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Case Report

Burned-Out Testicular Tumor: A Rare Entity which Can't Be Missed

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Abstract

Burned-out testicular tumors refer to the presence of a metastatic tumor with no visible primary testicular cancer. It's a rare entity difficult to diagnose with a potential negative impact on treatment and prognosis.

We describe the case of a 28-year-old male who presented with a right cervical mass and hemoptysis complaints. Clinical exam of genitals was normal but with a bilateral gynecomastia. A thoracic CT scan revealed well-defined spherical nodules scattered over both lungs and a large right cervical adenopathy. Blood markers showed elevated β-human chorionic gonadotropin (β-hCG) but normal Lactate Dehydrogenase (LDH) and α-Fetoprotein (AFP). Biopsy of the cervical adenopathy revealed a germ cell tumor metastasis. Ultrasound showed three small hypoechoic foci associated with micro calcifications in the left testicle. A left radical inguinal orchidectomy was performed and pathology concluded to a burned-out testicular tumor. The patient was first treated with four cycles of BEP with incomplete response followed by 2 cycles of high doses of Carboplatin and Etoposide. The FDG-PET-CT showed three new hepatic lesions and a new scheme of 4 cycles of GOP were started. The last FDG-PET-CT shows a complete metabolic response on hepatic and pulmonary lesions and partial response on the right supraclavicular adenomegaly.

Introduction

Testicular cancer is a rare disease mainly affecting young and middle-aged men but with an excellent prognosis. The «burned-out» testicular cancer is a very rare distinct entity indicating the spontaneous and complete regression of a testicular tumor in the presence of distant metastases at the diagnosis and generally difficult to diagnose [1]. Since 2016 the International Society of Urological Pathology (ISUP) warned pathologists to consider the risk of a germ cell tumor regression when a testicular scar is detected [2]. The treatment-related toxicity is high with a poorer prognosis [3].

We report the case of a young patient with a large right cervical adenopathy, bilateral gynecomastia and hemoptysis.

Case Presentation

A 28-year-old man with no significant medical history complained of a right cervical mass since two weeks and hemoptysis. His physical exam revealed a large right supraclavicular mass and a bilateral gynecomastia with a normal genital exam.

The radiological work up showed a voluminous right basic-cervical adenomegaly (5.1 cm × 7.9 cm) and well-defined spherical nodules scattered over both lungs characteristics of cannonball metastases. The patient's testicular tumor plasmatic markers revealed an elevated β-human chorionic gonadotropin (β-hCG: 73386 UI/L) but normal lactate dehydrogenase (LDH: 447 UI/L) and α-fetoprotein (AFP: 1.14 UI/mL). A biopsy of the cervical mass was performed and revealed testicular germ cell neoplasia. Subsequently, the patient had a CT scan of the abdomen and a brain MRI showing no metastatic disease. A scrotal ultrasound showed three small hypoechoic foci associated with a lot of microcalcifications in the left testicle.

A left radical inguinal orchiectomy was performed and the macroscopic examination showed three areas of scar tissue and three fibrotic regions without any evidence of neoplastic cells on microscopy (Figure 1 and 2).

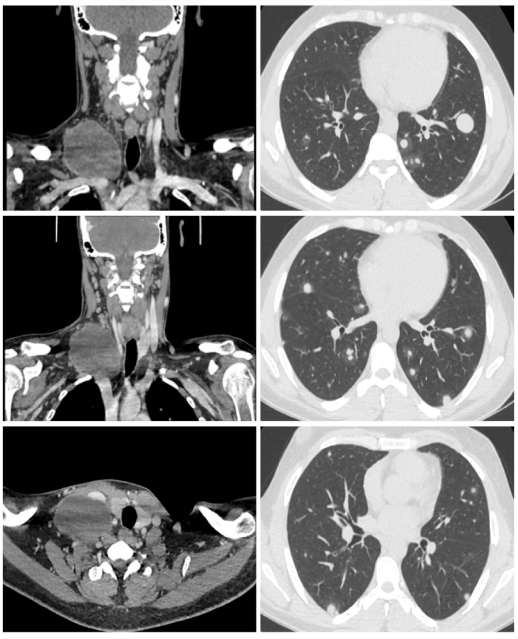

Figure 1: Initial CT-scan of the neck and the thorax prior to starting chemotherapy showed a large cervical mass and a «cannonball» metastasis.

