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Metastatic Serous Carcinoma of the Testis: A Case Report and Review of the Literature

Yuk Ting Ma Shalini Chaudhri Michael H. Cullen

The Cancer Centre, University Hospital Birmingham NHS Foundation Trust,
Birmingham, UK

Key Words

Chemotherapy · Metastatic · Serous carcinoma · Testis

Abstract

Serous tumours of the testis and paratestis are rare, with fewer than 50 cases reported in the literature. The majority of the reported cases have been borderline serous tumours, and these tend not to recur or metastasize. Conversely, serous carcinomas can metastasize but this is often a late event. The presence of invasion in an otherwise borderline tumour has also been associated with the development of metastatic disease several years later, thus highlighting the importance of extensive sampling of all cases of borderline serous tumours. We report a case of a young man diagnosed with serous carcinoma of the testis, occurring 18 years after first diagnosis of a testicular germ cell tumour in the contralateral testis. This pattern has not previously been reported.

Introduction

Ovarian-type epithelial tumours of the testis and paratestis are rare [1]. Serous tumours are the most common subtype, but these remain rare with fewer than 50 cases reported in the literature so far [1–3]. We report a case of a young man diagnosed with serous carcinoma of the testis, occurring 18 years after first diagnosis of a testicular germ cell tumour in the contralateral testis. To our knowledge, this is the first time this pattern has been reported.

Case Presentation

A 16-year-old Caucasian man was diagnosed with a metastatic non-seminomatous germ cell tumour of the left testis in 1988. He underwent a left orchidectomy followed by 4 cycles of bleomycin, etoposide and cisplatin (BEP) chemotherapy and subsequent retroperitoneal lymph node dissection (RPLND); he achieved a complete response. In 1997 (9 years later), he developed recurrent metastatic disease in the retroperitoneum. This was treated with combination chemotherapy using cisplatin, vincristine,

methotrexate, bleomycin, actinomycin D, cyclophosphamide and etoposide (POMB/ACE) followed by RPLND. Microscopically, only a differentiated teratoma was identified and the resection was complete. In 2001 (4 years later), he developed relapsed disease in the mediastinum. This was treated with 3 cycles of chemotherapy using paclitaxel, ifosfamide and cisplatin (TIP) followed by a thoracotomy; complete clearance of the mediastinal disease was achieved.

In 2006 (a total of 18 years later), he presented with a several-month history of swelling of the right testis. The tumour markers α -fetoprotein (AFP), β -human chorionic gonadotropin (HCG), and lactate dehydrogenase (LDH) were normal. Testicular ultrasonography demonstrated a large cystic mass in the right scrotum with more solid nodular components and also foci of calcification, suggestive of a testicular germ cell tumour. CT scanning of the thorax and abdomen demonstrated no evidence of metastatic disease. He underwent a right orchidectomy. Macroscopically, there was a small papillary lesion on the surface of the tunica albuginea measuring 10×6 mm, and the testicular parenchyma underneath this lesion comprised a poorly circumscribed grey/white area measuring $20 \times 5 \times 10$ mm. Microscopic examination of the papillary area on the surface of the tunica albuginea revealed a complex papillary branching pattern with small, detached clusters of neoplastic cells at the tip of the papillae. The cells lining the papillae were columnar with epithelial stratification and showed vesicular nuclei with atypia and mitoses. Psammoma calcification was also noted. The solid area continuous with the papillary area contained poorly differentiated cells in clusters and sheets with associated stromal desmoplasia. Both the papillary and solid components were positive for the epithelial markers EP4, carcinoembryonic antigen (CEA), cytokeratin 7 (CK7), epithelial specific antigen and epithelial membrane antigen (EMA), and negative for the mesothelial markers calretinin, thrombomodulin, CK20 and vimentin. A histological diagnosis of serous carcinoma of the testis arising from a papillary serous tumour of borderline malignancy was made. Following surgery, he remained well and was eventually discharged from regular follow-up in 2009.

