Case Report

Adrenal histoplasmosis in an immunocompetant Individual– a rare case report

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Abstract

Introduction: Disseminated histoplasmosis is common particularly among immunocompromised individuals, alcoholics and extremes of age. It is of concern because of the associated high morbidity and mortality. Asymptomatic adrenal involvement has also been described in 30 to 50% of patients with disseminated histoplasmosis. We report a case of adrenal histoplasmosis in an immune-competent subject presenting with adrenal insufficiency. **Case Report:** A 49 years old Indian male presented with three months history of weakness, nausea and vomiting, mucocutaneous pigmentation and weight loss. Past medical history was significant for tuberculosis, for which he was incompletely treated. He was non-smoker, non-alcoholic and non-diabetic, with no history of sexual promiscuity. USG of abdomen revealed bilateral suprarenal mass, with right adrenal (48.3x 39.3mm) and left adrenal measuring (42.4 x 38mm). CECT scan of upper abdomen showed enlargement of both adrenal glands with right adrenal 44x25x43mm, left adrenal measuring 53x37x46 mm suggestive of adrenal adenoma. CT guided FNAC of right adrenal mass showed necrosis, many budding yeast cells having the morphology of Histoplasma capsulatum. The patient was treated with liposomal form of amphotericin– B 150 mg / day for two weeks followed by oral itraconazole 200mg BD for one year and hydrocortisone 25 mg/day in divided doses. After one month, there was gain in weight, pigmentation faded and nausea & vomiting disappeared. **Conclusion:** Adrenal histoplasmosis usually occurs in a setting of immunocompromised condition and in endemic area, but it does occur in immunocompetant hosts from non-endemic area, as in our case. It is prudent to obtain a cytological or histopathological examination to confirm the diagnosis so that appropriate treatment can be instituted to avoid fatal complications.

Keywords: Adrenal histoplasmosis, Non-immunocompromised, Disseminated histoplasmosis

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Introduction

Histoplasmosis is a fungal infection caused by Histoplasma capsulatum [1]. Two major risk factors for developing the disease normally the first is working in high risk occupation and second risk factor is having a compromised immune system. High risk jobs include-construction worker, farmer, pest controle worker, demolition worker, roofer, land scaper. Conditions associated with weakened immunity include-being very young or very old, having HIV or AIDS, taking strong anti-inflamatory medications like corticosteroids, undergoing chemotherapy for cancer, taking TNF inhibitors for conditions such as rheumatoid arthritis, taking immunosuppressant drugs to prevent a transplant rejection. Disseminated histoplasmosis is common particularly among immunocompromised individuals,

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alcoholics and extremes of age and it is of concern because of the associated high morbidity and mortality [1]. India is considered to be a non-endemic area for histoplasmosis. Nevertheless, the first case of histoplasmosis being reported by Panja and Sen (1994) from India [2]. Since then many cases of disseminated histoplasmosis have been reported mostly from the eastern parts of the country [3]. Asymptomatic adrenal involvement has also been described in 30 to 50% of patients with disseminated histoplasmosis [4]. Here we report a case of adrenal histoplasmosis in an immune-competent subject who had adrenal insufficiency at presentation.

Case Report

A 49 years old Indian male presented to us with a 3 months history of weakness, nausea and vomiting, mucocutaneous pigmentation and weight loss. Patient gives history of tuberculosis in the past for which he was incompletely treated with four drug ATD for a month from DOTS. He is non-smoker, non-alcoholic and non-diabetic; there was no history of sexual promiscuity.

On physical examination, the patient appeared ill. His supine BP was 90/70 mmHg and standing position BP was 76/60 mm of Hg without any postural drop. Axillary temperature was 37ºc, weight was 54 kg, height was 164 cm, and BMI was 20.14 kg /m² and blackish pigmentation of oral cavity, knuckle, palmer creese and both legs.

