Burned-out testicular tumor with pulmonary and retroperitoneal metastases: a case report

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ABSTRACT

The term “burned-out” in reference to the testes refers to complete and spontaneous regression of a testicular tumor to fibrous tissue that is in a metastatic state. Described for the first time in 1927, there are several small series and isolated cases reported in the literature. A clinical case of burned-out testicular tumor is presented here. Patient is a 19-year-old male who presented with chest pain. Cardiopulmonary and genital examinations were unremarkable. Alpha-fetoprotein, human chorionic gonadotropin, and lactate dehydrogenase levels were elevated. Testicular ultrasound revealed intraparenchymatous calcifications in the left testis, chest X-ray and computed tomography showed pulmonary metastatic lesions, and abdominopelvic computed tomography revealed retroperitoneal tumor activity. Retroperitoneal lesion was biopsied, patient received four bleomycin, etoposide, and platinum chemotherapy cycles, and left radical orchiectomy was performed. Histopathological biopsy study reported mixed non-seminomatous germ cell tumor (choriocarcinoma 33.3%, endodermal sinus tumor 33.3%, and embryonic carcinoma 33.3%). Histopathological study of left radical

RESUMEN

El término tumor “burned out” o quemado de testículo, define la regresión espontánea y completa de un tumor testicular a tejido fibroso, encontrándose el mismo en estadio metastásico. Descripto por primera vez en 1927, existen algunas series cortas y casos aislados publicados en la bibliografía médica. Se presenta un caso clínico inherente a este tema. Se trata de un varón de 19 años de edad, que presentaba dolor torácico. La exploración cardiopulmonar y genital sin relevancia. Alpha feto proteína, gonadotrofina coriónica humana fracción beta y deshidrogenasa láctica elevadas; ultrasonido testicular con calcificaciones intraparenquimatosas en el testículo izquierdo; teleradiografía de tórax y TC de tórax con lesiones metastásicas pulmonares; TC abdomino-pélvica con actividad tumoral retroperitoneal. Se tomó biopsia de lesión retroperitoneal, se proporcionaron cuatro ciclos de BEP y se realizó orquiectomía radical izquierda. El estudio histopatológico de la biopsia retroperitoneal informó sobre un tumor germinal no seminomatoso, de componente mixto (coriocarcinoma 33.3%, tumor de senos endodérmicos 33.3% y carcinoma embrionario 33.3%); el estudio histopatológico de la
pieza derivada de la orquiectomía radical izquierda, no mostró actividad tumoral. En el seguimiento ulterior, los marcadores tumorales fueron negativos, sin progresión de lesiones metastásicas y ECOG 0.

Esta entidad se presenta usualmente con síntomas secundarios a la disseminación metastásica. Ante un paciente con adenopatías retroperitoneales y examen físico testicular normal, debe sospecharse un tumor “burned out” hasta demostrar lo contrario.

La quimioterapia previa a la orquiectomía radical, es el tratamiento más efectivo para esta patología.

Palabras clave: Tumor testicular quemado, metástasis retroperitoneal, orquiectomía, México.

INTRODUCTION

It is difficult to differentiate between primary retroperitoneal germ cell tumor and metastatic disease from an undetected gonadal tumor or a tumor that has spontaneously regressed. As a matter of fact, there are authors that doubt the existence of true primary retroperitoneal germ cell tumors. However, the distinction between the two tumor origins is thought to be fundamental since it involves important differences in treatment as well as in prognosis. In 1927 Prim reported the presence of testicular scarring in an autopsied patient with retroperitoneal tumor. Various other cases were reported in the years that followed. Azzopardi et al stated that some testes that were normal when palpated presented with scar tissue or small tumor foci in the histological report. The question arose as to whether some primary extragonadal germ cell tumors could be metastatic. In 1951, Friedman observed regressive changes or evident testicular tumor in 23 of 29 patients presenting with primary retroperitoneal germ cell tumor.

A clinical case is presented here of a burned-out tumor, that is to say, a non-seminomatous retroperitoneal tumor of testicular origin, despite the fact that no tumor was found in the anatomopathological study of the gonads but alterations were found in ultrasound (US) and anatomopathological studies of the testis.

