

Primary Histoplasmosis of Oral Mucosa: A Rare Case Report

Neelu Gupta¹, Surendra Prakash Vyas², Dharm Chand Kothari³

1- Associate Professor and Head of department, Department of Pathology, Sardar Patel Medical College, Bikaner, Rajasthan, India. 2- Associate Professor, Department of Pathology, Sardar Patel Medical College, Bikaner, Rajasthan, India. 3- MD, Department of Pathology, Sardar Patel Medical College, Bikaner, Rajasthan, India.

Correspondence to:

Dr. Dharm chand Kothari, Room No.- 40, Hostel No.- 4, Vivek Hostel, UG Hostel Medical College Campus, Bikaner PIN- 34001, Rajasthan, India.

ABSTRACT

Primary cutaneous or mucosal histoplasmosis is a very rare disease and it is benign and self-limited in duration. Infection may follow inoculation of spores through skin and mucous membranes as in laboratory infection and presents as a chancriform syndrome, with a nodule or ulcer at the site of inoculation and associated lymphangitis and lymphadenitis. Its spores are infectious to humans. We report a case of histoplasmosis in a 80 year old immunocompetent male present with non-healing ulcer of buccal mucosa. We present this case because of primary histoplasmosis of oral mucosa was rare presenting sign of histoplasmosis.

KEYWORDS: Buccal Mucosa, Histoplasmosis, Non-healing ulcer

INTRODUCTION

A highly infectious mycosis caused by *Histoplasma capsulatum* and affecting primarily the lungs, where it is generally asymptomatic.¹ The fungus is dimorphic intracellular, parasitizing in the reticulo-endothelial system and involving the many organs like spleen, liver, kidney, central nervous system.^{2,7} The disease caused by *H. capsulatum* var. *capsulatum*, referred to here as histoplasmosis (small-form histoplasmosis), is widely distributed throughout the world, occurring in some temperate and tropical countries in the Americas, Africa and Australasia.^{1,3} Oral histoplasmosis usually occurs in association with the chronic disseminated form of the disease rarely it may present as the initial or the only mucocutaneous manifestation of the disease.⁴ We report this case because histoplasmosis rarely present as a solitary non-healing ulcer of oral mucosa.

CASE REPORT

80 year old male farmer from a village was visited to surgical OPD with the chief complains of painful non healing ulcer on right side of buccal mucosa and adjacent gingiva since 2 months. He notice pain during eating before two month and difficulty in mastication. Gradually he also face problem in speech and deglutination. No long term drug history was present. Personal, past and family history was not contributory. Also, there were no systemic symptom like pulmonary, diabetes or hypertension or any other immune mediated disease was present.

On examination an ulcer measuring 3x2x1 cm in size, was present on right side of buccal mucosa adjacent to the molar teeth. The ulcer was inflamed and margin was

indurated.

His routine investigation like Complete Blood Count, Urine biochemical and microscopy, liver and renal function test was normal. His haemoglobin was 12.5gm%, total leucocyte count 8500/mm³, platelet 160000/mm³, and DLC was Neutrophil 63% and lymphocyte 34%. His bleeding and clotting time was normal. Random blood glucose was 110 mg%. HIV, and VDRL was negative by ELISA method. X-ray of oral cavity shows there is some degree of mandibular bone destruction with soft tissue swelling and ulcer. X-ray of chest was normal.

So on the basis of clinical presentation a provisional diagnosis of squamous cell carcinoma was made. Incisional biopsy was taken from the ulcerative lesion and sent for histopathological examination to our department.

Microscopic Examination

Section from representative areas shows inflammatory cells like neutrophil lymphocytes with macrophages containing oval yeast like bodies (Figure-1). Yeast like bodies are present in macrophages at eccentric as well as centric position (Figure-2). Grocott silver methamine stain and PAS stain was positive (Figure -3,4).

Mucicarmine stain was done for *Cryptococcus* capsule which was negative. So on the basis of clinical, histopathological and special staining a diagnosis of primary histoplasmosis of oral mucosa was offered. We did not culture the fungus due to lack of facility.

After diagnosis of histoplasmosis antifungal drug

How to cite this article:

Gupta G, Vyas SP, Kothari DC. Primary Histoplasmosis of Oral Mucosa: A Rare Case Report. Int J Dent Med Res 2015;1(6):91-93.

itraconazole 400mg daily oral was given to the patient for fifteen days. After treatment patient was markedly improved. so treatment was continued for six week ulcer was completely healed. Treatment was stop after three months.

