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Case Report

Low-grade serous carcinoma (mullerian/ovarian type) of the paratestis presenting as diffuse metastatic disease of unknown origin: Case report of an uncommon tumor with an unusual presentation

Alexander Filatenkov, MD[1], Amanda Strickland, MD[1], Matthew Karpowicz, DO, Franto Francis, MD PhD*

Department of Pathology, University of Texas Southwestern Medical Center, Dallas, TX 75390, USA

A B S T R A C T

Low-grade (papillary) serous carcinoma of ovarian type is rare in males and histologically identical to low-grade serous carcinomas in female patients. We present a case of paratesticular low-grade serous carcinoma in a 42 year old male, with the highly unusual initial presentation as diffuse metastatic disease in the abdomen. Imaging revealed a cystic lesion of the right testis/hemiscrotum. Biopsy of the abdominal metastasis and subsequent right orchiectomy specimen showed invasive papillary serous carcinoma, low grade, with prominent psammomatous calcifications. Immunohistochemically, both tumors were positive for PAX8, WT-1, CA-125 and cytokeratin CK7, and negative for mesothelioma markers (calretinin, D2-40) and CD10. The main differential diagnoses for this tumor include mesothelioma of tunica vaginalis, adenocarcinoma of rete testis/epididymis and metastases. Although rare as an initial presentation, ovarian type serous carcinoma of the paratestis should be included in the differential diagnosis in males with abdominal metastases that show papillary architecture, psammomatous calcifications and PAX8 immunoreactivity.

1. Introduction

Low-grade (papillary) serous carcinoma of ovarian type is rare in the testis and paratestis. The tumor is histologically identical to serous carcinoma arising in the ovary, and uses the same descriptive nomenclature. There have been < 40 cases described in the literature, and the majority have been locally confined tumors of either borderline type with non-invasive histology, or showing focal invasion in a background of predominantly borderline histology. On follow-up, the majority of these tumors have not recurred after treatment, with very rare cases showing distant metastasis on long term follow-up [2]. Most of these cases present as hydrocele and/or testicular swelling and the mean age of presentation for invasive tumors is 31 years (range: 16–42 years) [1–3].

We report what we believe is the first case of this rare tumor presenting initially with widespread abdominal metastases. Because of the rarity of this tumor in males and absence of reported initial metastatic presentation, pathologists need to be cognizant of this entity when evaluating metastases of unknown origin. Once the entity is included in one's differential diagnostic consideration, it is relatively easy to identify because of the tumor's distinctive features (histomorphology, psammoma bodies, immunohistochemistry). In our case, immunohistochemical workup further identified the tumor as analogous to low grade type of papillary serous carcinoma of the ovary.

2. Materials and methods

2.1. Case report

A 42 year old undocumented male immigrant with no significant medical history presented to clinic at the county hospital, complaining of constant, pressure like left flank abdominal pain. He also reported unintentional 20 lbs. weight loss over the previous six months and recently noticed a mass near his right testicle. No elevation of serum markers for testicular germ cell tumors was detected on blood testing.

Testicular ultrasound showed an extratesticular cystic structure with peripheral nodularity on the right side, without apparent central vascularity. At this point the differential diagnosis included

* Corresponding author: Department of Pathology, 5201 Harry Hines Blvd., Dallas, Texas 75235, USA.
E-mail addresses: alexander.filatenkov@phhs.org (A. Filatenkov), amanda.strickland@phhs.org (A. Strickland), matthew.karpowicz@utsouthwestern.edu (M. Karpowicz), Franto.Francis@utsouthwestern.edu (F. Francis).
[1] These authors contributed equally to this work.

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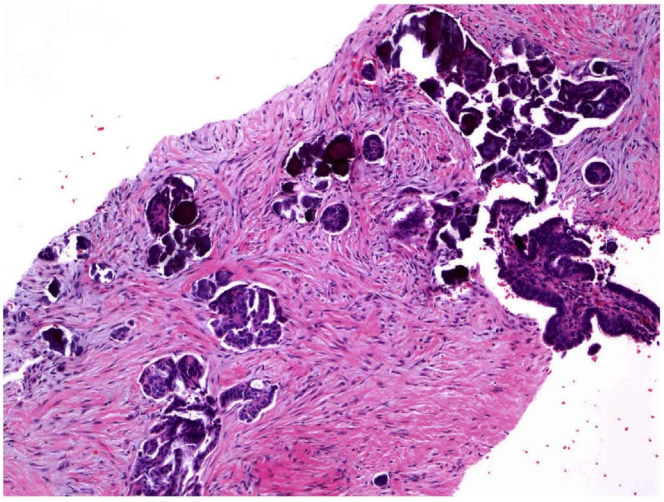

Fig. 1. CT guided needle biopsy of abdominal lesion revealed invasive carcinoma with papillary architecture and prominent psammoma bodies in a dense fibrous stroma.

posttraumatic/infectious hydrocele or a large exophytic epididymal cyst. Computed tomography (CT) revealed multiple nodules in the chest/pleura, partially calcified masses in the abdomen, low-density lesions in the liver and malignant ascites. Additionally, a partially calcified and cystic lesion was noted in the right hemiscrotum (Fig. 2A).