In 2010, he presented to his local hospital with ascites. The full blood count and tumour markers (AFP, HCG and LDH) were normal. The biochemical profile showed mild renal impairment which had been present for several years. CT scanning of the thorax and abdomen revealed significant abdominal ascites but no other evidence of metastatic disease. Laparoscopic examination demonstrated the presence of multiple peritoneal 'seedlings', and a biopsy was taken. Histological examination of the peritoneal biopsy revealed metastatic serous carcinoma. The ascites was drained and he was commenced on palliative chemotherapy using carboplatin (AUC 2) and paclitaxel (60 mg/m^2) administered on days 1, 8 and 15 of a 28-day cycle. After 3 weeks of treatment he suffered a grand mal seizure, and MRI of the brain demonstrated focal leptomeningeal carcinomatosis. Unfortunately, he continued to deteriorate and chemotherapy was discontinued.

Discussion

Serous tumours of the testis and paratestis are rare, with fewer than 50 cases reported in the literature [1–3]. These tumours usually occur in young to middle-aged adults and typically present as a testicular mass [4]. Our patient is unusual because he had previously been diagnosed with a testicular non-seminomatous germ cell tumour in the contralateral testis. This association has not previously been reported. The majority of the reported cases have been borderline serous tumours, and these tend not to recur or metastasize. Conversely, serous carcinomas can metastasize but this is often a late event [4, 5]. This was also a feature of our patient; he presented with ascites due to peritoneal disease 4 years after the original diagnosis of serous carcinoma of the testis. Although the association of serous carcinoma with areas of borderline differentiation within the same tumour is rarely seen in the ovary, a combined pattern is not uncommon in serous carcinomas of the testis [4, 6, 7]. This was also seen in the original testicular lesion in our case. The presence of invasion in an otherwise borderline tumour has been associated with the development of metastatic disease several years later, thus highlighting the importance of extensive sampling of all cases of borderline tumours [4].

The main differential diagnosis is malignant mesothelioma of the tunica vaginalis, which is characterized by an aggressive natural history [8]. The immunohistochemical profile can help to differentiate these tumours. Serous carcinomas of the testis are usually positive for the ovarian epithelial markers EMA, CA125, CK7, EP4 and S-100, and negative for the mesothelial markers calretinin, thrombomodulin and vimentin.

The origin of ovarian-type epithelial tumours of the testis and paratestis remains uncertain. Some authors have suggested that serous tumours may arise from epithelial cells of müllerian duct remnants, such as the appendix testis, or müllerian duct remnants along the testiculoepididymal groove [9]. This theory is supported by the occurrence of many tumours in this location. Alternatively, it has been suggested that cells derived from the mesodermal epithelium may retain their ability to differentiate into different cell types when they undergo neoplastic transformation. This hypothesis helps to explain why serous tumours can arise from structures such as the tunica albuginea, the tunica vaginalis and the spermatic cord [9].

Due to the rarity of serous carcinomas of the testis and paratestis, there has been minimal clinical experience worldwide in the management of these tumours. It has been suggested that adopting the same treatment used for serous carcinomas of the ovary is a reasonable approach; we treated our patient with weekly carboplatin and paclitaxel.

In summary, serous tumours of the testis and paratestis are rare. Extensive sampling of borderline tumours should be undertaken, and patients with any invasive component should be followed up for several years as metastatic disease is often a late event.

Disclosure Statement

All the authors declare that they have no competing interests.

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Erratum

In the article ‘A Retroperitoneal Dedifferentiated Liposarcoma Producing Granulocyte Colony-Stimulating Factor Accompanied by Spontaneous Rupture: PET/CT Imaging of a G-CSF-Producing Tumor’ (Case Rep Oncol 2011;4:236–241) by Hara, the following authors also contributed to the paper:

Tomohiro Fujii, Yoshimasa Jo, Teruhiko Yokoyama, Yoshiyuki Miyaji, Atsushi Nagai

Department of Urology, Kawasaki Medical School, Kurashiki City, Japan