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CASE STUDY

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Testicular non-seminomatous germ cell tumour presenting as a neck lump: a diagnostic and therapeutic challenge

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Abstract: Testicular tumours are one of the most prevalent cancers in young males. Teratoma is one type of testicular tumour, which carries a good prognosis if treated appropriately. We describe a case of a 37 year old man, diagnosed with testicular non-seminomatous germ cell tumour in 2005. He underwent left orchidectomy and radical chemotherapy with Bleomycin, Etoposide and Cisplatin. He had involved retroperitoneal lymph nodes at the time of diagnosis and underwent retroperitoneal lymphadenectomy in 2007. He made a good recovery but presented with a left neck lump in 2009, the appearance of which suggested differentiated teratoma on fine needle aspiration cytology. The neck lump was excised without any complications and histology confirmed the mass to be mature teratoma with no undifferentiated elements. He has remained disease free since then and remains under oncological surveillance, in keeping with current protocols.

Keywords: Testicular germ cell, Tumours, Neck lump, Metastasis, Surgery, Follow-up

Key learning points:

1. Following complete remission of testicular germ cell tumours, neck lumps can in fact be a late presentation of metastatic disease and should always be considered when a patient with a previous testicular tumour presents with a neck lump
2. Enlarged left supraclavicular lymph node (Virchow's node) may also represent metastatic testicular malignancy rather than the more common gastrointestinal or lung cancers
3. Surgery to excise the testicular germ cell neck metastasis is the standard of care
4. These patients need long term follow up

1. Introduction

Testicular tumours are not very common in the male population with only 1-2% of all cancers in males being testicular, however, it is the most common cancer in young males [1]. The incidence of this type of tumour varies greatly depending on where the patient is from[1]. Testicular germ cell tumours can be divided into two main types: seminomas and non-seminomatous germ cell tumours. Teratoma and embryonal carcinoma are both subtypes of non-seminomatous germ cell tumour and reassuringly are very treatable with good cure rates following surgery and chemotherapy [2].

The incidence of cervical metastasis in testicular germ cell tumours is about 5% [3]. Metastatic disease from the testis first involves the retroperitoneal lymph nodes, and then spreads via the thoracic duct to its emptying site near the junction of the left internal jugular and subclavian veins. Hence, the left supraclavicular region is one of the possible places where testicular germ cell tumours can metastasize [4]. Late relapses, defined as recurrence occurring more than two years after initial complete response to primary treatment, are uncommon but can occur in many anatomical sites and have been seen in the retroperitoneum, abdomen, pelvis, liver, mediastinum, scrotum, inguinal regions, adrenal gland, chest wall and buttocks [3, 5]. A detailed literature review reveals a few published articles on this rare occurrence [3, 6-9].

2. Aim

We aim to present our case report along with relevant literature summary to raise awareness about this condition.

3. Case presentation

A 37 year old Caucasian male presented with a few weeks history of a left neck lump without any associated red flag symptoms. His past medical history included a testicular non-seminomatous germ cell tumour (histologically, embryonal carcinoma) for which he had undergone a left Orchidectomy in December 2005. At the time of presentation, his retroperitoneal lymph nodes were involved by metastatic disease. He completed radical chemotherapy with Bleomycin, Etoposide and Cisplatin through to February 2006 and underwent retroperitoneal lymphadenectomy in December 2007; histologically, there was differentiated teratoma in the resected retroperitoneal specimen.

On examination, there was a 6x5 cm firm swelling in the left supraclavicular fossa abutting the posterior surface of the sternocleidomastoid muscle. It was non-tender on palpation and no other cervical lymphadenopathy was noted. Fine needle aspiration cytology (FNAC) raised the possibility of differentiated teratoma. All his tumour markers were within normal limits.

4. Investigations

The patient underwent routine blood tests and specific tumour markers were checked. The neck lump was subjected to FNAC and a staging CT scan was carried out confirming an isolated left lower neck swelling (Figure 1).

