Bilateral adrenal lymphoma

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ABSTRACT

Introduction: Noninvasive, high resolution imaging methods have enabled a greater number of asymptomatic adrenal masses (incidentalomas) to be diagnosed. Incidence is reported at 0.35-4.4% in computed tomography and 1.4-5.7% in autopsy series. In relation to etiology, the most frequent tumors are adrenal adenomas, with a frequency of 36-94%. Adrenal cancer frequency is 1.2-11% and metastases to other organs is 0-21%. Infections, granulomatous processes, and lymphomas are rare.

Genitourinary apparatus disease can be secondary to lymphoma and primary disease is uncommon. One case of lymphoma that presented with bilateral testicular and adrenal masses was found in the literature.

Keywords: Adrenal lymphoma, incidentaloma, bilateral, Mexico.

RESUMEN

Introducción: El empleo de métodos de imagen no invasivos y de alta resolución han permitido diagnosticar un mayor número de masas adrenales asintomáticas (incidentalomas). De acuerdo a su etiología, los tumores más frecuentes son los adenomas adrenales con una frecuencia de 36% a 94%. Los linfomas pueden afectar al aparato genitourinario de manera secundaria y la afectación primaria es rara. Se presenta el caso de un paciente masculino de 55 años, sin antecedentes de importancia, quien inició su padecimiento con pérdida de peso de 12 kg en cuatro meses, acompañado de lumbalgia derecha. Acudió con el médico quien le realizó Tomografía Axial Computarizada (TAC) y Resonancia Magnética Nuclear (RMN) abdomen-pélvicas, en las que se identificaron lesiones suprarrenales bilaterales. Se le realizaron mediciones de catecolaminas y metanefrinas en orina las cuales se reportaron dentro de parámetros normales. Con sospecha clínica de carcinoma adrenal derecha y adenoma izquierdo se le propuso realizar inicialmente adrenalectomía derecha por vía laparoscópica. Por presencia de lesiones óseas en columna y manifestaciones clínicas asociadas, fue tratado con radioterapia (RT) paliativa 30 Gy en 10 sesiones en campos oblicuos posteriores y un campo anterior y quimioterapia adyuvante.

Palabras clave: Linfoma adenal, incidentaloma, bilateral, México.
INTRODUCTION

The use of noninvasive and high resolution imaging methods has enabled the diagnosis of a large number of asymptomatic adrenal masses (incidentalomas). Their incidence in computed tomography (CT) is reported to be from 0.35-4.4% and from 1.4-5.7% in autopsy series. In relation to etiology, the most frequent tumors are adrenal adenomas, with a frequency of 36-94%, adrenal cancer, with a frequency of 1.2-11%, and metastases to other organs, with a frequency of 0-21%. Incidence of infections, granulomatous processes, and lymphomas is not common.¹

Lymphomas can affect the genitourinary tract in a secondary manner and primary disease is rare. One case of lymphoma presenting as bilateral testicular and adrenal masses was found in the literature.²

Image 1. Bilateral adrenal tumors: right tumor measures 5 cm x 6 cm, left tumor measures 3 cm x 3 cm and both have solid aspect with no interior fat and no regional adenopathy.

Image 2. NMR image of the spinal column. 50% invasion of the medullary canal with dural sac compression and thoracic spine instability.
CASE PRESENTATION

Patient is a 55-year-old man with no important past medical history. Illness began with 12 kg weight loss in four months, accompanied with right lumbalgia. He sought medical treatment and physician ordered abdominopelvic CT scan and nuclear magnetic resonance (NMR) imaging study (Image 1) that identified bilateral adrenal lesions: a 5 cm x 6 cm tumor dependent on the right adrenal gland and a 3 cm x 3 cm tumor dependent on the left adrenal gland. Both tumors had solid aspect, were not hyperintense, and had no evidence of interior fat. There was no evidence of regional adenopathy and the rest of the intra-abdominal organs had no alterations. Catecholamines and metanephrines in urine were measured and were within normal parameters. Patient was referred to the Instituto Nacional de Cancerología (National Institute of Cancerology) where CT scan of the cranium and chest were done and they showed no evidence of brain or lung neoplastic lesions. Change in thoracic vertebral body density was observed and so MIBG scintigram was done and was negative. With the clinical suspicion of right adrenal carcinoma and left adenoma, initial right laparoscopic adrenalectomy was proposed. Patient later presented with bone pain in the thoracic spine and paresthesia in the lower hemiabdomen. His lower extremities remained strong, with sensitivity and reflexes, but his gait was ataxic. Suspecting medullary canal compression, NMR study was ordered and showed lithic lesions secondary to neoplastic invasion in T1-T5, pathological fracture in T4 and T8, and invasion of 50% of medullary canal with dural sac compression and thoracic spine instability (Image 2).

Patient was treated with palliative radiotherapy (RT) of 30 Gy in 10 sections in posterior oblique fields and one anterior field. Corpectomy, medullary decompression, and lateral support placement were carried out. Patient presented with significant bone pain reduction and had normal gait after RT. Control NMR study showed complete response of spinal lesions (Image 3).

In relation to vertebral column findings and the presence of afternoon fever with peaks of up to 39°C, the following differential diagnosis of fungal or microbacterial infection affecting the adrenal glands and dorsal column was considered. Staining for microbacteria and fungi, purified protein derivative (PPD), and cultures were done and results were negative. Tomography-guided biopsy of right adrenal tumor was performed and histopathological study reported non-germinal center diffuse large B-cell lymphoma. Immunohistochemistry studies with CD 20, MUM1, Bcl 6, and CD 3 were positive. Treatment was begun with R-CHOP chemotherapy (rituximab, cyclophosphamide, doxorubicin, vincristine, prednisolone) and patient is currently undergoing his second cycle (Image 4).

DISCUSSION

Adrenal tumor evaluation is based on establishing the difference between malignant and benign masses and functioning and non-functioning masses. While autopsy reports show that 25% of lymphomas affect the adrenal...
glands, primary disease is rare and more than 90% are B-cell tumors. Bilateral disease can cause adrenal failure and rapidly progress to extranodal disease. Despite treatment, few patients survive for more than two years. Average survival is considered to be 15 months, however, one case was reported with 8-year survival with adrenalectomy and adjuvant radiotherapy treatment. There are only a few cases in the literature reporting on bilateral adrenal disease (19 of 27 published cases are on primary adrenal lymphoma).

This disease is more common in men than in women (19:8); symptoms are non-specific although they can be related to adrenal failure (12 of 27 cases). Tumors are usually discovered incidentally in ultrasound or CT imaging studies. Differential diagnosis is difficult and includes metastasis and adenoma. These tumors are believed to arise from hematopoietic tissue inherent in the adrenal gland.

**BIBLIOGRAPHY**