

CASE REPORT

A RARE CASE REPORT OF ORAL CHROMOBLASTOMYCOSIS IN INDIA

Shahana Mohammed¹, Mahaboob Khan², Harish Kumar Kasetty³

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ABSTRACT: Chromoblastomycosis, caused by various dematiaceous (pigmented) fungi, is a chronic fungal infection of the skin and subcutaneous tissue, belong to the heterogeneous group of subcutaneous mycoses. These dematiaceous fungi exist as saprophytes in soil and gain entry into the skin via traumatic implantation. The characteristic sclerotic bodies are thick walled dark brown single or multicelled fungal clusters seen in tissue. Infection usually follows trauma involving the exposed areas, very rarely mucosa. The lesions may be warty plaques, nodules which ulcerate. We report a rare case of oral chromoblastomycosis in a 63yr old female patient involving only the tongue.

KEYWORDS: Chromoblastomycosis, dematiaceous fungi, verrucous plaques, copper penny bodies, medlar bodies.

INTRODUCTION: Chromoblastomycosis is a chronic granulomatous mycotic infection caused by different species of dematiaceous fungi common being *Fonsecaea pedrosoi*, *Cladophialophora carrionii*, *Fonsecaea compacta*, *Phialophora verrucosa* and other species.¹ Chromoblastomycosis begins as a small papule or warty growth and slowly spreads by the growth of satellite lesions.² There is a high prevalence of chromoblastomycosis in countries with a tropical or subtropical climate and more common amongst the rural population.³

Fonsecaea pedrosoi is the commonest causative agent implicated. These saprophytic fungi are in the environment in the soil debris, vegetation, wood splinters and thorns.⁴ It typically occurs on the exposed surfaces, mostly the lower limbs following any trauma.⁵ Usually it is an occupation related infection, predominantly seen in males, frequently affecting the agriculturists, labourers and those walking bare foot.^{2,3} Generally chromoblastomycosis usually affects the lower limbs, in some rare instances it involves the upper limbs and face. It mostly occurs as an opportunistic infection in immunocompromised individuals. The sclerotic bodies are thick walled, brownish, septate sclerotic bodies in the tissues, which are the characteristic and diagnostic finding of the disease.

The rare involvement of oral mucosa in chromoblastomycosis can cause some diagnostic difficulties. Hence, we report a rare case of oral chromoblastomycosis in a 63yr old female patient involving only the tongue, successfully managed with itraconazole.

CASE HISTORY: A 63yrs female housewife presented with complaints of rash over the face, swelling of the lips, difficulty in swallowing and opening of the mouth since five days. [Figure 1] On examination there was maculopapular rash over face, erosions and oedema of both the lips and bilateral conjunctivitis. Oral examination could not be done as the patient was unable to open her mouth.

The patient was on anti-retroviral therapy zidovudine 300mg, lamivudine 150mg and nevirapine 200mg once daily since two months with a CD4 count of 65cells. A clinical diagnosis of Stevens-Johnson syndrome was made and all the drugs were stopped and the patient was started on

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I.V decadran 2cc BD which was tapered to 1cc BD over a period of two weeks and then started on oral prednisolone 20mg OD which was tapered to 5mg OD over another 2weeks. All other investigations were normal. As the patient improved we noticed few thick whitish discrete, firm and non-tender verrucous plaques over the dorsum of the tongue. [Figure 2] The number of plaques were increasing in spite of the clinical improvement. KOH mount showed scattered copper coloured bodies (Medlar bodies) which was suggestive of chromoblastomycosis. Histopathology of these plaques showed chronic non-specific inflammatory cell infiltrate along with multiple brown pigmented muriform cells in clusters which confirmed chromoblastomycosis. [Figure 3, 4] On culture with Sabouraud's dextrose agar typical dark green colonies of chromoblastomycosis were seen. [Figure 5] Patient was started on oral itraconazole 100 mg twice daily for four weeks initially with complete clearance of the lesions. The treatment was continued for two more months. At six month follow-up, the patient had no new lesions. [Figure 6]

DISCUSSION: Chromoblastomycosis is a systemic fungal infection caused by traumatic implantation of a specific group of dematiaceous fungi. The infection mostly occurs on the exposed areas, being more common in men. The sites most commonly involved are lower extremities, uncommonly it may affect the hands, arms and buttocks, very rarely may also involve the face.

Thomas et al. first reported two cases of chromoblastomycosis from India in 1957.⁶ Literature search has only showed cutaneous chromoblastomycosis in India. There have been no reports of oral chromoblastomycosis so far. Chandran et al. reported 35 new cases of cutaneous chromoblastomycosis from central Kerala.⁷ In 2014 Asit M et al reported a new case of cutaneous chromoblastomycosis from Udaipur involving the forearm.⁸ Extensive literature search revealed that in 2012 Fatemi MJ et al reported a case of oral chromoblastomycosis involving the hard palate.⁹ We hereby report a very rare case of oral chromoblastomycosis involving only the tongue, which could probably be the first case of oral chromoblastomycosis from India.