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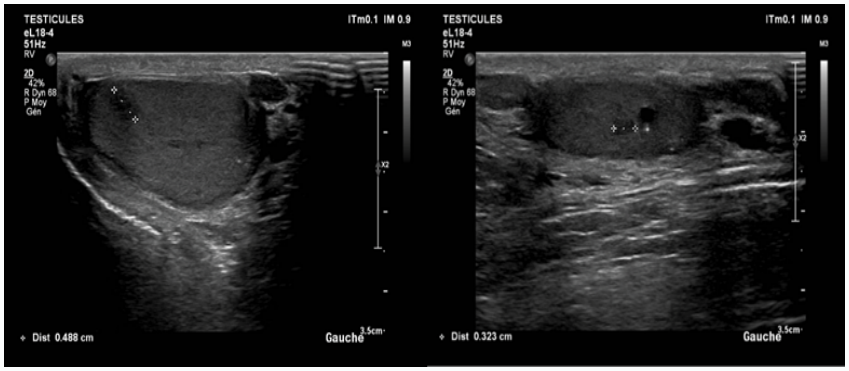

Figure 2: Testicular ultrasound showed three hypoechoic nodules in the left testis.

The patient was treated with four cycles of BEP (bleomycin 30 mg, etoposide 200 mg and cisplatin 50 mg) without any treatment-related side effect. β-hCG blood level increased slightly afterwards with a persistent cervical mass (4.7 cm × 7.1 cm) but a decrease of the lungs nodules.

Intensive chemotherapy with 2 cycles of high doses of Carboplatin and Etoposide and stem cell transplant were administered. The β-hCG level drop down within normal limits but the FDG-PET-CT scan showed three new hepatic lesions (Figure 3).

A new scheme of chemotherapy with 4 cycles of GOP (Gemcitabine-Oxaliplatin-Taxol) was then started. The last FDG-PET-CT shows a complete metabolic response on hepatic and pulmonary lesions and a partial response on the right supraclavicular adenopathy (Figure 4).

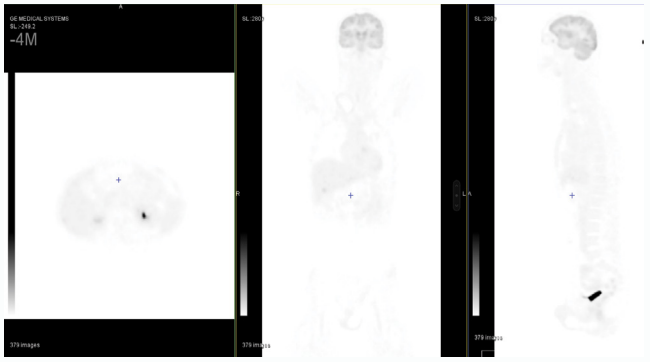

Figure 3: The FDG-PET-CT showed three new hepatic lesions.

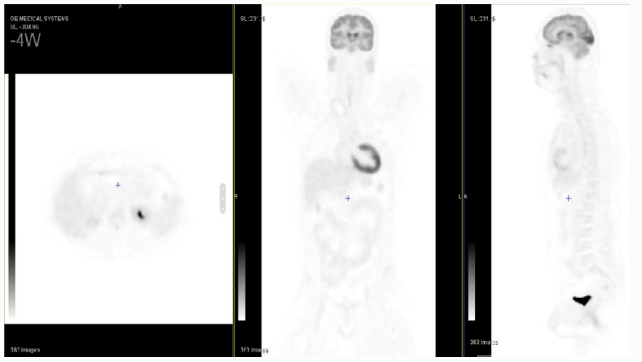

Figure 4: The last FDG-PET-CT showed a complete metabolic response on hepatic and pulmonary lesions.

Discussion

The «burned-out» testicular cancer designation refers to a GCT in extra-gonadal tissues with spontaneous regression of the primary testicular tumor [1]. Most of extra GCT is found in the retroperitoneal, supraclavicular, cervical and axillary lymph nodes and less often in the lung and the liver [4].

A burned-out tumor is characterized by a fibrous scar with no tumor in the testis. Several descriptions have been reported with disseminated choriocarcinoma or embryonic carcinoma [5,6].

How to explain this spontaneous regression? Some hypotheses support a spontaneous regression of the primary after metastasis of the germ cell tumor due to an immune response or ischemic phenomena caused by the high metabolic rate of the metastatic lesions exceeding the blood supply of the primary [7]. Other is in favor of a de novo development in extra-gonadal location [8].

Burned-out tumor may be misdiagnosed and as reported secondary tumors can often be confused with a primary tumor. Meticulous clinical examination and a careful scrotal ultra sound are mandatory. Any abnormality is a call for orchiectomy [9].

High-resolution ultrasound of the scrotum can allow the detection of small, highly echogenic foci, hypoechoic zones, microlithiasis or microcalcifications as reported in this case [10].

Burned-out tumors are more aggressive than primary testicular cancer and more often resistant to first line chemotherapy regimen. If an extra-gonadal seminoma remains sensitive to chemotherapy and is similarly treated with three or four cycles of BEP, extra-gonadal non-seminoma is frequently chemoresistant with much lower 5-year survival rates [11].

Conclusion

Burned-out testicular tumors are rare often difficult to diagnose. Considering this clinical entity should be noted in young patients presenting with gynecomastia or unusual metastasis sites, despite the absence of testicular mass. Plasmatic markers could be of help and testicular ultrasound is mandatory.

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