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Case Report

Bilateral Adrenal Histoplasmosis presenting with Primary Adrenal Insufficiency in a tertiary care hospital of East India: An incidental finding reshaping the appropriate therapeutic guideline

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Abstract
Histoplasma capsulatum is a dimorphic fungus causing opportunistic infections in immunocompromised host. Here we present a case of bilateral adrenal histoplasmosis in a 55 years old male, who presented with features of primary chronic adrenal insufficiency.
The patient presented with asthenia and weight loss with persistent hyponatremia and hyperkalemia with a previous history of hospitalization with features of fever, vomiting, abdominal pain and shock. Plasma cortisol level was low with high plasma ACTH; and failure to rise of cortisol level after ACTH challenge test confirmed Primary Adrenal Insufficiency. FNAC from bilaterally enlarged adrenal revealed intracytoplasmic yeast forms of capsulated fungus morphologically diagnosed as Histoplasma.
An overall diagnosis of Primary adrenal Insufficiency resulting from bilateral adrenal Histoplasmosis was made; which prompted initiation of a definitive therapy using systemic antifungals and steroid replacement.
Keywords: *Adrenal Insufficiency, Histoplasmosis in India.*

Introduction

Histoplasma capsulatum is a dimorphic fungus causing opportunistic infections in immunocompromised host. Here, we present a case of bilateral adrenal histoplasmosis in a 55 years old male, who presented with features of primary chronic adrenal insufficiency in the out patients' department of our institution. Incidental discovery of fungal etiology during the review of fine needle aspiration cytology (FNAC) slides from adrenal; a totally unexpected diagnosis in Indian scenario; led to the discovery of the cause of adrenal insufficiency- hitherto obscure to the clinician. The diagnosis was crucial and ingenious for framing appropriate therapeutic intervention for this patient.

Case Report

A 55 year old male, previously non diabetic, non hypertensive, presented with features of asthenia and significant weight loss in the last 3 months. History revealed that he was admitted to the

emergency in a peripheral government hospital few days back; with features of shock, vomiting, abdominal pain and fever; was managed conservatively and then discharged. Clinical examination revealed, the patient is mildly anemic with blood pressure being 90/64 mm Hg and all other routine investigations were apparently normal except persistent marked hyponatremia and hyperkalemia. Serology for HIV was non reactive, no evidence of tuberculosis found (either pulmonary or extra-pulmonary) even after detailed work up. Neither there was any evidence of occult malignancy, except bilateral enlargement of adrenal glands which were suspicious for adrenal SOL. An adrenal FNAC was planned, and within this period, hormonal assays were sent. The report revealed a low level of plasma cortisol, with high plasma ACTH; failure in response to ACTH stimulation test confirmed the diagnosis of Primary adrenal Insufficiency. The previous episode of emergency was explained as a case of Addisonian crisis in the moment of increased stress.

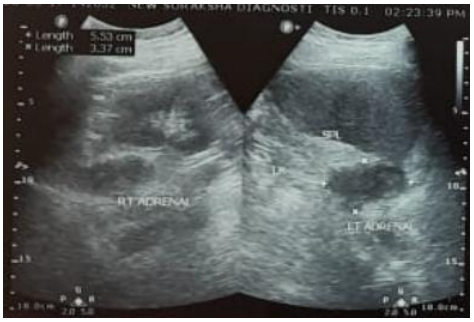

Fig 1: Ultrasonography finding showing bilateral adrenal enlargement

The USG guided FNAC procedure was performed in a outside Diagnostic centre and a vague report of Acute on chronic organizing inflammation was offered. The slides were sent for review in our department.

The cytology smear was pauci-cellular compising round to oval epithelial cells arranged in acini as well as in discrete fashion admixed with inflammatory cells in a background of blood elements. No granuloma or atypical cell suggestive of metastasis was noted. On closer examination, multiple inclusion bodies were noted within the cytoplasm as well as scattered in the background. Under oil immersion objective, the yeast forms of a capsulated fungus were observed-morphologically consistent with the diagnosis of Histoplasma capsulatum. Serolgical tests and culture were advised for corroboration.

The discovery in this case changed the entire treatment modality. First of all, it confirmed that there is actually no SOL, it is merely the hyperplasia of bilateral adrenal glands in response to increased ACTH. It also paved the way of a definitive chance of cure for the patient with initiation of systemic anti fungal treatment-otherwise guided solely by the previous report, the patient was destined to land in a blind alley of uncertainity.

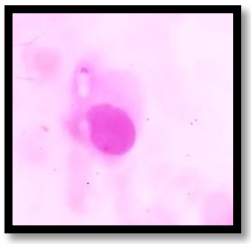

Fig 2

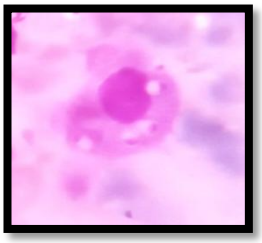

Fig 3

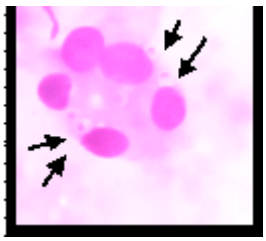

Fig 4

Fig 2,3,4: Photomicrographs showing fungal bodies of Histoplasma capsulatum within cell cytoplasm, magnification: 1000x, Leishman-Giemsa.

Discussion

Initially discovered in 1905 by Samuel T Darling, Histoplasma is endemic in the states bordering the ohio rever and lower missisipi valley. The reservoir being soil, infective microconidia get aerosolised and may spread via bat and bird droppings. In contray to normal belief, Histoplasma infections are not very uncommon in India particularly in the Gangetic plain and West Bengal[1]. In fact, in the review article by Randhawa et al largest number of cases in India are reported from West Bengal[2].

Histoplasma is an opportunistic dimorphic fungus, recognised by 3-5 micron intracellular yeast forms with characteristic narrow based budding and can be cultured in Saboraud's Dextrose Agar. The primitive Histoplasmin test is rarely used now-a-days. Various tests based on serological detection of Histoplasma antigen or antibody relying upon ELISA technique is now available. Serological diagnosis is mainly based on the identification of anti H and anti M antibodies and can be performed using complement fixation or immunodiffusion. Molecular diagnosis based on Nested PCR, though has been developed recently, is not available in most clinical establishments.

Three known forms of diseases caused by Histoplasma sp are known. The common pulmonary form, the disseminated form and a primary cutaneous form. Adrenal involvement by Histoplasmosis is rare, and most of the cases reported so far, showed bilateral involvement. In contray to our previous belief, publications are there reporting occurrences of disseminated Histoplasmosis in otherwise immunocompetent individuals[3].

Vyas et al[4] reported two cases of bilateral adrenomegaly diagnosed as adrenal histoplasmois by fine needle aspiration from PGIMER, Chandigarh. Similar case reports are there from different corners of the world[5,6] as well as from India itself[7,8]. Randhawa et al[2], in particular, is pointing towards the trend of increased incidence of Histoplasmosis in India in recent days after performing extensive literature review.

Commonly the patients are treated with Liposomal Amphotericin B and Itraconazole, but the prognosis is not always good in case of disseminated histoplasmosis particularly involving adrenals.

In our case, the therapy has been started just few days before;- we are eager to follow up the case for later developments and hopeful towards a good clinical outcome.

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