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Oncology

Ovarian type epithelial tumor of the tunica vaginalis with abdominal metastasis

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Introduction

Malignant paratesticular tumors are rare. We describe the first documented case of a paratesticular serous ovarian type epithelial tumor (OTET) associated with abdominal metastasis and its treatment.

Case presentation

A 36-year-old African American male was referred to urology clinic for a two year duration of left sided scrotal swelling. Outside ultrasound demonstrated a left sided simple fluid collection surrounding the testis consistent with hydrocele. The patient reported that at rest he was not bothered by his hydrocele, however his left hemiscrotum was painful when bumped. He denied fever, chills, dysuria, history of STI, and recent sick contacts. He had no significant past medical or surgical history. He reported daily tobacco use, smoking of only 5 cigarettes per day, occasional alcohol use and denied illicit drug use. He was adopted and therefore family history was unknown. Physical exam was unremarkable except for an enlarged left scrotum approximately the size of a softball.

Subsequent to findings of enlarged left sided scrotum on exam, a scrotal ultrasound was performed. This demonstrated a left sided hydrocele with concerns of peripheral cystic mass. A hydrocelectomy with intraoperative scrotal exploration and possible scrotoplasty was recommended.

In April of 2015 the patient underwent a left sided hydrocelectomy and scrotoplasty. Intraoperative findings included murky hydrocele fluid, thickened tunica vaginalis, and small polypoid masses studding the tunica vaginalis diffusely. Intraoperative frozen section analysis was reported as bland epithelial growth. Permanent analysis of this specimen revealed a low-grade infiltrating neoplasm of uncertain histogenesis into the tunica vaginalis that had some pathologic resemblance to an adenomatoid tumor. This specimen was negative for WT-1, D2-40, Calretinin and AFP. It was, however, positive for Pan-Cytokeratin and Ber-EP4 and thus was immunophenotypically more consistent with an epithelial neoplasm. P63 was equivocal and the Ki-67 labeling index was approximately 5%.[5]

The case was discussed at multidisciplinary tumor board and inguinal orchiectomy was the consensus recommendation. Preoperative tumor markers for B-hCG, AFP and LDH were all within normal limits. In July of 2015 the patient underwent a left sided radical orchiectomy, excision of the hemiscrotal scar, and insertion of a left sided testicular prosthesis. The pathological specimen was read as serous ovarian type epithelial tumor (OTET) of the tunica vaginalis with a positive spermatic cord margin and lymphatic metastasis (Fig. 1). Studies on the orchiectomy specimen included positive staining for PAX8, a result most often seen in tumors of the female urogenital tract - ovarian and endometrial carcinomas.

Further workup included a CT of the chest abdomen and pelvis to assess for metastatic disease that demonstrated retroperitoneal lymphadenopathy compatible with nodal metastases. His case was again discussed at a multidisciplinary tumor board and the consensus was to proceed with retroperitoneal lymph node dissection with possible adjuvant chemotherapy or radiotherapy.

In September of 2015 the patient underwent an open retroperitoneal lymph node dissection with excision of the left gonadal vein stump. This was performed using the full bilateral template. His hospital course was without complication and he was discharged on POD #5. Subsequent pathological analysis of the retroperitoneal nodes demonstrated that 10/19 lymph nodes were positive for metastatic OTET. He was offered adjuvant chemotherapy postoperatively for further treatment.

After the first cycle in November of 2015, the patient was admitted to the hospital due to an MRSA port site infection requiring port removal. In January of 2016 he was medically stable enough to resume his adjuvant chemotherapy and a second cycle of carboplatin and paclitaxel was initiated. He continued the remainder of the cycles without incident.

In 2011 there were reportedly fewer than 50 case reports of serous tumors of the testis and paratestis. Therefore, clinical experience with this type of serous carcinoma is minimal and data regarding chemotherapy is scant at best. Second opinion obtained at Memorial Sloan Kettering, which provided input regarding chemotherapy treatment after our patient's family contacted them. They recommended adjuvant

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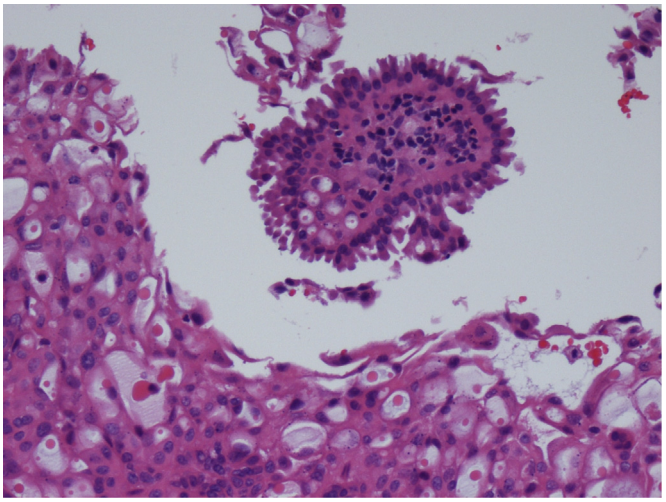

Fig. 1. H&E sections from the original local excision reveal an epithelial neoplasm predominantly composed of cells with abundant eosinophilic cytoplasm. Cytoplasmic vacuoles containing eosinophilic globules are prominent. There are distinct papillary foci.

paclitaxel and carboplatin in this case. Our patient had matted nodes and separate tumor nodules present at RPLND. Therefore, he was treated with paclitaxel 175 mg/m2 IV over 3 hours on day 1 followed by carboplatin (AUC 6) IV over 30 minutes on day 1 every 21 days. He had a repeat CT scan after 3 cycles of chemotherapy that revealed no measureable disease, and so he received 3 additional cycles of paclitaxel and carboplatin to achieve remission.[4]

Discussion

Paratesticular tumors are a rare occurrence, but have been documented in the literature. A malignant ovarian type epithelial tumor of the tunica vaginalis with abdominal metastasis treated with RPLND and adjuvant chemotherapy has never been reported in the literature. We present the first reported case of this rare tumor with rare characteristics that was successfully treated to the point of remission with a combined surgical and medical chemotherapy approach.[1]

The majority of paratesticular tumors are benign (70%) and are of mesothelial or soft tissue origin. The most common of these is the benign adenomatoid tumor of mesothelial derivation. This case was negative for all mesothelial markers (calretinin, WT-1 and D2-40), and stained with epithelial markers (BerEP4) and the Mullerian marker PAX-8. These staining characteristics fall into the category of ovarian type epithelial tumors (OTET). OTETs are often seen in association with hydrocele - as seen in this case.[2] Based on the initial histological finding of focal stromal invasion, the low grade appearance of the tumor and 5% Ki-67 labeling index, we initially favored that this was the most common variant of OTET - the serous borderline tumor. The subsequent finding of extensive nodal involvement pushed the diagnosis into the low grade serous carcinoma category, and indicated the need for adjuvant chemotherapy.[3,5]

Conclusions

In the case of this patient, a rare low grade Mullerian serous carcinoma (OTET) with abdominal metastasis was successfully treated with orchiectomy, RPLND and adjuvant chemotherapy with no known recurrence of disease to date. It demonstrates the feasibility and efficacy of a multidisciplinary approach to acheive remission. To our knowledge this is the first report of such a case in the literature.

Appendix A. Supplementary data

Supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.eucr.2018.05.007.

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