

CASE REPORT

Bilateral Adrenal Lymphoma: A Diagnostic Challenge

Youssef Kharbach ^a , Siham Alaoui Rachidi ^b, Zakaria Bakali Issaoui ^c, Abdelhak Khallouk ^d 

^a MD, Urology assistant professor, Urology department, Tanger University Hospital, Faculty of Medicine and Pharmacy, Abdelmalek Essaâdi University, 90000 Tangier, Morocco

^b MD, Radiology assistant professor, Radiology department, Tanger University Hospital, Faculty of Medicine and Pharmacy, Abdelmalek Essaâdi University, 90000 Tangier, Morocco

^c MD, Urology resident, Urology department, Tanger University Hospital, Faculty of Medicine and Pharmacy, Abdelmalek Essaâdi University, 90000 Tangier, Morocco

^d MD, Urology professor, Urology department, Tanger University Hospital, Faculty of Medicine and Pharmacy, Abdelmalek Essaâdi University, 90000 Tangier, Morocco

ABSTRACT

Bilateral primary adrenal lymphoma is rare with no specific clinical or imaging features. We report herein the case of a 44-year-old man presenting with massive bilateral adrenal tumors associated with normal adrenal function. Computed tomography findings were nonspecific. Because pheochromocytoma was ruled out and the patient had a normal coagulation test, we performed an ultrasound-guided adrenal biopsy for diagnostic purposes. Pathology confirmed adrenal non-Hodgkin's lymphoma. By reporting our case, we aimed to propose a practical approach for this rare entity and to show that there are still situations in which biopsy is necessary to make a diagnosis.

KEYWORDS: Lymphoma; Adrenal Gland; Image-Guided Biopsy.

Correspondence: Dr Youssef Kharbach. Urology department, Tanger University Hospital, Faculty of Medicine and Pharmacy, Abdelmalek Essaâdi University, 90000 Tangier, Morocco. Email: ykharbach@uae.ac.ma

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INTRODUCTION

Primary adrenal lymphoma (PAL) is rare with no strict definition and is rarely suspected in bilateral adrenal masses due to the absence of specific clinical or imaging features.^{[1],[2],[3]} This presents a diagnostic challenge. Authors report herein the case of a 44-year-old man with a PAL presenting as massive bilateral adrenal tumors associated with normal adrenal function. By reporting our case, we aimed to propose a practical approach for this rare entity and showed the feasibility of adrenal ultrasound-guided biopsy.

CASE HISTORY

A 44-year-old man was admitted to our hospital because of progressive nonspecific bilateral lower back pain, anorexia, nausea, weight loss, fatigue, and fever (38°C) for 4 months. The abdominal examination found nonpainful masses in both sides, which were difficult to delineate. No palpable lymphadenopathies were found. The patient had no symptoms of adrenal insufficiency, such as pigmentation on skin and mucous membrane or constipation.

Abdominal ultrasound suggested bilateral adrenal masses. Only plain computed tomography (CT) was performed because the patient was allergic to contrast material. CT scan of the abdomen and pelvis revealed huge masses in bilateral adrenal area. These masses were necrotic in their center and had regular contours. They did not appear encapsulated and measured 16.2 x 11.6cm x 16.7 cm on the right and 10.1cm x 8.7cm x 12cm on the left [Figure 1]. CT scan also showed one para-aortic lymphadenopathy of 25mm.

A complementary thoracic CT scan was performed which did not objectify other secondary lesions or lymphadenopathies.

No laboratory abnormalities were identified. The results of the adrenal function tests were normal.

Because pheochromocytoma was ruled out and the patient had a normal coagulation test, we performed an ultrasound-guided adrenal biopsy for diagnostic purposes. Pathology confirmed non-Hodgkin's lymphoma (NHL) composed of high grade malignant and activated diffuse large B- cells [Figure 2].

The patient underwent a gastro-intestinal (GI) endoscopy and a bone marrow biopsy, which found no anomalies.

The diagnosis of bilateral PAL was confirmed, in view of no prior history of lymphoma, the poor involvement of

lumbo-aortic lymph nodes, and the negative bone marrow biopsy.

As surgery is not recommended in these patients, we referred the patient to an oncologist for further chemotherapy and follow-up.

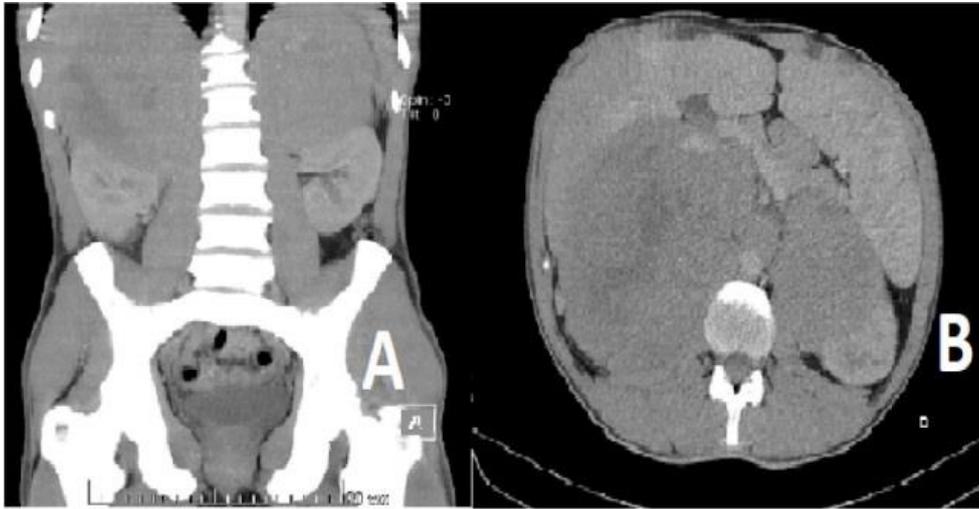


Figure 1: CT scan of the abdomen and pelvis revealing bilateral adrenal masses with necrosis and regular contours. A: axial image. B: coronal image.

