Case Report

Teratoma Malignant Transformation to Adenocarcinoma in a Retroperitoneal Metastasis of a Germ Cell Tumor in a HIV Acquired Immunodeficiency Syndrome Patient: Case Report

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Abstract:

We report the case of a HIV-positive patient with teratoma malignant transformation in a retroperitoneal metastasis of testicular germ cell carcinoma, submitted to chemotherapy with complete laboratorial response and retroperitoneal lymph node dissection. The pathological analysis of the specimen demonstrated an admixture of non-germ cells within it (adenocarcinoma) with teratoma. Other primary malignancies were excluded. After 48 months, no disease recurrence or other primary neoplasm were diagnosed.

Keywords: Testicular carcinoma; Teratoma; Neoplastic cell transformation

Introduction

Teratoma with malignant transformation (TMT) is defined as a non-germ cell malignancy arising within a teratoma (1). We report the case of a HIV-positive patient with TMT diagnosed at the time of retroperitoneal lymph node dissection (RPLND) after chemotherapy.

Case Presentation

A 28 year-old male presented with a palpable abdominal mass and an enlarged lymph node in the left supraclavicularis fossa. An ultrasonography disclosed a 0.8 cm irregular, hypoechoid lesion with calcification in the left testis and an abdominal CT scan demonstrated a retroperitoneal bulky lesion, measuring 8.0 centimeters. The serum markers were elevated to S2 levels (DHL was 3.5 normal level, beta-hCG 12,500 mIu/ml and AFP 3450 ng/ml). Both Elisa and Western-Blot anti-HIV tests were positive. The patient was considered as AIDS C3 due to the CD4+ lymphocytes count and the diagnosis of a previous *P carinni* infection.

Left radical orchiectomy and excision of the supraclavicular node were performed. The pathologic examination revealed intratubular neoplasm with minor foci of embryonal cell carcinoma in the testis and embryonal cell carcinoma in the supraclavicular lymph node. It was staged as pT1N3M1S2 (stage IIIb).

Chemotherapy was performed with three cycles of PEB (bleomycin, 30 mg/ m² and etoposide, 100 mg/m², days 1-5, plus cisplatin, 20 mg/m²) every three weeks and due to suboptimal response, two cycles of VIP (vinblastin 0,22 mg/Kg plus ifosfamide 1,2 g/ m², plus cisplatin, 20 mg/m², days 1-5) were added. Afterwards, serum markers were down to normal levels.

Correspondence: Hugo Fontan Kohler Rua Santana, 142, Sala 42 18130555, São Roque - Brazil Phone: 55 11 99170230 E-mail: hkohler75@uol.com.br An abdominal CT scan revealed residual disease in the retroperitoneum. A RPLND was then performed.

The pathologic study of the specimen revealed a teratoma with an admixture of non-germ cells within it (Figure 1). Immunohistochemical analysis confirmed



Figure 1 – Microscopic section of the retroperitoneal teratoma, showing a focus of adenocarcinoma. The specimen is colored with hematoxylin-eosin.

the hypothesis of an adenocarcinoma arising within a teratoma. Tumor cells within the teratoma stained for carcinoembryonic antigen (CEA) and cytokeratin 20 (CK 20). The patient was submitted to endoscopy and colonoscopy to exclude a second primary malignancy.

The patient was alive without evidence of disease 36 months after surgery.

Discussion

Teratoma malignant transformation is infrequent. It occurs in approximately 3% to 6% of those treated with platinum-based chemotherapy. In the specific subset of patients with residual teratoma, up to 14% have non-germ cell malignancies ¹. Transformation occurs most frequently for squamous cell carcinoma and less commonly to sarcomas or adenocarcinomas. Two mechanisms have been proposed to explain its occurrence. The first proposes a malignant transformation of the totipotential embryonal carcinoma cell and the second, a malignant transformation of mature teratoma elements¹⁻². Transformation would be facilitated by chemotherapy or radiation therapy.¹⁻² It may be divided in two types: induced by chemotherapy or radiation therapy; and spontaneous, with different clinical behavior in each case. The induced TMT occurs earlier and behaves more aggressively while the sporadic type develops in long-standing teratomas with a more benign clinical course.¹⁻² Its treatment is based upon complete surgical excision.³

Although testicular carcinomas are not considered as non-AIDS-defining cancer, its incidence is statistically higher in these patients than in the general population.⁴In an analysis of patients with testicular seminoma, this risk is higher for patients with disseminated disease, suggesting a role for immunosupression and may be associated with AIDSrelated testicular atrophy.⁵ There is no previous report of the association between TMT and HIV-infection/ AIDS, although we may suppose that impaired tumor immunosurveillance could facilitate adenocarcinoma differentiation within the teratoma.

TMT is a poor prognostic sign when found in metastatic sites and clinicians should be aware of it when there is disease progression with lowering of serum markers or a mixed response to treatment, with regression at one site and progression at another⁶. In patients with TMT, recurrence rates range from 59% to 86%, with an overall survival of 58% to 65%.^{1,6} It also must be remembered that long-term dedifferentiation of teratomas to malignant neoplasms may occur, requiring prolonged surveillance of patients with teratoma after chemotherapy for nonseminomatous germ cell tumors of the testis or testicular teratomas diagnosed in infancy.^{3,7}

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