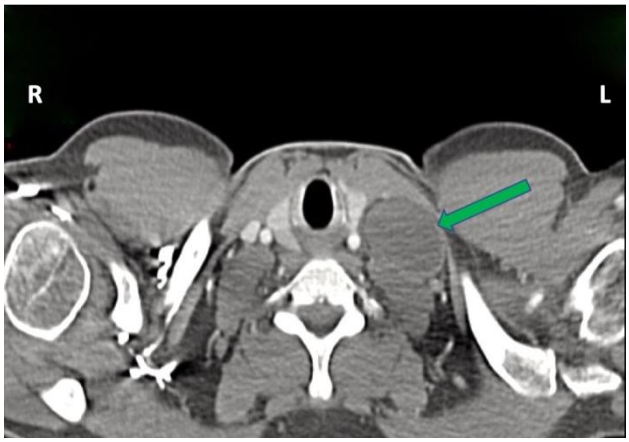

Figure 1 CT scan showing left neck lymphadenopathy (green arrow).

The pathology report indicated that much of the mass of differentiated teratoma appeared to be excised although there was not much surrounding normal tissue. Focally, inflammation in the wall of some of the cystic spaces extended to the peripheral margins. Histologically, the mass comprised a variety of differentiated elements, including fibroconnective tissue, fat, lymphoid tissue, cartilage and keratinising squamous epithelium which lined cystic spaces (Figures 2-5).

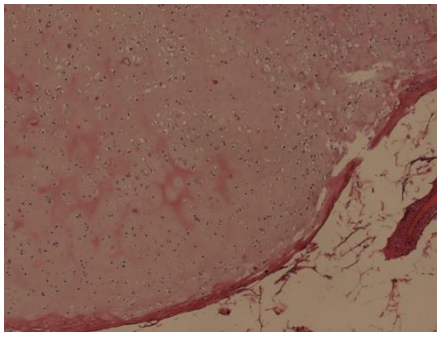

Figure 2 Histopathology of the resected neck mass showing cartilage

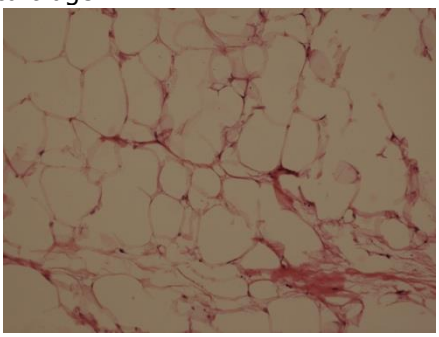

Figure 3 Histopathology of the resected neck mass showing mature fat.

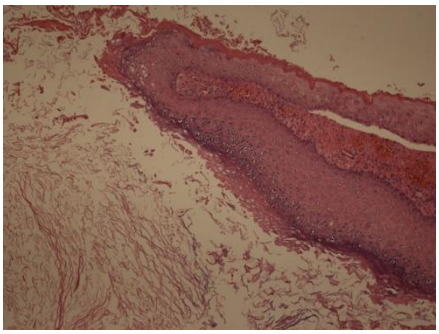

Figure 4 Histopathology of the resected neck mass showing keratinising squamous epithelium.

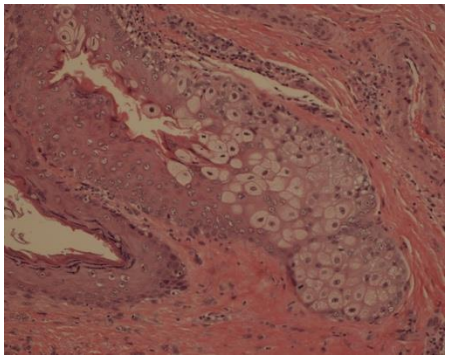

Figure 5 Histopathology of the resected neck mass showing a hair follicle.

