[10] Ma Yuk Ting et al. treated their metastatic testicular serous carcinoma patient with weekly carboplatin 2 AUC and paclitaxel 60 mg/m^2, but after 3 weeks, he suffered from a seizure, and magnetic resonance imaging (MRI) of the brain demonstrated leptomeningeal carcinomatosis and chemotherapy was discontinued.[11] Conversely, Vaughn DJ et al. showed that these tumors with aggressive features may respond to chemotherapeutic regimens used in ovarian cancer and also CA-125 may be useful for surveillance.[2]

In conclusion, serous testicular tumors of male genital tract are very rare pathological diagnosis but according to current literature they may be similar to their ovarian counterparts. As such, management similar to that used in ovarian counterparts may be preferred.[12] As there are no standard management options for these entities, sharing experience through case reports is essential.

Disclosures

Informed Consent: Written informed consent was obtained from the patients' family for the publication of the case report and the accompanying images.

Peer-review: Externally peer-reviewed.

Conflict of Interest: None declared.

Authorship Contributions: Concept – E.A.; Design – E.A.; Supervision – E.G.; Materials – B.S.K.; Data collection &/or processing – S.T.; Analysis and/or interpretation – S.Ş.; Literature search – E.A.; Writing – E.A.; Critical review – E.G.

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