Balkan Med J 2014;31:191-2 © 2014



# A Small Cell Carcinoma in the Testis Associated with Testicular Teratoma

Esma Türkmen, Bülent Erdoğan, Hilmi Kodaz, Sernaz Uzunoğlu

Department of Medical Oncology, Trakya University Faculty of Medicine, Edirne, Turkey

Dear Editor,

Neuroendocrine neoplasms of the testis are extremely rare, accounting for 0.2% of all testicular neoplasms. Although the origin of the primary testicular neuroendocrine tumour is controversial, components of mature teratoma, leydig cells and intratubular germ cell neoplasia have been reported as precursor lesions (1, 2). Although a case of small cell carcinoma (SmCC) originating from an ovarian teratoma has been reported (3), to our knowledge there are no reports in English including SmCC of the testis originating from a testicular teratoma. In this paper, we present a metastatic SmCC of the testicle associated with testicular teratoma, along with its pathological properties, clinical course and treatment results.

A 44-year-old male patient with a painless testicular mass underwent left radical orchiectomy in another hospital in 2010. The histopathological diagnosis was a 12 cm mixed germ cell tumour (GCT) composed of mature teratoma and embryonic cell carcinoma. Beta-human-chorionic-gonadotropin, alpha-fetoprotein levels and lactate-dehydrogenase levels were normal. Multiple pulmonary nodules (the largest 6 cm) and paraaortic lymphadenopathy (the largest 1.5 cm) were detected by computed tomography (CT).

The patient was relatively old for non-teratoma non-seminoma GCT and tumour markers were normal; thus the orchiectomy material was reexamined in our centre to exclude other neoplasms besides GCT. Immunohistochemical and morphological findings were consistent with SmCC originating from mature teratoma (chromogranin, synaptophysin, neuron-specific enolase positive, CD56, MIC-2, GFAP negative).

Although the patient had a diagnosis of SmCC, we treated him according to the principles of GCT treatment. Based on the knowledge that extrapulmonary SmCCs may behave in a different way than pulmonary SmCCs in terms of the organ of origin, we opted for germ cell carcinoma treatment rather than classical SmCC. As the chemotherapy protocol (cisplatin/etoposide/bleomycin) that is effective against GCT involves similar agents and doses as the cisplatin/etoposide protocol used against SmCC, we applied four cycles of cisplatin/etoposide/bleomycin. After partial response had been achieved, two additional cycles of cisplatin/etoposide were applied (to protect against bleomycin toxicity). After six courses of chemotherapy, the paraaortic lympadenopathy in the abdominal CT disappeared and a large number of lesions disappeared on the thorax CT, but three lesions (the largest 2 cm) remained. Three months after the completion of chemotherapy, pathological FDG involvement was not observed in residual pulmonary lesions on PET/CT.

In accordance with the GCT treatment approach, metastasectomy oriented to teratoma and microscopic residual disease was planned. However, the patient declined surgery. The patient has since been followed without recurrence and metastasis for 15 months. Although it was not possible to apply surgical treatment oriented to lung metastasis in our case, the efficiency of our treatment led us to believe that the prognosis and treatment of testis SmCCs may be applied in the case of GCTs.

Primary testiculary neuroendocrine neoplasms are frequently associated with a teratomatous component (1). Abbosh et al. observed Isochromosome 12 p on the carcinoid tumour tissue in all four cases of testicular carcinoid tumour arising in mature teratoma. This study shows that testicular carcinoid tumours associated with teratoma, a germ cell neoplasm that shares the same clonal origin as yolk sac, embryonal carcinoma and choriocarcinoma, are of germ cell origin (4).

The case of SmCC arising in a mature ovarian cystic teratoma reported by Lim et al. (3) also received six courses of combination chemotherapy with cisplatin/etoposide/bleomycin after surgical intervention for a huge ovarian mass. Recurrence was not reported in that case 34 months after primary diagnosis, suggesting that SmCC with a germ cell origin could be treated in the same way as GCT with a good prognosis.

Address for Correspondence: Dr. Esma Türkmen, Department of Medical Oncology, Trakya University Faculty of Medicine, Edirne, Turkey.
Phone: +90 543 406 79 96 e-mail: esma_a143@hotmail.com
Received: 27.09.2013 Accepted: 02.05.2014 • DOI: 10.5152/balkanmedj.2014.13086
Available at www.balkanmedicaljournal.org

Although we were not able to determine the histopathological nature of the residual tumour, given the favourable clinical prognosis of the patient we think that testicular SmCCs may be treated using the same principles as for GCTs.

**Ethics Committee Approval:** N/A.

**Informed Consent:** Informed consent was obtained from patient who participated in this case.

**Peer-review:** Externally peer-reviewed.

**Author contributions:** Concept - B.E.; Design - H.K.; Supervision - E.T.; Resource - S.U.; Materials - H.K.; Data Collection&/or Processing - B.E.; Analysis&/or Interpretation - S.U.; Literature Search - E.T.; Writing - E.T.; Critical Reviews - H. K.

**Conflict of Interest:** No conflict of interest was declared by the authors.

**Financial Disclosure:** The authors declared that this study has received no financial support.

## REFERENCES

1. Mazzucchelli R, Morichetti D, Beltran AL. Neuroendocrine tumours of the urinary system and male genital organs:clinical significance. *BJU Int* 2009;103:1464-70. **[CrossRef]**
2. Merino J, Zuluaga A, Gutierrez-Tejero F. Pure testicular carcinoid associated with intratubular germ cell neoplasia. *J Clin Pathol* 2005;58:1331-3. **[CrossRef]**
3. Lim S-C, Choi SJ, Sun CH. A case of small cell carcinoma arising in a mature cyctic teratoma of the ovary. *Pathol Int* 1998;48:834-39. **[CrossRef]**
4. Abbosh PH, Zhang S, Maclennan GT. Germ cell origin of testicular carcinoid tumors. *Clin Cancer Res* 2008;14:1393-96. **[CrossRef]**