Blood reports revealed haemoglobin-12.3gm%, WBC- 6500/³mm, platelet-1.8 lac/³mm, paired 8 AM serum cortisol- 2.15 µg/dl and 8AM ACTH - 561pg /ml which is suggestive of primary adrenal insufficiency. Other biochemistry reports demonstrated plasma renin activity 3.1 ng/ml/hr in supine (high normal), serum aldosterone 1.85ng/dl in supine (in low normal range), urea 32mg/dl, creatinine 1.13 mg/dl, serum potassium 5 mmol/l, blood for sodium 124 mmol/l, FBS 70.2 mg/dl, PPBS 105 mg/dl, blood for HIV serology was non reactive. Investigation for tuberculosis included Mantoux test-15mm induration after 72 hours, sputum for AFB for 2 days – negative, chest X-ray PA view – no abnormality detected. USG of the abdomen revealed bilateral suprarenal mass, with right adrenal measuring (48.3x 39.3mm) and left adrenal measuring (42.4 x 38mm). CECT scan of upper abdomen showed enlargement of both adrenal glands with right adrenal measuring 44x25x43mm and left adrenal measuring 53x37x46 mm suggestive of adrenal adenoma. CT guided FNAC from right adrenal mass showed mainly necrosis, many budding yeast cells having the morphology of *Histoplasma Capsulatum*. ECG demonstrated low voltage complex.

The patient was treated with liposomal form of amphotericin– B 150 mg / day for two weeks followed by oral itraconazole 200mg BD for 1 year and hydrocortisone 25 mg/day in divided doses. During follow up visit after one month in endocrine OPD, pigmentation begins to fade, patient gain weight, nausea vomiting disappear.

