Borderline Serous Papillary Tumour of the Testis: A Case Report and Review of the Literature

AMAR S. IBRAHIM[1], CHENG LI[2] and MOHAMMAD S. AL-JAFARI[2]

[1]*Laboratory Medicine and Pathobiology, Toronto General Hospital, University Health Network, Toronto, ON, Canada;*
[2]*Department of Histopathology, Warrrington Hospital, Warrington, U.K.*

Abstract. *Borderline serous tumour of the testis and paratestis is an uncommon entity. We report a case of borderline serous tumour of a 59-year-old male, who presented with a right testicular swelling which was clinically suspicious of carcinoma. Radical orchidectomy was performed and a cystic lesion was identified in the testis. Macroscopically the tumour was composed of a unilocular cyst with excrescences in the inner surface. The histological features were identical to the ovarian counterpart of borderline serous papillary tumour. The excrescences were formed by stratified columnar epithelium, which exhibited mild nuclear pleomorphism and mitotic activity, with a fibrovascular core and scattered psammoma bodies. There was no lymphovascular or stromal invasion. The lesion was surrounded by a dense fibrous wall. On immunohistochemistry, the lining epithelial cells expressed cytokeratin AE1/AE3 but not carcinoembryonic antigen or calretinin. Following the removal of the tumour, the patient was followed up and no recurrence or metastasis has occurred to date. This case highlights the need for clinicians and pathologists to be aware of this rare entity and to develop the best approach for patient management.*

Serous epithelial tumours are common tumours of the ovary. The epithelium forming the tumours histologically resembles the epithelial lining of the Fallopian tube. Such tumours are rare in the testis and thus it is a generally unfamiliar entity to clinicians and pathologists. The existing literature suggests that approximately fifty cases have been reported worldwide. Most cases occur in young to middle-aged adults and usually present as a testicular mass (1, 2). The case we report here is of a 59-year-old man resented as a suspicious malignant tumour in the right testis.

Correspondence to: Mohammad S. Al-Jafari, Department of Histopathology, Warrrington Hospital, Warrington, WA5 1QG, U.K. Tel: +44 1925275536, Fax: +44 1925662043, e-mail: msaljafari@gmail.com

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Case Report

On physical examination, the patient had right testicular swelling which was suspicious of a malignant tumour. The blood biochemistry was within normal limits: urea: 5.5 mmol/l, creatinine: 80 μmol/l, sodium: 141 mmol/l, potassium: 4.4 mmol/l. The markers were: human chorionic gonadotropin (hCG): <2 U/l (normal 0-2), alpha-fetoprotein (AFP): 6 ng/ml (normal <7), lactate dehydrogenase (LDH): 264 U/l (normal 90-500). Computerised tomography (CT) scan of chest, abdomen and pelvis showed no significant intra-abdominal lymphadenopathy. No abnormality was seen in the liver, spleen, right kidney, pancreas or adrenal glands. There was no mediastinal lymphadenopathy or destructive bone lesion. Radical orchidectomy was performed and the right testis with the spermatic cord was excised.

The specimen consisted of a testis and spermatic cord covered by *tunica vaginalis*. The testis measured 40×35×28 mm. The epididymis and the attached spermatic cord were all normal. Slicing of the testis revealed a unilocular cyst measuring 28×23×22 mm lying within the testicular parenchyma. The cyst contained thick coffee-colored fluid. There were excrescences on the inner surface of the cyst, which was surrounded by a thin fibrous capsule (Figure 1). The background testicular tissue appeared macroscopically normal.

The tumour was processed and submitted for histological examination in its entirety. Histologically, the cystic lesion was composed of densely fibrous stroma, resembling ovarian type fibrous stroma, lined by partially ciliated cuboidal to columnar epithelium. There were numerous pigmented histiocytes underlying the epithelial lining, suggestive of previous haemorrhage (Figures 2 and 3). The excrescences were composed of papillary tufts lined by stratified columnar epithelium. A mild nuclear pleomorphism and scanty mitotic figures were found within the lining epithelium. Foci of calcification and psamomma bodies were seen. There was no evidence of lymphovascular or stromal invasion identified (Figure 3). Immunohistochemically, the epithelium of the papillary tufts expressed cytokeratin AE1/AE3 but not

