CASE REPORT

An unexpected finding of a rare intrascrotal lesion: the sertoliform cystadenoma of the rete testis

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Dr Peter G Vaughan-Shaw,
pvaughan-shaw@nhs.net**SUMMARY**

A 19-year-old man presented with a right testicular swelling. Testicular ultrasound demonstrated areas suspicious for malignancy and so it was decided to proceed for a right radical inguinal orchiectomy. Initial histological examination revealed a multiloculated cystic lesion at the area of the rete testis yet normal testicular parenchyma elsewhere. Specialist histological opinion was sought from the regional teratoma multi-disciplinary team which confirmed the diagnosis of a sertoliform cystadenoma, an extremely rare benign testicular neoplasm. The recognition of the benign nature of the mass enabled complete reassurance to be offered to the patient and avoided further oncological treatment.

BACKGROUND

There has been an ever-increasing worldwide incidence of testicular tumours in young men over the last 40 years. The exact aetiology behind this rise is likely to be an early recognition with ultrasonography in industrialised countries though several studies do put forward an environmental hypothesis.¹ While the vast majority of testicular neoplasms are germ cells in origin, it is important to appreciate that alternative pathology may exist. A sertoliform cystadenoma is rarely encountered and has been infrequently reported in the literature. However, it is important that such lesions are recognised, as given their benignity, they do not necessitate further oncological treatment. Furthermore, the physical and psychological implications of a benign diagnosis, albeit postorchidectomy, are absolutely paramount in this relatively young age group. Clinicians should consider this rare diagnosis and seek a specialist histopathological opinion in case the diagnosis is not conclusive.

CASE PRESENTATION

A 19-year-old man was referred to the urology outpatient's clinic by fast track referral from his general practitioner. He presented with a 6-month history of a 'lump in his right testicle.' It had not changed in size, texture or location during that time. Upon examination a non-tender, smooth lesion was identified within the body of the right testis. The patient was fit with no significant medical or family history. He was not on any regular medication.

INVESTIGATIONS

Testicular ultrasonography was performed and was reported as showing an abnormal area in the right testis. Within this area, several cysts were identified; the largest two were of about 1 cm diameter with a

possible coexistent solid area. This was reported as highly suspicious of malignancy. Tumour markers were normal with a lactate dehydrogenase of 326 IU/l, α -fetoprotein levels of 1 μ g/l and β -human chorionic gonadotropin (HCG) levels of <1 mIU/ml. Routine blood tests were unremarkable.

DIFFERENTIAL DIAGNOSIS

Given that the majority of testicular tumours are germ cells in origin, differential diagnoses are limited, particularly in this case. The most important differential diagnosis is that of a sertoli cell tumour of the testis. In total, 12% of these have malignant potential.²

TREATMENT

The patient received appropriate preoperative counselling and proceeded to a right radical inguinal orchidectomy and testicular prosthesis insertion.

OUTCOME AND FOLLOW-UP

The patient made an uncomplicated recovery and was discharged the following day. The removed testis was analysed by the histopathologist as outlined below.

On sectioning the testis, a multiloculated cystic lesion was identified at the area of the rete testis measuring 22×12×8 mm. The rest of the testicular parenchyma was macroscopically unremarkable.

Upon microscopy, there was a proliferation of columnar and cuboidal cells with a tubular architecture (figures 1–3). Nuclei were monomorphic with occasional visible nucleoli. Focally there was cytoplasmic vacuolation. The lesion was located at the rete testis with a predominantly intraluminal pattern of growth and subsequent dilation of rete testis lumen. Tumour cells appeared to be embedded within

Figure 1 Testis, H&E, ×25. Note intraluminal pattern of growth, cystic dilatation of rete testis and focal stromal hyalinisation.

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Figure 2 Testis, H&E, x50. Note intraluminal pattern of growth and tubulocystic arrangement of cytologically bland columnar and cuboidal cells.

hyalinised stroma. Mitoses were sparse (<1 mitosis/10 hpf). No lymphovascular or perineural invasion was demonstrated. Background testicular parenchyma was unremarkable with no evidence of intratubular germ cell neoplasia (ITGCN).

