Case Report

Burned-out Testicular Germ Cell Tumor Mimicking Lymphoma

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Kuwait Medical Journal 2006, 38 (3): 235-237

ABSTRACT

Burned-out germ cell tumor of the testis is a rare entity which, with its metastatic spread often mimics other conditions such as lymphoma. Scrotal sonography is pivotal in the initial diagnosis of such a neoplasm with extragonadal metastases.

KEYWORDS: burned-out tumor, germ cell tumor, sonography, testis

INTRODUCTION

Germ cell tumors constitute the large majority of all testicular tumors, which overall are relatively rare, and are mainly encountered in young adults and teenagers[1]. The "burned-out" form of germ cell tumor refers to the situation in which widespread metastases are on the clinical frontline whereas the primary neoplasm has involuted[2]. Its mode of presentation may simulate other neoplasms[3,4]. We report such an example for which a systematic scrotal sonography rectified the initial erroneous diagnosis of lymphoma.

Case Report

A 17-year-old boy was transferred to our institution for staging / work-up of Hodgkin's lymphoma. He presented one month earlier to his physician with complaints of right-sided abdominal pain, back pain, night sweats, fever, fatigue, anorexia and weight loss of eight pounds over one month. A sonographic examination at the local hospital revealed retroperitoneal lymphadenopathy. There was no significant past medical history apart from smoking since twelve years of age.

On clinical examination, he was found to be short of breath, with inspiratory and expiratory wheezing, extensive non-tender cervical lymphadenopathy and hepatosplenomegaly. Examination of the scrotum revealed no abnormalities. The WBC count was 14.3 x 10^9/l (normal: 4.5-13.0 x 10^9/l), with 11% neutrophils and 86% monocytes. ESR was 37 mm/hr (normal: 3 -13). Electrolytes were normal. A bone marrow aspirate showed no evidence of malignant cells. Abdominal sonography showed enlarged prevertebral soft tissue masses (Fig. 1). CT of the chest / abdomen / pelvis showed extensive left supraclavicular, mediastinal lymph node enlargement. Retroperitoneal lymphadenopathy involved the inter-aortocaval chain and the left para aortic nodes, some of which had a hypodense centre (Fig. 2).

As part of the search for a primary malignancy a scrotal sonogram was done and it demonstrated no testicular enlargement, but a focus of coarse calcification (0.3 cm) with no associated obvious mass in the left testis (Fig. 3). The clinical working diagnosis was burned-out testicular primary tumor with metastatic spread. Bone scan was normal and the Lactate Dehydrogenase (LDH) level was elevated (1,089 IU/l; normal: 105-333 IU/l). A Gallium scan showed focal increased uptake at the left supraclavicular area (Fig. 4). The serum alpha-fetoprotein level was normal but the beta HCG level was elevated (293 IU/l; normal: 0-4 IU/l).

At surgery, a radical left orchidectomy was performed along with a left cervical lymph node biopsy. Testicular histology specimen revealed diffuse intratubular germ cell neoplasia including focal intratubular seminoma and focal extratubular extension, foci of mature teratoma components as well as foci of regressed (burned-out) germ cell lesion with fibrosis and dystrophic calcification. The left cervical lymph node showed a metastatic malignant mixed germ cell tumor with predominant embryonal cell carcinoma and focal choriocarcinoma. The therapy of this stage three malignant mixed germ cell tumor included a left orchidectomy and a

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Fig. 1: Abdominal sonography showing a cluster of enlarged lymph nodes in the prevertebral region (between cursors)

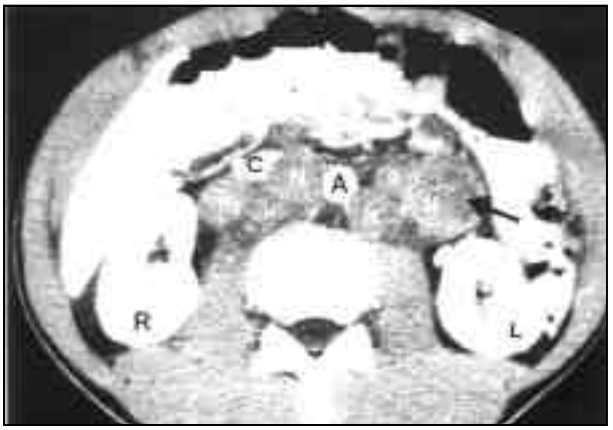

Fig. 2: Abdominal CT Scan at the lumbar level demonstrating lymphadenopathy (N) adjacent to the major vessels, with central hypodensity in some lymph nodes (arrow). (A = Aorta, C = Cava or inferior vena cava, L= Left, R = Right)