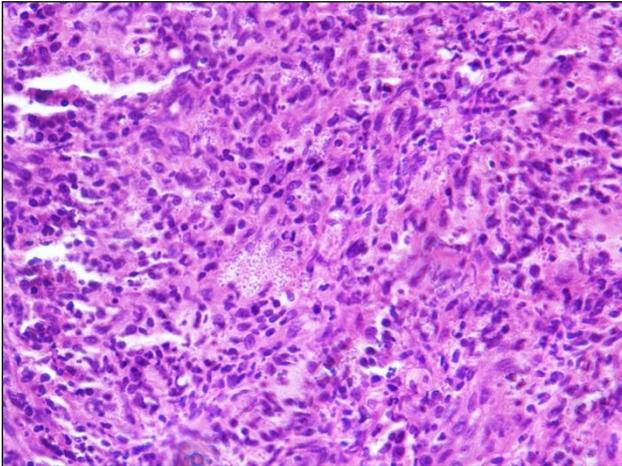


Figure 1- Section shows scattered inflammatory cells with macrophages containing oval yeast like bodies (H&E, 40x)

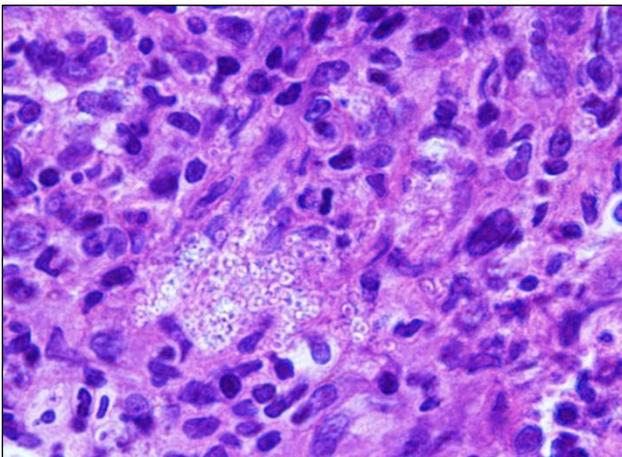


Figure 2- Section shows scattered inflammatory cells with macrophages containing oval yeast like bodies (H&E, oil immersion).

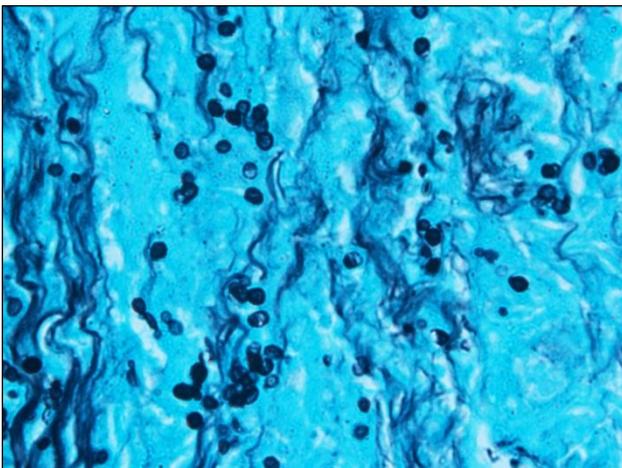


Figure 3- Section shows Grocott silver methamine stain macrophages containing oval yeast like bodies of Histoplasmosis (H&E, 40x).

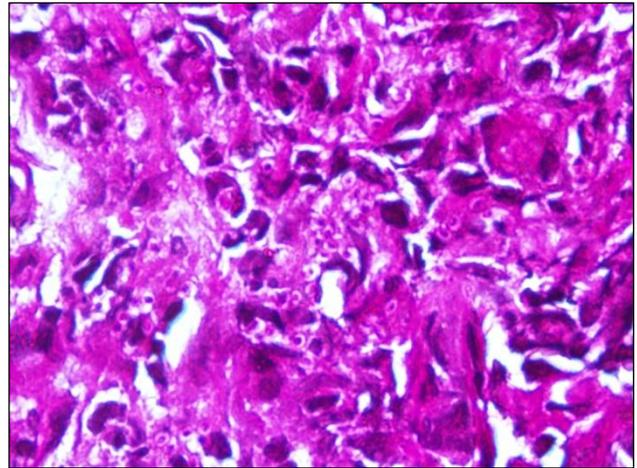


Figure 4- Section shows PAS stained oval yeast like bodies of Histoplasmosis (H&E, 40x).

DISCUSSION

Histoplasmosis, also called as Darling's disease or Histoplasmosis capsulati is caused by the dimorphic fungus, *H. capsulatum* is a granulomatous lesion.^{3,5,6}

Histoplasmosis is rarely reported in India, because of its various clinical presentation and lack of awareness among dermatologists but incidence of histoplasmosis are raising in India and it is the second most common opportunistic infection associated with HIV nowadays.^{4,5} Mucocutaneous histoplasmosis is common in AIDS patients or immunocompromised patient but it is rare in immunocompetent hosts.⁷

Two varieties of the fungus were differentiated—var. capsulatum, or the *H. var. Duboisii* on the basis of the yeast phase cell sizes.^{2,3,5}

Infants and children are frequently infected, and among adults the rate is highest in male agricultural workers. *H. capsulatum* exists as a saprophyte in nature and has often been isolated from soil particularly when contaminated with chicken feathers or droppings. Other birds, such as starlings, and bats have also been implicated in the establishment of saprophytic reservoirs of infection. Its spores are infectious not only to humans, but also to small animals such as dogs, cats and rats. The disease is not transmitted from human to human or from animal to human, but by the inhalation of air-borne conidia.

