# Primary pure carcinoid tumour of the testis: A case report and review of the literature

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**Summary**Primary testicular carcinoid tumours (TCT) are very rare, and a large tumour size and the presence of carcinoid syndrome predict a malignant course. Histologically, it is difficult to differentiate between benign and malignant TCTs. We report a case of a primary pure TCT with an unusual presentation in a 23-year-old man, who had an asymptomatic, enlarged scrotum on the right side for 7 years. On gross examination, the tumour was 9.6 cm in diameter. The Ki-67 labelling index was 19.8%. High inguinal orchidectomy was performed, and 30 months after surgery the patient remains asymptomatic.

**KEY WORDS:** Primary pure carcinoid tumour; Prognosis; Testicular cancer.

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### Introduction

Testicular carcinoid tumours (TCTs) are rare neoplasms, accounting for 0.2-1% of all testicular tumours (1, 2). To date, approximately 60 TCT cases have been described in the literature (2, 3). TCTs can be divided into three subgroups: primary pure TCTs, carcinoid tumours associated with teratomas (~20% of cases), and carcinoid metastasis to the testis (~9% of cases) (4). Upon detection of a TCT, a multimodal approach should be taken to exclude the possibility of a metastasis from another organ. The mean age of patients with TCTs at diagnosis is 46 years (range, 10-83 years (4). Patients with carcinoid tumours may present with a self-detected testicular mass or testicular ache (as occurs with common testicular tumours) or, more infrequently, carcinoid syndrome with red-hot flushing (face, neck, and upper chest), severe and debilitating diarrhoea, abdominal pain, palpitations, and bronchospasms (5).

Herein, we report on a young patient with a primary pure TCT having a high malignant potential, and discuss the clinicopathological features, diagnosis, treatment, and prognosis in relation to the relevant literature.

### **C**ASE REPORT

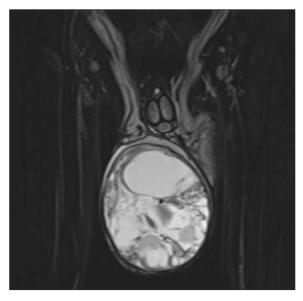
A 23-year-old man had an asymptomatic, enlarged scrotum on the right side for 7 years. He did not have a his-

tory of testicular trauma, haematuria, undescended testis, systemic symptoms, or weight loss, and there was no family history of testicular cancer. Physical examination revealed a hard, non-tender testicular mass in the right testis. The left testis appeared normal. Magnetic resonance imaging revealed a multilocular cystic tumour, measuring  $8.5 \times 7.8 \times 9.6$  cm in the right testis (Figure 1). Betahuman chorionic gonadotropin and alpha-fetoprotein lactate dehydrogenase levels were normal. Computed tomography scanning of the chest, abdomen, and pelvis revealed no significant para-aortic or iliac lymphadenopathy, no pulmonary abnormality, and no intestinal tumour. These findings favoured the diagnosis of a primary testicular tumour. The patient underwent high inguinal orchidectomy of the right testis.

Gross examination revealed that the right testis measured  $9.0 \times 8.5 \times 9.6$  cm.

Cut sections of the tumour demonstrated a lobulated cystic lesion with more than one yellowish septum, and regions of haemorrhage and necrosis.

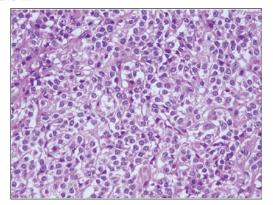
**Figure 1.** Magnetic resonance imaging revealed a multilocular cystic tumour, measuring  $8.5 \times 7.8 \times 9.6$  cm in the right testis.



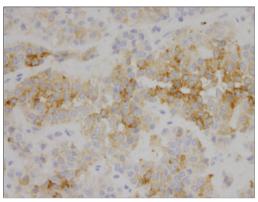
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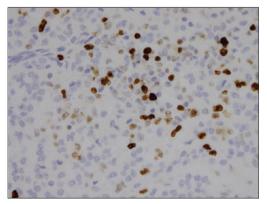
Figure 2.



**A)** Microscopic appearance of the tumour showing monomorphic cells arranged in a nested trabecular pattern (haematoxylin & eosin-stained, ×40 magnification);



**B)** Tumour cells exhibiting strong immunopositivity for synaptophysin (×40 magnification).



C) Ki-67 positive cells (×40 magnification).

The tumour was confined to the testis and epididymis without lymphovascular invasion. Histology revealed monomorphic cells arranged in a nested trabecular pattern (Figure 2a). The tumour cells were circular or polygonal with round to oval nuclei, a distinct nuclear membrane, granular cytoplasm, and 'salt and pepper'-like chromatin. An immunohistochemical examination revealed the tumour cells to be strongly immunopositive for synaptophysin, facilitating the diagnosis of a neuroendocrine tumour (NET) (Figure 2b), but negative for inhibin,

haematopoietic progenitor cell antigen, epithelial membrane antigen, and calretinin. The Ki-67 labelling index was 19.8% (Figure 2c).

No teratomatous elements were identified, and a diagnosis of primary pure TCT was confirmed. At 30 months after surgery, the patient remains asymptomatic.

## Discussion and Supplementary References are posted on www.aiua.it

### CONCLUSION

Our case adds to the other reports in the literature of primary TCTs having a high malignant potential. Our patient should undergo frequent biochemical and radiological examinations and long-term monitoring for recurrence and the development of metastases.

### REFERENCES

- 1. Rathert M, Ubrig B, Atkins DJ, Roth S. Carcinoid tumor of the testis. Urologe A. 2011; 50:340-2.
- 2. Alsharif S, Al-Shraim M, Alhadi A, et al. Primary neuroendocrine tumor of the testis. Urol Ann. 2014; 6:173-5.
- 3. Zavala-Pompa A, Ro JY, el-Naggar A, et al. Primary carcinoid tumor of testis. Immunohistochemical, ultrastructural, and DNA flow cytometric study of three cases with a review of the literature. Cancer. 1993; 72:1726-32.
- 4. Stroosma OB, Delaere KP. Carcinoid tumours of the testis. BJU Int. 2008; 101:1101 5.
- 5. Merino J, Zuluaga A, Gutierrez-Tejero F, et al. Pure testicular carcinoid associated with intratubular germ cell neoplasia. J Clin Pathol. 2005; 58:1331-3.

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