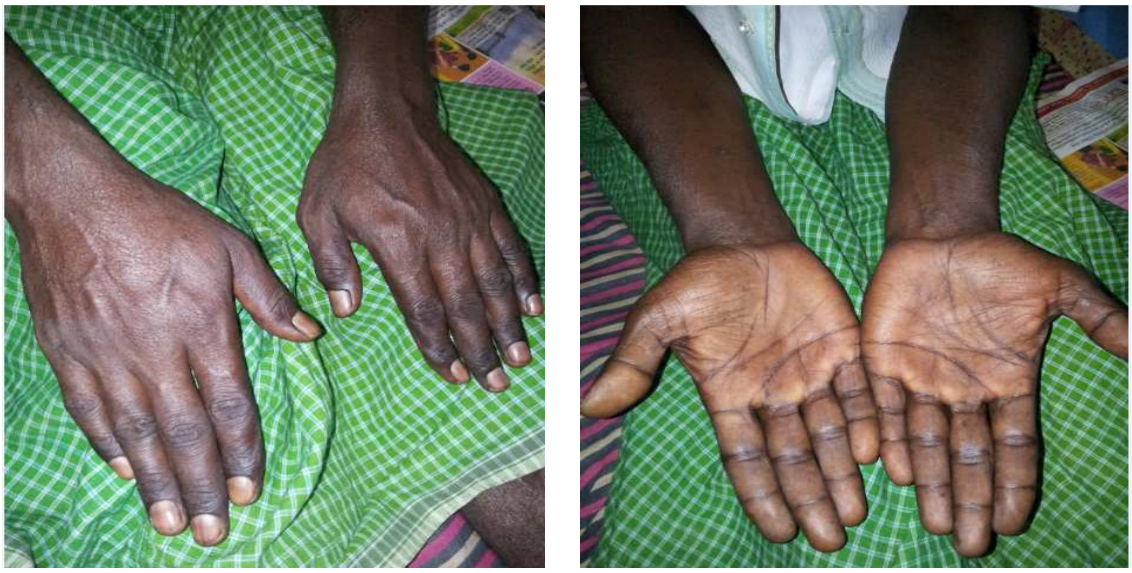

Figure-1: Blackish pigmentation of oral cavity, knuckle, palmer creese

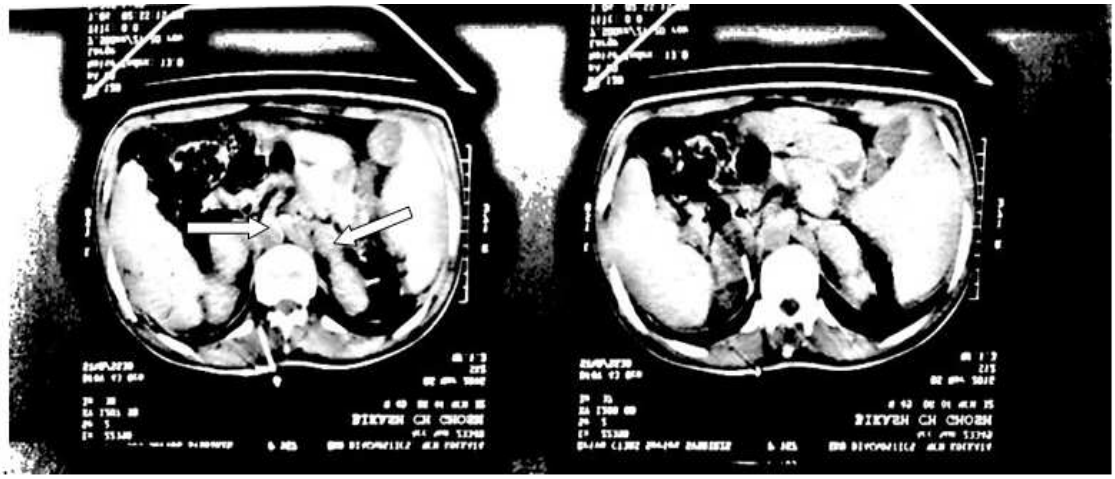

Figure-2: CECT of adrenal showing bilateral heterogenous enlargement

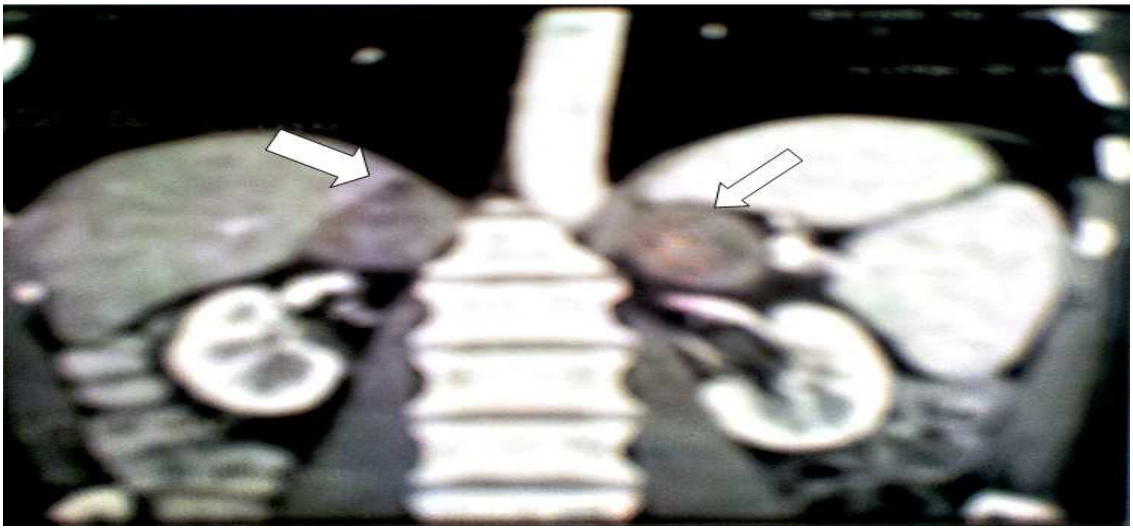

Figure-3: Showing bilateral adrenal enlargement

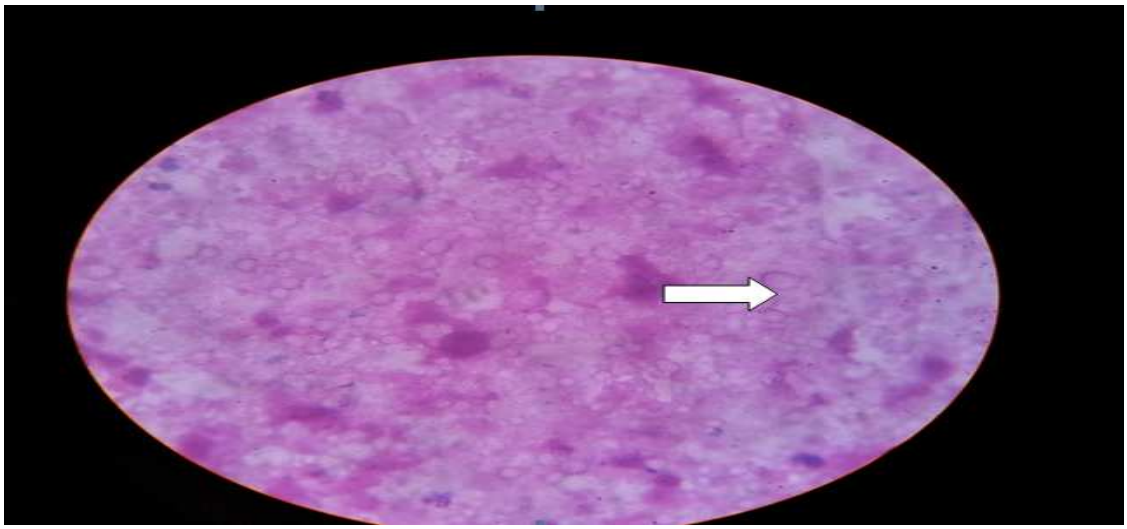

Figure-4: Adrenal histology showed mainly necrosis, many budding yeast cells having the morphology of histoplasma capsulatum.

Discussion

This case of middle aged male, complaining of weight loss, vomiting, hyper pigmentation, fatigue suggestive of primary adrenal insufficiency. CECT of abdomen revealed bilateral adrenal enlargements. The differentials would be benign or malignant adrenal tumors, metastatic tumor, subacute adrenal hemorrhage, disseminated infections such as tuberculosis, histoplasmosis, cryptococcosis, coccidioidomycosis, and paracoccidiodomycosis [4, 5, 6, and 7] and tuberculosis being the commonest. In Brazilian series of 131, 466 post mortem examinations there were 254 cases of adrenalitis of which 43.7% were caused by tuberculosis, 33.8% by paracoccidomycosis and 1.2% by histoplasmosis [8]. CT features of adrenal histoplasmosis vary depending on the stage of the disease. Typical features include bilateral symmetrical enlargement of adrenal gland with preservation of normal outline, peripheral enhancement and central hypodense area with calcification seen in healing phase [9]. Other infectious causes namely para-coccidomycosis, is