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Case Report

Oral Histoplasmosis Masquerading as Squamous Cell Carcinoma

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Abstract

Histoplasmosis is a systemic, deep mycotic infection caused by the dimorphic fungus, *Histoplasma capsulatum*. Systemic histoplasmosis has emerged as an important opportunistic infection in human immunodeficiency virus (HIV) patients and those in endemic areas. Reported cases of histoplasmosis have been low in India with less than 50 cases being reported. We are reporting a case of disseminated histoplasmosis with isolated oral involvement in an HIV seronegative patient. Treatment with oral itraconazole led to remission of the oral lesions.

Keywords: Oral Histoplasmosis; Histoplasma capsulatum; Oral Lesions

Introduction

Histoplasmosis is a granulomatous systemic mycotic infection caused by thermal dimorphic fungus, *Histoplasma capsulatum*. It is also known as Darlings disease as Samuel Darling while working in the Canal Zone in Panama in 1905 found it out. He first described its clinical disseminated disease form in a fatal case from Martinique after 30 years of finding it [1].

Histoplasmosis is endemic in the central eastern United States, especially the Ohio and Mississippi River Valleys, in Central and South America, and Africa, but is less frequently reported in Asia and Europe [2,3].

Reported cases of histoplasmosis have been low in India with less than 50 cases being reported [4].

Histoplasma develops as a branching hyphal form in the soil and a yeast form in the host tissue [5]. The organism is commonly found in warm, humid environment that contains bird and bat excreta. H. capsulatum is acquired by inhalation of airborne spores from mycelial fragments of the fungus.

Histoplasmosis may occur in three forms: (i) Primary acute pulmonary form, (ii) chronic pulmonary and (iii) disseminated form(DH) [6]. Occurrence of disseminated form of histoplasmosis is rare in immunocompetent individual though in immunocompromised and HIV positive patients, 95% of histoplasmosis appears as disseminated infection [3].

The oral lesions may occur in any part of the oral cavity and the lesions vary from nodules to painful shallow or deep ulcers [3]. Around 30 - 66% of patients with DH have oral lesions, frequently presenting as the initial sign. Generally, lesions of histoplasmosis in the oral cavity are the local manifestation of pulmonary or disseminated disease [7]. but rarely may they be the primary or even the only manifestation of the disease [8].

In India, histoplasmosis is rare and is endemic only in small regions in West Bengal and Maharashtra [9,10].

We hereby present a case of oral histoplasmosis from the state of Bihar in an otherwise immunocompetent, HIV negative young male without other subjacent underlying disease, with lesions exclusively in the bilateral retromolar trigone area of oral cavity. Bihar is a non-endemic region for histoplasmosis in India.

Case Report

The patients complained of symptoms such as occasional fever, asthenia, lassitude, lethargy, weight loss, some difficulty in speech and odynophagia for more than 2 months. On examination, the oral lesions appear as nodular ulcerative lesions of size approximately 2 cm × 2 cm that were painful, localized bilaterally in the retromolar trigone area of buccal mucosa. The ulcers have raised borders covered by a reddish yellow ulcerative necrotic membrane, resembling aphthous ulcers, traumatic ulcers, carcinoma or tuberculosis.

The ulcer did not bleed and showed no discharge. The lesions were initially budding, papule like and then developed gradually into ulcerating, indurated and painful lesions. The histopathology features were also consistent with those of a chronic specific infection characterized by infiltration of predominantly lymphocytes, formation of the ill formed granulomas composed of histiocytes, containing variable numbers of yeast-like oval structures, measuring 3 - 5µm in diameter morphologically looking like - confirmed

by PAS stain. On physical examination, no lymphadenopathy or organomegaly was found.

On systemic examination, no any relevant finding could be elicited. The patient does not report any risk factor for histoplasmosis. There was no history of oral trauma. He was non-alcoholic, non-smoker, non-diabetic and no past history for tuberculosis. He did not report a history of travelling and never went out of the local district.

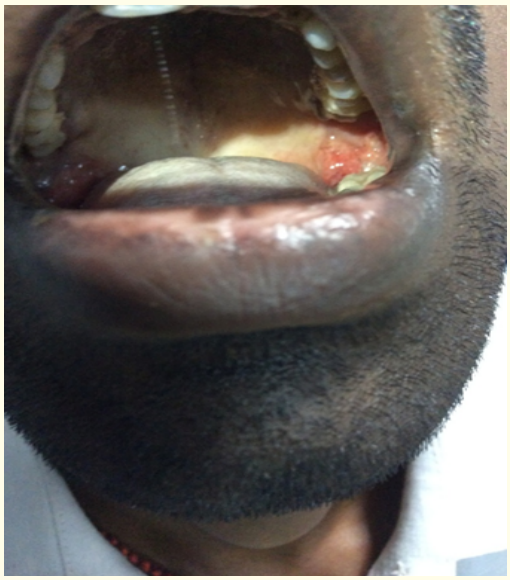

Figure 1: Gross Photograph of the Patient's Oral Ulcer Before (A) and after Treatment.