CT guided needle biopsy of an abdominal lesion was performed and revealed an invasive carcinoma with papillary architecture and prominent psammoma bodies within dense fibrous stroma (Fig. 1). Immunohistochemical staining of this tumor showed positivity for PAX-8, WT-1 and cytokeratins CK7 and AE1/AE3, and non-reactivity with TTF-1, CDX2, PSA as well as D2-40 and calretinin stains. The case was signed out as metastatic low-grade serous carcinoma with psammoma bodies, with a note that immunohistochemistry argued against the possibility of a lung, colon, prostate or pleural primary.

Because imaging suggested the likelihood of a paratesticular/testicular lesion, a right radical orchiectomy was performed shortly after the biopsy (Fig. 2B). A 70.5 g orchiectomy specimen was received, comprising of the testis, epididymis and spermatic cord. On opening the specimen, an approximately 4.0 cm cystic lesion filled with clear transparent fluid was identified within the tunica vaginalis, in the testiculo-epididymal groove region. The cyst showed multiple small tan-yellow papillary excrescences and hard nodules, as well as a subjacent, possibly contiguous 2.0 cm tan-brown mass. The rest of the testis was tan-brown and spongy with unremarkable parenchyma.

Microscopic examination of the specimen showed an invasive carcinoma with prominent papillary features and abundant psammoma bodies (Fig. 3). The neoplastic papillae were lined by moderately atypical, serous cuboidal and columnar cells. The mitotic count was low (< 3 per 10 high power fields) and no tumor necrosis was seen. Portions of the tunica vaginalis cyst lining away from the invasive tumor showed serous borderline tumor morphology, i.e. complex branching architecture and epithelial atypia, but without stromal invasion (Fig. 3). The tumor appeared to arise multifocally in the paratesticular region, from within the tunica vaginalis cyst and adjacent testiculo-epididymal groove region. The tumor extended into the epididymis, tunica albuginea and focally involved testicular parenchyma and hilar fat. Tumor cells were immunoreactive for PAX8, WT-1, estrogen receptor, cytokeratin 7, Ber-Ep4 and CA-125 stains, and negative for mesothelioma markers (calretinin & D240) as well as for CD10 immunostain, among other markers. Patchy to focal immunoreactivity was seen with p53 immunostain, i.e. “wild type” of p53 staining pattern, analogous to low grade type of papillary serous carcinoma in the ovary. Overall, the histomorphology and immunoprofile of the tumor were consistent with invasive, low-grade papillary serous carcinoma of mullerian/ovarian type arising in the paratestis, and morphologically similar to the lesion seen in the abdominal biopsy.

2.2. Follow-up

Following orchiectomy and diagnosis, the patient was scheduled for clinic visit to discuss initiation of chemotherapy, but did not show up. At two years following surgery, the patient had still not presented for continuation of care.

3. Discussion

In this case report, we report an uncommon tumor of the testis/paratestis - ovarian-type serous carcinoma- with a highly unusual initial presentation: widespread abdominal metastases. The majority of ovarian type serous tumors of the paratestis/testis reported in the literature have been non- invasive, and none presented initially with widespread metastases. Serous carcinomas of ovarian type are capable of metastatic spread, but they typically present as testicular hydrocele, or testicular fullness or mass [1,4]. The mean age of presentation for invasive tumors is 31 years (range: 16–42 years). Ovarian type epithelial tumors can be serous, mucinous, endometrioid, clear cell, transitional, [5,6] or squamous [2,7–9], with serous tumors being most common. Serous tumors can in turn be benign, borderline, or malignant (carcinoma). It has been suggested that ovarian type serous tumors in the testis/paratestis arise from either mullerian metaplasia of tunica vaginalis [11] or mullerian rests in paratesticular soft tissue or the appendix testis [9].

The current tumor was cystic and showed prominent papillary architecture and psammomatous calcifications, and was associated with

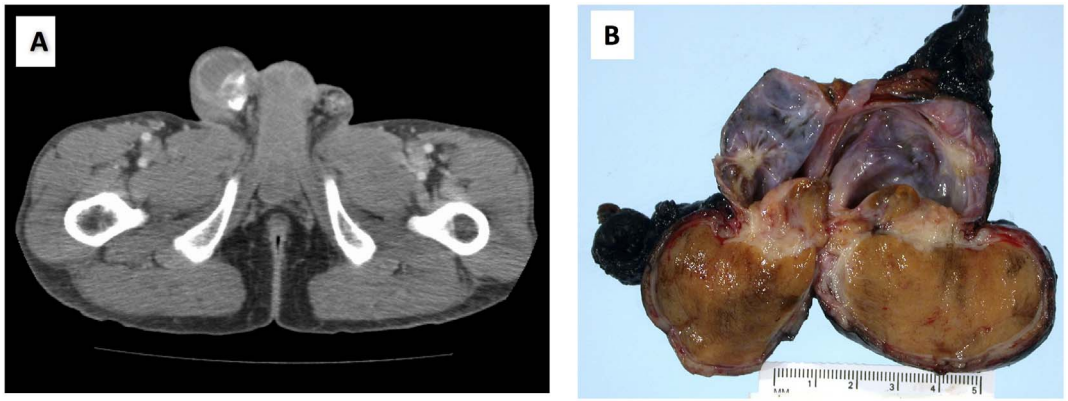

Fig. 2. A. Partially cystic, partially calcified mass noted in right hemiscrotum on CT. B. Gross exam showed 4.0 cm tunica vaginalis cyst with subjacent ~ 2.0 cm tan-brown mass and yellow-tan nodules in the cyst wall. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

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