

## Case Report

### Orofacial histoplasmosis in a patient with AIDS: a case report

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#### ABSTRACT

Histoplasmosis, also known as Darling's disease, is a fungal infection caused by *Histoplasma capsulatum* and is common among patients with AIDS and is mainly found in North, Central and South America and sub-Saharan Africa. It manifests as pulmonary, progressive disseminated histoplasmosis involving reticuloendothelial system, liver, spleen, lymph node, bone marrow, adrenal glands and primary cutaneous histoplasmosis. In HIV infection, 10% cases of histoplasmosis have cutaneous manifestations. Only 12 reports of AIDS associated histoplasmosis are described in the Indian literature. In this report, we describe an additional case of histoplasmosis in 19-year-old male in Kutch district of Gujarat state.

**Keywords:** Histoplasmosis, Oral, Cutaneous, HIV, AIDS.

#### INTRODUCTION

Histoplasmosis, a fungal infection caused by *Histoplasma capsulatum* is also known as Darling's disease.<sup>[1,2]</sup> It is endemic in the central eastern united states, especially the Ohio and Mississippi River Valleys.<sup>[3,4,5]</sup> In India, the first case of histoplasmosis was reported in 1954, thereafter from 1968 to 1992 only 25 sporadic cases have been reported.<sup>[4]</sup> A majority of cases have been reported from the eastern parts of the country, especially along the belt of Ganges and Brahmaputra. Due to migration and increased urbanization, cases are being reported from all over the country. *Histoplasma* grows in soil enriched with bird droppings, reaches human alveoli through inhalation, and can cause acute or chronic pulmonary infection to progressive disseminated histoplasmosis.<sup>[6]</sup> Oral histoplasmosis usually occurs in association with the chronic disseminated form of

the disease.<sup>[7]</sup> Disseminated histoplasmosis is the most common form associated with AIDS.

#### CASE HISTORY

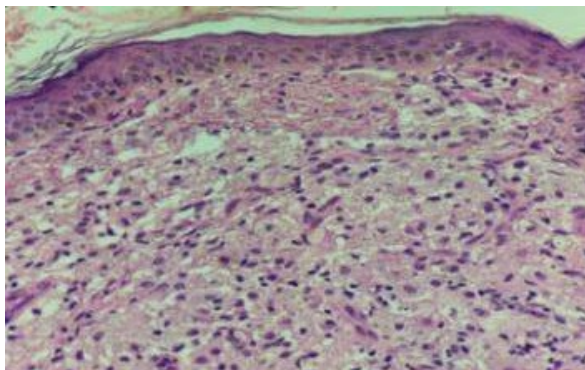
A 33-year-old male presented with multiple ulcers in oral cavity. The onset of the eruptions was sudden and gradually progressive. Clinically patient had multiple, well defined, erythematous nodules on soft palate, face and lips. [Figure 1] On general examination, he was moderately built and poorly nourished. His vital signs were within normal limits. There was no lymphadenopathy. His complete blood count showed haemoglobin of 10.3gm/dl; WBC count: 6290/ $\mu$ L; differential count: N-79%, L-11%, E-02%, M-08%; absolute lymphocyte count 692/ $\mu$ L. Chest X-ray was normal. CT contrast of paranasal sinuses showed possibility of invasive fungal infection. The nasal swab and biopsy from

tonsils were negative for fungal organisms. The records of CD4 and CD8 counts and viral load are not available. A biopsy of the soft palate and skin over the face showed similar features comprising of proliferation of foamy histiocytes beneath the lining epithelium and presence of small, uniform, oval intracellular budding yeast within it. [Figure 2] Periodic acid Schiff stain showed pink capsular staining of intracellular budding yeast. [Figure 3] Gomori's Methenamine Silver stain showed black coloured intracellular fungal organisms. [Figure 4] The patient was started on highly active antiretroviral therapy (HAART) and Itraconazole (200mg BD) for 15 days. The oral and cutaneous lesions of the patient completely disappeared within 20 days of the treatment. [Figure5]

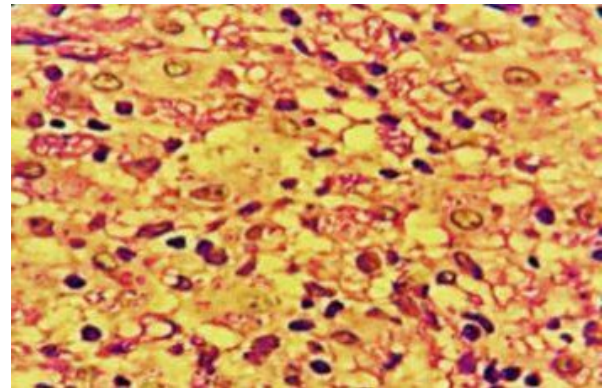
**Figure 1:** shows nodular umbilicated lesions over face and ulcers on soft palate and lips.



