

Extremely rare presentation of burned-out testicular tumor

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ISSN: 1658-3639

PUBLISHER: Qassim University

ABSTRACT

It is commonly accepted to consider retroperitoneal germ cell tumors as the metastasis of a viable or burned-out testicular tumor. In such cases, orchiectomy should be performed since the burned-out site in the testis could continue to harbor malignancy despite systemic chemotherapy. A 45-year-old male presented as an outpatient with complaints of back pain. He was diagnosed with a retroperitoneal mass and a palpable testis mass. He underwent radical orchiectomy. Pathological study revealed a burned-out tumor. Following chemotherapy, retroperitoneal lymph node dissection was performed. Despite the residual and enhanced retroperitoneal lymph nodes in his computed tomography scan, none of the resected lymph nodes showed a viable tumor. This is the first reported case of a palpable and hypochoic burned-out testicular tumor.

Keywords: Burned-out, chemotherapy, lymphadenectomy, testis, tumor

Introduction

In case of diagnosing a retroperitoneal midline mass in a male patient, germ cell tumors (GCT) should be taken into consideration, and due to their rare primary nature, further investigation aimed at finding other primary sites should be performed.^[1-3] Systemic chemotherapy is the most widely used treatment for extragonadal tumors.^[2,3] Burned-out testicular mass or Azzopardi tumor is a known condition which is accompanied by retroperitoneal metastasis, yet the primary tumor regresses, and only small foci of calcification remain as signs of its presence.^[4]

Orchiectomy is the treatment of choice in such cases since the burned-out site in the testis could continue to harbor malignancy despite systemic chemotherapy.^[2,3] Herein, we report a middle-aged man who was diagnosed with a retroperitoneal mass and had evidence of a burned-out tumor in ultrasound study. He responded to chemotherapy partially and underwent retroperitoneal lymph node dissection (RPLND).

Case Report

A 45-year-old male, married with two children, a computer engineer, 40-pack year smoker, presented to our clinic as an outpatient with complaints of back pain over the previous 3 months.

Physical examination revealed a small firm area in the upper pole of the right testis.

In ultrasound study, a 107 mm * 80 mm exophytic mass was diagnosed neighboring the middle part of the right kidney, proximal to the right ureter with compression effects on the right pyelocalyceal system, causing mild right hydronephrosis. Scrotal ultrasound study revealed a few hypochoic masses in the right testis with a maximum size of 15 mm * 8 mm.

In laboratory study for tumor markers, β -human chorionic gonadotropin (β HCG) level was 510 (normal range in non-pregnant women <20), lactic dehydrogenase (LDH) was 1248 (normal range <480), and α -fetoprotein (AFP) was 0.1 (normal range <10).

In the next step, pre-operative spiral abdominopelvic computed tomography (CT) scan with and without intravenous contrast (IVC) was performed. CT showed a 100 mm * 80 mm enhancing mass between the aorta and the right kidney with compression effect on the right kidney. Moreover, thrombosis inside the IVC, below the mass and the perinephric lymph node was also reported [Figure 1].

He underwent radical orchiectomy and right DJ insertion. The pathological study showed two foci of scar tissue with no evidence of viable tumoral cells (Burned-out tumor/Azzopardi tumor): Intratubular germ cell neoplasia was not identified. One of the scar tissues was indicative of a rete testis.

Tumor markers were rechecked after surgery showing the following results: LDH: 1180, β HCG: 277, and AFP: 0.6. Chest CT scan was also performed which was normal.

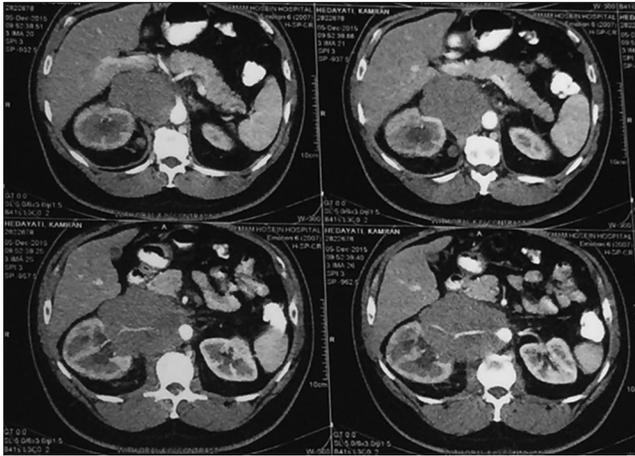


Figure 1: Computed tomography scan images before chemotherapy show thrombosis inside the intravenous contrast, below the mass and perinephric lymph node

Chemotherapy was initiated and 4 cycles of the bleomycin, etoposide, cisplatin regimen, 6 sessions at each cycle, was administered. By the end of chemotherapy, the abdominopelvic CT scan with IVC showed a 58 cm * 40 mm mass with enhancement, IVC invasion and compression effect on the right renal artery and right renal pelvis. CT also showed thrombosis inside the IVC which had extended to both the common iliac veins.

At the end of the chemotherapy course, tumor markers returned to normal; β HCG: 0.9, AFP: 3.1, and LDH: 327. On the ninth admission day, he underwent RPLND at our center. During surgery, multiple adhesive lymph nodes were dissected from the paraaortic and paracaval areas. The pathological study reported extensive necrosis with no evidence of malignancy.

The retroperitoneal fluid was also sent for cytologic examination, and the result was negative for malignant cells. Today, the patient is well and healthy living with no complaints or complications.

We have the ethical approval of Hasheminejad Hospital Research Deputy and patient's consent form in this study.

Discussion

When a retroperitoneal mass is diagnosed in adults or adolescents, one should always consider GCT as an important differential diagnosis, especially in the absence of an imaging clue for its origin,^[3-5] to avoid misdiagnosis and to apply the best treatment schedule for the patient.

However, in the presence of elevated tumor markers such as β HCG and AFP, treatment by chemotherapy can be initiated,

and no further evaluation is warranted, as the diagnosis of extragonadal GCT is definite.^[6] Furthermore, in the presence of ultrasound evidence of a burned-out tumor or when the pattern of lymph node involvement is consistent with a right or left side testicular tumor, inguinal orchiectomy should be performed during the treatment course.^[2]

Nevertheless, our case had a different presentation both at initial diagnosis and during the follow-up after chemotherapy. To the authors' knowledge, the presence of a palpable and hypoechoic burned-out tumor has not been mentioned in previous case reports. This finding could be a sign of a previous huge tumor which was burned-out, leaving an area of hypoechoic lesion with a diameter of 2 cm. It shows that irrespective of the size of the primary tumor, in case of a metastatic burned-out mass, the original tumor cells will regress and die. This once again brings up the hypothesis that these kinds of testicular masses may be associated with different genetic abnormalities which can lead to their different behaviors. Further investigation in this field might lead to the better understanding of the biology of such tumor cells besides enlightening the path to novel treatment options.

Another interesting aspect of our case was related to the imaging findings after chemotherapy and the surgical pathology report. Although CT scan with IVC is not the standard imaging for evaluating post-chemotherapy activity of retroperitoneal lymph nodes, enhancement of such nodes could be suggestive of tumor viability. As we described, the pathologic report of the resected lymph node revealed a necrotic and nonviable tumor. Our explanation for this finding is based on the fact that previous neovascularization in the retroperitoneum may have remained intact after chemotherapy while the tumor cells all died during this treatment, resulting in the enhanced tumoral residue.

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