Bilateral Wunderlich syndrome secondary to synchronous bilateral testicular germ cell tumor. A case report

Salgado Sánchez Luis Eduardo a,*, Martínez Garfías Arturo Enrique a, Juárez Ávila Joel a, Martínez Cornelio Andrés b, Ramírez González Laura Denisse b

a Department of Urology, Oncology Hospital, Mexican Institute of Social Security, Mexico City, Mexico
b Department of Pathology, Oncology Hospital, Mexican Institute of Social Security, Mexico City, Mexico

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ABSTRACT

Testicular cancer accounts for 1% of male neoplasms, the most common histology is bilateral presentation is reported in 1–2% of cases, and germ cell tumor histology represents 90–95% of cases. Specifically, choriocarcinoma as a pure component represents 0.3–1% of these neoplasms and as a mixed component, 8%. We present a 26-year-old male patient with bilateral Wunderlich syndrome secondary to renal metastases from testicular choriocarcinoma.

Introduction

Testicular cancer accounts for 1% of male neoplasms and 5% of urological tumors; The reported worldwide incidence is 71,105 new cases per year, with a mortality of 9507 cases per year, with an incidence of 4603 new cases in Mexico in 2018 generating 571 deaths in the same cause; At the time of diagnosis, patients present 1–2% of cases bilaterally with a histological predominance of 90–95% of germ cell tumors.

Case report

In this study we present a 26-year-old male patient who enters the emergency department, referring left orchalgia and increased ipsilateral testicular volume; On physical examination it reveals the painful left testicle, with increased volume and induration and the right testicle without alterations to palpation; Testicular ultrasound demonstrates bilateral testicular tumor, the left testicle of 5 cm in diameter with heterogeneous echogenicity lesion, the right of up to 3 cm with hypoechogenic multifocal lesions. A CT scan of the chest, abdomen and pelvis contrasted with evidence of bulky left perirenal hematoma, and multiple bilateral pulmonary metastatic lesions, without evidence of retroperitoneal adenopathies, is performed as an extension study; Tumor markers reported Alpha fetus protein (AFP) of 0.84 ng/ml, human chorionic gonadotropin beta fraction (BHCG) 1800 ng/ml and lactic dehydrogenase (LDH) of 897 IU/L. The rest of the results of laboratory tests, without other alterations, hemoglobin without decrease and with hemodynamic stability.

Bilateral radical orchiectomy is performed with findings in the left testicle of the left spermatic cord with tumor-free hypervascularity, the left testicle of approximately 10 × 8 cm stony and the right testicle of 4 × 3 cm, the spermatic cord free of tumor involvement.

The histopathological result of the left testis reveals mixed germinal tumor with choriocarcinoma in 98% and mature teratoma in 2% with dimensions of 5 × 4.5 cm, rete testis invasion, negative surgical margins and positive lymphovascular invasion (Fig. 1). The right testicle had a histopathological report of pure seminoma.

Due to the clinical stability of the patient associated with non-expansive perirenal hematoma, without evidence of a decrease in hemoglobin, it is decided to leave under surveillance.

The patient subsequently receives chemotherapy based on Bleomycin, Etoposide, Cisplatin (BEP), presenting on the fourth day of treatment asthenia, adynamia, dyspnea as well as lumbar pain with right predominance, performing control laboratories documenting a progressive decrease in hemoglobin values of 3 g in 24 hours. A control CT scan is performed with evidence of bilateral retroperitoneal hematoma.

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and hemoperitoneum (Fig. 2) associated with hemodynamic instability. Exploratory laparotomy is performed, with drainage of left retroperitoneal hematoma of 3500 ml associated with metastatic kidney involvement, for which left renal metastasectomy is performed, additionally metastatic commitment to mesentery is evidenced by performing intestinal resection and whole-entire lateral mechanical anastomosis and packing; Admission to intensive care unit for hemorrhagic shock grade IV, acute liver failure and acute renal failure, presenting poor evolution with subsequent death at 24 hours; The histopathological result of kidney and mesentery metastases was choriocarcinoma (Fig. 3).

Fig. 1. Left orchiectomy. Syncytiotrophoblast cells composed of big multinucleated cells and big irregular nuclei.

Fig. 2. CT scan showing bilateral perirenal hematoma.

Discussion

Within the group of germ cell tumors, pure choriocarcinoma is found in 0.3%–1% of these neoplasms and mixed form in 8%; this type of tumor has an aggressive oncological behavior with high metastatic capacity such as choriocarcinoma syndrome which causes bleeding in metastatic sites and significant morbidity and mortality; Alvarado/Hernandez et al. reported a series of 15 patients with choriocarcinoma as a pure or predominant component, finding 100% of cases with metastases (pulmonary 66%, hepatic 60%, brain 20% gastrointestinal tract 13% and renal 6%); There are few studies or reports in the literature of retroperitoneal bleeding or better known as Wunderlich syndrome in association with testicular cancer, finding publications such as
Yee-Huang Ku et al., in 2018 reporting a case of spontaneous retroperitoneal bleeding secondary to testicular choriocarcinoma,\(^3\) or as published in 1992 by Huang et al. reporting another case of choriocarcinoma-type testicular carcinoma with metastatic retroperitoneal involvement associated with Wunderlich syndrome\(^5\); in our case, the high aggressiveness of this type of testicular tumors is clearly evidenced, documenting metastasis to the kidney and mesentery, confirming with the histopathological result the commitment to choriocarcinoma, which is of very little presentation in the world literature, therefore, an early onset of chemotherapy is required in order to reduce this rate of metastasis and high morbidity and mortality.

**Conclusions**

Bilateral Wunderlich syndrome secondary to choriocarcinoma-type testicular tumor is a very poorly documented entity in the literature with a high rate of major complications and high mortality, which despite its low association should be considered at the time of evaluation of these patients in order to start timely treatment and avoid major complications.

**Conflicts of interest**

The authors declare that they have no conflict of interest.

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