Upon immunohistochemistry (figures 4 and 5), tumour cells were strongly positive for inhibin and calretinin. There was focal weak positivity for p53 immunostain. The proliferation index (Ki-67) was less than 5%. Cells were negative for HCG, placental alkaline phosphatase (PLAP), CD56, synaptophysin, chromogranin, PSA, CD117, α fetoprotein, pancytokeratin, epithelial membrane antigen (EMA) and carcinoembryonic antigen (CEA). CD30 immunostain was non-contributory.

Specialist histopathological opinion was sought at the regional teratoma multi-disciplinary team meeting where the morphological and immunohistochemical features were confirmed as those of a sertoliform cystadenoma of the rete testis. The patient was informed of the benign nature of the lesion as an outpatient appointment and continued recovery to complete health.

DISCUSSION

The majority of testicular cancers are germ cells in origin and these are further subdivided into seminomas (40%) and non-seminomas (60%). While a relatively uncommon cancer, it is the most common cancer in young men. Growths of the testicular duct collecting system are rare and often clinically misdiagnosed as growths of surrounding structures. One site of potential para-testicular tumour is the rete testis.

Lesions of the rete testis can be further subclassified into non-neoplastic and neoplastic. The neoplastic lesions are most

Figure 3 Testis, H&E, x200. Note bland nuclear features of lesional cells and focal cytoplasmic vacuolation.

Figure 4 Testis, calretinin stain tumour cells strongly immunoreactive for calretinin immunostain.

commonly advancement of tumours from the testicular body itself or adenocarcinoma of the rete testis. Sertoliform cystadenomas are a rare benign neoplastic growth of the rete testis, with only six cases identified in the literature.²⁻⁴

Cystadenomas present as unilateral scrotal masses range from 1.5 to 3.6 cm and can occur between the ages of 21 and 79.⁵ Discrimination based on radiological investigation is rarely useful. Orchidectomy and histological examination is therefore required to exclude a malignant process and confirm the diagnosis. Histologically, it is recognised by tumour cells within the cystic areas of the rete testis which are confined in epithelium. The sertoliform variety is such named as there are tall columnar cells with basal nuclei within the proliferations of tubules. These bear close resemblance to the architecture of the sertoli cells.

Immunohistochemical stains are similar for the tumours and hence the relevance is once again to allow differentiation upon location; sertoli cell tumours being intratesticular and sertoliform cystadenomas located within the rete testis. The diagnosis has a huge clinical benefit as the main differential and the more common finding is that of a sertoli cell tumour, which can undergo malignant change and requires further oncological treatment.² A prompt diagnosis of a benign testicular lesion avoids undue patient anxiety, further oncological treatment and unnecessary postoperative surveillance.

Figure 5 Testis, inhibin stain tumour cells strongly immunoreactive for inhibin immunostain.

With increasing availability and resolution of MRI, it has been suggested that such a modality could be employed where ultrasound is inconclusive, for example where a paratesticular lesion has been suggested and to avoid unnecessary orchidectomy.^{6 7} An organ-sparing approach would involve histological confirmation of a benign diagnosis from a biopsy or intraoperative frozen section sample and a period of surveillance. However, such an approach may still risk a missed malignant process. Given the rarity of this lesion we believe that neither radiologists nor pathologists could confidently exclude malignancy preoperatively and so an organ-sparing approach is neither practical nor without risk. Therefore, we conclude that there is no realistic way of confirming the diagnosis of benign para-testicular tumour preoperatively and as such clinicians should continue to manage these cases with radical orchidectomy as a standard of care until improved modalities of preoperative diagnosis is achieved.

Learning points

- ▶ With the increasing incidence of testicular neoplasms knowledge of particular tumour types is crucial.
- ▶ All suspicious testicular lesions should be treated as malignant. Radical orchidectomy is the standard primary treatment.
- ▶ Specialist histopathological examination is often required owing to the rarity of this lesion and should be sought early, usually via specialist regional multi-disciplinary team meeting.
- ▶ Benign testicular lesions require no oncological treatment or postoperative surveillance.

Competing interests None.

Patient consent Obtained.

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