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RESEARCH ARTICLE

PRIMARY ADRENAL LYMPHOMA B A LARGE CELLS (ABOUT TWOCASES)

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ABSTRACT

Bilateralprimary non-Hodgkin'slymphoma (NHL) of the adrenals is uncommon. 136 cases have been described in the literature. The symptoms of disease are variable and depend on the tumor size and the presence of adrenal insufficiency we report 2 cases of adrenal lymphoma, discovered following bilateral back pain and deterioration of general conditions. Hormonal exploration demonstrated adrenal insufficiency. Imaging explorations showed a large and bilateral adrenal mass. Percutaneous ultrasound guided biopsy of the adrenal and search for extension revealed primary bilateral adrenal lymphoma. After glucocorticoid substitution, treatment was based on a CHOP regimen chemotherapy, our two patients died uring their first year of treatment. The diagnosis of primary adrenal non-Hodgkin lymphoma should be investigated in patients with a rapidly growing bilateral adrenal mass associated with adrenal insufficiency.

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INTRODUCTION

Adrenal lymphoma is an exceptional etiology of bilateral adrenal masses, it represents less than 1% of all cases of non-Hodgkin's lymphoma (4), in fact 136 cases of adrenal lymphoma have been published of which 80% were bilateral (1).

OBJECTIVE OF THE WORK: To describe the different epidemiological, clinical, paraclinical and therapeutic aspects of this condition in the light of the data in the literature.

PATIENTS AND OBSERVATIONS

Observation 1: Patient aged 42 years, without any particular pathological history, hospitalized for abdominal pain and low back pain of progressive onset evolving for 4 months with an important alteration of the general state. The clinical examination found a pale patient, in poor general condition, orthostatic hypotension, bilateral lumbar contact. The biological examinations: have objectified an inflammatory syndrome with a VS to 100mm, normochrom normocytic anemia and an elevation of LDH: 1640, a hypocalcemia to 86 mg/l.

The endocrine workup that we were able to perform showed: a normal urinary methoxylated derivatives assay, thus ruling out a pheochromocytoma, and a low cortisol level. The search for a tubercular origin was negative. The uroscanner showed bilateral adrenal tumor lesions measuring respectively (7.7-12 cm), (10-15 cm) of regular contours, heterogeneous density, central hypodensity, related to a central necrosis. Both masses were enhanced after injection of contrast medium, the Washout was positive (figure1). In view of the anemia, an osteomedullary biopsy with immunohistochemical study showed a marrow rich in lymphocytes with dysmyelopoiesis of the three lineages. A biopsy of the mass showed a lymphoid proliferation suggestive of a diffuse large cell B lymphoma (Figure 2, 3). The patient received chemotherapy and died during the first year of treatment.

Observation 2: Patient aged 24 years, without any notable pathological history, complains of right lumbar pain with marked alteration of the general state, hospitalized for shock with lumbar pain and marked alteration of the general state, her clinical examination finds a patient in bad general state, tachycardia, hypotensive, dehydrated, pale with presence of melanoderma on the elbows and the oral mucosa. The biological examinations show an inflammatory syndrome with a VS at 150 mm, an elevation of LDH and an adrenal insufficiency.

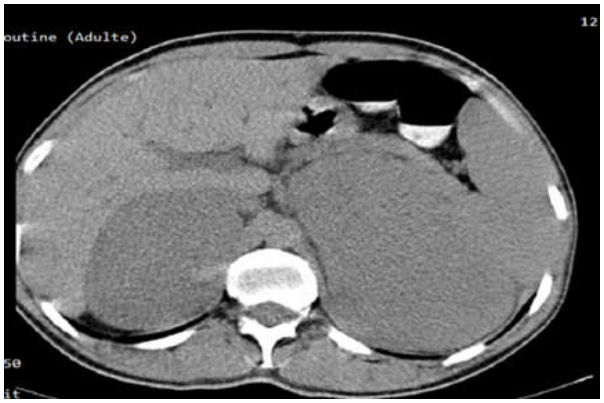

Figure 1. Scannographic section showing the two adrenal masses measuring 10.43 x 7.44 cm on the left side and 16.06 x 9.70 cm on the right side

Figure 2: Immunohistochemistry with CD20 labelling of tumour cells

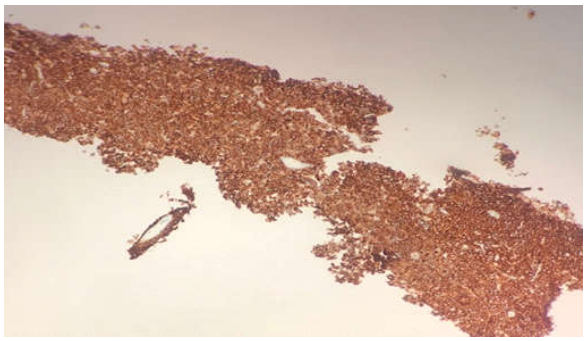

Figure 3. Immunohistochemical study with diffuse nuclear labelling by Ki67

The abdominal scan shows two bilateral adrenal masses more voluminous on the right side and having an intimate contact with the liver, compressing the kidney and the right excretory tracts below, they are associated with a magma of lombo-aortic mesenteric adenopathies and renal hilum, free ascites of medium abundance (Figure 4). The biopsy of the adrenal mass shows a large cell B lymphoma. The patient died a few months after being put on chemotherapy.