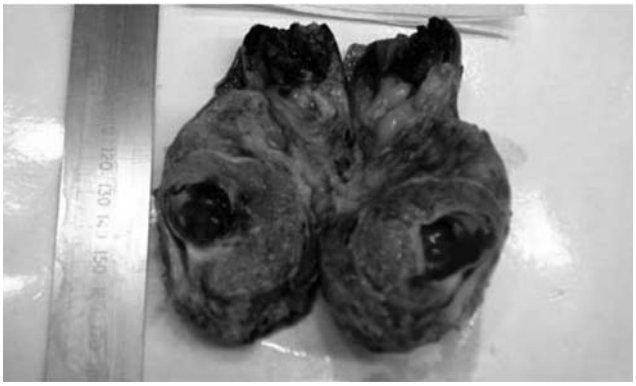

Figure 1. *Gross appearance of right orchidectomy specimen. The unilocular cystic lesion can be seen to lie in the testicular parenchyma and contains coffee-coloured fluid with excrescences on the inner surface.*

carcinoembryonic antigen and calretinin. The morphology and immunoprofiles are those of a borderline serous papillary tumour. The surrounding testicular tissue was normal and there was no evidence of intra-tubular germ cell neoplasia. Following the removal of the tumour, the patient was followed up and no recurrence or metastasis has occurred to date.

Discussion

All the known types of epithelial tumour that are morphologically identical to the ovarian surface epithelial tumours have been reported in testicular and paratesticular tissues. The most commonly reported are serous tumors with the majority of the cases being of the borderline type (1, 2). Other types of ovarian tumours have also been reported in testicular and paratesticular tissues including mucinous, endometrioid, clear cell and transitional (Brenner) tumours and serous carcinoma (3-7). Borderline serous tumours tend not to recur or metastasize. Conversely, serous carcinoma can metastasize and is associated with an unfavourable prognosis. Furthermore, the presence of invasion in an otherwise borderline tumour has been associated with the development of metastasis several years later (8). Therefore, extensive sampling of all cases of borderline tumour is important. The borderline serous tumour we report here was processed in its entirety for histological examination. No stromal invasion was identified and there has been no evidence of recurrence or metastasis.

The histogenesis of these epithelial tumours in testicular and paratesticular tissues is a matter of considerable discussion. Several hypotheses exist in the literature. Some suggest that these tumours arise from the remnants of Müllerian ducts found in the male appendix testis, epididymis, connective tissue between testis and epididymis and spermatic cord. Others suggest Müllerian metaplasia of the surface lining mesothelium and metaplasia of mesothelium within testicular parenchyma are the sources of origin. The latter provides a reasonable explanation of the pathogenesis of such tumors arising in the *tunica vaginalis* (2, 3, 8).

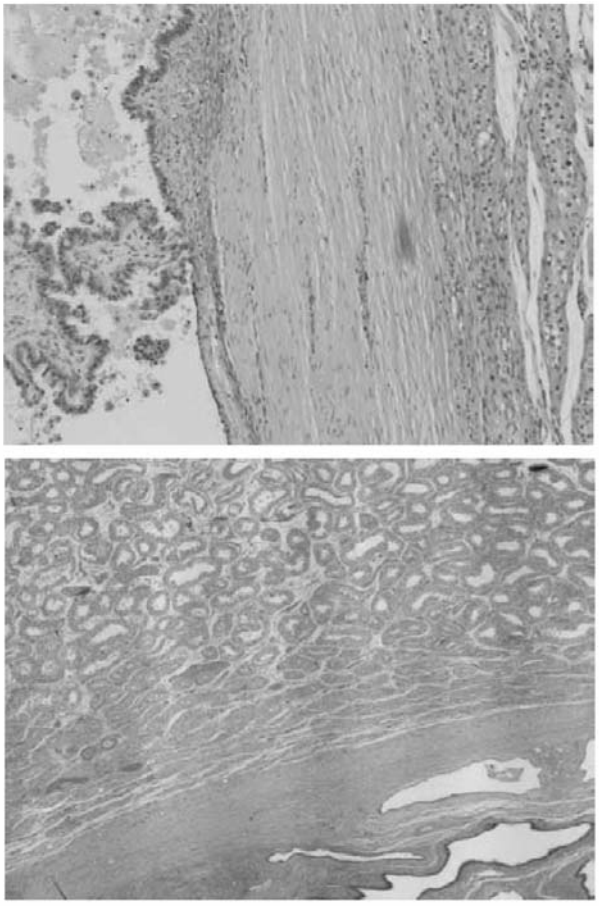

Figure 2. *Histological appearance of the serous borderline tumour. The tumour consists of a fibrous wall with exophytic papillary excrescences, with adjacent normal testicular tubules. (×40).*

These tumours usually occur in young and middle-aged adults. The mean age of patients with borderline tumours is 56 years (range 14-77 years) and for invasive tumours, 31 years (range 16-42 years) (4-6). Dull pain, swelling, palpable mass and associated hydrocoele are presenting signs and symptoms. Cancer antigen 125 (CA-125) levels may be elevated (4).

