Testicular Choriocarcinoma: diagnosed on cervical lymph node biopsy

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Abstract
Choriocarcinoma is a very rare germinal testicular tumour and in literature its incidence has been reported to be 0.3% of all germinal testicular tumours. An important tumour marker is serum ß-hCG which not only helps in establishing diagnosis but also in assessing response to chemotherapy. In this study we present a case of testicular choriocarcinoma, who presented with abdominal pain, cough, generalized weakness and left sided cervical mass. Incisional biopsy of cervical mass was performed. Histopathology revealed metastatic choriocarcinoma. Serum ß-hCG levels were 1227 ng/mL. Patient received intravenous cycles of PEB (cisPlatin, Etoposide, Bleomycin) chemotherapy but he had progressive disease both radiologically and on tumour marker monitoring. He was planned for salvage chemotherapy but was lost to follow up there after. It is concluded that in males, choriocarcinoma carries a very dismal prognosis and a very poor response to chemotherapy and radiotherapy; surgery has no role in the management.

Keywords: Choriocarcinoma, Male, Serum ß-hCG, Chemotherapy, Prognosis.

Introduction
Choriocarcinoma in males is considered to be a highly malignant variant of teratoma. The testes are the most common primary site for this lesion in males, although primary tumours of the mediastinum, subcutaneous tissue, pancreas and retroperitoneal areas have also been reported. Like other germ cell tumours (GCTs), choriocarcinoma typically affects young men but unlike other germ cell tumours, patients of choriocarcinoma of the testes do not usually present with local symptoms and the prognosis of these patients is very dismal.

In most reports, the tumour responds poorly to radiation and chemotherapy and carries a high mortality rate. Surgery is usually limited to tissue diagnosis.1 Choriocarcinoma recapitulates placental tissue development. For unknown reasons, it metastasizes early via haematogenous routes to the lung, liver, and brain.1,2

In the literature review of 10,000 cases of testicular germ cell tumours, only 54 (0.5%) cases of pure choriocarcinoma were reported. The peak incidence was found in men between the ages of 20-30 years.3

Usually the initial presenting symptoms are from metastases, or symptoms from elevated levels of ß-hCG, although the patient may present with painful testicular swelling. Serum ß-hCG levels are usually markedly elevated in pure choriocarcinoma. Syncytiotrophoblastic cells are responsible for the production of ß-hCG. Serum ß-hCG levels are not only helpful in establishing the diagnosis but can also help in assessing response to treatment.

We describe an unusual case of metastatic choriocarcinoma in a young male diagnosed on cervical lymph node biopsy and the literature review of this rare disease.

Case report
A 27 year old male presented with 3 months history of generalized fatigue, weight loss, fever, cough and abdominal pain. Patient was non-smoker and non addict. There was no family history of any malignancy in the family. He had never received radiation to any part of body and he did not have any known congenital chromosomal abnormality or mumps in childhood. Patient was completely oriented in time, space and person with good performance status (ECOG-0).

He had received symptomatic treatment from different general practitioners during the course of his illness but his symptoms did not improve. Patient sought advice from a surgeon and a CT scan of abdomen was done that showed massive abdominal lymph adenopathy. Meanwhile he developed left sided cervical lymphadenopathy along with severe backache. His respiratory symptoms also aggravated with time.

Cervical lymph node was biopsied and specimen was sent for histopathology. It was reported as "metastatic malignant neoplasm suggestive of Choriocarcinoma".
Immunohistochemical stain for ß-hCG was done on histopathology specimen which was equivocal.

Patient was referred to an oncologist for further management. After detailed history and physical examination, his metastatic workup was done. On physical examination, he had hard slightly enlarged asymptomatic left testes, left cervical lymphadeonopathy, hepatosplenomagaly, several palpable masses within the abdomen and scrotal swelling. His haematological and biochemical profiles were within normal limits. His chest radiograph showed multiple coin shaped lesions which were suggestive of metastasis. Abdominal and pelvic ultrasonography also showed para aortic lymphadenopathy. Bone scan was suggestive of metastatic disease in spine. His serum ß-hCG levels were 1227ng/mL.

Patient was given IV cycles of cisPlatin, Etoposide, Bleomycin (PEB) chemotherapy but he had progressive disease clinically and radiologically (Figure) with persistently elevated tumour markers. The patient was planned for salvage chemotherapy but was lost to follow up there after.

**Discussion**

Non-seminomatous germ cell tumours are highly aggressive malignant tumours of the testes. Most of the patients present with metastatic disease. Histologically, approximately 70% of the non-seminomatous germ cell tumours are composed of more than one germ cell components (mixed germ cell tumours). These various components include embryonal carcinoma, seminoma, yolk sac tumour and teratoma; choriocarcinoma is relatively rare. Less than 8% of the testicular germ cell tumours contain a component of choriocarcinoma, and pure choriocarcinoma accounts for only 0.3% of the primary testicular germ cell tumours.

The common presentation of testicular tumours is testicular swelling along with systemic symptoms, if metastatic. As choriocarcinoma in males has a very high propensity to spread haematogenously, it usually presents with systemic symptoms, like haemoptysis in case of respiratory involvement, abdominal pain and haematemesis in GI involvement and bone pains in case of skeletal metastasis. Our case was exceedingly unusual with cervical lymph node enlargement as initial presentation.

The diagnosis in this rare case was very difficult and involved thorough clinical evaluation, serum ß-hCG levels and most importantly histopathological examination of
biopsy specimen. Choriocarcinoma in males is an extremely aggressive disease with very dismal outcome and poor prognosis. Its syncytiotrophoblastic cells contain plasma chorionic gonadotrophin (ß-hCG), which is used as a tumour marker for diagnosis, grading and evaluation of response to treatment.

In most cases these tumours respond poorly to radiotherapy and chemotherapy and carry a high mortality rate. Triple drug chemotherapy gives an average survival of 3-4 months, the longest survival on record being 17 months in case of mediastinal choriocarcinoma. Surgery is usually limited to tissue diagnosis.

Standard chemotherapy for poor-risk and some good-to-moderate risk patients include IV cycles of BEP (i.e., bleomycin, etoposide, cisplatin). Other chemotherapeutic agents which have shown some activity in first line treatment or as salvage therapy in these cases include vinblastine and ifosfamide.

These tumours, although rare should be considered in the differential diagnosis of cervical masses.

References