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DISSEMINATED (LINGUAL AND ADRENAL) HISTOPLASMOSIS IN IMMUNOCOMPETANT HOST: AN UNCOMMON PRESENTATION.

Dermatology

Dr Amisha Kukreja JR Dept of DVL Dr RP Govt Medical College Tanda HP
Dr Sujaya Manvi SR Dept of DVL Dr RP Govt Medical College Tanda HP
Dr K S Mehta* Prof & Head Dept of DVL Dr RP Govt Medical College Tanda HP *Corresponding Author
Dr P S Chauhan Assoc Professor Dept of DVL Dr RP Govt Medical College Tanda HP
Dr Sheenam Hooda JR Dept of DVL Dr RP Govt Medical College Tanda HP

ABSTRACT

Histoplasmosis is a systemic fungal infection caused by dimorphic fungus, Histoplasma capsulatum presenting most commonly as pulmonary, primary cutaneous and progressive disseminated (PDH) forms. Disseminated histoplasmosis is a rare presentation most frequently documented in immunocompromised individuals such as those with acquired immunodeficiency syndrome. A case of disseminated lingual and adrenal histoplasmosis in an immunocompetant middle age man is reported here who initially presented with non specific symptoms of fever, weight loss and headache and later on developed oral lesions.

KEYWORDS

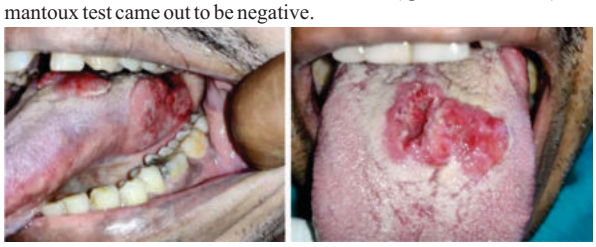
Oral Histoplasmosis, H.capsulatum, Adrenal involvement, Immunocompetent host.

INTRODUCTION

Histoplasmosis is an infective condition caused by a dimorphic, saprophytic fungus, *Histoplasma capsulatum* and is acquired by inhalation of its spores. Soil rich in bird and bat dropping is its natural habitat, and it exists as a mycelium in the atmosphere. The fungus enters the body through the respiratory tract in the form of microconidia, which are transformed into intracellular yeast-like structures in the lungs before disseminating hematogenously. Primary infection is usually asymptomatic and self-resolving. Immunosuppressed patients, mainly those with altered cellular immunity, may have disseminated disease with variable mucocutaneous involvement characterized by papules, nodules, gummas, or ulcers with a granulomatous base [1]. Dissemination occurs frequently to liver, spleen, lymph node, bone marrow and adrenals. Adrenal involvement is usually a sequelae of previous infection [2]. Despite being a rare infection in immunosupressed individuals, it is even rarer in immunocompetant hosts.

Case Report

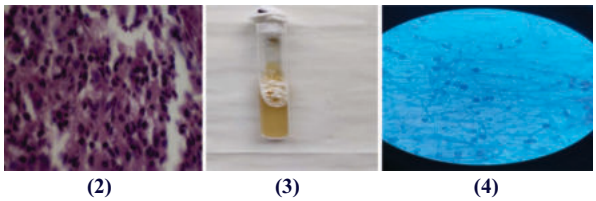
A 46-year old-HIV-negative man, resident of Sirmaur district of Himachal Pradesh, electrician by occupation, presented to dermatology outpatient department with painless oral ulcerative lesions over dorsum and lateral margin of tongue of 5 months duration. He was also having complaints of headache, cough, intermittent low grade fever, loss of appetite for 6 months and weight loss of about 3-4 kgs in last 2 months for which he was under evaluation in Medicine department. He was an occasional smoker, non alcoholic, non-diabetic and non hypertensive. Mucocutaneous examination showed two well defined, discrete ulcerative plaques with irregular borders and erythematous base with whitish slough at places, each over dorsum and left lateral margin of the tongue of size 7×4.5×0.2 cm and 5×4×0.2 cm respectively. The lesions were non tender and the surrounding area was normal. Laboratory investigations showed raised erythrocyte sedimentation rate of 48 mm in the first hour, normal hematological profile, hepatorenal tests and urinalysis. Chest x ray was normal and viral markers, acid fast bacilli stain, CBNAAT (sputum and tissue) and mantoux test came out to be negative.



(A) (B)

Fig.1: At baseline:Well defined erythematous plaques over (a) lateral aspect of tongue and (b) dorsum of tongue

Histopathological examination showed dense inflammatory infilterate of predominantly mononuclear cells with few eosinophils in the dermis with PAS positive intra- and extracellular tiny spores. Fungal culture showed growth of Histoplasma capsulatum.



(2) (3) (4)

Fig.2: Dermis: dense inflammatory infiltrate. PAS stain: PAS positive extracellular tiny spores (H n E)

Fig.3: Histoplasma capsulatum colonies on sabouraud dextrose agar incubated at 37°C showing white colonies with yellow tinge

Fig.4: Lacto phenol cotton blue (LPCB) mount showing numerous hyaline hyphae admixed with micro- and macro- conidia

Ultrasound abdomen was planned to rule out systemic involvement which showed diffusely hypoechoic enlarged bilateral adrenal glands. CT scan abdomen showed bilateral adrenal adenoma consistent with adrenal histoplasmosis. MRI abdomen further confirmed the diagnosis of adrenal histoplasmosis showing enlargement of both the adrenals with peripheral rim and septal enhancement.