On gross examination, borderline tumours of the testicular and paratesticular tissues are almost always cystic with a thin fibrous capsule, whereas invasive carcinomas are usually non-cystic and more infiltrative (4). Microscopically, features of borderline serous testicular tumours are identical to the morphology of the same tumours encountered in the ovarian counterparts. These tumours usually reveal papillae with

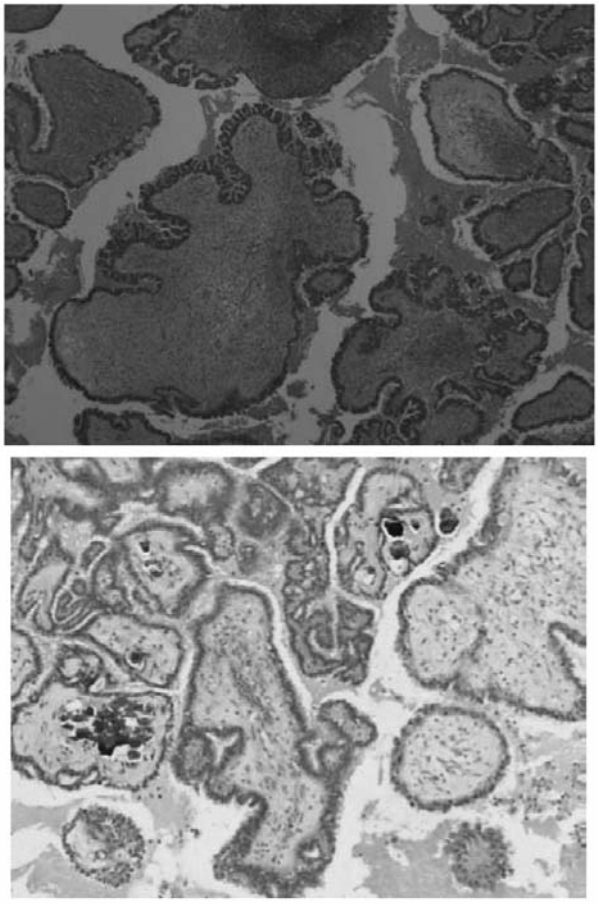

Figure 3. *Histological appearances of the borderline serous tumour. The papillary tufts were lined by stratified columnar epithelial cells with fibrovascular cores. Psammoma bodies and calcification were visible (×100).*

fibrovascular cores lined by stratified cuboidal to columnar epithelium with minimal cytological atypia and no stromal invasion. Variable mitotic activity and psammoma bodies, ciliated and hobnail cells are usually present (4-6).

The scarcity of these tumours in the testis has led to difficulty and confusion regarding their diagnosis. Such tumours can be confused with other tumour types such as mesothelioma. Some criteria have been useful in differentiating borderline serous tumours from mesothelioma. Localized and nondestructive gross appearance, the presence of ciliated lining epithelium and psammoma bodies, as well as positivity for cytokeratin CK7, estrogen, and negativity for CK20 and calretinin, all favour the diagnosis of serous borderline tumours. Positivity for calretinin favors the diagnosis of mesothelioma (7, 8).

In summary, borderline serous tumours of the testis and paratestis are rare, and clinicians and pathologists should bear this entity in mind when dealing with uncommon testicular tumours. The identification of invasive components in an otherwise borderline tumour is of prognostic significance, therefore, thorough sampling should be undertaken. The prognosis for patients after complete excision of borderline tumours is excellent, nevertheless, clinical follow-up is necessary for at least several years. Furthermore, relatively insufficient clinical information and experience in managing such patients owing to the rarity of these tumours highlight the need to develop the best management approach for patients with borderline serous tumours of the testis and paratestis.

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