Some of these cystic spaces were also partly lined by granulation tissue, chronic inflammation and foamy macrophages (Figure 6).

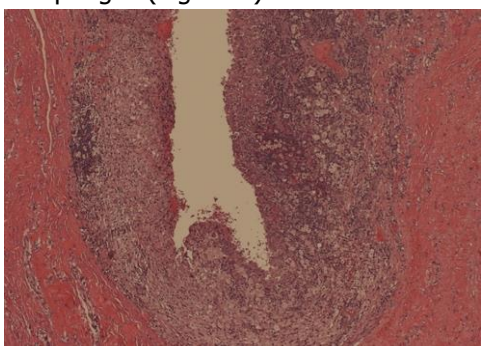

Figure 6 Histopathology of the resected neck mass showing an inflammatory reaction to cyst contents.

Other spaces were lined by cuboidal columnar epithelium with goblet cells. No undifferentiated elements were identified.

In the post-pubertal setting, differentiated teratoma is still regarded as malignant. There was no evidence in the neck mass of the embryonal carcinoma that constituted his original testicular tumour. Metastatic testicular non-seminomatous germ cell tumours are well recognised to have variable histological appearances that may differ from the original testicular tumour histology.

5. Differential diagnosis

Neck lumps are a common presentation in our head and neck clinics. The commonly encountered pathologies include reactive lymph nodes, branchial cleft cyst, thyroglossal duct cyst, thyroid nodule, salivary gland tumours, lymphoma and metastatic carcinomas. Testicular teratoma would not be the first thing a clinician would think of but with relevant history it should be considered as a potential diagnosis.

6. Treatment

The patient was admitted in September 2009 and the neck lump was excised under general anaesthetic successfully without any immediate complications. He developed a post-operative seroma that required needle aspiration in the clinic and oral antibiotics. He made a complete satisfactory recovery. The patient attended the head and neck clinic for his post-operative care.

7. Outcome and follow-up

The patient has continued to attend the oncology clinic where he was closely monitored for any disease recurrence. Fortunately, clinically, biochemically and radiologically he remained disease free. He remains under surveillance by oncology

8. Discussion

Testicular teratomas are a type of non-seminomatous germ cell tumour and contain tissues developing from endoderm, mesoderm and ectoderm. They most commonly present in the 25-35 age group [3]. Germ cell tumours account for 0.8% of all cancers in males and are the most common type of testicular neoplasm (95%) [10].

Cervical metastasis from testicular non-seminomatous germ cell tumours is extremely rare but can even occur as a first presentation of the testicular malignancy. Metastatic spread from non-seminomatous germ cell tumours usually starts in the retroperitoneal nodes, further spreading via the thoracic duct to the junction of the left internal jugular and subclavian veins. Relapses have been seen in anatomical sites including but not limited to the retroperitoneum, abdomen, pelvis, liver, mediastinum, scrotum, inguinal region, adrenal gland, chest wall and buttocks [5].

When a patient presents with a suspicious neck swelling then fine needle aspiration cytology (FNAC) is usually the first line investigation as happened in our case. Fine needle aspiration is the recommended diagnostic tool for head and neck cancers by SIGN and they state that imaging should be used for staging only [10].

Detailed literature review was carried out using the MESH terms teratoma, non-seminomatous germ cell tumour, chemotherapy, adjuvant, testicular neoplasms, incidence, tumour markers biological, cervical and neoplasm metastasis. There were only a few published articles identified confirming the rarity of this occurrence. Mustafa *et al.,* described the management of a 30 year old man who received surgery on his right testicle followed by chemotherapy. Four months later he developed involvement of his retroperitoneal nodes by metastatic teratoma successfully treated with laparoscopic lymphadenectomy. However, 2 months later he developed left supraclavicular lymphadenopathy suspicious for metastasis. He was treated with selective left neck dissection confirming the metastatic testicular teratoma [6]. Our patient, however, presented nearly 4 years after his orchidectomy with neck metastatsis supporting the recommendation that such patients need long term follow up.