DISCUSSION

Primary adrenal lymphoma is extremely rare, defined as malignant neoplastic proliferation of lymphoid cells exclusively in the adrenal glands (9),the most common histologic type is diffuse large B-cell lymphoma (1) . A total of 136 cases have been published between 1983 and 2015 (1). It occurs twice as frequently in men as in women, with a peak age of 62 years, and a predominance of bilateral forms in 80% of cases (1).

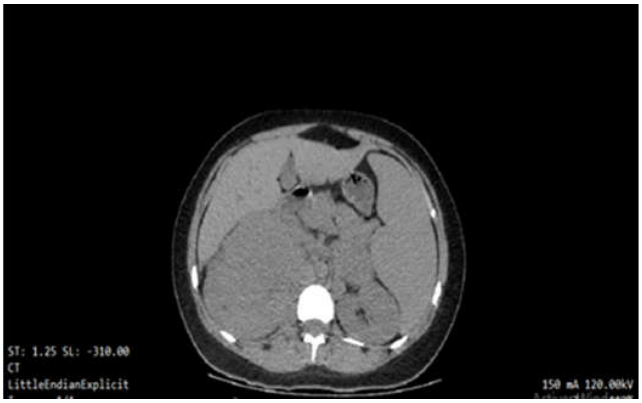

Figure 4. Two bilateral adrenal masses larger on the right side and having intimate contact with the liver, compressing the kidney and excretory tract with a magma of lumbo-aortic mesenteric adenopathies and renal hili.

Clinically, it is manifested by poor and non specific signs, related to the lymphoma it self (fever, weight loss), the tumor volume (abdominal pain, dorsolumbar pain) or more often adrenal insufficiency (vomiting, orthostatic hypotension, skin pigmentation) (1,3). Our two patients presented an abdominal pain associated with an altered general state and orthostatic hypotension. In lymphomas, the biological abnormalities is not remarkable, we can find: an inflammatory syndrome, an elevation of LDH or hypercalcemia and most often a total adrenal insufficiency. Our two patients presented an increase in sedimentation rate and LDH as well as adrenal insufficiency and they were under replacement therapy. Radiologically, the ultrasound may show hypervascularization, association of liquid areas corresponding to hemorrhage or necrosis with tissue areas: a sign suggestive of lymphomatous origin. The Adrenal scan shows the classic aspect of bilateral symmetrical masses well limited without fatty content relatively homogeneous, size >5cm, high density and low contrast after injection of the contrast product, slowing down of the washout of the contrast medium, local or distant lymph node extension (8,9), MRI does not allow to differentiate between lymphoma and metastasis, about 70% of lymphomas appear in hypo signal T1 and in hyper signal T2 (8,9).

In fact, there is a cluster of arguments that should make one think of the diagnosis of adrenal LMNH: the increase in LDH, the CT data and the adrenal insufficiency. However, the definitive diagnosis can only be made by a histological sample taken by laparotomy or by fine needle biopsy guided by ultrasound or CT scan, the latter must be performed after having formally ruled out a pheochromocytoma or a coagulation anomaly. Our two patients benefited from ultrasound-guided biopsies allowing to make the diagnosis.
In order to establish the primary nature of the adrenal lymphoma, a careful extension work-up must be performed, which should not show any other localization; it is both clinical (no adenopathy, liver or splenomegaly) and paraclinical: chest X-ray or, better still, a thoracic-abdominal-pelvic scanner. The treatment of adrenal lymphoma is essentially based on chemotherapy. Different protocols have been used, but due to the rarity of the disease, there are no retrospective studies evaluating the results of chemotherapy. CHOP (cyclophosphamide, doxorubicin, vincristine, prednisone) remains the most prescribed treatment protocol. Immunotherapy with the anti-CD20 monoclonal antibody (rituximab) as an adjunct to chemotherapy is currently being evaluated in aggressive type B NMH.

Surgery does not appear to be beneficial. Radiation therapy has been used by some authors as an adjunct to chemotherapy (1, 2,5). The prognostic results of published cases are rare (1), generally considered pejorative because the disease is aggressive and progresses rapidly A mean survival of one year in only 20% have been reported; however, because of the rarity of this disease, prognostic factors are difficult to elucidate (5). Our two patients died during their first year of treatment.

CONCLUSION

Adrenal lymphoma is a rare cause of adrenal mass, the involvement is more commonly bilateral and symmetrical, the clinical presentation is poor, This explains the late discovery. Imaging allows a certain restriction of the diagnostic range, without differentiating lymphoma from metastases. The diagnosis remains histological guided by cross-sectional imaging. Treatment is based on chemotherapy, and the prognosis remains pejorative.

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