CASE PRESENTATION

Patient is a 19-year-old male. He underwent appendectomy at 13 years of age. He was evaluated for clinical symptoms of 2-month progression of chest pain predominantly in the left hemithorax and 4 kg weight loss. Physical examination showed patient to be thin with no cardiopulmonary or genital alterations. Chest X-ray revealed multiple pulmonary nodules (Figure 1). Pulmonary and abdominopelvic computed tomography (CT) showed metastatic pulmonary lesions and retroperitoneal tumor activity (Figures 2, 3 and 4). Tumor marker results reported alpha-fetoprotein >35000 ng/mL, beta fraction human chorionic gonadotropin 5105.30 mIU/mL and lactate dehydrogenase 2517 IU/mL, and testicular US showed intraparenchymatous calcifications in left testis (Figure 5). Retroperitoneal lesion was biopsied and histopathological report stated mixed germ cell tumor (embryonic carcinoma 33.3%, endodermic sinus tumor 33.3%, and choriocarcinoma 33.3%) and histopathological study of testicular biopsy reported microcalcifications and stromal fibrosis of left testis. Chemotherapy was initiated with 4 cycles of bleomycin, etoposid, and cisplatin (BEP regimen) after which left radical orchiectomy was performed. Histopathological study of surgical specimen reported no tumor activity.

Presently patient is at 6-month postoperative follow-up. His clinical performance status is ECOG.
Table 1. ECOG performance status.6

<table>
<thead>
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<th>Description</th>
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<tr>
<td>0</td>
<td>Asymptomatic and completely active</td>
</tr>
<tr>
<td>1</td>
<td>Symptomatic but ambulatory</td>
</tr>
<tr>
<td>2</td>
<td>Confined to bed less than 50% of waking hours (less than 8 hours/day)</td>
</tr>
<tr>
<td>3</td>
<td>Confined to bed or chair more than 50% of waking hours (more than 8hr/day)</td>
</tr>
<tr>
<td>4</td>
<td>Confined to bed or chair 100% of the time</td>
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<tr>
<td>5</td>
<td>Dead</td>
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Recent reviews have concluded that so-called primary extragonadal tumors are very rare or practically non-existent and that the majority of these cases are actually cases of metastases of viable or burned-out testicular tumors, given that 76% of patients present with pathological findings in testicular biopsy.

BEP regimen treatment is the criterion standard in advanced disseminated germ cell tumor management worldwide.8,9 Over the last few years multiple therapeutic regimens have been utilized based on combinations of cisplatin. The most common are cisplatin, vinblastine and bleomycin (PVB) and cisplatin, etoposide and

**DISCUSSION**

This entity usually presents with symptoms that are secondary to metastatic dissemination. Burned-out testicular tumor should be suspected in all men presenting with retroperitoneal adenopathy in whom physical examination of the testes is normal.7

**Figure 1.** Chest X-ray showing multiple heterogeneous radiopaque images (cannon balls).

**Figure 2.** Contrasted abdominopelvic tomography showing normal kidney function, extensive para-aortic and paracaval lymph node conglomerate, along with retroperitoneum with large sized metastasis and homogeneous liver.

**Figure 3.** Abdominopelvic CT view showing large heterogeneously dense image on the right side of the pelvic cavity.
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CONCLUSIONS
Primary retroperitoneal germ cell tumors are very rare and many authors even doubt their existence. When there is retroperitoneal tumor, testicular alterations seen by means of ultrasound despite normal testicular palpation, burned-out syndrome should be considered. Bilateral testicular biopsy is obligatory in patients presenting with retroperitoneal germ cell tumor since there are pathological findings in nearly 100% of cases and viable tumor or scarring in 76% of cases.

The possibility that there can still be viable tumor in the testes after chemotherapy (sanctuaries) has been clearly demonstrated, making surgical exploration, and even orchiectomy, options to be taken into consideration when evaluating each patient.

ifosfamide (PEI). These regimens are therapeutically equivalent and vary only in their levels of toxicity. 8,9

Figure 4. Chest CT with view of the lung showing multiple hyperdense nodular images, homogeneous in both pulmonary fields, compatible with metastatic lesions.

Figure 5. Right testicular US showing two homogeneous hypechoic images.