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RESEARCH ARTICLE

ACUTE ADRENAL INSUFFICIENCY OF TUBERCULOSIS ORIGIN: CLINICAL CASE

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Abstract

AdrenalTuberculosis is arare and difficult condition to diagnose. It issometimesdiscoveredduring the etiologicalassessment of adrenalinsufficiency. Hereafter, is a report of a rare case of adrenaltuberculosis revealed through acute adrenal insufficiency, associatedwithcutaneoustuberculosiswithout an active pulmonary component.

Introduction:-

Addison's disease (AD) or Primary Adrenal Insufficiency (PAI) was first described in 1855 by THOMAS ADDISON in patients with adrenal tuberculosis. It is a rare disease with an incidence of 4/1,000,000 cases per year in Western countries and has potentially fatal consequences[1] and itssymptoms usually vary dependingon the extent of glucocorticoid and mineralocorticoid hormone deficiency.

In regards to the etiology of this disease, studies suggest that in developed countries, about 75-80% of cases are caused by autoimmune destruction, i.e. autoimmune adrenalitis, while in developing countries Tuberculosis (TB) is the most common cause [2].

Case Presentation

A 41 years oldmale, was admitted to the emergency roomin a state of hypovolemic shock with ahypotension of 60/40 mmHg, signs of intra- and extracellular dehydration, vomiting and diffuse abdominal painwith nonotable neurological disorders.

The patient has no particularpathologicalbackground, specifically no Tuberculosis.

Upon questioning, the patient reported gradual onset physical, psychological and sexual asthenia, predominantly in the evenings, associated with anorexia and a significant weight loss estimated at 20 kg in 5 months.

Clinical examination revealed skin hyperpigmentation on the face, elbows and soles of the feet with accentuated slate patches on the oral mucosa (Figure 1) suggesting melanoderma.

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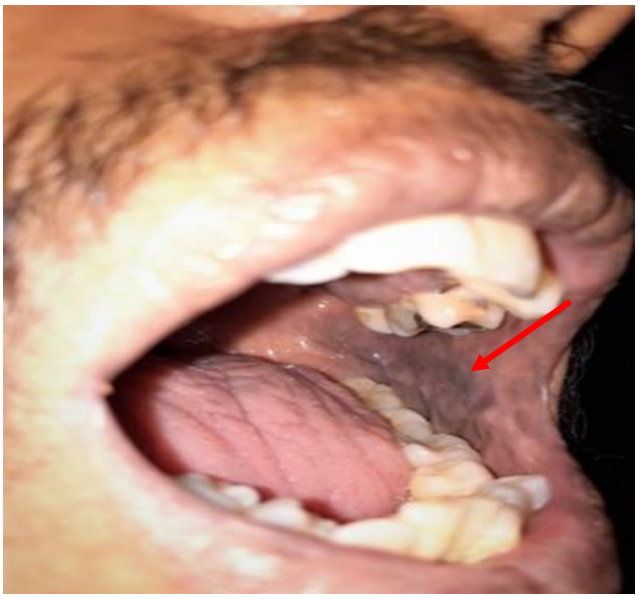

Figure 1:- accentuated slate patches on the oral mucosa.

Incidentally, we have also found a non-pruritic skin lesion on the right lumbar region that has recurred for 6 years, corresponding to a soft painless scarring plaque centered by a skin orifice.

The biological assessment showed a hyponatremia at 130mmol/l, a hyperkalemia at 5.35mmol/l, altered renal function with a GFR of 45.34ml/min, a biological inflammatory syndrome: CRP: 174 mg/l, Sedimentation rate: 125 mm during the first hour; fasting blood glucose at 0.80g/l. Regarding blood count, it showed a normocytic normochromic anemia and a normal leukocyte count. The 08h Cortisolemia was low at 45.30 nmol/l (normal is 101 to 535nmol/l), ACTH determination was not done due to lack of means.

At this point, the clinical and biological status of the patient was strongly suggestive of primary adrenal insufficiency in acute decompensation, and the patient was treated with intravenous hydrocortisone.

As part of the etiological assessment, an abdominal ultrasound was done and it described a tissue mass of the right adrenal cavity with irregular heterogeneous contours of 33.7 mm long axis. This was confirmed by the abdominal CT scan that described two tissue lesions largely necrotic involving the 2 adrenals (Figure 2). Afterwards, an abdominal MRI was done revealing bilateral adrenal hypertrophy with nodular lesions in T2 hypersignal enhancing peripherally in an annular fashion delimiting areas of necrosis (Figure 3).

Chest X-ray was normal, but the Quantiferon ^{R} test(for immunological diagnosis of tuberculosis by measuring the release of gamma interferon from sensitized T cells in response to highly specific Mycobacterium Tuberculosis Antigens) was positive.

Biopsy of the lumbar skin lesion revealed a granulomatous dermatitis compatible with cicatricial tubercular gum, althoughsputum and urine BK tests were negative on direct examination and culture.

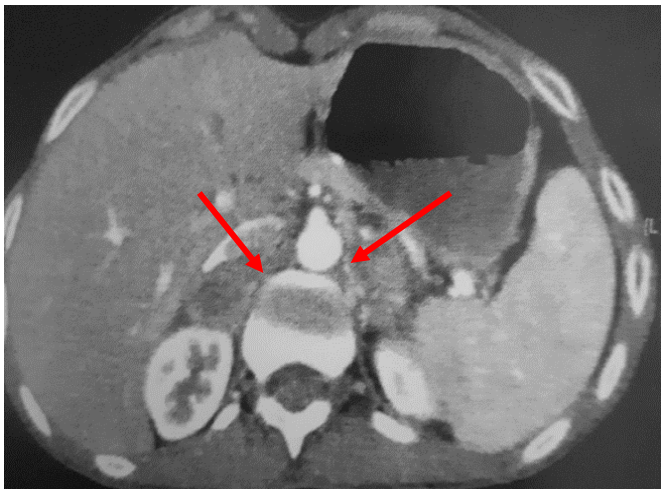

Figure 2:- 2 necrotic tissue lesions involving the 2 adrenal glands.

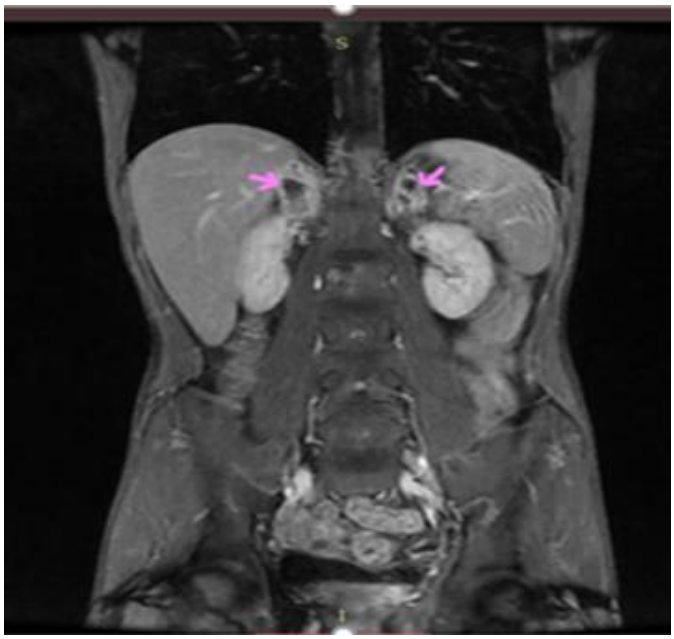

Figure 3:- Bilateral adrenal hypertrophy in T1 hyposignal with peripheral enhancement delimitingareas of necrosis.

Given the context of cutaneous tuberculosis, the diagnosis of primary adrenal insufficiency of tuberculosis origin was retained. Consequently, the patient was treated with anti-bacillary drugs according to the regimen established by the nationalanti-tuberculosis program, and by hormonal substitution: hydrocortisone 40mg/day in 2 doses at 8H and 16H with a good clinical and biological evolution, the dose of hydrocortisone was doubled because of the association with the enzymatic inducer: Rifampicin.

Fludrocortisone was not administered because the patient did not develop hypotension or hyponatremia after starting Hydrocortisone.

The patient was informed of the stress dosage, was equipped with an ADDISONIEN card and a glucocorticoid preparation for emergency parenteral administration.

Anti-bacillary drugs were stopped after 6 months of treatment, and the dose of hydrocortisone was then lowered to 20mg/day without recurrence of adrenal insufficiency signs.

The 6-month check-up showed clinical improvement with a weight gain of 5kg, the hormone levels remained low.

Discussion:-

One of the common sites of extrapulmonary tuberculosis is the adrenal glands and PSI occurs due totheirdestructionby caseous necrosis. For adrenal insufficiency to occur, at least 90% of the glands must be destroyed.Therefore, adrenal function rarely recovers even after anti-bacillary therapy is taken [3].

The adrenal glands are in 70% of cases bilaterally involved, which is the result of hematogenous or lymphogenous spread from the primary site of infection [3].

In a large autopsy study, the adrenal glands were the only site of infection in 25% of patients with adrenal TB [1,5], in consequence they might me the only infected organs with TB.

During the installation of Addison's disease, the adrenal glands are enlarged by the infiltration of inflammatory cells and granulomas in the cortex. This inflammation can calcify over time. Adrenal calcifications may be observed radiographically in 50% of patients [4].

The periodbetween non-adrenal tuberculosis and the onset of Addison's disease, may range from 0 to 50 years with a mean of 31.9±14.9 years [4]. In the present case, the discovery of Addisson's disease was concomitant with that of cutaneous tuberculosis, but the dermatosis had been recurring for 6 years.

When diagnosing PSI, patients with isolated adrenal damage don’t show any specific clinical signs. They usually present with general signs, abdominal pain or a feeling of heaviness... The signs of adrenal insufficiency are late and only appear in the chronic or inactive phase [5].

During the radiological assessment, Ultrasound will show a Uni or Bilateral adrenal mass; however, CT scans are more sensitive and allow better characterization.

During the acute phase, a CT scan will show adrenal hypertrophy often bilateral with areas of central necrosis (caseous necrosis) objectified by contrast injection. In this phase, most patient are almost always asymptomatic (incidentaloma).

The evolution during the chronic phase, is commonly characterized byatrophy and calcifications [5], though the absence of calcified adrenal glands does not exclude tuberculosis as a cause of adrenal insufficiency.

The diagnosis of certainty is based on an ultrasound or scannoguided biopsy with a histological study. The finding of a epitheliocellular granuloma with caseous necrosis is a specific confirmationof tuberculosis.

It is imperative to eliminate a pheochromocytoma before any diagnostic procedure. This being said, a biopsy is not necessary in patientsalready diagnosedwith extra-adrenal tuberculosis [6].

Concerning treatment plans, concomitant management of PSI and TB may be difficult since rifampin is a potent inducer of the cytochrome P450 (CYP) system, which is involved in the metabolism of glucocorticoids. This interaction results in decreased plasma levels of steroids, which can be potentially dangerous [4].

In the absence of exact guidelines for dose adjustment, it is advisable to make adjustmentswhen the enzyme inducer is started and after it’s stopped. In our patient, we doubled the dose of steroids when initiating anti-bacillary therapy and decreased it when stopping it.

Conclusion:-

Tuberculosis is still endemic in Morocco. PAI caused by tuberculous adrenalitis has non-specific symptoms and its onset is often insidious, which is why the diagnosis is often overlooked, andthe risk of acute adrenal insufficiency increases.

In the etiological assessment of chronic adrenal insufficiency, tuberculosis should be considered on the basis of a combination of clinical, biological and radiological evidence.

The treatment of TB infection simultaneously with PAI may be difficult to manage because of the interaction between rifampin and glucocorticoid drugs.

Despite the administration of anti-bacillary drugs, adrenal function does not recover in most cases of PSI of tuberculosis origin.