Burned Out Seminomatous Testicular Tumor with Retroperitoneal Lymph Node Metastasis: A Case Report

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Abstract Germ cell tumor is the most common malignancy in men aged 15–35 years. About five percent of the malignant germ cell tumors are extragonadal in origin. However metastatic deposit of seminoma in retroperitoneal lymph node without presence of any tumor in the testes is uncommon. We present a case of metastatic retroperitoneal seminoma as the initial presentation of a burned out testicular primary. The patient presented with pain in abdomen and back. Ultrasonography of the testis showed a lesion, which after high inguinal orchidectomy was reported as fibrosis only, without any tumor, i.e. burned out phenomenon. A laparotomy biopsy of the retroperitoneal lymph node mass was taken and reported as metastatic seminoma after immunohistochemistry. After 14 months of chemotherapy there is a residual lymph node of less than 3cm, serum tumor markers are normal and the patient is under observation.

Keywords Seminoma · Burned out phenomenon · Testis · Extragonadal germ cell tumor

Case Report

A 33 year old Indian male presented to our Institute with pain in abdomen and back intermittently, for 3 months duration. Patient is a tobacco chewer and hypothyroid under treatment. There was no significant history, physical examination was normal.

Ultrasonography (USG) of abdomen revealed a heterogeneous mass in the retroperitoneum. Contrast enhanced computed tomography (CECT) of abdomen showed a rim enhancing lesion 6.1×5.0 cm at aortic bifurcation [Fig. 1a]. Trucut biopsy was done and histopathological examination (HPE) reported as poorly differentiated carcinoma or Non-Hodgkin's lymphoma. Immunohistochemistry (IHC) showed CD117 positivity in the tumor cells, suggestive of seminoma. The tumor cells were negative for pancytokeratin, CD30 and CD45.

To find out the primary, USG of bilateral testes was done, which showed a heterogenous lesion of 1.5×1.2 cm in the left testicle [Fig. 1b]. Testicular tumor serum markers human chorionic gonadotropin (hCG), alphafetoprotein (AFP) and lactate dehydrogenase (LDH) were normal.

Left high inguinal orchidectomy was done. On gross, a whitish scar like lesion was noted [Fig. 1c]; microscopically no tumor was found; only fibrosis was seen. Re-grossing and re-examination of the specimen was reported as same.

Laparotomy and biopsy from the retroperitoneal lymph nodal mass was done. HPE showed neoplastic cells in sheets and nests with intervening thin fibrous septae and scattered lymphocytes. The cells had vesicular nuclei, prominent nucleoli and pale cytoplasm [Fig. 1d], suggestive of (1) metastatic/primary seminoma, (2) metastatic/primary poorly differentiated carcinoma. No definite lymph nodal architecture could be

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Fig 1 **a** CECT of the Abdomen showing a rim enhancing lesion, **b** the lesion in left testicle, **c** cut section of the orchidectomy specimen, **d** microscopic view (40x) of classical seminoma, inset—tumor cells showing CD 117 positivity

identified. The diagnosis of seminoma was confirmed with IHC, whose results were same as before. Final diagnosis was metastatic seminoma with ‘burned out’ primary testicular tumor.

Patient received three cycles of chemotherapy with Bleomycin, Etoposide and Cisplatin (BEP). After 14 months of follow-up, patient is asymptomatic, serum tumor markers are normal; there is a solitary lymph node of 2.2 cm x 2.1 cm below the aortic bifurcation. He is under observation.

Discussion

Germ cell tumor is the most common malignancy in men aged 15–35 years. Retroperitoneal seminoma without involvement of the testis is uncommon [1–3]. Two to five percent of the malignant germ cell tumors (GCT) are extragonadal in origin [1, 4].

Extragenadal germ cell tumor (EGGCT) is by definition a germ cell neoplasm displaying one of the histology associated with gonadal origin, but located outside of the gonads [4].

Although there is no clear explanation regarding the histogenesis of EGGCTs, two embryologic theories have been proposed. The first most accepted theory posits a failure of some primordial germ cells to complete their migration from the urogenital ridge to scrotum. The second theory proposes there are rests of totipotent cells left

behind during embryonic development, which become neoplastic [5].

EGGCT are typically found in the midline, the most common location being the mediastinum and retroperitoneum. [4, 6].

The burned out phenomenon in GCTs refers to a GCT in extragonadal tissue with spontaneous regression of an intragonadal tumor [2, 7, 8]. It presents by metastases to the retroperitoneum, mediastinum, lymph nodes, lungs and liver [3]. The mechanism for the regression of a primary gonadal tumor is unclear, but it has been suggested that the regression results from immunologic or ischemic change, as reported in patients with renal cell carcinoma, breast cancer, lymphoma, and malignant melanoma [2, 7]. One hypothesis suggests that common tumor antigens can be recognized after repeated exposure, by cytotoxic T lymphocytes, which are subsequently replaced by fibrosis [2].

Cytogenetically one or multiple copies of short arm of chromosome 12p with the loss of long arm of chromosome 12 is seen in nearly all germ cell cancers of primary gonadal and extragonadal origin [4].

FNAC or histology through percutaneous or open biopsy may be reported as seminomatous, non-seminomatous malignancy or only ‘undifferentiated carcinoma’ by the pathologist. Serum markers and IHC in the tissue confirm the diagnosis in almost all cases [4]. To establish a diagnosis of regressed testicular GCT certain features are helpful, which include scar formation, intratubular calcifications,

lymphoplasmacytic infiltrate, hemosiderin-laden macrophages and testicular atrophy [2].

The burned out tumors of the testis need to be distinguished from true EGGCT because the primary removal of the testicular tumor leads to a satisfactory outcome. These malignancies are treated surgically with or without adjuvant chemotherapy or radiotherapy [1, 2]. Seminomatous EGGCT have an equivalent prognosis as their primary gonadal counterpart [1, 4].

The diagnosis and treatment of these cases may be delayed due to nonspecific initial presentation and regression of the primary lesion. Hence awareness amongst clinicians is necessary for proper management and patient survival.

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