indistinguishable from histo-plasmosis on imaging. Furthermore metastasis may mimic infection because central necrosis is common in both the condition which is a challenging entity for a clinician [10, 11]. Percutaneous biopsy or FNAC using either CT or USG guidance is necessary for evaluating adrenal lesion, which exhibit typical microscopic features of histoplasma capsulatum. Histoplasma capsulatum is an intracellular dimorphic fungus which is usually seen in the macrophages cytoplasm and exhibits narrow based budding [12, 13].

The mortality in untreated disseminated histoplasmosis patient was as high as 80-100%, but if treated with antifungal these very high mortality rate is remarkably reduced to less than 25% [14]. In our patient primary antifungal therapy instituted was liposomal amphotericin B 3mg/kg body weight for two weeks followed by oral itraconazole 200 mg BD for 1year. Treatment duration of 1 to 2 years is recommended to reduce recurrence [15].

Conclusion

Primary adrenal insufficiency due to histoplasmosis in India is rare but encountered in clinical practice. Usually Adrenal histoplasmosis do occur in a setting of immunocompromised condition and in endemic area, but it does occur in immunocompetant hosts from non-endemic area, as in our case. It is prudent to obtain a cytological or histopathological examination to confirm the diagnosis so that appropriate treatment can be instituted to avoid fatal complications.

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