Recurrence following resection and the chemotherapy regime used in our patient usually occurs within the first 2 years and so his late recurrence is considered rare [5]. However, the literature has shown that there are exceptions with one recurring after 17 years [3]. This shows that late recurrence is rare but possible.

A study by Michael et al found that teratoma was the most common type of neoplasm in late recurrence [5]. This study also found that late recurrence comprising only teratoma had a relatively good prognosis [5]. 1% of patients in a retrospective study of 96 patients with advanced germ-cell tumours, who relapsed after cisplatin-based chemotherapy, had an isolated supraclavicular node [9].

A 24 year old man in India was reported by Hemalatha *et al.,* [11] who presented with an enlarged left supraclavicular node (Virchow's node). He had undergone a high orchidectomy 6 months prior for a testicular tumour. FNAC of the neck node revealed a mixed germ cell tumour (teratoma, yolk sac tumour and embryonal carcinoma) [11]. In our patient, however, the neck lump occurred 4 years after the primary tumour, and due to this late recurrence further investigations were needed for full staging and to exclude any other source of malignancy.

Further cases of recurrence in the cervical region were reported by O'Connor, Dias and Timon, who mentioned a 17 year old patient with metastatic testicular carcinoma presenting with a palpable left neck mass [7]. Two cases of germ cell tumours presenting as a left supra-clavicular mass with no other metastasis were reported by N J Slevin, P D James and D A Morgan [8].

Tumour markers also play an important role in the diagnosis and monitoring of testicular non-seminomatous germ cell tumours. Alpha fetoprotein and beta human chorionic gonadotrophin are the most commonly employed tumour markers [12]. It is recommended that tumour markers (AFP and βHCG) be monitored regularly [9]. Our patient had regular monitoring of tumour markers along with physical examination and CT scans in the follow up period. However, it is important to note that pure testicular teratomas do not increase alpha fetoprotein and human chorionic gonadotrophin levels. Our patient did not have any raised tumour markers throughout his follow up.

As for the chemotherapy regime, our patient originally received chemotherapy using Bleomycin, Etoposide and Cisplatin (BEP) which is the recommended treatment regime for non-seminomatous testicular germ cell tumours [13]. Along with surgical resection, this treatment regime has a 90% success rate [6]**.** Therefore, he received the most appropriate treatment available at the time. It has been found in a study by Dearnaley *et al.,* that this chemotherapy regime is good for low risk cases but not for high risk cases [14]. The same study also found that 18% of patients fail to respond to BEP or develop recurrent disease after BEP [14]. For the neck lump, he received complete excision of the lump avoiding the traditional neck dissection. In the absence of widespread cervical lymphadenopathy in our patient, it was deemed appropriate to carry out targeted excision of the left neck node thereby avoiding the risks associated with neck dissection. The patient has been kept under close clinical, serological and radiological follow up since his neck surgery.

9. Conclusion

A patient presenting with a neck lump should be seen in a dedicated head and neck clinic. If the patient has a past medical history of treated cancer a high index of suspicion should be maintained to rule out metastatic disease. Although rare, as shown in our patient, following complete remission of testicular germ cell tumours, neck lumps can in fact be a late presentation of metastatic disease and should always be considered when a patient with a previous testicular tumour presents with a neck lump. Management in a multidisciplinary team setting is recommended and the long term prognosis remains good.

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Authors Contribution

Data collection, analysis, drafting and finalising the final manuscript (MH, LD, SKD & AC). Designing the study, data collection, analysis, drafting and finalising the final manuscript (MS).

Data sharing statement

No additional data are available

Ethics Approval

Approval was sought and granted by the Departmental Ethics Committee.

Informed consent

Informed written consent obtained from the patient

Conflict of interest

The authors declare no conflict of interest.

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