Histoplasmosis present as cutaneous lesion only in 6% of patients.^{2,3} Most commonly, they present as primary ulcers, often with annular, heaped up borders. They may also present as papules, nodules, or large plaque like lesions. Lesions are purpuric or crusted or may develop pustular caps and ulcerate which may be tender, red nodules due to inflammation or panniculitis.⁵

Upper alimentary canal histoplasmosis usually associated with systemic disease, especially in immunocompromised patients, such as in human immunodeficiency virus (HIV) infection.⁴ Isolated oral histoplasmosis as presenting sign,

without systemic involvement, with underlying HIV is very rare.^{3,4,5} Oral mucosal lesion present in half of the disseminated histoplasmosis with cutaneous lesion. Oral lesions are present in 30-50% patient in disseminated disease, and it may involve all part of the oral mucosa, most commonly affected sites are the tongue, palate and buccal mucosa.⁵

In our case patient present with oral mucosal lesion as presenting sign. Lesions of the oral mucosa usually start as painless papular swellings and later develop ulcer. The contiguous skin may also be involved. Biopsy of a mucosal or cutaneous lesion is the most rapid and specific diagnostic method for rapid institution of lifesaving therapy as culture may require up to a 4-week incubation period.^{2,3}

Histoplasmosis can be diagnosed by combined approach on the basis of clinical signs and symptoms with lab investigation like biopsy, cultures, serologic test, including compliment fixation test, immunodiffusion, and histoplasmin skin test and culture of fungus. Fungal culture is strongest diagnostic evidence for histoplasmosis.^{2,3,4}

The spores of *H. capsulatum* are visualized in sections stained with H&E, Gram, or Giemsa. Silver impregnation stains and electron microscopic studies show that *H. capsulatum* does not possess a capsule and that the inner portion of the clear space represents the cell wall of the fungus and the clear space itself is filled with granular material that separates the cell wall of the fungus from the cytoplasm of the macrophage.^{2,3} When inhaled, the latter sprout and transform into small budding yeasts that are 2 to 5 μm in diameter.⁶ In cultures at a temperature of 37°C, the organism also grows in the yeast like form.^{2,3}

CONCLUSION

Primary histoplasmosis of oral mucosa is a rare infectious disease. It is more common in HIV infected or immunocompromised patient as opportunistic infection.

So clinician always kept in mind as a differential diagnosis in cases of non-healing ulcer of mucosa or skin.

REFERENCES

1. Kauffman CA. Histoplasmosis: a clinical and laboratory update. *Clin Microbiol Rev.* 2007 Jan;20(1):115-32. Review. PubMed PMID: 17223625; PubMed Central PMCID: PMC1797635.
2. Hinshaw M., Longley B.J., Fungal Diseases. In: Elder D.A., Lever' S *Histopathology of the Skin* 10th ed. Lippincott Williams & Wilkins, a Wolters Kluwer business; 2009. P. 611-613.
3. Hay RJ, Ashbee HR. Mycology. In: Burns T., Breathnach S, Cox N, Griffiths C. *Rook's Textbook of dermatology.* 8th ed. Blackwell Publishing; 2010. p. 1736-1750.
4. Patil K, Mahima VG, Prathibha Rani RM. Oral histoplasmosis. *J Indian Soc Periodontol.* 2009 Sep;13(3):157-9. doi: 10.4103/0972-124X.60230. PubMed PMID:20379415; PubMed Central PMCID: PMC2848788.
5. Bhagwat PV, Hanumanthayya K, Tophakhane RS, Rathod RM. Two unusual cases of histoplasmosis in human immunodeficiency virus-infected individuals. *Indian J Dermatol Venereol Leprol.* 2009 Mar-Apr;75(2):173-6. PubMed PMID: 19293507.
6. Vijayan C, Suprakasan S, Kumar GN, Nair PS, Jayapalan S. Primary mucocutaneous histoplasmosis presents as oral ulcer . *Indian J Dermatol Venereol Leprol.* 2007 May-Jun;73(3):209. PubMed PMID: 17563929.
7. Sood N, Gugnani HC, Batra R, Ramesh V, Padhye AA. Mucocutaneous nasal histoplasmosis in an immunocompetent young adult. *Indian J Dermatol Venereol Leprol.* 2007 May-Jun;73(3):182-4. PubMed PMID: 17558052.

Source of Support: Nil
Conflict of Interest: Nil