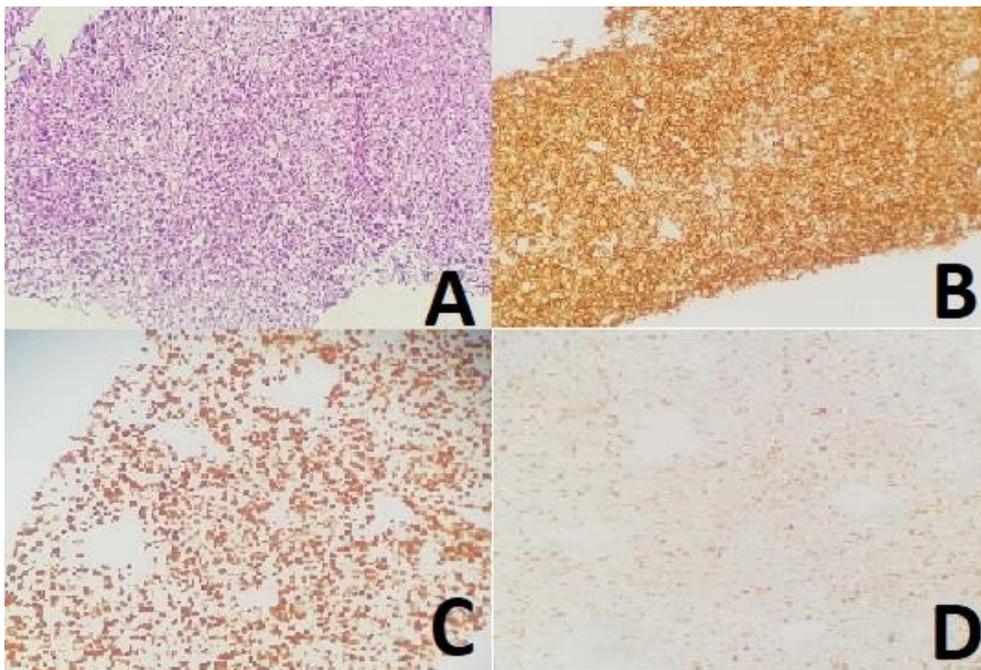


Figure 2: Histopathologic photography. A: Lymphomatous proliferation (H&E staining, original magnification at 20x). B: Lymphoma cells staining positively for CD20. C: Lymphoma cells showing intensive staining for Ki-67. D: Lymphoma cells staining positively for Mum1

DISCUSSION

Primary adrenal lymphoma is a rare form of non-Hodgkin lymphoma (<1%),^[1] with elderly male predominance.^[2, 3] It is bilateral in about 75 % of cases,^[3] and there is no specific clinical or imaging features.^[1,4] Adrenal insufficiency is found in 61% of cases.^[3, 4]

Actually, bilateral adrenal masses are rare with variable clinical presentation from asymptomatic patients to severe systemic clinical presentation.^[2] They require a different approach comparing to unilateral tumors.^[2]

Thus, bilaterality is non specific and suggests, in addition to adrenal lymphoma, many diagnoses such as metastasis, bilateral pheochromocytoma, bilateral cortical adenomas, adrenocortical carcinoma and infections such as tuberculosis.^[2, 5, 6] Subsequently, physical examination should be undergone to look for hormonal oversecretion.^[2]

First, the functioning status of adrenal masses should be explored by a hormonal evaluation including testing for subclinical Cushing's, pheochromocytoma and hyperaldosteronism.^[2, 5]

In hypo- or normocortisolaemic patients, CT characteristics usually lead to the diagnosis and guide treatment.^[2] If not, further characterization with magnetic resonance imaging (MRI) using chemical shift imaging is usually performed.^[7] However, both CT and MRI examinations have low specificity in diagnosing malignancy.^[8]

When imaging characteristics and biochemical findings do not lead to the diagnosis, adrenal biopsy become of great interest.^[2, 8-10] Multiple imaging modalities may guide percutaneous biopsies and the choice is based on equipment availability, cost, lesion conspicuity, and physician preference.^[8, 10] It seems to us that ultrasound-guided (US) biopsy in large adrenal tumors, as in our patient, is feasible and safe. US guidance allows real time visualization, absence of radiation, low cost and rapid availability.^[8] The biopsy should be performed in the most comfortable position for the patient.^[8] Core biopsy is preferred to fine needle aspiration in lymphoma,^[7] and should be performed after ruling out coagulation disorder and pheochromocytoma.^[8, 10]

Adrenalectomy is useless in PAL and the treatment is based on polychemotherapy.^[1] The prognosis is poor with rare durable remission after chemotherapy.^[3]

CONCLUSION

There are many etiologies of bilateral adrenal masses. Clinical and imaging features could lead to suspect the diagnosis. Adrenal biopsy is indicated when

pheochromocytoma and coagulation disorder are ruled out and imaging characteristics are nonspecific. Also, US guidance is feasible and safe in large tumors such as in our patient.

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We want to acknowledge our patient from whom we learn.

AUTHORS' CONTRIBUTIONS

The participation of each author corresponds to the criteria of authorship and contributorship emphasized in the [Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly work in Medical Journals of the International Committee of Medical Journal Editors](#). Indeed, all the authors have actively participated in the redaction, the revision of the manuscript, and provided approval for this final revised version.

COMPETING INTERESTS

The authors declare no competing interests with this case.

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None.

PATIENTS CONSENT

Written informed consent was obtained from the patient for the publication of this case report.

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