

Chromoblastomycosis with skeletal involvement: A case report

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Abstract Chromoblastomycosis is a chronic cutaneous and subcutaneous infection of tropical and subtropical climates, caused by dematiaceous (black pigmented) fungi. Infection is thought to be secondary to trauma or autoinoculation. It rarely involves other organ systems of the body especially underlying bones. Many cases with similar and additional features have been reported. We describe one case of this infection in a 20-year-old female with skeletal changes along with review of literature.

Keywords

Chromoblastomycosis, dematiaceous fungi, skeletal involvement.

Introduction

Chromoblastomycosis, also known as ‘Fonseca’s disease’ ‘Pedroso’s disease’ or ‘Verrucous dermatitis’, was first reported in Brazil in 1914 by a German physician, Max Rudolph.¹ After one year in 1915, Lane and Medlar, described the sclerotic cells which were then called as Medlar bodies/sclerotic bodies/copper-penny bodies or muriform cells. Chromoblastomycosis is caused by pigmented fungi like *Fonsecaea compacta*, *F. pedrosoi*, *Phialophora verrucosa*, *Cladophialophora carrionii* and *Rhinocladiella aquaspersa* and occasionally by *Exophiala spinifera*, *Aureobasidium pullulans* and *Chaetomium funicula*.^{2,3}

It is usually caused by traumatic inoculation usually on lower limbs like foot, legs, and arms, by contaminated soil or injury with thorns or wood splinters.^{4,5} The disease spreads slowly

from initial papule to warty plaques and tumorous, giving it a cauliflower-like appearance. Occasionally, the lesions are suppurative which may be multiple in immunosuppressed hosts.

As in all fungal diseases, the diagnosis requires a high index of suspicion. It is frequently delayed, probably due to rarity of the infection, and failure to elicit the usual history of exposure. Sometimes, culture for chromoblastomycosis organisms is negative but the diagnosis is still made on clinical picture supported by typical histological findings. A prolonged course of antifungal therapy is curative in most superficial cases but rarely surgical intervention is required.⁶⁻¹¹ We report a case of chromoblastomycosis which had infected multiple bones along with review of literature.

Case Report

A 20-year-old female, resident of Lahore presented at Department of Dermatology, Services Hospital, Lahore, with complaints of hyperpigmented, thickened plaques on both sides of face for the last 8 years. Initially, the

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Figure 1 Hyperpigmented verrucous plaque over face.



Figure 2 Second left toe showing resorption.



Figure 3 Resorption of left index and little finger.

lesions started as papules which gradually progressed to hyperpigmented verrucous plaques. There was no history of pain, fever, or itching. Similar scattered lesions appeared on ears, arms, dorsum of right hand, trunk and legs over the period of 5 years. There was also history of scaling, crusting and ulceration on the lesions. She took treatment from various hospitals but there was no satisfactory response to any therapy.

Physical examination revealed no abnormality. Cutaneous examination showed multiple well-demarcated hyperpigmented, hyperkeratotic verrucous plaques involving both sides of face and ear (**Figure 1**). Multiple skeletal findings were noted like telescoping of left second toe (**Figure 2**), absorption of terminal phalanx of left index and left little finger (**Figure 3**).

On laboratory investigations, blood, urine and stool examinations were normal. Gram staining and smear for Leishman-Donovan (LD) bodies were negative. The bacteriological examination of pus showed no AFB or other micro-organism. Hepatitis B surface antigen, anti-HCV and HIV screening were negative. X-ray chest, CT scan and abdominal ultrasonography were normal.

Radiological findings showed lytic lesions in terminal phalanx of left index finger, loss of terminal phalanx of little finger, lytic lesions of second metatarsal and digits of left second toe. Bone imaging studies showed two small focal lesions in left distal index and little fingers. Non-homogenous tracer distribution was noted in palm of right hand. Delayed images showed multiple foci of moderately increased tracer uptake involving distal phalanges of left index and little fingers, proximal and distal shaft of left tibia, left ankle joint and foot. Conclusion of bone imaging studies was multi-focal bone pathology.

Histopathology of skin revealed ulcerated stratified squamous epithelium with underlying dense chronic inflammatory cell infiltrate and extensive giant cell reaction. Numerous pigmented fungal hyphae were also seen. Fungal smear/KOH preparation showed numerous pigmented septate hyphae. Fungal culture was positive for *Phialemonium* species. *Phialemonium* species represents an intermediate genus between *Acremonium* and *Phialophora*. Diagnosis was confirmed as chromoblastomycosis. The patient is currently on oral anti-fungal (voriconazole), oral and

topical antibiotics, vitamin D3 and calcium supplementations with satisfactory response.

Discussion

Most of the cases are seen in middle age group especially agricultural workers, people walking barefoot and is rarely reported in children.^{4,5,12,13} Satellite lesions are formed due to autoinoculation by scratching. Complications include serous discharge, ulceration, lymphedema, elephantiasis and secondary infection and very rarely squamous cell carcinoma. The disease has a good prognosis, but recurrences are common. The key elements in the diagnosis of this infection are taking a detailed history, clinical findings, microscopic demonstration of pathognomonic, fungal culture and tissue biopsy for histopathology.¹⁴ In our case, cutaneous and mucosal involvement was noted along with skeletal involvement. The condition has to be differentiated from some other clinical entities like tuberculosis verrucosa cutis, leishmaniasis, sporotrichosis, blastomycosis, paracoccidioidomycosis, tertiary syphilis and leprosy.

In our case, well-demarcated hyperpigmented verrucous plaques with discharge, histopathology report, positive fungal scraping and positive fungal culture favored the diagnosis of chromoblastomycosis. Rarity of this case was skeletal involvement and very few cases have been reported in the past with extracutaneous spread.^{15,16,17} It was confirmed by bone X-rays and ^{99m}Tc-MDP bone imaging studies.

Tuberculosis verrucosa cutis clinically mimics this infection but it was ruled out due to the absence of typical histopathology and negative bacteriological examination for AFB. Where leishmaniasis and sporotrichosis are endemic, they must be excluded. The clinical features of both conditions are similar to

chromoblastomycosis but negative report of smear for LD bodies and histopathology report led to the exclusion of these conditions.¹⁸

Blastomycosis and paracoccidioidomycosis were ruled out by typical history, characteristic histology and absence of any systemic illness in our patient. In tertiary syphilis and leprosy, characteristic features are nodular or ulcerated lesions but no neurological or cardiovascular involvement and negative serological tests for syphilis are the key points to differentiate this condition from chromoblastomycosis infection.¹⁸

Conclusion

Chromoblastomycosis is not uncommon in our country. An early diagnosis and prompt treatment can save the patient from deeper involvement and complications.

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