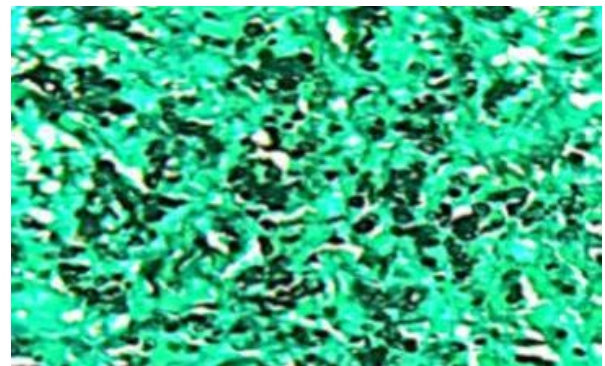
**Figure 2:** shows proliferation of histiocytes beneath the epidermis containing small, round to oval intracellular organisms with clear space around the organism. (H & E x 400)



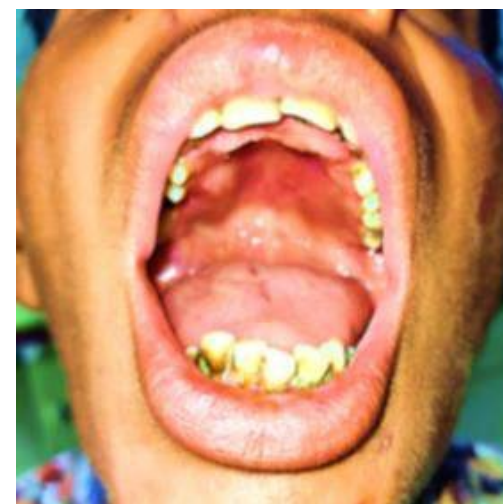
**Figure 3:** shows pink coloured capsular staining of *Histoplasma capsulatum*. (PAS x 1000)



**Figure 4:** shows black stained organisms in the histiocytes. (GMS x 1000)



**Figure 5:** shows disappearance of lesions over face, soft palate and lips after treatment.



**Table 1:** shows reported cases of Histoplasmosis in HIV infected patients in India.

Sr. No.	Author/Year and region of Publication/Ref. No.	No. of Cases	Age/Sex	Organs involved	Other manifestations
1	Singh T, 1996, Manipur <sup>[10]</sup>	1	F/38	Lymph node & Bone marrow	Lymphadenopathy, hepatosplenomegaly
2	Mohanchand R, 2000, Assam <sup>[8]</sup>	1	M/28	Palate, Bone marrow, LN	Lymphadenopathy, Hepatosplenomegaly
3	Patil K, 2003, Karnataka <sup>[11]</sup>	1	M/28	Palate	Lymphadenopathy
4	Joshi SA, 2006 Maharashtra <sup>[12]</sup>	1	M/45	Bone marrow & spleen	Hepatosplenomegaly
5	Wadhwa A, 2007, New Delhi <sup>[13]</sup>	1	No details	Lymph node	Lymphadenopathy, hepatosplenomegaly
6	Bhagwat P, 2009, Sirisi, Karnataka <sup>[4]</sup>	1	1 <sup>st</sup> - F/35	Face and abdomen	Lymphadenopathy, Hepatosplenomegaly
		1	2 <sup>nd</sup> - M/40	Tongue, buccal mucosa	Lymphadenopathy
7	Vishwanath S, 2009, Manipal, Karnataka <sup>[2]</sup>	1	F/38	Lymph node & Bone marrow	-
8	Patil K, 2009, Karnataka <sup>[7]</sup>	1	F/45	Tongue	Lymphadenopathy
9	Chande C, 2010, Maharashtra <sup>[3]</sup>	1	F/25	Face, tongue and buccal mucosa	Lymphadenopathy, hepatosplenomegaly
10	Mohammed S, 2012, Karnataka <sup>[9]</sup>	1	M/35	Tongue	Lymphadenopathy, consolidation lung
11	Tyagi R, 2016, Punjab (African male) <sup>[14]</sup>	1	M/20	Lymph node	Lymphadenopathy, hepatosplenomegaly

## DISCUSSION

Respiratory tract is the usual portal of entry for histoplasma and the lesions generally occur in lungs. The disseminated disease is the most commonly reported form in immunocompromised individuals.<sup>[3]</sup> It can involve the reticulo-endothelial system, intestine, skin and liver, and may present with leukopenia and anaemia. It is usually a late manifestation when associated with HIV infection and has an almost 20% mortality rate.<sup>[2]</sup> Skin lesions are usually the manifestations of disseminated histoplasmosis. The primary cutaneous histoplasmosis is very rare.<sup>[3]</sup> The endemic areas of Histoplasmosis include part of the USA, West Indies, Central and South America, Africa, India and the Far-East.<sup>[8]</sup> Studies from endemic areas have shown that 4% to 7% of HIV-infected patients present with disseminated histoplasmosis. Some studies have shown orofacial lesions in 66% patients with disseminated histoplasmosis.<sup>[9]</sup> In India, in the last two decades, histoplasmosis has been reported with increased frequency in patients with HIV.<sup>[3]</sup>

Literature search showed 12 reports of HIV associated histoplasmosis in India. [Table-1] The youngest patient was 20-year-old male<sup>[14]</sup> and oldest patient was 45-year-old male.<sup>[7,12]</sup> In 7/12 patients had orofacial histoplasmosis while other 5 cases had disseminated histoplasmosis (lymph nodes, bone marrow, spleen and skin). The present report is the first case of AIDS associated histoplasmosis from the Kutch district of Gujarat state. Even though prevalence of histoplasmosis is less in India, it should be considered in the differential diagnosis of oral and cutaneous lesions in patient with AIDS.

## CONCLUSION

Histoplasmosis should be considered in the differential diagnosis of an unusual, exaggerated, oral and cutaneous ulceration, when encountered in HIV infected patients. The histopathological diagnosis has implication on treatment of the patients with HIV/AIDS. Treatment with Amphotericin B or Itraconazole is effective however the mortality approaches 50% or more in those with multi organ failure.

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