CASE REPORT

A rare association of extragonadal seminoma with synchronous papillary carcinoma of the thyroid

Craig Barrington,¹ Gianfilippo Bertelli,² Rhodri Evans³¹Department of Medicine, ABMU, Swansea, UK²Department of Oncology, ABMU, Swansea, UK³Department of Radiology, ABMU, Swansea, UK**Correspondence to**Dr Craig Barrington,
craig.barrington@wales.nhs.uk

Accepted 19 February 2014

SUMMARY

A 45-year-old man presented with a large para-aortic retroperitoneal tumour, a smaller second mediastinal tumour and elevated lactate dehydrogenase (LDH). Biopsy established a diagnosis of extragonadal seminoma. Treatment with cisplatin and etoposide resulted in complete resolution of the mediastinal mass, reduction of the size of the retroperitoneal mass and normalisation of LDH. Postchemotherapy positron emission tomography (PET) scan showed a small residual focus of uptake in the retroperitoneal mass and an unexpected focus in the left side of the neck. This was initially thought to represent residual active disease, but an ultrasound (US) scan and US-guided core biopsy of a cervical lymph node demonstrated metastatic papillary thyroid cancer rather than seminoma. A small (1 cm) primary papillary tumour in the thyroid was identified subsequently. The patient received consolidation radiotherapy to the retroperitoneum and underwent total thyroidectomy and neck dissection followed by radioiodine treatment. He is currently in complete remission from both cancers.

BACKGROUND

Testicular and thyroid malignancies are rare with the age-standardised incidence rates of 6.9 and 1.7 per 100 000 population, respectively, in the UK.¹ The incidence of multiple primary neoplasms in patients with cancer is less frequent, occurring in 3.2–4.6% of all cases, while the synchronous appearance of two primary malignancies is even more unusual.² Developing one primary neoplasm gives an individual 1.29 times the relative risk of developing a secondary independent primary tumour compared with a person who has never had cancer.³ The 5-year survival rates of testicular and thyroid cancer in men in the UK is 97% and 74%, respectively, and improving.⁴

The testis is the most frequent site of presentation of malignant germ cell tumours with the retroperitoneum second and the mediastinum third most frequent.⁵ Second neoplasms are a well-recognised complication postradiotherapy and/or postchemotherapy,^{5–9} but we have only found one other documented case of a synchronous germ cell tumour and malignant thyroid tumour in the literature.⁹ There is, however, no documented case of an extragonadal germ cell tumour with a malignant thyroid tumour. We present a case of a patient with advanced extragonadal seminoma where the presence of a synchronous thyroid cancer caused uncertainty of the true extent of response to chemotherapy. An ultrasound (US)-guided core

biopsy was decisive in establishing that he did not need second-line chemotherapy and he eventually achieved complete remission of both primary neoplasms. The fact that the CT-positron emission tomography (PET) scan highlighted a potential metastasis within the neck when the primary disease was apparently responding well to treatment raised the suspicion that there was a potential alternative diagnosis. This case highlights two major teaching points:

- The importance of seeking a tissue diagnosis when there is clinical doubt as to whether or not there is a second primary tumour or metastasis from a second primary.
- The use of US-guided core biopsy to establish a histological diagnosis of metastatic disease from a second primary tumour.

CASE PRESENTATION

A 45-year-old white man presented with a 7-month history of lower back pain and weight loss of 20 kg. Clinical findings included a palpable abdominal mass and cachexia. CT scan showed a 12×18×16 cm retroperitoneal mass enveloping the aorta and a 3 cm posterior mediastinal mass. Serum α fetoprotein, carcinoembryonic antigen, CA19-9 and β -human chorionic gonadotropin were all normal but lactate dehydrogenase (LDH) was raised at 1116 U/L. Clinical and US examination of the testes did not reveal any abnormality. Biopsy of the retroperitoneal mass confirmed a poorly differentiated malignant tumour of germ cell origin: immunohistochemistry was suggestive of an extragonadal seminoma.

He was started on etoposide and cisplatin for four cycles, during which serum LDH normalised and his performance status returned to normal. Postchemotherapy CT scan showed reduction in the abdominal mass to 11.5 cm and complete response of the mediastinal mass. PET scan was obtained to assess whether there were still metabolically active areas in the para-aortic mass. The PET scan showed only a small residual area of uptake in the para-aortic region confirming complete response in the mediastinum. There was, however, an unexpected abnormal uptake in the left neck region. This was found on US scanning to correspond to a 1.4 cm left cervical lymph node, which in retrospect was present on the initial CT scan and had reduced in size during chemotherapy. US-guided fine needle aspirate (FNA) of the node showed malignant cells, and the initial interpretation of the findings was that this represented metastatic seminoma with incomplete response to first-line chemotherapy.

