Case Report

Chromoblastomycosis: a case report with literature review

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ABSTRACT

Chromoblastomycosis is a chronic progressive cutaneous fungal infection caused by several naturally pigmented fungi that is frequently misdiagnosed clinically because of its polymorphic clinical presentation. We present a case of cutaneous chromoblastomycosis in a 52 year male in the right leg that was mimicking mycetoma/cutaneous tuberculosis clinically. Later, it was diagnosed as chromoblastomycosis in histopathological examination on identification of characteristic sclerotic/medlar bodies. We found combination of chemotherapy and wide local excision was effective in the management of localized chromoblastomycosis with favorable outcome. To conclude, chromoblastomycosis should be considered in the differential diagnosis of chronic cutaneous infections and one has to look for sclerotic bodies particularly in granulomatous lesions as it has specific treatment.

Keywords: Chromoblastomycosis, Sclerotic bodies, Medlar bodies, Granuloma

INTRODUCTION

Chromoblastomycosis is a slowly progressive cutaneous mycosis caused by dematiaceous pigmented fungi in tropical and subtropical climates. It was first described in 1914, in Brazil by Max Rudolph, a German physician.1 The most common causative organisms are Fonsecaea pedrosoi, Phialophora verrucosa, Fonsecaea compacta and Cladophialophora carrioni. The fungi are usually found in soil, wood, and rotting vegetables and infection often results from trauma such as puncture from a splinter of wood. The lower limbs and hand are commonly affected and usually present as nodular verrucous lesions.2 In 1915, Medlar and Lane described the characteristic histologic picture of sclerotic bodies or medlar bodies.3

CASE REPORT

We report a case of cutaneous chromoblastomycosis in a 52 year male, a farmer who developed a nodular lesion over the right leg following a trauma during the field work. Subsequently, it gradually increased in size and number with ulcerations as shown in Figure 1 over a period of 3 months. On examination he had multiple warty and nodular lesions measuring 1-2 cm in diameter with ulcerated surface covered with exudates and tiny brownish black dots over the anterior aspect of lower part of the right leg. No constitutional symptoms were seen. He had no inguinal node enlargement.

Initial skin biopsy was reported as suppurative lesion elsewhere and was not responding to conventional antibiotic treatment. Subsequently he developed multiple lesions. He is not a diabetic or hypertensive. His complete blood counts, basic biochemical investigations were within normal limits. ELISA for HIV was negative. Chest X-ray was within normal limits. Clinically, cutaneous tuberculosis and mycetoma was suspected. As the lesion was progressive, non-healing and not responding to conventional antibiotics a wide excision biopsy was done.
round to oval golden brown sclerotic/medlar bodies were seen in single and clusters resembling ‘copper penny’ within the giant cells and also lying freely in the abscesses as in Figure 3. Septation was also noted. Ziehl Neelsen stain for acid fast bacilli was negative. As the pigmented copper penny sclerotic bodies were easily found a diagnosis of chromoblastomycosis was made.

He was put on oral itraconazole 200 mg/day for 3 months and the raw area was covered with delayed skin graft as in Figure 4a. The lesion healed well as in Figure 4b and there was no recurrence during one year follow up.

**DISCUSSION**

The term chromoblastomycosis is used for those chronic granulomatous fungal infections which characteristically
produced pigmented hard yeast cells called muriform cells or sclerotic bodies and belong to the group of fungal infections called phaeohyphomycosis caused by pigmented fungi. *Fonsecaea pedrosoi* is the commonest agent of chromoblastomycosis and its identification requires fungal culture studies.4

The lesion is commonly seen in bare footed agricultural workers in the lower extremities as seen in our case.4 Other unusual sites of involvement are upper extremities, face, penile shaft, vulva, tonsil, pleural cavity, ileocaecal region, laryngotracheal area and ala of nose.4,6 It usually follows a traumatic implantation of the etiologic agent beneath the epidermis and subsequently it spreads to the adjacent skin, thus causing satellite lesions as seen in our case.5,7 Hematogenous spread is rare, commonly occurs with *Cladosporium Trichoides*.3 The disease commonly occurs in middle-aged males (30-50 years). However, its occurrence in children and elderly was also reported.5,8,9 Chromoblastomycosis is concentrated in tropical and subtropical climates. Madagascar of African continent has the highest incidence in the world followed by Brazil. In India, it was first reported by Thomas et al in 1957, from Assam. Since then many cases have been reported across the country, majority from the Himalayan belt.6

We report this case from Tamilnadu state of South India.