A small, raised, erythematous asymptomatic papule develops slowly at the site of implantation, overtime producing a warty nodule, limited to the skin and the subcutaneous tissue. The most common complications are ulceration, secondary infection and scarring. Rarely malignant transformation to squamous cell carcinoma occurs.

Diagnosis is made by demonstration of thick walled sclerotic cells or pigmented fungus in the lesions. In culture on Sabouraud's dextrose agar all species are dark olive green to grey black velvety colonies. Fungal scraping study is a simple and non-invasive test for a bedside diagnosis.¹⁰ On direct microscopic examination of scrapings with 10% KOH typical, thick walled, globe-shaped, sclerotic cells, known as medlar bodies, resembling copper pennies are seen.

Histopathology also shows pigmented fungi with sclerotic or muriform bodies, which are thick-walled single cells or cell clusters seen as brown-colored "copper pennies." In our case numerous copper penny sclerotic bodies seen scattered which are confirmatory.

Drug therapy consists of long courses of high dose antifungals like Itraconazole and terbinafine given for a minimum of 6–12 months. The newer broad-spectrum triazoles such as posaconazole and voriconazole are promising drugs for treating deep mycoses.

Though oral chromoblastomycosis is very rare, it is important to consider it as one of the differentials for oral lesions. All the oral lesions should be investigated for deep fungal infections, especially in the immunocompromised, as chronic chromoblastomycosis may lead to malignancy.

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We report a rare case of oral chromoblastomycosis involving only the tongue as an opportunistic infection in a retro positive patient. Our patient was a housewife from a rural area without any history of trauma and a very good response to oral itraconazole 100 mg twice daily.

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Fig. 1: Picture showing swelling of both the lips with erosions, bilateral conjunctivitis and maculopapular rash over the face.



Fig. 1

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Fig. 2: Thick whitish plaques of chromoblastomycosis on the tongue prior to the treatment.



Fig. 2

Fig. 3: (A) Histopathology showing sclerotic bodies (H&E, 4x) (B) & (C) Histopathology showing sclerotic bodies in clusters which confirms chromoblastomycosis. (H&E, 40x).

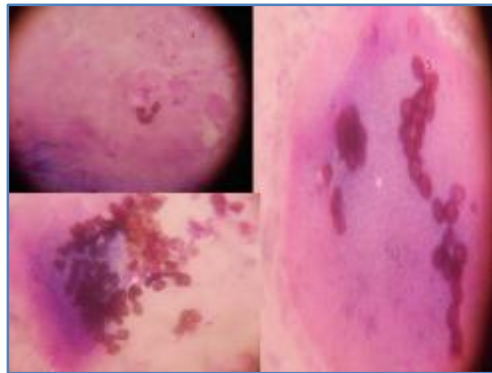


Fig. 3

Fig. 4: (A) Histopathology showing sclerotic bodies in clusters which confirms chromoblastomycosis. (H & E, 40x) (B) Histopathology showing sclerotic bodies (H & E, 4x).



Fig. 4

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Fig. 5: Culture showing typical dark green colonies of chromoblastomycosis.

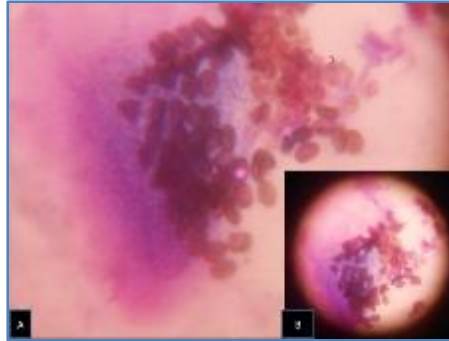


Fig. 5

Fig. 6: Complete healing of the lesions following therapy.



Fig. 6

AUTHORS:

1. Shahana Mohammed
2. Mahaboob Khan
3. Harish Kumar Kasetty

PARTICULARS OF CONTRIBUTORS:

1. Assistant Professor, Department of DVL, Gandhi Medical College and Hospital, Secunderabad.
2. Professor, Department of Pulmonology, Gandhi Medical College and Hospital, Secunderabad.
3. Senior Resident, Department of DVL, Gandhi Medical College and Hospital, Secunderabad.

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NAME ADDRESS EMAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Shahana Mohammed,
House No. B-32, 1-8-450/1,
Indian Airlines Colony,
Begurapet, Secunderabad, India.
E-mail: drshahena@gmail.com

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