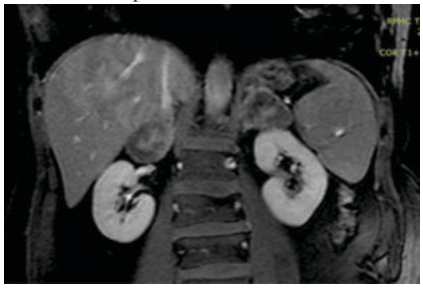


Fig.5: MRI abdomen: B/L enlarged adrenals with peripheral rim and septal enhancement

Serum ACTH levels were done in view of adrenal involvement and were found to be raised (432pg/ml; normal value<= 46pg/ml) but serum cortisol was normal.

Patient was initially started on oral Itraconazole 200mg twice daily and subsequently, in view of adrenal histoplasmosis, he was shifted to Amphoterecin B. A total of 9 injections of liposomal Amphoterecin B

in a dose of 3mg/kg were given on alternate days along with mineralocorticoid replacement. The lingual lesion responded completely by the end of 6 months and the patient improved systemically.

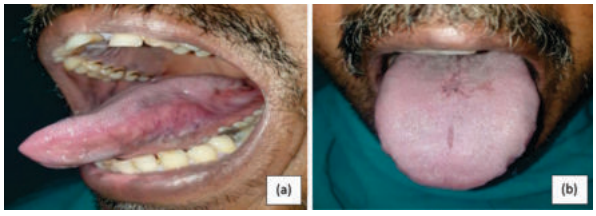


Fig.6: Complete clearance of lesions after 6 months of therapy (a)over lateral aspect of tongue and (b)dorsum aspect of tongue

DISCUSSION

Histoplasma capsulatum is a dimorphic fungi manifesting as mold form in soils and converts to yeast form when infecting a human host. Route of transmission is via inhalation of airborne spores. Soils contaminated by bat and bird droopings have always been linked to outbreaks of histoplasmosis, particularily if the environment is disturbed in form of man-made construction or natural processes. The rich soils of the Ohio and Mississippi rivers valleys are endemic with the fungi but histoplasmosis is not limited to the United States and can be found in parts of Central and South America, Africa, Asia, and Australia[3]. In India, histoplasmosis has been mostly reported from the regions of West Bengal and Assam, the Gangetic Plains, western parts of India and few southern and northern Indian states. Apart from some isolated cases, Himachal Pradesh is not endemic for histoplasmosis[4].

Immunocompetant individuals if infected generally do not show any signs or symptoms of infection. Besides the weakened immune status of the host, the severity of the disease may increase with large number of spores inhaled. After inhalation, these microconidia are easily able to travel down the repiratory tract to finally reach the alveoli. In alveoli, the mycelial is converted to yeast form and is the pathogenic form of *H. capsulatum*. It must evade innate immunity, to establish its niche within macrophages that act as carriers for dissemination of the disease to multiple organs. Organ affected are spleen, adrenal glands, lungs, liver, lymph nodes, gastrointestinal tract, central nervous system, kidneys, and oral mucosa[5].

Oral histoplasmosis is still a rare entity in Indian scenario and easily misdiagnosed especially in an immunocompetant host. The oral lesions of disseminated histoplasmosis are most frequently found on the tongue, palate, and buccal mucosa but can occur at other sites also. Disseminated histoplasmosis frequently (80 %) affects the adrenal glands, however hypoadrenalism is rarely reported. It involves both the adrenal glands with patients most commonly presenting with bilateral adrenal mass[6].

The clinical differential diagnosis of a lesion of oral histoplasmosis is broad ranging from benign entities such as non-specific ulcers to malignancy. Main differentials to be kept in mind are tuberculosis, leishmaniasis and bacterial infections including sexual transmitted infections (*Neisseria gonorrhea* and *Treponema pallidum*). Other fungal diseases such as candidiasis, blastomycosis and cryptococcosis also need to be considered and can be differentiated from one another by using special stains. Fungal culture remains the gold standard. *Histoplasma capsulatum* is classically described as elliptical, narrow based uninucleate budding yeast forms 2–4 μm in diameter[7].

This case here is distinct in itself showing dissimilarities from the previous reported cases of histoplasmosis in terms of patient's apparently well immune status. Also, the lesions in our patient were painless in contrast to other reported cases. Another interesting point in our case is the initial presentation of the patient with symptoms of headache, weight loss, loss of appetite, chronic cough and intermittent low grade fever creating clinical confusion with tuberculosis in Indian setup. As the signs and symptoms of adrenal involvement manifested almost a month earlier than the appearance of lingual lesion, it can cause delay in diagnosis and treatment which can further lead to catastrophic adrenal insufficiency.

Declaration Of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands the names and initials will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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