Fig 3: Left scrotum sonography: focal calcification (boxed) with acoustic shadowing in the lower pole of the left testicle

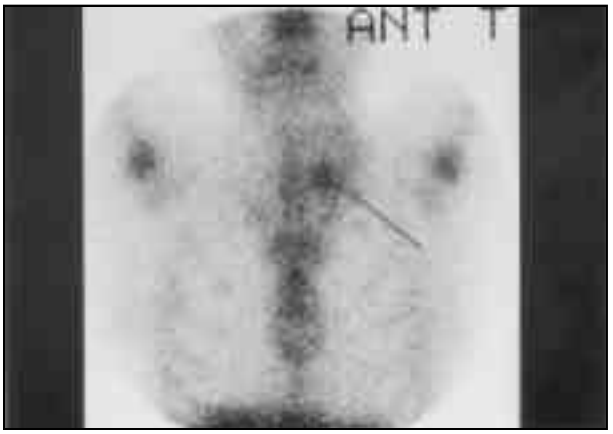

Fig 4: Gallium scan showing increased uptake at the left supraclavicular area (arrow)

chemotherapy regimen of bleomycin, etoposide, amifostine and cisplatin followed by extensive intra-abdominal lymph node resection. The histology specimen of the latter showed similar pathology to the specimen of the left cervical lymph node.

DISCUSSION

Clinical presentation of a burned-out tumor of the testis is often due to its metastatic spread to cervical, axillary, supraclavicular lymph nodes as well as to the mediastinum and retroperitoneum. CT and MRI are optimal for assessing the extent of the disease. The clinical impression is of a process such as lymphoma or even a rhabdomyosarcoma, besides the possibility of germ cell tumor. Extragonadal germ cell tumors are only about 5% of all germ cell tumors[5]. Metastases from a gonadal primary tumor are highly likely. It is known that palpation of the testicles is not very sensitive at detecting small testicular masses. The burned-out tumor is hence an occult gonadal germ cell neoplasm with primary metastases. Two pathologic types are described. The first variety includes testicular atrophy, and small intratesticular scars with no histologic sign of neoplasm. The scar has, however, some stigmata of neoplastic origin such as the presence of hematoxyphilic bodies, hemosiderin pigmentation and calcium phosphate deposits[6]. The second form is the *in situ* testicular tumor with intratubular localization of the neoplasm. Chemotherapy may not be as effective because of the blood-testis barrier, which is incriminated in the possible recurrence of local tumor[5] or the metachronous contralateral involvement[4]. The non-seminoma component has the highest risk of regression of the primary neoplasm[2]. The mechanism of regression of the tumor is yet to be elucidated. However, immune response, versus ischemia mechanism due to a high metabolic rate of the neoplasm outgrowing its blood supply has been implicated[7].

High resolution sonography of the scrotum with linear high frequency transducers allows the detection of small highly echogenic foci, hypoechoic zones, microlithiasis or macrocalcifications as in our patient[3-5,8]. The hyperechogenic foci are related to non-tumoral lesions such as hemorrhage,

infarction or fibrotic scar. The slightly echogenic areas may correspond to the tumor itself. Testicular microlithiasis has been associated with testicular germ cell neoplasm or intratubular germ cell neoplasia but the clinical significance and malignant potential of microlithiasis are still under investigation[9].

Multiple histologic types are often found together, as a mixed germ cell tumor, because besides the seminoma component, totipotential cells can develop along several pathways, either in undifferentiated embryonal carcinoma or in differentiated embryonic teratoma or extraembryonic choriocarcinoma. Metastatic spread pathway is mainly lymphatic except for choriocarcinoma which favors a hematogenous pattern. This explains why histologic characteristics of metastases can be different from features of the primary neoplasm[1,7].

In summary, in the work up of a male patient presenting with widespread lymphadenopathy, it is mandatory to perform a testicular ultrasound examination, despite a normal physical examination of the scrotum. The sonographic detection of a primary testicular tumor is crucial for the therapeutic and prognostic parameters.

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