CrossMark

To cite: Barrington C, Bertelli G, Evans R. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-202073

The patient was started on second-line chemotherapy with the VIP regimen (vinblastine, ifosfamide and cisplatin) but after the first cycle results of a US-guided core biopsy of the lymph node became available. The biopsy showed H&E and immunohistochemical features completely different from the original retroperitoneal biopsy, suggesting the presence of metastatic papillary thyroid carcinoma. Further US scan of the neck identified an 8 mm hypoechoic mass containing microcalcification (typical of a papillary carcinoma) in the upper pole of the left thyroid lobe, corresponding to a synchronous primary papillary thyroid tumour. Chemotherapy was discontinued and the management plan was changed to consolidation radiotherapy to the retroperitoneum, total thyroidectomy and neck dissection followed by radio-iodine treatment.

OUTCOME AND FOLLOW-UP

The patient completed all treatments by February 2013 and is currently in complete remission for both cancers.

DISCUSSION

The literature describes cases of patients with second primary malignancies following treatment for germ cell tumour. Our case highlights the possibility of two primary malignancies occurring synchronously and poses the question as to whether there are any genetic or environmental risk factors for both. There are no known environmental risk factors currently known that may explain an association of germ cell cancer and thyroid cancer; however, genetic alterations at a molecular level in both have been described involving the oncogene K-RAS.^{10–13}

PET scans are useful to assess response to chemotherapy in patients with advanced seminoma, where residual radiological masses are often present on conventional imaging even when no viable malignancy has been left. Obtaining a baseline prechemotherapy PET scan is also appropriate when possible, but in our patient this was not done because of the need to start chemotherapy urgently. Our case illustrates the fact that one of the benefits of PET scans is to highlight the presence of areas of abnormal uptake, which would be missed on conventional radiology. While in the majority of cases these represent additional metastases from the original tumour, it is important to consider the possibility of other primary cancers. In our patient, a PET scan and FNA initially gave the misleading impression that he had chemotherapy-resistant seminoma, which has a poor prognosis. A US-guided core biopsy enabled the correct diagnosis of concomitant second primary thyroid carcinoma, which has now been treated with curative intent. This case highlights the importance of considering additional biopsies in patients with germ cell cancers when confronted with unusual localisations or areas that do not seem to behave consistently with the effects of chemotherapy on the rest of the disease.

Learning points

- ▶ Synchronous germ cell tumours and thyroid neoplasm are rare.
- ▶ Our case adds to the scant existing literature on this subject.
- ▶ Physicians who manage patients with germ cell tumours should consider a biopsy of unusual areas of uptake on a positron emission tomography scan as this may reveal a different pathology and alter management.
- ▶ Ultrasound-guided biopsy provides a rapid and safe technique for establishing a histological diagnosis in cervical lymphadenopathy.

Contributors GB is responsible for patient care. RE reviewed the staging of CT scans and performed the ultrasound-guided biopsies. CB worked with GB at the time the patient first presented.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Cancer Research UK. *UK cancer incidence statistics*. London, UK: Cancer research, 2009. <http://www.cancerresearchuk.org/cancer-info/cancerstats/incidence/> (accessed 4 Nov 2012).
- 2 Dieckmann KP, Loy V, Klän R. Seminoma testis and concomitant ipsilateral renal cell carcinoma. *Urol Int* 1988;43:49–52.
- 3 Schottenfeld O. Multiple primary cancers. In: *Cancer epidemiology and prevention*. Philadelphia: WB Saunders, 1982:1025–35.
- 4 Cancer Research UK. *UK cancer survival statistics*. London, UK: Cancer research, 2009. <http://www.cancerresearchuk.org/cancer-info/cancerstats/survival/> (accessed 4 Nov 2012).
- 5 Appetecchia M, Pucci E. A rare association between malignant mediastinal seminoma and other malignant neoplasms. *J Endocrinol Invest* 2002;25:373–6.
- 6 Travis LB, Curtis RE, Honkey BF. Second malignancies after testicular cancer. *J Clin Oncol* 1995;13:533–4.
- 7 Syed AA, Jones NA, Bliss RD, et al. Metachronous testicular teratoma, testicular seminoma and papillary thyroid carcinoma occurring in a single individual: a report of two unrelated cases. *Eur J Cancer Care* 2010;19:701–2.
- 8 Fung C, Fossa SD, Beard CJ, et al. Second malignant neoplasms in testicular cancer survivors. *J Natl Compr Canc Netw* 2012;10: 545–56.
- 9 Ayoub JP, Amato RJ, Chiu A, et al. Synchronous appearance of germ cell tumour and papillary carcinoma of the thyroid. *Am J Otolaryngol* 2000; 21:416–20.
- 10 Strohmer TG, Slamon DJ. Proto-oncogenes and tumour suppressor genes in human urological malignancies. *J Urol* 1994;151:1479–97.
- 11 Wang LC, Vass W, Gao CL, et al. Amplification and enhanced expression of the c-Ki-ras-2 proto-oncogene in human embryonal carcinomas. *Cancer Res* 1986;47:4192–8.
- 12 Fagin JA. Molecular genetics of human thyroid neoplasms. *Annu Rev Med* 1994;45:45–52.
- 13 Fukunaga FH, Yatini R. Geographic pathology of occult thyroid carcinomas. *Cancer* 1975;35:1095–9.

Copyright 2014 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.

BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow