AACE Clinical Case Rep. 8 (2022) 45–46

Contents lists available at ScienceDirect

AACE Clinical Case Reports

journal homepage: www.aaceclinicalcasereports.com

Visual Vignette

Adrenal Histoplasmosis-A Therapeutic Restoration of Adrenal Morphology

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ARTICLE INFO

Article history:
Received 10 March 2021
Received in revised form
16 March 2021
Accepted 16 March 2021
Available online 26 March 2021

Case Presentation

A 42-year-old man presented with generalized weakness, easy fatigability, hyperpigmentation, low appetite, and significant weight loss of 10 kg and low grade fever over the last 6 months. He did not have cough, abdominal pain, or vomiting. He had hypertension for 5 years but had not required his antihypertensive medications for the last 2 months because of normal blood pressure values. General examination revealed hyperpigmentation on knuckles, oral mucosa, and palmar creases. Systemic examination was unremarkable. The patient had a serum sodium of 128 mmol/L, serum potassium of 5.2 mmol/L, along with a serum cortisol (8 AM) of 4.55 mcg/dL. His plasma adrenocorticotropic hormone (ACTH) was 174 pg/mL. Following an ACTH stimulation test, the serum cortisol was 7.5 mcg/dL. Other investigations, including complete blood count and liver function test, were normal. Chest x-ray was normal. The patient underwent a computed tomography (CT) scan of the abdomen (Fig. 1). Based on the findings shown in Figure 1, a CT-guided biopsy was done, and histopathology is shown in Figure 2. Patient was started on treatment and there was significant clinical improvement. CT abdomen after 1 year of treatment is presented in Figure 3.

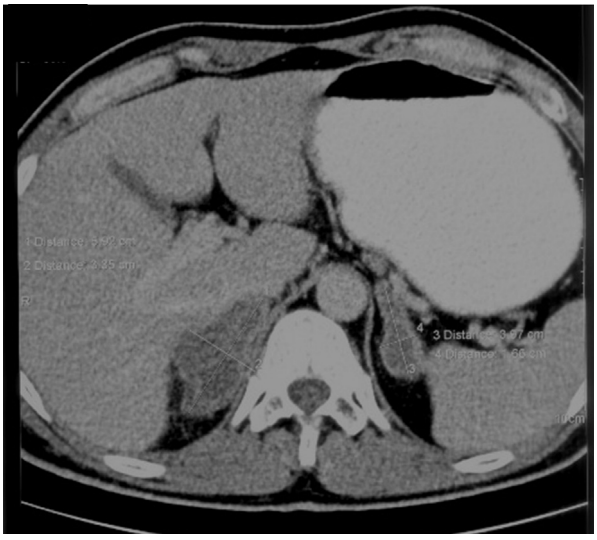

Fig. 1.

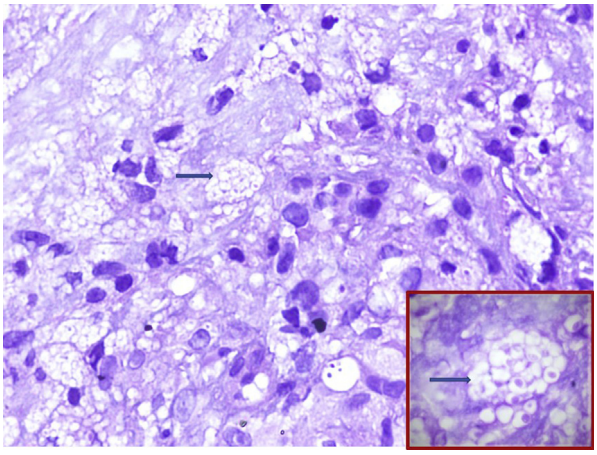

Fig. 2.

Abbreviations: ACTH, adrenocorticotropic hormone; CT, computed tomography.

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https://doi.org/10.1016/j.aace.2021.03.006

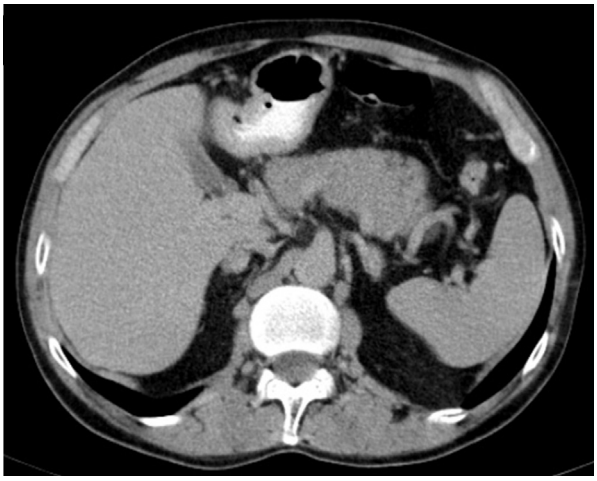

Fig. 3.

What is the diagnosis?

Answer

Primary adrenal insufficiency. Probable bilateral adrenal histoplasmosis.

The weight loss, hyperpigmentation and normalization of blood pressure were suggestive of primary adrenal insufficiency. This was confirmed biochemically with the presence of hyponatremia, hyperkalemia, and hypocortisolemia with an elevated plasma ACTH level. CT of the abdomen (Fig. 1) revealed homogenous enlargement of bilateral adrenal glands. A biopsy of the right adrenal gland revealed necrotizing granulomatous inflammation with colonies of encapsulated and budding yeast forms and pseudohyphae suggestive of histoplasmosis (Fig. 3). Culture from the biopsy specimen confirmed histoplasma capsulatum. Acid-fast bacilli culture was negative. The patient was started on prednisolone (5 mg/day), fludrocortisone (50 mcg/day), and itraconazole 200 mg twice a day. On follow-up at 12 months, the patient regained the lost weight and imaging revealed a marked decrease in the size of adrenal mass, though it was still not normal when compared to the crus of the diaphragm. (Fig. 2) The patient was continued on antifungal therapy and steroid replacements and asked to review after 6 months. Histoplasmosis is an infective condition caused by a dimorphic, saprophytic fungus. Adrenal involvement is seen in disseminated disease, but sometimes it may be the only site of demonstrable disease.[1] The differential diagnoses of bilateral adrenal enlargement are pheochromocytoma, metastasis, lymphoma, sarcoidosis, and infections such as histoplasmosis, tuberculosis, coccidioidomycosis, and blastomycosis.[2] Treatment of histoplasmosis involves systemic antifungal therapy; ketoconazole or itraconazole for mild to moderate forms and amphotericin B for severe infections.[3] Though the duration of antifungal therapy is about 12 to 18 months in different cohorts, there is a high relapse rate of about 7.5%.[4] However, most patients require long-term glucocorticoid replacement.[4]

Disclosure

The authors have no multiplicity of interest to disclose.

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