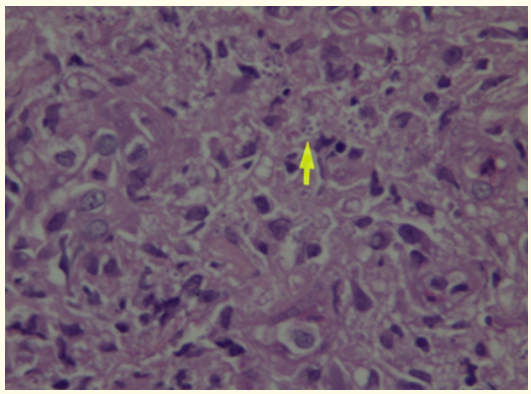

Figure 2: Microphotograph Showing Inflammatory Cell Infiltrate and Macrophages Containing the Yeast form of H.Capsulatum (H&E X 40).

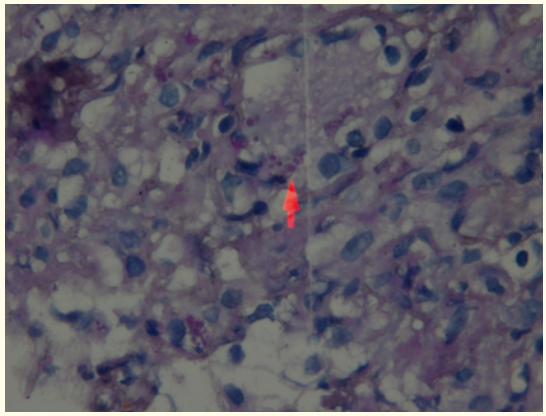

Figure 3: Yeast-Like Cells were Found when Stained with Periodic Acid-Schiff Stain (PAS X100).

Discussion

Histoplasmosis infection occurs through the inhalation of conidial forms present in the environment. The fungus is dimorphic intracellular, parasitizing the reticule-endothelial system and involving many organs like spleen, liver, kidney, central nervous system [11,12]. It is primarily a pulmonary disease, and the environmental reservoir is soil. There are two varieties of *H. capsulatum* that are pathogenic to humans, *H. capsulatum* var. capsulatum and *H. capsulatum* var. duboisii, and a third variety that is an equine pathogen, *H. capsulatum* var. farciminosum, which exists in Africa [13].

Oral lesions of histoplasmosis can manifest in a variety of forms as ulcerative, nodular, verrucous, vegetative, granulomatous, and plaque-like lesions involving the lips, gingiva, tongue, and palate [14,15,16].

Frequently, in cases of disseminated histoplasmosis, oral lesions appear as the initial clinical manifestation. Since 1946 only a few cases of apparently initial mucocutaneous histoplasmosis in patients without detectable systemic involvement have been reported [17]. According to one school of thought oral lesions result of hematogenous spread from an unknown infectious focus, and according to another school of thought primary oral lesions can occur by direct inoculation of the fungus in the mucosa [18,19].

Some investigators suggested that, when no systemic sign or symptom can be detected, oral histoplasmosis should be considered a localized disease, and treated as such (i) Clinical mani-

festations of histoplasmosis range from asymptomatic lesions to life-threatening disseminated infections, although the disease is self-limiting in immunocompetent patients [20].

Azole antifungal drugs like ketoconazole 200 mg tablet and itraconazole 100 mg capsule have off late been the drugs of choice, in most healthy and non-HIV infected Patients [21]. In the present case itraconazole 200 mg once a day has brought about complete clinical remission of the lesion in just 10 days, though medicine is to be continued for one month and patient has been suggested to be in follow up for at least 9 months. The patient's haematological values along with liver and renal function test were within the normal limits; thus, showing the drug was well tolerated. Facility for serum itraconazole assessment is not available in our set up, so it was not done.

The gold standard for the diagnosis is the isolation of the *H. capsulatum* in culture with observation of the conidia, as well as its conversion to yeast forms at 37°C. Diagnosis is usually made by a combination of culture, detection of the organism in tissues, measurement of antibodies, and detection of antigen.

Conclusion

Many of the patients do not have any identifiable risk factors for Histoplasmosis particularly in non-endemic area. Non-healing ulcer in oral region should not only be looked upon as malignant lesion rather infectious ethology like tuberculosis and histoplasmosis should also be kept in differential diagnosis.

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