The lesions are polymorphic ranging from ulcer to papulonodular or verrucoid hence clinically mistaken for other lesions.2,4,6 Secondary bacterial infections are common and repeated infections may lead to lymphatic fibrosis and elephantiasis of the legs. Recurrences are common and this disease has a potential to predispose for the development of squamous cell carcinoma.5,7

Chromoblastomycosis must be differentiated from tuberculosis verrucosa cutis, leprosy, leishmaniasis, mycetoma, sporotrichosis, psoriasis and tertiary syphilis.2,4,9 In our case it was mimicking mycetoma/verrucous tuberculosis. The diagnosis is based on the demonstration of sclerotic bodies in the tissue sections/KOH mounts and on the isolation and identification of the causative agent.

Diagnosis can be made easily in H&E stained sections when the organisms are in good numbers by demonstrating diagnostic ‘sclerotic bodies’. Because of their natural pigmentation the sclerotic bodies can be seen even in unstained sections and they retain their pigment even after ‘destaining’ the H&E stained sections.5 In our case, we found sclerotic bodies easily and it could be made out in unstained sections also. The sclerotic bodies are brown round to polyhedral approximately 5-8 µ, resembling copper penny with mature thick wall and characteristic septation.

Examination of unstained and destained sections is easy and less cumbersome, cost effective than special stains and also removes the possibility of artifacts with special stains. Sclerotic bodies must be distinguished from hemosiderin or formalin pigment in tissue sections. Artifactual pigments are irregular, dense brown colored and don’t show characteristic division, whereas sclerotic bodies have definite morphology with intracellular division. Histology has limitation in identifying the genus and exact species, as all of them look similar morphologically we have to rely on culture studies.4

Tissue responses to chromoblastomycosis includes pseudoepitheliomatous hyperplasia, intraepidermal and dermal microabcess, giant cell response, epithelioid granulomas, lymphoplasmacytic infiltration.4,5 We found all the above histopathological findings as given in Figure 2 and 3 in our case. There are major changes in cell wall content while adapting from hyphae to sclerotic bodies with rhamnose, palmitic, oleic acid and arachidonic acid being major composition in the latter compared to glucose, mannose, galacto-fructose, glucosamine, linoleic acid and ergosterol in the former.4

The verrucous lesions typically show small tiny black dots on the surface of each papilla and they represent microabcesses seen in histology and is ideal to sample these sites for histology or cytological examination. These microabcesses contain pathogenic spores, which are in the process of transepithelial migration by which the spores are released to the external environment and their autoinoculation and or lymphatic spread could explain the ‘satellite lesions’ as seen in our case. Similar transepithelial migration is also seen in rhinosporidiosis, perforating granuloma annulare.

Many cases are missed cytologically due to lack of clinical suspicion. Cytology smears show mixed inflammatory infiltrate and the fungal spores look orange to reddish brown and are of the size of RBCs, dispersed in singe unlike red cells which appear pink and are seen in clusters. Septation is a characteristic but not a consistent feature for chromoblastomycosis.5

Surgery was considered the treatment of choice for chromoblastomycosis before the advent of triazole antifungal agents. However, currently with the availability of potent antifungal agents, chemotherapy has become the first-line of treatment with itraconazole and terbinafine being the drugs of choice, while surgery is used only for limited or small lesions. Antifungal therapy should be continued until complete clinical resolution. A combination of fortnightly liquid nitrogen cryotherapy and pulsed monthly itraconazole was shown to shorten the duration of therapy and therefore could be a cost-effective approach for treatment of chromoblastomycosis.9 Various treatment modalities like cryotherapy, thermotherapy, laser therapy and surgical excision are available.7,8

In our case diagnosis was made in the wide excision biopsy specimen, then chemotherapy with itraconazole 200 mg/day for 3 months was given and the wound was covered with delayed split skin graft which completed
healed as given in Figure 4a and 4b. The patient is under follow up and there was no recurrence for the past one year. So, this combined chemotherapy with surgical management is found to be effective and reduces the duration of treatment with a favorable outcome.

**CONCLUSION**

Chromoblastomycosis, although infrequent, must be considered in the differential diagnosis of chronic skin lesions particularly cutaneous tuberculosis in patients from tropical and sub-tropical regions so that an early appropriate therapy can be instituted. Chromoblastomycosis spreads very slowly, is rarely fatal, and usually have good prognosis; but is a therapeutic challenge. In our case we found combination of chemotherapy and surgery was effective in management of localized disease.

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